Case Report

Adenoid cystic carcinoma of alveolar mucosa: A rare case report

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Abstract

Adenoid cystic carcinomas are rare epithelial malignancies of salivary glands that show slow growth and local invasion with recurrences seen many years after diagnosis. Upto 50% of tumors occur in intraoral minor salivary glands usually in the hard palate. Tumors on the alveolar mucosa are extremely rare. We present a case of adenoid cystic carcinoma on the right alveolar mucosa in a 35 year old female. The case is been presented for its rarity.

Keywords: Adenoid cystic carcinoma, alveolar mucosa, fine-needle aspiration cytology

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INTRODUCTION

Adenoid cystic carcinoma (ACC) is a rare epithelial malignancy of the salivary glands, lacrimal glands, ceruminal glands and occasionally excretory glands of female genital tracts.[1] It accounts for the fifth most common epithelial neoplasm of the salivary glands.[2] These are clinically innocuous lesions usually characterized by slow evolution, small size, slow growth,[3] locally invasive growth with high propensity for local recurrence and distant metastasis.[4] Majority of the cases arise in minor salivary glands, particularly the palate. Rarely, these lesions appear in the alveolar mucosa. We report a case of ACC on the right alveolar mucosa in 35-year-old patient with diagnosis based on fine-needle aspiration cytology (FNAC) and histopathology.

CASE REPORT

A 35-year-old female reported with the complaint of painful swelling in the right alveolar mucosa of 4 months' duration. The swelling gradually increased in size and was associated with pain which was mild and continuous in nature. On general examination of the patient, the patient was moderately built, well-nourished and well-oriented with all vital signs within the normal limits.

On intraoral examination, there was a diffuse, ill-defined swelling on the right alveolar mucosa in the molar region. Palpation revealed tender, movable swelling, bluish red, 1 × 2 cm and soft to firm in consistency [Figure 1]. The patient reported no significant medical history or any addiction. Orthopantograph revealed no significant bony changes. The

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patient was referred to the Department of Oral Pathology for FNAC and blood investigations [Figure 2]. Laboratory blood studies revealed normal results. Cytological smears wet fixed and stained with hematoxylin and eosin showed well-delineated, tightly cohesive clusters of basaloid cells surrounding mucoid and hyaline globules [Figure 3]. These findings were suggestive of adenoid cystic sent for the histopathological diagnosis. Histopathological examination revealed multiple pseudocystic spaces of variable sizes with cuboidal cells demonstrating oval nuclei and scant cytoplasm surrounded with the eosinophilic material [Figures 4]. All these features were diagnostic of ACC.

DISCUSSION

ACC is an aggressive malignant tumor characterized by indolent, locally invasive growth with high propensity for local recurrence and distant metastasis with a unique natural history since its first description by 2 Frenchmen, Charles Robin and Alexandre Laboulbene in 1853.[5] Its deceptively benign histologic appearance has been recognized for more than 100 years. It was first described as cylindroma by Theodor Billroth in 1856.[6,7] Krompecher named it as basiloma in 1908.[2] Later Spies in 1930 and Foote and Frazell in 1953 coined the term "cylindroma."[2] They credited the late James Ewing for having used that term for many years. Hence, the tumor has been variously referred to as "cylindroma," "basiloma," "adenocystic basaloid carcinoma," and "adenoepithelioma."[8] The term "ACC" is in use till date.[9]

The World Health Organization defines ACC as a "basaloid tumor consisting of epithelial and myoepithelial cells in various morphological configurations, including tubular, cribriform and solid patterns. It has a relentless clinical course and usually fatal outcome."[10] It comprises of 5%–10% of all salivary gland tumors, which account for 2% to 4% of all head and neck malignancies.[8,11] About 31% are most frequently located in minor salivary glands predominantly the palate. According to the previous

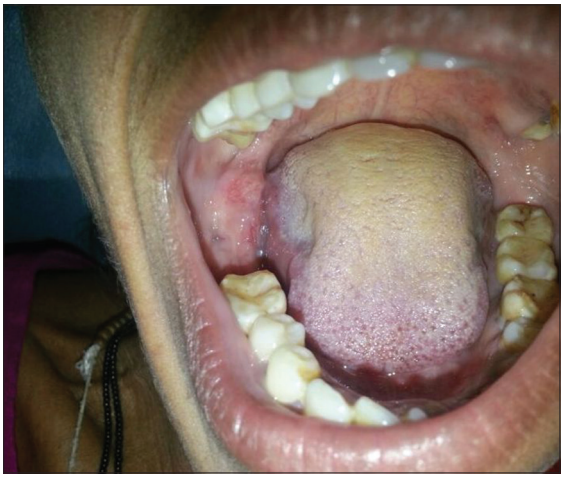

Figure 1: A diffuse ill-defined movable bluish red swelling on right alveolar mucosa in the molar region

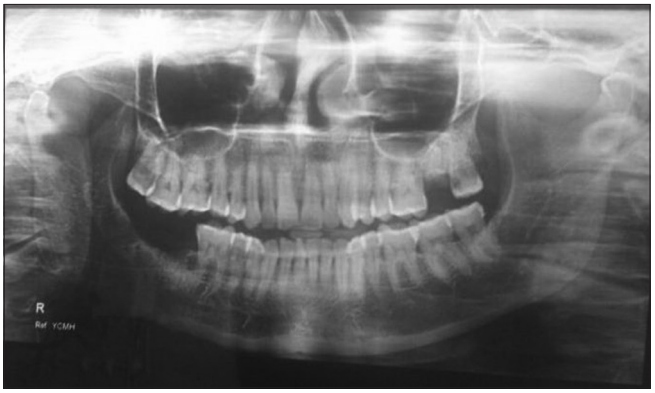

Figure 2: Orthopantograph revealed no significant bony changes

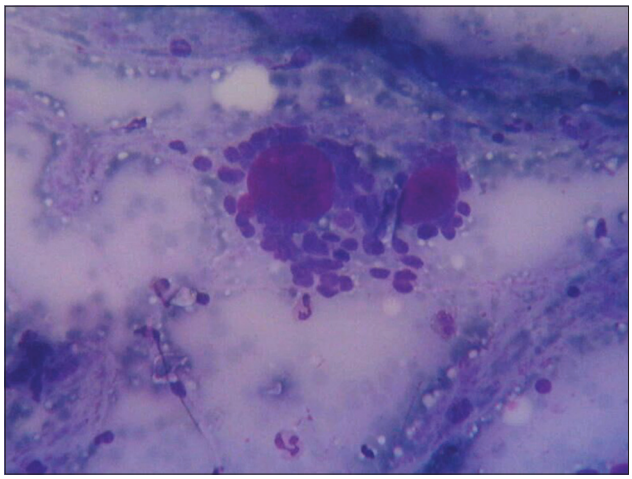

Figure 3: Fine-needle aspiration cytology stained smear showed well-delineated, tightly cohesive clusters of basaloid cells surrounding mucoid, hyaline globules

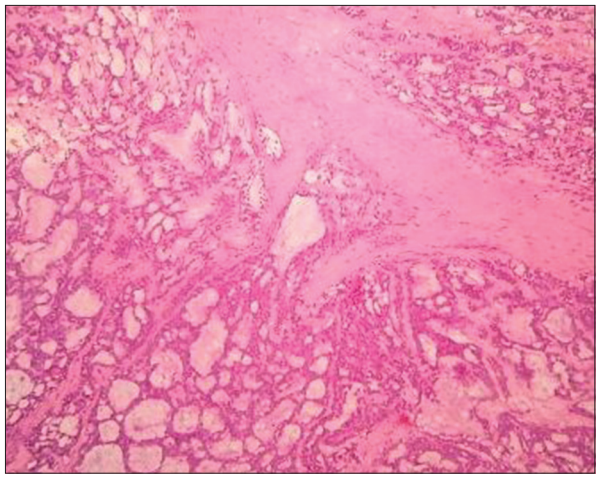

Figure 4: Photomicrograph revealed multiple pseudocystic spaces of variable sizes filled with eosinophilic material

literature reported, few cases are also encountered in the submandibular gland and parotid gland.[11] Till date, very few cases are reported in the alveolar mucosa, the present case is reported in this review for its rarity.

The clinical presentation of ACC involves a slow growing, firm and unencapsulated mass.[12] Pain is an important symptom of the condition due to its propensity for perineural spread[10] occasionally occurring before clinical evidence of the disease. Pain is often continuous and dull aching in nature.[12] The clinical course is characterized by an initial period of slow and indolent growth that is usually asymptomatic. In most cases, the tumor goes unnoticed until it has invaded local nerves and structures causing varying symptoms depending on location.[12] Thus, ACCs have a long clinical course and questionable prognosis with minor salivary gland ACCs having a worse prognosis than those of the major salivary glands. Local lymph node involvement is rare.[10] Lymphatic spread occurs less commonly than with other malignant epithelial tumors. Blood spread to distant sites occurs late in the course of the disease and distant metastasis is common particularly in the lung, and usually whenever the primary tumor has been inadequately treated.[8]

Histopathologically, three histological growth patterns have been identified and described: cribriform, tubular and solid. Cribriform is the most common histologic subtype. Perineural invasion along cranial nerves is a pathognomic factor of ACC and is believed to be responsible for the high rate of local recurrence.[8] Necrosis and vascular invasion are present at a lower rate.[13] Assessment of the histologic grade is of significance in predicting the likelihood of tumor recurrence and survival. In one series of studies, 5-year recurrence rates of 100%, 89% and 59% were reported for tumors with solid, cribriform, or tubular growth patterns, respectively.[8]

Various treatment modalities that have been proposed in ACC which include surgery, radiotherapy, chemotherapy and combined therapy. Surgical excision with wide margins is the treatment of choice. We have followed a similar treatment protocol.[10]

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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