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ADENOID CYSTIC CARCINOMA OF MANDIBLE MANIFESTING AS POST EXTRACTION LESION

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ABSTRACT

Aim: To present a case of adenoid cystic carcinoma of mandible manifesting as post extraction lesion.

Summary: A 56 years old female patient came with chief complaint of pain and cheek bite in right posterior teeth region of mandible since last 2 Months. She has undergone extraction of lower right 3rd molar before 1 year. Intra oral examination reveals a soft fibrous growth distal to right lower 2nd molar. A radiograph shows radiolucency distal and apical to distal root of right lower 2nd molar. A soft tissue at the site of extraction was excised and sent for histo-pathological report , which reveals adenoid cystic carcinoma.

Key learning points:

• Adenoid cystic carcinoma affecting the mandible may mimic a periapical lesion. Proper diagnosis of such a lesion is dependent on thorough clinical, radiographic and microscopic examinations.

• Such a case highlights the benefits of biopsy and histological examination of collected tissues.

• Diagnosis of lesions in the mandible should include salivary gland tumours.

INTRODUCTION

Adenoid cystic carcinoma (ACC) represents 7.5% of all carcinomas and 4% of both benign and malignant epithelial salivary gland tumors.ACC, a malignant epithelial neoplasm of salivary gland origin, was originally described by Robin & Laboulbene in 1853 (Tauxe et al. 1962). It most commonly affects adults,with a peak incidence in the 4th to 6th decades. The most commonly involved sites are the parotid gland,submandibular gland and palate, whereas the lower lip,retromolar-tonsillar pillar region and sublingual gland are affected less frequently.[1] very rarely,ACC may arise centrally within the jawbones, usually in the posterior mandible of adults, causing pain due to perineural invasion.[2] The present paper reports a case of primary intraosseous ACC of mandible without metastasis in 52 years old female patient.

CASE REPORT

A female patient aged 56 years presented at our clinic at gandhidham,Gujarat, with chief complaint of *mild* pain and cheek bite in lower right back teeth region since last 2 Months.

Patient was relatively asymptomatic before 2 Months. Then she started feeling pain in posterior mandibular teeth region which was dull, aching and intermittent in nature which subsides after taking analgesics(Tab.combiflame). Before 4 days she has bitten on her cheek which again aggravate her pain.she has under gone extraction of lower right 3rd molar before 1 year due to pain. Not significant medical history, absence of lymphadenopathy and no history of use of any kind of tobacco and alcohol.

Intraoral examination revealed a soft tissue growth distal to 2nd molar (figure 1)which was soft in consistency, fluctuant , and not attached to underlying bone having size of 1 cm in diameter. Missing lower right 3rd molar and tender on percussion 2nd molar. Panaromic radiography revealed multilocular radiolucent lesion distal and apical to 2nd molar.(figure 2)

Our provisional diagnosis was irritational fibroma distal to 2nd molar and periapical abscess in relation to(i.r.t)2nd molar.

Soft tissue was surgically excised,right lower 2nd molar was extracted ,socket was curettage and whole collected specimen was sent for histo-pathological examination.

Microscopically the lesion showed a solid Cribriform pattern with basaloid-type tumor cells arranged in cords and sheets.(figure 3). Islands of epithelial cells containing numerous spherical spaces showing a characteristic “Swiss cheese ”pattern were present.(figure 4).The tumour cells are small and cuboidal exhibiting deeply basophilic nucleous and little Cytoplasam, Based on microscopic findings, a diagnosis of intraosseous adenoid cystic carcinoma was made.

Patient was scheduled for further investigation but she refused for other investigations. So only chest x-ray was taken which was clear with out any lesion.

DISCUSSION

Salivary gland neoplasms affecting the jaw bone would appear to be very uncommon, with most cases being mucoepidermoid carcinomas. ACC occurring within the periapical region would also appear to be extremely rare [3]. Only 17 cases reported previously of Intraosseous adenoid cystic carcinoma[4]. Radiographically, most reports pertaining to intra-osseous salivary gland tumours appear to describe their presentation as unilocular or multilocular osteolytic radiolucencies [3]. For this case, a multilocular radiolucency located in a periapical area was observed. In addition, local pain appears to be a common finding for such lesions [3], as in this case. It can be misinterpreted as of dental origin infection. The correct diagnosis was only derived from the histological examination of the biopsied tissue. Consequently, the present case represents an intra-osseous ACC masquerading as a lesion of apical infection indicating that a proper diagnosis of such a lesion is dependent not only upon thorough clinical and radiographical examinations, but also upon the accurate interpretation of biopsied material. Furthermore, this particular case highlights the importance of the detailed histological examination of any tissues taken from the periapical region. In addition, central salivary gland tumours should be included in the differential diagnosis of lesions in the mandible, particularly for those located in the periapical area. The diagnostic criteria for primary intra-osseous salivary gland neoplasms have been proposed previously [5](Table 1).

Table 1 Diagnostic criteria for primary intra-osseous salivary gland neoplasms [5]

Diagnostic criteria
1. Radiographical evidence of osteolysis
2. Presence of intact cortical plates
3. Presence of an intact mucous membrane overlying the lesion
4. Absence of any primary tumour within the major or minor salivary glands
5. Histopathological confirmation of the typical architectural and morphological features of a salivary gland tumour

For the case reported here, all but the fourth diagnostic criteria were satisfied, as the patient refused an extensive work-up and examination.

The pathogenesis of intra-osseous salivary gland tumours remains controversial and several theories explaining the possible mechanisms for these tumours have been described [6,7] (Table 2) but no single theory of histogenesis is applicable to all cases of central salivary gland tumours.

Table 2 Possible mechanisms of intra-osseous salivary gland [6,7]

Possible mechanisms
1. Ectopic salivary gland tissue resulting from the entrapped minor salivary glands, or inclusions of embryonic rests of submandibular and sublingual glands or seromucous glands displaced from the maxillary sinus into the maxilla
2. Neoplastic transformation of the mucous-secreting cells commonly found in the epithelial lining of odontogenic cysts

True cases of central ACC do not arise from a metaplasia of odontogenic cysts and therefore, quite possibly, originate from the ectopic salivary gland tissues[3].Brookstone & Huvos (1992) reported that salivary gland neoplasms arising within the boundary of the jaw bones could behave differently to conventional salivary gland tumours found in the major and minor salivary glands, and therefore proposed a new clinical staging I–III for such lesions. The present case can be categorized as being a stage I disease, which has a fairly good prognosis.

Intra-osseous salivary gland tumours do not differ microscopically from their soft-tissue counterparts. Histologically, there appear to be three recognizable patterns of growth: solid, cribriform and tubuloductal.The main treatment for central ACC is excisional surgery [3]. A wide surgical resection paying particular attention to obtain clear margins around regional nerves was the suggested treatment as this tumour demonstrates a propensity for perineural growth. Furthermore, for such a condition, long-term follow-up is essential because this tumour type is notorious for its late and persistent recurrence and distant metastases, most commonly to the lung, brain and bone, in nearly 70% of patients at the time of their death [8].

CONCLUSION

In conclusion, central ACCs of the mandible occurring in the periapical area are rare, and they are a diagnostic challenge to every clinician in that they may easily be confused with lesions of odontogenic origin. The primary treatment objective in Adenoid cystic carcinoma patients is local control, normal functionality and distant metastasis prevention. For this purpose, early detection by the team of dental specialists is a pre-requisite, in order to enable a more favourable prognosis and better quality of life. The therapy involving combination of surgery & radiotherapy remains the modality of choice in most cases.

Figure:1

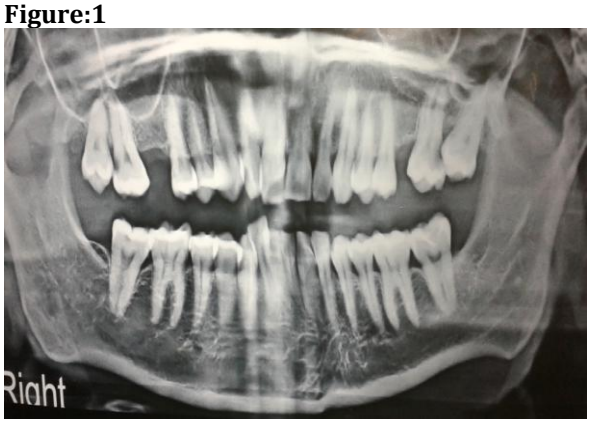

Figure:2

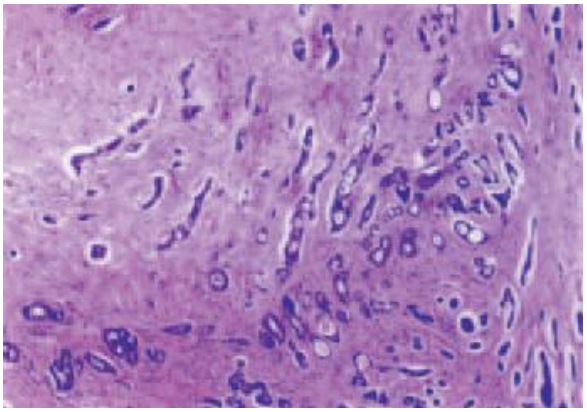

Figure:3

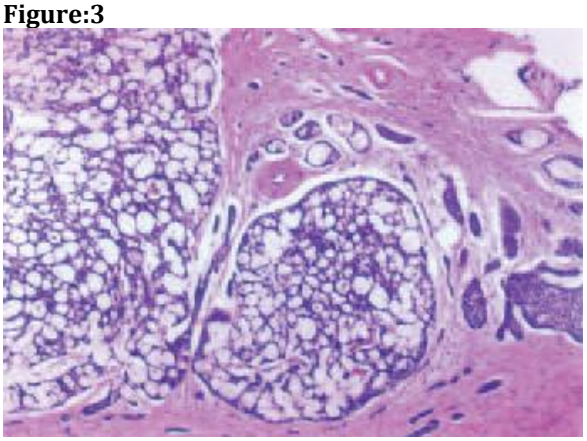

Figure :4

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