CASE REPORT

A CASE REPORT OF ANEURYSMAL BONE CYST IN THE ANTERIOR MANDIBLE OF A 13-YEAR-OLD: A RARE INCIDENTAL FINDING WITH CHARACTERISTIC HISTOLOGY AND NO CLINICAL MANIFESTATION

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The Aneurysmal bone cysts (ABCs) are neoplasms of a non-malignant nature with blood-filled cystic spaces and an array of skeletal involvement, though they are rarely found in the jaws. ABC presented atypically with jaw involvement, requiring a comprehensive assessment to distinguish it from other jaw lesions, including benign and malignant tumours. We report a 13-year-old female patient who presented with no visible swelling, but an orthopantomogram (OPG) later revealed a large unilocular radiolucency in the anterior of the lower jaw. The radiolucency depicted well-defined margins and a marked cortical expansion, resulting in a 'ballooned' or 'blown out' appearance. Curettage with biopsy confirmed the diagnosis by histological investigation. Histopathological data supported the diagnosis of anterior mandibular ABC in this instance. This case demonstrates the many clinical manifestations of ABC and underlines the importance of multidisciplinarity, comprising radiologists, pathologists, and clinicians, all for the correct diagnosis and appropriate care. Further research is needed to understand the pathophysiology and best treatment techniques for ABC, particularly in less common places such as the jaw.

Keywords: Aneurysmal Bone Cyst; Histopathology; Jaw Neoplasms; Mandibular Diseases

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INTRODUCTION

ABCs are benign neoplasms with blood-filled cystic cavities and varying degrees of skeletal involvement.[1] The term 'bone cysts' refers to a fluid-filled hollow cavity, whereas 'aneurysm' refers to the bone's puffed-out enlarged contour.[2] ABCs was originally recognized by Jaffe and Leisntein in 1942. ABC is mostly present in long bones and vertebrae, with only 1.8% occurrence in jaws, making them a rare sight.[3,4] There is a 3:1 occurrence ratio for the ABCs within the head and neck region, comparing the mandible to the maxilla, with ascendancy for the body of the mandible, ramus and mandibular angle.[4] The ABC mostly presents in children and adolescents during the first 2 decades of life with equal distribution among males and females. [1] No specific cause has been identified for the occurrence of ABC but it was historically thought to arise from either previous traumatic or vascular events or hemodynamic perturbations.[5] The previous literature refers to USP-6 gene translocation as the aetiology for ABCs that is present in the long tubular bones. Still, the details in connection with the ABCs of the jaw are indistinct.[6] ABCS can either occur solitarily or in conjunction with other neoplasms of the bone, namely giant cell tumour, chondroblastoma, ossifying fibroma, solitary bona cyst, fibromyxoma, giant cell reparative granuloma and osteoblastoma.[7,8] Mostly, ABCs are symptomless, but the manifestation of swelling and localized pain has also been reported.[9] Radiologically ABC presents as a lytic, expansile lesion giving a blown-out appearance on the radiograph.[10] Histologically, ABC can present as a cystic space filled with mostly blood, lacking an epithelial lining. It is characterized by diffuse infiltration by fibroblasts, giant cells and woven bone. At the molecular level, given the recurrent relocation of the gene USP6 (chr.17p13.2 locus), ABC has been designated as a neoplasm rather than a reactive lesion.[1]

This case report emphasises the importance of radiographic and histopathological examination in detecting asymptomatic jaw lesions, contributing to the existing literature on ABCs in Pakistan and focusing on the need for further research and awareness about this rare condition in the local population.

CASE REPORT

A 13-year-old female presented to our outpatient department for an orthodontic consultation regarding her retained upper left deciduous tooth. Extra orally no visible swelling was seen. After a clinical examination and history, she was advised to get an OPG for further evaluation. The patient was a healthy child with no significant chronic medical or surgical issues. Orthopantomography reveals a large unilocular radiolucency at the left lower border of the jaw, specifically the anterior mandible. The radiolucency was present around the roots of the lower teeth and extending from #35 to #42, crossing the midline. The radiographic margins were well defined on account of notable cortical expansion with the afflicted bone 'thinned out', reflecting the 'ballooned' or 'blown out' appearance of the bony contours. These radiographic findings were highly corresponding to a benign cystic lesion, likely an Aneurysmal bone cyst, Stafne cyst or a giant cell neoplasm or granuloma, furthermore, a plan for curettage and incisional biopsy of the lesion was devised. Grossly, multiple spongy haemorrhagic soft tissue fragments were received collectively measuring 2x1.5x0.5 cm. These multiple bits of the specimen were processed, sectioned, and stained for histopathology reporting. The final histological examination of the tissue showed a cystic lesion showing multiple spaces filled with haemorrhage, separated by cellular septae and flat attenuated lining, having characteristic hallmark features of ABC. The stroma showed infiltration with fibroblasts, giant cells, and woven bone. The tumour bulk showed giant cells which were scattered in the background of the fibrovascular stroma. No evidence of atypical cytological features was seen. On clinicopathological correlation, the lesion was histopathologically reported as ABC. Since the lesion revealed blood-filled cystic spaces, all other possibilities such as giant cell granuloma and Stafne bone cyst were ruled out. The clinical, radiological and histopathological findings are typical of an Aneurysmal bone cyst.

Outcome: Post complete removal of the lesion, the patient was kept on ANSAID with antibiotic cover for some time. No recurrence has been reported as yet.

Ethical Approval:

The case report was prepared retrospectively, focusing on an incidental finding that was managed according to standard clinical practice. As such, it did not involve any new interventions or experimental treatments that would necessitate prospective ethical approval. The patient's data was anonymized to maintain confidentiality, and informed consent for publication was obtained.

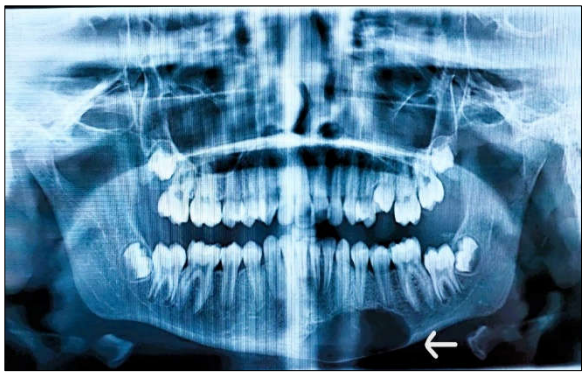

Figure-1: Radiographic image of a 13-year-old patient showing a large unilocular radiolucency at the left lower border of the jaw. The image presents a well-defined margin with marked cortical expansion, characteristic of an aneurysmal bone cyst (ABC) in the anterior mandible.

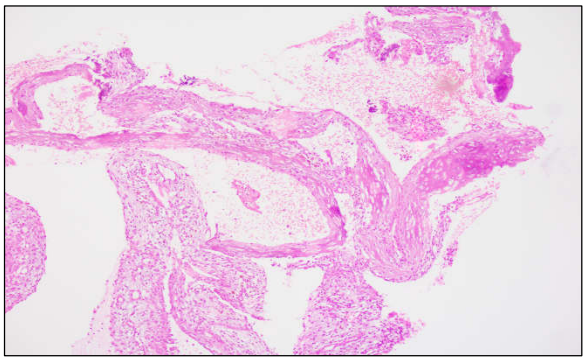

Figure-2 (a): Histopathological low-power image (x4) showing blood-filled cystic spaces separated by fibrous septa. The stroma shows infiltration with fibroblasts, giant cells, and woven bone, confirming the diagnosis of aneurysmal bone cyst (H&E staining)

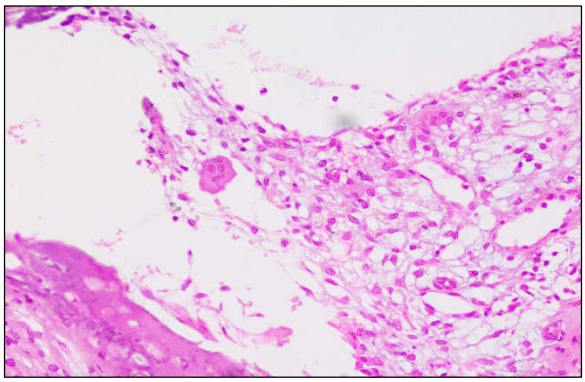

Figure-2 (b) Histopathological high-power image (x100) showing multinucleated giant cells scattered within the fibrovascular stroma (H&E staining)

DISCUSSION

Aneurysmal bone cysts (ABCs) are categorized as non-neoplastic, benign bone lesions, which are fast-developing, destructive, benign bone tumours initially recognized by Jaffe and Lichtenstein in 1942.[11]. ABCs are relatively rare osteolytic diseases of unclear cause. They are distinguished by pseudocystic cavities teeming with RBCs, which are divided with sepate of fibrous connective tissue. The majority of ABCs occur in the tibia as well as the femur, with only twelve percent affecting the craniofacial region. The ABCs are seldom seen within the gnathic region.[12] The mandible is more often compromised than the upper jaw, on account of the most commonly impacted areas being the molar region and the ramus (back part) of the mandible.[11,13] The clinical history of jaw ABCs varies from slowly growing lesions that heal on their own and are detected on routine radiographs to progressive pain that can lead to bone fractures.[12]

Although ABCs can affect people of all ages, but is mostly a condition of young adults. Approximately 50 to 70 percent of aneurysmal bone cysts develop during adolescence, with 70–86 percent affecting individuals younger than 20 years.[14] ABC lesions have sharp, well-defined borders, no periosteal response, and go through different stages of development. Small lesions may first exhibit a permeative pattern, followed by fast, destructive development that mimics malignancy. Finally, the lesions induce substantial bone growth and thinning, resulting in a "blown-out" look with persistent osteolysis and bone enlargement.[15] ABC has two major variants: primary and secondary. Primary ABC can be congenital, resulting in arteriovenous malformations and early tooth growth, or acquired, which is frequently associated with trauma. Secondary ABC is often caused by the breakdown of the existing conditions, including cysts, tumors, or fibrous and osseous diseases.[16] The theory of ABCs suggesting them as a reactive process has been challenged by research evidence pointing to cytogenetic abnormality, specifically the t(16;17) (q22; p13) chromosomal translocation.[17]

Conventional radiographs of ABCs reveal expansive, well-defined bone lesions exhibiting a radiographic pattern resembling soap bubbles and fluid-like opacity. However, the X-rays often fail to provide a definite diagnosis. The MRI paired with biopsies remains the gold standard for diagnosing ABCs. Treatment recommendations for ABCs should be based on the lesion's location, stability, and safety.[10,18] Careful curettage is regarded as an appropriate therapy for Jaw cysts. Studies show a recurrence rate of 13.3% with little difference between surgical techniques (curettage, 15.2%; resection, 11.8%).[19] Accurate recognition of the primary ABC is critical for successful therapy. Management options include curettage, bone grafting or replacement, and adjuvants like phenol, liquid nitrogen, and argon laser. Recent methods also explore sclerotherapy and monoclonal antibodies such as denosumab, though it must be used cautiously in children due to risk of hypercalcemia.[20]

CONCLUSION

Aneurysmal bone cyst (ABC) is an idiopathic condition primarily involving bones, with rare occurrences in the anterior mandible. It may mimic aggressive lesions radiographically due to its appearance and growth. Hence, ABC should be considered in the differential diagnoses of jaw radiolucencies. Image-guided core biopsy is advised for accurate diagnosis before procedures like curettage. A multidisciplinary approach is essential to ensure accurate diagnosis and treatment. More research is needed to understand its pathophysiology and therapeutic strategies, especially in uncommon presentations.

Statement of Informed Consent:

Informed consent for the publication of this case report was obtained from the patient and/or their legal guardian. No identifying information has been included in the manuscript to maintain patient confidentiality.

Disclaimer: The abstract has not been previously presented or published in any conference, and this manuscript is not part of any research, PhD, or thesis project. No other relevant information applies.

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