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Case Report

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Adenoid Cystic Carcinoma: A Case Report

Abstract

Adenoid cystic carcinoma (ACC) accounts for approximately 10 % of all salivary gland tumors. It is the most common malignant tumor of submandibular and minor salivary glands. The most common location is the palate, generally in the area of the greater foramen. ACC is generally characterized by a slow growth rate, and it is often present for several years before the patient seeks treatment. A 28 year old man reported with a swelling on the left side of palate since one and a half years which was diagnosed histopathologically as ACC. A detailed description of the case along with review of literature is presented here with an aim to focus on the importance of detailed or advanced investigations as in the present case.

Keywords: Salivary gland tumors, Computed Tomography, Swiss cheese.

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Introduction

ACC is a rare malignant tumor that affects the major and minor salivary glands, the lacrimal glands, ceruminous glands and occasionally the excretory glands of the female genital tract. 30-40% of these carcinomas occur as head and neck tumors. Approximately 50 to 70% of the reported cases occur in minor salivary glands. ACC is well known for its prolonged clinical course and the tendency for delayed onset of distant metastasis. The tumor displays a distinct cribriform

histomorphology often described previously as Swiss cheese or Sieve like but this terminology has proved inadequate in expressing the wide spectrum of histological diversity that may be seen. Current treatment recommendations for ACC include complete surgical resection and postoperative radiation therapy. Tumor recurrence rates vary in the literature but reportedly can be as high as 42%. Although the prognosis is poor, the course of disease is often indolent and patients with ACC survive for many years [2]. The aim of the present case reported

here, is to focus on the importance of detailed investigations.

Case report

A 28 year old male patient reported to the Department of Oral Medicine and Radiology with a chief complaint of painless swelling on the left side of the palate since one and a half years. Patient noticed the swelling six months after extraction of a decayed upper molar tooth. The swelling was slow growing, painless and persistent. There was no reduction in size of the swelling since the patient had noticed it. Personal history was negative for tobacco consumption, cigarettes or alcohol. On Extra oral examination no apparent abnormality was detected. Intraoral examination revealed a well defined; dome shaped swelling measuring 3x4cm. The swelling was seen to be extending from rugae area to junction of hard and soft palate posteriorly and mediolaterally from the mid palatine raphe to the palatal aspect of tooth number's 24,25,26,27 region.(Fig.1) The overlying mucosa was intact and of normal colour. Posterior aspect of the lesion appeared to be fluctuant. On hard tissue examination, tooth number 26 was missing. The provisional diagnosis was Residual cyst i.r.t. 26 while the differential diagnosis was Benign salivary gland tumor, Mucoepidermoid carcinoma, Pleomorphic adenoma, Adenocarcinoma. Radiographic investigations including Intraoral periapical radiograph, maxillary topographic occlusal and Paranasal sinus view were not suggestive of any apparent pathology involving bone or dentition of the left side of maxilla.

Fine Needle Aspiration Cytology (FNAC) was performed and Sero-sanguinous fluid was aspirated. (Fig.2).Smear showed many clusters of glandular epithelial cells and eosinophilic globules along with blood cells. Findings were suggestive of a benign salivary gland tumor. Plain and Post Contrast Axial and Coronal View (Computed Tomography) of facial bones was done and revealed peripherally enhancing well defined round hypo dense areas in the left hard palate anteriorly to the inner cortex of the maxilla most likely representing a benign mixed tumor.

An excisional biopsy was performed under local anesthesia. After reflection of a mucoperiosteal flap, the lesion was enucleated in total and submitted for histopathological evaluation. The lesion appeared macroscopically as an encapsulated soft tissue lesion that was separated from the adjacent tissue, without evidence of bony involvement.

Histopathological Examination revealed islands of hyperchromatic basilooid epithelial cells containing multiple cyst like spaces filled with hyalinized eosinophilic product forming cribriform and tubular structures. The tumor cells showed infiltration into the normal appearing stroma. The above features were suggestive of ACC. Since the report revealed ACC a malignant salivary gland tumor, the patient was referred to a higher centre for Radiation therapy. Patient was not willing to undergo radiation therapy. Patient was reviewed periodically but no recurrence was noted clinically after a year.

Discussion

ACC is a rare tumor of the head and neck region. However, it is the most common malignant tumor of minor salivary glands [3] of the palate, as in our case. The tumor affects men and women equally and usually occurs in the fifth decade of life, but in the present case the patient was 28 years old. In a similar case report, patient aged 30 years also had ACC [3]. Most Authors report common sites such as the parotid, sub maxillary and the accessory glands in palate and tongue and the lesser common sites are vulva, cervix, Cowper's glands, esophagus, external auditory canal, middle ear and nasopharynx .Rarely, it may also present as a primary intraosseous tumor of the mandible and maxilla [4].

Clinical presentation is often as an asymptomatic mass, however, this tumor is more likely than others to present with pain or paresthesia and numbness [5]. This tumor has a marked tendency to invade nerves and is seen in about 80% of all cases. Perineural invasion was clinically not evident in the present case. Facial nerve paralysis may be seen more frequently with ACC than with other tumors. Minor salivary gland involvement

is characterized by a submucosal mass with or without pain and ulceration. This is an interesting tumor with two distinct clinical entities [6]. One group has a relentless fulminating course with early metastasis and fatal outcome within a short period of 2-3 years. The second group has an insidious natural history and long survival period despite local recurrences. Most of the cases fall in the second category & is termed as “the patient and the tumor were existing in symbiosis” type. Lymphatic spread is very rare. Lymph nodes, however, in very extensive cases, may be involved by direct extension [7]. In long standing cases distant metastasis occurs via the blood stream most commonly to the lungs and bones.

Confirmatory diagnosis of ACC is primarily based on the characteristic histological features which play a significant role not only in diagnosing the tumor but also helps to determine treatment and prognosis. Three histological patterns of growth have been described. The typical ACC has a cribriform pattern-nest and columns of cells of rather bland appearance are arranged concentrically around gland-like spaces which are filled with hyaline Periodic acid schiff positive material as in our case. Some have a predominantly tubular pattern while a few others have a solid pattern.

Radiological investigations, especially Computed Tomography scans are important to delineate the tumor, to plan extent of surgery and to look out for recurrences as a follow up postoperatively. Treatment of these tumors includes surgical excision and postoperative radiation therapy as in the present case [8, 9, 10].

Conclusion

The primary treatment objective in ACC patients is local control, normal functionality and distant metastasis prevention. For this purpose, early detection by the team of dental specialists is a pre-requisite, in order to enable a more favorable prognosis and better quality of life. The role of the usefulness of various diagnostic modalities like FNAC, biopsy and advanced diagnostic imaging techniques like Computed Tomography has been mentioned in the present case. The

therapy involving combination of surgery and radiotherapy remains the modality of choice in most cases.

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Conflict of Interest

No conflict of interest associated with this work.

Contribution of Authors

We declare that this work was done by the authors named in this article and all liabilities pertaining to claims relating to the content of this article will be borne by the authors.

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