

## Case Report

# Adenoid cystic carcinoma in maxillary antrum - A rare presentation

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## Abstract

Adenoid Cystic Carcinoma is a rare malignant salivary gland tumor comprising <1% of all malignancies of head and neck and 10-15 % of the malignant tumors of salivary glands. Most of it arises in minor salivary glands nearly about 60%. Intraorally, about 50% of adenoid cystic carcinomas occur in the palate. Here by, we present a case of adenoid cystic carcinoma affecting the left maxillary antrum and palate on the same side in 30 years old male along with a brief review of literature.

## Key words

Adenoid Cystic Carcinoma, Malignant salivary gland tumor, Maxillary antrum, Palate.

## Introduction

Adenoid Cystic Carcinoma is rare tumor of head and neck comprising 10-15% of malignant tumors of the salivary glands [1]. AdCC involving the nose, paranasal sinuses and maxillary antrum are very rare and carry the worst prognosis [2]. Parotid gland is the most common site origin of AdCCs nearly about 25% in head and neck. Most of the AdCCs arise in the minor salivary glands (60%) [3]. AdCCs causes wide local infiltration, perineural spread, local recurrence and distant metastasis. Complete surgical resection of the tumor with postop

radiotherapy in case of close margins, perineural invasion, extensive primary tumors (stages T3, T4) or high grade histology [3, 4].

Hereby, we present a case of 30 years old male who presented with swelling involving maxillary antrum and palate which on incisional biopsy and histopathological examination was diagnosed as adenoid cystic carcinoma of maxillary sinus.

## Case report

A 30 years old male presented to ENT OPD with a swelling of size 5x5 cm, firm to hard over left

maxillary areas associated with pain and left side nasal blockage. One year back the swelling was smaller but associated with pain for which he got his molar extracted. Intraorally it involved left side of the palate of size nearly 3x3 cm, firm to hard, irregular margins extending from medial side of first premolar to tuberosity involving the palatal area but not crossing the mid line. The molars and premolars of that side were not seen. Oral hygiene was poor and teeth were stained as he was a tobacco chewer. There was no cervical lymphadenopathy. His laboratory investigations included hemoglobin 10 mg/dl, total leukocyte count - 11000/microl, platelet 1 lakhs/micro liter. Serum sodium, potassium, calcium and phosphorus were all within normal limits.

CT scan of the left maxillary sinus showed inhomogenously enhancing mass obliterating the sinus and infiltrating the surrounding bone. He was sent to the department of Pathology for FNAC of the swelling. The smears on examination were cellular with hyaline spherical globules of varying size with adherent tumor cells with scanty cytoplasm, high nuclear: cytoplasmic ratio, nuclear molding and naked nuclei. Incisional biopsy of palate and sinus lining, on histopathological examination revealed moderately collagenous connective tissue stroma infiltrated by numerous cystic spaces lined by basaloid cells in cribriform pattern. Duct like areas were filled with eosinophilic, coagulum. Basaloid cells demonstrated pleomorphism and occasional mitotic figures. There was no evidence of perineural invasion. A diagnosis of AdCC (cribriform pattern) was established. The patient was treated by wide surgical excision with clear margins and hemi maxillectomy of left maxillary region with postop radiotherapy. The patient is under regular follow up (**Figure – 1 to 5**).

### Discussion

Adenoid cystic carcinoma is a rare, slow growing malignant tumor of salivary gland with higher chances of recurrence [5]. It is the most common malignant tumor of minor salivary gland [5]. It

mostly involves the palate. It can occur at any age but commonly in age group 24-78 years with female predilection (F:M = 1.2:1) [6]. Intraorally 50% of AdCCs, occur on the palate followed by lower lip, retromolar tonsillar pillar area, sublingual gland, buccal mucosa and floor of the mouth [7]. The nose and paranasal sinuses represent the next most common sites [8]. There are 3 patterns of growth - cribriform, tubular and solid [9].

Grading of AdCCs [10]

Grade	Type
I	Well differentiated, Tubular and Cribriform, no solid pattern
II	Pure cribriform + <30% of solid pattern
III	Predominant solid

**Figure – 1:** Patient with swelling of size 5x5 cm in left maxillary area.



**Figure – 2:** Intra oral lesion of size 3x3 cm. with irregular margins.



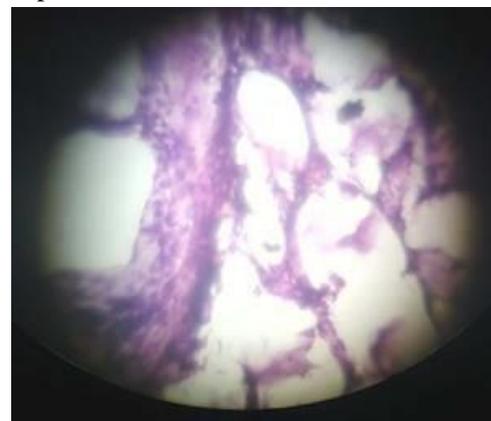
**Figure – 3:** Gross specimen of tumor. Cut section showed solid and infiltrative growth.



**Figure – 4:** H & E Stain, 10X. Collagenous connective tissue stroma infiltrated by columns surrounding glandular spaces filled with eosinophilic material.



**Figure – 5:** H & E Stain, 40X. Collagenous connective tissue stroma infiltrated by columns surrounding glandular spaces filled with eosinophilic material.



Grossly, it has solid appearance and infiltrative pattern of growth [5]. The main ultrastructural

features of AdCC are pseudocysts intercellular space abundant basal lamina and true glandular lumens [5]. Cytological smears show cellular smears with cells arranged singly and in clusters. Hyaline stromal globules are the most striking features of this neoplasm with adherent tumor cells. Tumor cells have scanty cytoplasm, high nuclear: cytoplasmic ratio, nuclear molding and naked nuclei [11]. On histological examination clusters of small basaloid cells with small amount of cytoplasm and uniform ovoid or round deeply basophilic nuclei exhibiting only slight atypia separated by thick strands of homogenous hyaline eosinophilic basement membrane like material. There are many round or ovoid clear vacuoles or gland like spaces between tumor cells are seen [12]. Immunohistochemically, it is positive for keratin, LEA, lysozyme lactoferrin, alfa - antichymotrypsin, S-100 protein and CD - 117 (c KIT) [5]. p 53 over expression is related to resistance to radiotherapy and chemotherapy [13]. Its aggressive behavior by its ability to invade and metastasize is influenced by urokinase type plasminogen activator receptor expression which is a negative prognostic factor [14].

Treatment of choice is total resection of tumor with clear margins [2]. Postop radiotherapy causes regression of tumor and relieves symptoms [2]. Prognostic factor depend on site, stage, perineural invasion, grade and pattern of growth. Tubular and cribriform type has better prognosis than solid type [15]. Minor salivary gland tumor has worse prognosis due to early local infiltration and invasion of surrounding tissue [15].

### **Conclusion**

Adenoid cystic carcinoma is mainly malignant tumor of major and minor salivary gland. Treatment of choice is total resection of the tumor with clear margins. Cribriform type has better prognosis than solid type. It has remarkable capacity of recurrence and perineural invasion. Postoperative radiotherapy causes

regression of tumor and relieves symptoms. Role of Chemotherapy is controversial.

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