***Case report***

# ANEURYSMAL BONE CYST OF MAXILLA MIMICKING DENTIGEROUS CYST: A RARE CASE REPORT


**Abstract:**

Aneurysmal bone cyst (ABC) is a rarebenign intraosseous lesion occurring most commonly in long bones and spine and characterized byblood-filled spaces of various sizes.The incidence of aneurysmal bone cyst in the craniofacial skeleton is rare which may remain undiagnosed for a long period prior to becoming symptomatic. In the jawbones, the mandible is involved more commonly than the maxilla.We present a rare case of aneurysmal bone cyst in maxilla in 16 year old male patient reporting to our department with complaint of painless swelling in upper right anterior region of jaw. Enucleation was done and histopathologically diagnosis was confirmed.We conclude that practitioners must have knowledge of aneurysmal bone cyst for correct diagnosis, investigation and effective management.



**Keywords:**

Aneurysmal bone cyst, Case report, Cyst, Enucleation, Impacted tooth, Maxilla,




## Introduction :

Aneurysmal bone cyst (ABC) has been described for the first time in 1942 by Jaffe and Lichtenstein.[1] In 2002, an ABC was defined by theWorld Health Organisation (WHO) as ‘a benign cysticlesion of bone composed of blood filled spacesseparated by connective tissue septa containing fibroblasts,osteoclast-type giant cells and reactive wovenbone’ [2]

Aneurysmal bone cysts (ABC) are typically found in long bones and the spine, but rarely can be seen in the craniofacial region. The incidence is 2% and 1.3% in the craniofacial region and maxillary region, respectively. [3] Available literature suggests that the mandible is affected more frequently than the maxilla, the proportions varying from 2:l to 11:9.[4]The etiology and pathogenesis of ABCs are still uncertain but are thought to be reactively related to previous trauma or pathology. The leading hypothesis etiology in ABC pathology is vascular and affected by local bone changes.[5]It may be clinically asymptomatic or may arise as a quickly advancing lesion enlarged to cause a pathological fracture.[6]Diagnosis should base on clinical symptoms, radiological, and histopathological examinations.A diagnosis of dentigerous cyst is usually entertained when impacted tooth is associated within a cyst. However, we report a rare case of aneurysmal bone cyst of maxilla with impacted maxillary right central incisor tooth within the cyst causing diagnostic dilemma. The purpose of this case report is to emphasize the importance of ABC diagnosis and effective treatment.

**Case Report:**

**Clinical history:**A 16 years old male patient reported to the department of oral medicine andradiology with complaint of painless swelling in upper right front region of jaw since 2 -3 months. Swelling was gradually increasing in the size to attain the present size. There was no history of trauma and no significant past medical or family history or known allergy.

**Clinical findings:**on extra oral examination, there was a diffuse swelling of right mid face of normal skin colour extending mediolaterally from ala of the nose towards tragus of ear and superoinferiorly from 1.5 cm below inferior orbital fissure till 1.5 cm below alatragus line. With obliterated nasolabial fold. Absence of extraoral fistula.On palpation swelling was nontender, soft in consistency,with smooth surface, aferbrile, nonfluctuant , noncompressible, nonreducible, nonpulsatile. No signs of paraesthesia reported. Intra oral hard tissue

examination showed missing teeth with 11, palatally displaced with 12, grade ii mobility with 11, 21, crowding with 21,22,23 and over retained with 63.Soft tissue examination revealed solitary, dome shaped, bluish red coloured swelling of size approximately 3 x 3 cm present on attached gingiva of 11,12,13. Obliteration of right upper labial vestibule was evident. Absence of any discharge with smooth surface texture and surrounding tissue appears to be normal. On palpation swelling was smooth, sessile, nontender, firm in consistency, nonfluctuant, noncompressible, nonreducible, nonpulsatile. [fig.1]

**Figure 1:**  **Preoperative image**

Based on the history and clinical examination a provisional diagnosis of dentigerous cyst with 11 was made. The differential diagnoses included AOT, ameloblastoma, aneurysmal bone cyst, CGCG, AV shunt.

**Diagnostic assessment:** FNAC was done in which 5ml brownish fluid smear revealed numerous foamy macrophages against the background of peripheral blood smear. No epithelial cells seen. [fig. 2]

**Figure 2: FNAC with 5ml brownish fluid**

Routine panoramic radiograph revealed well-defined unilocular radiolucency in maxillary right anterior region, roughly oval in shape of size approximately 4cm mesiodistally and 3 cm superoinferiorly. Internal structure of lesion shows impacted 11. Displacement of 12, 13 is evident. [fig 3]

**Figure 3: OPG**

CBCT scans overall radiographic impression was osteolytic lseion w r t right maxillary anterior and posterior region. [fig 4]

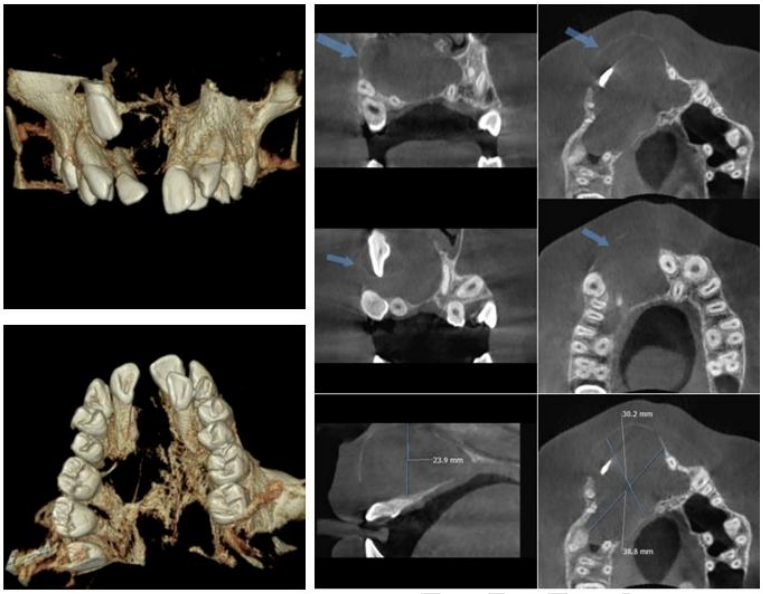
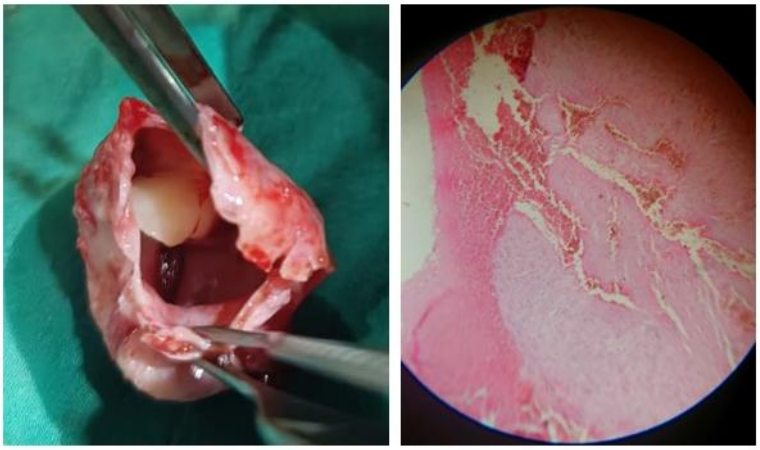


**Figure 4: CBCT**

D/d-adenomatoid odontogenic tumor, dentigerouous cyst, aneurysmal bone cyst, unicystic ameloblastoma

**Therapeutic intervention:**haematological investigations were within normal range. The treatment planned included extraction of 12 followed by enucleation of cyst under general anaesthesia.Resected tissue was then sent for histopathological examination which revealed no epithelial lining and multiple blood-filled spaces with stroma consisting of spindle shaped cells and multinucleate giant cells which was suggestive of aneurysmal bone cyst. [fig 5] Hence a final diagnosis of ABC was made. Patient condition was improved in follow up after 20 days.



(a) (b)

**Figure 5: (a) Excised tissue, (b) Histopathology**

**Discussion:**

An aneurysmal bone cyst (ABC) is a pseudocyst due to the absence of a cyst epithelial. Bernier and Bhaskarreported the first ABC case containing the maxillofacial skeleton in 1958.[5] Jaffe and Lichtenstein defined it in 1942 as blood-filled lesions in which giant cells and irregular bone trabeculae.[1] More than 90% of ABC found in the jaws occur inTheposterior areas, especially in the mandible.[7]in our case it was found in maxillary anterior region. The pathogenesis of ABC is still controversial. The role of trauma has been suggested by few authors. [1]Some authors favored intra medullary hematoma as the cause of ABC. [8]Local circulatory abnormalities leading to increased venous pressure and resulting in dilation of the local vascular network is one of the most widely accepted theories available in literature.[9]Recently, chromosomal translocation *t* (16;17)(q22;p13) as a recurrent cytogenetic abnormality has also been suggested as a responsible factor for primary ABCs.[10] Aneurysmal bone cysts are located most commonly in the shaft of a long bone or in the vertebral column. It is generally found in patients younger than the age of 20. No significant gender predilection is noted.[11]present case was16 year old male patient. These lesions can have multiple clinical features, from an asymptomatic lesion discovered on routine X-ray to a sometimes expansive lesion. However, the main symptom is a painless swelling of the jaw. Additional symptoms, such as limited mouth opening, loosening of teeth, nasal obstruction

and lip paraesthesia are described in literature.[12]Radiology is suggestive but not diagnostic for aneurysmal bone cyst.[11] Radiographically it may appear as expansive, osteolytic, unilocular or multilocular radiolucent lesion, with expansion and thinning of the surrounding cortical bone. This variegated appearance in imaging could be due to the various types of aneurismal bone cyst (solid, vascular and mixed).[13] The radiographic appearance of maxillary aneurysmal bone cysts is even less characteristic than that of mandibular lesion. CT scan ABC may appear unicystic, multilocular, or moth eaten, causing expansion, perforation, or extensive destruction of the bony cortices.[14] Histologically ABC is characterized by blood-filled spaces of various sizes and channels divided by connective tissue septa, which can contain osteoid tissue, osteoclast-like giant cellsand woven bone.[7] Three types of ABC have been described based on histopathological features.[15] Solid type (5% of the cases) is characterized by a dense stroma, scanty sinusoids, few blood vessels and caverns, bone expansion (instead of perforation), and without severe bleeding during surgery. Vascular variant (95% of cases) is characterized by a loose scanty stroma, numerous engorged blood filled sinusoids and caverns. Brisk bleeding during surgery and extensive bony destruction with spread in the soft tissues are also obvious. The mixed type lies between the 2 previous variants.[15] Our case was of solid variant type as it was not associated with severe bleeding during surgery. Surgical therapy is the most frequently applied treatment of ABCs,[6] although embolization, cryotherapy, and wait and see strategy have also been used as other treatment modalities.[4] Recurrence rates range from 20% to 30% according to different series and seems to occur most frequently within the first year after surgery.[4,11] Several authors recommend immediate reconstruction of the defect with autogenous grafts in cases of aesthetic deformity and in cases with high risk of fractures.[11]Considering the aggressive nature of the lesion and age of the patient, a conservative surgical resection of the lesion was done.

**Conclusion:**

ABCs of maxilla are rarer. Impacted tooth within aneurysmal bone cyst leads to diagnostic dilemma. biopsy and aspiration may help in a definitive diagnosis over its confusing clinical and radiological features which are similar to various other pathologic entities of the jaws. Conservative surgical resection is preferable to curettage as the latter is associated with much higher rate of recurrence.

**CONCENT:**

Patient concent was taken

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