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RESEARCH ARTICLE

UNICYSTIC AMELOBLASTOMA IN 5 YEAR OLD CHILD: A RARE CASE REPORT

[1]Dr. Padmashree, S., [2]Dr. Mahesh, [3,*]Dr. Shibin Shaju and [4]Dr. Padma Pandeshwar

[1]Professor and Head of the Department, Oral Medicine and Radiology, Vydehi Institute of Dental Sciences and Research Centre, #82, EPIP Area, Nallurahalli, Whitefield, Bangalore
[2]Senior Lecturer, Oral Medicine and Radiology, Vydehi Institute of Dental Sciences and Research Centre, #82, EPIP Area, Nallurahalli, Whitefield, Bangalore
[3,4]Oral Medicine and Radiology, Vydehi Institute of Dental Sciences and Research Centre, #82, EPIP Area, Nallurahalli, Whitefield, Bangalore

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ABSTRACT

Ameloblastoma is a most common benign odontogenic neoplasm which effects the maxillofacial region and more frequently affecting the mandible. It exhibits various clinical, radiological and histopathological features based on different types of ameloblastoma. Among these types, unicystic ameloblastoma (UA) is the least seen variant of ameloblastoma, with good prognosis and less recurrence if detected at the earliest. UA refers to those cystic lesions that show clinical, radiographic, or gross features of a jaw cyst but on histological examination show a typical ameloblastomatous epithelium lining the cystic cavity, with or without luminal and/or mural tumour proliferation. UA commonly occurs in 2nd and 3rd decades of life but only about 10% of cases are reported in children and less than one third of those occur in children below 10 years. We report a rare case of 5 year old male patient, presenting with a swelling in the left lower jaw region which was diagnosed as UA by correlating the clinical, radiological, and histopathological findings. Thus early diagnosis of UA in children is mandatory as it affects the growth of the jaw. The incidence rate, behavior and prognosis of the tumour in children make the surgical consideration different from adults.

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INTRODUCTION

Unicystic Ameloblastoma (UA), a variant of ameloblastoma first described by Robinson and Martinez in 1977, refers to those cystic lesions that show clinical and radiologic characteristics of an odontogenic cyst but in histological examination show a typical ameloblastomatous epithelium lining part of the cyst cavity, with or without luminal and/or mural tumor proliferation (Gulten *et al*., 2008; Isha Goel *et al*., 2014; Richa Wadhawan, 2016; Shally Gupta, 2011). It accounts for 15% of all intraosseous ameloblastomas, and often affects the younger population with half of the cases occurring in the second and third decade of life (Shally Gupta *et al*., 2011; Shelly Arora *et al*., 2013; Zainab Chaudhary, 2012). UA have a slight male predilection and frequently originate from the posterior mandible.[3, 4] Mandible is more affected than maxilla at the ratio of 13: 1 (Shally Gupta *et al*., 2011). UA are characterized as a slow growing and relatively locally aggressive cystic lesion.

Corresponding author: **Dr. Shibin Shaju,**
Oral Medicine and Radiology, Vydehi Institute of Dental Sciences and Research Centre, #82, EPIP Area, Nallurahalli, Whitefield, Bangalore.

Radiographically, the lesions commonly show expansive unilocular radiolucency with a well-demarcated border. Approximately 50 to 80% of cases are associated with an impacted or unerupted tooth (Isha Goel *et al*., 2014; Ongkila Bhutia, 2013). Therefore, the clinical and radiographic presentations of UA are sometimes indistinguishable from those of dentigerous cysts (Ming-Hsuan Hsu, 2014). UA are further classified into three groups: 1) simple or luminal type (unilocular cyst lesion with lining epithelium showing features of an ameloblastoma); (2) intraluminal type (cystic lesion comprising intraluminal tumour nodules and odontogenic epithelium with a plexiform pattern, which resembles the one seen in the plexiform solid ameloblastoma; hence this lesion is also been termed as 'plexiform unicystic ameloblastoma' by several authors); and (3) mural or intramural type (with the presence of ameloblastomatous epithelium tumour islands in the cyst wall, which may (group 3b) or may not (group 3a) be attached to the cyst lining) (Seintou, 2014).

CASE REPORT

A 5 years old male patient came to the outpatient department of Oral Medicine and Radiology of Vydehi Institute of Dental

Sciences and Research Centre with the chief complaint of swelling and pain in the left back tooth region since 2 months. Patient's father gave the history of fall from bicycle 3 months back, followed by swelling and pain in left lower back tooth region. Swelling was initially very small in size and gradually increased to present size. Pain was dull aching and intermittent in nature. Pain was aggravated on chewing food and there were no relieving factors. He also gave the history of loss of tooth after a week of trauma in the same region which was firm before. On general physical examination the patient was moderately built and nourished and all the vital signs were within the normal limits.

On extra oral examination (Fig1), no abnormalities were seen on inspection. On palpation mild expansion of buccal cortical plate in relation to left side of mandible was noted. On intraoral inspection (Fig 2), 73 was missing which was exfoliated one week after trauma. A solitary sessile growth measuring of size 1 x 1 cm in dimension was seen in the edentulous space of 73 with surface having small erythematous areas. A Mild diffuse swelling was noted in relation to 74, 75 region in buccal sulcus measuring 1.5cm x 1.5cm in size with obliteration of buccal vestibule. The overlying and surrounding mucosa of the swelling appeared normal. On palpation, the soft tissue growth in relation to 73 and swelling in relation to 74, 75 were firm in consistency and non tender in nature. A mild buccal and lingual cortical plate expansion in relation to 74, 75 region was present. There was no bleeding present. The adjacent teeth were non tender, non carious and not mobile. Considering the above findings, provisional diagnosis of dentigerous cyst for swelling and reactive gingival hyperplasia for the growth in the 73 region were given.

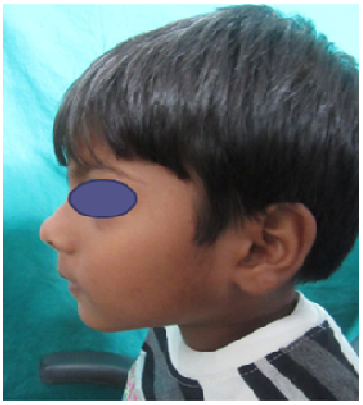

Fig 1. A mild diffuse swelling in the left lower 1/3rd of the face

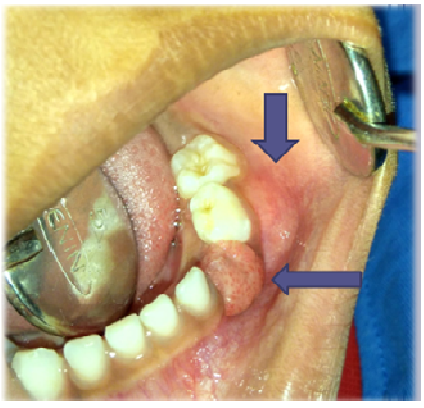

Fig 2. Solitary sessile growth seen in the edentulous space of 73 and mild diffuse swelling noted irt 74,75 region

The digital orthopantomogram (Fig 3) showed a cystic lesion with unilocular radiolucency involving developing crown of 33,34 extending superoinferiorly from alveolar crest to lower border of mandible, mesiodistally from 72 to 74 region measuring 3 x 2.5 cms in dimensions with scalloped borders with sclerotic margin in mesial aspect and diffused margin distally. Also showed root resorption of 74, 75 and developing tooth buds of 33, 34, 35, 36 and 37. Displacement of 33, 34, 35 was also noted.

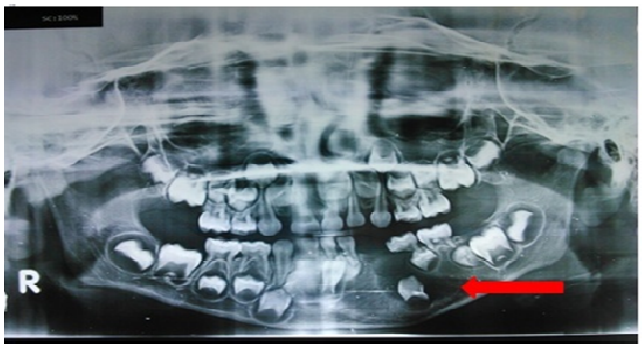

Fig 3. Orthopantogram showing unilocular radiolucency involving developing crown of 33,34 extending superoinferiorly from alveolar crest to lower border of mandible, mesiodistally from 72 to 74 region

Reformated panoramic CBCT (cone beam computed tomography) image (Fig 4) revealed inferiorly pushed nerve canal. Axial view (Fig 5) at the level of cementoenamel junction of all the teeth showed osteolytic lesion with maximum anteroposterior dimension of 26.0mm and buccolingual dimension of 15.7mm with both buccal and lingual cortical plate expansion and thinning. Perforation of buccal cortical plate in the premolar region also noted. Coronal view (Fig 6) at the level of 33, 34 showed the embedded 33 in the osteolytic lesion and buccally pushed 34 with maximum superioinferior dimension of 18.9mm with both buccal and lingual plate expansion and thinning.

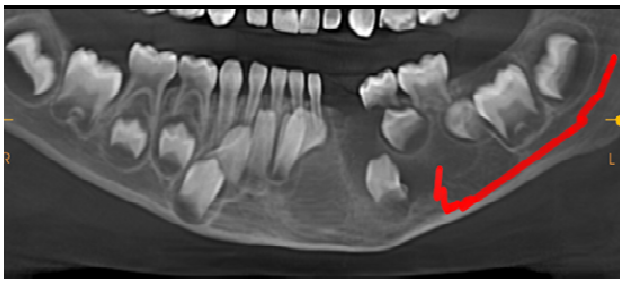

Fig 4. Reformated panoramic CBCT shows inferiorly pushed inferior alveolar nerve canal

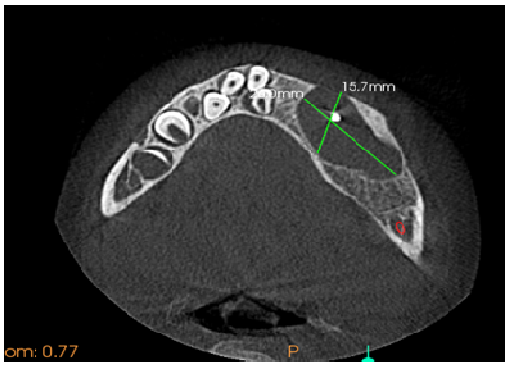

Fig 5. CBCT Axial view shows osteolytic lesion with both buccal and lingual cortical plate expansion and thinning. Perforation of buccal cortical plate in the premolar region also noted

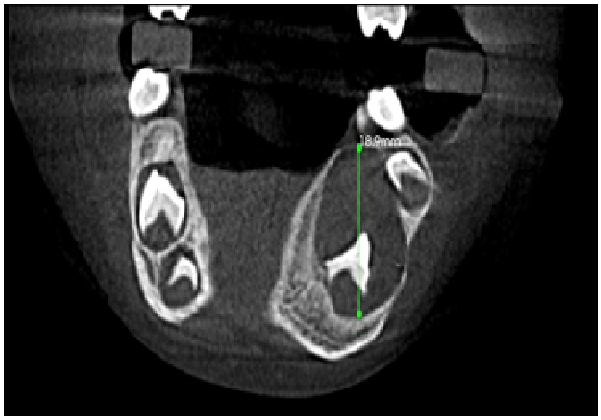

Fig 6. CBCT coronal view shows embedded 33 in the osteolytic lesion and buccally pushed 34 with both buccal and lingual plate expansion and thinning

Considering all radiological findings, the radiological differential diagnosis of dentigerous cyst and unicystic ameloblastoma were made. Excisional biopsy was done. The histopathological report showed all the 3 histopathologic variants of the unicystic ameloblastoma such as luminal, intraluminal and mural type. In luminal type (Fig 7) tumour was confined to the luminal surface of the cyst. Intraluminal type (Fig 8) showed nodular proliferation into the lumen without infiltration of tumour cells into the connective tissue wall. In mural type (Fig 9) invasive islands of ameloblastomatous epithelium in the connective tissue wall which are not involving the entire epithelium were seen. Based on these features, the final diagnosis of UA was made. The patient was under follow up for 19 months without any recurrence till date.

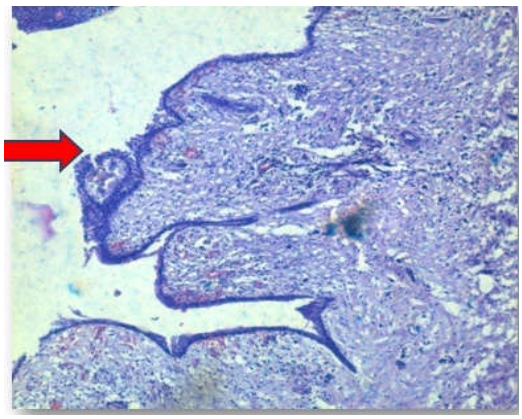

Fig 7. Photomicrograph of 10x shows Tumor confined to the luminal surface of the cyst

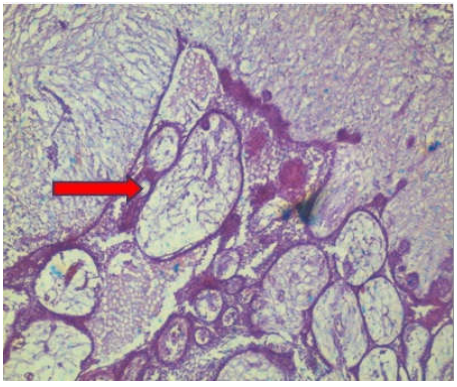

Fig 8. Photomicrograph of 10x shows nodular proliferation into the lumen without infiltration of tumor cells into the connective tissue wall

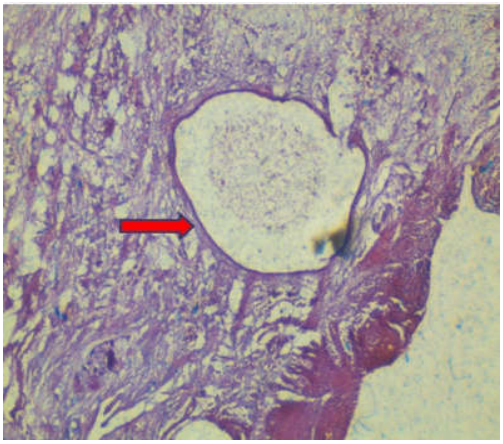

Fig 9. Photomicrograph of 10x shows invasive islands of ameloblastomatous epithelium in the connective tissue wall not involving the entire epithelium

DISCUSSION

Ameloblastoma is a benign, locally aggressive and infiltrative odontogenic neoplasm with a rare capacity to metastasize which comprises only 1.3% of all jaw cysts and tumours and 2nd most common odontogenic neoplasm constituting 10 % of neoplasm of odontogenic origin (Saravanakumar *et al*., 2014). UA is a disorder of odontogenesis with common clinical and radiographical manifestations with other odontogenic lesions such as dentigerous cyst, odontogenic keratocyst which makes diagnosis difficult (Unawane *et al*., 2011). It is a variant of ameloblastoma comprising of 10% to 15% of all intra bony lesion. Leider *et al* proposed three pathogenic mechanisms of evolution of UA:

- Reduced enamel epithelium associated with a developing tooth undergoes ameloblastic transformation with subsequent cystic development.
- Ameloblastomas arise in dentigerous cyst or other types of odontogenic cysts in which the neoplastic ameloblastic epithelium is preceded temporarily by non-neoplastic stratified squamous epithelial lining.
- Solid ameloblastoma undergoes cystic degeneration of ameloblastic islands with subsequent fusion of multiple microcysts and develops into a unicystic lesion (Shelly Arora, 2013).

It has been suggested that it arises as a result of neoplastic transformation of the epithelial lining of dentigerous cyst or any other type of odontogenic cyst. A high percentage of these lesions are associated with impacted tooth and most commonly cited provisional diagnosis is dentigerous cyst. The recurrence rate is low and thus indicating less aggressive nature. UA is characterized by one or more of the following features of Ackermann criteria.

Ackermann criteria (Sudhakara K Reddy, 2011):

Group 1: Luminal type (tumour confined to luminal surface of the cyst)

Group 2: Intra luminal type (nodular proliferation of the lumen without infiltration of tumour cells into the connective tissue wall)

Group 3: Mural type (invasive islands of ameloblastomatous epithelium in the connective tissue wall without involving the entire epithelium).

Commonly seen associated manifestations are painless swelling, facial asymmetry, and unilocular lesion with defined sclerotic borders, tooth impaction, displacement, mobility, root resorption, root divergence, occlusal interference and extrusion of tooth (Unawane, 2011). Thus the dentists are often get confused and turn into potential complications associated with incorrect diagnosis and complicated treatment of unicystic ameloblastoma (Unawane, 2011). Dentigerous cyst, odontogenic keratocyst, residual cyst, adenomatoid odontogenic tumor, giant cell lesion and sometimes solid ameloblastoma can be the commonly considered differential diagnosis for UA.

It is difficult to differentiate dentigerous cyst from UA.[7] However, following manifestations favors UA such as defect in the wall of cyst, unilocular cystic lesion extending into the ramus, expansion of both the buccal and lingual cortical plates. Tumour usually grows buccolingually, whereas the cyst grows towards most dependent part i.e. buccally, presence of erythematous and granulomatous tissue at the marginal gingiva. (Mucosal ulceration) with absence of bony cortex. Keratocyst usually spread anterio-posteriorly and seldom shows cortical expansion. On aspiration, keratocyst shows large amount of keratin. Residual cysts are associated with missing teeth that have been extracted. Adenomatoid odontogenic tumor is more seen in anterior maxilla whereas central giant lesion often arises anterior to first mandibular molar. Solid ameloblastoma is multilocular and seen uncommonly in patients less than 30 years of age.

UA is treated conservatively with decompression, enucleation and peripheral ostectomy as well as periodic long-term follow up. A more aggressive surgical approach may be considered when the condition recurs more than twice or by the patient's wish (Rafaela Scariot, 2012). The treatment of ameloblastoma is controversial. In children, the treatment is complicated by three factors: a) continuing facial growth, different bone physiology (greater percentage of cancellous bone, increased bone turnover and reactive periostium) and presence of unerupted teeth; b) difficulty in initial diagnosis; and c) predominance of the unicystic type of ameloblastoma (Rafaela Scariot, 2012). Unicystic ameloblastoma has been considered to be lesion with a comparatively less recurrent potential than the solid type, but the various subtypes of unicystic ameloblastoma have different prognostic features, the intraluminal subtype seem to be less aggressive than the intramural or mural subtype (Zainab Chaudhary, 2012).

Treatment modality of UA is being divided into following types.

- Enucleation alone can results in more recurrence rate among all treatments (30.5%). A better conservative approach is enucleation with application of Carnoy's solution and the extraction of closely related adjacent teeth which has the recurrence rate of 16%. The success of the application of Carnoy's solution after enucleation was thought to be due to both its penetration and fixation action. The usual practice is to apply the solution with cotton applicators or ribbon gauze for 3-5 min, rinse the bony cavity. The recurrence rate will lower more if the closely related teeth with tumour are extracted (Ongkila Bhutia, 2013).
- Marsupialization with other treatments can cause 18% of recurrence rate (Ongkila Bhutia, 2013).

The incidence rate, behavior and prognosis of the tumour in children make the surgical consideration different from adults. Hence conservative treatment should be the first choice for treating ameloblastoma in children. The treatment should be performed as early as possible after diagnosis in order to prevent possible proliferation into the adjacent tissues. Long-term follow-up is mandatory for UA since recurrence may take place years after removal. More than 50% of cases recur within 5 years after the surgery. Frequent postsurgical radiographical examinations favor early detection of recurrence.

Conclusion

Thus early diagnosis of UA in children is mandatory as it affects the growth of the jaw. The diagnosis of UA in children is difficult because most of the lesions radiographically resemble dentigerous cyst as 70 – 80% of cases are associated with unerupted tooth. It is a very rare event to see all three histological varieties in the same lesion like in our case which makes the case a rare entity. Only 2% of cases occur before the age of 10 years as our patient was of 5 years old. Hence as oral physicians, we must be vigilant regarding the rare presentation of such benign tumour in young age.

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