10.5005/jp-journals-10031-1104

CASE REPORT

Unicystic Ameloblastoma

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ABSTRACT

The term ameloblastoma was first coined by Churchill in 1934. Robinson and Martinez in 1977 described ameloblastoma as being a tumor that is usually unicentric, nonfunctional, intermittent in growth, anatomically benign and clinically persistent. WHO in 2005 classified ameloblastoma as a benign tumor with odontogenic epithelium with mature fibrous stroma without odontogenic ectomesenchyme. International classification of diseases numbers ameloblastoma as ICD-O 9310/0. Shafers classified it as a being odontogenic tumor presently thought to be as a result of alterations or mutations in the genetic material of cells that embryologically preprogramed for tooth development. It has six histologic variants of which unicystic ameloblastoma is a distinct clinical variety. This article is a report of one such case.

Keywords: Ameloblastoma, Unicystic, Intraluminal, Intramural.

How to cite this article: Dave PM, Galinde JS, Poonja KS. Unicystic Ameloblastoma. J Contemp Dent 2015;5(1):40-42.

Source of support: Nil

Conflict of interest: None

INTRODUCTION

The unicystic variant of ameloblastoma is a relatively new category that has been created because of the differences between it and the conventional type. Its incidence is approximately 6% of the total ameloblastomas reported. Mostly noted in the second and third decades of life, mean age being 22 years at the time of diagnosis which is approximately one and a half decades earlier than the conventional type. Its typical radiographic presentation is that of a well-defined, unilocular radiolucency associated with the crown of an unerupted tooth, usually a mandibular third molar. Therefore, it is difficult to differentiate it from a dentigerous cyst or an odontogenic keratocyst. It may also appear as a unilocular radiolucency that is nonspecific. Correlation of cystic lesion along with histopathological findings of a unicystic structure lined by ameloblastic epthelium gives the definitive diagnosis. They probably arise from neoplastic alteration of a preexisting cyst or develop *de novo* as a unicystic neoplasm from remenants of primitive dental lamina. The differential diagnosis clinically may be of other ameloblastic odontogenic tumors, dentigerous cyst or keratocyst odontogenic tumor (KCOT). Surgical management ranges from simple curettage to more agressive treatment of the surrounding bone according to the depth of infiltration of the ameloblastic epithelium into the surrounding tissues. The recurrence rate for true unicystic ameloblastoma is approximately 14%. Long-term follow-up is required for both this type and the conventional type.[1]

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CASE REPORT

A 29-year-old male presented with the complaint of persistent swelling on his right lower face since 2 months. Slight bulging of the right side of the face could be discerned. Intraorally, swelling in the right posterior vestibule was noted next to the lower molars and in the retromolar region (Fig. 1). The dentition was essentially intact, but the bite was in edge to edge occlusion. This condition was said to have remained unchanged during recent months. Lip sensation was normal. Radiographic examination showed well-defined unilocular radiolucency in the right mandible extending from the distal aspect of mesial root of first molar to the angle of the mandible (Fig. 2). Expansion of the mandible in all directions and thinning of the inferior cortex of the mandible were also noted. Fine needle aspiration biopsy was done to rule out malignant cells. CT scan reports

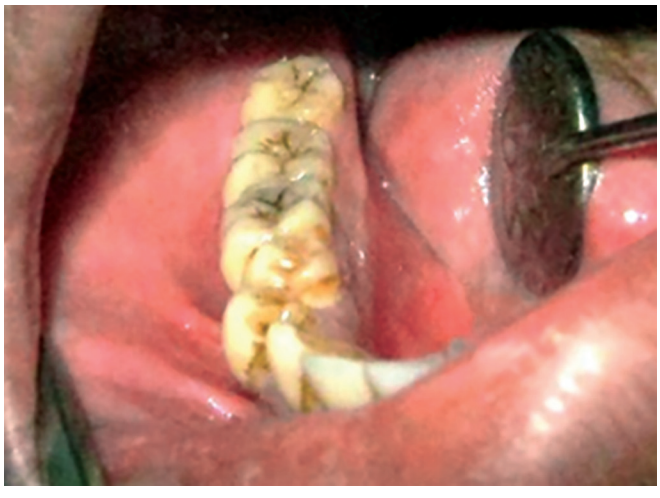

Fig. 1: Intraoral view showing obliteration of the right buccal vestibular space

were suggestive of ameloblastoma (Fig. 3). Segmental mandibulectomy and immediate reconstruction with reconstruction plate were planned as the bening tumor had thinned out the inferior cortex making it vulnerable to fracture post enucleation. Tumor was approached from the submandibular incision. Segmental resection with wide margins of the mandible from the angle up to the second premolar was then performed with a motorized saw (Fig. 4). A portion of the overlying soft tissue was excised along with the bony segment. Reconstruction of the segmental defect was done with the reconstruction plate (Fig. 5). Microscopic histopathological examination showed intramural and intraluminal type of unicystic ameloblastoma. Infection of the graft was controlled by systemic and local antibiotic treatment, as well as by daily wound care. A numb sensation of the right lower lip and chin was reported.

DISCUSSION

Unicystic ameloblastoma was first described as a distinct clinical entity in 1977 by Robinson and Martinez.[2] The unicystic ameloblastoma was thought to occur in a younger population (third decade) than its solid counterpart (fourth decade) It was most commonly encountered in the posterior mandible and is commonly associated with impacted teeth. In 1988, Ackermann et al classified unicystic ameloblastoma into three histological subsets depending on whether they had a cystic lining composed of simple odontogenic epithelium, a cystic lining showing intraluminal plexiform proliferation of the epithelial lining (intraluminal unicystic ameloblastoma), or a cystic lesion with epithelial invasion of the supporting connective tissue in either a follicular or plexiform form (mural unicystic ameloblastoma).[3] Following this reclassification, it was suggested that the first two subgroups were nonaggressive and could be treated by enucleation, but the third (the intramural group) required more aggressive treatment. However, this differentiation can generally only be made retrospectively from histological material once the lesion has been removed.[4] It is often difficult to differentiate a unicystic ameloblastoma from a multicystic ameloblastoma because many solid

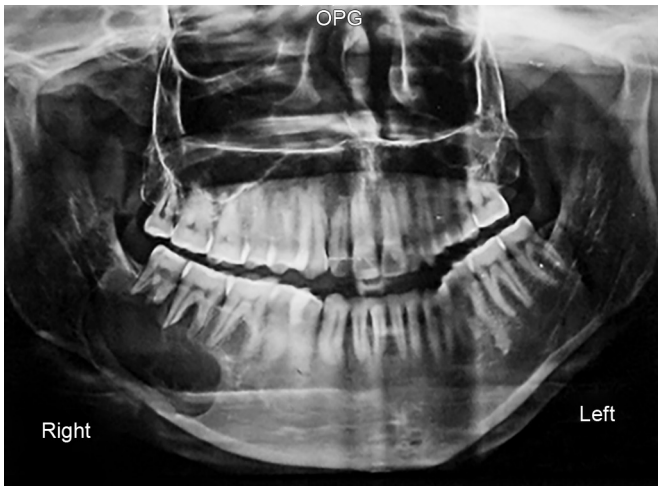

Fig. 2: Orthopantomogram showing unicystic radiolucent lesion in right posterior mandible

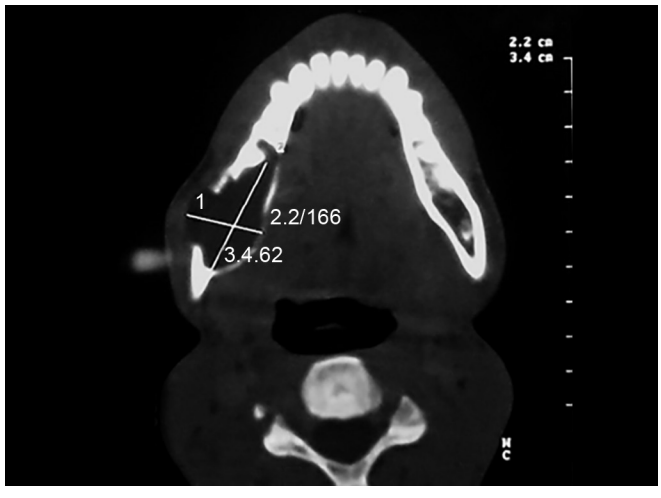

Fig. 3: CT scan axial slice through mandible showing radiolucent osteolytic lesion with cortical expansion

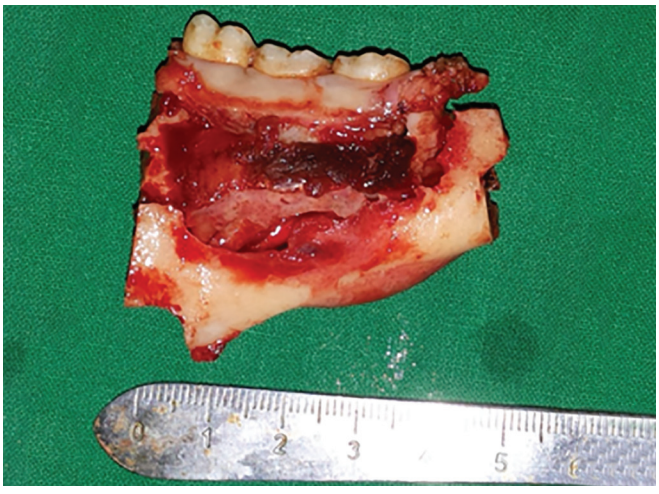

Fig. 4: Specimen showing tumor proper

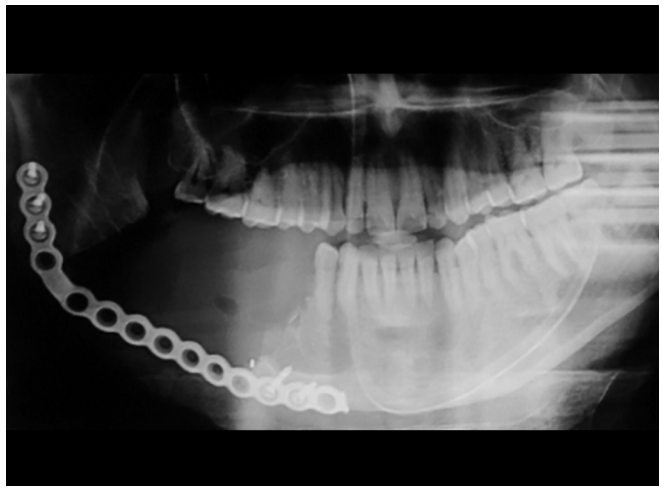

Fig. 5: Immediate postoperative OPG

ameloblastomas have a cystic component, and it is likely that the multicystic lesions behave more like a solid ameloblastoma. It is probably for these reasons that more recent studies have shown that simple enucleation of the so-called unicystic ameloblastoma is associated with a recurrence rate that may be as high as 60% and is similar to historical recurrence rates noted from enucleation of solid and multicystic ameloblastomas.[5-9] Unless the lesion has been suspected to be an ameloblastoma preoperatively, it is usually removed by enucleation and curettage as a non-neoplastic odontogenic cyst. Since final diagnosis can only be made after histological examination of the entire specimen, the treatment strategy proposed by Chapel et al in 2004 is the most rational. Unilocular cystic lesions in the maxilla or mandibular body should be enucleated and submitted for histological examination. If diagnosis is UNAM grade 1 (intralining) or grade 2 (intraluminal), no further treatment should be done immediately, but long-term follow-up (10-15 years) of the patient is required. If the diagnosis is UNAM grade 3 (intramural) or solid/multicystic ameloblastoma, the treatment should be partial maxillectimy or marginal/segmental resection immediately after primary surgery. In case of unilocular cystic lesion in the retromolar trigone and the ascending ramus of the mandible enucleation of the lesion with excision of the overlying mucosa should be done possibly with supplementary treatment of the cavity with liquid nitrogen or chemical cauterization with Carnoy's solution preserving the inferior alveolar nerve as possible.[10]

CONCLUSION

Ameloblastoma is a common bening tumor occurring in the maxillofacial area with atypical presentation, any one line of treatment strategy cannot be standardized and each case needs to be catered accordingly.

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