Adenoid cystic carcinoma of palate

Dhaval N. Mehta, Shilpa J. Parikh[1]

Department of Oral Medicine and Radiology, Karnavati School of Dentistry, Uvarsad, Gandhinagar, Gujarat, [1]Department. of Oral Diagnosis and Radiology, G.D.C H, Ahmedabad, India

Address for correspondence:
Dr. Dhaval N. Mehta, Sunita Society, Beh. C. N. Vidhalaya, Bhudarpura Road, Ambawadi, Ahmedabad - 380 006, Gujarat, India. E-mail: drdhaval80@gmail.com

Abstract

Adenoid cystic carcinoma is a rare tumor arising from the minor salivary glands;, the palate being the commonest site. Distant metastasis and perineural invasion areis common in adenoid cystic carcinoma. Diagnosis of adenoid cystic carcinoma is made usually with the help of clinical features, radiographic features and histologic features. We reported a case of adenoid cystic carcinoma of palate involving left maxillary sinus. The diagnosis of the case and brief review of literature of adenoid cystic carcinoma is discussed. The aim here is to highlight the importance of diagnosis, treatment and long-term follow-up of the patients with adenoid cystic carcinoma.

Key words: *Adenoid cystic carcinoma, cylindroma, salivary gland tumor*

INTRODUCTION

Adenoid cystic carcinoma is a malignant neoplasm that may affect either the major or minor salivary glands of the oral cavity.[1,2] Adenoid cystic carcinoma was originally described by Lorain and Laboulbene in 1853. In 1859, Billroth suggested the name cylindroma. In 1930, Spies suggested the term adenoid cystic carcinoma to replace cylindroma and this name has been widely accepted. Until 1940s, the tumor was thought to be a benign variant of the mixed salivary gland tumor. In 1943, Dockerty and Mayo emphasized the malignant nature of this

tumor.[1,3,4] Adenoid cystic carcinoma is a rare tumor of head and neck region. It accounts for <1% of all head and neck malignancies and about 4-10% of all salivary gland tumors. However, it is the commonest malignant tumor of the minor salivary glands. [1,3-7]

CASE REPORT

A 60-years-old female patient had a chief complaint of left intra-oral swelling since $2^1/_2$ months. The patient gave a history of exfoliation of maxillary left first molar before $2^1/_2$ months. Patient She had a habit of snuff inhalation 3-4 times/a day since 13-14 years.

Intra-orally, 3 × X 3 cm sized firm and tender swelling was present, which extended from maxillary left first premolar to maxillary left third molar region obliterating maxillary left buccal vestibule and palatally extending in the same region without crossing the midline, involving both soft and hard palate with normal overlying mucosa. Mobility of the involved teeth [maxillary left first and second premolar-Grade I, maxillary left second and third molar- Grade III] were present without any intra-oral sinus [Figure 1]. Left submandibular lymph nodes were enlarged, palpable, mobile and tender. OPG showed irregular radiolucency extending from maxillary left first premolar to maxillary left third molar with poorly defined border giving floating teeth appearance and angular (apical) root resorption of maxillary left first and second premolar, maxillary left second molar (mesial root). It also showed involvement of the left maxillary sinus [Figure 2]. PNS view showed homogenous haziness in left maxillary sinus without erosion/or destruction of lateral walls of maxillary sinus [Figure 3]. On the basis of clinical and radiographic findings, we thought of ameloblastoma converting into squamous cell carcinoma, squamous cell carcinoma of left maxillary sinus/or mixed tumor of minor salivary glands.

For further investigations, punch biopsy was taken

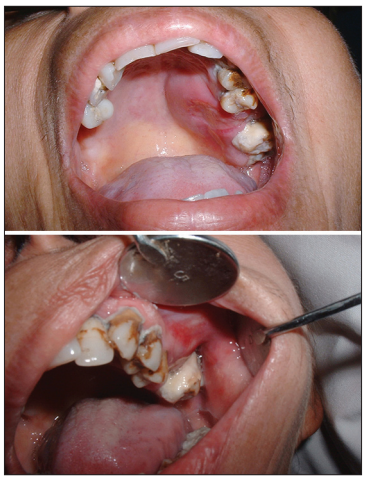

Figure 1: Intra-oral swelling of hard palate with the site of biopsy and obliteration of left upper buccal vestibule

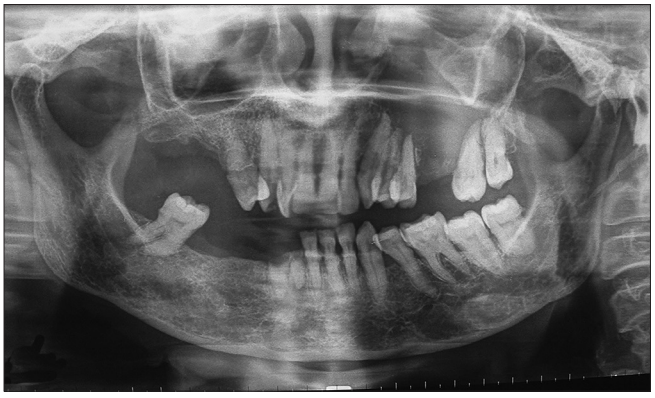

Figure 2: OPG showing radiolucency with poorly defined borders, floating teeth appearance of maxillary left first and second premolar, maxillary left second molar and involving left maxillary sinus

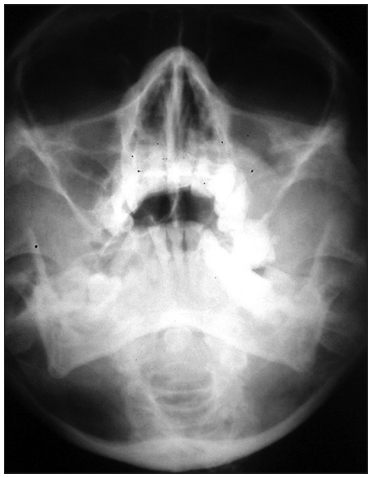

Figure 3: Haziness in left maxillary sinus without erosion/or destruction of lateral walls of maxillary sinus in PNS

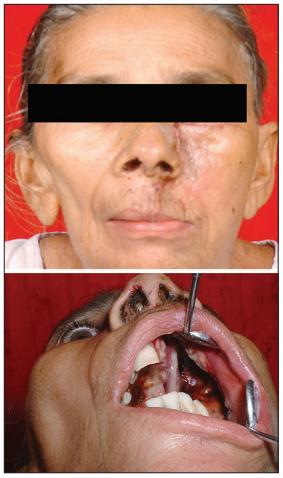

Figure 4: Post-surgical scar on the left side of face with intraoral post-surgical defect

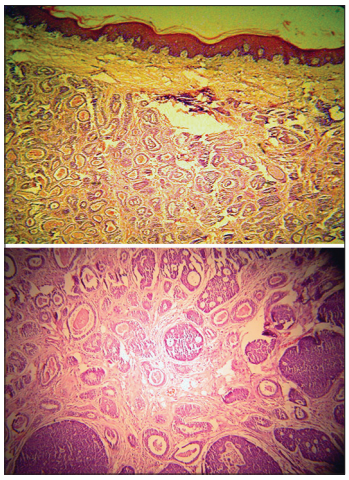

Figure 5: Photomicrograph showing Swiss-cheese pattern suggestive of adenoid cystic carcinoma

from the palate. Histologic features were suggestive of pleomorphic adenoma of minor salivary gland. Patient was examined preoperatively. No evidence of spread or distant metastasis was found. After surgical removal of tumor with partial maxillectomy and resection of left side of hard palate, preoperatively prepared obturator was adjusted to cover the defect [Figure 4]. Histologically, excisional biopsy showed the features of adenoid cystic carcinoma of palate without lymphovascular invasion [Figure 5]. No metastasic nodes were seen. Patient was advised for post-surgical radiotherapy. Patient was given total radiotherapy dose of 60 Gy in 30 fractions (200 cGy/fraction for 6 weeks). Patient was advised for follow-up every 6 months.

DISCUSSION

Adenoid cystic carcinoma is a rare tumor of head and neck region. Palate is the most common site of intra-oral lesion. It is most commonly found in the 5th to 6th decade of life with no specific sex predilection. In the major salivary glands, adenoid cystic carcinoma most commonly affects the parotid and submandibular glands, while in the minor salivary glands, palate is the most common site; other sites are floor of mouth, tongue and lip. Rarely, it may also present as primary intraosseous tumors of the maxilla and mandible. Most frequent clinical feature of adenoid cystic carcinoma affecting major salivary gland is reported to be the presence of tumor-usually 2-4 cm at its greatest diameter and intraoral adenoid cystic carcinoma seldom larger than 3 cm at its greatest diameter.[1,2,4,8] In the presented case, a female patient of 60 years of age had adenoid cystic carcinoma, which affected the minor salivary glands of her palate was reported. The lesion is uncapsulated and infiltrative; invasion of underlying bone is common. Incidence of cervical metastasis is low. Distant metastasis occurs through blood stream to lung and bones. Direct extension of lesion of the base of skull has been reported as a cause of death.[3,4] Three histologic patterns of growth have been described by some authors.[1,3,5,7,8] (1) Cribriform pattern: "Honeycomb" or "Swiss-cheese" pattern. Stromal connective tissue becomes hyalinized and surrounds the tumor cells, forming a structural pattern of cylinders from which the lesion derived the name cylindroma. (2) Tubular pattern: Tthe cribriform type has a very aggressive course compared too tubular form. (3) Solid pattern: Having worst prognosis. Presented case draws the attention that the marked pleomorphism of the lesion display histologically may be the most striking and confusing feature commonly seen in pleomorphic adenoma arising in minor salivary glands. In the presented case, punch biopsy was suggestive of pleomorphic adenoma and excisional biopsy showed the classical features of adenoid cystic carcinoma. This may be because of lack of availability of deep tissues of the lesion which had characteristic features of adenoid cystic carcinoma.[9] The choice of treatment offor Aadenoid cystic carcinoma is surgical resection with post-surgical radiotherapy (not less than 60 Gy), which enhances the local and regional control in adenoid cystic carcinoma. The role of chemotherapy for adenoid cystic carcinoma is still controversial.[7,10] For the presented case, partial maxillectomy was performed and post-operative radiotherapy [60 Gy (200 cGy/fraction – 30 fractions in 6 weeks)] was given to the patient. Adenoid cystic carcinoma of the minor salivary gland has a better prognosis than of major salivary glands because intra-oral lesions are diagnosed and treated earlier and therefore less likely to have advanced neural involvement and metastasis to regional or distant sites and the prognosis of adenoid cystic carcinoma of palate is reported to be better than the lesion located anywhere else.

SUMMARAY AND CONCLUSION

Salivary gland tumors should be considered in the differential diagnosis of aggressive lesions in maxilla and mandible and especially when the aggressive lesion is involvinginvolves palate, adenoid cystic carcinoma involving minor salivary gland tumors should be considered in differential diagnosis. Confirmatory diagnosis should be made with excisional biopsy having deeper tissues.

REFERENCES

1. Mader C, Gay WD. Intraoraladenoid cystic carcinoma. J Am Dent Assoc 1979;99:212-4.

2. Tarpley TM Jr, Giansanti JS. Adenoidcystic carcinoma-analysis of fifty oral cases. Oral Surg Oral Med Oral Pathol 1976;41:484-9.
3. Clark JM, Triana RJ, Meredith SD. Uncontrolled central adenoid cystic carcinoma: Case report. Ear Nose Throat J 2000;79:785-6.
4. Dutta NN, Baruah R, Das L. Adenoidcystic carcinoma-clinical presentation and cytological diagnosis. Indian J Otolaryngol Head Neck Surg 2002;54:62-5.
5. Pandey R, Singh PK, Khan L. Adenoid cystic carcinoma: A rare presentation as a nasal and hard palate mass. J Cytol 2008;25:159-60.
6. Cleveland D, Abrams AM, Melrose RJ, Handlers JP. Solid Adenoid cystic carcinoma of the maxilla. Oral Surg Oral Med Oral Pathol 1990;69:470-8.
7. Eneroth CM, Hjertman L, Moberger G. Adenoidcystic carcinoma of the palate. Acta Oto-laryngologica 1968;66:248-60.
8. Gondivkar SM, Gadbail AR, Chole R, Parikh RV. Adenoid cystic carcinoma: A rare clinical entity and literature review. Oral Oncol 2011;47:231-6.
9. Adkins KF, Campbell AF. Adenoid cystic carcinoma of the palate. Oral Surg Oral Med Oral Pathol 1970;30:734-41.
10. Martínez-Rodríguez N, Leco-Berrocal I, Rubio-Alonso L, Arias-Irimia O, Martínez-González JM. Epidemiology and treatment of adenoid cystic carcinoma of the minor salivary glands: A meta-analytic study. Med Oral Patol Oral Cir Bucal 2011;1:16:e884-9.

How to cite this article: Mehta DN, Parikh SJ. Adenoid cystic carcinoma of palate. J Nat Sc Biol Med 2013;4:249-52.

Source of Support: Nil. **Conflict of Interest:** None declared.

Access this article online

Quick Response Code:	Website: www.jnsbm.org
	DOI: 10.4103/0976-9668.107319