International Journal of Oral & Maxillofacial Pathology. 2013;4(1):60-63
Available online at http://www.journalgateway.com or www.ijomp.org
ISSN 2231 – 2250

Case Report

Salivary Duct Carcinoma of Minor Salivary Gland

Nishat Shah, Sandip Kulkarni, Rajeev Desai

Abstract

Salivary duct carcinoma is a rare invasive malignancy arising in the ductal epithelium of the salivary glands. These are uncommon but distinct group of highly malignant salivary gland tumours, usually seen in the major salivary glands, especially the parotid gland. Pathomorphologically, these tumours show great similarities to ductal carcinomas of the female breast, which is why it has been described as "salivary duct carcinoma." Salivary duct carcinomas affecting the minor salivary glands have been reported in only four percent of the salivary duct carcinoma cases and constitute two percent of all the salivary gland malignant neoplasms. Although neoplasms of this type are infrequent in minor salivary glands, they are less aggressive and may lead to early diagnosis before distant metastases could occur. The present article reports a case of salivary duct carcinoma of the palate in a 45-year-old male patient with aggressive clinical and histopathological behaviour.

Keywords: Salivary Duct Carcinoma, Malignancy; Salivary Glands; Ductal Epithelium; Palate.

Nishat Shah, Sandip Kulkarni, Rajeev Desai. Salivary Duct Carcinoma of Minor Salivary Gland. International Journal of Oral & Maxillofacial Pathology; 2013:4(1):60-63.

Received on: 06/03/2013 Accepted on: 22/04/2013

Introduction

Salivary duct carcinoma (SDC) is a rare tumor accounting for 1% to 3% of all malignant salivary gland tumors.[1] It is an aggressive adenocarcinoma, resembling a high-grade mammary duct carcinoma. Salivary duct carcinoma occurs predominantly in males, with a M:F ratio of 2:1. The age range of patients is from 23-80 years, with more cases being reported in the fifth and sixth decades of life. Neoplasms of this type are infrequent in minor salivary glands and have been reported in only 4% of the SDC cases. Here we present a case of salivary duct carcinoma of the palate in a 45-year-old male patient with aggressive clinical and histopathological behaviour.

Case Report

A 45-year-old male patient presented with a chief complaint of swelling in the roof of his mouth on the left side for the past 2.5 years. The swelling started as a small nodule and gradually progressed to the present size. It was painless initially and patient developed pain only in the last few weeks. Patient had undergone extraction of teeth from maxillary left quadrant because of mobility. Nasal obstruction and discharge was absent. There was no history of smoking, tobacco chewing and alcohol.. His routine blood investigations were within normal. Extraoral examination revealed a diffuse swelling of left side of face measuring 3 x 3cm in size and obliterating the nasolabial fold. The swelling extended from infraorbital margin to the corner of mouth and from ala of nose to the malar eminence mediolateraly. Overlying skin appeared normal and on palpation the swelling was bony hard with mild tenderness (Fig 1a).

On intraoral examination a diffuse swelling was seen extending from midline to left maxillary second molar. Buccal cortical plate was expanded with obliteration of buccal vestibule. Palatally the swelling extended from gingival margin to the midpalatine raphe. The overlying mucosa appeared stretched with few engorged blood vessels (Fig 1b). Computed tomography (CT) scan showed an expansile destructive lesion with a huge heterogeneously enhancing soft tissue component in alveolar region, hard palate and left maxillary sinus. Destruction and demineralisation of maxillary alveolar ridge, hard palate, left maxillary sinus, floor of the orbit, anterior part of nasal septum and nasal turbinates was seen. Loss of underlying teeth and few calcifications were noted (Fig c & d). No lymph node involvement was noted on CT scan. The tumor extension was T3N0M0 with the tumor in stage III as per TNM classification.[2] Based on these findings a provisional diagnosis of salivary gland malignancy was made.

The histopathological features revealed numerous infiltrating islands and cords of

neoplastic glandular epithelium in fibro vascular connective tissue (Fig 1e). Intraductal cribriform growth patterns were seen in abundance with central "comedonecrosis" (Fig 1f). The neoplastic cells were cuboidal to polygonal in shape and exhibited cellular pleomorphism, eosinophilic cytoplasm, nuclear hyperchromatism and prominent nucleoli. A definitive diagnosis of salivary duct carcinoma was then made. The lesion was surgically excised completely followed by radiotherapy of 60 Gy in 30 fractions on the primary site. Local recurrence or metastasis was not reported within the 5 years of follow-up period performed by taking chest x-ray at regular intervals. Patient was advised to maintain long term follow up.

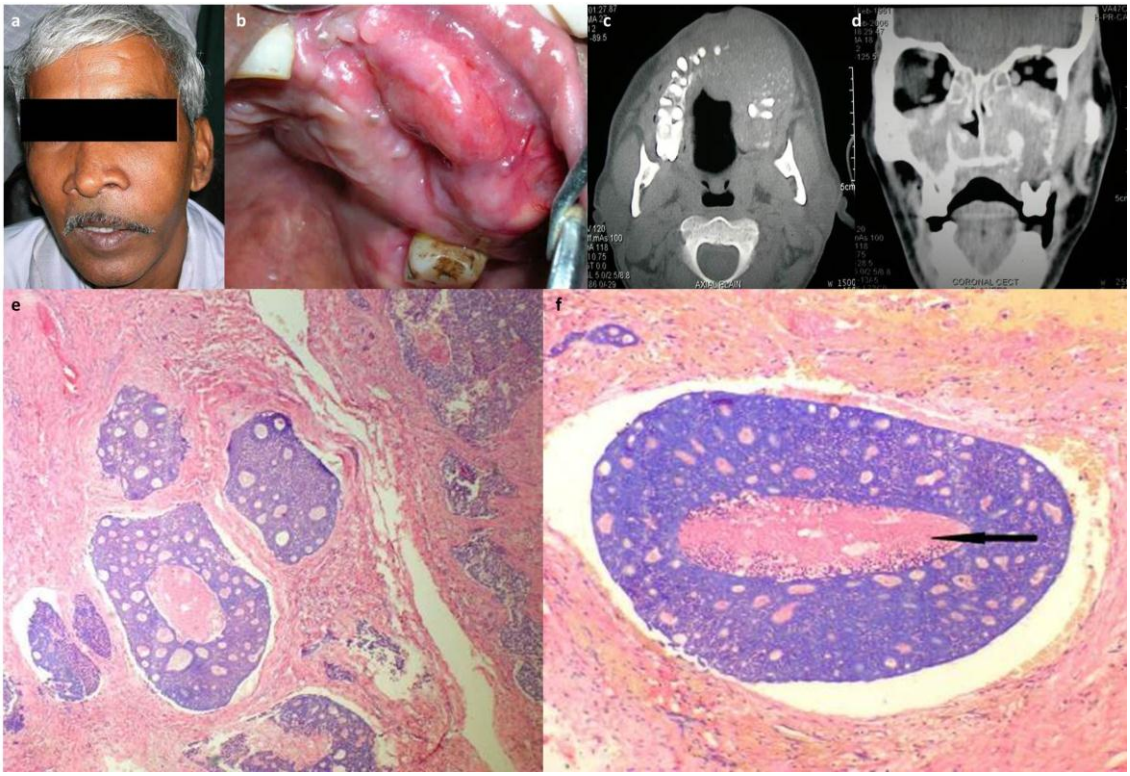

Figure 1: The clinical photograph showing solitary diffuse extraoral swelling on left side of face (a) and Intraorally showing expansion buccal as well as palatal cortical plates with obliteration of the buccal vestibule (b). The computed tomography scan of axial view showing expansile lesion in left maxilla along with loss of underlying teeth (c) and coronal view of the same showing destruction of left maxillary alveolar ridge, maxillary sinus, nasal septum, nasal turbinate and orbital floor (d). The photomicrograph showing intraductal cribriform growth pattern in fibrovascular stroma at 10X magnification (e) and island of neoplastic epithelium with central comedonecrosis at 45X (f).

Discussion

SDC was first described by Kleinsasser in 1968[3] and was further defined by several authors. It was only recently recognized as a distinct clinic-pathological entity. Pathomorphologically, these tumors showed great similarities to ductal carcinoma of the female breast, which is why they described this tumor as "salivary duct carcinoma." However, the designation, 'salivary duct carcinoma' has gained acceptance because it is used in the WHO classification of tumours.[4]

It represents a rare tumor with an estimated incidence of 1% to 3% of all malignant salivary gland tumors.[1] Salivary duct carcinoma accounts for 0.9% to 6% of all parotid tumors.[5] Rarely, submandibular glands and minor salivary glands are concerned. Intra-orally, the common sites of occurrence are palate, followed by buccal mucosa /vestibule, upper lip, and maxilla / mandible.[6] SDC occurs predominantly in males (M:F = 2:1), with more cases being reported in the fifth and sixth decades of life. It presents as a rapidly growing mass, which develops aggressively with possibilities of early distant metastases, local recurrence, and high mortality. Sometimes it may develop in pre-existing pleomorphic adenoma, but it can also occurs de novo. Lymph node metastases have been reported in 22% of the SDC cases in the minor

ISSN 2231 - 2250

salivary glands, as compared to 83% in the SDC of the major salivary glands.[7] Imaging findings, especially CT scan and Magnetic resonance imaging (MRI) are non-specific but they can indicate the malignant nature of the tumor by showing ill defined borders or an infiltration of the adjacent tissue. Positive diagnosis is based on histologic examination.

Macroscopically, the aspect is of a well-contoured yellowish or grayish-white tumor that may be nodular, multinodular, cystic or infiltrating, surrounded by fibrosis with haemorrhagic areas, necrosis, cystic degeneration and intratumoral calcification.[8] SDC exhibits a wide range of histological appearances.[9]The tumor is composed of an intraductal and invasive components. Intraductal component is cribriform, papillary, and solid with comedo-like central necrosis. Tumour cells are polygonal in shape, with granular eosinophilic cytoplasm, enlarged hyperchromatic, pleomorphic nuclei and prominent nucleoli. 'Comedonecrosis' means a type of necrosis occurring in the glands, in which there is central luminal inflammation with devitalized cells, which usually occurs in the breast in intraductal carcinoma. The infiltrative component is made of glands, cords of cells with desmoplastic reaction. Vascular invasion and perineural infiltration have been reported in some cases. Atypical mitotic figures are seen in most of the lesions. Dystrophic calcifications are seen in some cases. In present case calcification were seen on CT scan.

Delgado et al,[10] suggested that cribriform growth pattern and comedonecrosis are more frequently encountered in high-grade than in low-grade SDC. Progression is evidenced by the acquisition of higher cytologic grades such as, monomorphous cellular composition, well defined cytoplasmic borders and rigid cellular arrangement. In our case, cribriform growth pattern and comedonecrosis were prominent and also higher cytologic grades were observed. Differential diagnosis includes mucoepidermoid carcinoma, adenocarcinoma NOS, metastatic adenocarcinoma, oncocytic carcinoma, papillary cyst adenocarcinoma, papillary cyst acinic cell carcinoma and the most relevant morphologic feature is the presence of an "intraductal" component which is specific of the diagnosis.[11]

The treatment of this lesion involves the radical surgical excision of the lesional tissue and its associated structures, with concomitant neck dissection, followed by post-operative radiation therapy. For patients with minimal residual disease after surgery a dose of 60-66 Gy photons in daily fractions of 2 Gy over 6 weeks is advisable. Patients with postsurgical macroscopic disease with unresectable primary tumours or with inoperable recurrent tumours should be given doses of 60 Gy photons. An additional dose of 10 Gy is usually given through reduced portals to the volume of known residual disease. In these selected patients an optional mixed-beam therapy consisting of photons and a neutron boost can be applied. Irradiation of the adjacent neck lymph nodes should be given with 50-60 Gy photons if there is tumour involvement. After a neck dissection, irradiation of the neck is optional.[12-14]

Conclusion

The present report is a rare case of salivary duct carcinoma originating in minor salivary glands with aggressive clinical behaviour and high grade carcinoma. A long term follow up is essential to rule out regional and distant metastasis.

Acknowledgement

We would like to thank all the staff members for their support.

Author Affiliations

1.Dr.Nishat Shah, Reader, Department of Oral Pathology, MIDSR Dental College, Latur, 2.Dr.Sandip Kulkarni, Reader, Department of Oral Pathology, Terna Dental College, Nerul, Navi Mumbai, 3.Dr.Rajeev Desai, Professor, Maharashtra University of Health Sciences, Department of Oral Pathology, Nair Hospital Dental College, Mumbai Central, India.

References

1. Seifert G, Caselitz J. Epithelial salivary gland tumors: Tumor markers. In: Fenoglio-Preiser CM, Wolff M, Rilke F, editors. Progress in surgical pathology. Vol. 9. New York: Field and Wood; 1989. 157-87p.
2. UICC. UICC (International Union against Cancer). TNM classification of malignant tumours. Sobin LH, Wittekind Ch, editors. 6th ed. New York, Chichester, Weinheim, Brisbane, Singapore, Toronto: Wiley-Liss. 2002 [Medline]
3. Kleinsasser O, Klein HJ, Hubner G. Salivary duct carcinoma. A group of salivary gland tumors analogous to mammary duct carcinoma. Arch Klin Exp

Ohren Nasen Kehlkopfheilkd 1968;192:100-5.
4. Skalova A, Nagao T. Salivary duct carcinoma. World Health Organization Classification of tumours, Pathology and genetics of head and neck tumours. In: Barnes L, Eveson JW, Sidransky D, editors. Lyon: IARCC Press; 2003. p. 236-8.
5. Kumar NS, Ranganathan K, Nathan JA, Saraswathi TR. Salivary duct carcinoma of minor salivary gland. J Oral Maxillofac Pathol 2004;8:87-90.
6. Lewis JE, Mc Kinney BC, Weiland LH, Ferreiro JA, Olsen KD. Salivary duct carcinoma: Clinicopathologic and immunohistochemical review of 26 cases. Cancer 1996;77:223–30.
7. BenJelloun H, Maazouzi A, Benchakroun N. et al. Salivary duct carcinoma: report of two cases and literature review. Cancer Radiother 2004;8:383–6.
8. Hosal AS, Fan C, Barnes L, et al. Salivary duct carcinoma. Otolaryngol Head Neck Surg 2003;129:720–25.
9. Delgado R, Vuitch F, Albores-Saavedra J. Salivary duct carcinoma. Cancer 1993;72:1503-12.
10. Delgado R, Klimstra D, Saavedra AJ. Low grade salivary duct carcinoma: A distinct variant with low grade histology and a predominant intraductal growth pattern. Cancer 1996;78:958-67.
11. Anderson C, Muller R, Piorkowski R, Knibbs DR, Vignoti P. Intraductal carcinoma of major salivary gland. Cancer 1992;69:609-14.
12. Borthne A, Kjellevold K, Kaalhus O, Vermund H. Salivary gland malignant neoplasms: treatment and prognosis. Int J Radiat Oncol Biol Phys 1986;12:747-54.
13. Elkon D, Colman M, Hendrickson FR. Radiation therapy in the treatment of malignant salivary gland tumors. Cancer 1978;41:502-6.
14. Harrison LB, Armstrong JG, Spiro RH, Fass DE, Strong EW. Postoperative radiation therapy for major salivary gland malignancies. J Surg Oncol 1990;45:52-5.

Corresponding Author
Dr. Nishat Shah,
Flat no.104, Building E1,
Madhuban Co-op Housing Society,
Kalas Road, Vishrantwadi,
Pune – 15, Maharashtra, India.
Ph: +91 9922440971
Email:shah_nishat_n@yahoo.com

Source of Support: Nil, Conflict of Interest: None Declared.