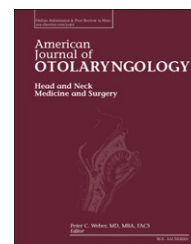


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Neuroendocrine adenoma of middle ear with new bone formation and review of literature [☆]

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ABSTRACT

Neuroendocrine adenoma of the middle ear has been described in literature as middle ear adenoma with neuroendocrine differentiation or carcinoid tumor. While there have been several case reports describing imaging features of carcinoid tumors of the ear and middle ear adenomas, in our literature review, we have not found a single case where bone formation is described as a prominent radiological feature. We report a first documented case of middle ear carcinoid tumor with new bone formation demonstrated on CT imaging and performed a review literature regarding the tumor. A differential diagnosis of neuroendocrine adenoma of the middle ear should be considered when there is prominent bone formation with a soft tissue mass in the middle ear on CT imaging.

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1. Introduction

Neuroendocrine adenoma of the middle ear has been described in literature as middle ear adenoma with neuroendocrine differentiation or carcinoid tumor. While there have been several case reports describing imaging features of carcinoid tumors of the ear and middle ear adenomas, in our literature review, we have not found a case where bone formation is described as a prominent radiological feature. We report a first documented case of middle ear carcinoid tumor with new bone formation demonstrated on CT imaging.

2. Case review

A 39 year old Chinese female presented to us with gradual left ear hearing loss over 8 months duration. There was no prior history of trauma or exposure to unusually loud noise. The patient has a history of bilateral ear discharge but did not have any tinnitus, otalgia or giddiness. Clinical examination of the cranial nerves was normal. Otoscopic examination revealed an opaque left tympanic membrane which was retracted. There was erythema noted in the pars flaccida and a slight bulge was seen laterally. Nasal endoscopic findings

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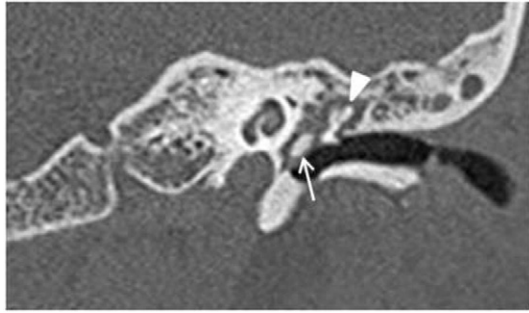


Fig. 1 – Coronal CT image of left temporal bone shows the abnormal dense bone formation separate from the ossicles (arrowhead) and promontory. Abnormal soft tissue within the middle ear with bone formation is demonstrated (arrow).

were normal. Audiogram showed moderate to profound mixed hearing loss in the left ear and profound high frequency sensory neural hearing loss in the right ear.

An unenhanced CT scan of the temporal bones was performed in coronal and axial views for further evaluation. CT scan shows a soft tissue lesion within the left middle ear cavity with an associated irregular bony structure close to the tip of the handle of the malleus and separate from the promontory (Figs. 1 and 2). The left stapes and incus are normal in configuration with an intact incudo-stapedial junction. No bony erosion of the ossicular chain is seen. There is mild sclerosis of the left temporal bone with a retracted tympanic membrane. Given the features of chronic otomastoiditis of the left temporal bone, the soft tissue lesion within the middle ear was thought clinically to be granulation tissue. The irregular bony structure was initially thought to be related to post inflammatory ossicular fixation of the fibro-osseous type.

The patient subsequently underwent a left transcanal excision of middle ear tumor. Intra-operative findings revealed a polypoidal left middle ear mass with a bony mass formed between the malleus and incus inferiorly. The bony structure was free from the surrounding walls of the middle ear and the ossicles (Fig. 3). The ossicular chain is intact and

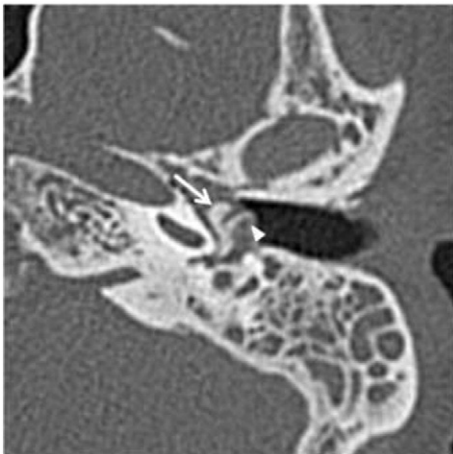


Fig. 2 – Axial CT image of the left temporal bone showing abnormal dense bone formation (arrows) which is adjacent but separate from the malleus handle (arrowhead).

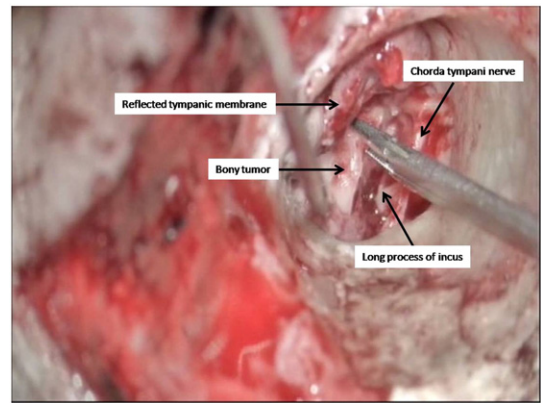


Fig. 3 – Bony lesion was separate from the incus.

in continuity. The promontory was free of tumor. The middle ear tumor and bony mass were removed completely without disruption to the ossicular chain.

Pathological examination indicated that the tumor has an infiltrative growth pattern. A piece of dense cortical ossicle-like bone was present which was surrounded and focally infiltrated by tumor cells. Tumor tissue was positive for NSE, synaptophysin, AE1/3 and CK7 and negative for chromogranin, CD 56, S100, p63 and CD 117. The final diagnosis was carcinoid tumor (middle ear adenoma with neuroendocrine differentiation) based on the morphological and immunohistochemical findings.

3. Discussion

Middle ear adenomas are rare. It can have both neuroendocrine and epithelial differentiation, depending on which stage of differentiation it is at. Adenomas of the middle ear are divided to mixed or papillary. Mixed adenomas are found in the middle ear and mastoid, rarely involving the facial nerve. Papillary types tend to be more invasive and can invade the petrous apex, the facial nerve, the temporal or posterior fossa. Hence they are sometimes referred to as low grade carcinoma [1,2].

Patients with neuroendocrine adenoma usually present with progressive unilateral hearing loss, tinnitus and a feeling of fullness. Pain and vertigo may not be present. Facial nerve involvement and bony erosion or destruction indicate a poorer prognosis [3].

The radiographic manifestation of a neuroendocrine adenoma on CT is as a well-circumscribed soft tissue mass without evidence of bone erosions. It may sometimes be embedded between ossicles [4]. Bone destruction and invasion of surrounding bone are a rare finding [5]. While ossification has been reported with carcinoid tumor in gastric and bronchial carcinoid tumor [6,7], this imaging feature has not been described in carcinoid tumors in the middle ear.

New bone formation in the middle ear cavity can be the result of chronic inflammation or neoplasm, the former being the most common cause. In patients with chronic otomastoiditis, post inflammatory ossicular fixation can result in conductive hearing loss. There are three pathological manifestations of post inflammatory ossicular fixation such as fibrous tissue fixation, tympanosclerosis and fibro-osseous sclerosis. Fibrous tissue fixation presents as soft tissues

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