



Salivary Gland Choristoma of the Middle Ear: A Case Report

Keyvan Aghazadeh¹, Ebrahim Karimi¹, Amirsina Sharifi¹

1- Head and Neck Surgery Research Center, Tehran University of Medical Sciences, Tehran, Iran

Received: 15 July 2015

Revised: 12 August 2015

Accepted: 22 October 2015

ARTICLE INFO

Corresponding author:
Amirsina Sharifi

Email:
a.s_sharifi@yahoo.com

Keywords:
Salivary gland choristoma,
Middle ear mass,
Hearing loss,
Ossicular chain malformation

ABSTRACT

Salivary gland choristoma of the middle ear is a rare mass characterized by the presence of normal salivary gland tissue in the middle ear cavity. This mass is usually accompanied with ossicular chain and facial nerve anomalies. Total surgical excision is a treatment of choice if the facial nerve is intact. Here, we describe a case of salivary gland choristoma of the middle ear, and discuss our experience. Our patient was a 41-year-old man presented with 6 months history of hearing loss. Intra-operatively, we detected a soft, pinkish mass with evidence of ossicular chain malformation. Patient showed hearing development in following up after three months.

Citation: Aghazadeh K, Karimi E, Sharifi A. **Salivary Gland Choristoma of the Middle Ear: A Case Report.** Case Rep Clin Pract 2016; 1(1): 15-8.

Introduction

Since 1961 that Taylor and Martin reported the first case of salivary gland choristoma of the middle ear, there are few reports of this mass in the literature (1, 2). Ectopic location of a histologically normal salivary tissue is the definition of salivary gland choristoma (3, 4). Given this scarce prevalence, the proper diagnosis would be difficult; as in most cases, patients present with non-specific symptoms (5, 6). Here, we present a 41-year-old man with initial symptoms of conductive hearing loss and serous otitis media.

Case Report

A 41-year-old man was referred to our

hospital complaining hearing loss initiated six months prior to admission. He had no previous history of hearing lost. His history was negative for otorrhea, otalgia, vertigo and facial nerve palsy. He had also negative family history of any hearing related symptoms. Right ear otoscopic examination revealed opaque tympanic membrane with no demonstrable perforation. There was also subtle retraction in right tympanic membrane. Left ear was perfectly normal in otoscopic examination. There was no evidence of facial nerve palsy, and he did not show any remarkable abnormal features in the head and neck examination.

Pure tone audiometry demonstrated 60 dB

conductive hearing loss in the right ear. Air bone gap was 40 dB. Computed tomography (CT) scan of the temporal bone (Figure 1) showed soft tissue density surrounding the ossicles that filled the epitympanum cavity. Mastoid antrum was hypoplastic in right side and mastoid air cells were opaque. In addition, displacement of ossicle chain was seen in right side. The CT scan findings were in accordance with normal structure in the left side.

Patient underwent general anesthesia and facial nerve monitoring. Exploratory tympanotomy was performed through trans-

canal tympano-meatal flap. Middle ear was full of glue and serous liquid and there was no evidence of cholesteatoma. Ossicle chain was displaced and stapes was missing. A soft, rubbery, pinkish mass measuring near $15 \times 5 \times 3$ mm in size was found on promontory. There was no evidence of mass involvement of the facial nerve as any abnormalities in the course of the facial nerve. Histopathologic examination of the resected mass reported seromucinous glands covered by normal mucosa coated with respiratory epithelium.

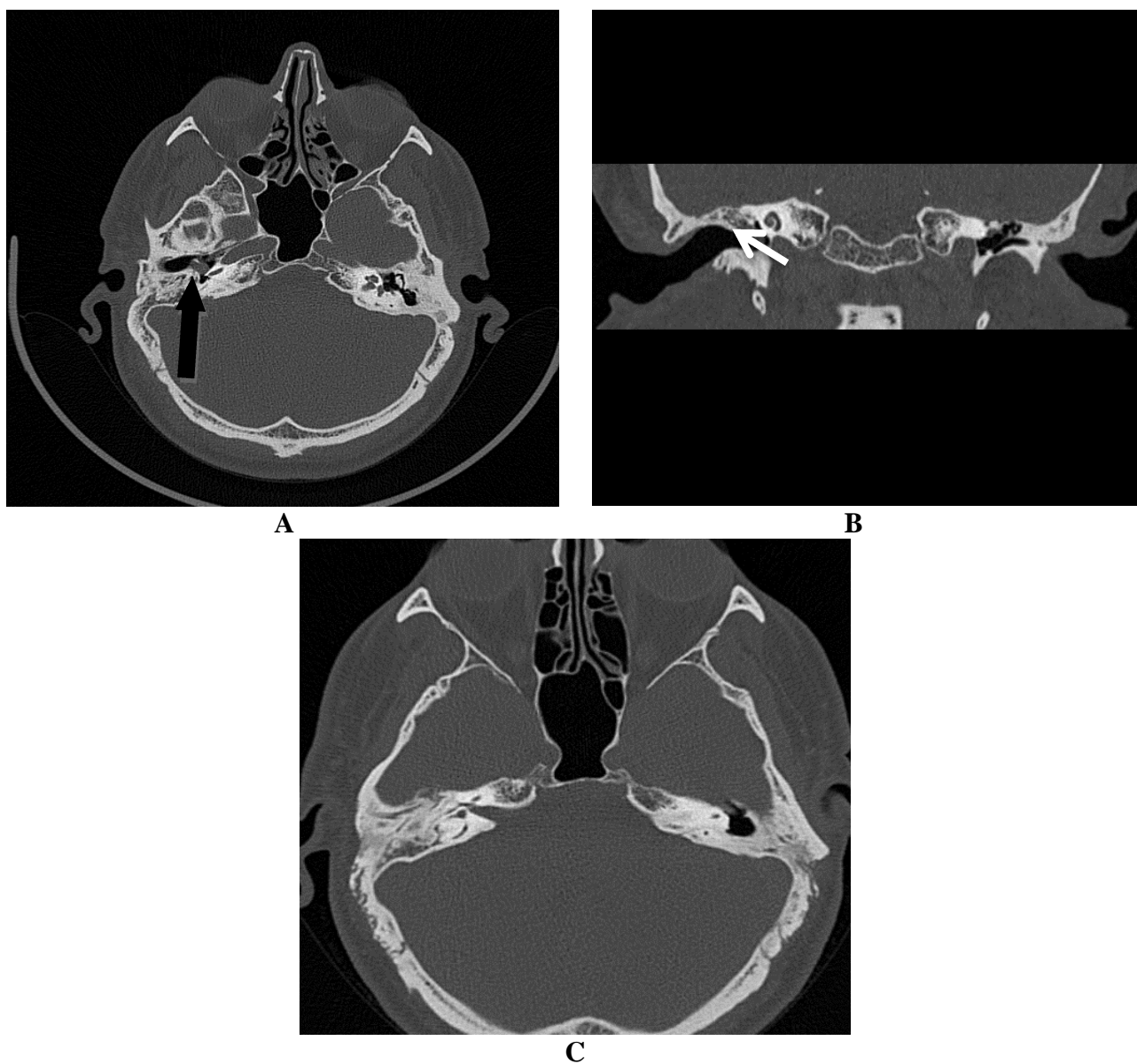


Figure 1. Computed tomography (CT) scan of salivary gland choristoma in the right middle ear cavity; A. Temporal bone CT scan shows a soft tissue mass occupying the right middle ear cavity (thick arrow); B. Axial plane on CT scan shows soft tissue attenuation in the mastoid air cells, representing the presence of mastoiditis (thin arrow); C. The course of the facial nerve is intact (arrow head).

There was no postoperative complication. Three months after the operation, pure tone audiometry showed improvement and patient's hearing was slightly better (10 dB improvement of air bone gap in three-month follow-up).

Discussion

Ectopic salivary gland tissue may be found in the various sites of the head and neck area such as middle ear, gingival, lower neck and anterior chest wall (7-11).

Although several studies tried to elucidate the pathophysiology of ectopic salivary gland tissue in middle ear cavity, the exact mechanism in embryologic development of this mass has not been determined (12). So far, based on the Patten theory, the ectopic expansion of the remnant parotid epithelium or pharyngeal endoderm seems to be associated with salivary gland of the middle ear cavity (5). In this regard, Buckmiller et al. suggested that salivary gland choristoma, hearing loss, ossicular chain abnormalities, facial nerve anomalies and branchial arch anomalies are components of a syndrome (10). Besides, it has been reported that ectodermal anomalies such as hemifacial atrophy and deformed auricles are accompanied with salivary gland choristoma (3).

Salivary gland choristoma of the middle ear have been seen in patients ranging from 11 months to 52 years of age with a mean age of 17.5 years (3, 13).

Thus, it can be added to the differential diagnosis of the middle ear cavity masses in every age group; beside congenital cholesteatoma, glomus tumor, neuroma, dermoid cyst, teratoma and lymphoma which are presenting with unilateral hearing loss without perforation of the tympanic membrane (8, 11, 12).

Hearing loss and tinnitus are defined as the most common symptoms. In addition, 24% of the patients with salivary gland choristoma of the middle ear cavity showed serous otitis media (2, 9). In our patient, six months of the hearing loss forced him to seek medical attention.

Although the literature review revealed an

overwhelming predominance of left-side involvement (14), but in our case, the choristoma was located in right middle ear cavity.

The most challenging step in treatment of salivary gland choristoma of the middle ear is high incidence of facial nerve involvement. Thus, conservative management of this mass is highly emphasized in the literature (4, 12, 15). In our case, there were no evidences of tumor involvement in the facial nerve in the preoperative evaluation using the CT scan.

Considering the fact that choristoma is a normal tissue and there is no report of recurrence even when it was removed incompletely, the total excision would be the treatment of choice if the facial nerve is intact (13, 16). This treatment would also prevent further possible development of malignant tumor, which is reported to arise from choristoma (17, 18).

Conclusion

Salivary gland choristoma of the middle ear is an extremely rare mass with nonspecific symptom of unilateral conductive hearing loss. Considering the fact that the radiologic study of the head and neck region cannot provide definite diagnosis and only postoperative histopathological evaluation shows the nature of the mass, it should be considered as a differential diagnosis of middle ear cavity masses.

Conflict of Interests

Authors have no conflict of interests.

Acknowledgments

This paper was not supported by special grants.

References

1. Taylor GD, Martin HF. Salivary gland tissue in the middle ear. A rare tumor. *Arch Otolaryngol* 1961; 73: 651-3.
2. Chen S, Li Y. Salivary gland choristoma of the middle ear. *Ear Nose Throat J* 2015; 94(2): E9-E12.
3. Ha SL, Shin JE, Yoon TH. Salivary gland choristoma of the middle ear: a case report. *Am J Otolaryngol* 2000; 21(2): 127-30.
4. Fois P, Giannuzzi AL, Paties CT, Falcioni M. Salivary gland choristoma of the middle ear. *Ear*

- Nose Throat J 2014; 93(10-11): 458-64.
5. Toros SZ, Egeli E, Kilicarslan Y, Gumrukcu G, Gokceer T, Noseri H. Salivary gland choristoma of the middle ear in a child with situs inversus totalis. *Auris Nasus Larynx* 2010; 37(3): 365-8.
 6. Yazici D, Cetik F. An infrequent mass of the middle ear: salivary gland choristoma. *Arch Otolaryngol Head Neck Surg* 2006; 132(11): 1260-2.
 7. Shin CE, Kim SS, Chwals WJ. Salivary gland choristoma of the anterior chest wall. *J Pediatr Surg* 2000; 35(10): 1506-7.
 8. Ookouchi Y, Honda N, Gyo K. Salivary gland choristoma of the middle ear in a child: a case report. *Otolaryngol Head Neck Surg* 2003; 128(1): 160-2.
 9. Supiyaphun P, Snidvongs K, Shuangshoti S. Salivary gland choristoma of the middle ear: case treated with KTP laser. *J Laryngol Otol* 2000; 114(7): 528-32.
 10. Buckmiller LM, Brodie HA, Doyle KJ, Nemzek W. Choristoma of the middle ear: a component of a new syndrome? *Otol Neurotol* 2001; 22(3): 363-8.
 11. Rinaldo A, Ferlito A, Devaney KO. Salivary gland choristoma of the middle ear. A review. *ORL J Otorhinolaryngol Relat Spec* 2004; 66(3): 141-7.
 12. Lee DK, Kim JH, Cho YS, Chung WH, Hong SH. Salivary gland choristoma of the middle ear in an infant: a case report. *Int J Pediatr Otorhinolaryngol* 2006; 70(1): 167-70.
 13. Vasama JP, Ramsay H, Markkola A. Choristoma of the middle ear. *Otol Neurotol* 2001; 22(3): 421-2.
 14. Enoz M, Suoglu Y. Salivary gland choristoma of the middle ear. *Laryngoscope* 2006; 116(6): 1033-4.
 15. Hintz EB, Yeane GA, Buchberger GK, Vates GE. Intracranial salivary gland choristoma within optic nerve dural sheath: case report and review of the literature. *World Neurosurg* 2014; 81(5-6): 842-4.
 16. Morimoto N, Ogawa K, Kanzaki J. Salivary gland choristoma in the middle ear: a case report. *Am J Otolaryngol* 1999; 20(4): 232-5.
 17. Kartush JM, Graham MD. Salivary gland choristoma of the middle ear: a case report and review of the literature. *Laryngoscope* 1984; 94(2 Pt 1): 228-30.
 18. Peters BR, Maddox HE, Batsakia JG. Pleomorphic adenoma of the middle ear and mastoid with posterior fossa extension. *Arch Otolaryngol Head Neck Surg* 1988; 114(6): 676-8.