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Adenoid Cystic Carcinoma - A Case Report

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Abstract:

The diagnosis of salivary gland malignancies has always posed a challenge for the oral pathologist. The vastness and varieties of the salivary gland malignancies have added to unpredictable nature of the lesions; hence a precise and timely diagnosis is required for a better prognosis. Here, we report a case of adenoid cystic carcinoma of the palate in a 40-year-old female.

Keywords:	*Salivary Gland Carcinomas, Adenoid Cystic Carcinoma, Maxilla, Palate*
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1. Introduction

Adenoid cystic carcinoma (ACC) (formerly known as cylindroma) is a slow growing but aggressive neoplasm with a remarkable capacity of recurrence. It is characterized by proliferation of ductal (luminal) and myoepithelial cells in cribriform, tubular, solid and cystic patterns.{1}

The salivary gland most commonly involved by this tumor is the parotid, the submaxillary salivary gland followed by the accessory gland of the palate and tongue. The adenoid cystic carcinoma can occur in any salivary gland, however 50% develop in the minor salivary glands. ACC usually occurs as a slowly growing tumor. Patients usually complain of a constant, low-grade, dull ache, which gradually increase in intensity. Facial nerve paralysis may develop in parotid tumors. Palatal tumors can be smooth surfaced or ulcerated.{2}.

Here, we report a case of adenoid cystic carcinoma in a 40-year female in which the palatine salivary glands are involved.

2. Case Report

A 40–year old female patient reported to the outpatient department with a swelling on her upper right palate for two months. Patient gave a past history of pain in her upper right back tooth, which was sharp and intermittent pain which lasted for two days.

The pain subsided on its own without any medication. However, after a month of the pain the patient felt a swelling on the palatal aspect of the same side which was sudden in onset, consistent in size and not associated with any suppuration. Her past medical, surgical and dental history was non-contributory.

Intraoral examination revealed the presence of all permanent teeth on all the four quadrants except for the third molars. The solitary swelling seen on the right palatal aspect extended from the mesial side of 15 to distal region of 17. (Figure1) It was well defined with erythematous shiny surface covering a portion of the palatal surface. On palpation swelling was non tender, firm in consistency without any fluid thrill, non-compressible and non-suppurative. The submandibular lymph nodes (bilaterally) were palpable, non-tender and measured 0.5x0.5 cm in size approximately.

Radiographic examination revealed an irregular radiolucency with diffuse radiopacity extending from the mesial aspect of 15 to distal aspect of 17. (Figure 2). It was provisionally diagnosed as salivary tumor.

Histopathological examination revealed lesional cells arranged in cords, nests, islands, cribriform areas and few ductal structures containing central eosinophilic coagulum. The neoplastic cells were uniform, basaloid in nature with hyperchromatic nuclei. Few areas showed epi-myoepithelial islands. The islands are separated from the overlying parakeratinized stratified squamous epithelium by the dense fibrous connective tissue. (Figure 3 & 4).

Correlating the clinical, radiographic and histopathology findings, a final diagnosis of adenoid cystic carcinoma was made.

Treatment was partial maxillectomy with adequate surgical margins. Excisional biopsy was also confirmatory for ACC.

3. Discussion

The ACC is classified as malignant tumors of the salivary glands [3] according to WHO classification 2017. The gene mutation for ACC is t (6;9) (q22-23; p23-24) MYB-NFIB which is present in 90-100% of ACC [4]

ACC is a relatively uncommon neoplasm representing about1 % of all the malignant tumors in the head and neck region and is the fourth most common malignant salivary gland tumor accounting for 10% of all such tumors [5]

ACC is slightly more prevalent in females than males and occurs most commonly during the fifth- sixth decades of life.[6] Our case also presented in a 40-year-old female.

In general, salivary gland tumors are usually best depicted on MRI. The involvement of cranial nerves and tumoral infiltration around the nerves and osseous structures is optimally assessed via non – contrast T1 –weighed and contrast – enhanced, fat suppressed T1-weighted MR sequences.[7]

Grossly, ACC tends to be poorly circumscribed, unencapsulated, and firm with a white to gray-white cut surface and significant variability in size.[8] Hemorrhage and necrosis are not common features, but if observed should suspect a high grade of malignancy.

Histologically, the tumor is heterogenous with main three distinct patterns: The tubular pattern is the least common type and lowest grade with small nests of ductal cells surrounded completely by a second myoepithelial cell layer in a back ground of eosinophilic hyalinized stroma.

The cribriform pattern is most common and is comprised of invasive islands of basaloid cells with many cyst-like spaces, referred to as pseudocyst forming a "swiss cheese "or "sieve –like" pattern. The solid pattern demonstrates large islands and cords of carcinoma with areas of necrosis, increased mitotic activity and perineural spread.{9} Our case also presented with a mixture of cystic and solid areas.

The likelihood of lymph node metastases increases by 5-10-fold in patients with high-grade variants of ACC occurring in 43-57% of patients with tumors showing these histologic features.{10}

The optimal treatment of ACC has not yet been fully established. Most authors advocate the use of surgical excision and postoperative radiotherapy. Some series have found no statistically significant difference between patients treated with combination therapy and those treated with surgery alone.{11}Therefore, proper physical examination followed by surgery and radiation with close surveillance and careful attention to quality of life issues are the cornerstones of management of this disease. The differential diagnosis includes polymorphous adenocarcinoma , carcinoma ex pleomorphic adenoma, and basal cell adenocarcinoma.{12}

4. Conclusion

We present a case of adenoid cystic carcinoma of palatal salivary gland with foci of high-grade transformation / dedifferentiation. This presentation usually warrants a combined therapy for better outcome.

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Figures & Legends:

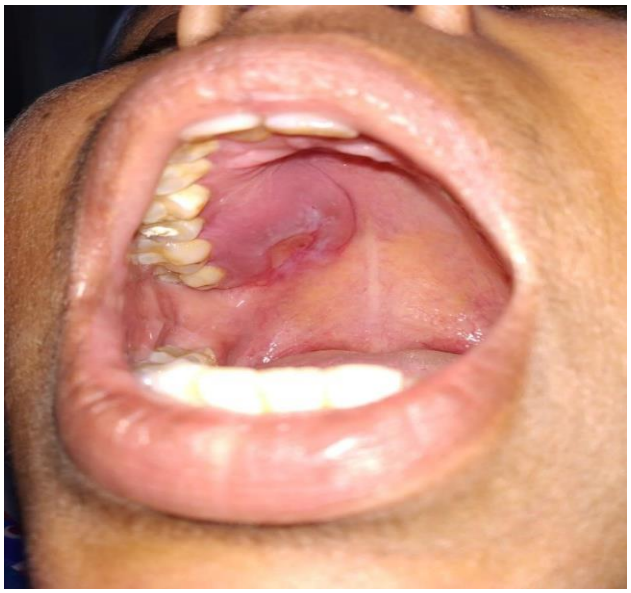

Figure 1: the clinical picture showing the swelling in the right palate

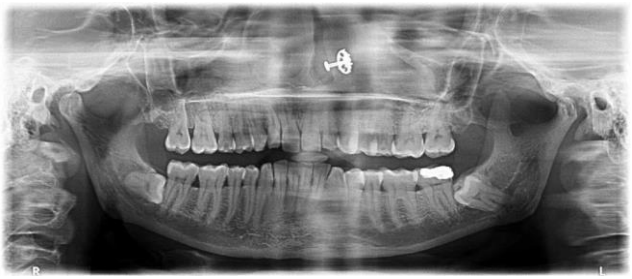

Figure 2: the orthopantomograph showing a radiolucent lesion in the right maxillary posterior region

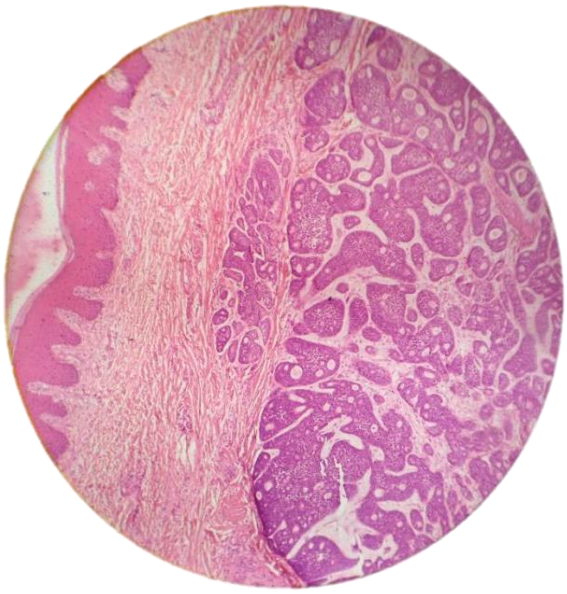

Figure 3: the photomicrograph showing the separation of tumor islands from the mucosal surface. (H & E 20x)

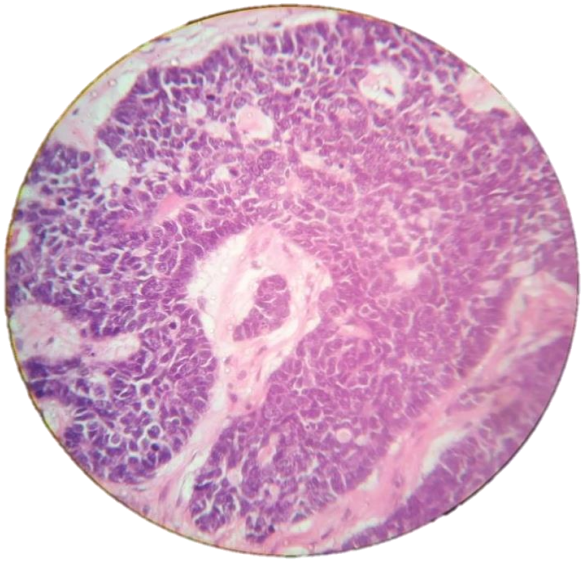

Figure 4: the photomicrograph showing the solid and cystic areas of basaloid tumor cells (H & E 40x)