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Case Report

Dual odontogenic tumors unicystic ameloblastoma and compound odontoma in a 24 year old male: A case report

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ABSTRACT

Unicystic ameloblastomas is one of imperturbable jaw tumor. It is a quirky and unique cystic variant of ameloblastoma with clinical and radiological features having kinship with odontogenic cysts of developmental and inflammatory origin and has a favorable prognosis rate. This paper presents a case of a unicystic ameloblastoma in a 24-year-old male in his right angle and ramus of mandible region along with compound odontome in left posterior maxilla.

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1. Introduction

Ameloblastoma, a belligerent clinical entity was firstly described by Cusack in 1827. Its capacity to expand and grow into full blown size with resulting bone deformity makes it important for knowledge of health care professionals.¹ The first explicit & comprehensive description was given by Falkson in 1879. Later on Ivey and Churchill coined the term 'ameloblastoma' in 1930.² Its clinical demenaor includes slow-growing, assailing and locally aggressive nature. It accounts merely 1% of all oral tumors with incidence reaching its pinnacle in thirty and forty years of life and no gender predilection has been evident.³ Categorization of ameloblastomas was done into four categories according to the World Health Organization (WHO) Classification (2005) of Head and Neck Tumours: multicystic, peripheral, desmoplastic, and

unicystic.⁴ In 1977 Robinson and Martinez proclaimed unicystic ameloblastoma as a distinct entity.⁵ Unicystic variant is tranquil in nature and usually occurs in younger years of life than the solid or multicystic variant. It predominantly affects mandible with more than 90% of cases have been reported in angle ramus region. About 5-15% prevalence rate of Unicystic ameloblastoma has been reported among all intraosseous ameloblastomas and half of cases occur in the second decade of life.⁶ Paul Broca in 1867, firstly described odontoma & this term was originally used for any tumour of odontogenic origin. Broca defined the term as 'tumours which were formed by the overgrowth or the transitory of the complete dental tissue'.⁷ They are the most common odontogenic tumours & occurrence rate has been reported as 22% of all the odontogenic tumours of the jaws.⁸ They are generally asymptomatic, incidentally found on radiograph and are seldom diagnosed before the second decade of life. Impaction or delayed eruption of permanent

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teeth is a common consequence of this pathological entity.⁹ In this report, we present a case of unilocular unicystic ameloblastoma in a 24 year-old male patient involving the right side mandibular angle region, extending up middle of ramus and compound odontome in posterior maxilla.

2. Case Report

A 24 year old male came to department of Oral Medicine, Diagnosis & Radiology, Institute of Dental Education & Advance Studies, Gwalior, Madhya Pradesh with chief complaint of pus discharge in his right lower back jaw since 1 year. It was associated with dull, intermittent, and non radiating pain. Pain relieved with over the counter analgesics taken as and when required. There was no other clinical presentations such as difficulty in mouth opening, chewing and occluding teeth. His medical history was non significant. On extraoral examination, no facial asymmetry was evident. On intraoral examination,

Teeth present 18, 17, 16, 15, 114, 13, 12, 11, 21, 22, 23, 24, 25, 26, 27,

48, 47, 46, 45, 44, 43, 42, 41, 31, 32, 33, 34, 35, 36, 37, 38

Erythema was present in gingiva distal to 48 region. The mucosa was intact and there were no evidence of sinus opening. On palpation no vestibular tenderness and obliteration was evident in 46 47 48 region (Figure 1). Tender on percussion was negative in relation to 46 47 48.No tooth mobility was present. Therefore intraoral periapical radiograph was advised.IOPA taken in relation to 46 47 48 region showed diffuse radiolucency involving distal root of 48 (Figure 2). Panoramic radiograph was further advised for localization and enlarged view of lesion. It revealed a solitary, well-defined radiolucency of size 2.5 cm × 2 cm extending anteroposteriorly from distal root of 48 to ramus of the mandible, superior inferiorly 2 cm below the sigmoid notch to 2 cm away from inferior border of the mandible (Figure 3). A provisional diagnosis of unicystic ameloblastoma was given with differential diagnosis of odontogenic keratocyst. Panoramic radiograph also revealed well defined aggregate of radiopaque mass resembling tooth like denticles in relation to 28 region (Figure 3). A provisional diagnosis of compound odontome was also given for left posterior maxillary lesion. As this lesion was asymptomatic it was not surgically retrieved for histopathological examination. Patient was referred to department of oral and maxillofacial surgery & cone beam computed tomography was advised by them. Axial (Figure 3), sagittal (Figure 4), coronal (Figure 6) slices were obtained and they show an ovoid shaped radiolucency in distal aspect of 48 of size scaling from 17 mm superior-inferiorly and 23 mm antero posteriorly and 11mm bucco lingually with smooth, uniform and corticated borders. Mild bucco lingual expansion was appreciable along with perforation of alveolar crestal bone and

thinning of lingual cortical bone can be seen. Antibiotics were prescribed prophylactically. Surgery commenced with crevicular incision under local anesthesia. A full thickness mucoperiosteal flap was raised buccally from the right third molar was retrieved. Third molar was extracted. The lesion was removed from the mandible meticulously and in toto using a range of chisels and curettes. The specimen was sent for histopathological examination.Hematoxylin and eosin stained material showed epithelial lining with ameloblast-like cells and adjacent connective tissue stroma. There was no luminal proliferation of epithelium, suggestive of intraluminal ameloblastoma (Figure 7). The patient's postoperative course was uneventful.



Fig. 1: Intraoral photograph of patient revealing 46, 47, 48



Fig. 2: Intraoral periapical radiograph of patient depicting an ovoid shaped radiolucent area extending from distal root of 48 towards ramus of mandible



Fig. 3: Panoramic radiograph revealing ovoid shaped homogeneous radiolucent area at right angle of mandible and radiopaque a collection of tooth-like structures arranged in floral pattern in posterior maxilla

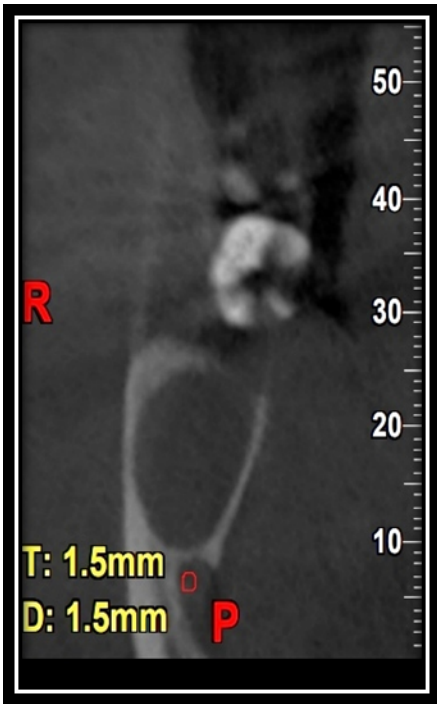


Fig. 4: Cone beam computed tomography (Axial view)

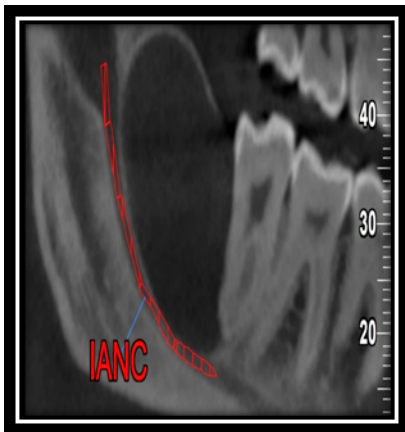


Fig. 5: Cone beam computed tomography (Sagittal view)

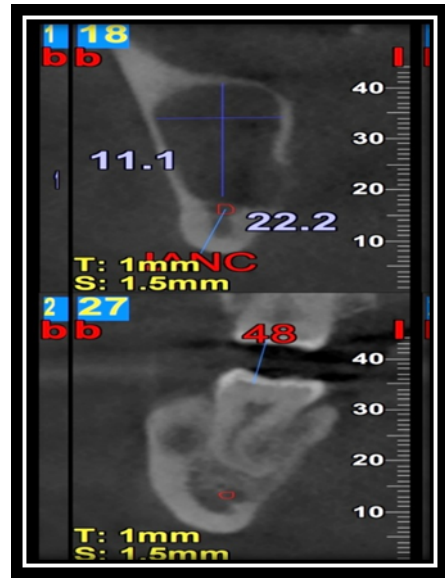


Fig. 6: Cone beam computed tomography (Coronal view)

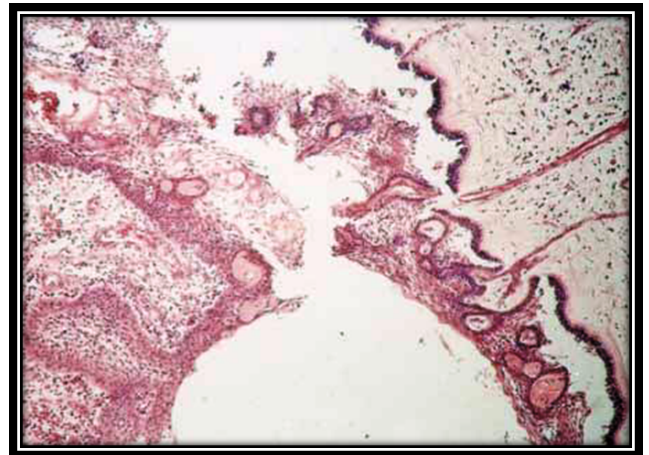


Fig. 7: Microscopic revealing intraluminal ameloblastoma

3. Discussion

Various theories have been postulated about origin of unicystic ameloblastomas. Literature documented over years suggest that it may arise from reduced enamel epithelium associated with developing tooth or it may develop in a preexisting dentigerous cyst or other types of odontogenic cyst or solid ameloblastoma may undergo cystic degeneration, resulting in its formation. However, no sufficient evidences are present to prove these theories.¹⁰ Roentographically, multilocularity is exhibited by majority of the conventional ameloblastomas, whereas unicystic ameloblastomas presents as a solitary large unilocular radiolucency. Its association with unerupted third molars has been proven in numerous studies done over years and may be espied during the course of routine radiography.¹¹

The preponderance ameloblastomas arise in the mandible, and the majority of these are found in the angle and ramus region. Our present case was also found at right angle and ramus region. Clinical presentation of most patients includes swelling and grotesque facial appearance and pain being an occasional presenting symptom.¹² In present case no swelling and facial asymmetry was present. Due to persistent growth of tumor it may sometimes lead to mucosal ulceration. Smaller detriments are incidentally discovered on routine radiographic investigations or as a result of local effects like tooth displacement, deranged occlusion and failure of teeth eruption in oral cavity produced by the tumor.¹³ In present case no such clinical findings were evident. Unicystic ameloblastoma according to prognostic and therapeutic implications is classified into three subgroups by Ackermann et al in 1988.¹⁴

1. Group I: Luminal (tumor confined to the luminal surface of the cyst)
2. Group II: Intraluminal/plexiform (nodular proliferation into the lumen without infiltration of tumor cells into the connective tissue wall)
3. Group III: Mural (invasive islands of ameloblastomatous epithelium in the connective tissue wall not involving the entire epithelium).

Philipsen and Reichart gave histologic subgrouping:¹⁵

1. Subgroup 1: Luminal
2. Subgroup 1.2: Luminal and intraluminal
3. Subgroup 1.2.3: Luminal, intraluminal, and intramural
4. Subgroup 1.3: Luminal and intramural.

Conservative enucleation can be done for first two groups, whereas latter two groups showing intramural growths require more aggressive procedures such as marginal or segmental resection.¹⁶ In 1988 judicious use of Carnoy's solution was suggested by Stoelinga and Bronkhorst in order to prevent recurrence after conservative surgical management of unicystic variant & cauterization with this solution is advocated for first two subgroups. It should be used cautiously as it can render some damage to the periodontium of the adjacent tooth. After enucleation robust curettage of the bone should be avoided as it may embed ameloblastic foci deeper into bone.¹⁷ Various reports in literature concluded that barely about 25 % or slightly less recurrence rates for unicystic ameloblastoma have been evident after conservative surgical treatment in comparison to solid multicystic ameloblastoma where recurrence rates was found to be on higher end about 50%–90%.¹⁸ Recurrence following treatment after interval of about seven years or more is commonly seen and has been documented in literature. Recurrence also depends on histologic subtypes of unicystic ameloblastoma. Those pathological entities invading the fibrous wall having recurrence rate of 35.7%, but others without invasion have recurrence rate

only about 6.7%.¹⁸ Recurrence rates are also impacted by type of preliminary treatment done for management of lesion. Lau and his co workers analysed various cases and concluded recurrence rates of 3.6% for resection, 30.5% for enucleation alone, 16% for enucleation followed by Carnoy's solution application, and 18% by marsupialization followed by enucleation.¹⁹ Odontomas are non-combative, hamartomatous developmental malformations or lesions of odontogenic origin. They may occur solitary or multiple radioopaque lesions ranging from small to giant size. Odontomas has been classified in two categories by WHO in 2005 has classified on basis of histopathological findings.²⁰ Complex odontomas, in which well formed dental tissues are discernible exhibiting an amorphous and more or less disorderly arrangement, and the compound odontomas, in which arrangement of dental tissues are in an orderly pattern, but their morphological features such as size and configuration are modified, producing multiple small teeth like conformations called odontoids or denticles.²¹ The teeth affected in order of frequency are canines, maxillary central incisors and third molars. In majority of cases teeth remain impacted but in a very few instances odontomas are related to missing teeth. By and large these malformations are intraosseous, but at times they may erupt into the oral cavity.²² In present case odontome was asymptomatic and associated tooth maxillary left third molar was missing and odontome was far below alveolar crest and unerupted in oral cavity. It can be differentiated from the Odontoameloblastoma can be considered as differential diagnosis and can be differentiated by the fact that it is well circumscribed and usually separates easily from its bony bed.²³

4. Conclusion

A thorough knowledge of biologic, roentographic and histopathological features is essential for diagnosis of unicystic ameloblastoma. It is a tumor with a penchant for recurrence. Diligent examination of tissue sections should be done by the pathologist in order to determine histological staging and level of progression and invasion of ameloblastoma or not so that the complications can be minimized & appropriate therapeutic strategies can be planned.

5. Source of Funding

None.

6. Conflict of Interest

None.

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