Case Report

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Pathology Section

Apocrine Carcinoma of the External Auditory Canal: A Rare Case Report

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ABSTRACT

Apocrine carcinoma of the External Auditory Canal (EAC) is an extremely rare malignant neoplasm arising from modified apocrine (ceruminous) glands. We report a 41-year-old man with a long-standing recurrent EAC mass, previously excised multiple times and reported as papillary apocrine cystadenoma and ceruminous adenoma. He presented with progressive conductive hearing loss and a firm swelling occluding the EAC with extension into the temporal region. Imaging showed a large soft-tissue mass filling the canal and involving the temporal soft-tissue and mastoid air cells. Wide excision revealed an invasive tubulopapillary neoplasm with abundant eosinophilic cytoplasm, apocrine snouts, nuclear pleomorphism, mitoses and necrosis. Immunohistochemistry showed diffuse positivity for androgen receptor, GATA-3 and GCDFP-15 with a high Ki-67 proliferative index, confirming apocrine carcinoma. Re-excision with wider margins and selective neck dissection revealed lymph node metastasis and lymphovascular invasion. The patient subsequently received adjuvant radiotherapy. At the time of writing this report, he remains on regular follow-up with no evidence of local recurrence or metastatic disease. This case is notable for its unusual site, occurrence in a relatively young patient and progression from previously benign-reported ceruminous lesions. It highlights the malignant potential of recurrent ceruminous gland tumours and underscores the need for complete excision, thorough histopathological evaluation and long-term surveillance to detect early transformation or recurrence.

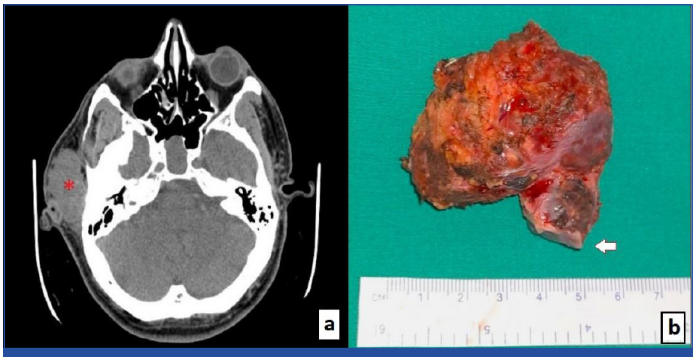
Keywords: Adenocarcinoma, Ceruminous glands, Ear neoplasms, Hearing loss, Sweat gland neoplasms

CASE REPORT

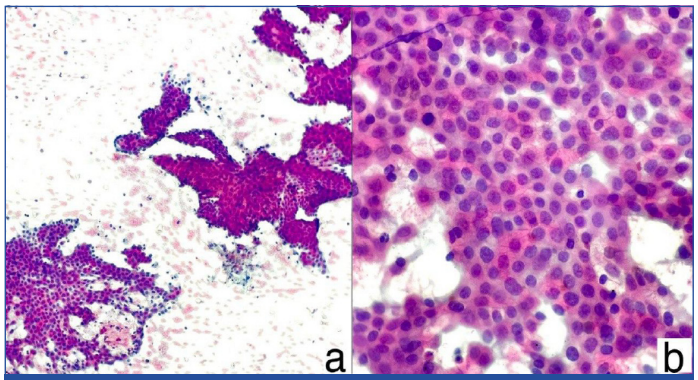
A 41-year-old man presented with a one-year history of progressive hearing loss and swelling involving the right EAC and temporal region. He had undergone three previous excisions at the same outside institution (2015, 2017 and 2018), with prior histopathology reports variably diagnosing papillary apocrine cystadenoma and ceruminous adenoma. Attempts to obtain the original slides and blocks were unsuccessful.

On examination, the EAC was completely obstructed by a firm, non-tender mass approximately 6 cm in size. Fine-needle aspiration cytology [Table/Fig-1a,b] showed a cellular neoplasm with oncocytic cells arranged as papillae and sheets. A diagnosis of oncocytic lesion with papillary structures was made and excision was suggested. Computed tomography [Table/Fig-2a] revealed a 6.0×5.0 cm soft-tissue lesion involving the superomedial pinna and extending into the EAC, temporal soft-tissue and mastoid air cells. No biochemical investigations were performed.

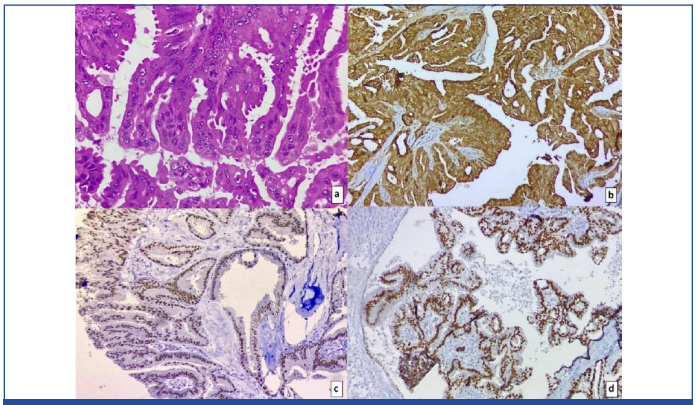
The mass was excised and measured 5.0×4.5×3.0 cm with a grey-yellow, partly haemorrhagic cut surface [Table/Fig-2b]. Microscopy revealed an infiltrative tubulopapillary neoplasm composed of eosinophilic cells with apocrine-type snouts, moderate nuclear atypia, mitotic figures and focal necrosis [Table/Fig-3a]. Tumour cells expressed androgen receptor, GATA-3 and GCDFP-15, and were negative for p63, CD117 and S100 [Table/Fig-3b-d]. Ki-67 proliferative index was approximately 40%, consistent with high-grade apocrine carcinoma.



[Table/Fig-2]: a) Contrast-enhanced CT showing a 6.0×5.0 cm soft-tissue lesion obstructing the External Auditory Canal (EAC) with extension into temporal soft-tissue and mastoid air cells; b) Gross specimen measuring 5.0×4.5×3.0 cm with grey-yellow, soft and haemorrhagic cut surface.



[Table/Fig-1]: a) Cytology smears showing a cellular neoplasm with oncocytic cells arranged as papillae and sheets. (PAP stain, 10x); b) Cells with a round nucleus with abundant eosinophilic granular cytoplasm (PAP stain, 40x).



[Table/Fig-3]: a) Invasive tubulopapillary carcinoma with apocrine features (H&E, 20×); b) Tumour cells positive for androgen receptor (IHC, 20×); c) GATA-3 nuclear positivity (IHC, 20×); d) GCDFP-15 cytoplasmic positivity (IHC, 20×).

Due to infiltration and close margin concerns, a second, wider excision was performed. This included resection of the temporalis muscle, superficial parotid tissue, tympanic membrane and level I-II lymph nodes. Two lymph nodes contained metastatic deposits, and lymphovascular emboli were present. The patient underwent adjuvant external beam radiotherapy (2 Gy×33 fractions). He continues regular follow-up and remains free of recurrence or metastasis.

DISCUSSION

Apocrine carcinoma is a rare malignant neoplasm arising from apocrine sweat glands and is most commonly reported in areas with a high density of apocrine glands, such as the axilla, anogenital region and scalp. Less frequently, it has been described in sites including the eyelid, face, trunk and extremities [1]. Apocrine carcinoma of the EAC is exceptionally rare. Its clinical presentation often resembles benign otologic conditions, and the scarcity of documented cases makes diagnostic interpretation challenging. Most available information comes from isolated case reports and small series. For this reason, the following is the published data that allow meaningful comparison with the present case in terms of behaviour, diagnostic challenges and therapeutic implications.

Wanner B et al., reported that malignant tumours of the EAC often show substantial local invasion and recur when margins are inadequate [1]. Yang Y et al., described the wide morphological spectrum of ceruminous tumours, contributing to diagnostic difficulty [2]. Samara P et al., noted that malignant lesions in this region are often detected late due to their resemblance to common benign ear disorders [3].

Apocrine carcinoma at cutaneous sites offers additional insight. Tsuruta S et al., described classic apocrine differentiation and the risk of lymph node metastasis [4]. Singh VY et al., documented ceruminous adenocarcinoma and showed that such tumours may behave aggressively despite appearing indolent clinically [5]. Thompson LDR et al., examined ceruminous adenomas and highlighted the need to identify subtle atypia that may suggest early malignant transformation, which is relevant given this patient's prior benign diagnoses [6]. Nagarajan P et al., observed that recurrent benign ceruminous lesions can develop more aggressive features over time [7]. Hollowell KL et al., identified nodal metastasis as an important unfavourable prognostic factor in apocrine carcinoma, aligning with the nodal disease in this case [8]. Paties C et al., described hallmark microscopic findings such as tubulopapillary structures and apocrine snouts, all of which were evident here [9]. Michel RG et al., detailed the infiltrative pattern typical of ceruminous gland malignancies, comparable to the invasion identified in our patient [10].

The histopathological differential diagnoses include a spectrum of ceruminous and non-ceruminous tumours of the EAC. Ceruminous adenoma and papillary apocrine cystadenoma show well-circumscribed, non-infiltrative lesions with bland cytology and low mitotic activity, lacking the overt atypia, necrosis and infiltrative growth seen in our case. Ceruminous adenocarcinoma, not otherwise specified, adenoid cystic carcinoma and mucoepidermoid carcinoma are important malignant differentials; these may show overlapping glandular or cribriform patterns but differ in cytologic features and immunoprofiles. Metastatic adenocarcinoma (particularly from breast or gastrointestinal tract) and other apocrine adnexal carcinomas must also be excluded, especially in older patients, by clinical correlation and immunohistochemistry [6].

The present case resembles published cases in showing a canal-filling mass, local extension and an immunoprofile consistent with apocrine carcinoma [2,3]. Two aspects make this case particularly noteworthy. The patient is younger than most individuals reported in the literature. Additionally, he had a well-documented history of benign ceruminous lesions before the development of carcinoma, supporting the likelihood of malignant transformation as proposed by Thompson LDR et al., and Nagarajan P et al., [6,7].

Management in earlier reports generally favours wide local excision or temporal bone resection, often combined with neck dissection when nodal involvement is suspected [7,8]. The present case patient underwent complete resection and nodal clearance followed by adjuvant radiotherapy due to lymphovascular invasion and lymph node metastasis. His favourable course and absence of recurrence thus far highlight the value of early and aggressive multidisciplinary management.

CONCLUSION(S)

Recurrent or long-standing ceruminous gland tumours of the EAC should be excised completely, evaluated thoroughly and followed over the long-term. Apocrine carcinoma may arise from pre-existing benign ceruminous lesions and can present with nodal metastasis. Early identification and prompt multidisciplinary treatment are essential for sustained disease control.

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