Report of Unicystic Ameloblastoma of Mandible in a Young Adult – A Radiological Perspective

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ABSTRACT

Unicystic ameloblastoma (UA) is a rarely encountered, less aggressive variant of ameloblastoma. It refers to those cystic lesions which radiographically exhibits features of jaw cyst but show a typical ameloblastomatous epithelial lining on histologic examination. Imaging is very important in diagnosing this tumour which has considerable similarities with odontogenic cysts. Here we report a case of an UA in an 18 year old male patient and discuss the role of panoramic radiograph, Cone beam computed tomography and Ultrasonography in the investigation and treatment.

KEYWORDS: Unicystic Ameloblastoma, Ultrasonography, Cone beam computed tomography of mandible

CASE REPORT

An 18 year old male patient reported to our department with a chief complaint of swelling on the right lower back tooth region since 3 months (Figure 1). Patient gave a history of trauma from the blunt end of knife on the right lower back tooth region 3 months back. He noticed a small pea size swelling after 3-4 days of trauma which gradually increased to present size. Medical and dental history were non- significant. On examining extra orally a solitary diffuse swelling was present on lower 1/3rd of right side of face of approximately 3 x 2.5 cm in size, extending in superior to inferior direction from 1 cm below the ala tragus line to base of mandible and in anterior to posterior direction from 2 cm posterior to corner of mouth to 1.5 cm anterior to pinna of ear. On palpation, swelling was soft to firm in consistency, non tender, no change in overlying skin with no rise in local temperature. Obliteration of buccal vestibule was seen in the region of 45, 46,47 due to the swelling. Mucosa over the swelling was normal. On palpation, there was expansion of the cortical plates in the region 45, 46, 47 along with grade II mobility in these teeth (Figure 2). Electrical pulp vitality test was performed in relation to 45, 46, 47, and they were non vital. Third molar in this quadrant was missing.

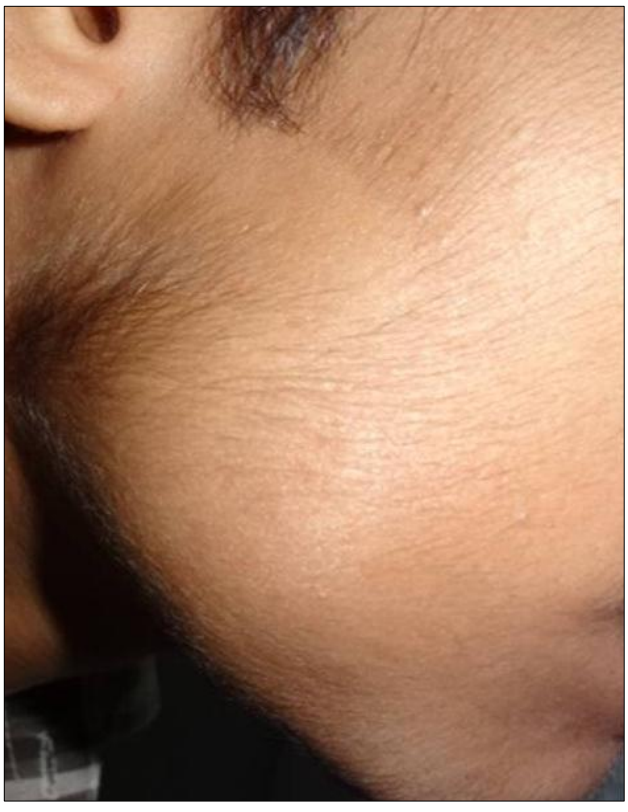

Figure 1: Extraoral photograph of the patient showing swelling on the right lower middle third of face.

A provisional diagnosis of aneurysmal bone cyst with a differential diagnosis of ameloblastoma was made. Fine needle aspiration cytology (FNAC) was performed, and the aspirate was a blood mixed fluid.

Panoramic radiograph (OPG), Ultrasonography (USG) and Cone beam computed tomography (CBCT) were advised for the patient. On the panoramic radiograph (Figure 3), a well- defined multilocular radiolucency on

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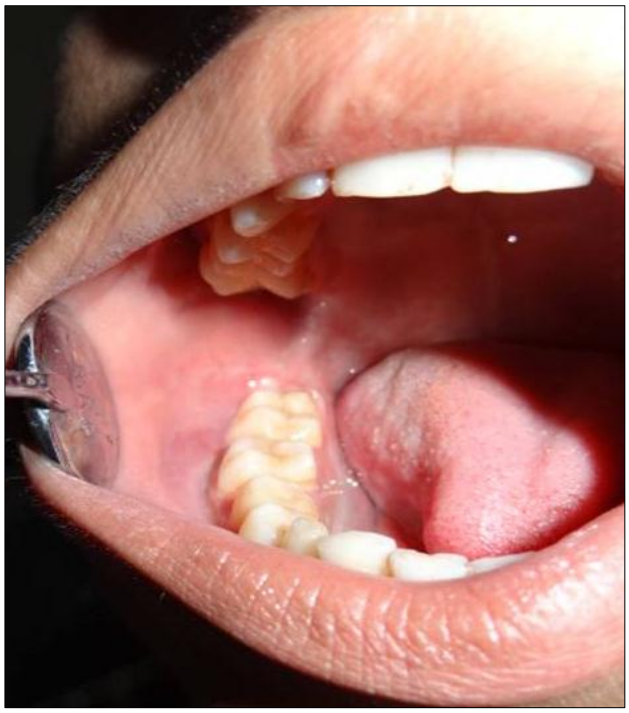


Figure 2: Intraoral photograph showing the swelling in the vestibule i.r.t 46, 47.

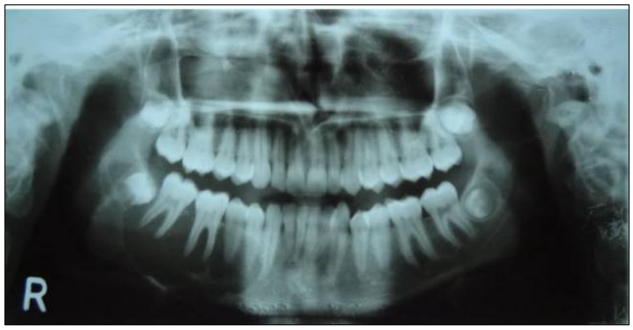


Figure 3: Panoramic radiograph showing the lesion on right posterior part of the mandible.

the right body of mandible extending from distal aspect of 43 to mesial aspect of 48 was seen. It had a well defined periphery without sclerotic border. The septae within the lesion gave an appearance of a spider web. The lesion caused resorption of roots of 44 and 45. Inferior border of mandible was intact along with intact cortices of mandible. 48 was impacted. An ultrasonographic examination with Color Doppler (Figure 4) using a High frequency Linear probe showed that the lesion mostly appeared hypoechoic with multiple internal echoes, well circumscribed in margins with posterior acoustic enhancement and was avascular. CBCT (Figure 5) revealed additional features which were not visible on panoramic radiograph like marked expansion, thinning of adjoining buccal, lingual and inferior cortices was appreciated with cortical effacement of the right inferior alveolar canal. The lesion had a radiolucent matrix with patchy fine bony septae in the peripheral sub-cortical region and centrally within the matrix, giving rise to multilocular appearance. Apical blunting and resorption of roots of 46 and 47 were also seen.

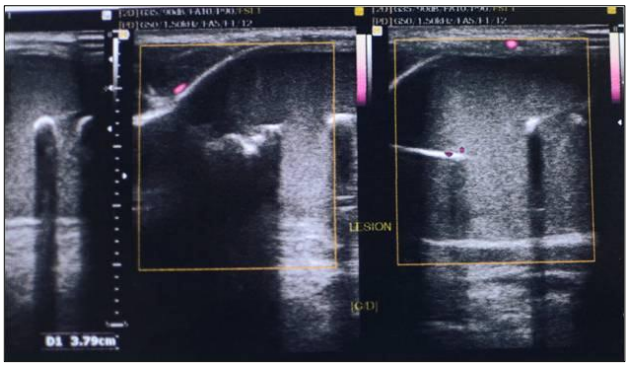
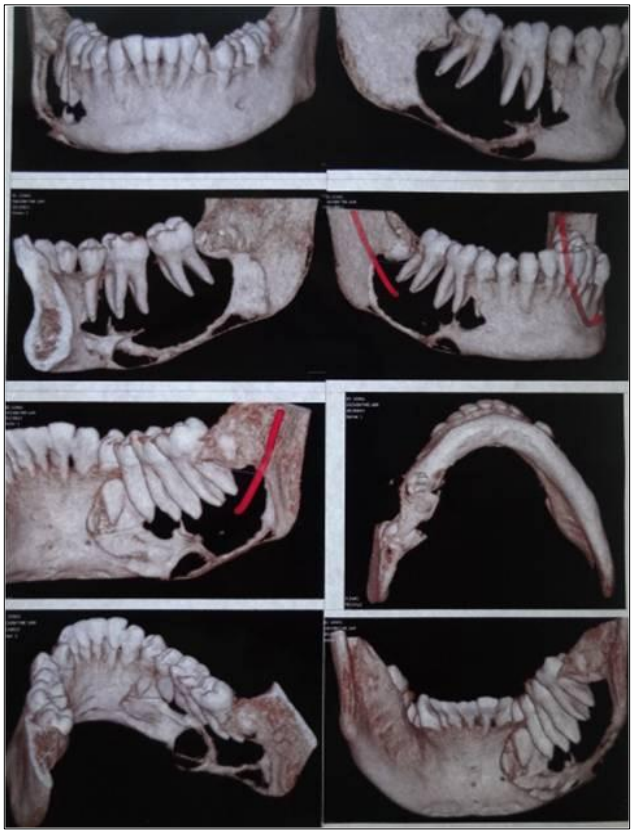
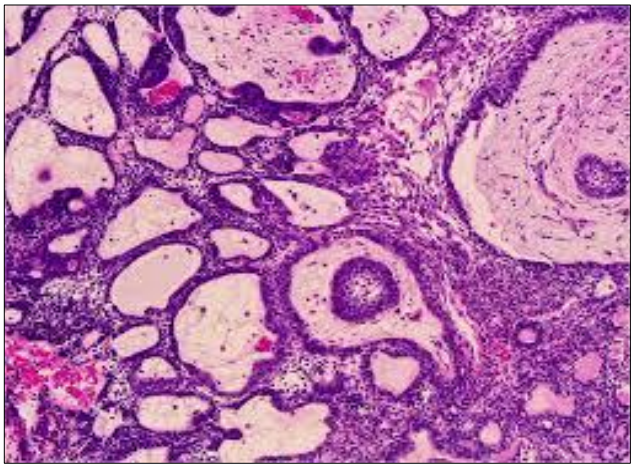


Figure 4: Picture showing the hypoechoeic and hyperechoeic areas on Color Doppler USG.

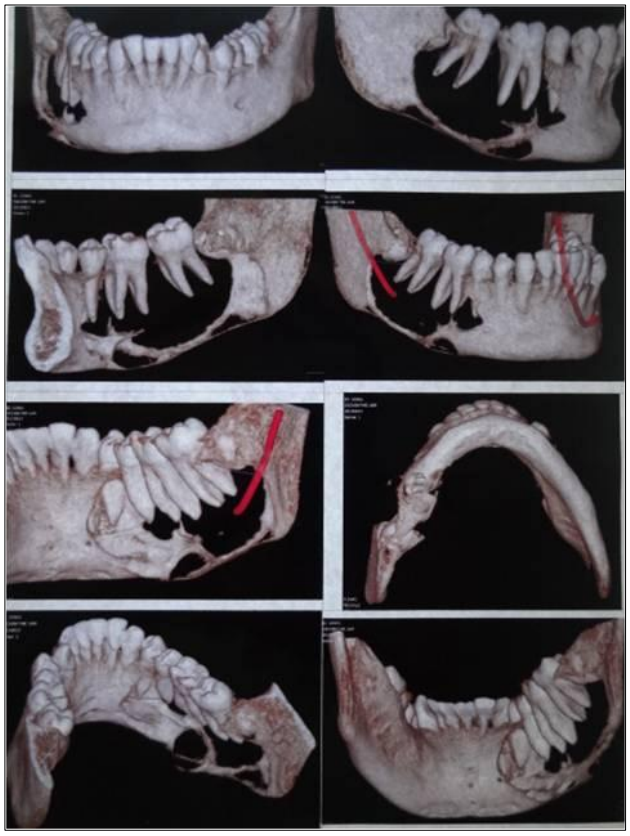


Picture 5: 3D CBCT reconstruction pre- op showing the extent of the lesion.



Picture 6: H&E stained section (x 40) showing ameloblastomatous epithelium.

Incisional biopsy was performed under LA in the department of Oral and Maxillofacial Surgery, and the H&E stained section (Figure 6) showed epithelium and connective tissue component. Epithelium was thin 2-4 cell layer thick in certain areas, and hyperplastic at other areas. Basal cells showed palisaded arrangement in few foci and superficial cells resembled stellate reticulum. There was proliferation of epithelium in the lumen at a few areas. Underlying connective tissue was fibrocellular in nature having thick bundles of collagen fibers, fibroblasts, blood vessels of varying sizes. Considering the clinical, radiological and histological findings a final diagnosis of unicystic ameloblastoma was given, and the patient was referred to Department of Oral and Maxillofacial Surgery, where marsupialisation of the lesion was done. The post operative CBCT (Figure 7) taken at 4 months showed evidence of formation of new endosteal bone with numerous small coarse new bone trabecullae along inferior, buccal anterior and posterior margins of the lesion and also residual lingual – crestal margins in posterior 1/3 of the lesion. There was early evidence of re-cortication along the buccal inferior margin of the lesion in 47, 48 region and slight re-cortication along the lingual margin in inferior third of lesion. Soft tissue thickening and internal air density were noted within the lesion which suggested the healing of the lesion. The patient is still under follow up.



Picture 7 : 3D CBCT reconstruction showing the post – op healing

DISCUSSION

Mandibular swellings of benign type can be classified as of odontogenic or of non-odontogenic origin. Ameloblastoma is the most common tumour of odontogenic origin developing from epithelial cellular elements and dental tissues[1], especially among the Asians.[2] It is slow growing in nature, persistent and locally aggressive neoplasm of epithelial origin accounting for 10% of the odontogenic tumours. It is commonly seen in the third to fourth decade of life with equal sex distribution. The majority of the ameloblastomas are seen in the mandible and the posterior part of mandible is the common site often associated with an un- erupted molar. There are three forms of ameloblastomas, namely unicystic, multicystic and peripheral. Multicystic is the most common represents 86% of the cases.[1] Unicystic ameloblastoma (UA) is a rare type of ameloblastoma, which account for about 6% of amelblastomas.[1] The term unicystic is derived both from the macro, and microscopic presentation of the lesion.[2] UA is one which has a large cystic cavity with luminal, intraluminal or mural proliferation of ameloblastic cells. It is of significance since it is a less aggressive variant which has a low recurrence rate.[3] It's usually seen in a younger age group, with about 50% of the cases occurring in second decade of life.[1] More than 90% of the cases are associated with the impacted tooth, and the mandibular third molar is the most commonly involved tooth.[1] The patient usually presents with a facial swelling or facial asymmetry, may be an occasional presenting symptom. Although mucosal ulceration is rarely found, it may be present due to continued growth of the tumour. Along with it the patient may also present with mobile tooth, alteration at the occlusal surface and failure in eruption of teeth caused by the tumour. Small lesions are discovered accidently on routine radiographic screening examinations. Diagnosis for UA on histopathological examination can be made if the lesion shows a cystic sac surrounded by an odontogenic epithelium which is seen in focal areas. As UA has a higher recurrence rate it should be differentiated from odontogenic cysts.[1]

H.M Worth has classically described 4 patterns for the radiographic appearance of ameloblastoma in which he says that the Unicystic type usually appears as unilocular radiolucency resembling a cyst. However, unlike a cyst, it causes a break or discontinuity in the peripheral cortex and may even show trabeculae within lumen.[4] In children and young adults, however, ameloblastomas may present as unilocular or as multilocular radiolucent lesions which can histologically be either unicystic or multicystic. Our case was of the multilocular unicystic variety. Radiological interpretation is paramount in this group of patients because it helps to standardize the surgical management. Ogunsalu C et al. have categorized ameloblastomas radiologically by studying 18 cases below 20 years of age.[5] Features such as presence /absence of root resorption and cortication at the periphery are useful in determining the aggressive nature of the lesion. Ameloblastoma in children and young adults are believed to be progressive in growth, especially those with less or no cortication. Also, patients in this age group may require a longer and more frequent evaluation

although unicystic ameloblastoma generally have less recurrence rate.[5] Conventional radiographs are useful only to study the interface between the tumor and normal bone and are quite useless to determine their extension into soft tissue. Here Conventional Tomography, Cone beam computed tomography, and Magnetic resonance imaging may play a significant role. But due to radiation safety issues CBCT is preferred over CT. Although the appearance on CBCT is essentially the same as in plain films, CBCT has an edge over the plain film and is highly recommended as it aids in better defining of the image and also in assessing the periphery and relationship to the adjascent structures. CBCT can also detect also if there is perforation in the outer cortex. As in our case, a follow up CBCT scan helped in appreciating the subtle changes seen in the endosteum during the post operative phase. Magnetic resonance imaging (MRI) scores more over CT for appreciating papillary projections into cystic cavity even though both are comparable for detecting the cystic components of the tumour and can provide better images of the nature and extension of the lesion. T1 weighted images have intermediate signal, and T2 weighted images have intermediate to high signals.[3]

Ultrasonographic studies of bony lesions are very few. But currently there is an emerging interest in expanding the applications of this non invasive, real time modality in Oral and Maxillofacial imaging to procure information of blood flow and also to distinguish cystic and solid components within the tumours. Although USG usually shows only bony surfaces and contours, in a neoplasm, the mandibular cortical bone effacement occurs thereby allowing penetration of ultrasonic beams. Hence, we can appreciate the internal structure of the tumour. Lu et al. have classified ameloblastomas into 4 types based on bony septum, compartment size and destruction features. They also observed that if the CDFI shows high blood flow signals it warrants a more aggressive surgical protocol.[6]

CONCLUSION

UA is a rare variant of ameloblastoma seenusually in the earlier decades of life. Our 18 year old patient was treated conservatively by marsupalisation. Imaging has an important role in assessing the aggressiveness of the tumour and helping in treatment planning especially in young patients. Newer imaging modalities like CBCT also aid in post-operative follow up. USG is evolving as a useful tool for diagnosing osseous tumours.

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