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UNICYSTIC AMELOBLASTOMA IN 11 YEARS OLD CHILD: A CASE REPORT

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ABSTRACT

Ameloblastoma is a benign odontogenic neoplasm, which frequently affects the mandible. The term ameloblastoma includes several clinico-radiological and histological types. Apart from the most commonly encountered clinico- pathologic models there are few variants, whose biological profile is unknown or not elicited. The reason for lack of understanding is the scarcity of case report published in the literature. Among the types, unicystic ameloblastoma is the least encountered either it presents as unilocular or multilocular radiolucency, but peculiar radiographic presentation of unilocular radiolucency in anterior maxillary region is extremely rare, which has not been reported yet. Here we report a distinctive case of unicystic ameloblastoma of right side of maxilla in a 11-year- old- boy with the radiographic presentation as mentioned above.

INTRODUCTION

Unicystic ameloblastoma (UCA) is considered as a variant of ameloblastoma and was described in the literature by Robinson and Martinez in the year 1977. It refers to a variant of cystic lesions that show clinical and radiologic characteristic similar to odontogenic cyst. Histologic examination typically represent ameloblastomatous epithelium lining entirely or part thereof, of the cystic cavity, which might show luminal and/or mural proliferation [1].

Tumors involving the jaws are usually benign and arise from odontogenic tissues or their remnants such as tissues of enamel organ, odontogenic rests of Malassez, reduced enamel epithelium and lining of odontogenic cysts [2]. Ameloblastoma is the most common form of aggressive benign tumors of the jaws [3]. This tumor is most commonly seen in molar ramus area of the mandible and is asymptomatic, slow growing and commonly associated with an impacted tooth [4]. The molar region is the common site for maxilla and might extend into the maxillary sinus, nasal cavity or base of the skull in isolated cases.[3] Its potential to grow to a copious size with severe bone deformity makes it a clinically important diagnosis.

It is more common in the second and third decade of life and is uncommon in children aged 10 years and younger [3,4]. Cystic variant behaves less aggressively and responds favorably to enucleation and curettage than its solid counterpart [4].

Case report:

An 11year old boy was brought by his parents to Department of Pedodontics and Preventive Dentistry. The parents were concerned about the appearance of a diffuse swelling over right side of child's face which gradually increased in size in past 6 months. The swelling appeared localized externally over the right nasolabial region and intraorally obliterating the nasolabial fold. The mass had expanded the maxillary bone and was hard on digital palpation due to the continuity of bony cortex (Figure 1).

A fine needle aspiration yielded a clear, straw-colored fluid. Histological analysis showed pus cells, few

red blood cells, and cholesterol crystals. The sample was inconsistent and biopsy was scheduled.

An orthopantomogram revealed a radiolucent lesion with well defined borders which included partially calcified tooth bud of the right maxillary lateral, canine, 1st and 2nd premolar (12,13,14,15) (Figure 2). The child was further subjected to a contrast enhanced computed tomography, to understand the extent of the lesion which also confirmed the cystic nature of the lesion, measuring approximately 2.8x1.7x1.6 cm at its maximum, and involved the region anteromedial to right maxillary sinus, cystic cavity was separated from right maxillary sinus by a thin bony septum (Figure 3a, 3b, 3c). The lesion seemed to expand the maxillary labial cortex and no expansion of the lateral nasal wall was noted. The lesion's posterior extent was not beyond the anterior border of vertical plate of palatine bone.

Considering the nature of the lesion, location, and age of the patient and radiological appearance a differential diagnosis was made that include dentigerous cyst, radicular cyst, primordial cyst and ameloblastoma. Local anesthesia was administered followed by extraction of 52,53,54,55 and marsupialization. Tissue obtained was sent for histopathological analysis. The site was left open for normal eruption of impacted tooth and an obturator was given to prevent any food lodgment. The biopsy report revealed a final diagnosis of Unicystic Ameloblastoma (UCA) with secondary infection.

A decision was made to enucleate the lesion under general anesthesia since its aggressive in nature. The entire cystic wall was excised along with the permanent tooth buds of 12,13,14,15 (Figure 4a, 4b). Due to high potential for its recurrence peripheral ostectomy was performed and Cornoys' solution was applied. The resected bony tissue was free of any ameloblastic infiltrations (Figure 5) and a functional removable partial denture was delivered and followed-up for 2 years (Figure 6a, 6b).

Figure 1. Preoperative intraoral view

Figure 2. Preoperative Orthopantomograph

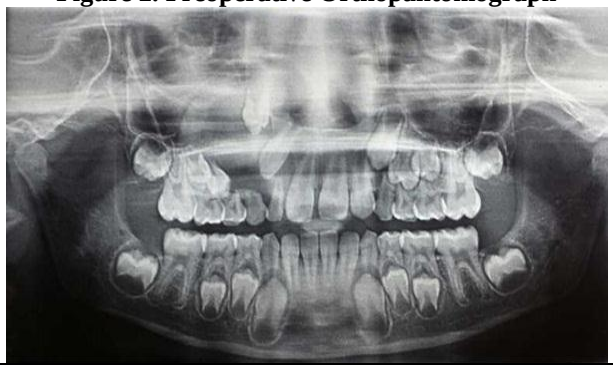
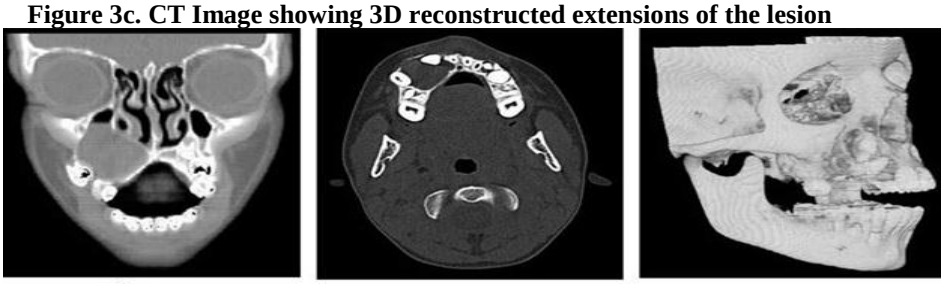
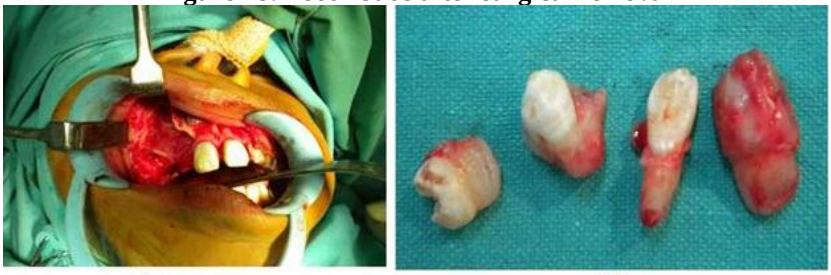


Figure 3a. CT Image showing superior-inferior extensions
Figure 3b. CT Image showing antero-posterior extensions
Figure 3c. CT Image showing 3D reconstructed extensions of the lesion



3 a 3 b 3 c

Figure 4a. Surgical defect after enucleation
Figure 4b. Tooth buds after surgical removal



4 a 4 b

Figure 5. Postoperative Orthopantomograph

Figure 6a. Surgical defect after healing
Figure 6b. Surgical defect corrected by prosthesis

6 a 6 b

DISCUSSION

Ameloblastoma is a true neoplasm of odontogenic epithelium. A review of 1,036 ameloblastomas of jaw found the average patient age being 38.9 years, with only 2.2% under 10 years, 8.7% between 10 and 19 years [5]. The unicystic ameloblastoma usually presents in the second decade of life, and the multicystic ameloblastoma in the third to fourth decades of life [6].

UCA occurs more commonly at younger age compared to multicystic ameloblastoma as in our case. The common representative site is in posterior mandible followed by parasymphysis region, anterior maxilla and posterior maxilla. The age is considerably lower and ranges from second to third decade (Reichart & Philipsen). In our case, the lesion occurred in the posterior maxilla in 11 year old, which is similar to the cases reported by Ackermann et al. and Paikkatt et al. [7].

UCA is a rare type of ameloblastoma, accounting for only 6% of all reported cases of ameloblastomas. Majority of cases are represented in the mandible with 50% to 80% of cases showing an association with impacted third molars. Patients present with swelling and facial asymmetry and pain not being a prominent symptom. Mucosal ulceration does not usually occur, but continued growth of the tumor will result in some patients. Small lesions are sometimes identified accidentally on routine radiographic screening or sometimes by clinical signs and symptoms like tooth mobility, occlusal aberrations and failure of tooth eruption inhibited by growing tumor [8].

The radiological features of ameloblastoma resemble many odontogenic and non-odontogenic cyst in the jaws. The bone is expanded, resembles a honeycomb or soap bubble and is eccentrically ballooned with a cystic appearance. There may be destruction or perforation of the cortex and a periosteal reaction with altered radiodensity. It may appear unilocular or multilocular radiolucent, radiopaque or mixed [6,8].

Histologically, the minimum criterion for diagnosing a lesion as UCA is the presence of a single cystic sac lined by odontogenic epithelium. UCA should be differentiated from odontogenic cysts because the former is far more aggressive and has a higher rate of recurrence [6]. Ackermann et al. Classified this entity into the following three histologic groups [3,6-8].

Group I: Luminal UCA (tumor confined to the luminal surface of the cyst)
Group II: Intraluminal/plexiform UCA (nodular proliferation into the lumen without infiltration of tumor cells into the connective tissue wall), and
Group III: Mural UCA (invasive islands of ameloblastomatous epithelium in the connective tissue wall not involving the entire epithelium).
Philipsen and Reichart classified histologically into subgroups [6,7].
Subgroup 1: Luminal UCA
Subgroup 1.2: Luminal and intraluminal
Subgroup 1.2.3: Luminal, intraluminal and intramural
Subgroup 1.3: Luminal and intramural

The UCAs diagnosed as subgroups 1 and 1.2 can be treated conservatively (careful enucleation), whereas subgroups 1.2.3 and 1.3 showing intramural growths require treated radical resection, as for a solid or multicystic ameloblastoma [3,4,8].

The present case showed features of Intraluminal proliferation but no infiltration of the cyst wall and hence was diagnosed as group Intraluminal UCA (Group II).

Several authors have thought the lesions with mural invasion should be treated aggressively with marginal or segmental resection whereas Group I and II are 100% cured by enucleation [3,4].

A study done by Ord et al. revealed that treatment of UCA in children is complicated by following factors:

- The continued facial growth.
- Different bone physiology (greater percentage of cancellous bone facilitating rapid spread, increased bone turnover, and reactive periosteum).
- Presence of unerupted teeth and difficulty in initial diagnosis [9].

CONCLUSION

Diagnosis and management of an unicystic ameloblastoma is significant because of its aggressive nature. A prompt diagnosis with histological confirmation in the early stages will reduce the extensive surgical intervention. The most common appearance can be deceiving and requires proper attention and ability to diagnose. In children, early management and regular follow up ensures the reduction of disfigurement and recurrence if any is managed on time.

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