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CASE REPORT

UNICYSTIC AMELOBLASTOMA IN ANTERIOR MANDIBLE: A RARE ENTITY- A CASE REPORT

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ABSTRACT

Ameloblastoma is a benign odontogenic neoplasm that frequently affects the mandible. The term ameloblastoma includes several clinical, radiological, and histological types. In many cases, it poses significant diagnostic and therapeutic challenges. Ameloblastoma is a benign tumor that invades locally with the most common site being the third molar region mandibular jaw. Most commonly ameloblastoma occurs in the mandibular third molar region up to 66%, followed by 11% in the mandibular premolar region, 10% in the mandibular anterior region, 6% in the maxillary anterior, and posterior region, and only 1% in the maxillary premolar region. Here we present a rare case of Unicystic ameloblastoma occurring in the anterior mandibular region crossing the midline in a 17-year-old-male.

INTRODUCTION

According to the 4 th WHO classification of ameloblastoma, the Unicystic type is classified as a benign epithelial odontogenic tumor (1). As the name implies, it can develop from the enamel organ, which does not differentiate to form hard tissue, residual epithelial tissue, or epithelial components of odontogenic cysts. The term Unicystic ameloblastoma (UA) refers to a type of cystic lesion that resembles any other jaw cyst clinically, radiographically, or grossly. A typical ameloblastomatous epithelium lining a cyst cavity is revealed by histologic examination (2). Unicystic ameloblastomas account for 10-15% of all intraosseous ameloblastomas (3). This paper illustrates a case of Unicystic ameloblastoma of the mandible in a 17-year old male.

Case Presentation: A male patient aged 17 years reported to the Department of Oral Medicine Radiology at M. A Rangoonwala Dental College, Pune, Maharashtra, India with a complaint of swelling in the lower front tooth and jaw region for the past month, with associated pain while swallowing. The swelling initially was insignificant, but later began as an asymptomatic enlargement of the lower right and front jaw. It slowly progressed over the past several days to attain its present size. The patient gave a history of trauma in the lower front region two years ago, associated with a cricket ball while playing. Derangement of lower front teeth was noted by the patient. The medical history stated that the patient underwent surgery for a left-hand fracture, six years ago while playing cricket.

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On extra oral examination, A diffuse, smooth surfaced swelling over the chin region in the lower third of the face on the right side measured about 30 x 20mm approximately. The swelling was observed, in the right para- symphysis region extending from the level of the angle of the mouth, to the lower border of the mandible, crossing the midline. The superior inferior extension was from the lower border of the lower lip to the base of the mandible. Anteroposterior extension from the midline to the commissure region. On palpation, the swelling was smooth, afebrile & nontender. The skin overlying the swelling was shiny, whitish red, and nonpinchable. On lymph node examination solitary left submandibular lymph node was tender, palpable, and firm in consistency (Fig 1). On Intra oral examination, a diffuse, grossly ovoid smooth surface swelling measuring about 30x 20mm approximately extending from the mesial side of 44 to the distal side of 32 on the other side obliterating the vestibule. Mucosa overlying the swelling was normal. The surface was smooth with well-defined borders. On palpation, margins were well-defined, firm in consistency, non-compressible, non-fluctuant, and nonreducible. Expansion of buccal and lingual cortical plate was associated with the involved teeth. Tendernesswas present i.r.t 44,43,42,41,31,32,33 (Fig *2*). On hard tissue examination labially placed 13, derangement associated i.r.t 43,42,41,31,32. Grade I mobility was present i.r.t 43,42,41,31,32. Class II div 1 malocclusion was noted. Displacement associated i.r.t 42. Calculus deposit noted i.r.t 43,42 along with marginal gingivitis. (Fig3,4). A chairside investigation was performed that included the pulp vitality test, aspiration (FNAC), and an occlusal radiograph to obtain a better two-dimensional view. The vitality test showed 44,43,42,41 were vital, whereas 31,32,33 were nonvital. Following the appearance of the swelling aspiration was carried. The aspirate showed straw-colored fluid (fig 5). Considering the noteworthy appearance of the aspirate further radiographic investigations were carried that included Occlusal radiograph (two-dimensional view).

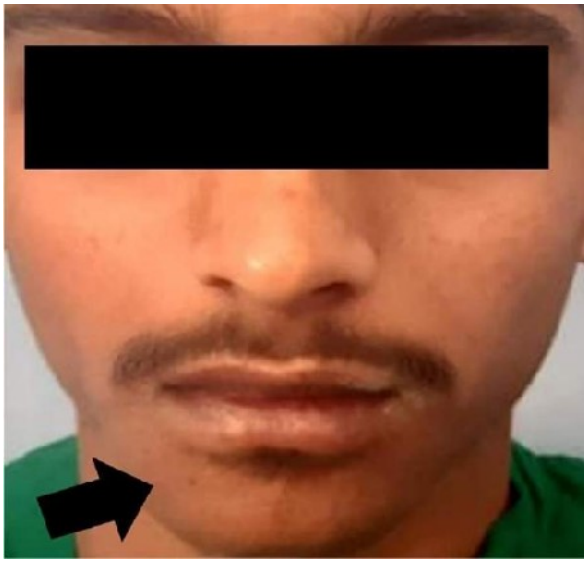

Figure 1. Swelling over the right side of the anterior mandible

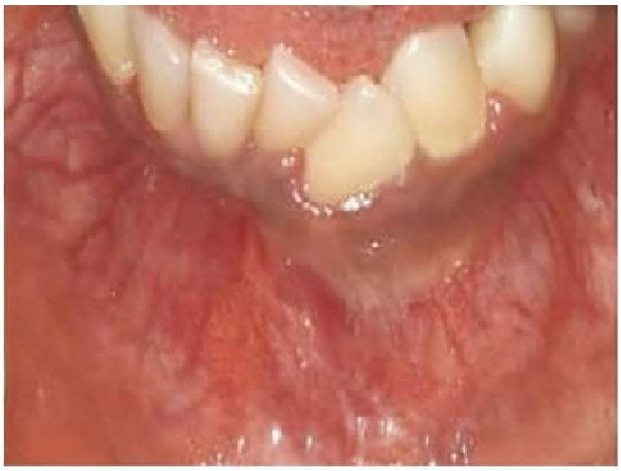

Figure 2. Area of chief complaint

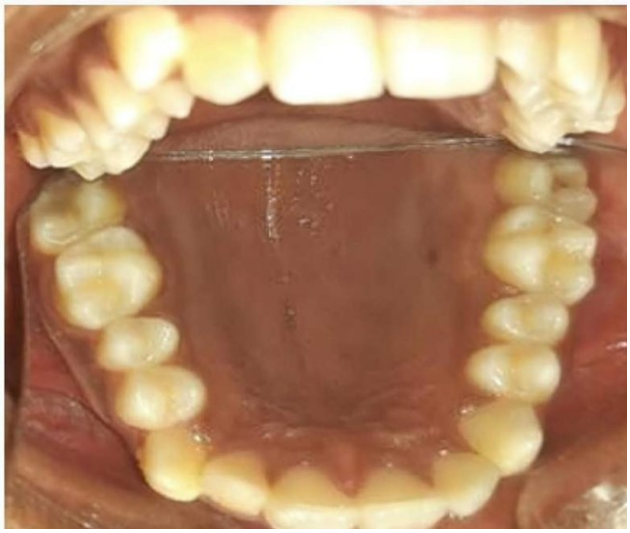

Figure 3. Maxilla Intraoral hard tissue examination

- Occlusal radiograph (two-dimensional view)
- CBCT (three-dimensional view)

Mandibular occlusal radiograph showed a multilocular radiolucency in the lower anterior region extending from the mesial surface of 33 to the distal surface of 45, thinning of the cortex in the buccal-lingual plane.

The size of the lesion was 30x20 mm approximately; Frank internal septa were noted from 45 to 32, giving a “soap bubble” appearance.

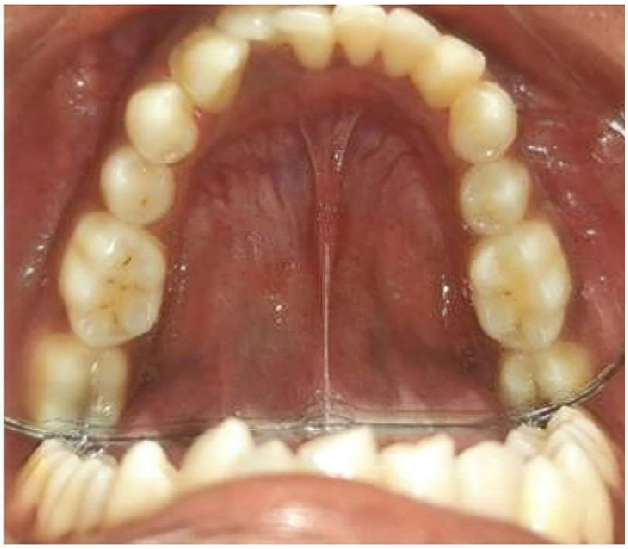

Figure 4. Mandible- Intraoral hard tissue examination

Displacement was associated with 42 which gave a floating appearance (Fig 6). CBCT (cone beam computed tomography) was performed to gain a better three-dimensional view. Cone beam computed tomography (three-dimensional) scan showed multilocular radiolucency inthe anterior region, an expansile cystic lesion crossing the midline in the anterior mandible completely destructing the buccal cortical plate, and thinning of the lingual cortex.

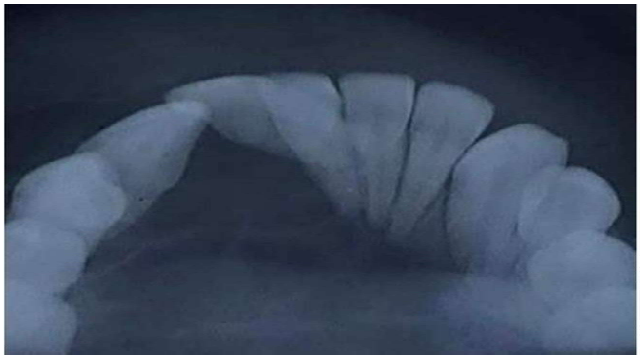

Figure 6. Mandibular anterior occlusal radiograph

There was flaring of the roots i.r.t 42. A provisional diagnosis of an infected radicular cyst was given. Differential diagnosis of benign odontogenic tumor such as ameloblastoma, calcifying epithelial odontogenic tumor (CEOT), and odontogenic keratocyst (OKC) was considered. The radiographic differential diagnosis of centralgiant cell granuloma, variant ameloblastoma was considered. Considering the clinical findings, chairside investigations, and radiographic features, enucleation was planned. After obtaining informed consent from the patient.

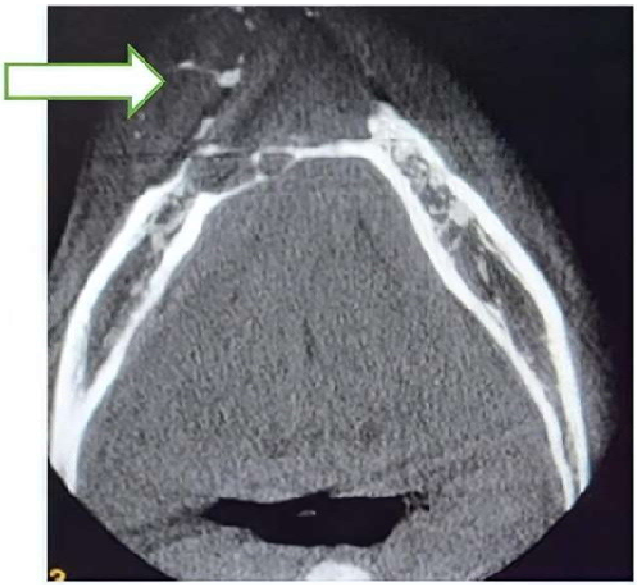

Figure 7. Axial View

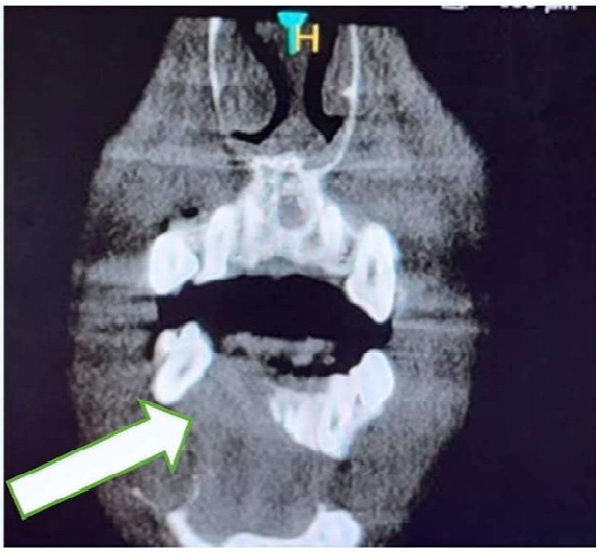

Figure 8. Coronal View

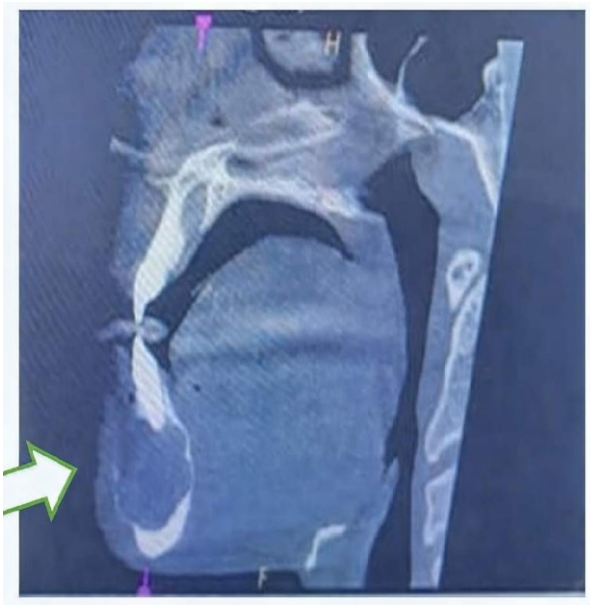

Figure 9. Sagittal View

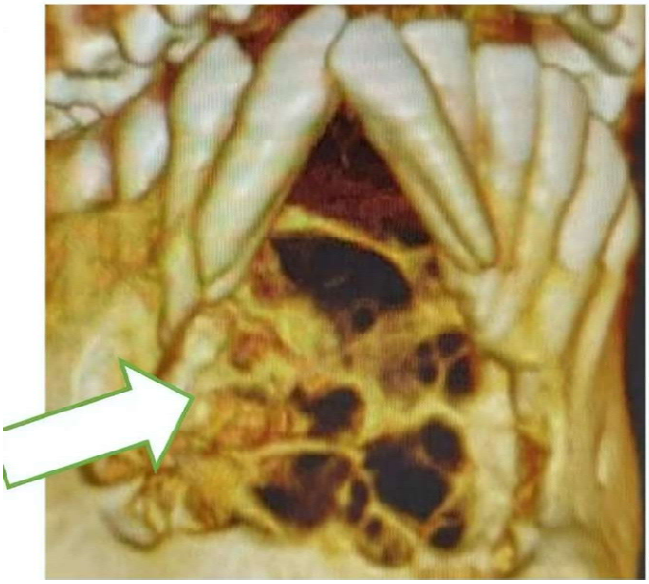

Figure 10. Three-Dimensional View

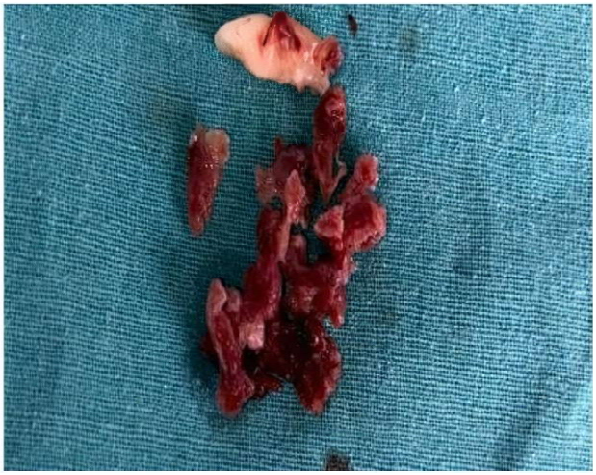

Fig. 11. The broken lobules and the extracted mandibular lateral incisor

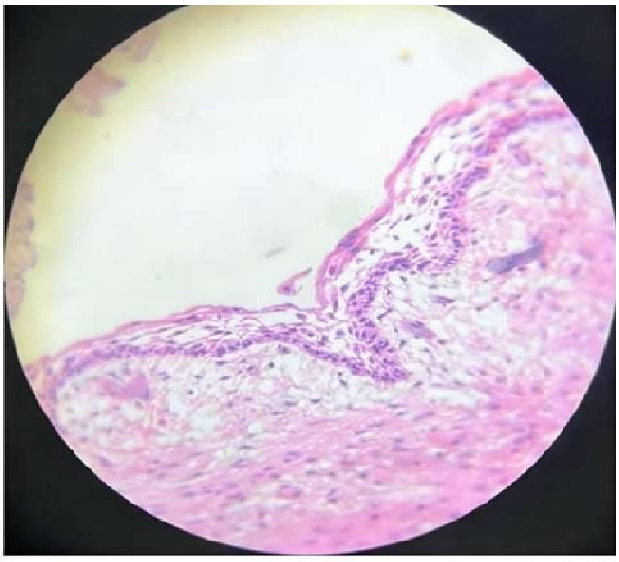

Figure 12. Unicystic Ameloblastoma

The lobules were broken and the lesion was decompressed; along with the extraction of the lower right lateral incisor (Fig11) and surgical rubber drain was placed. The specimen was further sent for histopathological evaluation.

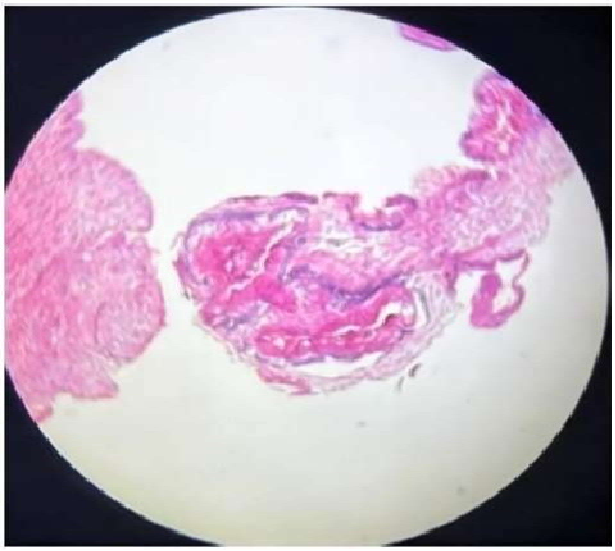

Figure 13. odontogenic epithelial islands present in connective tissue

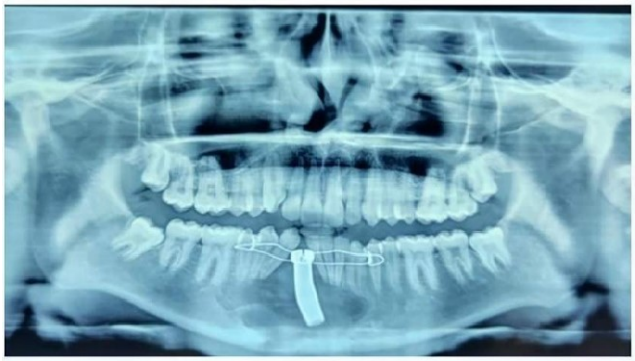

Figure 14. The post-operative orthopantomogram

The histopathological features noted were epithelial lining demonstrated a basal layer of cuboidal to columnar cells with hyperchromatic nuclei that showed reverse polarity and basilar cytoplasmic vacuolization. Overlying cells were loosely cohesive and resembled stellate reticulum-like cells. Connective tissue capsule consists of collagen fibers, extravasated RBC'S and odontogenic epithelial islands. Bone evidence was noted at the periphery. Overall findings were suggestive of Unicystic Ameloblastoma with odontogenic epithelial islands present in connective tissue (Fig 12). The post-operative orthopantomogram was taken to see the extent of rubber drain placed (Fig14). The nature of the tumor was explained to the patient and the patient is on regular follow-up visits.

DISCUSSION

Unicystic ameloblastoma accounts for 5-15% of all ameloblastoma types. Robinson and Martinez coined the term "UA" in 1977 (4). "Cystogenic Ameloblastoma" was also named by WHO in the second edition of the international histologic classification of odontogenic tumors (5). Unicystic Ameloblastoma typically affects young patients in their second decade of life (2).The development of a completely cystic ameloblastoma may be due to defects in desmosomal attachments or the degradation of the central zone of the enamel organ by metalloproteinases and serine proteinases after tooth development (6).Radiologically, Unicystic ameloblastoma appears to look like any other odontogenic cyst; however, histologically, it is distinguished by the presence of ameloblastic epithelial lining. It is classified into luminal and mural types. UA stands out in terms of prognosis. The recurrence rate is 6.7-35.7%, with an approximate average interval of 7 years (7).

CONCLUSION

The current case is indeed a unique variation of Unicystic Ameloblastoma. The location of the tumor in the anterior region and its association with trauma and swallowing pain is not commonlyseen in cases of ameloblastoma. This highlights the importance of thorough clinical examination and imaging studies in the diagnosis of these types of tumors, as well as the need for individualized treatment planning based on the specific features and characteristics of each case. In such a case, a multidisciplinary approach involving oral and maxillofacial surgeons, radiologists, and pathologists can be helpful in making an accurate diagnosis and developing an appropriate treatment plan. This can involve a combination of surgical resection, imaging studies, and biopsyto determine the extent and aggressiveness of the tumor, as well as the best approach for treatment.In addition, it's important to monitor the patient closely after treatment, especially in cases of anterior tumor location, as this area can have a significant impact on the patient's ability to eat and speak. Follow-up care and surveillance are crucial in ensuring the long-term success of treatment and reducing the risk of recurrence.

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Conflicts of interest: There are no conflicts of interest.

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