Pleomorphic Adenoma of the Middle Ear

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Citation

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Abstract

Pleomorphic adenoma of the middle ear is an extremely rare disease. This case report demonstrated a patient of recurrent pleomorphic adenoma of the middle ear with emphasis on differential diagnosis from middle ear adenoma. Possible sources of disease were discussed. Immunohistochemical examination plays significant role for making a defined diagnosis.

INTRODUCTION

Pleomorphic adenoma (mixed tumor) arising in the ear is rare, and extremely rare in the middle ear. Not much has been reported for this disease concerning the diagnosis, treatment and prognosis. We report a case of recurrent pleomorphic adenoma of the middle ear and review the literature.

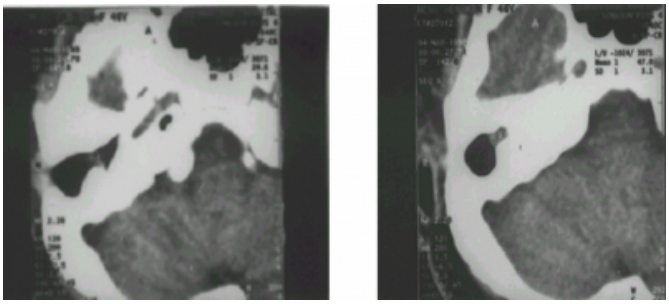
CASE REPORT

A 46-year-old woman first presented in 1979 with tinnitus and gradual hearing loss in the right ear. Physical examination showed bulging at both the posterior tense and flaccid parts of the right tympanic membrane, with no tympanic membrane perforation The x -rays of the right mastoid revealed mastoiditis without bone destruction. Electronic audiometric examination revealed the right ear air conduction as PTA25dB and normal bone conduction. Tympanoplasty type III of the right ear was performed. During surgery a 0.4- cm- diameter mauve-colored round and smooth mass was found at the tympanic entrance, which filled the epitympanic and middle tympanic cavity, adhering with the incus and tympanic membrane. The ossicles were intact. The tympanic cavity was not expanded. The mass was excised along with the incus, the malleus head was interposed. The postoperative histopathologic diagnosis of the tumor was middle ear adenoma.

In 1999,the patient had recurred tinnitus and hearing loss in the right ear. Computed tomography scan revealed an irregularly shaped soft tissue like mass within the middle ear with a CT value of 24-47Hu and sized 1x0.6x0.6cm. The right mastoid showed increased density and lacking septa between air cells and communication to middle ear cavity. The parotid gland was normal (fig1). Surgical resection was done. During surgery granulomatous tissue was found at the tympanic entrance and epitympanum, the stapes was wrapped by neoplasm. No tumor mass was found in the external auditorial canal. The neoplasm was completely excised. At 5-year's follow-up, no evidence of tumor recurrence was found.

Figure 1

Figure 1a and b: Unenhanced axial computed tomographic scan demonstrating a soft tissue like mass of the right middle ear(Arabic numeral 1)



HISTOPATHOLOGY AND IMMUNOHISTOCHEMISTRY

Intraoperative frozen sections of the tumor showed adenomatous proliferation and disrupted glandular structure. Postoperative histopathology HE staining showed that the tumor tissue was comprised of gland, duct, and epithelial cord, with disorganized glandular structure and slight focal epithelial atypia (fig2). Immunohistochemical staining revealed Ck (+)(fig3a), S-100 (+) (fig3b), -SMA (-), monoclonal antibody to chromograninA (cgA) was used to exclude carcinoid tumor and had negative staining. After consultation with several pathologists from different institutes, the lesion was diagnosed as pleomorphic adenoma of the middle ear.

Figure 2

Figure 2: Histological appearance of the tumor, showing adenomatous proliferation, comprised of gland, duct, and epithelial cord. (H & E,. x 350)

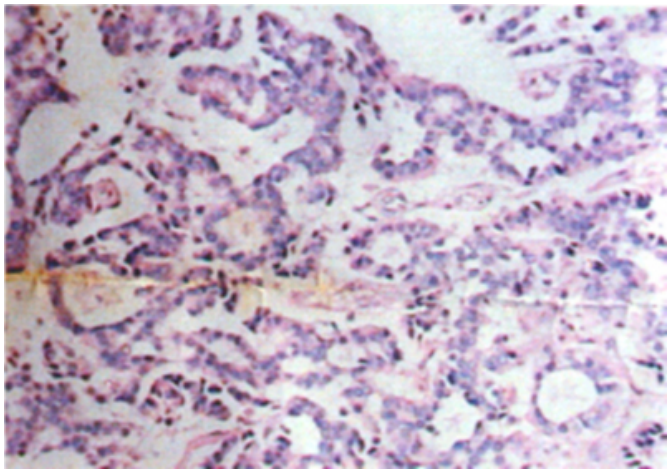
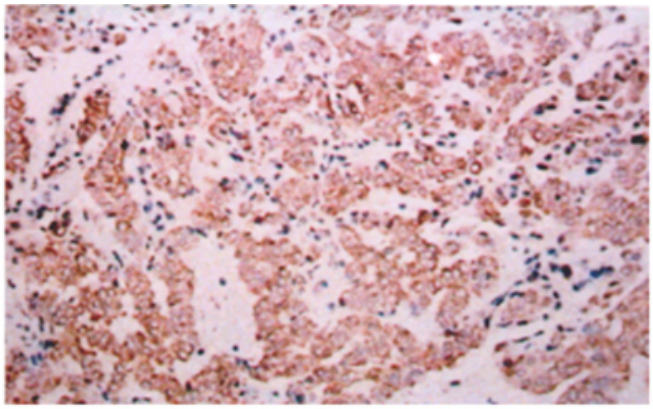
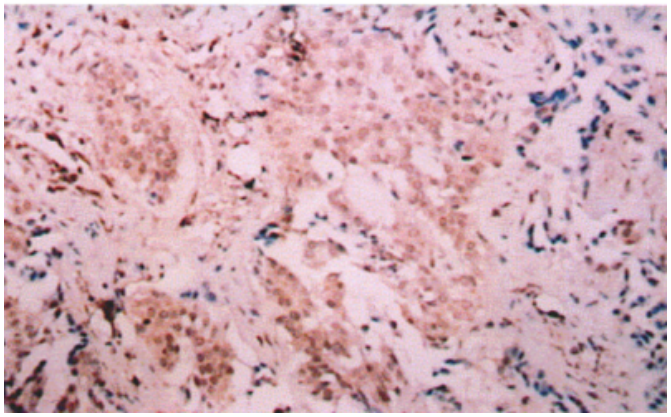


Figure 3

Figure 3: a. Immunohistochemical results showing positive staining of tumor cells to cytokeratin. b.Positive immunostaining of tumor cells to S-100 (DAB staining, x350).





DISCUSSION

Pleomorphic adenoma mainly occurs in major salivary glands such as parotid and submandibular glands as well as minor salivary glands at oral cavity and upper respiratory tract. Histopathologically the tumor is composed of glandular epithelial and myoepithelial cells, immunohistochemically demonstrate focal or widespread positivity to cytokeratin and S100 protein in both the epithelial and mesenchymal components of the neoplasm. Case reports of pleomorphic adenoma in sites other than the major salivary glands show their histopathological identification is not always straightforward just like this case; immunohistochemistry contribute significantly to the formulation of a definitive diagnosis [1]. Middle ear adenoma is composed of single layer epithelium and lacks myoepithelium, which may have origin in middle ear epithelium metaplasia [2,3]. Recent study suggested that middle ear adenoma have both epithelial and neuroendocrine differentiation.[4,5] which may be immunoreative with CK and chromogranin (cgA) .

The pleomorphic adenoma of the ear is rare and mainly reported to occur in the external auditory canal. The pleomorphic adenoma of the middle ear is extremely rare, only 2 case reports were obtained through Medline search [6,7]. One case was a 43-year-old female reported by Saeed, the tumor was located at the hypotympanum,3x3 mm in size. Another case was reported by Peters, the patient was a 51-year-old man with the tumor located at the mastoid and invaded middle ear cavity, posterior cranial fossa and facial nerve, 2.5cm in diameter .Both cases were considered originating from ectopic salivary gland.

The patient in this case was diagnosed having “adenoma”23 years before the present diagnosis of “pleomorphic adenoma”. Since immunohistochemical test was not done for the first diagnosis and the pathological sample was not available for reexamination, there is an uncertainty concerning whether this patient had two independent tumors or just the same tumor with inconsistent diagnostic terms. However the location of the tumor was at the same site of middle ear for both episodes, the invasion from tumor of the adjacent structures could also be excluded, the successive occurrence of adenoma and pleomorphic adenoma at the same anatomical site would be less likely.

Therefore it was reasonable to regard the two disease onsets as originated from the same tumor type. Possible explanation for the inconsistence of diagnosis might be due to less defined pathological terms for the middle ear adenomatous neoplasm and technique limitation in the past, which made

the first diagnosis inaccurate.

Middle ear pleomorphic adenoma might have three possible origins that is, the ceruminal glands of the external auditory canal , ectopic salivary gland or metaplasia of the middle ear mucosa.[6]. Since normal ceruminal glands have never been identified within the middle ear cleft[7]; ectopic salivary gland tissue has been reported to be present in the middle ear[1,8,9], and mucosal epithelium metaplasia is less likely to account for the mixed tumor elements in pleomorphic adenoma, most authors inclined to the origination from ectopic salivary glands. In this case of pleomorphic adenoma, there is no direct evidence to identify the histological origin, but the immunocytochemical results of S-100(+) and CK(+) suggested the origin from ectopic salivary gland is possible.

Case reports of pleomorphic adenomas in sites other than the major salivary glands show clinical and radiological signs of benignity[1]. In this case , the lesion was localized and non-invasive, its histopathological identification is not straightforward to differentiate from adenoma, immunohistochemistry is significant to the formulation of a definitive diagnosis .It also implies that middle ear pleomorphic adenoma may recur over 20 years after resection, an appropriate follow-up is recommended, resection at recurrence is effective.

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