International Journal of Health Sciences and Research
www.ijhsr.org ISSN: 2249-9571

Case Report

Adenoid Cystic Carcinoma Involving Palate: A Case Report

Ajmal M[1]*[@], Luqman M[2]*, Bagi AM[3]**, Bokhari K[3]**

*Assistant Professor, **Associate Professor
[1]Division of Oral Medicine & Radiology,
[2]Division of Oral Pathology,
[3]Division of Oral & Maxillofacial Surgery,
College of Dentistry, King Khalid University, Abha, KSA

[@]Correspondence Email: drmajmal@gmail.com

Received: 10/05//2012 *Revised: 01/06/2012* *Accepted: 05/06/2012*

ABSTRACT

Salivary gland tumors are uncommon. They are classically slow growing, with patients being aware of their presence over months or even years. The reported incidence is approximately 3% of all head and neck neoplasms. Tumors of minor salivary gland origin account for only 10–15% of all salivary gland neoplasms. Adenoid cystic carcinoma accounts for 21% to 42% of minor salivary gland tumors and occurs most frequently on the hard palate. This cancer is characterized by its infiltrative growth and perineural invasion. Surgery followed by radiotherapy remains to be the gold standard to treat this carcinoma. We present a case of adenoid cystic carcinoma involving the hard palate in a 40 year old male patient and brief review the literature for adenoid cystic carcinoma in terms of its biological nature, the histogenesis, pattern of spread and the treatment modalities.

Keywords: carcinoma, adenoid cystic carcinoma, salivary glands,

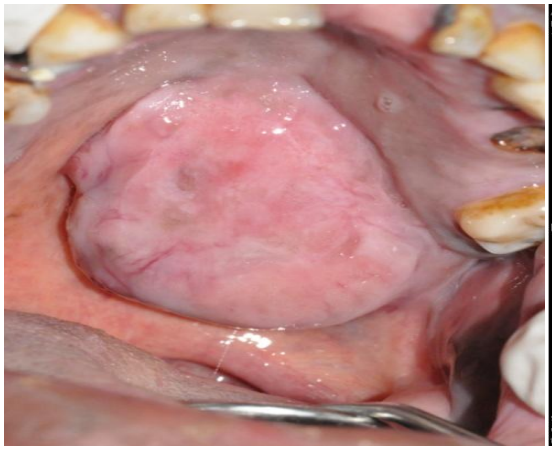
INTRODUCTION

Minor salivary gland tumors (benign and malignant) are a rare clinical entity. [1] Salivary gland carcinomas (major and minor) represent around 3 per cent of all head and neck malignancies. [2] Minor salivary gland tumors represent a heterogeneous group of neoplasm, with a broad range of histological types and growth patterns. Tumors originating in the minor salivary glands are infrequent, and represent less than 20% of all salivary neoplasms. [3] They are usually innocuous, asymptomatic and do not interfere with function till they attain large size. On the basis of appearance

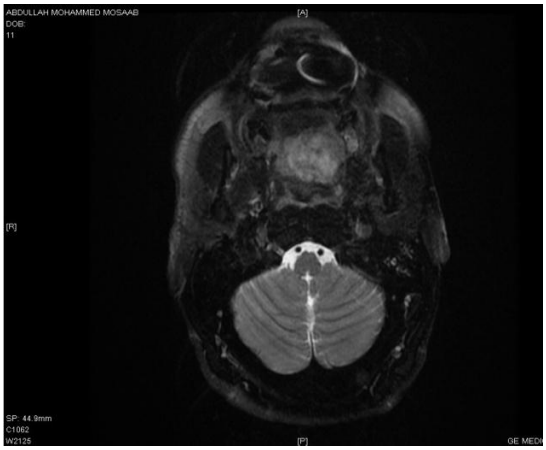
alone, it is not possible to differentiate between benign and malignant variants. The overlying mucosal ulceration is generally only secondarily, from either masticatory or denture trauma or pressure. [4] Age plays very little role in differential diagnosis of minor salivary gland neoplasia, as these lesions can arise at any age. Outcome of treatment of minor salivary gland carcinoma is closely related to the histological type and surgery followed by radiation treatment yield better control of local disease than either surgery or radiation alone. [5] We present a case of adenoid cystic carcinoma in a 40 year old male patient reported to our institution (King Khalid University, College of Dentistry) and review the literature for this carcinoma.

CASE REPORT

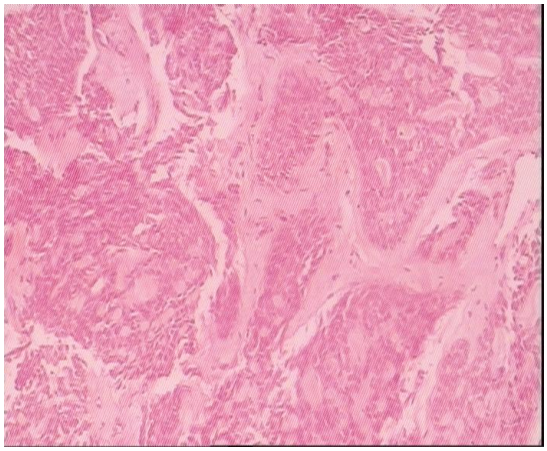
A 40 year old male patient reported to our institution (King Khalid University, College of Dentistry) with a chief complaint of swelling in the posterior hard palate. On clinical examination, intraorally, a 3×3 centimeters firm swelling was noticed which was extending from mid palatal region posteriorly up to the uvula **(Figure-1)**. The swelling progressed to the present size over a period of 3 months. Personal history of the patient in terms of deleterious habits was not significant. The patient complained of dysphagia and dysphonia due to the swelling.



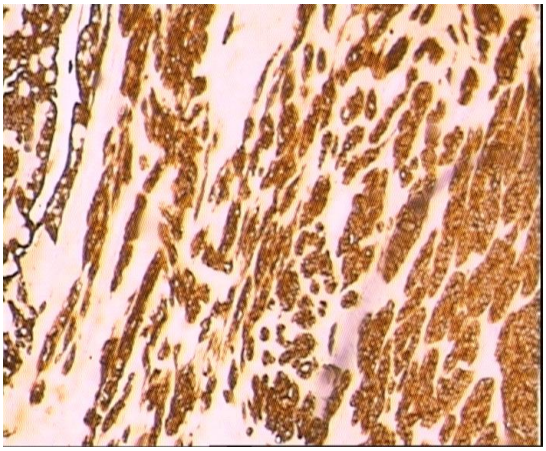
(Figure-1)



(Figure-2)



(Figure-3)



(Figure-4)

The anterior limit was of the lesion was almost up to the midpalatal region. There was no evidence of ulceration over the swelling. The tumor was firm in consistency on palpation with no mobility elicited thus confirming fixity to the underlying structures. Bilateral submandibular lymphnodes were palpable, hard in consistency and were fixed to the underlying structures. A provisional diagnosis of malignant tumor was made based on the rate of growth, fixity to the structures and pattern of involvement of lymph nodes. Patient was advised for an MRI scan **(Figure-2)** which showed 3×3 centimeter oval shaped mass in the mid palatal region with irregular border on right side. A radiological diagnosis of malignancy was reported. Incisional biopsy was planned under local anesthesia which confirmed the clinical and radiological diagnosis of malignancy. Histopathological diagnosis of adenoid cystic carcinoma from the minor salivary glands was reported. (**Figure-3)** shows baseloid epithelial cell nests forming multiple cylindrical cysts like cribriform pattern and solid patterns. Cribriform pattern is seen prominently in the histological section which suggests histological diagnosis as cribriform variant of adenoid cystic carcinoma. It also revealed perineural invasion of tumor cells which is the characteristic of this tumor. Immunohistochemistry CK (AE1, AE3) shows **(Figure-4)** positive staining especially for the luminal cells. Patient has not turned up for follow-up as he was not willing for any treatments.

DISCUSSION

Adenoid cystic carcinoma accounts for 21% to 42% of minor salivary gland cancers and occurs most frequently on the hard palate. It is characterized by its infiltrative growth and perineural invasion. Spread to regional lymphatics is relatively uncommon, but hematogenous spread occurs in 25% to 50% of cases which directly affects the prognosis. Systemic involvement is said to occur in the presence of uncontrolled disease at the primary site. [6] The three major subtypes of adenoid cystic carcinoma are cribriform, tubular, baseloid or solid variants. [7] The baseloid solid pattern is characterized by solid nests or sheets of cells representing areas of anaplastic tumor and carries worst prognosis. [8]

Several studies for adenoid cystic carcinoma have been reported in the literature elaborating the biologic nature, histogenesis, metastatic spread, treatment modalities for this tumor. Chaudhry AP *et al.,* in 1986 studied 12 cases of adenoid cystic carcinoma using light and electron microscopy and found the presence of myoepithelial cells in 9 out of 12 cases representing the histogenesis of this tumor. [9] Takakashi H *et al.,* in 2004 reviewed 200 cases of intraoral minor salivary gland tumors of which 73 were malignant. They reported adenoid cystic carcinoma as the most common malignant tumor with palate as the common site and mean age of occurrence as 47.2 years for females and 50.6 years for males. [10] Garden AS *et al.,* studied the influence of positive resection margins and nerve invasion in 198 patients of adenoid cystic carcinoma. They stated that perineural invasion is associated with an increased rate of local failure. [11] Regezi JA *et al.,* evaluated the clinical, histological and immunohistochemical data on 238 oral minor salivary gland tumors and concluded that adenoid cystic carcinomas stained positive for S-100 protein. [12] This finding of S-100 protein in adenoid cystic carcinoma is a helpful differentiating feature when canalicular adenoma is multifocal. Avery CME *et al.,* in their retrospective study of 15 cases of adenoid cystic carcinoma reported

the influence of combined treatment on adenoid cystic carcinoma and concluded that combination treatment with both surgery and radiotherapy has improved the control of local disease. [13] Beckhardt RN *et al.*, *reviewed* 116 malignant minor salivary gland tumors over a period of 46 years and stated that grade 3 tumor histology, tumor size greater than 3 cm, perineural invasion, bone invasion and positive surgical margins were associated with decreased survival. [14]

Salivary gland tumors are known for late recurrences. [15] In case of adenoid cystic carcinoma, which has a distinctive clinical and histological pattern of behavior, the rate of recurrence is relatively high. The margins of the tumor are usually ill-defined; with narrow columns of cells streaming off from the main mass along the tissue planes between specialized structures such as muscle fasciculi, and most markedly along perineural spaces. Perineural spread is noted microscopically in approximately two-thirds of patients with adenoid cystic carcinoma of the minor salivary glands. [16] The absence of host lymphocytic reaction to tumor cells, the frequency of "skip areas" with clumps of cells permeating the tissue beyond the apparent edge of the tumor, between muscle fibers, in Haversian systems makes the margins of clearance extremely difficult.

Adenoid cystic carcinoma is a characteristic tumor of minor salivary glands with most common site of occurrence in the hard palate. Although we could not do any treatment for this case as the patient was not willing, the biologic basis of this tumor with a characteristic feature of high perineural invasion needs to be considered in the treatment planning. A combination of surgery followed by radiotherapy is highly recommended to control the local disease and limit distant metastasis.

REFERENCES

1. Hyam DM, Veness MJ, Morgan GJ. Minor salivary gland carcinoma involving the oral cavity ororopharynx. Aust Dent J, 2004;49:(1):16-19
2. Vicente OP, Marqués NA, Aytés LB, EscodaCG. Minor salivary gland tumors: A clinicopathological study of 18 cases. Med Oral P Patol Oral Cir Bucal 2008;13(9):E582-8.
3. Jansisyanont P, Blanchaert Jr RH, Ord RA. Intraoral minor salivary gland neoplasm: a single institution experience of 80 cases. Int. J. Oral Maxillofac. Surg. 2002; 31: 257–261
4. MacIntosh RB. Minor salivary gland tumors: types, incidence, and management. Oral Maxillofac Surg Clin N Am 1995;7:573-89
5. Chou C, Zhu G, Luo M, Xue G. Carcinoma of the Minor Salivary glands: Results of surgery and combined therapy. J Oral Maxillofac Surg 1996;54:448-53
6. Anderson JN, Beenken SW, Crowe R, Soong SJ,Peters G, Maddox WA, Urist MM. Prognostic factors in minor salivary gland cancer. Head Neck 1995;17:480-86
7. Brookstone MS, Huvos AG, Spiro RH. Central adenoid cystic carcinoma of the mandible. J Oral Maxillofac Surg 1990;48:1329-33
8. Mc Gregor IA, Mc Gregor FM. Salivary Tumors. Cancer of the face and mouth; Churchill Livingstone, Edinburgh 1986;569-606
9. Chaudhry AP, Leifer C, Cuter LS, Satchidanand S, Labay GR, Yamane GM. Histogenesis of adenoid cystic carcinoma of the salivary glands: Light and Electron microscopic study. Cancer 1986;58:72-82
10. Takahashi H, Fugita S, Tsuda N, Tezuka F, Okabe H. Intraoral minor salivary gland tumors: a demographic and histologic study of

200 cases. Tohoku J Exp Med 1990;161:111-28
11. Garden AS, Weber RS, Morrison WH, Ang KK, Peters LJ. The influence of positive margins and nerve invasion in adenoid cystic carcinoma of the head and neck treated with surgery and radiation. Int J Radiat Oncol Biol phys 1995;32(3):619-26
12. Regezi JA, Lloyd RV, Zarbo RJ, Mc Clatchey KD. Minor salivary gland tumors: A histologic and immunohistochemical study. Cancer 1985;55:108-15
13. Avery CME, Moody AB, McKinna FE, Taylor J, Henk JM, Langdon JD. Combined treatment of adenoid cystic carcinoma of the salivary glands. Int J Oral Maxillofac Surg 2000;29:277-79
14. Bechardt RN, Weber RS, Zane R, Garden AS, Wolf P, Carrrillo R, Luna MA. Minor salivary gland tumors of the palate: Clinical and pathologic correlates of outcome. Laryngoscope 1995;105(11):1155-60
15. Garden AS, Weber RS, Ang KK, Morrison WH, Matre J, Peters LJ. Postoperative radiation therapy for malignant tumors of minor salivary glands. Cancer 1994;73(10):2563-69
16. Parsons JT, Mendenhall WM, Stringer SP, Cassisi NJ, Million RR. Management of minor salivary gland carcinomas. Int J of Radiat Oncol Biophys 1996;55:443-54.

****