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CASE REPORT

Atypical Proliferative Ameloblastoma with a Rare Histopathological Feature and Review of Literature

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ABSTRACT

The ameloblastoma, because of its aggressive clinical behavior and its histological feature apparently benign, constitutes a puzzling paradox. Some hypotheses concerning this strange clinicalhistologic contradiction are analyzed, as well as the additional paradox represented by the neoplastic parenchyma itself, in which a tissue consisting of cells nominally able to form enamel do not elaborate any of the calcified dental tissues. Here I am presenting a rare case report of ameloblastoma having a rare histopathologic picture of combination of multiple histopathologic types.

Keywords: Ameloblastoma, Plexiform, Unicystic, Odontogenic.

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INTRODUCTION

Ameloblastoma name implies a resemblance to cells of the enamel-forming organ. The general agreement that ameloblastomas are odontogenic in origin occurs largely on the basis of the histologic similarities of the tumor and the developing enamel organ.[1,2] It has been described very aptly by Robinson[8] as being tumor that is 'usually unicentric, nonfunctional, intermittent in growth, anatomically benign and clinically persistent.

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The term 'ameloblastoma' as applied to this particular tumor was applied by Churchill[1] in 1934 to replace the term 'Admantinoma' coined by Malassez[2] in 1885, since the later term implies the formation of hard tissue, and no such material is present in this lesion. The first neoplasm of this nature reported in the scientific literature is credited to Broca[1] in 1868, although Guzack[1] reported a tumor of the jaw in 1826 which may be the first recorded instance of an ameloblastoma.[2] It is the second most common odontogenic neoplasm after odontoma.[9] Excluding odontoma, the incidence of ameloblastoma is at least equal to the incidence of all the other odontogenic neoplasms combined.

However G Sriram and Shetty RP[5] based on an institutional study on 250 odontogenic tumors reported ameloblastoma to be the most common (61.5%) odontoplasmic neoplasm in India. Ameloblastomas can be of varied origin, although the stimulus initiating the process is unknown. It may be derived from—Cell rests of enamel organ, either remnants of dental lamina or remnants of Hertwig's root sheath, the epithelial rests of Malassez., Epithelium of odontogenic cysts, particularly the dentigerous cyst, and odontomas, disturbances of developing enamel organ, basal cells of surface epithelium of jaws,[2,14] heterotopic epithelium in other parts of body, as in pituitary gland, now it is thought that it is likely to be the result of alterations or mutations in the genetic materials of cells that are embryologically programmed for tooth development.[6]

The average age of occurrence is in range of 33 to 39 years, and most cases occur between ages 20 and 60 years.[7] No significant sex predilection has been reported. It occurs in all areas of jaws, but the mandible is most commonly affected.[3] Molar angle ramus area is involved three times more frequently than are the premolar and anterior regions combined. Maxillary tumors tend to occur in slightly older patients than do the mandibular lesions.[14]

CASE REPORT

A healthy 35-year-old Hindu male patient presented to the Department of Oral and Maxillofacial Surgery, in January 2011 with complaint of pain and swelling over left lower 3rd of face since 6½ months.

Patient was relatively asymptomatic 6½ months back when he noticed a small swelling over left lower 3rd of

face which increased gradually in the first 4 months and then rapidly in last 2 months. Swelling was associated with pain, which was dull aching, intermittent and increased during chewing. There was no history of trauma, fever, pus discharge or restricted mouth opening. The patient gave history of surgery for a swelling in the same region.

Extraoral examination revealed a single well defined oval swelling over left angle of mandible of approximately 7 × 8 cm extending from left preauricular region to lower border of mandible and 2 cm lateral to left corner of mouth to posterior border of left ramus (Fig. 1). The swelling was nonwarm, tender and soft to firm and was fluctuant in certain areas. Left Submandibular lymph nodes were tender and palpable. Paresthesia was present over the left body of the mandible. The swelling did not yield any aspirate.

Intraorally the lips, labial mucosa and the buccal mucosa were blanched and fibrosed. A single well defined swelling of size approximately 6 × 4 cm was present in lower left buccal vestibule extending from lower left premolar region running posteriorly to the anterior border of ramus of mandible obliterating the buccal vestibule. Expansion of buccal and lingual cortical plates could be appreciated.

X-ray OPG showed single well-defined multilocular radiolucency extending from left parasymphysis to just below left sigmoid notch with complete resorption of anterior border and extreme thinning of posterior border at angle region with soap bubble appearance of the lesion (Fig. 2).

Computed tomography scan showed lytic, expansile, moderately enhancing soft tissue density lesion involving left ramus and angle of mandible with extension into the adjacent soft tissues along with enlarged submandibular lymph nodes (Figs 3 and 4).

On the basis of the above clinical and radiographic findings a provisional diagnosis of recurrent ameloblastoma and differential diagnosis of malignant ameloblastoma and ameloblastic carcinoma was arrived at. Incisional biopsy of the mass was done under local anesthesia and it was reported as Plexiform and Follicular Ameloblastoma.

The patient was subsequently planned for mandibular resection under general anesthesia. As the CT scan was suggestive of soft tissue infiltration of tumor mass so reconstruction was deferred.

The tumor mass was exposed with an extended submandibular incision. The tumor was then noted to involve the whole of the left mandible measuring about 8 × 7 cm including the body, ramus and head of condyle. In view of this extensive tumor involvement, hemimandibulectomy whereby the mandible was split at mesial to lower right canine and the left mandible was removed *en bloc* with the tumor (Fig. 5). The wound was closed primarily without reconstruction. Postoperative period was uneventful sutures were removed on day seven. Postoperative intermaxillary fixation was given for 21 days (Fig. 6).

Postoperative histopathological (excisional biopsy) result revealed unicystic ameloblastoma with mural proliferation of follicular and plexiform pattern along with areas of acanthomatous changes and presence of areas of mitotic figures and large numbers of dilated engorged capillaries suggestive of atypical proliferative

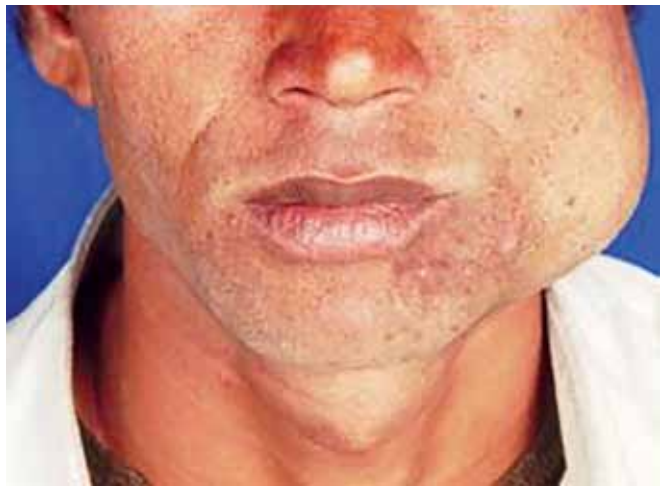

Fig. 1: Frontal view, showing swelling over left side of face

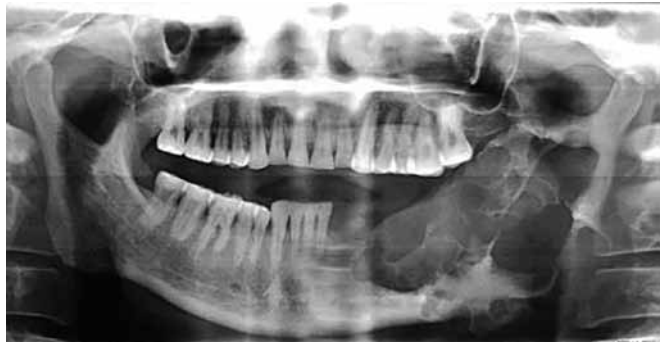

Fig. 2: Preoperative X-ray OPG

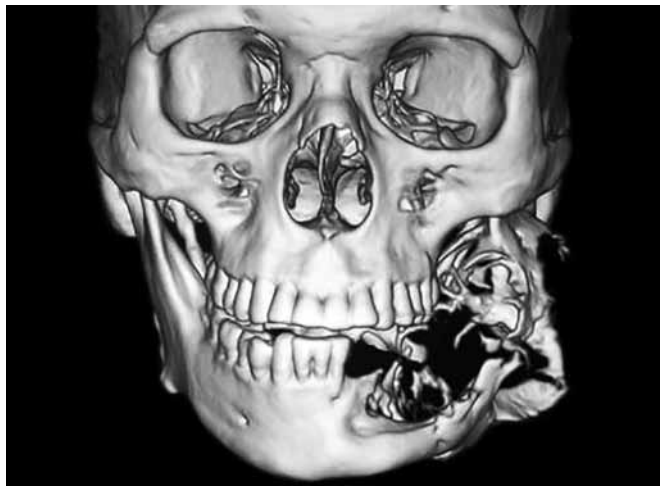

Fig. 3: 3D CT scan (frontal view)

ameloblastoma with a rare histopathological picture with the simultaneous appearance of three subtypes (Fig. 7).

DISCUSSION

Ameloblastoma is the commonest benign tumor of odontogenic origin which developed from epithelial cellular elements and dental tissues in their various phases of development.[13] It is generally a slow growing but locally invasive tumor. Reichart and Philipsen,[11] in an analysis of the three previously mentioned entities, state that the average age of the patients with ameloblastoma is 36 years. Gardner[12] criticizing such review, calculated a 39-year-old average age for the solid multicystic ameloblastoma, 22 for that unicystic ameloblastoma and of 51 years for the peripheral ameloblastoma. Equal incidences have been found in the two sexes by Reichart.

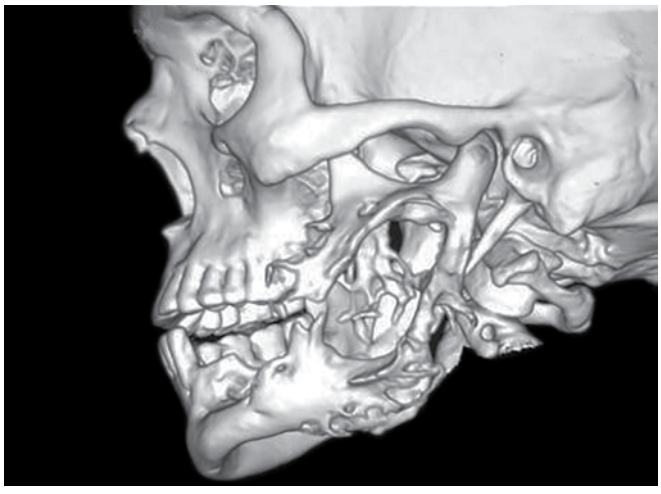

Fig. 4: 3D CT scan lateral view

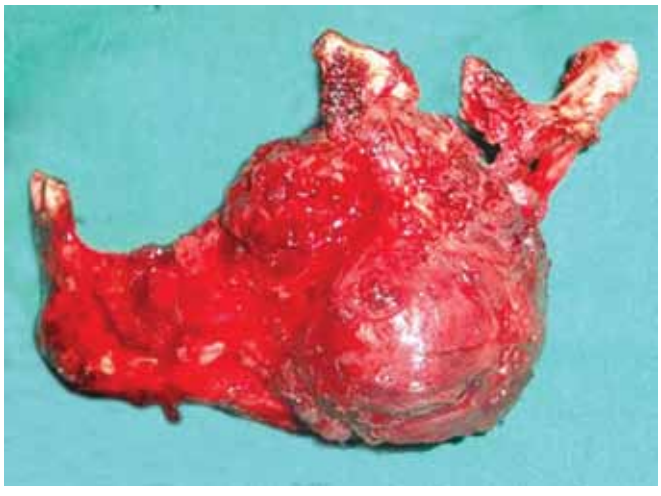

Fig. 5: Resected mandible

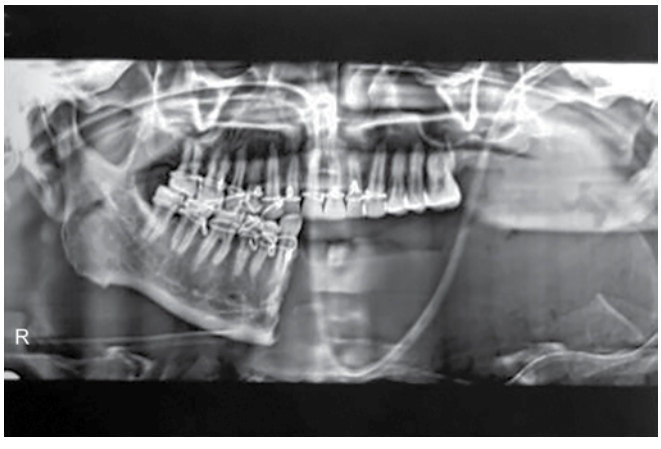

Fig. 6: Postoperative X-ray OPG

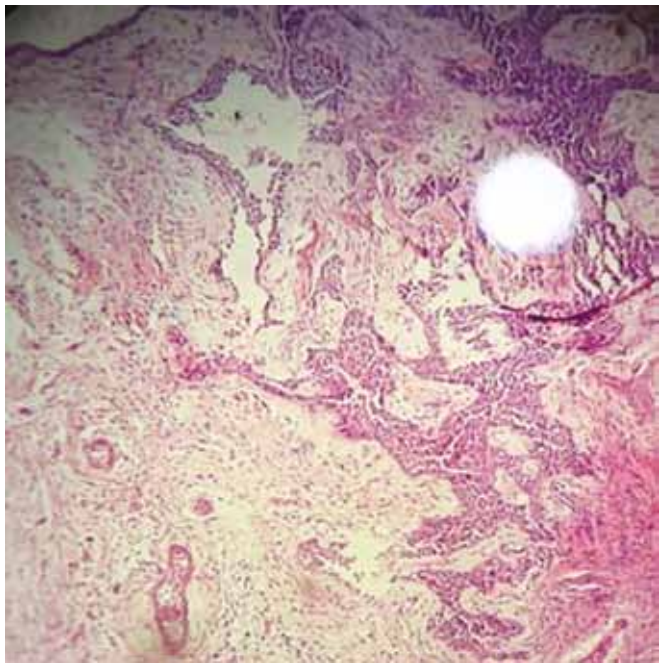

Fig. 7: Histopathologic view

It is usually asymptomatic, often presents as a slow growing, painless swelling, causing expansion of the cortical bone, perforation of the lingual and/or buccal plates and infiltration of soft tissue.[4] There is gradual facial asymmetry, with mobility and displacement of the involved teeth, pain and paresthesia may occur if the lesion is pressing upon a nerve or is secondarily infected.

Radiographically it usually has a well defined corticated border which is usually curved. The internal structure varies from totally radiolucent to mixed with presence of bony septa. The septa within the tumor are well developed and relatively coarse giving a gross honey comb appearance or soap bubble appearance.[9] There is pronounced tendency of ameloblastoma to cause extensive root resorption, though root displacement is also seen.

The six histopathologic subtypes of ameloblastoma include the follicular, plexiform, acanthomatous, granular cell, basal cell and desmoplastic types.[13] These subtypes can exist singly or in combination. The literature based retrospective study by Reichart et al[11] (1995) showed that the follicular type of ameloblastoma has the highest recurrence rate of 29.5%. The acanthomatous type showed only 4.5% recurrence and the plexiform is intermediate

between these two extremes 16.7%. The follicular type is the most commonly encountered variant and the basal cell type is rarest subtype.

Compared to its multicystic counterpart, the unicystic ameloblastoma tend to be less aggressive and has lower recurrence rate.[7] Even though some authors advocate a more conservative approach such as enucleation and curettage of this tumor, a large lesion with erosion of the mandibular cortex certainly requires a more aggressive approach for complete removal of the tumor.[10,15] Eppley[17] (2002) in his review of 60 mandibular ameloblastoma cases has shown that there was no recurrence of those cases treated via *en bloc* resection as compared to enucleation and curettage in which the recurrence rate was as high as 25 to 50%.

These characteristics of the ameloblastoma remember the basal-cell carcinoma, a known malignant neoplasm, although of low malignancy by its slow, invasive growth and by the fact that it only occasionally produces metastases. Ameloblastoma, on the contrary; known to be benign by its histological aspect, nevertheless, presents a highly aggressive behavior and, despite its slow growth, it is extremely invasive, as are malignant tumors, and produces occasional metastases.[16]

CONCLUSION

We think that the product of our experience, is useful for an efficient programming of treatment, taking in consideration numerous variables of this pathology and the indications found in literature. We believe that for the diagnostic phase the instrumental examinations (X-ray, CT or MRT) are essential; while intralesional biopsies are inefficient because they do not offer a whole vision of the tumor and could lead to diagnostic error. Therefore we think that it is advisable, considering: the site and extension, of the lesion, age and general conditions of the patient, to remove the lesion in a conservative manner in a first surgical step and according to the histolgical aspect evaluate a possible radical resection. But, the rare we are presenting here which 6 years before treated conservatively shows recurrence with more aggressive picture.

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