



Case Report

Ceruminous Adenoid Cystic Carcinoma of External Auditory Canal

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An adenoid cystic carcinoma (AdCC) of the ceruminous glands is very rare; its diagnosis is most often challenging, and simple biopsies may be misleading. Our paper describes a case of a circumferential mass of the left ear canal that was initially reported as a basal cell carcinoma on biopsies in the clinic and on frozen sections intraoperatively. The final pathology was an AdCC of the ceruminous glands of the external auditory canal. Our case reflects the difficulty in the diagnosis of an AdCC of the ceruminous gland and the importance of keeping broad differential diagnoses in mind when counseling patients with masses in the ear canals until final pathology is obtained.

KEYWORDS: Ceruminous glands, adenoid cystic carcinoma, auditory canal

INTRODUCTION

An adenoid cystic carcinoma (AdCC) of the ceruminous glands very rarely involves the external auditory canal (EAC), and in some instances, its pathology is misleading. We present a case of an AdCC in the ear canal that was initially diagnosed as a basal cell carcinoma (BCC) on performing clinical biopsies and frozen section procedures and discuss the treatment, outcomes, and misleading biopsy results.

CASE PRESENTATION

A 75-year-old woman presented to our head and neck oncology clinic with a 7-month history of a painless mass at the meatus of the left EAC that had been diagnosed as a BCC on biopsies in the clinic. The patient denied having otorrhea but complained of decreased hearing. She did not have any palpable cervical lymphadenopathy. A Computed Tomography scan of the temporal bone (Figure 1) showed complete obstruction of the meatus of the left EAC without involvement of the temporal bone or the parotid gland.

Intraoperatively, the tumor was found to extend to the tissue surrounding the tympanosquamous suture. Complete resection of the EAC and parotidectomy were performed, and negative margins were achieved. Intraoperative frozen sections were interpreted as being a BCC.

A histological analysis showed a basaloid neoplasm with a nested and cribriform architecture with numerous pseudoglandular tumor formations containing basophilic and eosinophilic materials. The tumor appeared to encircle the EAC in a 360° manner and showed extensive perineural invasion. The parotid gland showed microscopic tumor involvement at its periphery. The tumor did not appear to originate from the gland parenchyma (Figures 2–4). The patient recovered well and underwent adjuvant external beam radiation therapy and showed no evidence of disease recurrence during her follow-up visits. Exemption for review was granted by the Institutional Review Board.

DISCUSSION

Glandular neoplasms represent only 2.4% of all EAC neoplasms at a high-volume tertiary referral center^[1]. Malignant variants include adenoid cystic carcinomas, mucoepidermoid carcinomas, and ceruminous adenocarcinomas not otherwise specified, with benign forms usually consistent with pleomorphic adenomas^[1].

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Figure 1. Contrast-enhanced axial CT scan of the temporal bone showing the mass in the left external auditory canal.
CT: computed tomography

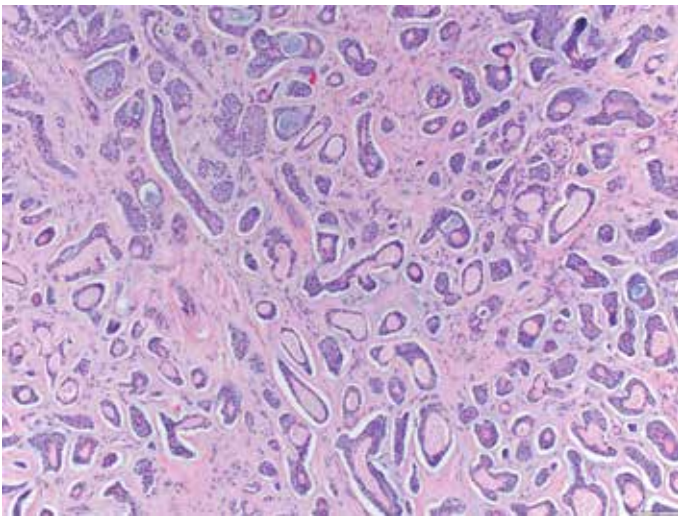


Figure 2. Hematoxylin and eosin staining showing the circumferential tumor in the left ear canal.

The histopathological characteristics of AdCCs in the EAC are identical to those found in the major and minor salivary glands. The tumor comprises duct lining cells and modified myoepithelial cells in various architectural patterns (most commonly cribriform, tubular, and solid nests). The tumor contains pseudoglandular/pseudocystic spaces filled with basophilic mucoid or hyalinized eosinophilic material. The eosinophilic connective tissue often occupies the extracellular stroma throughout pseudoglandular configurations. Tumor patterns containing nests or balls of cells may often be confused with a BCC^[2], which was the case in our patient. A BCC typically shows spindle-shaped nuclei, whereas an AdCC has rounded nuclei with a

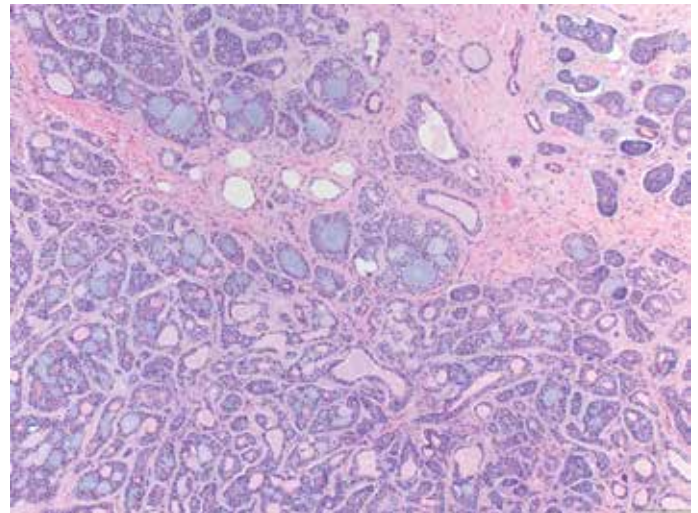


Figure 3. Hematoxylin and eosin staining, 100x magnification. Tumor with cribriform and tubular arrangements of small, bland, basaloid cells with scant cytoplasm. Pseudoglandular spaces contain basophilic and eosinophilic material.

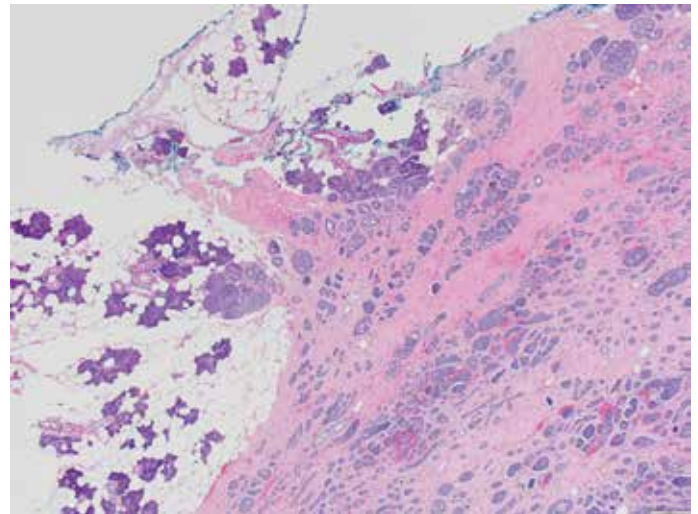


Figure 4. Hematoxylin and eosin staining, 200x magnification.

scarcity of mitotic figures^[1]. As with most AdCCs, those that originate from the ceruminous glands also show perineural invasion^[3].

The clinical presentation of ceruminous AdCCs is similar to that of other ear canal tumors. The main complaint is usually a mass within the ear canal and otalgia. Other symptoms include decreased hearing, bloody otorrhea, and otitis externa. Large asymptomatic lesions may grow until they obstruct the ear canal, allowing time for the tumor to become locally destructive or metastasize. Intracranial and pulmonary metastases have been reported in the literature^[4, 5]. As the ceruminous glands are found in the deepest portion of the canal dermis adjacent to the cartilage, the epidermis is rarely violated by the tumor. This significantly delays the diagnosis and hence the survival rate as a cutaneous neoplasm is not seen. The median time from the onset of symptoms to diagnosis has been reported to be around 2 years^[3]. The rate of local recurrence and distant metastasis reaches more 40% and 21%, respectively, despite lateral temporal bone resection^[3]. Our patient had negative neck/chest CT and PET-CT scan results in her follow-up visits.

Otolaryngologists should be vigilant when counseling patients with a BCC in the ear canal because this may very well be an AdCC of the ceruminous glands and as both conditions have different behaviors, treatments, and outcomes, with the ceruminous carcinoma having a less favorable natural history than a BCC. A very thorough follow-up with the timely recognition and diagnosis of AdCC in the EAC is necessary to improve outcomes and limit both regional and distant recurrence.

Informed Consent: The Institutional Review Board exempted this case from review.

Peer-review: Externally peer-reviewed.

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