



CASE REPORT

A CASE REPORT OF RECURRENT AMELOBLASTOMA OF MANDIBLE WITH LITERATURE REVIEW

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ABSTRACT

Ameloblastoma is an aggressive, benign odontogenic tumor of epithelial origin. It comprises about 1-3% of all odontogenic tumors of maxillofacial region. It is characterized by slow persistent growth, locally aggressive infiltrative lesion mainly occurring in mandibular ramus region. They are occasionally associated with the presence of an impacted third molar. Conservative to radical surgical approach is mainstay of treatment depending on the extend of lesion and its histopathological behavior. There may be a high local recurrence rate when the tumor is not properly removed, due to remaining tumor cells or seeding of tumor cells in adjacent regions. This article reports a case of a multicystic ameloblastoma that had been previously treated with conservative therapy which failed and resulted in recurrence eight years later. Radical surgical protocol with wide surgical margin of safety adopted to prevent the recurrence of these aggressive tumors.

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INTRODUCTION

Ameloblastoma is the aggressive, benign, second most common odontogenic tumor of the oral cavity that originates from remnants of the dental lamina and dental organ (odontogenic epithelium) (Sivapathasundharam, 2016). It presents as a slow and persistent asymptomatic growth with the main site of occurrence being the mandible. Its incidence peak is in the third and fourth decade of life, with no gender preference and it is most common in black individuals (Ramesh, 2010; Amzerin, 2011). The ratio of ameloblastoma of the mandible: maxilla is 5:1, with higher occurrence in the mandible (Ram, 2010). Some manifestations associated with this tumor were painless swelling, facial asymmetry, dental impaction, tooth displacement, tooth mobility, root resorption, divergence of roots, occlusal interferences and dental extrusion (Kalaskar, 2011). Despite its benign nature, ameloblastoma

carries high risk of loco-regional recurrence, mostly seen within 5 years after surgical procedure. Biopsy and radiological evaluation should be done to differentiate the subtypes of ameloblastoma for proper operative planning to minimize recurrence rate. Various modalities of treatment have been suggested in literature like curettage, enucleation, marsupialization, cryosurgery, electrocautery, radical en bloc resection, sclerotherapy and radiotherapy etc (Shear, 2007). The primary choice of treatment for most common variant, multicystic or solid ameloblastoma is radical surgical excision with 1-2 cm safety margin beyond the limits of radiographic lesion, while the unicystic type may be successfully treated by enucleation or marsupialization (Dolanmaz, 2011). The purpose of this article is to emphasize the importance of a radical approach for the treatment of a multicystic ameloblastoma, by reporting a case of recurrence that had been treated with conservative therapy.

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Case report

A 50 years old male patient was presented to the department of oral medicine and radiology complaining of pain in the lower right jaw region since 15 days. During anamnesis, the patient reported having already undergone surgery once in that region to remove lesion 8 years back, but the pain had persisted as mild since long after the surgery. There was no documentation of prior treatment nor biopsy or histopathological examination to verify the diagnosis. Clinically, the patient had mild facial asymmetry from corner of mouth to angle of mandible extraorally with the tender firm swelling and paresthesia. Intraoral examination of lower right quadrant shows obliteration of buccal and lingual vestibule with buccal cortical plate expansion extending from 33 to 47 region covered with mucosa of normal colour (Figure 1, 2, 3). Incisional biopsy was requested for histological examination and the diagnosis was follicular ameloblastoma.



Figure 1. Extraoral- Frontal View



Figure 2. Extraoral-Wormian View



Figure 3. Intraoral View

The radiographic orthopantomograph examination revealed multilocular radiolucency (with intertwining thin radio-opaque septae) extending anteroposteriorly from 42 to 47 and superoinferiorly from alveolar crest to lower border of mandible. Mild root resorption is appreciated with 42, 47 (Figure 4).



Figure 4. Preoperative Orthopantomograph

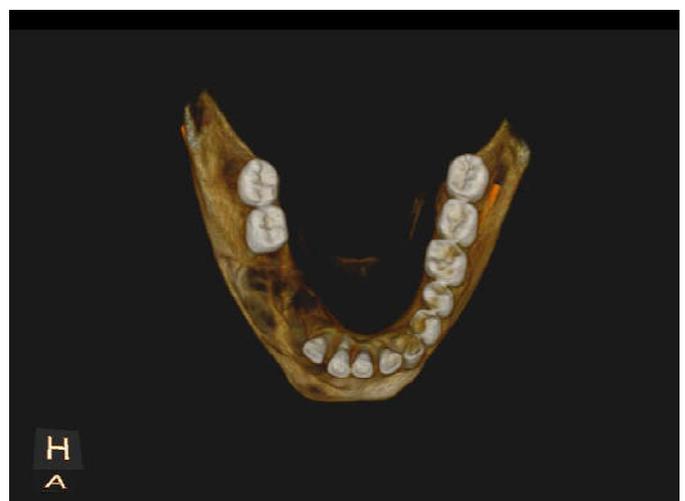


Figure 5. Occlusal 3 D View of Lesion in CBCT

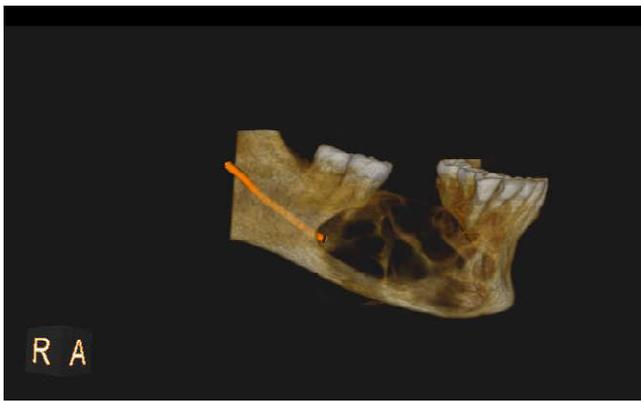


Figure 6. Lateral 3D View of Lesion in CBCT

A cone beam computed tomography further confirms OPG findings and revealed lesion causing slight ballooning expansion of the mandible in all the three planes with significant thinning of labial/buccal, lingual and inferior cortices but intermittently intact. Inferior alveolar canal is not traceable in first molar and premolar region (Figure 5, 6, 7).

DISCUSSION

Ameloblastoma is a rare benign odontogenic tumor, which was first described by Cusack. Malassez introduced term ‘‘adamantinoma’’ which is used for a rare form of bone cancer. Churchill renamed it as ameloblastoma (Sivapathasundharam, 2016).

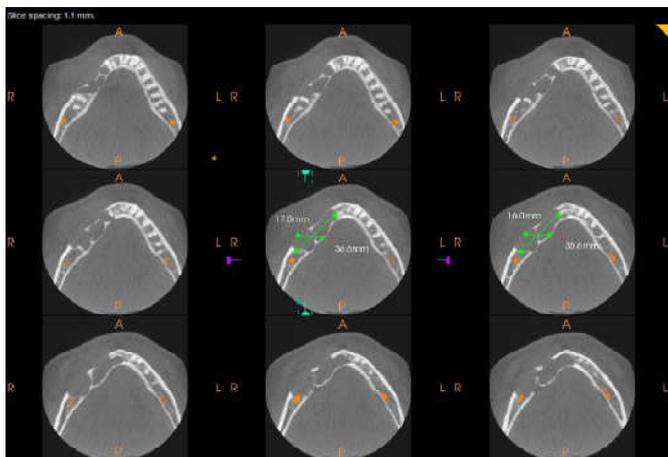


Figure 7. Axial View of Lesion in CBCT

Due to its locally aggressive growth pattern, ameloblastoma has a high recurrence rate. More than 50-72% % of the recurrences occur within the first 5 years after primary surgery (Reichart, 1995). But, here in our rare case report we noticed recurrence after 8 years which is very uncommonly reported in literature (Dolanmaz, 2011). Several factors governs this rate with the first and foremost being the clinicopathologic variant of the tumor. The solid or multicystic variant has the highest recurrence potential because of the great propensity for local infiltration. The second factor is the anatomic localization of the tumor. The most common anatomic location for ameloblastoma is the mandible; which has a dense cortical bone structure that prevents spreading for several years. Tumor can however extend beyond the radiological margin by spreading through the central cancellous bone. The third factor contributing to recurrence is the inadequacy of surgery (Adekeye, 1986). Most of the times due to limited extend or

small size of tumor, many operators prefer conservative surgeries like enucleation, marsupialization. To maintain lower intact border, marginal resection is done. But all this type of conservative approaches could be detrimental if safety margin of 1 cm beyond radiographic boundary not taken and would leads to recurrence. Thus, radical surgical excision is the most appropriate treatment that should be done for recurrent ameloblastoma as supported by literature (Kalaskar, 2011; Reichart, 1995; Collings, 1993). Tumoral excision and mandibular reconstruction with bone graft either vascularized or nonvascularized is the preferred choice for recurrent cases by most surgeons (Collings, 1993).

Conclusion

Ameloblastomas as having high propensity of loco-regional invasion and high risk of recurrence. Therefore, it is a paradox to recommend conservative treatment mainly in large sized expansile tumor mass. A radical surgical approach with a safety margin of at least 1 cm beyond the radiographically visible margins should be adopted to prevent tumor recurrence. This may be performed by either marginal or segmental mandibulectomy depending on the location and extent of lesion. Immediate primary reconstruction of the bone defect with local or distant free flaps followed by rehabilitation with implant-supported prostheses in a second stage will improve mastication and facial harmony of the patient.

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