

Case Report

A Case Report of Ceruminous Adenocarcinoma of External Auditory Canal

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Abstract: The external auditory canal contains ceruminous glands, which are modified apocrine sweat glands, along with sebaceous glands. Tumors that originate from ceruminous glands are very rare; thus, the classification, clinical behavior, and management of these tumors remain debatable. Here we present a case of ceruminous adenocarcinoma arising from the external auditory canal with all the mandatory histological features. Although most authors advise more aggressive therapy, our patient was treated with local en bloc resection of the tumor followed by intensity modulated radiotherapy.

Keywords: Ear neoplasms, adenocarcinoma, ear canal.

INTRODUCTION

The ceruminous glands are modified apocrine glands located within the dermis of the skin overlaying the cartilaginous portion of the external auditory canal (EAC) (Iqbal, A., & Newman, P. 1998). Watery secretions of ceruminous glands, along with sebaceous gland secretions, are drained into the hair sacs of fine hairs in EAC, together forming the cerumen (wax) (Thompson, L.D. *et al.*, 2004). Ceruminous gland-originated tumors of EAC are rare neoplasms. Because of the varied clinical and histological manifestations of these tumors, controversies currently exist regarding their nomenclature, classification, histopathological features, diagnosis and treatment.

Until Wetli *et al.*, (1972) proposed a classification based on histological features in 1972, these tumors were referred to as ceruminoma, a term which does not discriminate between benign and malign lesions. Ceruminous adenocarcinomas are malignant glandular tumors of EAC that do not show specific clinical symptoms. As most cases are advanced, treatment results are unsatisfactory (Wetli, C.V. *et al.*, 1972; Hicks, G.W. 1983). Herein we report our case of

ceruminous adenocarcinoma of EAC, which presented as a polypoid mass. The patient was treated with local en bloc resection of the tumor and followed by radiotherapy.

CASE PRESENTATION

A male patient aged 51 years, presented with history of left ear discharge of 1 year and left ear block for 3 years and without a previous history of surgery, or trauma and with a mild hearing loss in his left ear for about 2 months. He had no dizziness, tinnitus, vertigo, earache, or facial paralysis. Physical examination showed a reddish-violet polypoid, non-tender mass emerging from the anterior wall of left EAC that prevented the visualization of the tympanic membrane. Examination of the nose and throat showed no abnormality and no pathological lymphadenopathy on the neck was detected by ultrasonography. Pure-tone audiometry revealed a mild conductive hearing loss in the left ear. Magnetic resonance imaging (MRI) of the temporal bones revealed an isointense polypoid lesion, measuring approximately 23x10x7 mm (Figure 1). An incisional biopsy was performed and specimen was sent to our department for histopathology.

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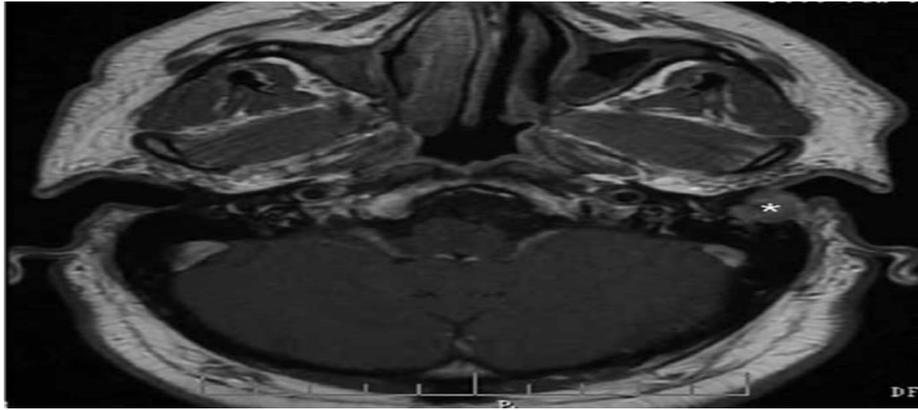


Fig1. Axial contrast-enhanced T1 MRI of the tumor on the left external auditory canal

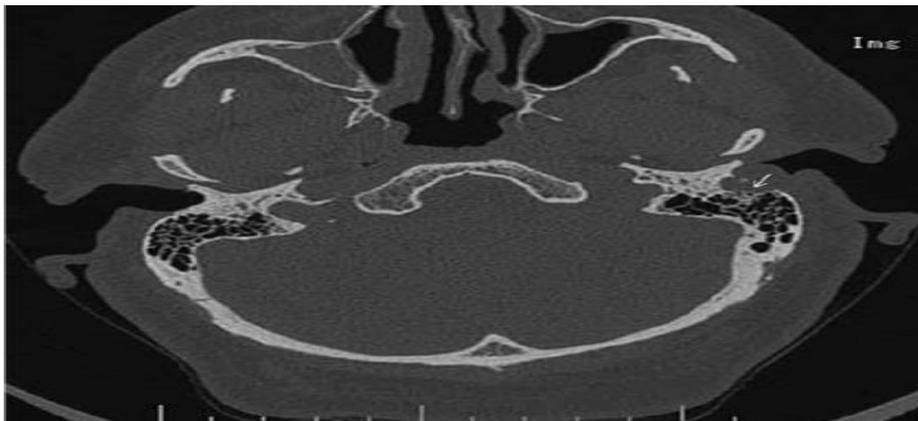


Fig2- Axial CT image of the tumor on the left external auditory canal

Macroscopy and microscopy

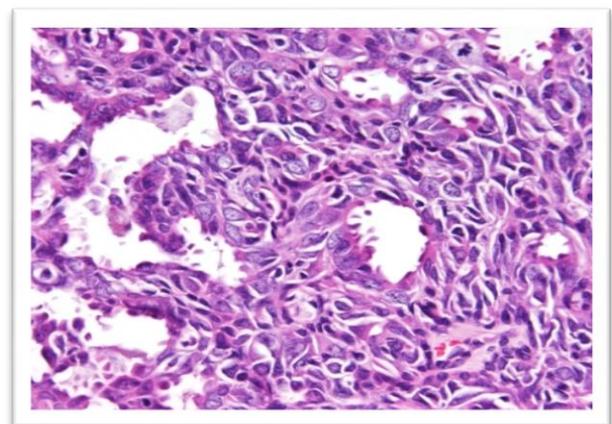
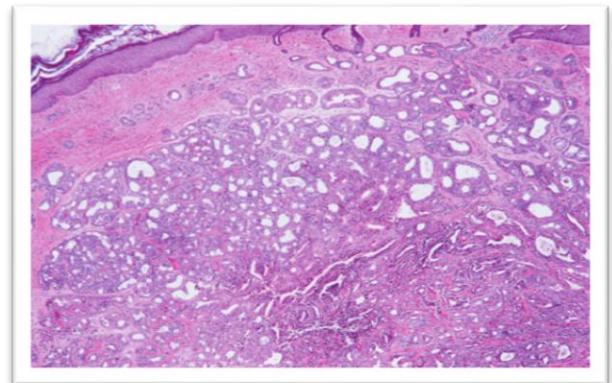
The specimen was received as multiple whitish soft tissue bits together measuring 15x10x5mm.

H and E stained sections showed tissue lined by stratified squamous epithelium, with subepithelium showing an infiltrating neoplasm composed of cells arranged in irregular glandular pattern, with focal cribriform arrangement separated by stroma showing extensive hyalinisation. Most of the glands are lined by single layer of cuboidal cells, some areas shows apocrine secretions. Cellular pleomorphism noted. Other glands show double layer of epithelium. Mitosis 7-8/10HPF and a few vascular tumour embolus noted. Areas of necrosis also noted.

IHC was performed with CK7, CD 117, p63 and Ki67.

The luminal cells show CK7 strong positivity; CD 117 was positive. The myoepithelial cells are p63 positive and Ki67- 10%.

A diagnosis of Ceruminous Adenocarcinoma was made



DISCUSSION

Ceruminous adenocarcinomas are rare malignant tumors that originate from the ceruminous glands of EAC. Differential diagnosis between ceruminous adenocarcinoma and ceruminous adenoma is oftendifficult. Although pathological evaluations of ceruminous adenocarcinomas demonstrate significantly more infiltration, perineural invasion, irregular gland formation, pleomorphism, prominent nucleoli, increased mitotic figures, atypical mitotic figures and tumor necrosis, only few of these features are usually observed, which complicates the distinction between adenocarcinoma and adenoma (Thompson, L.D. *et al.*, 2004). Primary lung, breast, or kidney cancer metastasis should be excluded when diagnosing this tumor (Soon, S.L. *et al.*, 2001).

Otalgia, mass, and hearing loss are the most common symptoms reported for ceruminous gland tumors in the literature (Crain, N. *et al.*, 2009). In addition, facial nerve involvement may result in facial nerve paralysis (Pulec, J.L. 1977).

Although cervical lymph node metastasis is uncommon (less than 4%) metastasis to the bones, lungs, and brain may be possible (Hicks, G.W. 1983; Soon, S.L. *et al.*, 2001). CT is helpful in detecting bone erosion and destruction as well as tumor extension (Mansour, P. *et al.*, 1992). The main treatment is surgery. Although recurrences usually occur within months after the initial treatment, it is reported that some may develop up to 7 years after the surgery (Hicks, G.W. 1983). Therefore, cautious long term follow-up on a 3-monthly basis is planned.. Long term

follow-up is planned for our patient and there was no evidence of recurrence till date after the surgery.

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