**A Saga of Two Aneurysmal Bone Cyst Cases: An Institutional Case Report**

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**Abstract**

Aneurysmal bone cyst (ABC) is a benign osteolytic lesion mainly seen in the long bones and vertebrae but very rare in jaw. It is associated with rapid growth pattern, asymmetry of the face, and variable radiographic appearance which makes histopathologic evaluation a very important tool in its precise detection. Here, we present two cases of ABC are being described one of which occurred in a 46-year-old female, in the left side of mandible which was treated with curettage and extraction of involved and resorbed teeth. The other one occurred in a 44-year-old woman in both sides of the mandible, treated by curettage and extraction of involved tooth followed by platelet-rich fibrin bone graft.

**Keywords:** Aneurysmal bone cyst, benign bone lesions, mandible, pseudocysts

**INTRODUCTION**

The World Health Organization defines aneurysmal bone cysts (ABCs) as “a benign tumor-like, expansive osteolytic lesions consisting of blood-filled spaces and channels divided by connective tissue septa that can contain osteoid tissue and osteoclastic giant cells. In 1893, ABC was first identified and described as ossifying hematoma by Van Arsdale.[1] The credit for recognizing ABC as an intraosseous osteolytic lesion chiefly affecting the metaphyseal region of the long bones and vertebrae goes to Jaffe and Lichtenstein. Bhaskar and Bernier were the first to report ABC in the jaws in 1958.[2,3] The ABC is a relatively rare, benign osteolytic lesion, and considered to be a pseudocyst due to the lack of epithelial lining.[4,5] Maximum (50%) cases of ABC arise in long bone, 20% in vertebral column and about 1.5% of ABCs are nonodontogenic, nonepithelial cysts of mandible.[6,7] Mandible is affected more than maxilla (3:1) with a preference for body, ramus, and angle of mandible.[8] It can be classified into three types: (1) Conventional or vascular type showing rapidly growing, expansive as well as destructive lesion causing cortical plate perforation and soft-tissue invasion. (2) The solid type is detected as small, asymptomatic lesion on the routine radiograph.[9,10] (3) Mixed type showing features of both type and may be due to transitory intermediate phase.[10]

**Case Reports**

**Case report 1**

A 46-year-old female patient reported to our Department of Oral and Maxillofacial Surgery with a complaint of intermittent pain and swelling in the left lower teeth region since 3 years, which gradually has attained the present size. Her medical and family history was not of much significance. History of trauma was provided by the patient in that region 5 years back. Extraoral examination revealed facial asymmetry and diffuse swelling involving the left side of the lower jaw measuring about 5 cm × 2 cm, which was firm and tender on palpation [Figure 1]. Intraoral examination showed obliteration of vestibular region along with buccal and lingual cortical bone expansion, extending from lower left canine to first molar region [Figure 2]. Grade 1 mobility was observed in the left lower first and second premolar. Radiographic examination revealed large unicocular radiolucency extending from left lower canine to first molar region along with root resorption [Figure 3]. After the removal of the involved...
teeth, curettage of the lesion was performed under general anesthesia [Figure 4]. On histopathologic evaluation of the specimen, numerous, variable sized vascular spaces lined by endothelial cells was seen, containing numerous red blood cells (RBCs) but the presence of giant cells could not be ascertained [Figures 5 and 6].

Postoperative 1-week review was uneventful and swelling of the left cheek was subsided. Subsequent regular reviews up to 1 year were uneventful with no sign of recurrence observed. The patient is still under regular follow-up.

**Case report 2**

A 44-year-old female patient reported to our Department of Oral and Maxillofacial Surgery with a complaint of intermittent pain and swelling in the left and right lower teeth region since 5 years. She had no relevant medical and family history. The patient had a history of trauma in the region of swelling 10 years back. Extraoral examination revealed facial asymmetry and diffuse swelling involving the left and right side of the lower jaw measuring approximately 10 cm × 2 cm, which was firm and tender on palpation [Figure 7]. Intraoral examination showed obliteration of vestibular region along with buccal and lingual cortical bone expansion, extending from lower left first molar region to lower right third molar region. Grade 1 mobility was observed in some tooth. Pericoronitis was present in left and right third molar region. Radiographic examination revealed large unilocular radiolucency extending from lower left first molar to lower right third molar region [Figure 8].

After the removal of the mobile teeth and third molars of both the sides, curettage of the lesion was performed under general anesthesia followed by the usage of platelet-rich fibrin (PRF) bone graft for the resultant bony defect [Figure 9]. Histopathologic evaluation of the specimen did not ascertain the presence of giant cells, but numerous, variable sized vascular spaces lined by endothelial cells, containing numerous RBCs was seen in Figures 10 and 11. Postoperative 1-week review was uneventful and the swelling was subsided. Subsequent regular follow-up for 2 years was uneventful with no sign of recurrence. The patient is still under regular follow-up.

**Discussion**

The word “aneurysmatic” is used to describe the blown-out appearance of the bone which is a common clinical finding in these type of lesions.

It is a well-known fact that due to lacking of an epithelial lining ABC is categorized as pseudocyst. ABC constitutes only 5% of the bony lesions occurring in maxillofacial region. The low occurrence rate of ABC in skull bones is thought to be due to the low venous...
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pressure of the skull bones. Because, ABC generally occurs in the areas of the skeleton with high venous pressure and high marrow content.\[12\] Many theories, describing the origin of ABC, have been proposed by researchers worldwide. The first theory was proposed by Jaffe and Lichtenstein according to which increased venous pressure leads to engorgement of vascular bed and finally resorption that is caused by alteration in local hemodynamics.\[13,14\] Steiner and Kantor\[15\] have suggested that ABC can occur as primary or secondary lesion associated with other bone diseases, whereas Struthers and Shear\[16\] have arrived to the conclusion that ABC can occur as a secondary phenomenon in a preexisting lesion and giant cell granuloma. According to Levy et al.,\[17\] an essential factor in the development of ABC is a history of trauma and subperiosteal hematoma formation. However, Tillman et al.,\[18\] have reported no history of trauma in 95 cases. According to Hernandez et al.,\[19\] ABC can be divided into primary and secondary - (i) primary could be congenital or acquired in adults with a history of trauma and (ii) the congenital is seen in children and young adults with no history. Degeneration of preexisting lesion such as cyst, tumor or fibro-osseous lesions is thought to be associated with the secondary.\[19\] In our first case, there was a history of trauma 5 years back, and 10 years back in our second case enabling us to designate it as etiologic factor. Researchers like Panoutsakopoulos et al., described chromosomal anomalies involving band 16q22 in three cases.

Others have reported familial incidence in their literature. Only 1.9% of ABCs are found to be occurring in jaws; the most common locations are long bones and vertebral column. There are some reported cases of ABC in unusual locations such as mandibular condyle and coronoid process.\[10,13\] ABC is known for its variable clinical presentation, ranging from a small, asymptomatic lesion to rapidly growing, expansible, destructive lesion causing pain, swelling, deformity, neurologic symptoms, pathologic fracture, and perforation of the cortical bone.\[10\] Radiographic appearances of ABC vary from unilocular to multilocular appearance, may be of soap bubble or honeycomb appearance. It may show radiolucency, radiopacity, or mixed appearance.\[11\] Root resorption of the affected teeth may be other radiological finding. Unilocular radiolucency along with root resorption of the affected tooth was observed in both our cases. Histological features of ABC comprise of numerous sinusoidal blood-filled spaces of variable shape, placed in a fibrous stroma with multinucleated giant cells and osteoid. Variable amounts of hemosiderin pigmentation may be present. These features were seen in both of our case except for giant cells which could not be located. Osteoid and
calcifying fibro-myxoid tissue is seen in solid type. Features of both vascular and solid type of lesion are seen in the mixed form. Currently, intralesional curettage is the standard of care and most widely employed treatment. However, tumor recurrence with curettage alone is common and has driven some to propose a multitude of adjuvants with varying efficacy and risk profiles. Historically, therapies such as en bloc resection or radiation therapy were utilized as an alternative to decrease the recurrence rate, but these therapies imposed high morbidity.\textsuperscript{14} En bloc excision though has the lowest rates of recurrence but costs high morbidity (postoperative pain, limb length discrepancies, muscle weakness, and decreased ranges of motion) to the patient.\textsuperscript{20} Recrudescence and inoperable ABC lesions has been historically treated by radiotherapy.\textsuperscript{21} However, radiotherapy entails a significant amount of risks, radiation-induced sarcoma has been reported in many cases. The standard of care for ABCs is curettage with or without bone-graft depending on the resultant void. Some studies have showing recurrence rates as high as 59% even with the best efforts at curettage.\textsuperscript{22} As a result, various adjuvants have evolved to reduce recurrence including the use of cement, high-speed bur, argon beam, phenol, and cryotherapy. No significant effect of adjuvant phenol or high-speed bur could be established in a recent study by Kececi \textit{et al.} in 85 patients.\textsuperscript{22} On the other hand, superior control rate of about 97% by the use of high-speed bur is reported by Wang \textit{et al.}\textsuperscript{23} In the same line, Garg \textit{et al.} demonstrated that recurrence rate can be significantly reduced by the use of a high-speed bur combined to electrocautery when compared to simple curettage.\textsuperscript{24} It is proven that local relapse can be better prevented by the use of bone cement (polymethylmethacrylate) than bone grafting.\textsuperscript{25} In a study done by Peeters \textit{et al.} in 80 patients has shown relapses in only 5% of the patients.\textsuperscript{20} Even more sophisticated techniques such as argon beam therapy which relies on a coagulation effect have also been used. This method has shown better elimination of the risk for
local recurrence as compared to simple curettage and. Yet, according to the observations of Steffner et al., the overall complication rate with argon beam laser was 19%, as compared to 6% when high-speed bur was used.[29]

**Conclusion**

ABCs are aggressive benign lesions with high rates of recurrence rendering its treatment uniquely challenging. The standard of treatment remains curettage and grafting to fill the bone void, but the adjuvant or alternative treatment methods to reduce recurrence are numerous.[29] Here, we discuss two cases of ABC of mandible in a 46-year-old female treated by curettage and extraction of involved and resorbed teeth and a 44-year-old female treated by curettage and extraction of involved teeth followed by PRF bone graft.

**Declaration of patient consent**
The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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**Conflicts of interest**
There are no conflicts of interest.

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