Case Report

Giant acanthomatous epithelial tumor of the mandible: A rare entity

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ABSTRACT

Ameloblastoma is a rare benign odontogenic tumor which arises from the odontogenic epithelium. It usually exhibits a range of histopathologic features, such as follicular, plexiform, acanthomatous, granular, basal cell, and desmoplastic variants, which are well recognized. This study reports a case of giant acanthotic ameloblastoma that developed at the anterior mandible in a 70-year-old male. Radiologically, it was diagnosed as ameloblastoma, benign tumor of the jaw which is rare. Biopsy was taken intra-orally, it was confirmed as acanthomatous ameloblastoma. When extensive squamous metaplasia, often associated with keratin formation occurs in central portions of the epithelial islands of follicular ameloblastoma, the term acanthomatous is sometimes applied.

Key words: Ameloblastoma, giant, mandibular tumors, odontogenic, squamous, X-ray

INTRODUCTION

Ameloblastoma is the best known and the most frequent form of odontogenic tumors. Odontogenic tumors comprise a heterogeneous group of lesions that ranges from hamartomas to benign and malignant neoplasms but locally aggressive odontogenic neoplasm has a high recurrence rate.[1-3] The reported recurrence rate of acanthomatous ameloblastoma (AA) after resection varies from 0% to 25%.[4] AA is considered as an aggressive tumor of the canine jaw, characterized by irregular verrucous masses adjacent to the tooth.[5] There have been 11 cases reported of extreme ameloblastoma till date.[6] We report a case of acanthotic type of ameloblastoma of size 10 cm × 14 cm × 11 cm, which is a rare entity. The radiolucent area of pediatric squamous odontogenic tumors shows the involvement of the roots of neighboring teeth which is associated with an unerupted canine. Radiological investigations such as X-ray and CT scan helps in diagnosis and in management especially where symptoms are non-specific.[7] Patients with extreme ameloblastomas are usually from rural areas of developing countries who delay the treatment due to fear of surgery. Because of its clinical behavior and diversity of histological features, ameloblastoma poses a challenge to radiologist and surgeon in view of management especially if it is very huge in nature as seen in our case.

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CASE REPORT

A 70-year-old male patient reported with a large swelling over the left side of the face since 2½ years. It was of small in size which slowly increased in size to the present size of 10 cm × 14 cm × 11 cm. There was no complaint of pain, loss of appetite, fever or weight loss. Vitals were stable. Local examination revealed a nontender firm swelling of the size 10 cm × 16 cm over the jaw region. Skin texture was normal. It was extending from the lower border of the mandible to the zygometric arch and from the preauricular area to the ala of the nose and angle of the mouth. Swelling was fixed to the underlying structures but was free from the skin. Intra-orally a small soft-tissue mass present sublingually involving the left side of the edentulous mandible [Figure 1].

All routine blood investigations were within normal limits. On postero-anterior, X-ray of the mandible is showing large expansile multiloculated lytic lesion involving the left side of the ramus of the mandible with cortical erosions at places [Figure 2]. A axial contrast-enhanced

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computed-tomography (CT) revealed a large heterogeneously enhancing soft-tissue mass arising from the left mandibular ramus causing destruction of mandible and new bone formation in the form of bony septae with calcifications [Figure 3a and b]. Biopsy took intra-orally, and it was diagnosed as squamous AA. The peripheral palisading was present with cystic changes along with focal areas of center squamous metaplasia with formation of the keratin pearls [Figure 4]. Peripheral part of the tumor showed bony trabeculae containing sheets of the tumor cells.

DISCUSSION

The term "giant" or "extreme" ameloblastoma is reserved for lesions that are truly large and that cause gross asymmetry and regional dysfunction. The tumor is classified histologically into two main types: Plexiform and follicular and further sub-classified into four types: Acanthomatous, granular cell, desmoplastic and basal cell carcinoma.[8]

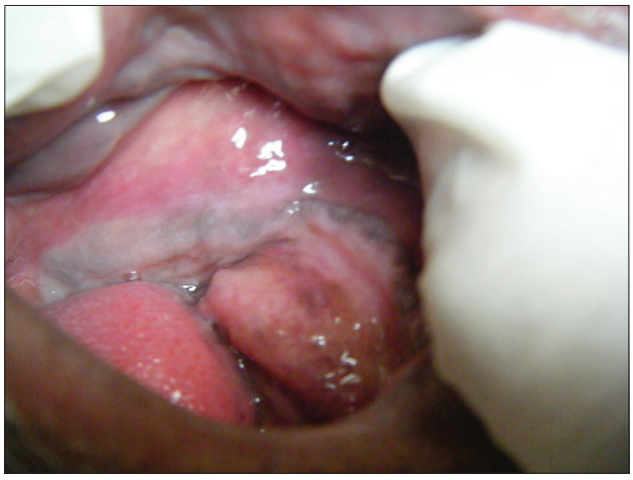


Figure 1: Gross appearance of the lesion showing intra-orally

Acanthomatous is benign locally aggressive tumor that invades the alveolar bone which is a rare subtype.[9] The etiopathogenesis of ameloblastoma is not clear, but it is suggested that the tumor may be originating from cell nests of the enamel organ, epithelium of odontogenic cyst or disturbance of the developing enamel organ. Squamous metaplasia such as that seen in AA may be attributed to chronic irritation due to calculus and oral sepsis.[10]

Squamous odontogenic tumors clinical and radiographic features are neither unique nor sufficient for diagnosis, as this tumor may be confused with a number of other pathologies. This tumor may occasionally be misdiagnosed as ameloblastoma, squamous cell carcinoma, verrucous carcinoma, and keratoacanthoma. CT findings include cystic areas of low attenuation with scattered isoattenuating regions, representative of soft-tissue components. The lesion can also erode through the cortex with extension into the

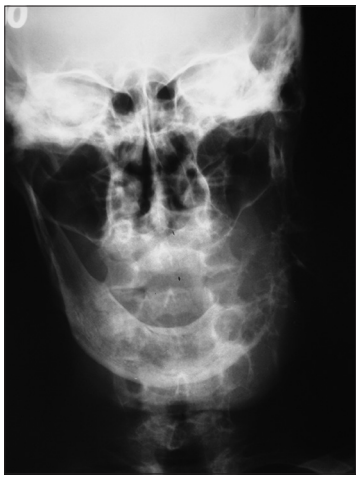
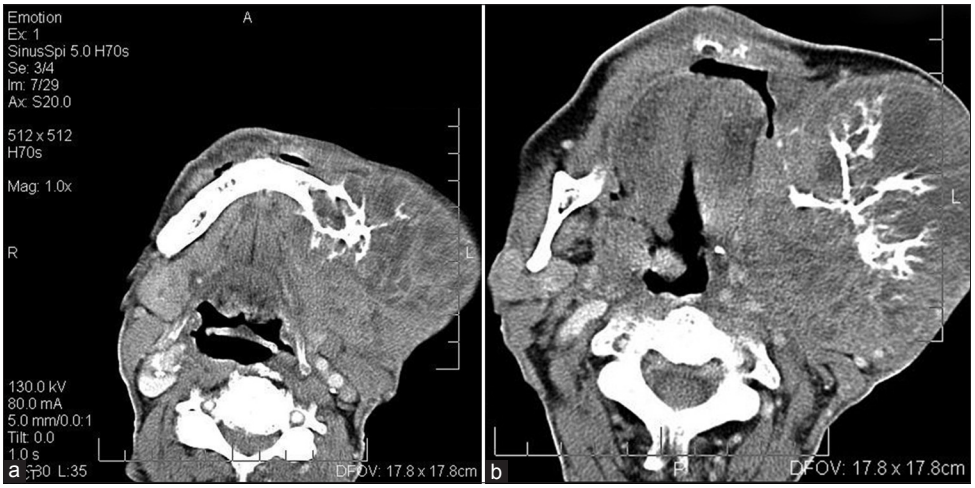


Figure 2: Postero-anterior radiograph of the mandible is showing large expansile multiloculated lytic lesion involving the left side of the ramus of the mandible with cortical erosions at places



Figures 3: (a and b) Axial contrast-enhanced computed-tomography is showing large heterogeneously enhancing soft-tissue mass arising from the left mandibular ramus causing destruction of the mandible and new bone formation in the form of bony septae

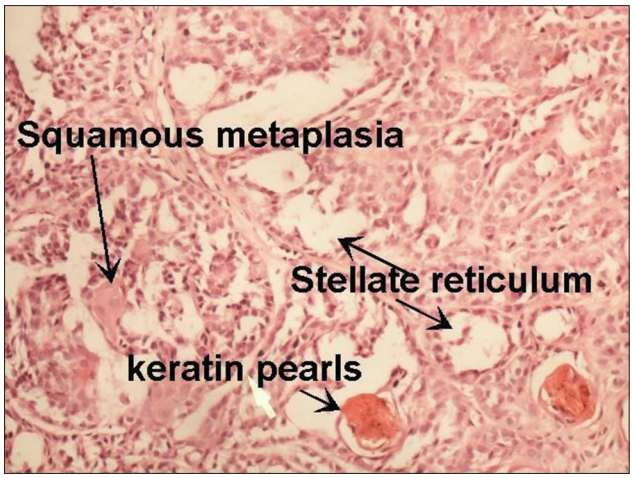


Figure 4: Histopathological (H and E, ×100) revealed solid epithelial cell nests with peripheral palisading tumor cells and central stellate reticulum with squamous pearls

surrounding oral mucosa. In addition, erosion of the roots of adjacent teeth is unique to ameloblastoma and indicates the aggressive behavior of the tumor.

The treatment of choice is complete surgical resection. If possible, conservative surgery may be used if an assured complete removal can be performed. In the present case, surgical resection of the lesion was done. In addition to low sensitivity of this neoplasm, the intraosseous location of the ameloblastoma prevents the use of radiotherapy as an effective therapeutic option because radiation induces the potential development of secondary tumors. Therefore, in all types of ameloblastomas, a thorough long-term clinical and radiographic follow-up is always recommended.[11] AA if left untreated can develop into an invading squamous cell carcinoma.

CONCLUSION

A definitive diagnosis requires that a more extensive biopsy be performed to obtain a larger portion of the lesion, but we have made the diagnosis on radiological investigations and was confirmed on biopsy. Clinical and microscopic correlations are, therefore, crucial because the aggressive odontoameloblastoma shares a high-risk of recurrence with ameloblastoma.

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