European Journal of Clinical Medicine
Vol 6 | Issue 2 | May 2025
ISSN 2736-5476

REVIEW

Adenoid Cystic Carcinoma of the External Auditory Canal: A Case Report and Literature Review

Driss Elidrissi[1,*], Youssef Lakhdar[1], Omar Oulghoul[1], Mohamed Rami[1], Mohamed Chehbouni[1], Othman Benhoummad[2], Youssef Rochdi[1], and Abdelaziz Raji[1]

ABSTRACT

Adenoid cystic carcinoma is an uncommon neoplasm that originates from the salivary glands, particularly when it developed in the external ear canal. Perineural invasion of this tumor requires aggressive treatment with surgical resection followed by adjuvant radiotherapy. We report a rare case of adenoid cystic carcinoma in a 71-year-old male patient, manifesting with symptoms of hypoacusis and a visible tumor of the external auditory canal. The patient underwent a successful surgery using a radical mastoidectomy followed by radiotherapy as part of her treatment regimen.

Keywords: Adenoid cystic carcinoma, external auditory canal, radical mastoidectomy, salivary glands.

Submitted: December 09, 2024

Published: May 22, 2025

10.24018/ejclinicmed.2025.6.2.364

[1] Department of ENT and HNS Surgery, Mohammed VI University Hospital, Morocco.
[2] Department of ENT and HNS Surgery, Faculty of Medicine and Pharmacy of Agadir, Ibn Zohr University, Morocco.

** Corresponding Author:*
e-mail: driss.elidrissi93@gmail.com

1. Introduction

Primary malignancies affecting the external auditory canal (EAC) are extremely rare, and the most common type is squamous cell carcinoma [1]. Adenoid cystic carcinoma (ACC) typically originates in the salivary glands accounting for 22% of all salivary gland malignancies [2], but may also appear in nasal cavity, the intraosseous mandible or maxilla, lacrimal glands, tracheobronchial tree, breasts, esophagus, and EAC [3]. ACC arising in the EAC is therefore a seldom encountered occurrence. Since 1894, Very few cases of EAC ACC have been reported in the English literature [4]. The natural progression of ACC in EAC is characterized by a slow growing, which can often lead to delayed diagnosis. However, ACC is a highly invasive cancer with a high recurrence rate [5]. Treatment aims to achieve complete surgical removal with clear margins due to the high risk of local recurrence. Distant metastases, primarily to the lungs, can occur over many years [6], [7]. Here, we report a rare case of adenoid cystic carcinoma of The EAC in a 71-year-old man. Literature is reviewed, and diagnostic procedures and surgical approaches are discussed.

2. Case Report

The patient was a 71-year-old man with a history of second type diabetes, who presented since a year, a budding lesion obstructing the entire external auditory meatus of the right ear (Fig. 1), complicated by hypoacusis, with no vertigo, facial paralysis, otorrhagia or other associated signs and without palpable lymphadenopathy.

Pure-tone audiometry showed moderate conductive hearing loss with an air-bone gap of 26 dB in the affected ear.

The CT scan found a tissue lesion on the posterior wall of the right external auditory canal, measuring 6 mm × 16 mm with bone lysis opposite the mastoid; the tympanic membrane and ossicular chain were intact (Fig. 2).

MRI reveals irregular, circumferential thickening of the partially stenosed right external auditory canal, with evidence of a nodular lesion adjacent to its posterior wall in T1 hyposignal and heterogeneous T2 and FLAIR hypersignal, in diffusion hypersignal with ADC restriction, heterogeneously enhanced after contrast injection, measuring 12 mm × 6 mm, arriving in intimate contact with homolateral parotid with no signal anomaly opposite, associated with a bony signal anomaly of the homolateral mastoid measuring 7 mm × 4.5 mm, probably related

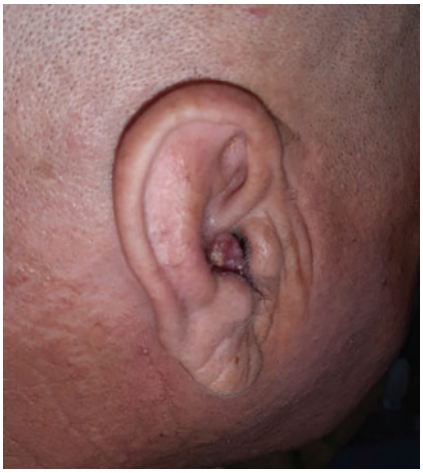

Fig. 1. The picture shows the mass protruding into the external auditory canal.

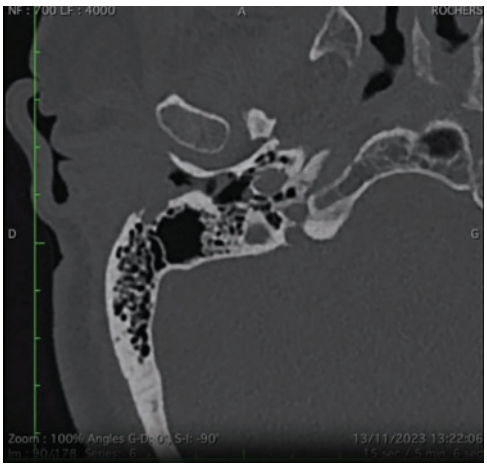

Fig. 2. The CT scan axial cuts showing a lesion on the posterior wall of the right external auditory canal, measuring 6 mm × 16 mm with bone lysis opposite the mastoid.

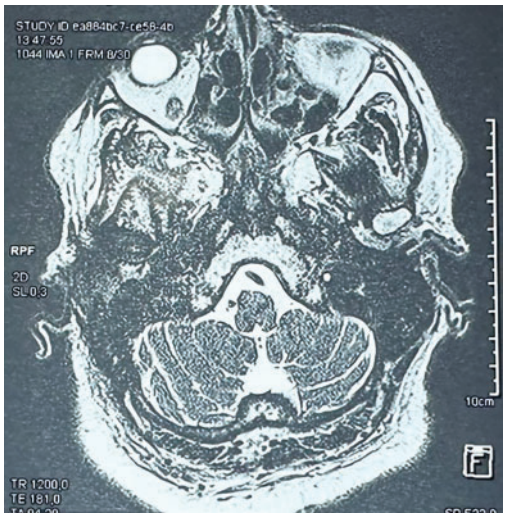

Fig. 3. T2 MRI axial cuts reveal irregular, circumferential thickening of the partially stenosed right external auditory canal, with evidence of a nodular lesion adjacent to its posterior wall, measuring 12 mm × 6 mm.

to bone lysis a T3 according to Pittsburgh classification (Figs. 3 and 4).

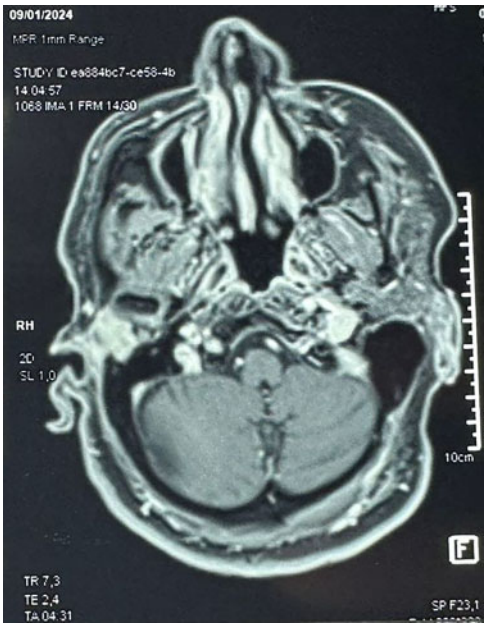

Fig. 4. T1 MRI axial cuts reveals irregular, circumferential thickening of the partially stenosed right external auditory canal, with evidence of a nodular lesion adjacent to its posterior wall, measuring 12 mm × 6 mm and an accidentally discovered cholesterol granuloma of the contralateral petrous apex.

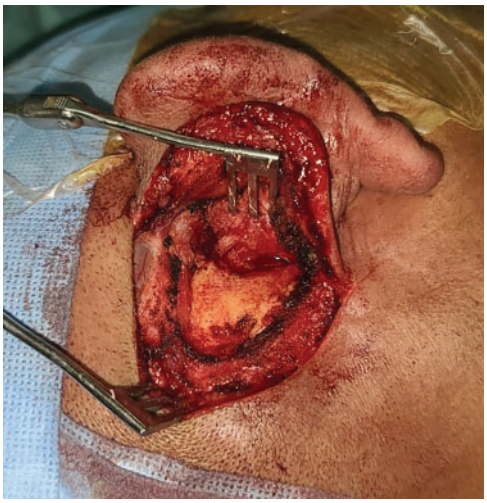

Fig. 5. The tumor infiltrating subcutaneous and muscular tissue flap.

Patient subsequently underwent a biopsy under local anesthesia through the external auditory canal, that revealed a cystic adenoid carcinoma.

The patient underwent a surgery via a retroauricular approach, we discovered a tumor obstructing the external auditory canal and infiltrating subcutaneous and muscular tissue (Fig. 5).

The visible part of the tumor was removed (Fig. 6), and the tympanic membrane was visualized intact. We then performed a radical mastoidectomy followed by conchomeatoplasty (Figs. 7 and 8).

In immediate post-operative follow-up, there was no bleeding or hematoma, no facial palsy, no cophosis or vertigo, and no neuromeningeal signs. The patient was monitored every two days until the sutures were removed on the tenth day. There was no infection, pus discharge or suture laceration. The clinical and CT scan follow-up was

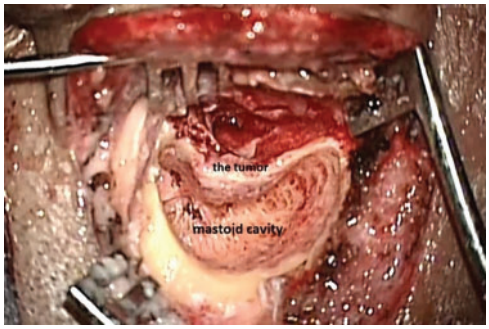

Fig. 6. Operative view before tumor removal.

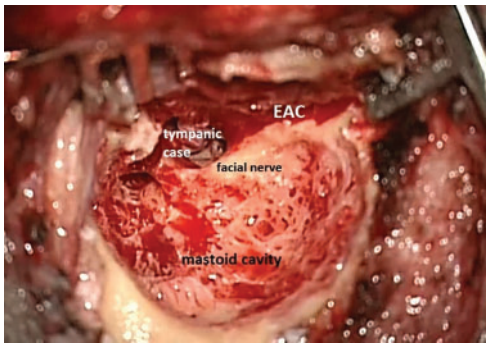

Fig. 7. Tympanic case, external auditory canal and mastoid cavity after the tumor removal.

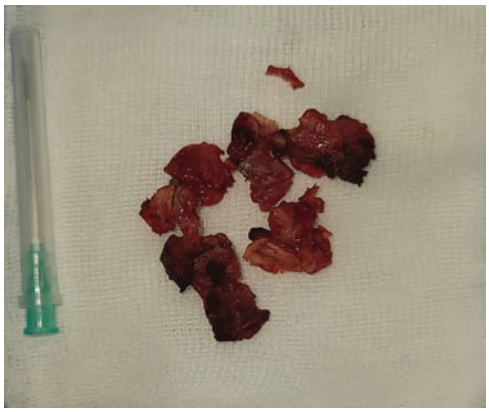

Fig. 8. The excision pieces.

unremarkable, with no evidence of tumor recurrence after two years.

Anatomopathological study revealed a 1.5 cm lesion that was in favor of a cystic adenoid carcinoma, with lesion-free resection margins.

3. Discussion

External auditory canal (EAC) tumors are infrequent, with only 20% originating from glandular tissue, predominantly presenting as squamous cell carcinomas [8]. Adenoid cystic carcinomas (ACC) are the most common glandular tumors. Their features resemble those of ACC originating from small salivary glands, including silent growth, local recurrence, perineural invasion, and delayed distal metastasis [9], [10].

Gender prevalence remains controversial in the literature. Triantafillidou *et al.* reported a higher frequency among women, while Lucia *et al.* observed a greater incidence among males [8], [9]. The tumor can manifest at any age, but peaks in occurrence are noted in the 5th and 6th decades of life [8]. Symptoms typically include pain, hypoacusis, otorrhea, and EAC nodules, with the tumor often growing for years before diagnosis [9], [10].

Diagnosis primarily relies on pathology examination, with fine needle aspiration occasionally assisting in preoperative diagnosis [4]. Treatment typically involves radical mastoidectomy combined with ear canal resection, with specimen freezing used to evaluate margins and perineural invasion [7], [8]. Post-operative radiotherapy is recommended to mitigate local tumor recurrence, significantly improving outcomes compared to surgery alone (86% against 11% when only surgery is done) [9], [10]. Some authors advocate radiotherapy solely for advanced tumors exhibiting specific features such as skull base invasion, neck metastasis, perineural invasion, solid histological type, or recurrent tumors [7]–[11].

Neck dissection is reserved for cases with positive nodules detected in the neck, occurring in approximately 4% of cases [12], [13]. Distal metastases are more frequent, affecting 48% of patients, especially when the primary tumor is incompletely excised [10]. Common sites of metastasis include the lungs, kidneys, and vertebrae [8]. Despite metastatic spread, patients often have prolonged survival, underscoring the importance of local tumor control to minimize morbidity and sustain reasonable quality of life [12].

Prognosis worsens in cases of local tumor recurrence, perineural, parotid, or bone involvement, as well as when margins are positive, and the tumor exhibits a solid histological type [11]–[13].

4. Conclusion

Adenoid cystic carcinoma is a rare tumor rarely found in the external auditory canal. Its management typically involves radical procedures aimed at enhancing local disease control and potentially decreasing distal metastasis occurrence.

Compliance with Ethical Standards

The study was conducted in compliance with ethical standards.

Funding

This research received no external funding.

Ethical Approval

While formal ethical approval was not obtained for this study, we ensured that all aspects of the research were conducted ethically and with respect for the rights and well-being of the participants.

Informed Consent

Informed consent was obtained from all participants involved in the study, and this information has been appropriately included in the manuscript.

Conflict of Interest

There are no conflicts of interest to declare related to this research.

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