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CASE REPORT

Maxillary Ameloblastoma: A Rare Case Report

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ABSTRACT

Ameloblastoma is a benign but locally aggressive odontogenic tumor. The knowledge about this tumor is gaining greater importance because of its emerging variants. It is very essential to know the clinical, radiological and histopathological features of all the subtypes of ameloblastoma along with their behavioral and prognostic characteristics. Worldwide, maxillary ameloblastoma is rare, but its late detection renders adequate treatment difficult. Majority occur in the mandible with about 5 to 20% occurring in the maxillary bone. Here, we report a case of follicular ameloblastoma of the anterior maxilla in a 60-year-old male.

Keywords: Ameloblastoma, Maxillary ameloblastoma, Odontogenic tumor.

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INTRODUCTION

Ameloblastomas (from the early English word amel, meaning 'enamel' and the Greek word blastos, meaning 'germ')[1] are true neoplasms of enamel organ type tissue which does not undergo differentiation to the point of enamel formation. It has been described very aptly by Robinson as being a tumor that is 'usually unicentric, nonfunctional, intermittent in growth, anatomically benign and clinically persistent'.[2] The term 'ameloblastoma' was suggested by Churchill in 1934 to replace the term 'adamantinoma', coined by Malassez in 1885. In any event, the first thorough description of an ameloblastoma is that of Falkson in 1879.[2]

It is the second most common odontogenic neoplasm after odontoma.[2] The estimated incidence of ameloblastomas is approximately 0.5 per million population per year. Most cases are diagnosed between 20 and 60 years of age.[3] Maxillary ameloblastomas occur 12 years later than that of its mandibular counterpart.[4] Approximately, 15 to 20% of ameloblastomas have been reported to originate in the maxilla; with just 2% arising anterior to premolars.[5] According to the literature, solid ameloblastoma occurred least frequently in the maxillary bone.[3,6]

Based on the recent classification of odontogenic tumors, by World Health Organization (WHO), benign ameloblastomas are recognized in four subtypes: solid/multicystic, desmoplastic, unicystic and the extraosseous/peripheral types. Solid ameloblastomas affect the mandible preferably, especially the posterior region with a proportion between the gnathic bones of 1:5.[6,7]

CASE REPORT

A 60-year-old male patient, a farmer by profession, reported to the Department of Oral Medicine and Radiology of Rajarajeswari Dental College and Hospital, Bengaluru, with the chief complaint of swelling on the left side of the face for 2 months.

The patient was apparently asymptomatic until he noticed a small swelling on the left side of the face for 2 months. The swelling gradually increased in size to the present dimension. Patient did not give any history of pain associated with the swelling. Patient gave past history of trauma to the face due to a road traffic accident 2 years back. His medical, surgical, dental, personal and family histories were noncontributory. General physical examination revealed no abnormality other than those related to the chief complaint.

Extraoral examination revealed detectable facial asymmetry due to solitary swelling in the left middle third of the face with diffuse borders, roughly oval in shape, measuring approximately 4 × 5 cm, extending from midline till 5 cm anterior to the tragus. Superiorly, it extended upto the infraorbital margin. Obliteration of nasolabial fold was noted (Fig. 1).

Intraoral examination showed buccal cortical expansion in the left maxillary quadrant vestibular area extending from central incisor to first molar. The swelling was nontender, firm and predominantly nonfluctuant, with an area of fluctuance in the distal aspect of the swelling. Hard tissue examination revealed displaced 21, 22 and 23. Grade I mobility of 11, 21, 22, 23, 24 and 25 was noted (Fig. 2).

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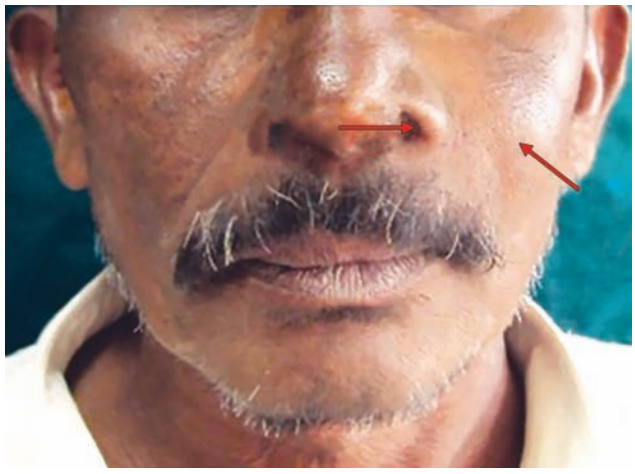


Fig. 1: Extraoral swelling on the left side of the face

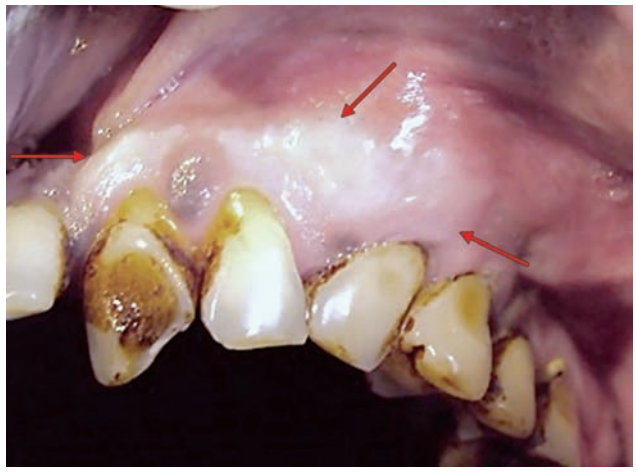
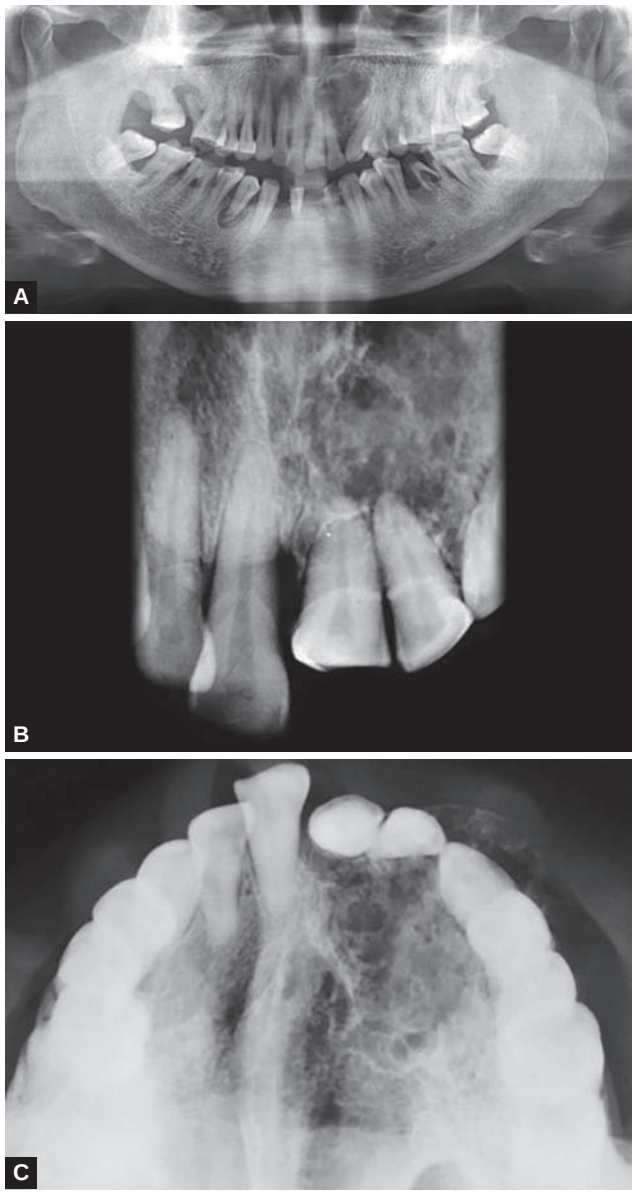


Fig. 2: Intraoral swelling showing buccal cortical expansion



Figs 3A to C: (A) Panoramic radiograph showing multilocular radiolucency extending from mesial aspect of 21 to mesial aspect of 23, (B) intraoral periapical radiograph of 11, 21, 22, 23, and (C) anterior maxillary occlusal radiograph showing multilocular radiolucency with soap bubble appearance extending from 21 to 26 with displaced 21, 22 and 23

Patient was then subjected to vitality test in relation to 11, 12, 13, 14, 15, 16, 21, 22, 23, 24, 25 and 26. All teeth were found to be vital except 22, 23 and 24. Needle aspiration was performed through the area of fluctuance which revealed yellow straw colored fluid. Based on the patient's history, clinical findings and the chair side investigations, a provisional diagnosis of radicular cyst in relation to 22, 23 and 24 was made.

Further, patient was subjected to radiographic examinations (panoramic radiograph, intraoral periapical radiograph of 11, 21, 22, 23 and anterior maxillary occlusal radiograph) which revealed a multilocular radiolucency with soap bubble appearance extending from 21 to 26 with displaced 21, 22, and 23. Haziness of left maxillary sinus was also noted (Figs 3A to C). Based on these findings, radiographic differential diagnosis of central giant cell granuloma, odontogenic myxoma, ameloblastoma were considered.

To know the clear extention of the lesion, cone beam computed tomography (CBCT) was performed along the anterior maxilla which revealed the presence of a large multilocular radiolucent lesion measuring about 28 mm superioinferiorly, 42 mm mediolaterally, 27 mm anterioposteriorly. The lesion extended from the midline, lateral to the nasopalatine canal (anteriorly), to the region of 26 (posteriorly). Buccopalatally, the lesion extended with expansion, thinning and perforation of the buccal cortex upto the canine region. Distal to canine the lesion extended subcortically causing expansion of buccal cortex but sparing the alveolar bone. Superiorly, the expansile lesion extended into the nasal cavity causing thinning and perforation of the lateral nasal floor. The lesion had a scalloped border and honey comb like multilocularity. The posterior part of the lesion showed central air filled cavity while anteriorly dense soft-tissue opacity was noted. The lesion appeared to be centered in between 22

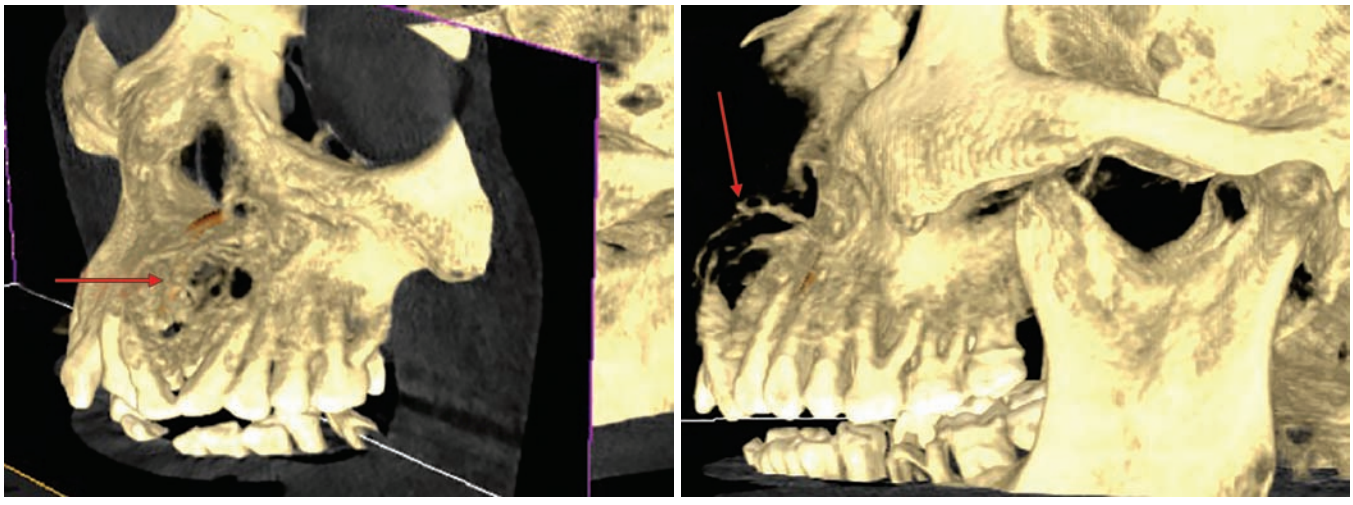


Fig. 4: Three-dimensional volume rendered image (arrows showing erosion and expansion of labial cortical plate irt 21 and 22)

and 23 with lateral and buccal displacement of the roots of these teeth along with 21. Loss of tooth structure with periapical radiolucency was present in relation to 25 and 26 (Fig. 4).

Incisional biopsy was performed and the histologic analysis of the specimen revealed numerous odontogenic follicles in a mature stroma where peripheral cells of follicles showed tall columnar cells with reversal of polarity and centrally stellate reticulum like cells were seen (Fig. 5). Few follicles showed juxtaepithelial hyalinization along with acanthomatous changes and follicular degeneration which was suggestive of follicular ameloblastoma.

Based on all investigations, a final diagnosis of follicular ameloblastoma was given. Surgical removal of the ameloblastoma along with the involved teeth was advised, but as the patient was not cooperative, the treatment could not be performed.

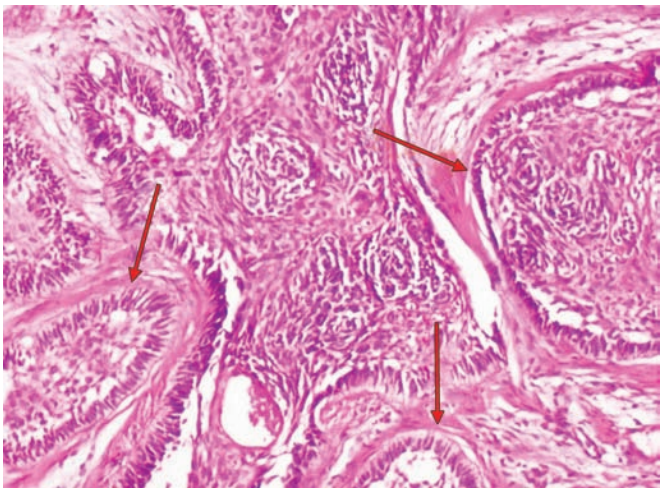


Fig. 5: Histological picture (40× magnification) showing numerous odontogenic follicles in a mature stroma. Peripheral cells of follicles are columnar showing loss of polarity with central reticulum like cells. Acanthomatous and follicular degeneration is also evident in a few areas

DISCUSSION

Globally, maxillary ameloblastoma is rarer than mandibular lesions. It is generally accepted that only 20% of ameloblastomas occur in the maxilla, although some reports indicate an incidence as low as 1% in the maxilla and of those 47% occur in the molar region, 15% in the antrum and the floor of the nose, 9% in the premolar areas, 9% in the canine regions and 2% in the palate.[8]

Ameloblastoma occurs over a broad age range; cases have been reported in children younger than 10 years through elderly adults older than 90. The average age at diagnosis consistently is reported in the age range of 33 to 39, and most cases cluster between ages 20 and 60 years,[9] as in our case. No significant sex predilection has been reported.[10]

Maxillary ameloblastoma has a predominantly painless and slow growth because of the lack of a thick cortical plate, plentiful cancellous bone and the proximity of the maxilla to the nasal cavity, nasopharynx, paranasal sinuses, orbits and skull base. There is commonly a delay in the recognition of the maxillary ameloblastoma extending into these structures and this itself may prove fatal in some cases. In addition, the more abundant blood supply of the maxilla provides another possible mode of spread. Sometimes, invasive maxillary ameloblastomas with extension into the orbit, frontal sinus, skull base, middle cranial fossa and petrous apex have resulted in the death of the patient.[8]

Patients with ameloblastoma most commonly present with chief complaints of swelling and facial asymmetry. Although, the swelling is typically asymptomatic, pain is an occasional presenting sign. A chief complaint of painless swelling often heralds a lesion of long duration and significant size. Continued growth of the tumor and enlargement of the involved area may eventuate

in ulceration of the mucosa overlying the lesion. Small lesions tend to be discovered more often on routine radiographic screening examinations or as a result of local effects produced by the tumor. Such local effects include: tooth mobility, occlusal alterations, and failure of eruption of teeth.[4,10,11]

Ameloblastomas may present on conventional radiographs as a unilocular or multilocular corticated radiolucency resembling a cyst. Bony septa may result in a honeycomb or soap-bubble appearance.[3,11] The lesion may remain asymptomatic before a facial swelling develops. Radiographs of our case had also revealed multilocular radiolucency. Computed tomography (CT) and magnetic resonance imaging (MRI) may be helpful in establishing the extent of the lesion, particularly when located in the maxilla.[3]

Six histopathologic subtypes of ameloblastoma are recognized: follicular, acanthomatous, granular cell, basal cell, desmoplastic, and plexiform.[4,11] Most tumors show a predominance of one pattern, but few lesions are found to be composed purely of one histopathologic subtype.[11] Mixtures of the different patterns commonly are observed. Lesions tend to be subclassified according to the predominant pattern that is present.[4] In this case, predominantly follicular pattern was seen with some amount of acanthomatous changes.

A number of modalities have been proposed in the treatment of ameloblastoma. Maxillary ameloblastomas are more difficult to treat because of the combination of the well vascularized, fragile, cancellous maxillary bones, presence of the paranasal sinuses, nasal and orbital cavities which readily facilitates tumor spread to the zygomatic bone, cranial base and paracranial structures and the pterygomaxillary fissure.[8] These are usually treated by resection with 10 to 15 mm safety margin of healthy bone. This may include resection of the alveolar ridge, hard palate and maxillary sinus and the lateral nasal wall.[12] The prognosis is dependent on the extension of the lesion and adjacent structures involved rather than the origin of the lesion.

Ameloblastoma has a persistent and slow growth, spreading into marrow spaces with pseudopods without concomitant resorption of the trabecular bone. As a result, the margins of the tumor are not clearly evident radiographically or grossly during operation and the lesion frequently recurs after inadequate surgical removal, showing a locally malignant pattern. Long-term follow-up is necessary, because this lesion has been shown to recur 25 and 30 years following primary treatment.[5]

CONCLUSION

Ameloblastoma is characterized by specific clinical, imaging and histological features. The clinician should be alert to the unusual presentation of this neoplasm and include ameloblastoma as a differential diagnosis in any lesion ranging from simple abscess to any fibro-osseous lesions/neoplastic growth presenting in anterior maxilla. The definitive diagnosis requires histopathological examination. With the potential for recurrence, such cases should always be kept under long-term follow-up post-treatment.

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