Case Report

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Aneurysmal Bone Cyst Of Maxilla With Palatal Perforation: A Rare Case Report.

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ABSTRACT

Aneurysmal bone cysts (ABCs) are rare, blood-filled bone lesions that grow as solid lumps within the jawbone. These cysts account for about 1.5% of nonodontogenic jaw tumors and mostly occur in long bones. The mandible is commonly affected, while ABCs in the upper maxilla are rare, comprising only 2% of cases. This report details an uncommon instance of ABC in a 25-year-old patient who presented with a painless swelling in the right upper back tooth and palatal area for two months. First described by Jaffe and Lichtenstein in 1942, ABCs in the jaws are typically seen in younger patients. In this case, treatment involved curettage of the cyst and reconstruction with a palatal rotational flap to cover the defect.

Keywords : ABC, Palatal perforation, giant cells.

INTRODUCTION

Aneurysmal bone cysts are infrequent osteolytic growths within the jawbone, manifesting as solid, hard lumps that continue to expand. They may cause discomfort or show no symptoms at all. It's common for the bone's outer layer (cortical bone) to expand and perforate. Teeth in the affected area may shift, yet they generally remain vital unless a secondary infection occurs. Initially, these cysts were referred to as "periosteal giant cell tumors" by Potts [2]. Based on Potts' case series involving 29 patients, the mandibular body and angle region are the most commonly affected sites, with the maxilla being less frequently involved. Additionally, the condition is more prevalent among females than males.
Radiographically, an aneurysmal bone cyst appears as a clearly defined, multi-chambered radiolucent area. Usually, fragile bone structures and partitions intricately mesh with cystic radiolucencies, producing a pattern resembling a honeycomb or soap bubble [3]. Shafer et al. (1974) noted that cortical bone destruction often accompanies periosteal reaction [4]. However, the radiographic appearance in jaw lesions doesn't offer a definitive diagnosis [1].
In this case report, we detail a case of an aneurysmal bone cyst located in the posterior maxilla, which responded positively to conservative therapy. We performed a procedure involving the curettage of the affected bone and then used a palatal rotational flap to cover the resultant defect. IRB exemption is received as No identity of pt. has been revealed and consent was taken prior to photography.

CASE PRESENTATION

A 25-year-old individual presented to our department in August 2022, complaining of painless swelling in the right upper back teeth region persisting for 2 months. Initially, a mild and painless swelling was noticed in the right upper back teeth region, which gradually increased in size. Despite the lack of serious symptoms, the patient did not seek medical attention for 2 months. The patient did not report loosening teeth, bleeding, pus discharge, trauma, dental intervention, high-risk behaviours, radiation exposure, systemic disease, family history, or any ENT-related issues.
Upon clinical examination, facial asymmetry was noted, characterized by an obliterated right nasolabial fold and a bulge on the right lateral wall of the nose. A diffuse swelling on the right side of the face was observed, measuring 3x4 cm, extending superoinferiorly between the line connecting the tragus and lateral canthi and the line from tragus to the corner of the mouth. Anteroposteriorly, it obliterates the right nasal fold and extends 3 cm in front of the tragus. The skin surface appeared normal, with a regular left nasolabial fold. The lips were competent, and normal jaw movements were observed, without clicking or crepitus during mouth opening or closure, with a mouth opening of 35 mm. No scar marks, inflammation, sinuses, or fistulas were observed over the swelling. Tenderness was present over the swelling, with no paresthesia, pulsation, and all inspection findings confirmed on palpation.

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On intraoral examination, the gingivopalatal and buccal sulcus on the right upper side were obliterated from the 13-16 region. The gingiva appeared pale and inflamed. The tongue, floor of the mouth, and retromolar trigone were normal. Tooth 15 exhibited mobility but was vital. Occlusion was intact. On palpation, all inspection findings were confirmed, and a hard, fixed mass was detected in the right upper gingivobuccal and palatal sulcus.

Computerized tomography (CT) revealed the presence of an osteolytic bony lesion in the right maxillary upper teeth region, with bulging and thinning of the cortex. Orthopantomography (OPG) demonstrated a multiloculated radiolucency with diffuse borders. Multiple lagoons with a vascular appearance were visible within the lesion, leading to a provisional diagnosis of aneurysmal bone cyst (ABC).

The lesion was surgically removed under general anaesthesia through an intraoral approach, involving a vestibular incision made with a Colorado needle, followed by curettage. During the curettage, the palate was accidentally perforated, necessitating closure with a palatal rotation flap.

A biopsy confirmed the presence of a multiloculated cystic lesion with blood-filled cystic gaps containing giant cells and fibroblasts, along with calcified basophilic material (resembling blue reticulated chondroid-like substance). While necrosis was rare, mitotic activity was detected, without evidence of cytologic atypia, suggestive of aneurysmal bone cyst.

After 20 days, the patient reported fluids leaking through the nose, a hole in the palate, and paresthesia on the right side of the face. An alginate impression was taken, and a palatal acrylic plate was fabricated, leaving the defect for self-healing. However, the patient found it difficult to wear the plate, prompting a second attempt at palatal fistula closure under local anaesthesia after a month. Sutures were removed after 10 days, and the recovery was uneventful.

An 8-month follow-up showed complete healing of the defect, improved radiographic results, reduced paraesthesia to a significant extent, and restored facial contour. Radiographs at the 8-month mark revealed bone formation in the area previously affected by the lesion.

DISCUSSION

An uncommon pathological condition affecting the jaws, termed an aneurysmal bone cyst, was first identified by Jaffe and Lichtenstein in 1942 [5]. Initially known as "periosteal giant cell tumor," it was described by Potts [2]. Potts' case series of 28 patients indicated that the maxilla is rarely affected, while the mandibular body and angle area are the most frequently affected sites. Additionally, there is a lower male predilection compared to females.

Various hypotheses have been proposed, but the exact pathophysiology of aneurysmal bone cysts remains unresolved. Although a history of trauma is often present, it's uncertain whether trauma contributes to the formation of these cysts. Lichtenstein's [6] theory in 1950 suggests that bone resorption is caused by persistent local hemodynamic alterations and elevated venous pressure, resulting in an engorged and dilated vascular bed. The cyst consists of giant cells that cause bone resorption, later replaced by new bone. Potential causes of hemodynamic alterations include arteriovenous aneurysms or venous thrombosis. However, Bernier and Bhaskar [7] in 1958 questioned the vascular origin and proposed a relation between giant cell granuloma and aneurysmal bone cyst, indicating an unorganized canalizing hematoma. They proposed that when injured blood vessels remain connected to hematoma it will result in aneurysmal bone cyst in absence of connection it will result in giant cell granuloma.

Biesecker et al. [8] in 1970 documented 66 instances of aneurysmal bone cysts and noted that 32% of them are associated with pathological bone lesions. According to their hypothesis, an arteriovenous fistula is caused by the initial pathological lesion, leading to subsequent reactive bone alterations due to hemodynamic pressures. They believed that this subsequent lesion was the aneurysmal bone cyst.

Histologically, this tissue consists of a lax network of fibrous connective tissue and young fibroblast cells. Additionally, there are abundant enlarged capillaries and blood-filled cavities present. Spindle-shaped cells sometimes line these spaces containing uncoagulated blood, suggesting circulation through vascular channels [9]. Connective tissue stroma contains giant cells, resembling giant cell granuloma. Additionally, areas of hemoglobin breakdown (haemosiderin) frequently coincide with regions of bleeding within the lesion. Shear in 1976 supported the idea that aneurysmal bone cysts may arise secondary to another pathological disorder such as fibro-osseous lesions [10].

No indications have been found to support the idea that aneurysmal bone cysts represent a type of cancerous growth[11]. Nevertheless, there have been cases of a malignant variation that has spread to the lungs [12], which could be indicative of a specific form of osteosarcoma. This underscores the significance of thorough cytological analysis [13].

A severe medical condition such as sarcoma could be identified in a youthful individual who exhibits a swiftly advancing abnormal growth in the jawbone or other parts of the face's skeletal structure. In the case of an aneurysmal bone cyst lesion, the affected teeth remain vital, responding to the carbon dioxide ice test, unless a secondary infection occurs. An aspirational biopsy of this lesion unveiled fluid with an amber hue. The total soluble protein content measured at 64 g/l, indicating that the fluid within the cyst originates from the blood in vascular channels [1].

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An elevated alkaline phosphatase level, although atypical for an aneurysmal bone cyst, may be associated with the patient's pregnancy [13,14]. Surgical enucleation and curettage or conservative excision are the most commonly employed treatment approaches for sizable lesions. Low-dose radiation has been employed [15]. Nonetheless, the authors express reservations about its use for oral lesions due to the considerable potential side effects associated with this treatment modality. Oral and facial lesions documented thus far have shown a relatively low recurrence rate. However, aneurysmal bone cysts outside the maxillofacial region tend to recur after incomplete treatment. Some researchers reported recurrence [11].

RESULT

The conservative approach to surgical treatment proved to be highly effective in achieving a favourable outcome for this patient. While the entire lesion was not formally removed, the bone has fully regenerated. According to Jaffe (1964), even when only partial curettage of the cystic area is performed, healing is likely to occur [9]. It appears that aneurysmal bone cysts in the jaw region can be successfully managed through conservative methods, with resection being reserved as a potential option only for extremely extensive cases.

Figure 1a : (FRONTAL PROFILE) an obliterated right nasolabial fold and a bulge on the right lateral wall of the nose.



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Figure 1b : (PALATAL VIEW) Gingivopalatal sulcus on the right side is obliterated from the 13 -16 region.



Figure 1c: (BUCCAL VIEW) buccal sulcus on the right side is obliterated from the 13 -16 region.



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Figure 1d: (3D CT FACE) Presence of an osteolytic bony lesion in the right maxillary upper teeth region.

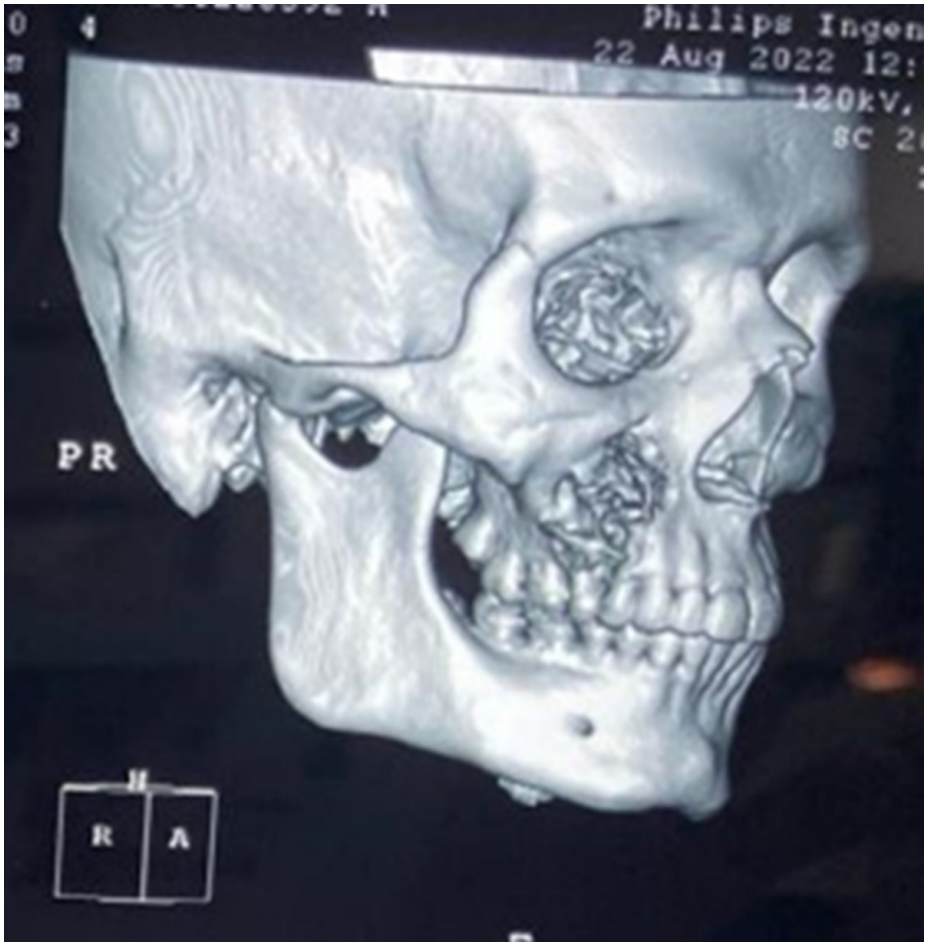
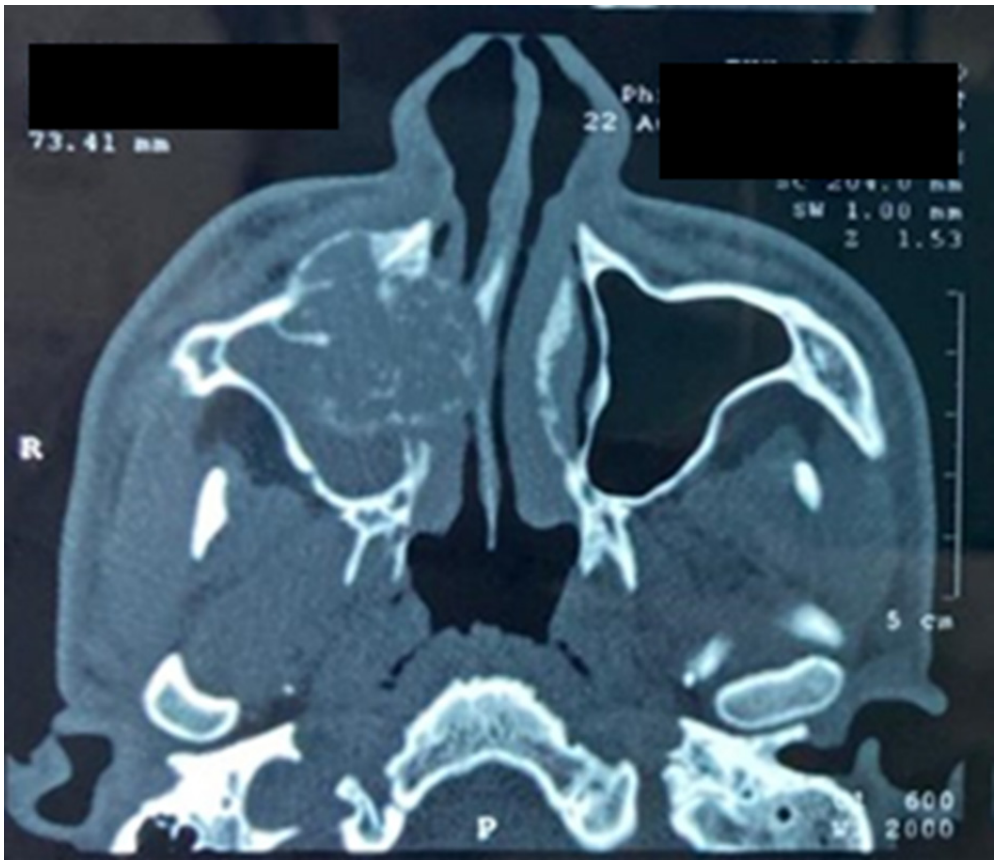


Figure 1e: (AXIAL SECTION OF CT) SHOWING bulging and thinning of the cortex on right side along with obliteration of sinus.



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Figure 2a: (PALATAL VIEW) surgical site after curettage and perforation of palate



Figure 2b: (BUCCAL VIEW) surgical site after curettage and perforation of palate.



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Figure 3a: (PALATAL VIEW): - a palatal fistula.



Figure 3b : (PALATAL VIEW): palatal fistula closure under local anaesthesia by palatal rotational flap (I MONTH FOLLOW UP)



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Fgure 3c : 8-month follow-up showed complete healing of the defect.



Figure 3d : 8-month follow-up showed improved radiographic results.



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CONCLUSION

Treatment frequently includes surgical removal through enucleation and curettage, or a more conservative excision approach for substantial lesions. In this case surgical enucleation and curettage or conservative excision was done for this huge lesion. Aneurysmal bone cysts in the oral and facial regions have a lower recurrence rate compared to those in other areas, especially when effectively treated with conservative measures alone. A close follow up of the same up to 1 year didn't observe any recurrence.

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