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Recurrence of Ameloblastoma of Maxilla in a 35 Year Old Male: A Case Report

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Case Report

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Abstract: Ameloblastoma or admantinoma is a benign odontogenic tumor of epithelial origin. It is rare tumour occurring in the jaws which constitutes 1–3% of all cysts and tumours of jaw with locally aggressive behavior & high recurrence rate. The tumor is by far more common in the mandible than in the maxilla. A unilocular or multilocular radiolucency with a honeycomb or soap bubble appearance is the most striking feature. Sometimes ameloblastoma is indistinguishable from a dentigerous cyst. We present a case report of ameloblastoma of right posterior maxilla in a 35 year old male.
Keywords: Ameloblastoma, Anterior maxilla, Neoplasms.

INTRODUCTION

Ameloblastoma is a benign, slow growing, locally invasive odontogenic tumor [1]. It is the second most common odontogenic neoplasm and accounts for 12% of all odontogenic tumors. It is prevalent in all age groups but most frequently occurs in third and fourth decades of life [2]. Cusack described it in 1827 and French physician Louis-Charles Malassez designated it as an adamantinoma in 1885 [3]. The first detailed description was given by Falkson in 1879 and renamed as ameloblastoma in 1930 by Ivey and Churchill [4]. Exact etiology of ameloblastoma is unknown. Tumor has been reported by far more common in mandible than maxilla [5]. In the mandible, the most commonly affected sites are molar and angle (70%) followed by premolar (20%), and rarely anterior region (10%). [6] The molar/ramus area is the most frequently involved in Japanese [7] and Whites [8]. In blacks, ameloblastomas occur more frequently in the anterior region of the jaws [9].

Ameloblastoma may arise from: Cell rest of enamel organ either remnant of dental lamina or remnant of Hertwigs sheath; epithelium of odontogenic cyst; and disturbances of developing enamel organ. The neoplasm is usually asymptomatic, painless, and present with bony deformity [10].

CASE REPORT

A 35 year old male patient came to department of oral medicine, diagnosis & radiology, Institute of Dental Education & Advance Studies, Gwalior, Madhya Pradesh with chief complaint of swelling in upper right back jaw region since 4 years. Patient gives history of trauma 12 years back due to accidental collision with wooden table. The swelling appears 3 years later and patient got it completely excised along with extraction of 14 15. Patient gives history of recurrence of gradually progressive swelling since 4 years. Paresthesia was also reported. Extra oral reveals facial asymmetry due to unilateral large diffuse swelling in right maxillary anterior measuring approximately 6× 5 cm in size which extended from ala of nose to outer canthous of eye antero posteriorly and 2 cm below right infraorbital margin to ala tragal line superior inferiorly. (Figure 1 & 2) Color of the overlying skin was normal. On palpation, the swelling was soft, compressible, and non tender. Intra-oral inspection showed a large firm swelling that extends in the right maxillary premolar region obliterating buccal vestibule with no displacement of adjacent teeth. (Figure 3a, b) Overlying skin was normal in color. Neither tenderness nor pus discharge was observed upon palpation. There was expansion of buccal cortical plate. Patient visited a private hospital 4 days back and was advised Paranasal sinus view (PNS) and contrast enhanced computed tomography (CECT) in axial, coronal and sagittal planes. Patient brought these radiographic investigations to us. PNS reveals large obliteration of right maxillary antrum. (Figure-4) CECT reveals well defined lobulated multiilocular radiolucency with a large expansile cystic lesion of size 4.4 x 4.1 cm arising from right maxillary alveolus in premolar area and extending into right maxillary antrum (Figure 5 & 6). The inferior fissure was not infiltrated and the lower

right concha of the nose was not distended and penetrated in any position. Provisional diagnosis of ameloblastoma was given with dentigerous cyst and giant cell tumour in differential diagnosis. Incisional biopsy revealed follicular ameloblastoma. The patient was operated under LA. Patient was called for follow-up. Histopathologic examination (Figure-7) revealed ameloblastic follicles in a fibrocellular connective tissue stroma. The peripheral cells of the follicles were tall columnar ameloblast like cells, demonstrating reversed nuclear polarity and central stellate reticulum like areain center suggestive of follicular ameloblastoma.

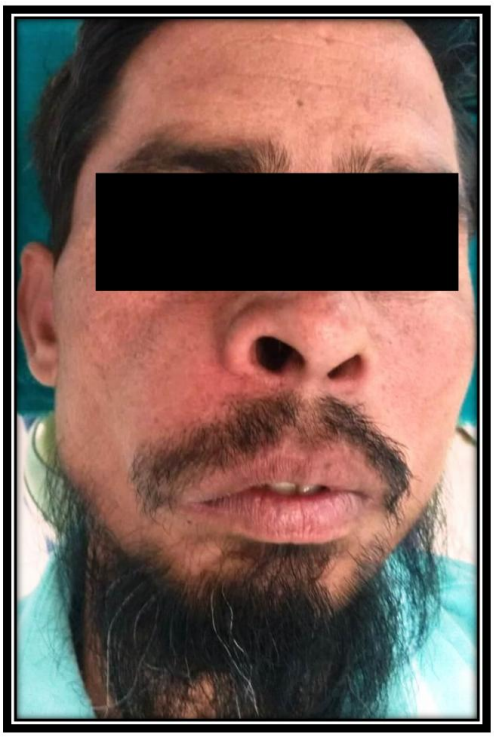

Fig-1: Frontal profile

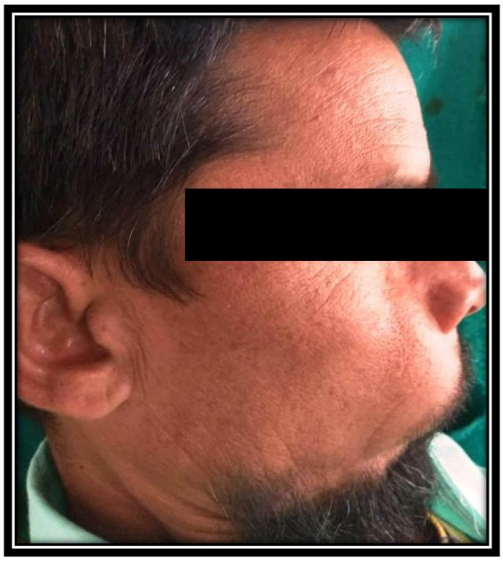

Fig 2 Lateral Profile

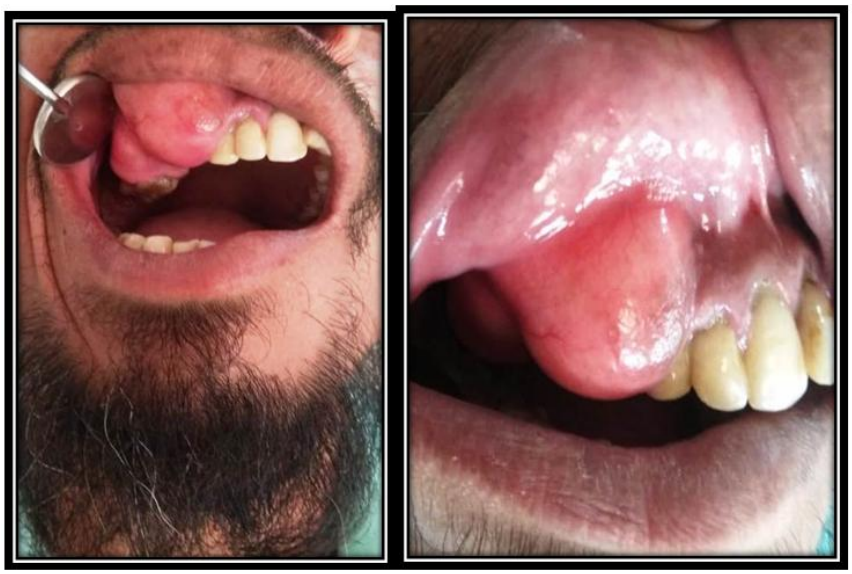

Fig-3 (a, b): Intraoral photograph of patient

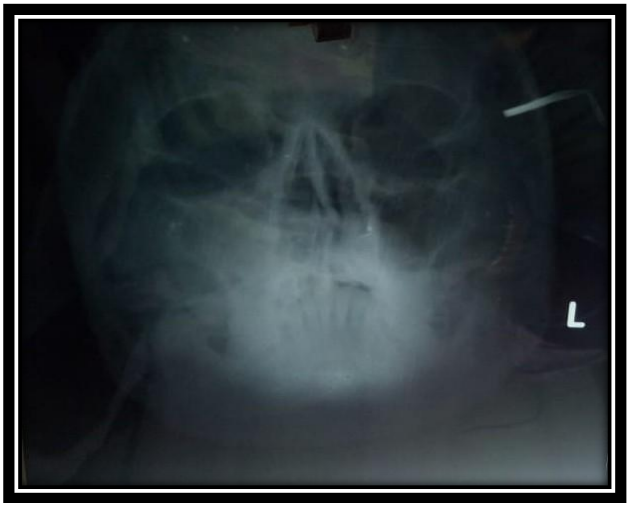

Fig-4: Paranasal sinus view

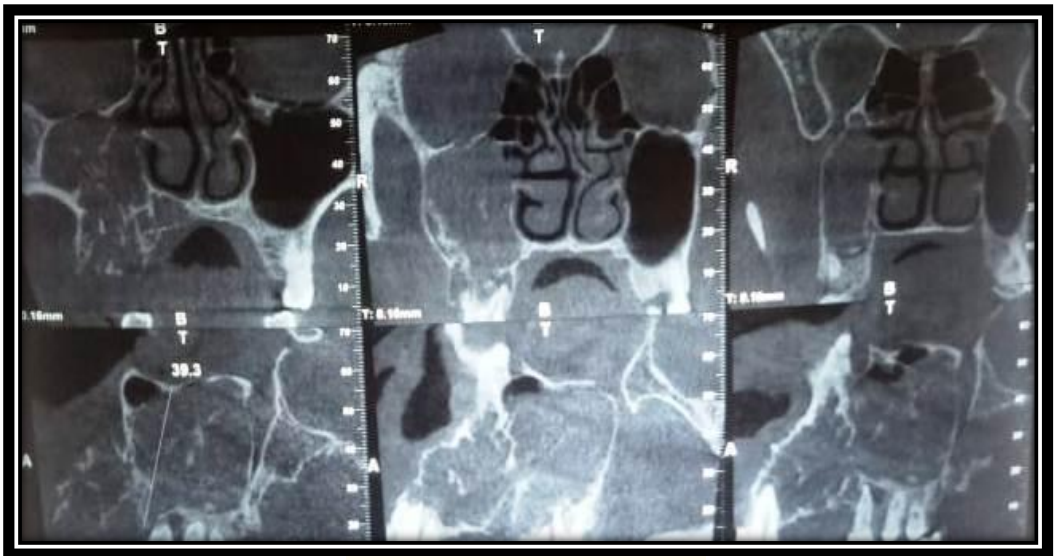

Fig-5: CECT scan-coronal view

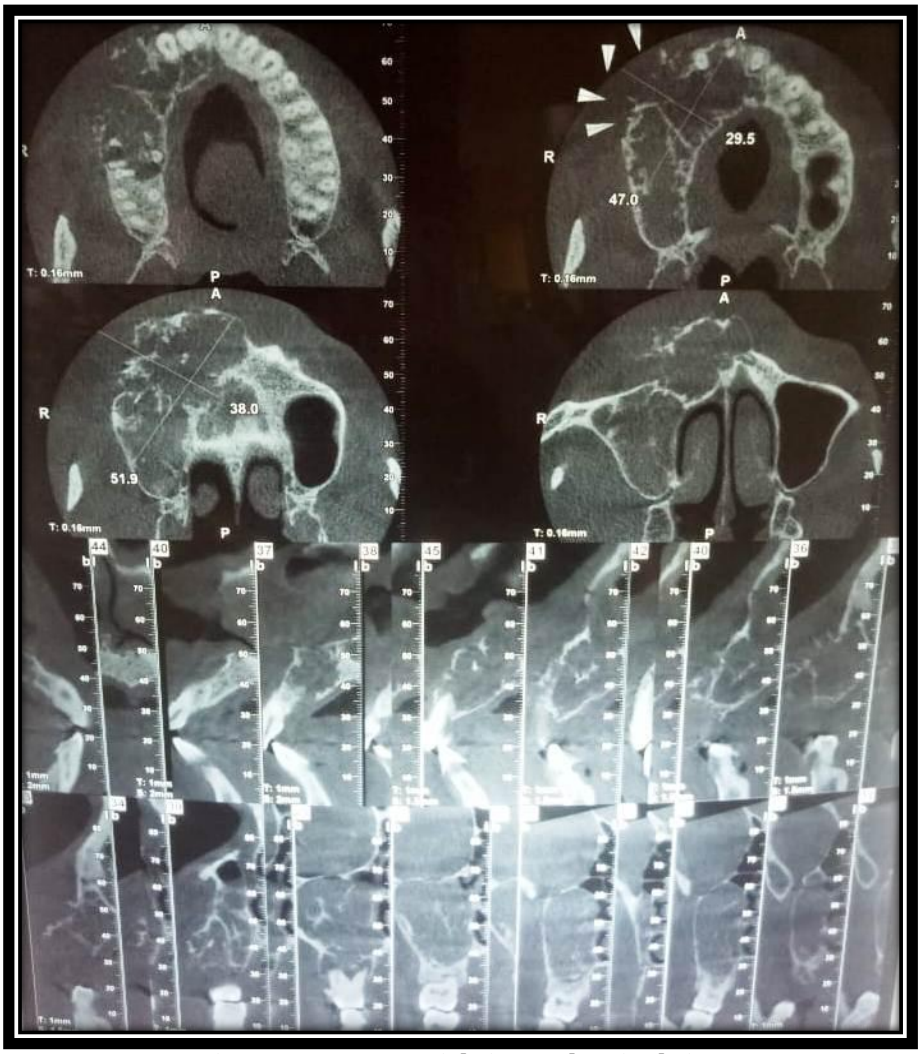

Fig-6: CECT scan-axial view and sagittal view

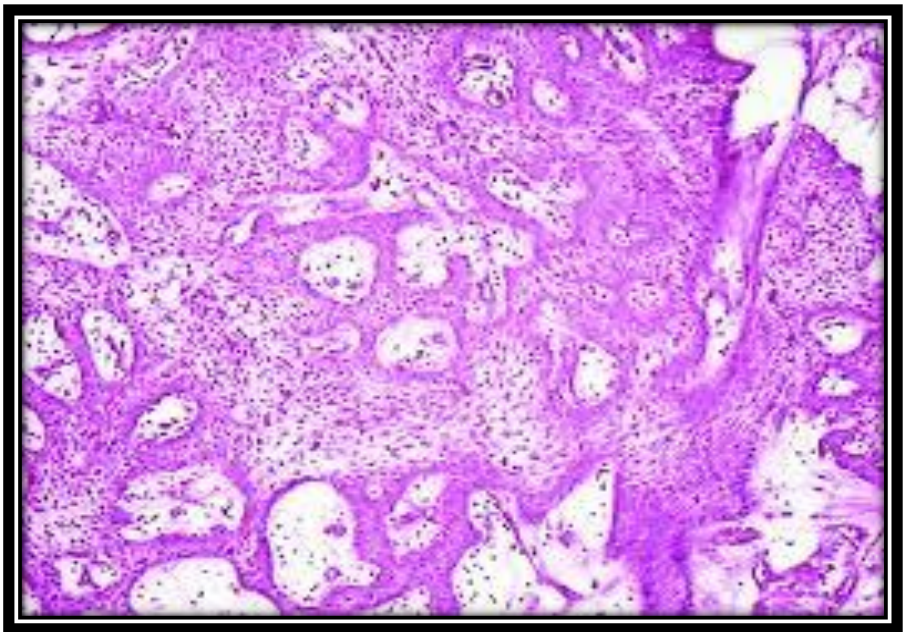

Fig-7: Histopathological picture

DISCUSSION

Ameloblastoma is a true neoplasm of enamel organ type. Robinson described it as unicentric, nonfunctional, intermittent in growth, anatomically benign, and clinically persistent [11]. Radiographically it can be unicystic, multicystic, or solid and peripheral type [12]. Out of all reported cases 86% are of multicystic or solid type. Unicystic ameloblastoma is of three subtypes: luminal, intraluminal, and mural. Our case is of multicystic type. Radiographically, ameloblastoma appears radiolucent as they are osteolytic and are frequently multilocular with well-defined sclerotic margins [13]. It causes a slow growing, painless expansion of jaw which causes thinning of cortical plates. Margins may or may not appear scalloped. Root resorption, tooth mobility, and paresthesia are featuresassociated with advanced cases of ameloblastoma. Follicular, granular, plexiform, desmoplastic, basal cell, and acanthomatous are histological variants of this pathological entity [14]. Calcifying epithelial odontogenic tumor (CEOT), odontogenic myxoma, central giant cell granuloma, or ameloblastic fibroma is considered as differential diagnosis of ameloblastomas. Management of lesion includes surgical excision, enucleation, curettage, cryotherapy, radiotherapy, and chemotherapy [15]. Wide surgical excision with adequate safe margins is the treatment of choice and it involves complete removal of tumor with negative margin of 15-20 mm. Recurrence rate of this lesion is high. Recurrence rates were 3.6% for wide resection, 30.5% for enucleation, and 16% for enucleation followed by use of Carnoy's solution as reported by Lau *et al*., [16] Clinicopathological variant of tumor, anatomic site, safe margins during surgery and histological variants influences recurrence rate of lesion. The solid variety has the greatest propensity for local infiltration and recurrence. The dense cortical bone of the mandible prevents the tumor from spread than maxilla [17]. Role of radiotherapy has not been established as a useful treatment modality in case ameloblastoma [18]. Role of chemotherapy is not yet well-defined, however, few reports showed little response with Cisplatin and Paclitaxel [19]. Prevalance rate of malignant transformations of ameloblastoma is less than 1%. Malignant ameloblastoma may arise de novo or transformation of pre-existing ameloblastoma. Ameloblastomas rarely metastasize. Most common site of spread are lung, cervical lymph nodes, and rarely brain. Long-term follow up is recommended due to high rate of recurrence irrespective of the treatment done [20].

CONCLUSION

It can be concluded that at present, histologic examination is the most sensitive tool for differentiating between odontogenic cysts and tumors. However, both clinical and radiologic findings share equal contribution to the final diagnosis. In order to make an exact differential diagnosis, it is of great significance for the radiologists to become familiar with and to investigate the radiographic images of this rare lesion, even in the literature. Thus, it is of utmost importance to correlate the histopathologic findings with clinical and radiographic features to achieve at a correct definitive diagnosis as all such lesions may have prognostically different biologic behaviours and the final diagnosis may alter the therapeutic decision significantly.

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