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Adenoid Cystic Carcinoma of the Ceruminous Glands of the External Auditory Canal: A Case Report

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Abstract **Case Report**

Introduction: Primary malignant tumors of the external auditory canal (EAC) are rare. Among them, adenoid cystic carcinoma (ACC) arising from the ceruminous glands is an exceptional entity, whose clinical and radiological presentation may mimic a benign lesion, thereby delaying diagnosis. ***Case report:*** We report the case of a 63-year-old woman who presented with an EAC mass evolving over 4 months, associated with otalgia. Otologic examination revealed a round, smooth, sessile mass of the outer third of the EAC, obstructing visualization of the tympanic membrane, which appeared normal on the visible portion. Preoperative computed tomography (CT) suggested an EAC lipoma based on a fat-density lesion. Surgical resection of the mass was performed. Histopathological examination showed a tumor proliferation with cribriform and tubular architecture, lined by monomorphic cells developing within a hyaline cylindromatous stroma. Immunohistochemical analysis concluded to an adenoid cystic carcinoma of the ceruminous glands. The patient was referred to the National Institute of Oncology for further management. Postoperative MRI showed no evidence of residual or recurrent tumor, a finding confirmed on a second MRI performed 7 months later. ***Discussion:*** This case illustrates the diagnostic difficulty posed by ceruminous gland ACC, whose radiological appearance can be misleading, as well as the essential contribution of histopathological and immunohistochemical examination to a definitive diagnosis. Management, mainly surgical, and prolonged clinical and radiological follow-up are discussed in light of the literature. ***Conclusion:*** Any EAC mass, even one with a radiologically benign appearance, warrants histological vigilance. Ceruminous gland ACC, although rare, must be considered and requires long-term follow-up given its potential for late recurrence.

Keywords: adenoid cystic carcinoma; ceruminous glands; external auditory canal; lipoma; immunohistochemistry.

1. INTRODUCTION

Primary tumors of the external auditory canal (EAC) are dominated by benign lesions (osteomas, exostoses, papillomas), malignant tumors being markedly rarer. Among primary malignant EAC tumors, squamous cell carcinoma accounts for more than 80% of cases, ACC representing only about 5% [4,5]. Among malignant EAC tumors, those arising from the ceruminous glands (modified apocrine glands located in the cartilaginous third of the canal) constitute a distinctive and uncommon histological entity [3,4].

Adenoid cystic carcinoma (ACC), also known as cylindroma, is classically a salivary gland tumor. Its ceruminous location is exceptional and poses diagnostic difficulties, both clinically and radiologically, owing to a presentation that often mimics a benign EAC condition [1,2]. Definitive diagnosis relies on histopathological examination, complemented by immunohistochemistry, which also helps rule out the main differential diagnoses, notably mucoepidermoid carcinoma and ceruminous adenocarcinoma [3].

We report, through this work, a case of adenoid cystic carcinoma of the ceruminous glands revealed by an EAC mass whose initial CT appearance was mistakenly suggestive of a lipoma, discussing the diagnostic, therapeutic, and prognostic particularities of this rare entity.

2. CASE REPORT

The patient was a 63-year-old woman, hypertensive on well-controlled medical treatment, with no other notable medical history, who presented with a mass of the external auditory canal evolving over 4 months. This mass was associated with otalgia, with no other associated otologic symptom reported (otorrhea, hearing loss, tinnitus, facial paralysis, cervical lymphadenopathy).

Otologic examination revealed a round, smooth, sessile mass located in the outer third of the external auditory canal. This mass obstructed adequate visualization of the tympanic membrane, but the latter appeared normal on the portion that remained visible.

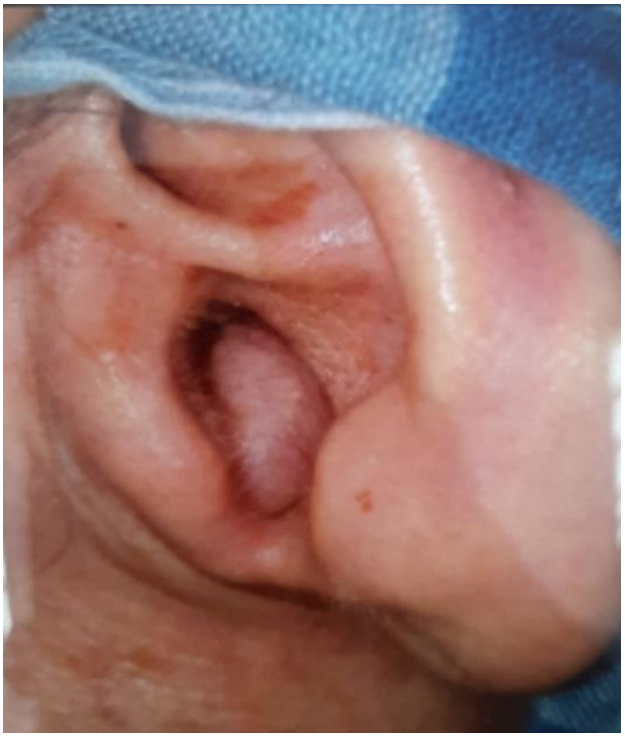

Figure 1: Otologic examination revealing a round, smooth, mass in the external auditory canal

Given this presentation, a computed tomography (CT) scan of the temporal bones was performed prior to any surgical procedure. It showed a fat-density lesion of the EAC, without associated bone lysis, an appearance primarily suggestive of an EAC lipoma.

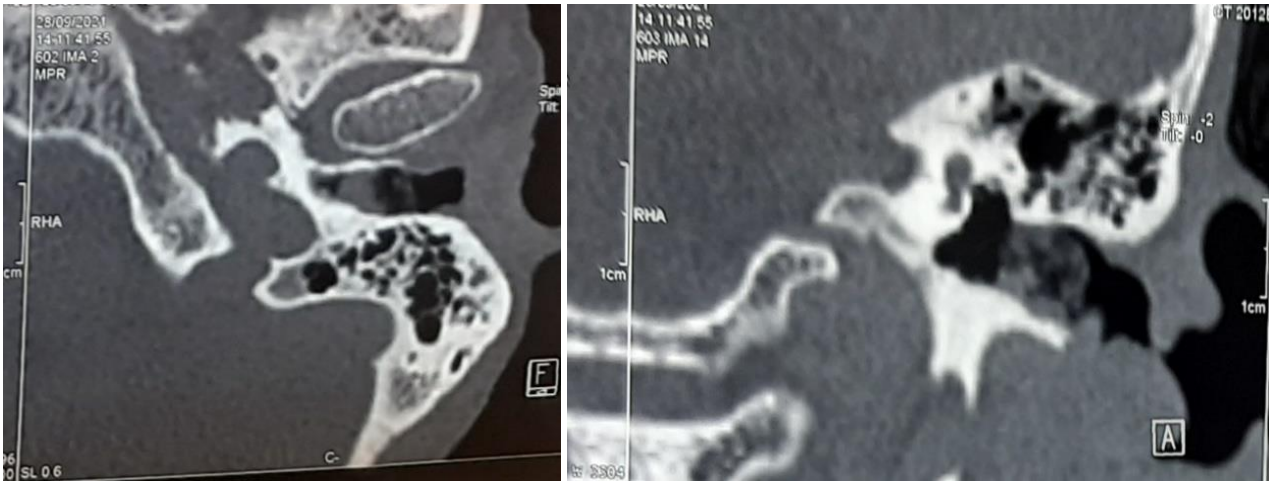

Figure 2: Axial and Coronal tomographic scan of the patient showing fat-density lesion of the left EAC

Given this reassuring radiological appearance, surgical resection of the mass was performed. Histopathological examination of the surgical specimen showed a tumor proliferation with cribriform and tubular architecture, lined by monomorphic cells developing within a hyaline cylindromatous stroma.

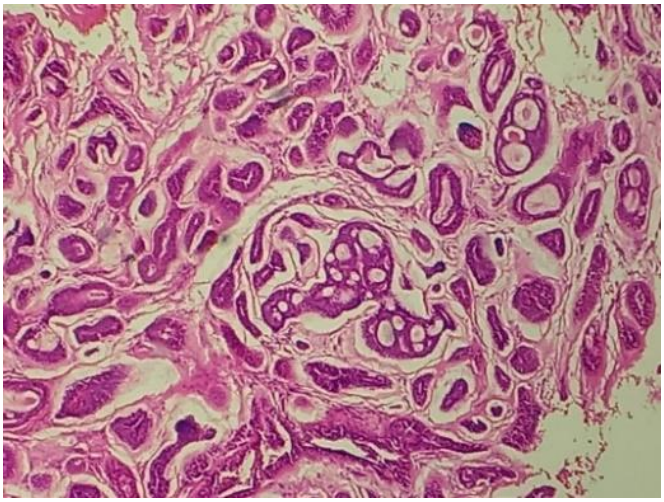

Figure 3: Photomicrograph showing a tumor proliferation with cribriform and tubular architecture, lined by monomorphic cells developing within a cylindromatous hyaline stroma. H&E, x10.

Immunohistochemical analysis showed the following results:

- Anti-CK7: intense and diffuse positivity in the luminal tumor cells;
- Anti-EMA: intense and diffuse positivity in the luminal tumor cells;
- Anti-CD117 (c-kit): intense and diffuse positivity in the myoepithelial cells;
- Anti-CK5/6 and anti-p63: intense and diffuse positivity in the myoepithelial cells;
- Anti-p53: positive in the tumor cells;
- Ki-67 proliferation index: estimated at 20%.

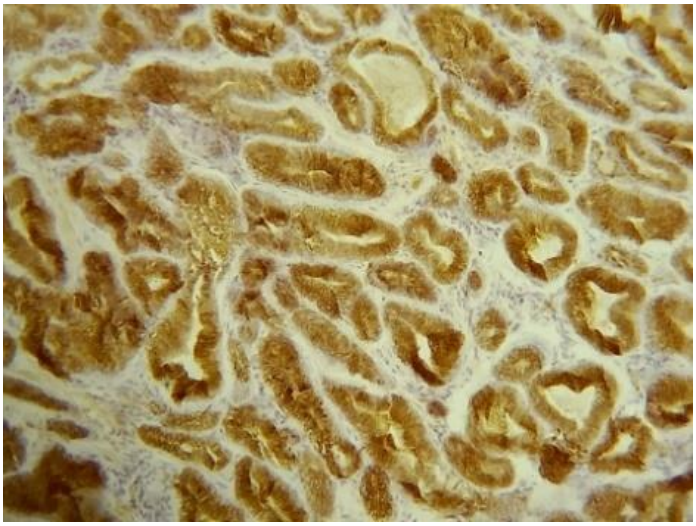

Figure 4: On immunohistochemical study the tumor cells diffusely and intensely express CD117

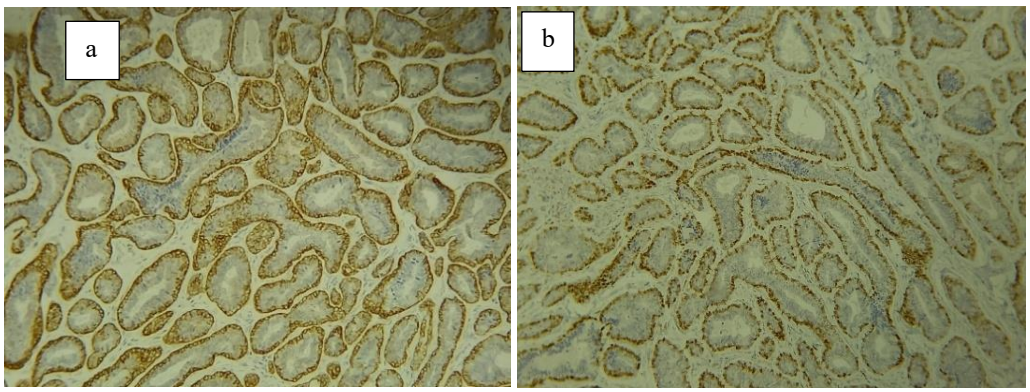

Figure 5: The basal cells express CK5/6 (a) and p63 (b)

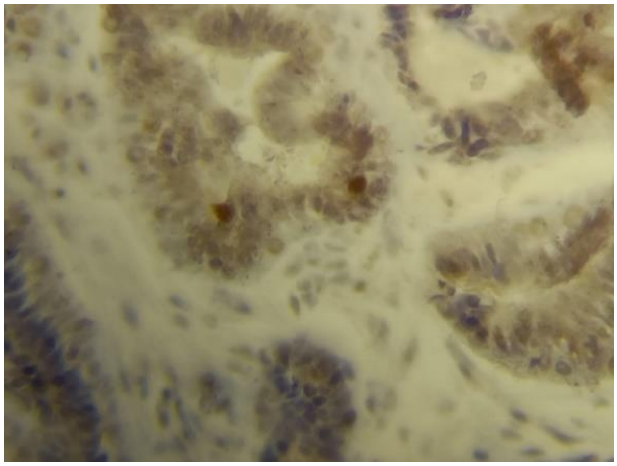

Figure 6: P53 was expressed by the tumor cells

This architectural pattern (mixed cribriform and tubular) together with the immunohistochemical profile (luminal CK7/EMA positivity, myoepithelial CD117/CK5-6/p63 positivity) allowed the diagnosis of adenoid cystic carcinoma arising from the ceruminous glands of the EAC to be established.

Given this diagnosis of malignancy, the patient was referred to the National Institute of Oncology (INO) for further work-up and management. Magnetic resonance imaging (MRI) performed in this context showed no evidence of residual tumor or local recurrence. A second follow-up MRI, performed 7 months after surgery, confirmed these same findings, with no evidence of locoregional progression.

The status of the surgical resection margins could not be determined. Given the absence of residual tumor demonstrated on the successive MRI studies, the multidisciplinary tumor board decided to pursue simple surveillance by MRI, with no indication for adjuvant treatment.

3. DISCUSSION

3.1. Background and Epidemiology

Ceruminous glands are modified apocrine glands located exclusively in the skin of the outer cartilaginous third of the EAC. Tumors arising from them are rare and span a spectrum ranging from benign ceruminous adenoma to malignant tumors such as ceruminous adenocarcinoma, mucoepidermoid carcinoma, and adenoid cystic carcinoma [3,6]. The latter preferentially affects middle-aged to older adults, with a female predominance reported in several series, although this point remains debated among authors [5,8].

3.2. Clinical and Diagnostic Features

The clinical presentation of ceruminous gland ACC is nonspecific: an EAC mass with a generally slow course, otalgia reported as the most frequent symptom, present in about three-quarters of patients in some series, conductive hearing loss, or even otorrhea [8]. ACC should be considered in any EAC mass associated with otalgia evolving for more than 6 months, particularly in middle-aged women [7]. Histologically, this cancer shows marked perineural tropism, found in more than two-thirds of cases in some series, although its presence is not always statistically correlated with the occurrence of otalgia [8].

Radiologically, ACC has no pathognomonic CT sign, which explains initial diagnostic errors, as in our observation where the fat-density appearance mistakenly suggested a lipoma. Lipomas of the otologic region are typically characterized on CT by markedly negative density (in the range of -40 to -110 Hounsfield units), without bone erosion or contrast enhancement, and by T1 hyperintensity that suppresses on fat-saturation MRI sequences [10,11]. A rare variant, osteolipoma, may additionally combine elements of bone density within the fatty mass [12]. The association with otalgia and a course of several months, as in our patient, could have constituted an additional warning sign warranting increased histological vigilance despite the reassuring CT appearance. MRI, particularly gadolinium-enhanced sequences, is more effective than CT in assessing tumor extension, notably perineural extension along the facial nerve, and for surgical planning [7].

3.3. Definitive Diagnosis: Contribution of Histopathology and Immunohistochemistry

Definitive diagnosis relies exclusively on histopathological examination. ACC is histologically characterized by a dual cell population, epithelial (luminal) and myoepithelial (basal), arranged in variable architectural patterns, cribriform, tubular (the two most common forms), or solid, the latter being associated with a worse prognosis, with frequent perineural tropism [9,14]. In our observation, the architecture was mixed, cribriform and tubular, with a characteristic hyaline cylindromatous stroma.

Immunohistochemistry plays a decisive role, both in confirming the biphasic nature of the tumor and in differentiating it from other ceruminous gland tumors, notably ceruminous adenocarcinoma and

mucoepidermoid carcinoma, which are histologically indistinguishable from a salivary gland primary, making it also necessary to exclude extension from the parotid gland by imaging or surgical exploration [6,14]. The typical immunohistochemical profile combines positivity of luminal cells for cytokeratin 7 (CK7) and epithelial membrane antigen (EMA), and positivity of basal-myoepithelial cells for C-kit (CD117), cytokeratin 5/6, and p63 [6,14]. This profile was precisely reproduced in our patient, with intense and diffuse CK7 and EMA staining in the luminal component, and intense and diffuse CD117, CK5/6, and p63 staining in the myoepithelial component, confirming the biphasic nature of the proliferation.

The Ki-67 proliferation index is generally moderate in this tumor, whereas p53 positivity has been reported in more than half of cases in some series [14]. In our patient, the Ki-67 index was estimated at 20%, and p53 staining was positive in the tumor cells, two findings that, while not diagnostic in themselves, may suggest a non-negligible proliferative activity and further justify caution during follow-up.

3.4. Therapeutic Management

The standard treatment for ceruminous gland ACC is surgical, based on wide excision with clear margins, which may range, depending on extension, from wide local excision or radical mastoidectomy to lateral or total temporal bone resection in advanced forms [8,15]. Because of the perineural tropism of this tumor, careful evaluation of extension along the facial nerve is essential before any procedure, and parotid or bone involvement should be systematically sought, as it affects prognosis [9,15].

Adjuvant radiotherapy is widely recommended by most authors, particularly in cases of incomplete resection, perineural extension, or solid histological pattern, with the aim of reducing the risk of local recurrence, which remains the most important prognostic factor [16]. A recent retrospective study of 60 patients treated with postoperative radiotherapy reported 10-year overall survival and local recurrence-free survival rates of approximately 84% and 80%, respectively, with advanced T stage remaining an independent risk factor for poorer survival [16].

3.5. Prognosis and Follow-up

ACC is known for its often-indolent course but also for its potential for local recurrence reported in about one-third of patients in some series and distant metastasis, mainly pulmonary, sometimes occurring very late, with an interval that can reach up to 14 years after initial treatment in the historical series by Perzin *et al.,* [8,9]. This justifies prolonged clinical and radiological (MRI) follow-up, well beyond the first 5 years. In our observation, the absence of residual disease or recurrence on the postoperative MRI and at 7 months is a reassuring finding in the short term, but does not preclude very long-term surveillance given the natural history of this tumor, particularly given the non-negligible Ki-67 proliferation index noted in our patient.

4. CONCLUSION

Adenoid cystic carcinoma of the ceruminous glands is a rare malignant tumor of the external auditory canal, whose clinical and radiological presentation can be misleading, as illustrated by this case initially oriented toward a lipoma. Definitive diagnosis relies on histopathological and immunohistochemical examination, making systematic histological analysis of any resected EAC mass mandatory, regardless of its radiological appearance. Management, mainly surgical, must be accompanied by prolonged follow-up given the tumor's potential for late progression.

Declarations

Conflicts of interest: The authors declare no conflict of interest.

Consent: Informed consent was obtained from the patient for publication of this case.

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