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Case Report

Rare Case of Primary Ceruminous Pleomorphic Adenoma of External Ear Canal

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Abstract

Pleomorphic adenoma of the external ear canal, also known as chondroid syringoma is a rare benign space-occupying lesion in this anatomical site.

We report a rare case of primary pleomorphic adenoma of the RT external auditory canal in a healthy 31-years-old male patient and discuss its clinical presentation, diagnosis, imaging study and treatment options.

A 31-years-old male presented with ongoing right ear fullness and hearing deterioration over a 2 -years period. He denied any ear drainage. A firm rubbery mass around the outer 3rd part of the external auditory canal was noted on physical examination.

High resolution CT scan of the temporal bone showed tumor mass with no evidence of bone destruction and without evident involvement of the tympanic membrane, MRI demonstrated clear separation between the RT parotid gland and the lesion.

The patient underwent surgical excision by a retroarticular approach for obtaining a complete resection of the tumor and the entire skin of the external auditory canal with wide canaloplasty.

Final histology and immunohistochemistry panels confirmed a diagnosis of primary ceruminous pleomorphic adenoma of the external auditory canal, and three months following the surgery, patient is free of any recurrent disease or hearing loss.

Keywords: Pleomorphic Adenoma; External Ear Canal

Introduction

Pleomorphic adenoma of the external ear canal is a rare entity and described in the literature at least in 36 studies up to date [1-18], the first case of a pleomorphic adenoma of the external auditory canal was reported by Mark and Rothberg in 1951 [19].

In the head and neck region, pleomorphic adenomas occur most frequently in the major salivary glands but may also arise from the minor salivary glands of the upper respiratory and alimentary tracts. Histologically they contain morphologic diversity with both epithelial and mesenchymal components in various proportions.

Approximately 5% of all external ear neoplasms are benign adenomas [4].

Here we report a case study of an interesting primary pleomorphic adenoma of the external ear canal and a literature review of this rare entity.

Case Report

We present a 31 years old healthy male with a 2-year progressive sensation of opacity and hearing loss in his RT ear, denies any tinnitus or vestibular symptoms, diagnosed with RT exostosis in HMO and referred to our center for clinical evaluation.

High resolution computer tomography of the ears includes coronal reconstructions revealed a soft tissue fullness with dimensions of 12 mm x 13 mm x 10 mm that seems to be arrived from the anterior wall of RT external auditory canal without evidence of any bone erosion and with proximity to the tympanic membrane without its involvement (Figure 1).

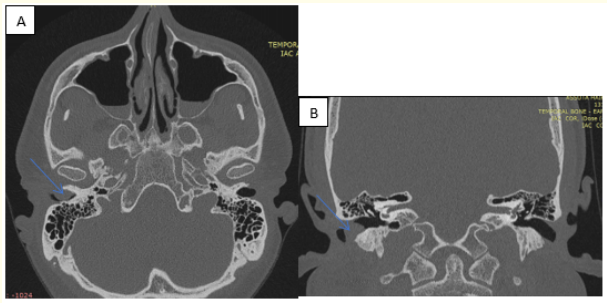

Figure 1: HRCT scans of the ears demonstrating the RT soft tissue fulness (arrow), A-axial, B-coronal view.

Audiometry demonstrated RT down slopping conductive hearing loss curve up to 80dB in 3KHZ (Figure 2).

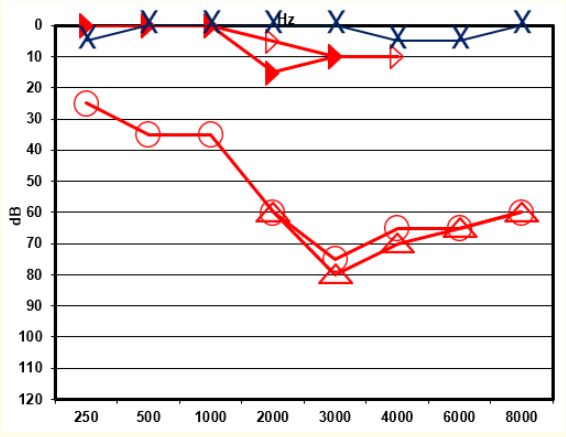

Figure 2: Audiometry confirmed RT conductive hearing loss.

MRI of the ears with contrast showed: homogeneous enhancement of the lesion with gadolinium on T1 sequence imaging. T2 sequence shows a clear separation between the parotid gland and the SOL described (Figure 3).

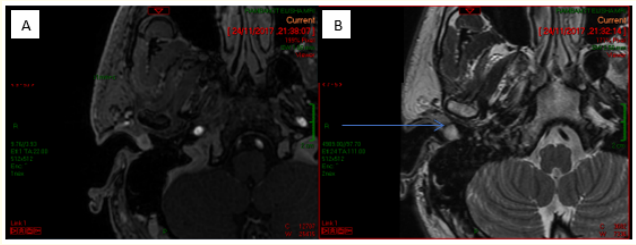

Figure 3: MRI (A) T1 with gadolinium and (B) T2 sequence, demonstrate aclear seperation between the parotid gland and the finding and homogenic enhanement of the lesion with gadolinium (arrow).

This neuro-radiological imaging consult concluded that this finding is highly suspected for primary pleomorphic adenoma that arrives from the ceruminous gland of the external ear canal.

Patient underwent excisional biopsy in the operating room under general anesthesia, specimen resected completely and wide canaloplasty was performed to ensure free margins of disease (Figure 4) and specimen sent to pathology lab in our institute that confirmed the diagnoses of pleomorphic adenoma by immunohistochemical staining showing positive cells with cytokeratin, actin and S-100 and a low ki67 proliferation index (Figure 5).

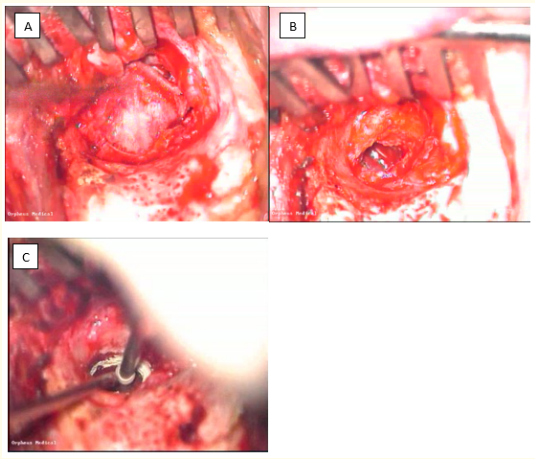

Figure 4: Intraoperative photos (A) before and (B) after the surgical exision, (C) wide cannaloplasty

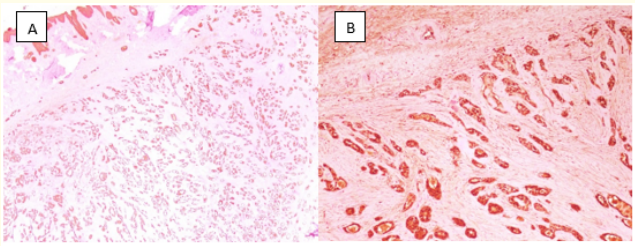

Figure 5: Histopathologic confirmation of ceruminous pleomorphic adenima. (A) tubules positive with cytokeratin immunostain, (B) tubules positive with S100 (myoepithelium).

Discussion

Pleomorphic adenoma also known as "mixed tumor" is a well-known benign neoplasm in the head and neck region, it is the most common benign neoplasm in the parotid gland (80%) [2], characterized by neoplastic proliferation of parenchymatous glandular cells along with myoepithelial components that has a malignant transformation potential of 6% [2].

Inside the external auditory canal this type of space occupying lesion described in a small number of articles only[6] and considered as a rare neoplasm.

Histologically there are 2 variants (epocrine and ecrine) [3], In the literature there is a controversy weather it originates from the ceruminous gland or ectopic salivary gland, recent studies strongly suggesting that this type of lesion arriving from ceruminous gland origin [6]. Diagnoses criteria suggested by presence of niches of polygonal cells, tubuloalveolar structure with glandular elements and ductal structure, occasional keratin cysts and chondroid matrix (basophil) or hyaline (eosinophil) [3].

The most common clinical presentation is a painless lump accompanied with fullness sensation and discharge, with least common hearing loss as the main complain (5 out of 25 patients) [11].

Since a malignant transformation is potential, the accepted treatment for this neoplasm is complete wide surgical excision that provides very low recurrence rate.

In our case the main complain of our patient was hearing loss, audiometry confirmed pure down sloping hearing loss curve up to 80 dB at 3 KHz, a wide surgical excision with canaloplasty was done to ensure complete excision of the lesion and to avoid any recurrence, follow up revealed no subjective hearing loss and external ear canal was free of neoplasm recurrence.

Conclusion

Pleomorphic adenoma of the external auditory canal is considered as a rare entity that was described in the literature only as case studies, our case report describing a 31 years old previously healthy man that was referred to our institute due to complains of hearing loss without any other ontological, vestibular symptom or complains, a pleomorphic adenoma of external ear canal was confirmed histopathological and resected completely with wide canaloplasty for ensuring free margins of disease for minimizing recurrence rate, several months later patient is without symptoms or any recurrence and reported of hearing back to normal.

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