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A Rare Variant of Hybrid Ameloblastoma: A Case Report

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Abstract: Ameloblastoma is a true benign neoplasm of odontogenic epithelial origin. It is the second most common odontogenic neoplasm. It comprises of approximately 1% of all oral tumors and 18% of all odontogenic tumors. Ameloblastomas present with a wide range of clinical and radiological behaviors which makes it the most significant odontogenic neoplasm. There are many histopathological variants of ameloblastoma amongst which hybrid ameloblastoma which is a combination of various histological variants is the rarest type. Up to our knowledge 26 cases have been reported in the literature so far. Generally in hybrid ameloblastoma there is a combination of desmoplastic and follicular variants histologically but sometimes three or more histological variants are also seen in this type. We present a case of 50 year old male patient who presented with a huge mandibular ameloblastoma which was resected along with hemimandibulectomy and reconstructed with iliac crest graft and reconstruction plate. Histopathological examination revealed presence of stromal desmoplsia and osteoplasia with classical follicular islands, granular and acanthomatous differentiation in collision with luminal and intraluminal unicystic ameloblastoma with features of COC ex ameloblastoma.
Keywords: Benign neoplasm, Iliac crest graft, Odontogenic tumor, Reconstruction.

INTRODUCTION

Ameloblastoma is an odontogenic tumor comprising of around 1% of all the cysts and tumors of the jaws. Among the six histolocal variants of ameloblastoma plexiform and folicullar are two most commonly encountered variants accounting to 28.2% and 32.5% respectively; followed by the acanthomatous subtype 12.1% while desmoplastic is extremely uncommon with incidence rates ranging from 4-13% [1]. A recently published biological profile based on 3,677 ameloblastoma cases, has clearly demonstrated that it is not suitable to describe ameloblastoma in any scientific study without specifying the type. Hence, based on clinical features, radiographical appearances, histopathology, and behavioral and prognostic features, subtypes or variants of ameloblastomas can be presently distinguished as follows

1. The classic solid/ multicystic ameloblastoma
2. The unicystic ameloblastoma
3. The peripheral ameloblastoma
4. The desmoplastic ameloblastoma, including the so-called hybrid Lesions [2].

In 1827 Cusak published a report describing what obviously was an ameloblastoma. The age group predilection peaks in the 20 to 30 years. Though Nagata K *et al*. presented a case of ameloblastoma in the mandible of an 82-year-old man [3].

CASE REPORT

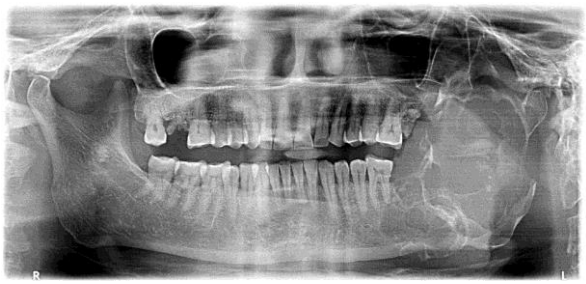
A 50- years-old male patient reported to Department of oral & maxillofacial surgery, Narsinhbhai patel dental college & Hospital, Visnagar with the chief complaint of swelling on the left side of the face since four years. Patient was apparently normal 4 years back, when pain started in lower left back tooth region of jaw which was sudden in onset, dull, intermittent in nature and aggravated while eating food and relieved by taking rest. Pt did not take any medication for that. He visited a local practitioner and underwent extraction of tooth. There was gradual decrease in mouth opening since four years. Extra-oral incision & drainage of swelling was done by general surgeon one year back. Any significant medical history or oral destructive habits were absent. Pt was not under any medication at the time of reporting.
Extra oral examination revealed facial asymmetry due to a well circumscribed swelling present on left middle & lower

1/3rd of the face, measuring approximately about 6×7 cm^2 in diameter, extending anteriorly from left corner of the mouth to posteriorly up to the tragus of the ear and superiorly just beneath the ala-tragus line and inferiorly 1 cm beyond the lower border of the mandible (Fig 1).

Fig–1: Pre-operative profile view

Incisional scar seen over the left side of swelling extending from zygomatic arch to the body of mandible inferiorly. The swelling was non tender on palpation, hard in consistency, skin over the swelling was normal and swelling was fixed to the underlying bony structures. Cervical lymphnodes were non tender and non palpable. Intraorally a diffuse swelling was present in the lower left posterior jaw region measuring 3×2 cm^2 in size approximately, extending anteriorly from buccal sulcus of 36, posteriorly up to retro molar region and ascending ramus region causing the obliteration of the buccal and lingual vestibule.

On radiological examination Orthopantamogram revealed multilocular radiolucency extending from 36 tooth, involving the entire ramus sparing condyle, inferiorly extending beyond the inferior border of the mandible. Distal root resorption of 36 was observed (Fig 2).



Fig–2: Radiolucency extending from body of mandible to condyle

Frontal and lateral view 3D Cone beam computed tomography (CBCT) reconstruction showed a multilocular radiolucency involving the entire ramus by sparing coronoid process and condyle, inferiorly extending beyond the inferior border of the mandible and causing the areas of bone resorption. Inferiorly thinning of the cortical border at the angle of the mandible was also observed (figure. 3 and 4).

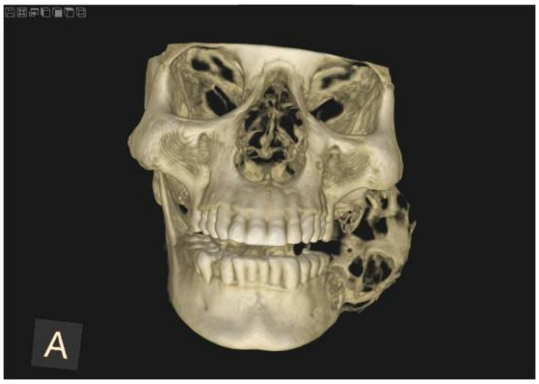


Fig-3: 3-D Computed tomography showing multilocular radiolucency involving entire ramus & destruction of buccal cortices

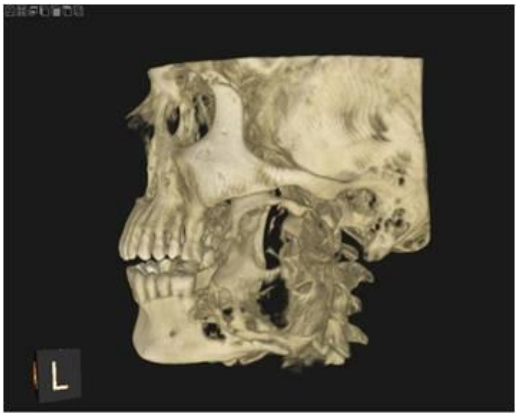


Fig-4: Multilocular radiolucency encroaching infra-temporal fossa

On the basis of clinical examination and radiological examination provisional diagnosis was multilocular ameloblastoma.

Incisional biopsy was performed and reported as ameloblastoma (Figure5).

Fig-5: FNAC shows thin dirty brown colored fluid

Surgical excision of the lesion along with hemimandibulectomy was planned. The patient was taken under general anesthesia for radical excision of the lesion. Extra oral submandibular incision was made

from midline to left angle region 2 cm below lower border. Layer by layer dissection was done to expose the lower border of the mandible. Hemimandibulectomy was performed with long solid tumor mass (12 x 9 cm) extending to infra-temporal fossa was done carefully and reconstruction of the defect was done using anterior iliac crest graft and stabilized with reconstruction titanium plate (Figure 6 7).

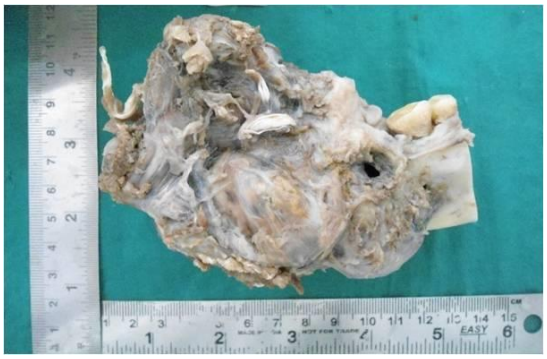


Fig-6: Specimen

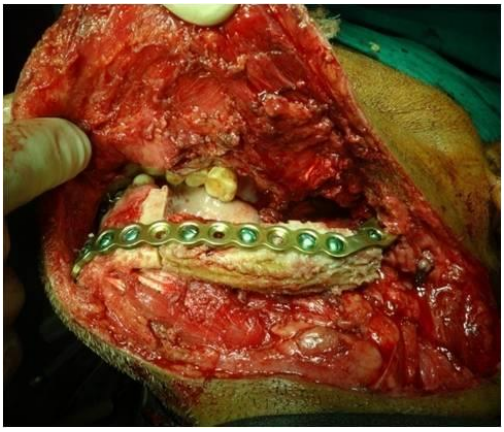


Fig-7: Reconstruction of mandible with iliac crest bone graft and titanium reconstruction plate

Layer wise closure was done after stabilization of iliac crest graft with titanium screws. Specimen was sent for Histopathological examination.

H&E stained section showed small and large ameloblastic odontogenic epithelial islands. Most of the follicular type of ameloblastic islands were lined by tall columnar ameloblast like cells and central granular cells with acidophilic granular cytoplasm. Few areas of central stellate reticulum like cells showed squamous metaplasia indicating acanthomatous differentiation. The luminal, intraluminal and intramural proliferation of the follicles were observed. There was presence of peripheral cystic lining showing large number of ghost cells simulating COC. Stroma showed desmoplasia along with marked osteoclastic activity. Few stromal areas showed large amount of blood vessels and numerous giant cells in vicinity. Multiple occurrence of the histologic features suggested Hybrid ameloblastma ex COC.

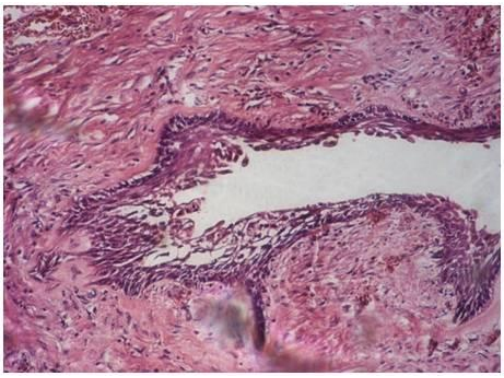


Fig-8A: Photo micrographs of H &E stained section shows Follicular type of islands

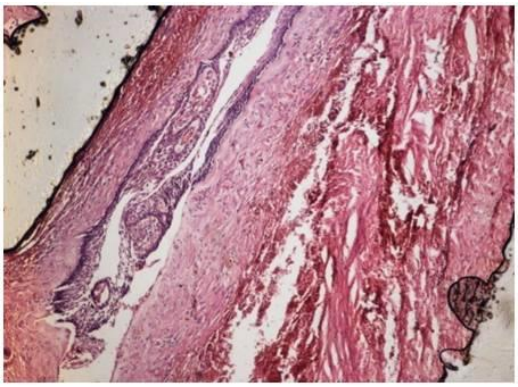


Fig-8B: Photo micrographs of H &E stained section shows Squamous metaplasia suggestive of acanthomatous transformation

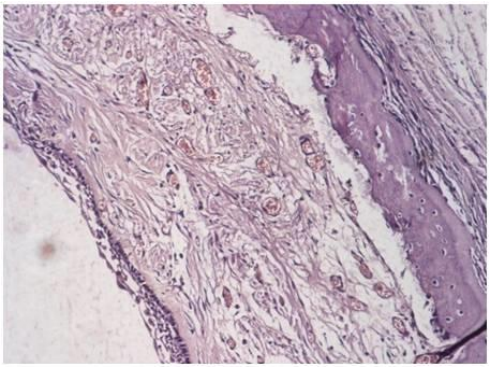


Fig-8C: Photo micrographs of H &E stained section shows Compressed ameloblastic follicle due to desmoplsia

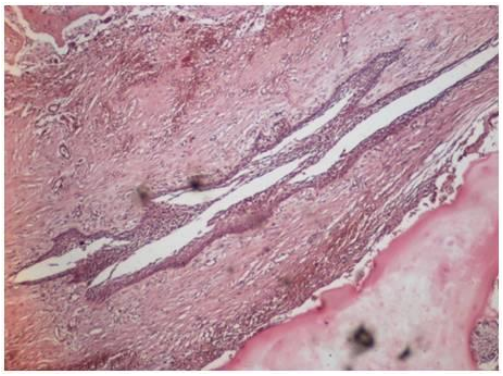


Fig-8D: Photo micrographs of H &E stained section shows Large ameloblastic follicle granular cells showing eosinophillic cytoplasmic granules

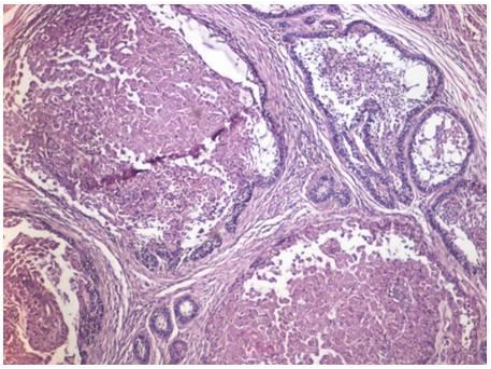


Fig-8E: Photo micrographs of H &E stained section shows Luminal unicystic lining with & Osteoid area

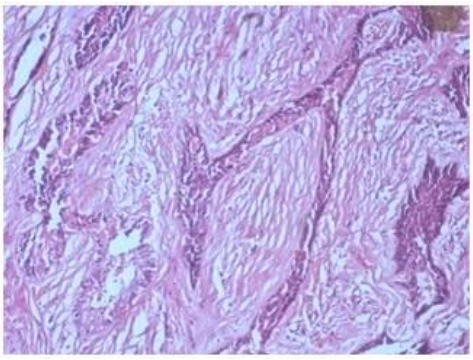


Fig-8F: Photo micrographs of H &E stained section shows Intraluminal proliferation of the cystic lining

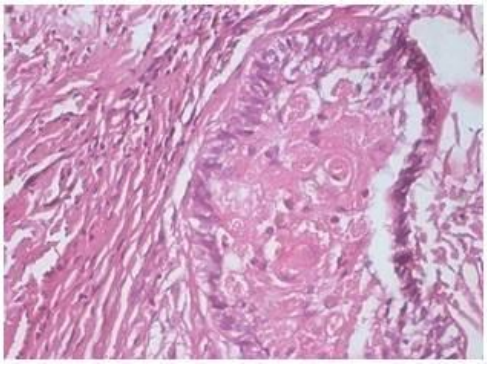


Fig-8G: Photo micrographs of H &E stained section shows Intraluminal unicystic lining

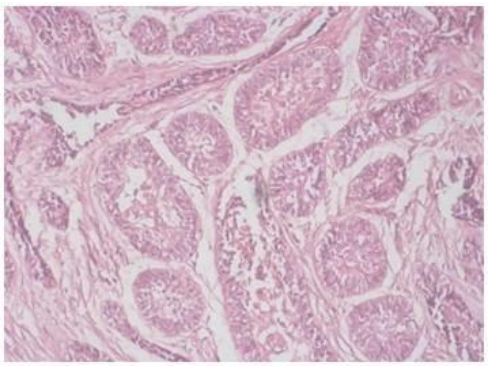


Fig-8H: Photo micrographs of H &E stained section shows Intramural proliferation of the unicystic lining

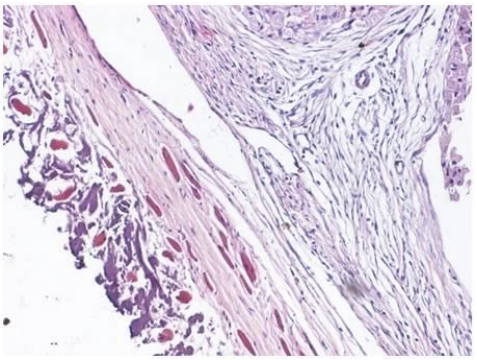


Fig-8I: Photo micrographs of H &E stained section shows Lining showing ghost cells typical of COC

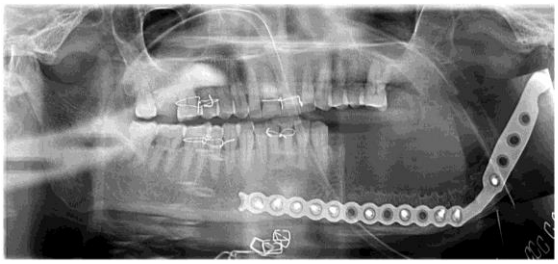


Fig-9: POST-OP OPG

DISCUSSION

The present case report is of a 50-year-old male patient with a classical clinical presentation of the lesion in the left posterior region of the mandible to be of ameloblastoma.

Radiographically, in most cases, ameloblastomas present characteristic but not diagnostic features. A unilocular or multilocular radiolucency with soap bubble or a honeycomb appearance is the most striking feature [4]. Ameloblastoma is an osteolytic lesion and does not produce mineralized components except in rare cases [5]. The present case showed multilocular radiolucency in 3D CT scan imaging & OPG with considerable bone resorption involving entire ramus extending to the coronoid process. Panoramic view also showed resorption of the root irt 36 which is not an uncommon finding regarding ameloblastomatous lesion.

Vickers and Gorlin in 1970 defined the histopathological features of ameloblastoma. Most of them present variable range of patterns. These patterns may include follicular, plexiform, granular cell, desmoplastic, acanthomatous, basal cell type, and unicystic types. In a large case study it was suggested that individually these pattern comprises 28.2%, 32.5%, 4.28%, 4-13%, 12.1%, 2.02%, and 6% of cases included respectively [6]. The present case showed histological picture with more than one type of histologic variant. Granular cell variant was predominant along with areas of follicular subtypes, acanthomatous differentiation, luminal intraluminal and intramural unicystic types with the areas showing desmoplasia and osteoplasia. Some areas showed typical lining resembling Calcifying odontogenic cyst (COC) with the presence of ghost cells which rendered the lesion direction of being ameloblstoma ex COC. It was added to the cases earlier reported with more than one histologic pattern. In the study conducted by Fulco GM *et al* [7] 75.5% of the solid lesions included were made up by more than one histological patterns. Similarly, Nonetheless, only 16.1% and 19.7% of the solid lesions evaluated by Reichart *et al* [8, 9] and Kim and Jang, respectively, showed more than one histological type. According to Adebiyi et al. ameloblastomas, especially the large ones, are made up of numerous histological patterns [1].

Waldron and El-Mofty first described the hybrid ameloblastoma (HA) as an individual histologic variant showing follicular or plexiform pattern coexisting with areas of characteristic desmoplastic ameloblastoma (DA) [10]. It was speculated that the desmoplastic changes occur secondarily in the stroma of preexisting Solid/Multicystic ameloblastoma. It has been suggested that the hybrid tumor should be considered a collision tumor. As rare incident Siar CH *et al*. presented a case which is an example of radicular cysts and ameloblastoma occurring as a collision phenomenon [11].

The preferred treatment modality for ameloblastoma is surgical exicision of the lesion along with free margin due to its high reccurence rate. So in the present case wide resection of the lesion followed by reconsruction was carried out. Long term follow-up is necessary for determination of recurrence of the lesion. The present case was followed-up for 24 months without any signs of recurrence. The final esthetic result was satisfactory.

CONCLUSION

Hybrid lesion in ameloblastoma is a proven histopathological variant. Usually such lesion is histologically present with desmoplasia with follicular, acanthomatous and basaloid patterns. Additionally, the present case showed granular cell type pattern, osteoplasia and unicystic lining with luminal, intraluminal and intramural proliferation which is not common. Wide Surgical resection of the lesion with normal margin along with reconstruction of surgical defect can yield best esthetic and functional outcomes as shown in this case.

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