

Adenoid Cystic Carcinoma - A Rare Case Report

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Abstract: Adenoid cystic carcinoma also known as cylindroma is a rare tumor, accounting for only 1% of all malignant tumors of the oral and maxillofacial region. It accounts for 22% of all salivary gland malignancies and involves both major and minor salivary glands but the percentage for involvement of minor salivary gland is more than that of major salivary glands.¹ It is a slow growing tumor and is often present for several years but the occurrence is relatively high with distant metastasis.² Both men and women are equally affected from the tumor and occurs usually in 5th decade of life. In this article we present a case of adenoid cystic carcinoma which occurred in a 35-year-old patient with swelling of right maxillary region and pain.

I. Introduction

Adenoid cystic carcinoma is a rare salivary gland neoplasm. Earlier it was also called as cylindroma but as the term cylindroma is also used for skin adnexal tumor so the term should be avoided. It has distinctive histopathological features.³ It affects both major and minor salivary glands but according to researchers it involves 15 to 30% of submandibular gland, 50% of minor salivary gland and 2 to 15% of parotid gland.⁴ Most common site for occurrence of this tumor is palate. It can also arise in other sites of head and neck also.² Adenoid cystic carcinoma presents as a swelling of long duration which can be painful and ulcerated. Both men and women are equally affected from the tumor and occur usually in 5th decade of life. In adenoid cystic carcinoma patients usually complaints of slow growing swelling with dull aching pain which is gradually increasing, there can be facial nerve paralysis also when it involves the parotid gland.^{5,6} The aim of this article is to report a new case of ACC, in an attempt to distinguish it from other salivary gland tumors.

II. Case Report

A 35 year old patient presented with gradually increasing swelling with pain in upper right back region of jaw for last 8 months. He visited a dentist 6 months back for similar complaint, where extraction was done which aggravated the swelling.

On extraoral examination a single swelling was present on right side of the face extending from lateral border of nose till 3 cm in front of the ear. It was tender on palpation. On intra oral examination 16, 17 and 18 were missing. Grade III mobility was present in relation to 13, 14 and 15. Swelling with vestibular obliteration was present extending from 14 to 18 medially it was extending till hard palate (**Figure 1 and Figure 2**). The swelling was tender on palpation and firm in consistency and well defined borders. Pus discharge was also present from sinus opening in relation to 14. On examination there was also destruction of nasal septum.

The patient was advised for panoramic radiograph. This radiograph showed destruction of inferior border of maxillary sinus with radiopacity in the sinus of right side. Root resorption was also seen in relation to 13, 14 and 15 (**Figure 3**). Patient was advised for computed tomography scan for obtaining a more precise view but patient was not willing to go for it because of financial problem. Incisional biopsy was done. The report of the biopsy revealed numerous tumor island showing cribriform or swiss chesse pattern. The tumor cells are darkly stained cells which display a basaloid appearance with angulated, hyperchromatic nuclei and scant, clear to eosinophilic cytoplasm. Few islands show tubular and solid pattern. Few areas of perineural invasion are also evident. Based on above features a diagnosis of Adenoid Cystic Carcinoma was made with a differential diagnosis of malignancy of maxillary sinus, mucoepidermoid carcinoma and adenocarcinoma.

Surgical resection was done and patient was recalled after 1 month. The swelling was reduced. Patient is still under follow up.

III. Discussion

ACC was first described by Robin and Laboulbène in 1853 and later it was named by “Billroth” as ‘cylindroma’ due to its characteristic histological appearance.⁶ It is a rare salivary gland neoplasm although it can appear at different body sites. It is the common malignancy of lacrimal glands and secondly it involves salivary glands. Most common site for occurrence of this tumor is palate.⁷ It can also arise in other sites of head and neck also. Adenoid cystic carcinoma presents as a swelling of long duration which can be painful and ulcerated. It is a gradually increasing swelling which can cause facial nerve paralysis. There will be insidious destruction of surrounding tissues and perineural invasion.⁸ There are three histological patterns of growth: solid, cribriform and tubuloductal. The World Health Organization (WHO) histological definition is ‘an infiltrative malignant tumor having a very characteristic cribriform appearance. The tumor cells are arranged as small duct-like structures or larger masses of myoepithelial cells disposed around cystic spaces to give a cribriform or lacelike pattern.’⁹ Again in 2005 WHO defined it as “A basaloid tumor consisting of epithelial and myoepithelial cells in various morphological configurations, including tubular, cribriform and solid pattern. It has a relentless clinical course and usually a fatal outcome.”¹⁰

The researchers reported a poor prognosis in adenoid cystic carcinoma. But in this patient there is no recurrence seen till now. In solid growth pattern reported a frequent recurrence rate approximately 100%. Tubuloductal architecture reported more typical growth rate and the lowest rate of recurrence at 59%, the cribriform architecture, which is associated with recurrence rate of 89%. The cribriform pattern is the most common form which consists of islands of basaloid epithelial cells that contains multiple cylindrical, cystlike spaces. In tubular pattern multiple small ducts or tubules are seen whereas in solid pattern larger islands or sheets of tumor cells are present. The cause of ACC is unknown no predisposing factors are reported. Patients often complain of slow growing mass which is recognized when it becomes large. The mass is usually asymptomatic and painless. Bone invasion can be seen.¹¹

Lymphatic spread of ACC is common in which cervical lymphatic metastasis is about 8%. Although spread to regional lymph nodes is rare, distant metastases, particularly to lungs and bone, are more common. CT and MRI are helpful in diagnosis of ACC, but in this patient he was not willing of CT SCAN.^{2,3}

Surgical resection with wide margin is the treatment of choice for better prognosis. In our patient we performed tumor resection with postoperative radiotherapy. The report of the biopsy revealed numerous tumor island showing cribriform or swisscheese pattern. The tumor cells are darkly stained cells of uniform size. Few islands show tubular and solid pattern. Few areas of perineural invasion are also evident. Based on above features a diagnosis of Adenoid Cystic Carcinoma was made.

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