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Unicystic Ameloblastoma of the mandible – A rare case report

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Abstract

Solid or multicystic ameloblastomas accounts for more than 80% whereas unicystic ameloblastoma and peripheral types forms the remaining 20%. The term unicystic ameloblastoma was coined by Ackermann et al. in 1988. The cystic lesions showing clinical, radiographical or gross picture of cyst of jaw bones but microscopically demonstrating a classical ameloblastomatous epithelium lining the cyst cavity, with or without luminal and/or mural tumor proliferation are unicystic ameloblastomas. This article highlights the surgical management of such unicystic ameloblastoma in a 50 year female.

Keywords: Unicystic ameloblastoma, Bicortical expansion, Surgical management

Introduction

Ameloblastoma is a benign neoplasm mainly made up of epithelial tissue which has an invasive and infiltrative nature locally with a high recurrence rate.[1] If left untreated, they are able to reach large sizes, causing facial disfiguration and functional problems. No sex predilection is noticed with increased incidence around 30 and 40 years.[2,3]

Newer WHO simplified version of classification suggests three types: conventional, unicystic and peripheral.[4] Unicystic ameloblastoma's presentation is similar to that of a cyst than conventional one. It accounts for 5-15% of all intraosseous ameloblastomas. UA is seen in young patients contrast to the conventional one which is seen in

middle age group. Radiographically it is unilocular but conventional presents as multilocular.[5]

Surgical management is the main mode of treatment based upon the clinical presentation. It varies from conservative management like segmental or marginal resection to complete resection of it if it is larger in size. But conventional treatment has been associated with the complications such as functional or esthetic deformities.[6] Hereby we are presenting a case of unicystic ameloblastoma with complete surgical management in a 50 year old female.

Case report

A 50 year old female patient reported to our hospital with the complaint of swelling in the left lower side of the face since 3 months. She had visited to the nearby dental clinic and was under antibiotics after which the swelling was gradually reduced in the size but not fully recovered. Medical, family histories were non-contributory. On extra oral examination, solitary firm and hard swelling noticed on the left lower third of the face measuring approximately 3X2cms anteroposteriorly in the lower border of the mandible. No pus discharge or surface changes noticed. Left submandibular lymphadenopathy was present. Intra oral examination revealed mild buccal and lingual cortical plate expansion in the left lower border of the mandible marginal mandibular and facial nerve intact. No mobility of the teeth noticed in that region and occlusion was not altered with normal mouth opening.

Panoramic radiograph showed unilocular radiolucency with scalloped borders and root resorption of left third molar and distal root resorption of left second molar. Loss in the continuity of the lower border of the mandible noticed (figure 1). The plain computed tomography (CT) report showed evidence of a large expansile lytic lesion below the left lower third molar tooth involving the angle and body of the left half of the mandible measuring 3.4X2.4X4.8cms causing bone remodeling, with scalloping of the borders and cortical thinning more so involving the lingual cortex. Heterogeneous contents seen within the lesion. Suspicious area of cortical breach seen involving the posterior cortex. Significant cervical lymphadenopathy noted involving bilateral level II, III, IV, V. CT report was suggestive of ameloblastoma or odontogenic keratocyst (figure 2).

Based on the investigative reports, biopsy was done and sent for histopathological evaluation. The histopathological reports suggested multiple sections showed cyst wall lined by odontogenic epithelium. This epithelium has basal cell with displaced nucleus basally and vacuolated cytoplasm. The central cells resemble stellate reticulum cells. Also seen are tissue bits composed of hemorrhage, fibrocollageous tissue with cholesterol clefts and foreign body giant cell reaction, trabecular bone. Three lymph nodes with hyperplasia were also observed. Features are suggestive of Ameloblastoma – unicystic type. En block resection and reconstruction with long plate was done (Figure 3). Healing of the lesion was noticed during the follow up of the patient (Figure 4).

Discussion

According to WHO in 2003, ameloblastoma was divided based on differences in biologic behavior, treatment plan and recurrence rate as classic solid or multicystic ameloblastoma, unicystic ameloblastoma, peripheral ameloblastoma and desmoplastic ameloblastoma, including the so-called hybrid lesions.[6] But they have given a new type which is more simplified than the previous classification into 3 types: conventional, unicystic and peripheral. New term conventional one has been introduced instead of solid or multicystic to avoid confusion with unicystic type. Desmoplastic ameloblastoma was also reclassified as a histological

subtype and not as a clinical-pathological entity, based on the fact that it behaves like any conventional ameloblastoma, although its clinical and radiographic characteristics are peculiar.[7]

UA only accounts for 6% of all the types being the rare one. Usually it is seen in young age group contradicting to the present case which was seen in fifth decade of life. The gender predilection is slightly more towards male accounting a ratio of 1.6:1.[9,10] Posterior mandible is the most common site of occurrence which was the site of occurrence in our presented case followed by parasymphysis, anterior maxilla and posterior maxilla.[9,11] The clinical presentation varies from painless swelling causing facial asymmetry to mucosal ulcerations or sometimes it will be asymptomatic. It presents more like a cyst clinically especially dentigerous cyst.[11,12] Few lesions are identified during routine radiography or as a result of local effects on surrounding structures like tooth mobility, altered occlusion or delayed eruption.[13] No such local effects on tooth or surrounding structures were noticed in our case. Only bicortical expansion was present clinically and presented as painless swelling which is classical feature of UA.

Different imaging modalities used includes with conventional intraoral radiographs followed by occlusal and panoramic imaging. But Computed tomography and Cone beam computed tomography gives us more accurate picture.[14] In the present case, we used CT imaging modality. Radiographically, there are mainly two presentations, either unilocular or multilocular associated with root resorption or displacement of tooth. In our case, it presented as unilocular expansile lesion with scalloped margin and thinning of the lingual cortex. However, Eversole et al. and Paikkatt et al. have discussed four different radiographic patterns of UA which includes unilocular, scalloped macromultilocular, pericoronal, interradicular, or periapical expansile radiolucencies.[15,16] Ackermann et al has classified the UA based on histopathological presentation as Group I: Luminal UA (tumor confined to the luminal surface of the cyst); Group II: Intraluminal/plexiform UA (nodular proliferation into the lumen without infiltration of tumor cells into the connective tissue wall), and Group III: Mural UA (invasive islands of ameloblastomatous epithelium in the connective tissue wall not involving the entire epithelium).[17] Whereas Philipsen and Reichart have subclassified into Subgroup 1: Luminal UA; Subgroup 1.2: Luminal and intraluminal; Subgroup 1.2.3: Luminal, intraluminal and intramural; Subgroup 1.3: Luminal and intramural.[7]

The management of ameloblastoma is still controversial because of its benign nature, locally aggressive and high recurrence rate. Factors which has to be considered for selecting the treatment includes age, size, location, duration, involvement of cortical bone and soft tissues. Based on these variables, the treatment may be conservative or radical.[14] Enucleation, enucleation with curettage or cryotherapy forms the conservative treatment options and Radical treatment includes marginal or block resection and immediate bone reconstruction.[14,18] En block resection and reconstruction with long plate was done in the present case.

Conclusion

Unicystic ameloblastoma is a different entity compared to the conventional one as it mimics like a cyst mainly dentigerous cyst. It is usually seen in young generation, asymptomatic swelling especially in posterior mandible. This article highlights a rare case presentation of unicystic ameloblastoma in the posterior mandible in 50 year old female with surgical management.

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Figure legends

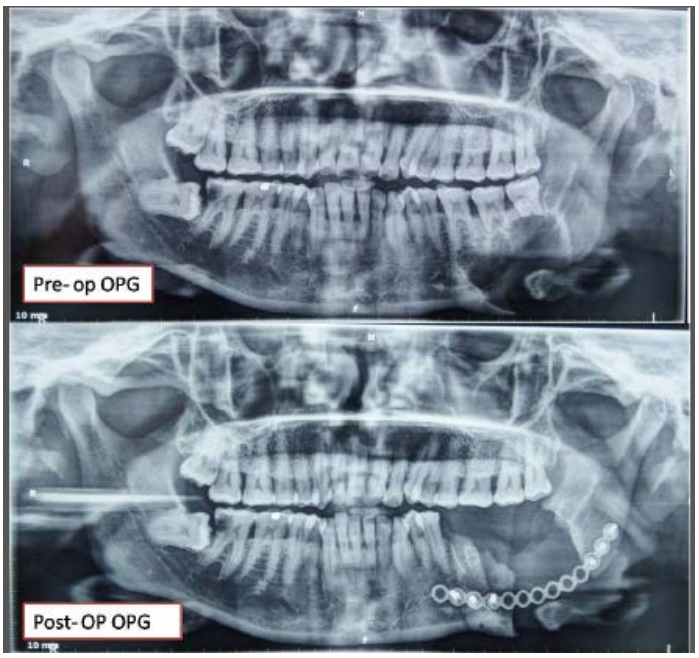

Figure 1: OPG (Pre –OP) showing unilocular radiolucency with scalloped borders and root resorption of left third molar and distal root resorption of left second molar and Post OP OPG showing surgery done with placement of long plate.

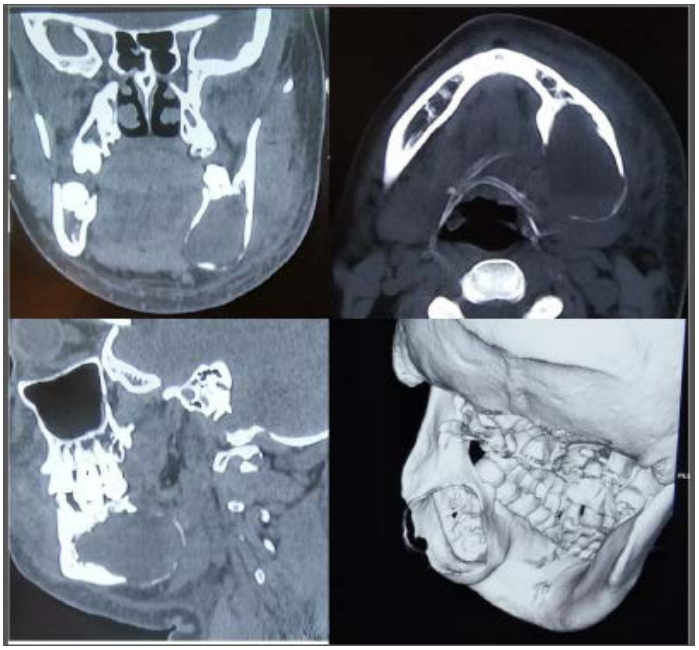

Figure 2 : The CT (Coronal, sagittal, axial and 3D sections) shows a large expansile lytic lesion below the left lower third molar tooth involving the angle and body of the left half of the mandible with scalloping of the borders and cortical thinning more so involving the lingual cortex.

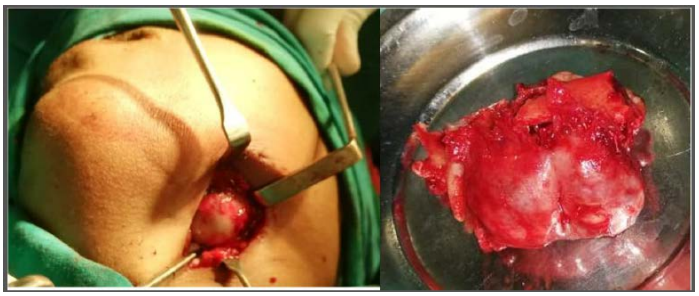

Figure 3: En block resection surgery

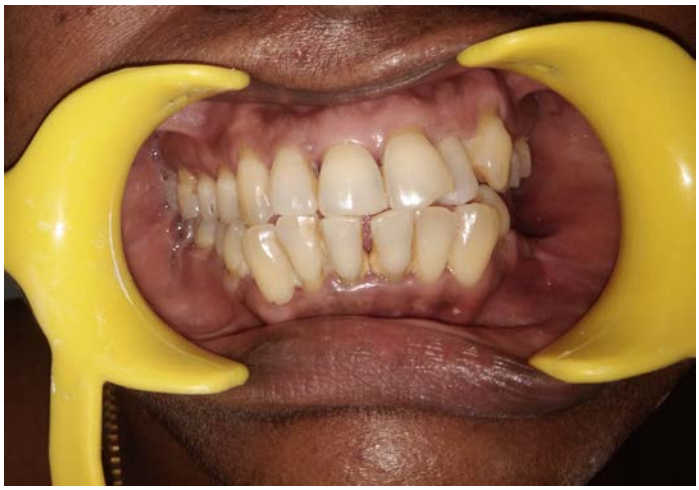

Figure 4: Post operated photograph