CASE REPORT

MULTICYSTIC FOLLICULAR AMELOBLASTOMA MIMICKNG LATERAL PERIODONTAL CYST- A RARE CASE REPORT

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ABSTRACT:
Ameloblastoma is a polymorphic neoplasm consisting of proliferating odontogenic epithelium which usually has a follicular or plexiform histologic pattern lying in fibrous stroma, commonly affects molar ramus areas of mandible and clinically classified into four types commonest being multicystic. We report a case of multicystic follicular ameloblastoma, clinicoradiographically suggestive of lateral periodontal cyst in a 40 year old woman who presented to us with solitary swelling in right canine premolar region, following biopsy histopathological examination revealed follicular ameloblastoma, further marginal resection of tumor was performed under general anesthesia. Another peculiar finding which was accidental in OPG of this patient was a circumscribed radiopaque mass in missing left maxillary third molar area suggestive of odontoma.
Key words: Multicystic follicular ameloblastoma, Neoplasm, Odontogenic epithelium.

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INTRODUCTION

Ameloblastoma is a epithelial odontogenic neoplasm that frequently affects the molar and ramus area of mandible.[1] Its relative frequency equals the combined frequency of all other odontogenic tumors excluding odontomas,[2] theoretically they may arise from rests of dental lanina , developing enamel organ, HERS, epithelial lining of odontogenic cysts or basal cells of oral mucosa; nothing is known about the exact cause and the reliable microscopic criteria for its early diagnosis have yet to be defined.[3] Although ameloblastomas generally are not classified as a malignant lesion it is extremely aggressive and infiltrative many have suggested that this lesion should be considered as a low grade or indolent malignancy.[4] They appear in four different clinicoradiographic situations having different therapeutic considerations and prognosis.

1. Solid/Multicystic
2. Peripheral
3. Desmoplastic
4. Unicystic.[4]

Radiographically typical ameloblastomas have a multilocular or unilocular radiolucent apperance with fairly well defined margins, adjacent teeth may be tilted or displaced and root resorption is commonly observed.[5] Histologically many variants have been described commonest form is follicular ameloblastoma followed by plexiform it is not necessary to be conserned with the histologic patterns exhibited by the lesion because these various histologic patterns have no bearing on the biologic behavior of the tumor.[6] Odontomas are the most frequently occurring odontogenic tumors;[7] they are composed of both epithelial and mesenchymal tissues which appear normal but have a deficit in structural arrangement. The level of differentiation may vary, creating various formations of dental tissues.[8] This report presents the case of a patient who was provisionally diagnosed with lateral periodontal cyst in lower right canine premolar area

and odontoma in upper left third molar area based on clinical and radiographic features before the final diagnosis of multicystic follicular type ameloblastoma was made based on histopathological features of lower lesion, we couldn't arrive at the final diagnosis of upper lesion since patient refused to take the treatment for asymptomatic upper lesion.

CASE REPORT:

A forty years old female presented to our hospital with a chief complaint of swelling in the lower right front tooth region for about two years. Initially it was peanut sized and no symptoms were present so patient didn't seek any medical aid, later it increased gradually to the present size. Intraorally a solitary painless swelling was noticed in lingual side between right mandibular canine and first premolar measuring about 2 x 2 cms extending from mesial aspect of canine to mesial aspect of first molar, swelling had well defined boundary, mucosa covering the swelling was healthy, and bone like hard in consistency, first premolar was migrated distally and both the premolars were tender on percussion with nonvital pulp in 2nd premolar and no sensory paralysis was noted (Figure :1).

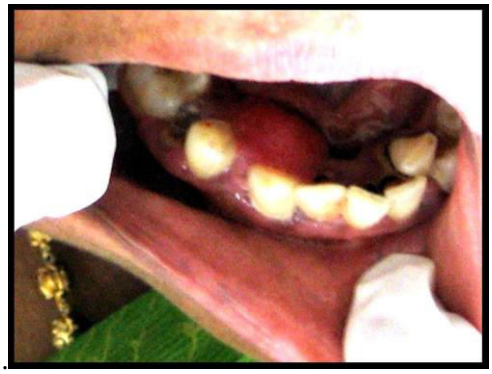

Figure 1: Extra oral photograph of patient

Patient's oral hygiene was poor, she had generalized bleeding on probing, gingival inflammation and shallow pockets, multiple root stumps were present in posterior regions of both the arches.

Intra oral radiographs showed a radiolucent area between canine and first premolar measuring about 1x1.5 cms with relatively distinct borders, expansion of the lesion had caused significant displacement of the first premolar distally. Dental caries was present in 2nd premolar causing gross destruction of tooth, where as in 1st premolar and canine neither caries nor root resorption was noted. Apart from this one more accidental finding observed in the OPG was a well circumscribed radiopaque mass surrounded by a thin radiolucent rim measuring about 2.5 cms in diameter in missing upper left third molar region. (Figure 2)

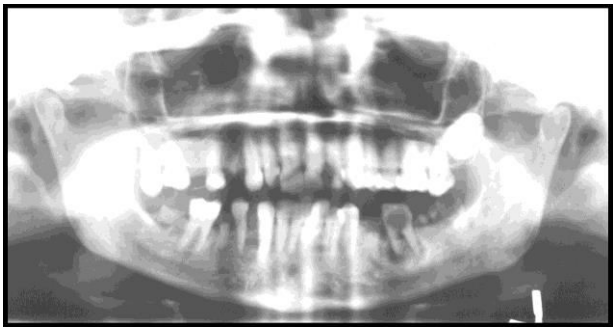

Figure 2: OPG exhibiting lower radiolucency in right canine premolar area and upper radiopacity in right third molar area

Based on the above findings the lower lesion was provisionally diagnosed as a lateral periodontal cyst with the following differential diagnosis Odontogenic keratocyst, Early calcifying odontogenic cyst, Simple bone cyst, Unicystic ameloblastoma and upper lesion was diagnosed as complex odontoma, since patient was not prepared for the treatment of odontoma it was not excised. Results of preoperative biopsy suggested follicular ameloblastoma, subsequently marginal resection of the lesion was performed under general anesthesia, the excised specimen was mixture of hard and soft tissue measuring about 2.5 X2.0X2.0cms, grayish brown in color with few cystic spaces with in the mass, along with two premolars and canine, further the specimen was decalcified with the help of 5% formic and 5% nitric acid, processed routinely and stained with H & E. Microscopy exposed similar features as that of preoperative biopsy; numerous follicles of odontogenic epithelium surrounded by moderate degree of inflammatory cell infiltrate predominantly lymphocytes. The follicles showed peripherally arranged tall columnar cells with the central core of stellate reticulum like cells, nuclei of columnar cells located away from the basement membrane, the central zones of some follicles exhibited foci of cystic degeneration and keratin pearl formation. Numerous extravasated RBCs and blood capillaries were also observed suggestive of Follicular Ameloblastoma. (Figure 3,4 and 5)

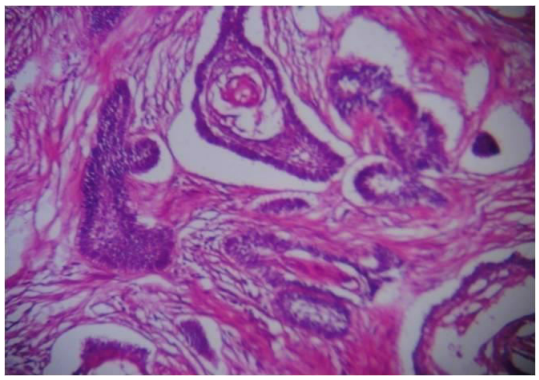

Figure 3: Photomicrograph showing follicles of ameloblastoma in mature fibrous connective tissue.

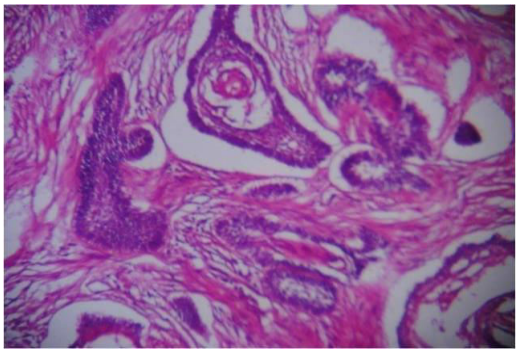

Figure 4: Photomicrograph showing follicles with connective tissue

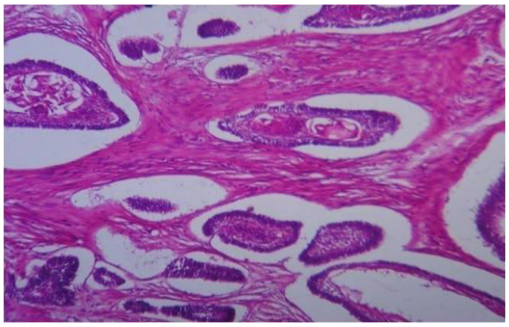

Figure 5: Photomicrograph demonstrating single layer of ameloblast like cells in the follicles showing Vickers & gorlins criteria

DISCUSSION

Ameloblastoma is known from 180 years, after the first case reported by Cusak in 1827. Broca in 1868 is the first person to give the detailed description and till date four clinicoradiographic variants have been identified among which solid/multicystic variant is commonest which was present in the present case. In 2003 WHO classification this lesion has been classified under benign tumors arising from odntogenic epithelium with mature fibrous stroma; odontogenic ectomesenchyme no present.[4]

Although no sex differences exists in the onset of this disease people between ages of 30 & 40 are most commonly affected[1]as in this case. Location of multicystic ameloblastoma in canine premolar region as in this present case have been reported in about 7.7% of cases in a study by Reichart et al, [4] location of tumor in jaws plays a very important role in prognosis; further the tumor is from vital structures less likely is to infiltrate them so ameloblastomas of body of mandible as in our case as well as in anterior maxilla are less dangerous than those of ascending ramus and posterior maxilla. Death has been reported from intracranial extensions.[3]

The radiographic presentation of solid/multicystic ameloblastoma is not always pathgnomic, but is frequently suggestive mostly present as cystic lesions which are unilocular in 47% of cases, multilocular in 37% and 16% soap bubble appearance in molar ramus areas[4] with or without bony expansions and root resorption.[5] In the present case root resorption was not observed and location was not where it is usually seen which lead us to think in different terms mostly as lateral periodontal cyst.

Lateral periodontal cyst is usually discovered during routine radiographic examination, is located mainly between the roots of vital mandibular canines and premolars, and seldom causes pain or other clinical symptoms. The defect appears on radiographs as a round or teardrop-shaped, well circumscribed radiolucency,[8] most of the features presented in our case were simulating these, such as canine and premolar region, well defined radiographic boundaries, vital teeth in the region of swelling and absence of root resorption were against multicystic ameloblastoma. Histopathology of solid/multicystic ameloblasoma closely resembles enamel organ and varies greatly, number of histological types have been described in this variant such as follicular, plexiform, granular, acanthomatous, clear cell variant, basal cell variant,[9] of these most commonest is follicular type. This shows histologically islands of epithelium resembling enamel organ epithelium in mature fibrous connective tissue stroma, the epithelial rests consists of a core of loosely arranged angular cells resembling stellate reticulum of enamel organ. A single layer of tall columnar cells surrounds central core and fulfilling Vickers &

gorlin criteria[2] all these classical features were evident in the present case. Although all features were evident of a multicystic follicular ameloblastoma the reports of this type in canine premolar region has been reported infrequently, since this region is commonly affected by desmoplastic variant unlike the present case.[4]

Even though the origin of ameloblastoma s are thought to be from dental lamina, developing enamel organ, HERS, epithelial lining of odontogenic cysts or basal cells of oral mucosa exact cause is not known either in humans or animals because experimental induction of ameloblastomas has not been successful and has not helped clarify the problem.[3] In this present case the origin could be probably from the cell rests of mallasez or could be from basal cells of epithelium since it was located in alveolar bone between teeth. The optimal method of treatment has a subject of controversy for many years. The conventional ameloblastoma tends to infiltrate between intact cancellous bone trabaculae at periphery of lesion before bone resorption becomes radiographically evident therefore the actual margin of tumor often extends beyond its apparent radiographic or clinical margin. Recurrance rates of 50 to 90% have reported in various studies after curettage and upto 15 % after marginal resection. So, later way is the most widely accepted treatment.[2] In this case marginal resection was carried out and till date i.e. after about one and half year of treatment no recurrance has been reported. However this short period of time doesn't indicate cure since recurrance often takes many year to become evident. Hence it has to be monitored regularly. Unlike ameloblastomas which does not undergo differentiation till enamel formation.[9] Odontoma is a odontogenic tumor which consists of enamel, dentin, pulp and occasionally cementum and appears radio dense surrounded by radiolucent rim in radiographs and most of the times it is an accidental finding like in this case.[7] Since patient didn't allow for excision of odontoma, we couldn't arrive at the final diagnosis of this lesion due to the lack of histopathology, since amleoblastic odontoma (odontoameloblastoma) also shows the radiographic features similar to that of complex odontomas.[9]

The unique feature of these two lesions in the present case is that we have never come across any case affected by both ameloblastoma and odontoma or ameloblastoma and odontoameloblastoma as discrete neoplasms. Ameloblastoma has been reported to occur in association with odontogenic cysts and other non odontogenic lesions[10] such as dentigerous cyst, radicular cyst, [11] calcified odontogenic cysts,[12] pindborg's tumor[10] on the other hand odontomas are known to present in association with calcified odontogenic cysts [14-16] and in gardners syndrome.[2,9,17] However gardner's syndrome which is known to present with odontomas have been reported by Patel H et al, in association with ameloblastoma,[17] but in our case none of the other features of gardner's syndrome were noticed. Since patient did not allow for excision of odontoma we could not arrive at the final diagnosis of odontoma.

CONCLUSION

Radiographic presentation of ameloblastoma is not always pathgnomic but is frequently suggestive most present as cystic lesions with either a unilocular or multilocular pattern with or without bony expansion but final diagnosis is incomplete without histopathology hence preoperative biopsy should be advised to reach final diagnosis and to plan appropriate treatment as for as relationship of ameloblastoma and odontoma is concerned coming to a conclusion without histopathology / final diagnosis on odontoma will be incorrect at this stage. If at all we consider this lesion to be a odontoma/odontoameloblastoma then these two tumors in two opposite sides of either jaws could be a co-incidence or might be some undescribed syndrome which awaits for further reports.

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