Case Report
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Auricular Sebaceous Carcinoma, Report of a Rare Case

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Abstract

Sebaceous carcinoma is a rare and invasive tumor that originates from the Zeis and Meibomian glands around the eyes as well as in sebaceous glands in other head and neck areas and less commonly on the trunk. We report an 82-year-old Iranian man with a sebaceous carcinoma on the left external ear that was successfully treated with radiotherapy.

Keywords: Adenocarcinoma, Sebaceous, Auricular cancer, Radiotherapy, Skin neoplasms

Introduction

Sebaceous carcinoma is a rare and invasive tumor that stems from the Zeis and Meibomian glands around the eyes as well as in sebaceous glands in other head and neck areas and less commonly on the trunk. It is more prevalent in women aged 60 to 70 years and in Asian populations.[1,2] It can mimic the clinical and histopathological appearance of other carcinomas, hence its diagnosis can be challenging.[3] Common risk factors contributing to the development of this carcinoma include older age, history of irradiation, immunosuppression after solid organ transplantation, and Muir Torre Syndrome. Factors that make the prognosis weaker include: multicentricity, size>1cm in diameter, poor differentiation, extensive tissue infiltration, vascular or lymphatic involvement, pagetoid change, and duration of symptoms >6 month. Tumors less than 6 months old have a very good prognosis. Mortality rate varies from 20 to 22%. The treatment of choice for well-differentiated sebaceous carcinoma is complete resection of the lesion with a safe margin of 5 to 6 mm. In poorly-differentiated tumors, treatment involves extensive removal of the lesion, followed with radiotherapy.[4] Given that there are few reports of this rare case, we decided to report it.

Case Presentation

The Ethics Committee of Mashhad University of Medical

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An 82-year-old Iranian man presented with an erythematous, exudative plaque with induration, which had begun a year before and increased in size over this time. On physical examination, there was a firm, painless, immobile and erosive plaque with a lobular surface on the patient's left external ear (Figure 1). Differential diagnoses such as squamous cell carcinoma, amelanotic melanoma, and malignant adnexal tumor were made. Deep incisional biopsy and histopathology study of the mass were performed, showing an epidermis with neutrophil-containing parakeratotic keratosis, ulcerative neoplastic proliferation consisting of multifaceted cell clusters and lobules with coarsely vacuolated eosinophilic cytoplasm, and atypical nuclei with multiple mitoses (which are indicative of sebocytic differentiation); a number of dyskeratotic and necrotic cells were further detected with extension to the edge and depth of fibrotic stroma with sunlight changes in collagen fibers, with the possible diagnosis of poorly-differentiated sebaceous gland carcinoma (Figure 2a,b,c).

To confirm the sebaceous carcinoma differentiation, immunohistochemical staining for epithelial membrane antigen (EMA) and adipophilin was performed.1 In our study, the EMA of the tumor cells was positive and the carcinoembryonic antigen was negative (Figure 3a,b). With a final diagnosis of auricular sebaceous carcinoma and due to the metastatic potential of the tumor, chest x-ray (CXR) and head and neck computed tomography (CT) scan without contrast were performed, indicating no evidence of metastasis or lesion invasion. In the case of our patient, surgical treatment was contraindicated as the definitive approach. Therefore, non-surgical treatment such as definitive radiotherapy was recommended. A total of 35 radiotherapy sessions with a total dose of 70Gy (centigray) for the left ear and 50Gy for the left side of the neck were carried out (CTV mean dose 71.92 Gy, GTV mean dose 72.95 Gy, and PTV mean dose 71.32 Gy), which yielded an acceptable therapeutic outcome. Follow-up was done daily after each initial session, with subsequent follow-up sessions every two weeks for three months and then, monthly during the treatment. There was no evidence of erosive mass at the end of the treatment, only a single wet patch was seen with desquamation. Unfortunately, the patient died at the end of the first year due to kidney failure; thus, it was not possible to follow him up for a recurrence of the tumor.

Discussion

Sebaceous carcinoma can mimic clinical and histopathological appearance of other carcinomas, resulting in a challenging diagnosis.[4] Immunohistochemistry can be used to differentiate sebaceous carcinoma from basal cell carcinoma and squamous cell carcinoma. The most useful immunostains in the differential diagnosis of PSC are EMA, Ber-EP4, androgen receptor (AR), and adipophilin.[3]

Pathology feature shows multivacuolated cells with clear cytoplasm and indented nuclei as evidence of sebaceous differentiation.[5] There are less than 15 known cases originating from the external auditory canal[6] and only 8 cases of the in-situ type have been reported in the extraocular

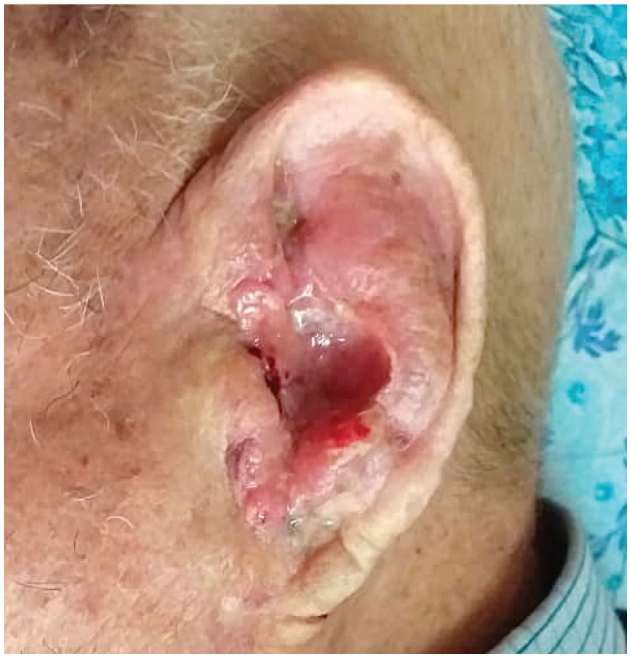

Figure 1. This figure shows the auricular sebaceous carcinoma: an erythematous exudative plaque with induration on the external ear.

area. Similar to our patient, a 2020 case report showed sebaceous carcinoma in the area of the external ear canal, which was completely removed by tumor surgery.[7] The treatment of choice for well-differentiated sebaceous carcinoma is complete resection of the lesion with a safe margin of 5 to 6 mm. In poorly-differentiated tumors, treatment involves extensive removal of the lesion, followed with radiotherapy.[6] Regular examination of the skin and lymph nodes and other organs should be performed periodically. Use of Mohs micrographic surgery has been reported with lower recurrence rates.[4] Previously, this rare cancer was shown not to be sensitive to chemotherapy; however recently, the role of chemotherapy has been evolving.[8] In our case, surgery was not possible due to an underlying disease, but radiotherapy resulted in complete tumor regression.

Conclusion

Sebaceous carcinoma is a rare but invasive tumor. Due to the lack of definite pathophysiology, diagnosis requires pathology and immunohistochemistry, imaging, determining family history in terms of accompanying Muir Toure syndrome, evaluation of lymph nodes, and, if necessary, sentinel lymph node biopsy.

The first-line treatment is surgery. Adjuvant treatment with radiotherapy is preferable to chemotherapy. In the present case, due to comorbidities such as kidney and heart failure, local radiotherapy was preferred, while no surgical treatment was implemented.

Informed Consent

The patient signed the informed consent prior to the treatment.

Conflict of Interest

None declared.

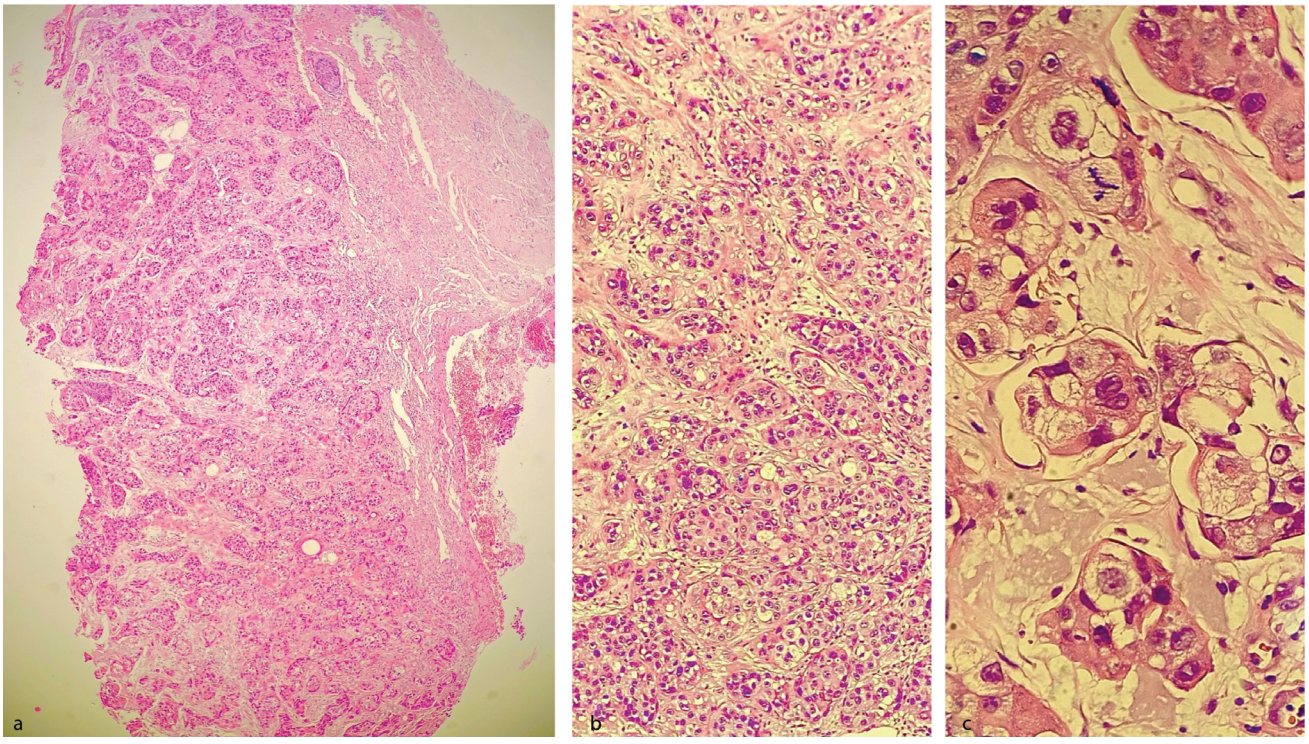

Figure 2. (a,b,c): Histopathologic feature of the tumor, hematoxylin and eosin stain (H & E) (magnification ×40, ×100, ×400) . a: Ulcerative neoplastic proliferation consisting of multifaceted cell clusters and eosinophilic lobules within the entire dermis. b: Large nests and jagged lobules with eosinophilic cytoplasm and mild inflammation in peripheral stroma. c: At high magnification, there are eosinophilic cells with atypical nuclei, coarsely vacuolated cytoplasm with obvious sebosytic differentiation. Multiple mitoses and pyknotic nuclei (arrow).

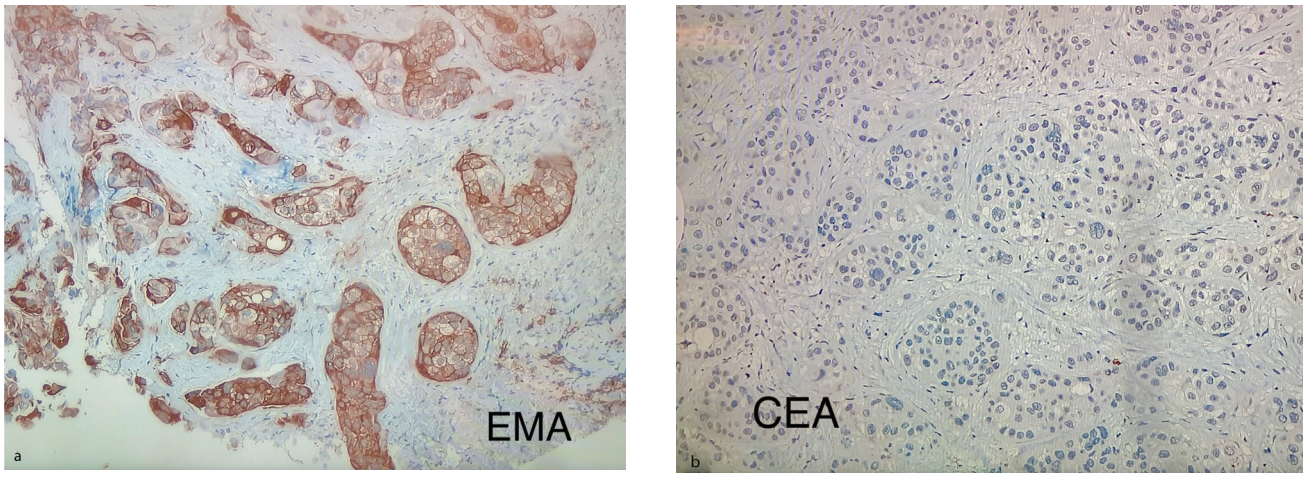

Figure 3. This figure shows the immunohistochemical stain for EMA and CEA. a: Positive for EMA (×400 magnification); b: Negative for CEA (×400 magnification); EMA (×400 magnification); b: Negative for CEA (×400 magnification).

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