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Case Report (Open Access)

AMELOBLASTOMA OF MANDIBLE – REPORT OF 3 CASES

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ABSTRACT
Ameloblastoma is a benign, locally aggressive and recurrent neoplasm arising from the odontogenic epithelium. Ameloblastoma comprises about 1-3% of all oral tumours and cysts of the jaws and 11% of all odontogenic tumors. Radiographically 3 types of ameloblastoma are Unicystic, Conventional, Peripheral (extra-osseous) variety. Histomorphologically ameloblastoma has multiple types which include Follicular, Plexiform, Acanthomatous, Granular, Basal cell & Desmoplastic. These types may pose a diagnostic challenge to pathologist while reporting especially in the biopsies.
We have studied 3 histopathological types of ameloblastoma and correlated them clinically and radiographically.

Keywords: *Ameloblastoma, Benign, Recurrence*

INTRODUCTION

Ameloblastoma is a benign, locally aggressive and recurrent neoplasm arising from the odontogenic epithelium (Suma *et al.*,2013). Ameloblastoma comprises about 1-3% of all oral tumours and cysts of the jaws and 11% of all odontogenic tumors (Singh *et al.,* 2019). The tumour is more common in the mandible than in the maxilla. Other rare locations include sinonasal tract, middle ear, temporal bone and infratemporal fossa (Suma *et al.*,2013). It is commonly seen in 3rd to 7th decades of life with equal male and female predominance (Formiga *et al.,* 2007). They are usually asymptomatic and may be detected incidentally during radiographical examination as cystic lesions. This may be unilocular or multilocular with expansile radiolucency and well defined corticated border (Reddy *et al.*,2015). According to World Health Organization (WHO) in 2005, four histological variants of ameloblastoma were described as solid/multicystic, unicystic, desmoplastic, and extraosseous/peripheral subtypes (Barnes *et al.*,2005). However; in 4th edition of World Health Organization, 2017, the name of solid/multicystic ameloblastoma changed to Ameloblastoma, conventional and the desmoplastic type was included under the common term of ameloblastoma (9310/0 ICD-O code) (El-Naggar AK *et al.*,2017). The clinico-radiological types of ameloblastoma are Unicystic, Conventional and Peripheral (extra-osseous) variety. Ameloblastoma has multiple histological variants of which, Follicular and Plexiform varieties are most common followed by Acanthomatous and Granular types. Less frequent variants of ameloblastoma includes Desmoplastic ameloblastoma, Clear cell ameloblastoma, Basal cell ameloblastoma and Keratoameloblastoma (Sharma *et al.,* 2016). The identification of the morphological subtypes of Ameloblastoma is important as some subtypes may behave aggressively with higher recurrence rate.
We have studied 3 histopathological types of ameloblastoma and correlated them clinically and radiographically.

Case Report 1

57 year female, came with gradually progressive swelling in right jaw since 1 year. Clinically, it was non tender and hard swelling fixed to right mandible. CT scan revealed well defined heterogeneously hypodense 4.4x4.2x5.2cm solid cystic lesion with peripheral calcification in the right side of body of mandible with erosion. We received segmental mandibulectomy specimen showing single expansile tumor measuring 5x4x4cm, pushing inner mucosal surface along the body of mandible. Cut surface was solid cystic showing multiple cysts. Microscopy showed multicystic tumor comprised of cystic spaces lined by odontogenic epithelium. The tumor was arranged in anastomosing strands and islands with peripheral columnar cells showing reverse polarity and palisading along with central loosely cohesive

stellate reticulum like cells showing predominant granular cell change. Diagnosis of Granular cell type of ameloblastoma was given. (Figure 1a,1b,1c)

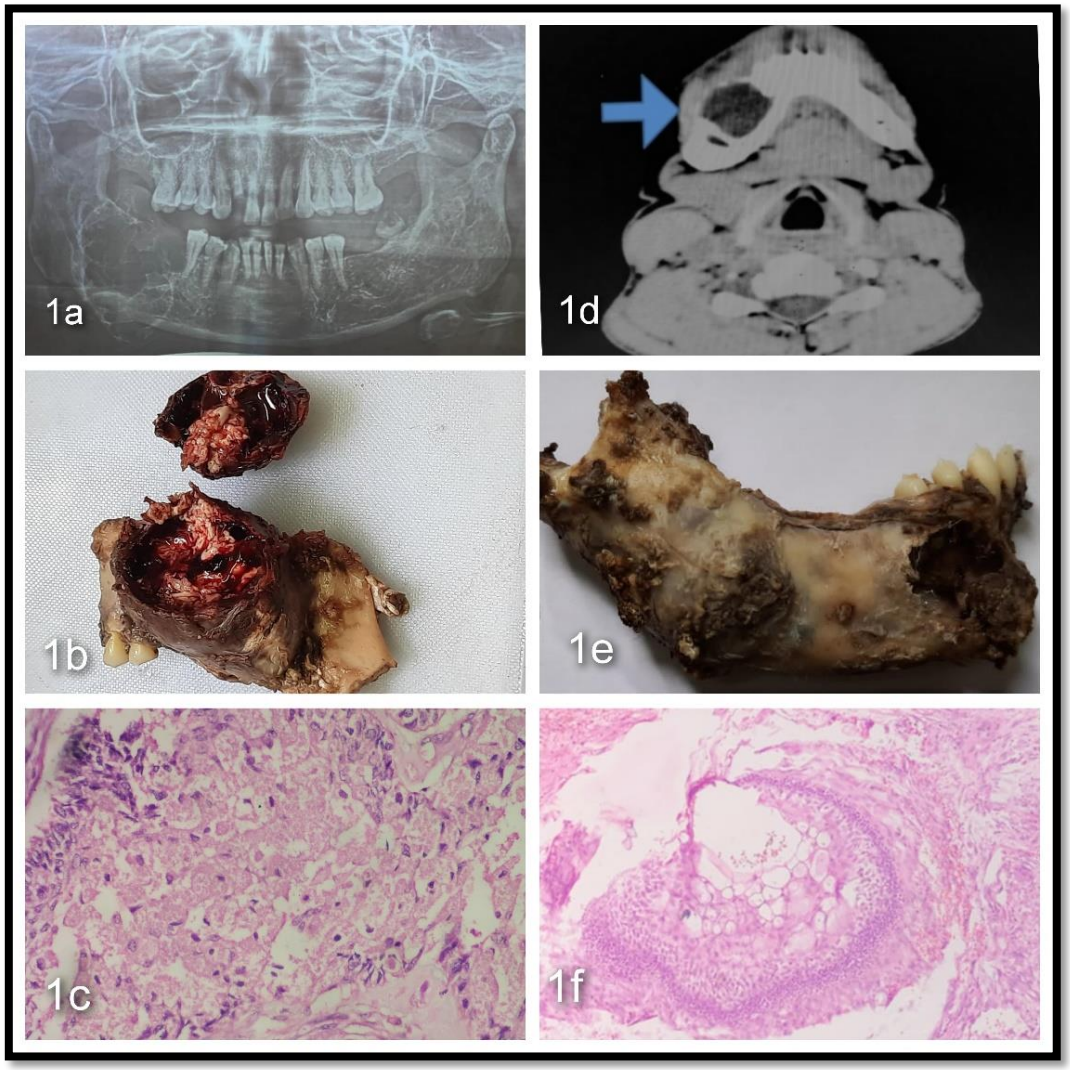

Figure 1: **Case 1 (Figure 1a, 1b, 1c) Granular cell type of ameloblastoma**. *Figure 1a -Case 1 showing well defined heterogeneously hypodense solid cystic lesion with peripheral calcification in the right side of body of mandible with erosion on CT scan; Figure 1b -Case 1 segmental mandibulectomy grossly showing solid cystic lesion; Figure 1c – Case 1 showing predominant granular cell change in Granular cell variant of Ameloblastoma.*
Case 2 (Figure 1d, 1e, 1f) Acanthomatous type of ameloblastoma. *Figure 1d -Case 2 showing well defined enlarged multicystic expansile lesion giving soap bubble appearance in ramus and body of right mandible on CT scan; Figure 1e -Case 2 hemimandibulectomy grossly showing greyish white multicystic lesion in body of the mandible expanding until angle of mandible; Figure 1f – Case 2 showing odontogenic tumor with squamous metaplasia and individual cell keratinisation in Acanthomatous variant of Ameloblastoma.*

Case Report 2
26 year female, came with complaints of swelling in cheek since 3 years, which was insidious in onset and gradually progressive associated with dull aching, non-radiating and continuous pain. NECT brain study showed well defined multicystic expansile lesion giving soap bubble appearance in ramus and body of right mandible, likely suggestive of ameloblastoma. We received right hemimandibulectomy specimen with greyish white multicystic lesion measuring 4.5x1x1 cm in body of the mandible

expanding until angle of mandible. Microscopy revealed odontogenic tumor arranged in strands, islands and isolated cells with squamous metaplasia and individual cell keratinisation. Diagnosis of Acanthomatous type of ameloblastoma was made. (Figure 1d,1e,1f)

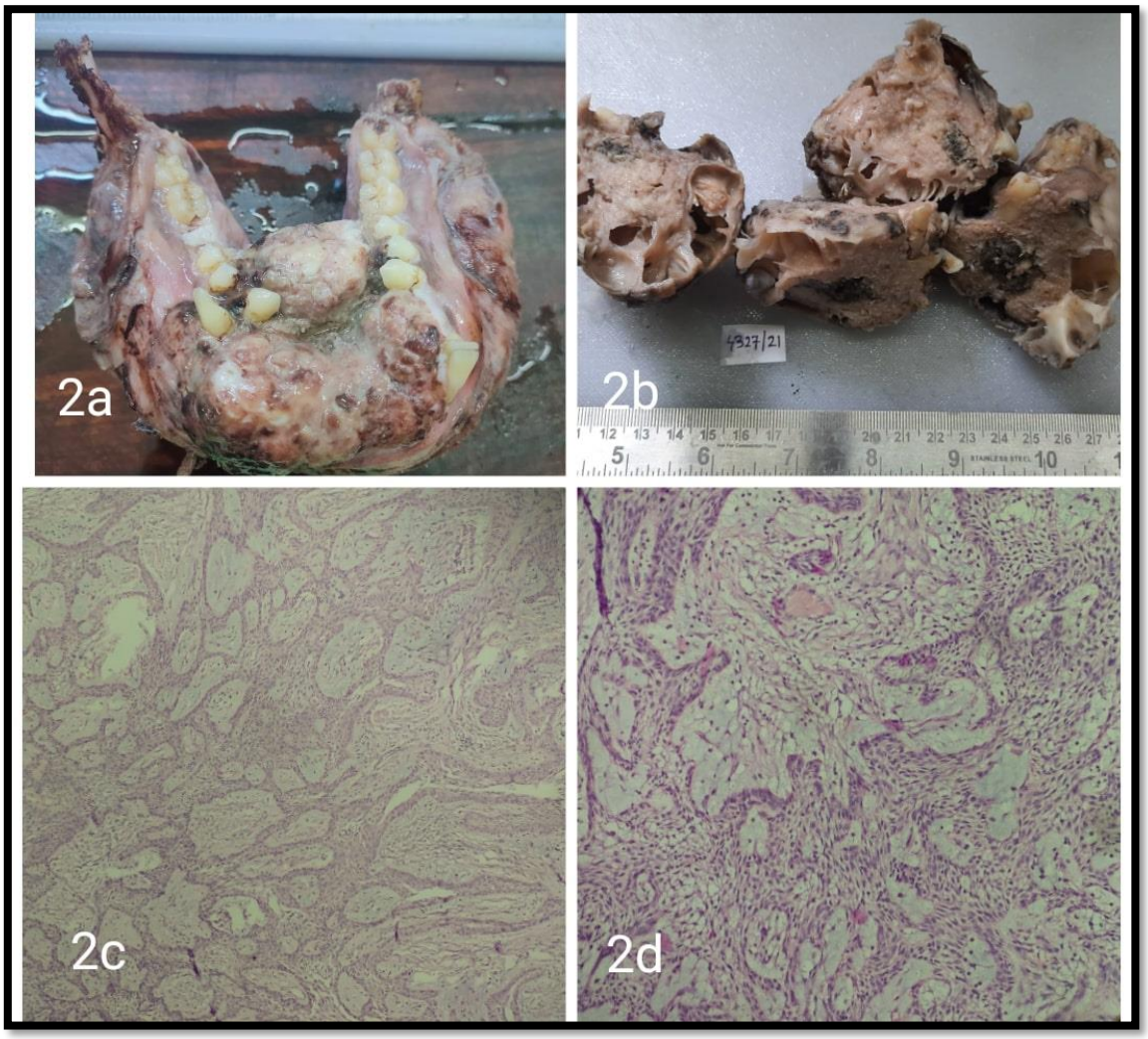

Figure 2: Case 3 (Figure 2a, 2b, 2c, 2d) Plexiform type of ameloblastoma. *Figure 2a and 2b - Case 3 bilateral segmental mandibulectomy showing giant ameloblastoma with multicystic tumor measuring 15x8x7 cm involving body of right and left mandible; Figure 2c and 2d- Plexiform type of Ameloblastoma showing strands and islands of odontogenic epithelium*

Case Report 3

18 years male, came with facial disfigurement and huge gradually increasing swelling of jaw since 4 years involving entire mandibular body till angle of mandible on both sides and measuring 13x10x7 cm. CBCT scan of mandible revealed well defined isodense multilocular expansile lesion in the mandible extending from mesial root of mandibular right second molar till distal root of mandibular left second molar antero-posteriorly. The lesion has caused thinning and expansion of buccal and lingual cortical plate with perforation of cortical plates at many areas. The differential diagnoses given on CBCT were central giant cell granuloma and ameloblastoma. Grossly we received bilateral segmental mandibulectomy specimen measuring 18x10x7 cm. On cutting open, multicystic tumor was identified measuring 15x8x7 cm; involving body of right and left mandible and alveolar region. On histopathology, odontogenic multicystic tumor showing mixed plexiform, acanthomatous and granular cell patterns was noted. There was predominant plexiform pattern with anastomosing strands and

islands of odontogenic epithelium. Diagnosis of Plexiform type of ameloblastoma was given. Patients close follow up was advised in view of local recurrence. (Figure 2a,2b,2c,2d)

DISCUSSION

Ameloblastoma is most often seen in third to fifth decades of life with age range of 10-90 years; in our series age range found to be 18-57 years (Lomi *et al.,* 2018). There are inconsistent results regarding sex predilection in ameloblastoma .Though gender predilection is equal in cases of ameloblastoma; our series showed female predominance with male to female ratio of 1:2. Studies by reddy *et al.,* and Lomi et al showed slight male predominance than females in their case series. (Reddy *et al.,*2015) (Lomi *et al.,*2018). Patients usually complain of a slow growing and painless swelling as seen in our 3 cases. Very large tumors may cause facial deformity, displacement of teeth, malocclusion with restricted mouth opening and airway obstruction. (Suma *et al.,* 2013). Histologically, there can be mixture of 1 or more patterns with predominance of single pattern. We found 3 different histological patterns in our series namely granular cell, acanthomatous and plexiform patterns. In general; solid and multi-cystic types are considered locally aggressive with high recurrence rates as compared to unicystic ameloblastoma which is less aggressive with lower recurrence rate and can respond better to conservative surgery (Neagu *et al.,*2019). Studies showed that plexiform pattern (due to higher levels of matrix metalloproteinases) and follicular pattern had higher rate of recurrence; varied in different studies of literature (Bwambale *et al.,*2022). The treatment includes marginal or segmental resection of mandible with 1cm resection margins. The recurrence rate is usually higher in patients undergoing conservative curettage or enucleation (90-100%) than patients undergoing radical surgery (13-15%) (Chapelle *et al.,* 2004). Giant ameloblastoma can occur in patients who delay treatment due to negligence because of the slow evolution of the tumor and low socio-economic level (Elmrini *et al.,*2021). We presented one case of giant ameloblastoma with size of 15cm in which patient was asymptomatic but presented with facial deformity and tooth malocclusion. Enucleation is inadequate for giant ameloblastoma due to their high rate of recurrence because of inadequately excised tumors. Surgical resection with bony surgical margins of 1 to 2 cm are recommended in these cases (Hughes *et al.,*1999). Annual follow-up usually for 10 years is generally recommended. Our patient right now doing well after 1 year of operation and no recurrence encountered in this patient.

CONCLUSION

Ameloblastoma is the unusual benign odontogenic tumour which is most common in the mandible and maxilla in the middle age group. There are many types of ameloblastoma based on histologic patterns. These morphologic types pose a diagnostic challenge to pathologist while reporting especially on biopsy material. It is always pertinent to consider clinical and radiological parameters which helps in the histopathological diagnosis. As ameloblastoma are locally aggressive tumors they should be diagnosed and treated immediately to avoid further complications. These patients usually need lifelong follow up as these tumors have chances of recurrence.

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