

Intraosseous acinic cell carcinoma

V Hiremath, N Mishra¹, SG Patil²

Departments of Oral Pathology and ¹Oral Medicine and Radiology, Mansarovar Dental College, Bhopal, ² Department Of Oral and Maxillofacial Surgery, H.K.E' S.N. Dental College, Gulberga, Maharashtra, India

Abstract

Acinic cell carcinoma is an uncommon low-grade malignant tumor of salivary glands. It was first described by Nasse in 1892, arising in parotid salivary gland. Salivary gland tumors are also known to develop within jaw bones, arising within the jaw as a primary central lesion, and are extremely rare with only a few cases reported.

We present a rare case report of 65-year-old woman with intraosseous acinic cell carcinoma of left side of the mandible.

Key words: Acinic cell carcinoma, central tumor of mandible, intraosseous acinic cell carcinoma

Date of Acceptance: 17-Dec-2011

Introduction

Acinic cell carcinoma is an unusual type of salivary gland tumor, it accounts for between 2% and 4% of all tumors of the parotid gland. In 99% of the cases, parotid gland is the primary site for acinic cell carcinoma. However, acinic cell carcinomas have been described in the mandible, accessory parotid gland, maxillary sinus, lacrimal glands, palate, buccal mucosa, and tongue. Most cases are unilateral; bilaterality is rare.^[1] Acinic cell carcinoma may be found in all age groups, including children, with peak incidence noted within fifth and sixth decade of life.^[2] Acinic cell carcinomas usually present as slow-growing lesions less than 3 cm in diameter, with pain being a frequent presenting symptom.^[3] Histologically, acinic cell carcinoma is frequently surrounded by a thin capsule, which may be composed of cells with varying degrees of differentiation; well-differentiated cells bear remarkable resemblance to normal acinar cells whereas less differentiated cells resemble embryonic ducts and immature acinar cells.^[2]

Treatment of acinic cell carcinoma in most cases is surgical. High recurrence rates are seen in tumors treated by enucleation and limited excision. Long-term follow-up is necessary.^[2]

Case Report

A 65-year-old female patient reported to the Department of Oral Medicine and Radiology, Mansarovar Dental College,

Kolar Road (Bhopal, Madhya Pradesh), India, with swelling of the left side of face of 6 months' duration. She reported pain in lower left quadrant of her dentition for the past 5 months. The swelling initially was of a small size and gradually increased up to the present size. Pain is dull and continuous, aggravated on eating food, and subsides on its own after some time. Extra-oral examination revealed a solitary, diffuse swelling present on the left side of face, measuring approximately 5 cm x 3.5 cm, the overlying skin was normal in appearance, swelling was extending superiorly from infra-orbital region and inferiorly upto lower border of mandible. Anteriorly, the swelling was from nasolabial fold and posteriorly up to tragus of ear. [Figure 1] On palpation, findings of inspection were confirmed. It was tender on palpation, firm to hard in consistency; there was no enlargement of submandibular lymph nodes. Upon intra-oral examination, inspection revealed solitary swelling in the buccal mucosa in relation to left lower back tooth region, measuring approximately 3 cm x 2 cm. Color of the overlying mucosa was similar to adjacent mucosa. Vestibular obliteration was seen in relation. [Figure 2] Upon intra-oral palpation, swelling was tender and firm in consistency. Buccal cortical plate expansion was present in relation to left lower back tooth region. Based on history and clinical findings, a provisional diagnosis of benign odontogenic tumor was given.

Address for correspondence:

Dr. Nishi Mishra,
Department of Radiology, Mansarovar Dental College, Bhopal, India.
E-mail: drnishi_mishra@rediffmail.com

Access this article online

Quick Response Code:



Website: www.njcponline.com

DOI: 10.4103/1119-3077.113470

PMID:

Investigations were carried out; Hematological findings were within normal limits. Panoramic radiograph [Figure 3] showed a solitary unilocular radiolucency, measuring approximately 3.5 cm x 2.5 cm, involving the left ramus and body of the mandible. Radiolucency was extending anteriorly from anterior border of ramus of the mandible posteriorly up to posterior border of mandible and superiorly involving entire height of ramus of the mandible. The lower border of mandible was thinned out and was discontinuous, suggesting a fracture. Teeth 36, 37, and 38 were missing. The patient was subjected to computed tomographic examination [Figure 4], which revealed a well-circumscribed soft tissue mass (hypodense lesion) measuring 46 mm x 42 mm x 51 mm, with an epicenter at the left angle of mandible, involving the entire ramus of mandible with extension into posterior part of body of mandible. Cortex on medial and lateral aspects of ramus appeared to be destroyed due to the mass. The above features suggested the possibility of malignancy with a differential diagnosis of a benign odontogenic tumor such as ameloblastoma. However, clinico-pathologic correlation was required.

Further, an intra-oral incisional biopsy was done. Histopathological examinations were carried out, (Photomicrograph [H and E] staining) [Figures 5] shows tumor mass of salivary gland. These tumor cells were forming ducts and follicles lined by cuboidal to low columnar epithelial cells. Some of the follicles were filled with eosinophilic mucinous secretion. Foci of hemorrhage were also seen. The above histopathological findings were suggestive of acinic cell carcinoma of left side of mandible.

Discussion

Acinic cell carcinoma is also known as acinic cell or serous cell adenoma or adenocarcinoma.^[2] Accessory salivary gland tissue is found in many sites throughout the head and neck region, including centrally within the maxilla and mandible. Neoplastic transformation of the salivary gland tissues in particular is a known albeit uncommon phenomenon. Mandible is the most common site for intraosseous salivary neoplasia, accounting for 75% of all cases.^[4] Two hypotheses for intraosseous



Figure 1: Swelling on left side of face



Figure 2: Intraoral swelling



Figure 3: Radiolucency in the OPG

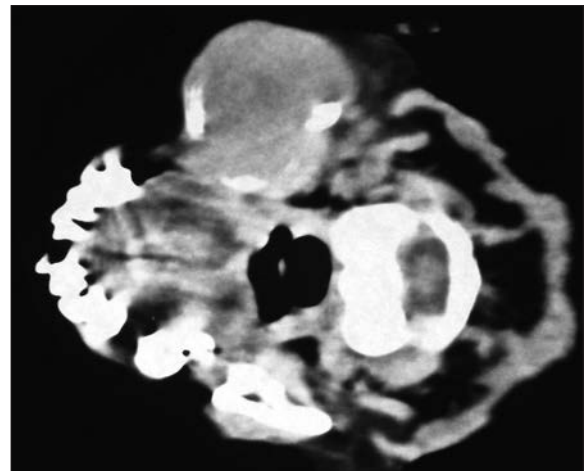


Figure 4: Computed tomography findings

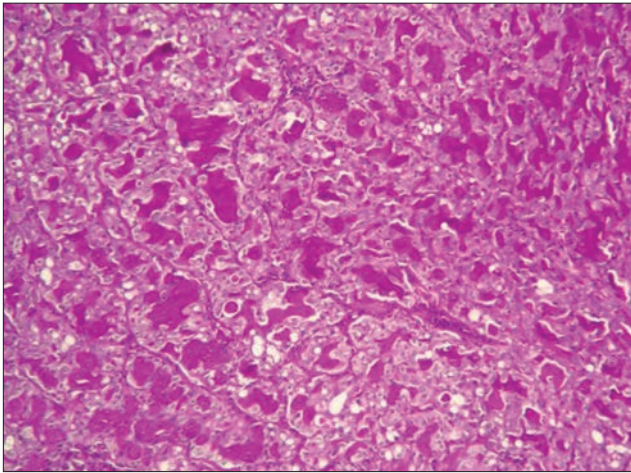


Figure 5: Photomicrograph with pas stain

glandular neoplasms can be proposed: one suggests that the salivary tissue may developmentally be entrapped within the jaws and the other suggests metaplasia of the epithelial lining of odontogenic cysts.^[5] In the present case, salivary tissue was entrapped in the mandible. Joseph A. Regzi^[3] documented that acinic cell carcinoma may be found in all age groups, including children, with peak incidence noted within fifth and sixth decade of life and that slow-growing swelling and pain are the presenting symptom, as in the present case where the patient was 65 years' old with complaint of swelling and pain. Nakazawa M *et al.*^[6] reported a case acinic cell carcinoma of mandible in female patient as in the present case where the patient was a female and mandible was involved. Hara I *et al.*^[7] reported a central acinic cell carcinoma of mandible in a 67-year-old woman, radiographic examination revealed a radiolucency from 2nd to 3rd molar of left mandible, preliminary diagnosis was odontogenic tumor, and histopathological examination disclosed diagnosis of acinic cell carcinoma. Similarly, a

provisional diagnosis of benign odontogenic tumor was considered in the present case, but the histopathological examination revealed diagnosis of acinic cell carcinoma.

Histopathologically, large lobules or nests of tumor cells with little intervening stroma are characteristic. The arrangement of neoplastic cells is quite variable. Generally, cells are arranged in solid masses with blunted or pushing margins. Other variations include microcystic, papillary cystic, and follicular forms.^[3] Surgery is the preferred treatment. They seldom metastasize, yet they have a strong tendency to recur.^[2,3]

We conclude that despite the rarity of intraosseous acinic cell carcinoma, the dental surgeon should be aware of this diagnostic possibility, emphasizing the need for histopathological analysis, defined and guided treatment, and adequate follow-up.

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How to cite this article: Hiremath V, Mishra N, Patil SG. Intraosseous acinic cell carcinoma. *Niger J Clin Pract* 2013;16:386-8.

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