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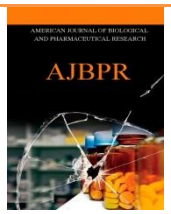

SOLID AND MULTICYSTIC FOLLICULAR AMELOBLASTOMA - A CASE REPORT

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ABSTRACT

Ameloblastoma is a slow-growing benign neoplasm that has a strong tendency to local invasion and that can grow to be quite large without metastasizing. Rare examples of distant metastasis of an ameloblastoma in lungs or regional lymph nodes do exist. Greatest period of prevalence is in the age range of 20 to 50 years. The majority of them occur in the mandible, and over two-thirds occur in the molar and ramus area. Microscopically, follicular, plexiform, and acanthomatous are the histological variants in which the appearances of basal cells, stellate reticulum (with varying degrees of cystic degeneration), and squamous metaplasia are reproduced. Here we are presenting a case of solid and multicystic follicular ameloblastoma in a 24yr old male patient.

INTRODUCTION

Ameloblastomas are benign tumors whose importance lies in its potential to grow into enormous size with resulting bone deformity. They are typically classified as unicystic, multicystic, peripheral and malignant subtypes [1]. The most common tumor of odontogenic origin is ameloblastoma which develops from epithelial cellular elements and dental tissues in their various phases of development. More than 80% of all ameloblastomas are solid or multicystic variants, with unicystic ameloblastoma being an important clinicopathologic form of ameloblastoma and occupying the remaining 20% of the cases along with peripheral ameloblastoma. Occasionally an ameloblastoma forms from the epithelial lining of a dentigerous cyst and this is called a *mural (within the wall) ameloblastoma* [2].

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This paper illustrates a case of follicular ameloblastoma in a 24yr old male.

Case Report

A 24yr old male patient had reported to the outpatient department of the hospital with a chief complaint of pain and swelling in relation to the left lower posterior region of jaw since 8-10 days (Figure 1). Patient was apparently well 8-10 back and noticed pain and swelling in left lower posterior region and also reports of paresthesia in the same region. Pain was dull in nature and aggravated on chewing food which subsided itself. Pain was not associated with fever and no medication was taken. Patient had a habit of chewing tobacco since for 3-4 times per day since 4years. On extra oral examination there was no facial asymmetry. The lymph nodes were non palpable. Intra oral examination on inspection: revealed an ulcerative growth in lower left molar region i.e. 36 and 37 associated with pseudomembrane in the region of 38. The growth was extending to the lingual vestibule in the region of 36 and 37 and involving the retromolar area of size

5x2cm. Swelling in buccal vestibule in the region of 38 was also present (Figure 2).

On palpation: It was soft to firm on palp7ation and was tender on palpation. Expansion of the lingual cortical plate and buccal cortical plate was present. No discharge was seen.

INVESTIGATIONS

IOPA radiograph (Figure 3) revealed tooth like radiopacity in the apical third region of 37 impinging on the inferior alveolar canal. Mandibular occlusal (Figure 4) radiograph revealed radiolucency involving 38 extending anteriorly upto mesial aspect of 37 causing expansion of lingual cortical plate. OPG radiograph (Figure 5) revealed a well-defined radiolucency in the left lower posterior region of size 5x5cm extending anteriorly from apical third of 37 and posteriorly upto the anterior part of the ramus of the mandible. Bowing down of radiolucency causing thinning of inferior border of the mandible was also seen. Tooth like radiopacity is seen within the radiolucent region in apical third of 37. CT image (Figure 6) revealed radiolucency in left posterior region of size 5x5cm.

Expansion of lingual and buccal cortical plates is seen. After radiographic examination a differential diagnosis of *dentigerous cyst, odontogenic keratocyst, unicystic ameloblastoma, odontogenic myxoma* was given.

Preoperative Biopsy was done and microscopic examination of H & E stained section shows follicular tissue with few areas of odontogenic epithelium. Lesional tissue shows follicles of consisting of tall columnar peripheral ameloblast like cells and central cells resembling stellate reticulum. Cystic space on coronoid area shows ameloblast linning with areas of solid tumors growth. Overall features were suggestive of solid and multicystic follicular ameloblastoma.

Treatment

The lesion was surgically excised, left hemimandibulectomy was done, the patients' appearance was reasonably restored (Figure 7). Intra- orally mandibular jaw bone was stabilized with reconstruction plates as seen in the radiograph (Figure 8).

POSTOPERATIVE BIOPSY (FIGURE 9)

H&E stained section shows lesional tissue composed of cords and follicles of odontogenic epithelium with diagnostic tall columnar cells resembling at periphery and central stellate reticulum like cells suggestive of follicular amelobalastoma. Cystic space on the coronoid area shows ameloblast like linning with areas of solid tumors growth.

Figure 1. Preoperative Profile Photograph showing swelling on left side of jaw

Figure 2. Intra Oral Photograph

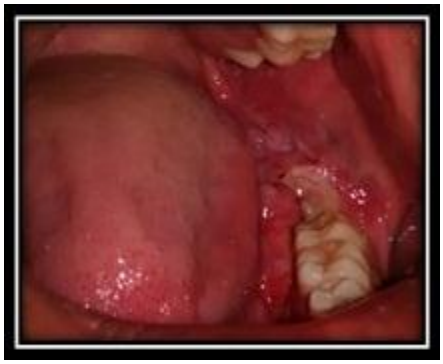


Figure 3. IOPA showing tooth like radiopacity in apical third region of 37 and impinging on the mandibular canal

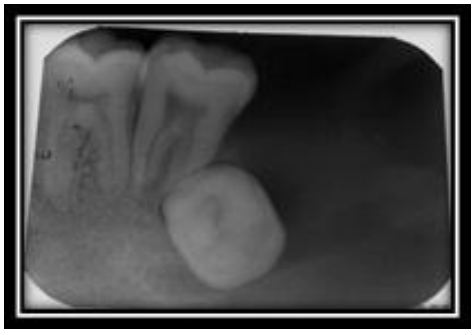


Figure 4. Mandibular occlusal radiograph showing radiolucency in molar region and causing expansion of lingual cortical plate

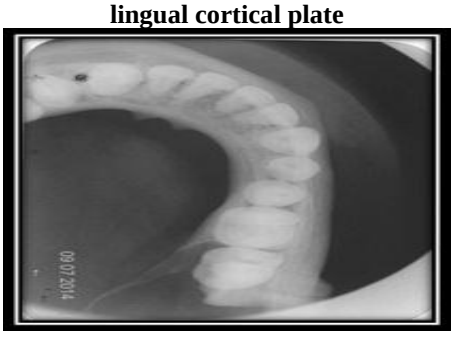


Figure 5. OPG showing well defined radiolucency which is seen extending from 36 to the ascending part of ramus. Bowing down and thinning of inferior border of mandible

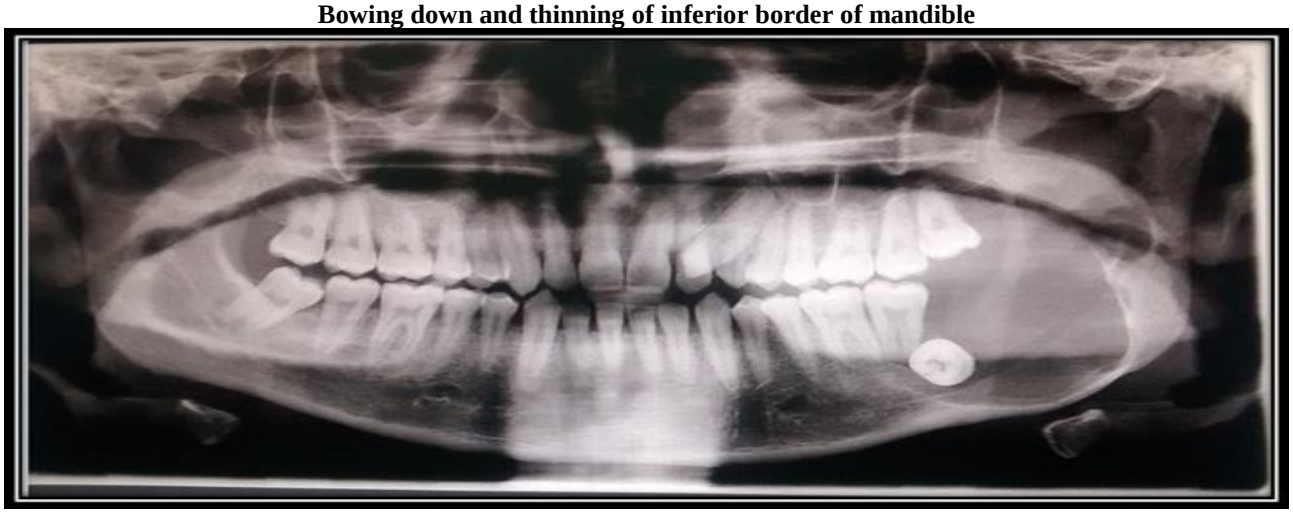


Figure 6. CT image showing the entire extent of the lesion with its greatest diameter of about 5cm. Expansion of lingual and cortical plates can be seen

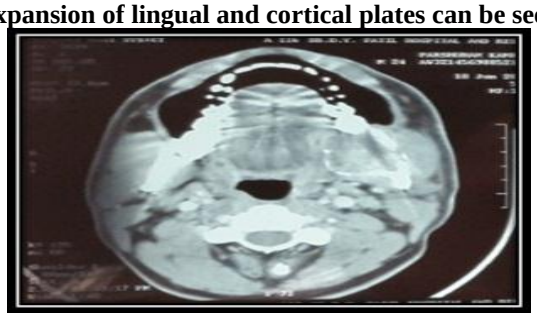


Figure 7. Postoperative profile photograph

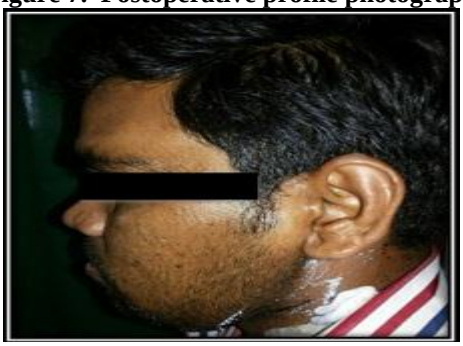


Figure 8. Post operative OPG , showing bone plates stabilizing mandible

Figure 9. Microscopic view with cystic lining showing stellate reticulum like cells

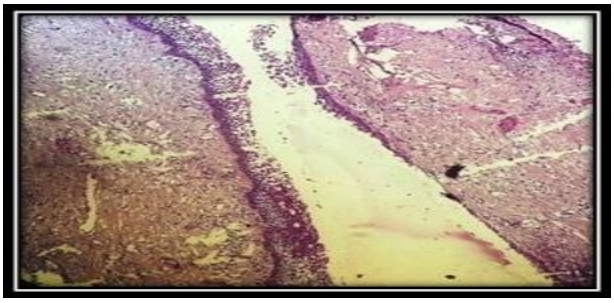
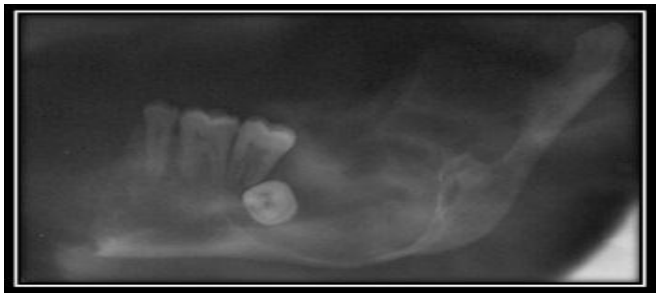


Figure 9. Radiograph of the specimen showing multicystic cavities



DISCUSSION

Ameloblastoma is a benign epithelial odontogenic tumor but is often aggressive and destructive, with the capacity to attain great size, erode bone and invade adjacent structures. Although the term ameloblastoma was coined by Churchill in 1933, the first detailed description of this lesion was by Falkson in 18797. It is the most common odontogenic tumor although it represents only about 1% of tumors and cysts of the jaws.

In the mandible (80% of ameloblastomas), 70% are located in the area of the molars or the ascending ramus, 20% in the premolar region, and 10% in the anterior region. About 10-15% of ameloblastomas are associated with a non-erupted tooth [3].

Ameloblastomas are rare in children; the greatest period of prevalence is in the age range of 20 to 50 years. Ameloblastomas occur with equal frequency in both sexes [4]. The majority of them occur in the mandible, and over two-thirds occur in the molar and ramus area. Cystic expansion can lead to parasthesia, root resorption and asymmetry of the face. Curettage of the unilocular or multilocular lesion is often followed by local recurrence, and block excision of the lesion with a good margin of unaffected bone is the treatment of choice and is rarely followed by recurrence [5]

In the present case, a large follicular ameloblastoma was found in the molar region and involving ascending ramus in left mandible. It was associated with non-erupted tooth and did not show any signs of egg shell crackling. Clinically, ameloblastoma frequently manifests as a painless swelling, which can be accompanied by facial deformity, malocclusion, ulceration and periodontal disease and paraesthesia of the affected area. In our case, clinical examination revealed a large, expansive mass in the ascending ramus and molar region of the mandible.

Radiologically, the lesions are expansible, with thinning of both buccal and lingual cortical plate. The lesions are classically multilocular with cystic with a soap-bubble or honey comb appearance. Occassionaly CT images reveal unilocular ameloblastomas resembling dentigerous cyst or odontogenic keratocyst.

There are seven histological types of ameloblastoma: follicular, plexiform, acanthomatous, granular cell, desmoplastic, basal cell, and unicystic variant, with the first two being the most common. The most common symptoms are facial swelling, pain, malocclusion, loosening of teeth, ill-fitting dentures, periodontal diseases or ulceration, oroantral fistulas and nasal airway obstruction [6].

Ameloblastoma can be either solid or multicystic, but they frequently demonstrate both characteristics. Although the majority of the tumours originate from within the maxilla or mandible, they can also be peripheral. The different histological variants do not significantly alter treatment considerations except for the unicystic and the peripheral types, which can typically treated with enucleation and curettage. The multicystic ameloblastoma has a recurrence up to 50% during the first 5 years postoperatively so long term follow up is must [7].

It has been postulated that the epithelium of origin is derived from one of the following sources:
(1) Cell rests of the enamel organ,
(2) Epithelium of odontogenic cysts
(3) Disturbances of the developing enamel organ,
(4) Basal cells of the surface epithelium or
(5) Heterotropic epithelium in other parts of the body.

Follicular pattern simulates the developing dental follicle and the enamel organ by arranging the epithelial cells to resemble stellate reticulum.

Lucas and Thackray (1952) attribute the formation of intrafollicular cystic cavities to a deficiency in absorption and diffusion of nutritive elements (coming from the perifollicular blood capillaries) to the centre of the cellular islands, causing their degeneration by nutritive insufficiency, since the neoplastic growth causes extremely large follicles. However, this same central degeneration could have been caused by the polarization of the nuclei at the cellular end facing the stellate reticulum. This probably causes the cells of the peripheral layer of the follicles to remove nutritive elements from the interior of these cellular islands and not from the connective tissue facing the other cellular extremity. This nutritive competition can cause metabolic deficiencies for the cells of the stellate reticulum, which can explain the degeneration of the central cells of the islands and the consequent formation of cystic cavities in its interior [8].

Treatment of ameloblastoma varies from curettage to en bloc resection. Bone grafts replace the surgically removed bone, with autologous bone grafting being the most desirable. The most commonly used grafting material is for alveolar ridge reconstruction is free autogenous iliac bone. However autologous calvarial bone grafts can also be used. It cannot be used in case of thickness of calvarial bone of less than 5mm [9].

CONCLUSION

Currently histologic examination is the most sensitive tool for differentiating ameloblastoma from odontogenic cysts. Careful examination of the whole specimen is essential with multiple sectioning. Thus, lesions which clinically and radiographically appear to be odontogenic cysts may prove to be ameloblastomas. The ability to predict this potential occurrence prior to surgery would greatly enhance therapeutic strategies for reducing the incidence [10]. A multidisciplinary approach, including oral surgery, orthodontics, and prosthodontics is able to provide a patient diagnosed with follicular ameloblastoma.

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