Case Report

AHB Advances in Human Biology

Adv Hum Biol. 2015;5(2):93-96.

Adenoid Cystic Carcinoma of Palate: A Case Report

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ABSTRACT

Background: Adenoid cystic carcinoma (ACC) comprises approximately 10% of all salivary gland tumors. It is the most common malignant tumor of sub-mandibular salivary gland and also of minor salivary glands. It is most commonly found in the area of the greater palatine foramen of the palate. One of the features of ACC is its slow growth rate & thus it is often present in the patient's mouth for several years before manifesting with symptoms. A detailed description of a case of ACC along with review of literature is presented here with emphasis to focus on the importance of detailed investigations, histo-pathological evaluation and post-operative rehabilitation.

Keywords: Adenoid cystic carcinoma, Salivary gland, Tumour, palatine.

INTRODUCTION

ACC is a rare malignant tumor that affects both major & minor salivary glands, ceruminous glands, the lacrimal glands and rarely the excretory glands of the female genital tract. Around 30-40% of these carcinomas occur in the head & neck region, of these, around 50 to 70% of the reported cases occur in minor salivary glands. ACC is known for its prolonged clinical course and its tendency for delayed onset of distant metastasis. ACC displays a distinct cribriform histo-morphology described previously as Swiss Cheese or Sieve like pattern, but this terminology has proved inadequate in expressing the wide spectrum of clinical diversity that this pathology presents. Current treatment recommendations for ACC include complete surgical resection and postoperative radiation therapy. High tumor recurrence rate of 42% also have been reported in the literature. The course of the disease is indolent and patients with ACC often survive for many years[1].

CASE REPORT

A 32 year old male patient reported to the Department of Oral and Maxillofacial Surgery with a chief complaint of painless swelling on the right side of the palate since one & a half years. Patient noticed the swelling six months after extraction of a decayed, mobile upper molar tooth. The swelling was painless and slow growing. There was no reduction in size of the swelling since the patient had noticed it and instead increased in size gradually to present size. Personal history was negative for tobacco consumption, alcohol and cigarettes smoking. There was no history of trismus, pus discharge, fever, malaise, paraesthesia, weight loss or loss of appetite. The medical, social and family histories were unremarkable. Systemic examination and physical examination revealed no abnormal findings and all the vital signs were in the normal limits.

Received: June. 19, 2014: **Accepted**: Oct. 3, 2014
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pISSN 2321-8568
eISSN 2348-4691

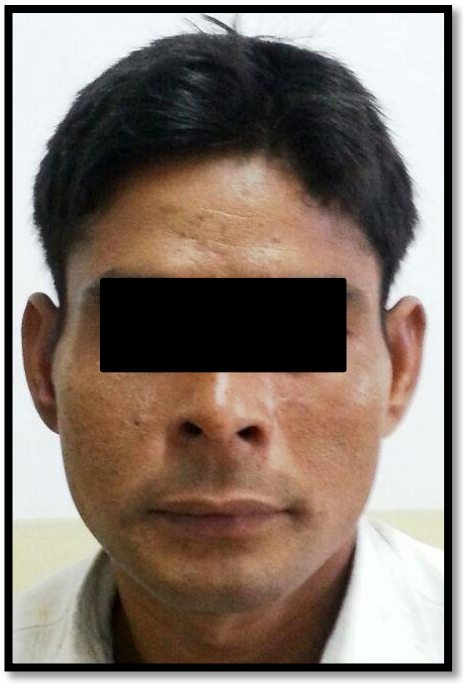

Fig 1: Extra-oral photograph showing patient's facial profile.

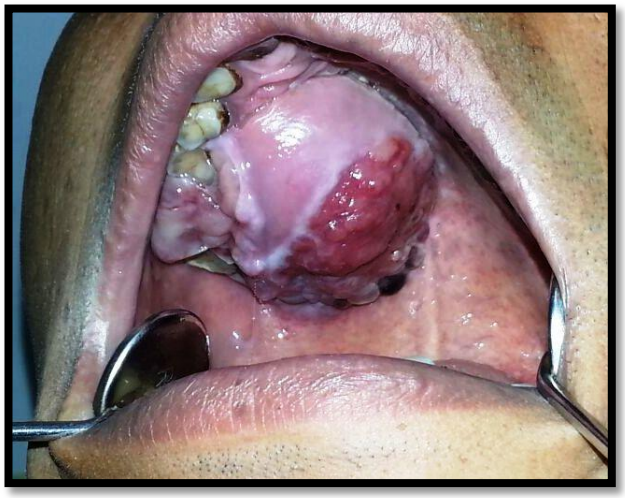

Fig 2: Intra-oral photograph showing swelling on the right side of the palate with tethering of overlying mucosa.

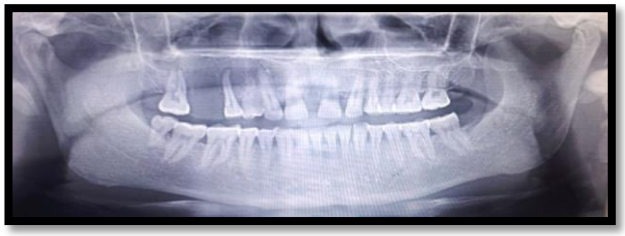

Fig 3: Pre-operative OPG radiograph.

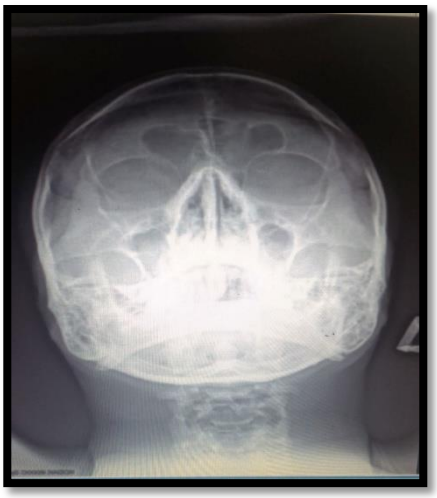

Fig 4: Pre-operative PNS radiograph.

CLINICAL EXAMINATION

On extra-oral examination no apparent abnormality was detected. Cervical lymph nodes were not palpable or tender bilaterally (Figure 1).

On intraoral examination, it (Figure 2) revealed a well-defined, solitary dome shaped swelling measuring 3x4cm. The swelling was extending from palatine rugae area to junction of hard and soft palate posteriorly & mediolaterally from the mid palatine raphe to the palatal aspect of 12,13,14,15,16,17 region. Tethering of overlying mucosa was noted with no discharge. Swelling was sessile, firm in consistency, non-fluctuant and non-compressible.

Mucoepidermoid carcinoma, Pleomorphic adenoma, Adeno cysic carcinoma, central giant cell lesions were considered. The patient underwent following investigations to reach to a probable diagnosis. Electric pulp vitality test of the involved teeth revealed that all the teeth in the vicinity were vital. A complete hemogram was also advised which showed all the values within normal range. Routine radiographic examinations, FNAC and incisional biopsy were done prior to surgical treatment.

RADIOGRAPHIC FEATURES

OPG & PNS view radiographs were not suggestive of any apparent pathology involving bone or dentition of the right side of maxilla (Figure 3 and 4).

Fine needle aspiration cytology was performed and serosangious fluid was aspirated. Smear revealed many clusters of eosinophilic globules and glandular epithelial cells along with various normal blood cells. Findings were suggestive of a benign salivary gland tumor.

HISTOPATHOLOGIC FEATURES

Incisional biopsy specimen revealed islands of hyperchromatic basiloid epithelial cells containing multiple cyst like spaces filled with hyalinized eosinophilic product forming cribriform & tubular structures. The tumor cells were found infiltrated into the normal appearing stroma. The above features were indicative of adenoid cystic carcinoma.

TREATMENT

Surgical excision of the lesion with clear margins through hemi-maxillectomy was performed and the defect was treated with immediate reconstruction using a temporary acrylic plate and split thickness skin graft. Definitive reconstruction after 6 weeks was done using permanent obturator. Patient has been followed up for the past 8 months with no signs of recurrence and satisfactory level of function and aesthetics and no neural deficit.

DISCUSSION

ACC is known to be a rare tumor of the head and neck region. However, it is the commonest malignant tumor of minor salivary glands[2] of the palate, as in the present case. Both men & women are equally affected by this tumor and it usually occurs in the fifth and sixth decade of life, but in the present case the patient was 32 years old. The common sites for its occurrence are the sub mandibular, parotid and the accessory glands in palate and tongue while the lesser common sites include cervix, esophagus, external auditory canal and middle ear. Rarely, it may also present as a primary intraosseous tumor of the mandible and maxilla[3].

Clinical presentation is often as a mass with no symptoms; however, this tumor can be present with pain or paresthesia and numbness[4]. It has a marked tendency to invade nerves & this can be seen in about 80% of all cases. Perineural invasion was not evident clinically in the present case. Facial nerve paralysis was found more frequently with Adenoid cystic carcinoma than with other tumors. Minor salivary gland involvement by a submucosal mass with or without pain and ulceration on the surface area can be found with this tumor. This is a tumor with two distinct clinical entities[5] where one group has a relentless fulminating course with early metastasis and fatal outcome within a short period of two to three years while the second group represents with an insidious natural history and long survival period despite recurrence of the tumor locally. Most of the cases fall in the second category and this is termed as the patient and the tumor both exist in symbiosis. Lymphatic spread from this tumor is found to be rare. Lymph nodes, however, in very extensive cases, may be involved by direct extension[6]. In long standing cases distant metastasis occurs via the blood stream most commonly to the lungs and bones.

Confirmatory diagnosis of Adenoid cystic carcinoma 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 its outcome. Three histological patterns of growth have been described. ACC histologically has a cribriform pattern in which nests and columns of cells of rather bland appearance are arranged concentrically around gland-like spaces which are filled with hyaline PAS positive material as in the present case. Some have a tubular pattern while a few others have a solid pattern. Radiological investigations, especially CT scans are important to diagnose the tumor, to plan extent of surgery and to look out for recurrences postoperatively.

Treatment of ACC includes surgical excision and may also involve postoperative radiation therapy[7,8]. Radiation therapy used alone has a high rate of local recurrence but may provide useful palliation in inoperable/disseminated disease[9]. Neutron radiotherapy has been studied in this disease with some authors suggesting a role for it[10]. Pulmonary metastasectomy for adenoid cystic carcinoma metastasis restricted to the lung does not seem to alter survival[11]. Chemotherapy using one or a combination of drugs (cyclophosphamide, 5-fluorouracil, mitomycin-C and cisplatin) has been used with some success and remission. However, definitive role of chemotherapy in the management of adenoid cystic carcinoma is yet to be

established[12]. The long natural history and indolent progression of adenoid cystic carcinoma make the status of therapeutic options less clear and "stationary disease" a poor criterion for treatment. The patient is on follow-up for the last eight months and has had minimal neural symptoms following surgery.

CONCLUSION

The primary treatment objective in Adenoid cystic carcinoma patients is control of the lesion, normal function and prevention of distant metastasis. For this purpose, early detection by the team of dental specialists is a pre-requisite, in order to enable a more favorable outcome of treatment and better quality of life. Various diagnostic modalities like biopsy, fine needle aspiration cytology & advanced diagnostic imaging techniques like Computed Tomography has been very useful for diagnosis of tumor as in the present case. The therapy which involves combination of surgery & radiotherapy remains the modality of choice in most cases.

CONFLICT OF INTEREST

No potential conflict of interest relevant to this article was reported.

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How to cite this article:

Gupta A, Gowri V, Sunil VSB, Shah G, Jain A, Thomas H. Adenoid Cystic Carcinoma of Palate: A Case Report. Adv Hum Biol. 2015;5(2):94-96.