

Case Report

Aneurysmal Bone Cyst of the Mandible: A Case Report

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Abstract:

Background: Aneurysmal bone cyst (ABC) is a rare, benign, non-neoplastic, expansile osteolytic lesion characterized by blood-filled spaces separated by fibrous septa. While ABCs commonly occur in the metaphyseal regions of long bones and the spine, involvement of the craniofacial skeleton is uncommon, representing approximately 1-2% of all reported cases. The pathogenesis of ABC remains incompletely understood, with proposed mechanisms including reactive vascular malformation, post-traumatic origin, and genetic susceptibility involving USP6 gene rearrangements.

Case Presentation: A 15-year-old Libyan female presented with an incidental finding of a painless swelling in the left posterior mandible during a routine dental examination. Panoramic radiography revealed a well-defined, unilocular radiolucent lesion extending from the left first molar to the left third molar, with no root resorption. Cone-beam computed tomography demonstrated cortical thinning and buccolingual expansion, without perforation or involvement of the inferior alveolar canal. Fine needle aspiration yielded blood, prompting CT angiography, which excluded vascular malformations. The patient underwent surgical curettage with primary closure under general anaesthesia. Intraoperatively, the lesion exhibited characteristic bluish discoloration and blood-filled spaces. Histopathological examination confirmed the diagnosis of ABC.

Conclusion: This case highlights the importance of a structured diagnostic approach combining appropriate imaging and, when indicated, CT angiography to exclude vascular lesions before surgical intervention. Thorough curettage remains an effective treatment modality for ABCs of the mandible, with excellent bone healing observed a six-month follow-up.

Keywords: Aneurysmal bone cyst, mandible, cone-beam computed tomography, case report, Libya

Introduction:

Aneurysmal bone cyst (ABC) is a rare, benign, non-neoplastic, expansile osteolytic lesion characterized by blood-filled spaces separated by fibrous septa containing multinucleated giant cells [1]. The term is descriptive: "aneurysmal" refers to the marked bony expansion, while "cyst" denotes the fluid-filled cavities. However, ABC is more accurately considered a pseudocyst because it lacks an epithelial lining on histological examination [2]. Earlier classifications, including the 2020 WHO system, described ABC as a benign osteoclastic tumor with prominent giant cells. In the more recent WHO classification of odontogenic and maxillofacial bone tumors, ABC is categorized under "giant cell lesions and bone cysts" [3]. ABCs predominantly affect the metaphyseal regions of long bones, particularly the femur and tibia, which account for over 50% of cases, followed by the spine at 12-30% [4,5]. Involvement of the craniofacial skeleton is notably rare, representing only 1-2% of all reported ABCs [1]. Within the jaws, the mandible is affected more frequently than the maxilla, with a reported ratio of approximately 2:1 to 11:9 [6]. The body of the mandible and the ramus are most commonly involved, while condylar and coronoid involvement is uncommon [7]. ABCs typically present during the first two decades of life and demonstrate a slight female predilection [6,8].

The exact etiology and pathogenesis of ABCs remain incompletely understood. Multiple theories have been proposed, including post-traumatic origin, reactive vascular malformation resulting from local circulatory disturbance, and genetic predisposition [9]. Notably, cytogenetic studies have identified chromosomal translocation t(16;17)(q22;p13) in a subset of cases, resulting in fusion of the USP6 gene with CDH11 [10]. Oliveira et al. demonstrated that approximately 70% of primary ABCs demonstrate USP6 rearrangements, with CDH11 being the most frequent partner gene [11]. Furthermore, ABCs may arise as primary lesions or develop secondarily in association with other bone pathologies, including fibrous dysplasia, giant cell tumor, osteoblastoma, and odontogenic cysts [12].

Clinical presentation of ABCs in the jaws varies considerably. Patients may be asymptomatic, with lesions discovered incidentally during routine radiographic examination, or may present with progressive swelling, pain, facial asymmetry, tooth displacement, root resorption, or paresthesia [13]. The growth rate can range from indolent progression over years to rapid expansion over weeks to months [14]. Pathological fracture, while less common in gnathic bones than in weight-bearing long bones, may occur in advanced cases [4].

Radiographically, ABCs typically appear as radiolucent, expansile lesions that may be unilocular or multilocular,

often with characteristic "soap bubble" or "honeycomb" appearances [6]. Cortical thinning and expansion are common, and root resorption of adjacent teeth may be observed [13]. Fluid-fluid levels, while not pathognomonic, are a suggestive finding on magnetic resonance imaging and may occasionally be visualized on computed tomography [15].

The differential diagnosis for radiolucent lesions of the jaws is extensive and includes odontogenic keratocyst, ameloblastoma, central giant cell granuloma, traumatic bone cyst, and vascular lesions such as central hemangioma and arteriovenous malformation [8]. Distinguishing ABC from these entities requires careful correlation of clinical, radiographic, and histopathological findings.

Therapeutic management of ABCs remains somewhat controversial, with treatment selection guided by lesion location, size, extent, and patient age [5]. Surgical options include simple curettage, curettage with adjuvant therapy, en bloc resection, and selective arterial embolisation [9]. Recurrence rates following curettage alone range from 10-59%, with lower rates reported when adjuvant measures are employed [9].

This case report presents the diagnosis and surgical management of an aneurysmal bone cyst involving the posterior mandible in a young Libyan adult. The report emphasises the critical role of CBCT in preoperative assessment, the importance of excluding vascular lesions before surgical intervention, and the characteristic

histopathological features that establish a definitive diagnosis.

Case Report:

A 15-year-old Libyan female presented to the dental clinic with an incidental finding noted during a routine dental examination. The patient reported awareness of a mild, painless swelling in the left posterior mandibular region but denied any history of trauma, significant discomfort, or sensory disturbance. Her medical and dental history was otherwise unremarkable, with no known systemic conditions or medications.

Clinical Examination:

Extraoral examination revealed no facial asymmetry, lymphadenopathy, or visible swelling. Intraoral examination demonstrated a subtle buccal expansion in the left posterior mandible in the region of the mandibular molars. The overlying mucosa appeared normal in color and texture, with no evidence of erythema, ulceration, or infection. All teeth in the affected quadrant responded positively to electric pulp vitality testing. No mobility, percussion sensitivity, or periodontal abnormalities were noted.

Radiographic Findings:

Panoramic radiography revealed a well-defined radiolucent lesion in the left posterior mandible associated with the apices of the mandibular left first molar extending posteriorly to the left third molar. The lesion appeared unilocular with well-defined borders and did not cause root resorption of the involved teeth (Figure 1).



Figure 1. Panoramic radiograph showing a well-defined, unilocular radiolucent lesion extending from the mesial root of the mandibular left first molar to the left third molar. Note the absence of root resorption of the involved teeth.

For detailed characterization, cone-beam computed tomography (CBCT) was performed. CBCT imaging demonstrated a unilocular radiolucent lesion measuring approximately $18 \times 14 \times 10$ mm with the following features: well-defined and corticated margins, scalloping between the roots of the mandibular first and second molars, buccolingual expansion with thinning of both buccal and lingual cortices, absence of cortical perforation, and no involvement of the inferior alveolar nerve canal.

Differential Diagnosis and Diagnostic Workup:

Based on the clinical and radiographic presentation, the initial differential diagnosis included traumatic bone cyst, odontogenic keratocyst, and aneurysmal bone cyst. Given granuloma was also considered. Fine needle aspiration (FNA) was carefully performed using a 22-gauge needle and yielded frank blood (Figure 2), raising concern for a vascular lesion.

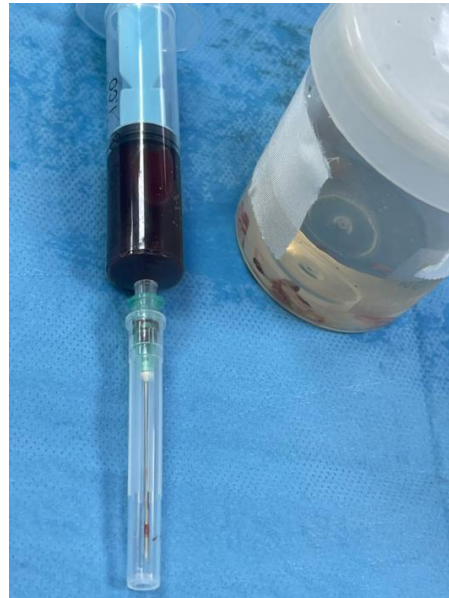


Figure 2. Fine needle aspiration of the lesion yielded frank blood, raising suspicion for a vascular lesion.

Consequently, CT angiography was requested to exclude central hemangioma and arteriovenous malformation. CT angiography confirmed the diagnosis of aneurysmal bone cyst and definitively excluded high-flow vascular lesions.

The technique: axial images were obtained through the mandible with the use of intravenous contrast.

Report: CT of the mandible showing a 3 cm left-sided non-enhanced lytic lesion, expansile, full of dense fluid (blood), preserving thin laminar cortical, affecting much of the left mandible, encasing the root of the molars. Findings

are consistent with an aneurysmal bone cyst of the mandible.

Surgical Management:

The patient underwent surgical intervention under general anaesthesia. Following standard aseptic preparation, a full-thickness mucoperiosteal flap was elevated to expose the buccal cortex. Intraoperatively, the lesion exhibited a bluish discoloration visible through the thinned cortical bone (Figure3). A bony window was created using a surgical bur under copious saline irrigation.



Figure 3. Intraoperative photograph showing bluish discoloration visible through the thinned buccal cortical bone prior to window creation.

Upon entering the lesion, the cavity was found to be filled with blood-filled spaces, consistent with the vascular nature of an aneurysmal bone cyst. The lesion lacked an epithelial lining and demonstrated active scalloping between roots, cortical thinning with buccolingual expansion, no cortical perforation, and no involvement of

the inferior alveolar canal (Figure 4). The lesion was meticulously curetted, and the surgical cavity was thoroughly irrigated with sterile saline. The specimen was submitted for histopathological examination. Primary closure was achieved with interrupted sutures.



Figure 4. Surgical exposure of the lesion demonstrating blood-filled cystic spaces, scalloping between roots, cortical thinning with buccolingual expansion, no cortical perforation, and no involvement of the inferior alveolar canal.



Figure 5. Immediate postoperative view after completion of suturing .

Histopathological Findings:

Gross examination revealed multiple fragments of soft tissue and bone, reddish-brown in color and hemorrhagic in appearance. Microscopic examination demonstrated blood-filled cavernous spaces of varying sizes separated by fibrous septa. These spaces lacked an endothelial or epithelial lining. The fibrous septa contained numerous

multinucleated giant cells, spindle-shaped fibroblasts, and areas of reactive woven bone formation. Hemosiderin pigment deposition was noted throughout the specimen. No cytological atypia or malignant features were identified (Figure 6). These findings were consistent with the diagnosis of aneurysmal bone cyst.

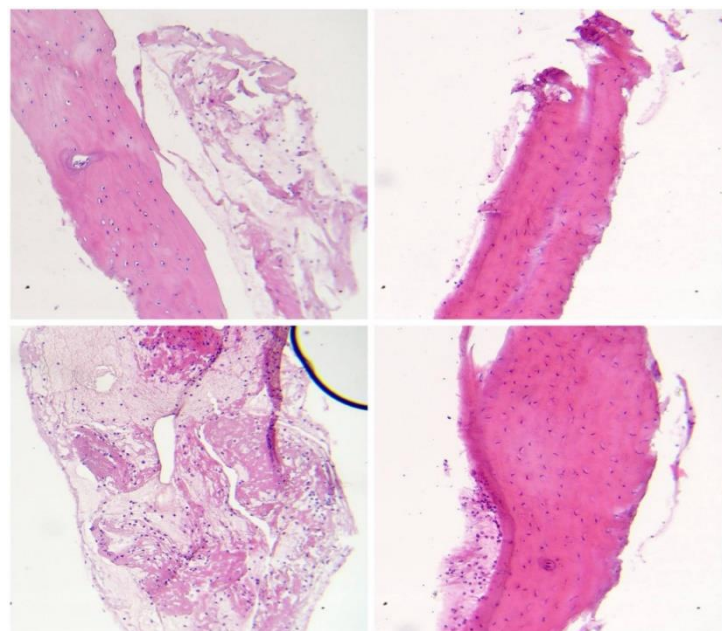


Figure 6. Histopathological photomicrograph demonstrating blood-filled spaces separated by fibrous septa containing multinucleated giant cells, consistent with aneurysmal bone cyst (H&E stain).



Postoperative Course:

The patient recovered uneventfully and was discharged on postoperative day one with appropriate analgesic coverage. At one-week follow-up, the surgical site demonstrated satisfactory healing with no signs of infection or dehiscence. At six-month follow-up, panoramic radiography revealed complete bone fill of the surgical defect with normal trabecular pattern. The patient remained asymptomatic with no clinical or radiographic evidence of recurrence.

Discussion:

Aneurysmal bone cyst of the jaws represents a diagnostic challenge due to its rarity and radiographic similarity to other osteolytic lesions of the maxillofacial region. With only

1-2% of ABCs occurring in the craniofacial skeleton, individual clinicians encounter these lesions infrequently, underscoring the importance of case reports in contributing to the collective understanding of this entity [1].

The pathogenesis of ABC remains a subject of ongoing investigation. The traumatic theory suggests that intraosseous hemorrhage following injury may initiate a reactive process leading to cyst formation [9]. Alternatively, the vascular theory proposes that local circulatory disturbances create increased venous pressure, resulting in expansion of the vascular network and subsequent bone resorption [4]. More recent evidence supports a genetic component, with the identification of USP6 gene rearrangements in a significant proportion of primary ABCs [10]. Oliveira et al. demonstrated USP6 rearrangements in 69% of primary ABCs analyzed, while no secondary ABCs showed these alterations [11]. This genetic distinction has diagnostic implications and supports the classification of primary ABC as a neoplastic rather than purely reactive process.

In the present case, the lesion was primary in nature with no evidence of an associated precursor lesion. The young age of the patient (15 years) aligns with the established epidemiology of ABCs, which predominantly affect individuals in the first two decades of life [6]. The posterior mandibular location is also consistent with the literature. Urs et al. reported that among gnathic ABCs, the mandible was involved in 71% of cases, with the body and ramus being the most frequent subsites [9].

The diagnostic approach in this case highlights several important considerations. First, the value of CBCT in characterizing jaw lesions cannot be overstated. CBCT provided detailed three-dimensional information regarding lesion extent, cortical integrity, root relationship, and proximity to the inferior alveolar canal, all of which informed surgical planning [2]. Second, the finding of blood upon FNA prompted CT angiography, a critical step that excluded vascular lesions before surgical intervention. Attempting curettage of a vascular malformation misdiagnosed as an ABC could result in catastrophic intraoperative hemorrhage. As noted by Sun et al., the presence of blood on aspiration should always raise suspicion for vascular lesions and prompt appropriate imaging [13]. Third, definitive diagnosis required histopathological examination, as radiographic features alone are insufficient to distinguish ABC from other osteolytic lesions.

Histopathologically, ABC is characterized by blood-filled cystic spaces lacking endothelial or epithelial lining, separated by fibrous septa containing multinucleated giant

cells, fibroblasts, and hemosiderin deposits [3]. The multinucleated giant cells in ABC are reactive osteoclast-like cells and should be distinguished from the neoplastic giant cells seen in giant cell tumor of bone. Solid variant ABCs, which contain a predominance of solid tissue with fewer cystic spaces, may pose additional diagnostic difficulty and require distinction from other giant cell-containing lesions such as central giant cell granuloma [12].

The intraoperative findings in this case, specifically the bluish discoloration visible through thinned cortical bone and the blood-filled cystic spaces encountered upon entry, are characteristic of ABC and consistent with descriptions in the literature [7]. The absence of cortical perforation and inferior alveolar canal involvement were favorable prognostic indicators that permitted conservative surgical management.

Treatment of ABCs in the jaws has evolved over time. Historically, radiation therapy was employed, but this modality has largely been abandoned due to the risk of post-irradiation sarcoma [4]. Currently, surgical curettage with or without bone grafting represents the mainstay of treatment for accessible lesions [5]. Adjuvant measures such as phenolization or cryotherapy may reduce recurrence rates by addressing microscopic residual disease. En bloc resection is reserved for extensive or recurrent lesions, or those involving expendable bone [5]. A systematic review by Cottalorda et al. reported recurrence rates ranging from 10% to 59% following curettage alone, with most recurrences occurring within the first two years following surgery [4]. Long-term clinical and radiographic follow-up is therefore essential.

Alternative treatment modalities have emerged for lesions that are surgically challenging. Denosumab, a RANKL inhibitor, has shown efficacy in reducing lesion size and facilitating subsequent surgery in ABCs of the spine and pelvis [14]. Selective arterial embolization has been successfully employed for ABCs in locations where surgery carries high morbidity [5]. Percutaneous sclerotherapy using polidocanol or doxycycline represents another minimally invasive option [15].

The present case demonstrated a favorable outcome with complete resolution following thorough curettage and primary closure. Immediate postoperative imaging confirmed complete removal, and at six-month follow-up, complete bone fill of the surgical defect was observed with no evidence of recurrence. Continued surveillance is planned to monitor for late recurrence, as cases have been reported up to two years postoperatively [9]. The successful outcome in this case reinforces that for appropriately selected ABCs of the jaws, conservative surgical curettage remains an effective and appropriate treatment modality.

Conclusion:

Aneurysmal bone cyst of the mandible, though rare, should be considered in the differential diagnosis of radiolucent jaw lesions, particularly in young patients. This case emphasizes the critical importance of a structured and systematic diagnostic approach encompassing clinical examination, vitality testing, appropriate imaging including CBCT, FNA when indicated, and CT angiography to exclude vascular lesions prior to surgical intervention. The characteristic intraoperative findings of bluish cortical discoloration and blood-filled cystic spaces, combined with confirmatory histopathology, establish the



diagnosis. Thorough curettage remains an effective and appropriate treatment modality for appropriately selected ABCs of the jaws. Long-term follow-up is necessary to monitor for recurrence and ensure optimal patient outcomes.

Conflict of Interest

The authors declare that they have no conflict of interest

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