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Case Report

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Prefabricated Implant Treatment for an Aneurysmal Bone Cyst of the Mandible: A Case Report and Review of the Literature

Mohammad Faisal[1], Virender Gombra[2], Deepika B Popli[3], Mandeep Kaur[4]

[1] Professor, Oral and Maxillofacial surgery, Faculty of Dentistry, Jamia millia Islamia-110025, New Delhi, India
[2] Professor, Oral Medicine and Radiology, Faculty of Dentistry, Jamia Millia Islamia-110025, New Delhi, India
[3] Professor, Oral Pathology and microbiology, Faculty of Dentistry, Jamia Millia Islamia-110025, New Delhi, India
[4] Professor, Oral Medicine and Radiology, Faculty of Dentistry, Jamia Millia Islamia-110025, New Delhi, India

Abstract

Aneurysmal bone cyst (ABC) is a benign, destructive and expanding bone lesion. Most commonly it is found in long bones and around 2-12% of cases are seen in head and neck region. ABC is an expansile osteolytic lesion, which consists of blood filled spaces separated by connective tissue septa and multinucleated giant cells. Aspiration, radiographic examination, and cross-sectional scans assist in ruling out other odontogenic or non-odontogenic pathologies and also help in locating the lesion. This paper documents case report of Aneurysmal bone cyst in a 28-year-old male patient. This article presents clinical diagnosis, investigations, and surgical management of the lesion, along with a review of literature. There are various treatment options for the management of aneurysmal bone cyst. This case management consisted of osteotomy and mandibular reinforcement with prefabricated implants using stereolithography models followed by histopathological examination, which further confirmed the diagnosis. This lesion has rare occurrence and should be considered in differential diagnosis for osteolytic and expansile swelling in mandibular region.

Keywords: Mandible, Non-odontogenic cyst, Mandibular tumours, Benign bone lesions, Mandibular pseudocysts.

INTRODUCTION

Aneurysmal bone cysts (ABC) are non-neoplastic , destructive , expansive , erosive benign bone lesion [1]. First described by Van Arsdale in 1893 as ossifying hamartoma, but renamed later by Jaffe and Liechtenstein, the aneurysmal bone cyst. In 1958, Bernier and Bhaskar reported the first case occurring in the jaws [2]. World Health Organization (WHO) in 1992 classified ABC in odontogenic and nonodontogenic cysts, this lesion was reclassified in 2017 under 'Giant cell lesions and bone cysts' [3]. WHO classification of tumors of bone (2020) classifies ABC as a benign osteoclastic giant cell rich neoplasm and considers it as pseudo cyst due to lack of epithelial lining [1]. WHO defined aneurysmal bone cyst as an expansile osteolytic lesion, which consists of blood filled spaces separated by connective tissue septa that contain osteoid material and multinucleated giant cells [4]. These lesions account for about 9.1% of all primary bone tumors. They commonly appear during second decade of life, with an average age of onset at 13 years, and 90% cases are diagnosed before 30 years [5]. Mostly it is found in the metaphysis of the long bones, mainly in "tibia and femur" and 2-12% of the lesions in the head and neck region. Among head and neck region 90% of the cases affect the posterior mandible, body of the mandible (40%), ramus (30%), angle (19%), symphisis (9%) and condyles (2%). There is a slight greater incidence in women (62%) [6]. Other facial regions include infratemporal region, zygoma and orbital floor [7]. Clinically, ABC may present with pain, swelling, or an associated pathological fracture of the bone. However, based on location of ABC tumor neurological deficits may appear due to spinal cord involvement and other symptoms include reduced hearing, headaches and visual disturbances [5]. Although, the etiopathogenesisis uncertain, but it may occur de- novo as a primary lesion, or as a secondary phenomenon associated to primary bone lesions, such as the ossifying fibroma, solitary bone cyst, osteoblastoma, fibrous dysplasia or central giant cell granuloma [5,8,9]. Various suggested theories are like congenital, post traumatic, reactive malformation, genetic predisposition, dilatation of local vascular network due to increased venous pressure caused by local circulatory abnormalities. Chromosomal translocation (16; 17) (q22; p13) is a cytogenic abnormality has been suggested for etiology of ABC [5,6,9]. The differential diagnoses for ABCs include but are not limited to ameloblastoma, the central giant cell granuloma, the myxoma, the traumatic bone cyst, the keratocystico Odontogenic tumor, chondroblastomas, fibrous dysplasia, giant cell tumors, telangiectatic osteosarcomas, and unicameral bone cysts.

***Corresponding author:**
Dr. Virender Gombra
Professor, Oral Medicine and Radiology, Faculty of Dentistry, Jamia Millia Islamia- 110025, New Delhi, India
Email: vgombra@jmi.ac.in

Imaging can help in identifying and also characterizing the extent of the disease [5,8]. Occasionally, its presentation can be a rapid expansive growth locally destructive that may be misdiagnosed as a high-aggressive or a malignant neoplasm. This rapid growth may result from erosion of the cortical plates of an asymptomatic slow growth lesion [9]. Management of these lesions is most commonly done by surgery [10]. Review of previous case reports showed that there is not much literature on ABC management using prefabricated implant and this case report presents aneurysmal cyst of mandible which was increasing in size and stereolithography models were made to plan surgery and prefabricated implant was placed for maintaining contour.

CASE REPORT

This case report has been written according to CARE case report guidelines. A 28 years old male reported with complaint of swelling on left side of face since one year with history of mild pain. Patient gave history of left side posterior molar extraction for same complaint and observed no relief. Medical and systemic history was negative. On examination a solitary hard swelling was observed on left side posterior mandibular angle region. Intraorally 36, 37 and 38 were missing and diffuse hard, tender swelling was present in left posterior alveolus region (Figure 1). Provisional diagnosis of benign odontogenic tumor was given. Panoramic view showed multilocular lesion on left side of mandible. Cone beam computed tomography (CBCT) evaluation showed an expansile, multilocular lesion involving left side mandibular body, angle, ramus region and alveolar crest to inferior border of the mandible. Mandibular canal cortical lining was indistinct within the lesion and deficient cortical bone in buccal, lingual and retromolar region. Approximate greatest dimension was 28.25 mm buccolingually, 47.66 mm anteroposteriorly x 47.12mm superoinferiorly.

Differential diagnosis considered for this lesion were Ameloblastoma, Odontogenic keratocyst, Cental giant cell granuloma and Aneurysmal bone cyst. (Figure-2) Complete blood count investigation was within normal range. Needle aspiration showed a blood filled lesion. Incisional biopsy also confirmed a blood filled lesion and histopathology showed blood-filled large cystic spaces lacking endothelial lining which were separated by fibrous septa containing spindle-shaped stromal cells, numerous osteoclast-type multinucleated giant cells, and areas of reactive woven bone formation. The fibrous septa also exhibited occasional hemosiderin-laden macrophages. No significant cytological atypia or abnormal mitotic activity was seen and it was suggestive of Aneurysmal bone cyst (Figure 3). CT scan of the lesion was done to localise soft tissue extent of the lesion.

It showed expansile lytic lesion with no significant extraosseous soft tissue extent. Treatment planning involved 3D planning for cutting guide and implant placement followed by surgery (Figure 4). Nasal intubation was done. Patient was scrubbed and draped under aspetic conditions and 5cm long incision was given in the left skin crease in submandibularregion ,extending superiorly till the mandibular angle region. Subplatysmal dissection was performed. The facial nerve (marginal mandibular) was identified and preserved and the facial aretry was identified, clamped and divided. The dissection was done till the inferior border of the mandible and subperiosteal dissection was done to expose the lesion. The pterygomassetric shin was divided . The lesion was exposed and osteotomy was done from 35-36 region till the ramus region with burs and chisel mallet. After currettage, the prefabricated implant was adapted, incision was given intraorally in the labial vestibule extending from 43 to 33 (Figure 5A). After achieving proper occlusion the implant was fixed with 2x8mm screws in the anterior region and 2x6 mm screws in the condylar region.There was insignificant blood loss and wound was closed in layers intraorally in addition to extraoral with 3-0 vicryl and 3-0 proline sutures.

The pterygomassteric sling was sutured to the implant in angle region. Throat pack was removed and pressure dressing was given. Patient was extubated and shifted to recovery room. Postoperative histopathology confirmed diagnosis of Aneurysmal bone cyst. The patient was evaluated clinically and radiographically for one year, no recurrence of the lesion was identified during that time; aesthetic and functional results were satisfactory. One year follow up showed no recurrence of the lesion.

Figure 1: A,B Extra-orally swelling on left side of mandibular angle and 1 C shows intraorally obliteration of mucobuccal fold in left side posterior mandibular alveolus region

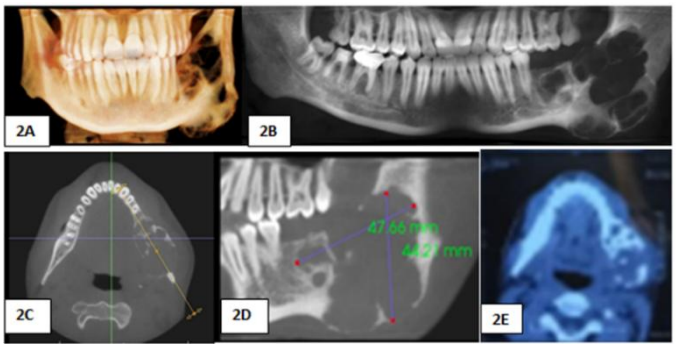

Figure 2: A shows 3 dimensional CBCT of the lesion, 2B shows panoramic view showing multilocular lesion, 2C shows cross sectional axial CBCT of the lesion, 2D shows Sagittal cross sectional image, 2E shows axial image of the CT scan

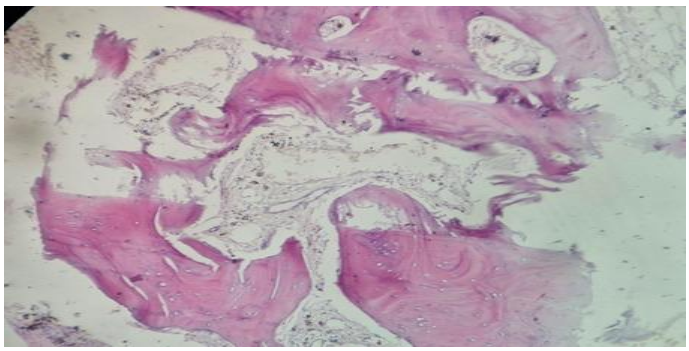

Figure 3: Shows histopathology of the lesion showing blood filled cystic spaces, giant cells and fibrous septa

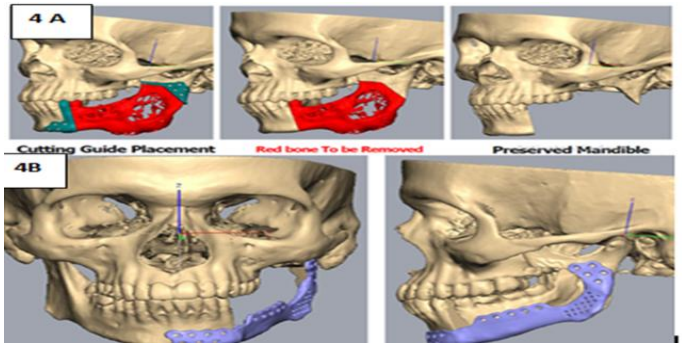

Figure 4: A shows cutting guide placement and 4B shows implant planning

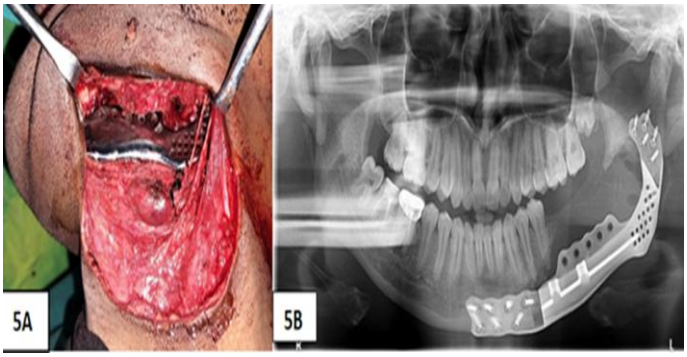

Figure 5: A surgical placement of implant and 5B shows Implant planning

DISCUSSION

Clinically, ABC most commonly appear as painful lesion but it may present as asymptomatic lesion [11]. Some other symptoms seen may include loose or displaced teeth, malocclusion, haemorrhage, pus discharge, nasal obstruction, lip paraesthesia and bone fracture. Aspiration of dark red or brownish haemorrhagic fluid is suggestive of ABC [8,12]. Recommended Pathogenesis for ABC is localised vascular proliferation in response to unknown stimulus or occurrence of an arterio-venous shunt in between the stroma and the medullary vessels [7]. Most of the ABC are primary and result from chromosomal rearrangement, and less commonly they are secondary, due to haemorrhage induced by aberrant venous circulation, resulting in local remodelling by osteoclasts [5]. Pulpal electrical Vitality tests may show vital teeth in ABC and rarely non vital teeth [5,13]. ABC mostly appears as rapidly growing swelling; radiographically appear as radiolucent lesion with variable presentation. Usually shows cortical expansion, erosion and malignant tumor like appearance. Clinical classification includes following varieties (1) Conventional or vascular; (2) Solid form is rare and usually observed during radiographic examination; and (3) Mixed form which shows both conventional and solid types [8]. Clinically most of the lesions present as facial symmetry and a distinct swelling due to expansile bony lesion. Usually lesions are slow growing till cortical bone is intact and shows a rapid growth after perforation of cortical bone. Facial deformity can lead to malocclusion. Pain can be occasionally present in these lesions. Sometimes these lesions may show root resorptions of teeth, dysaesthesia, and paresthesia. Maxilla involvement of these lesions may present clinically as epistaxis, nasal obstruction, proptosis, and diplopia. Solid type of ABC is usually asymptomatic whereas vascular type mostly invades adjacent soft tissues with rapid growth [9,13]. Extent of lesion can be evaluated in panoramic radiography, CT scan, CBCT and MRI and CT scan aid in soft tissue assessment of the lesion. Radiographic features may vary from radiolucent, mixed radio-opaque and radiolucent, moth eaten, unicystic or multilocular with ballooning expansion. Usually, it appears as radiolucent, multilocular, cystic with coarse septae, destruction of bony cortex, and may show periosteal reaction of sun ray appearance. Radiographically it may be present with impacted teeth [5,9 7]. Radiologically it often shows blowout type of expansion and may have honeycomb, soap bubble, or moth-eaten appearance due to osteolytic lesion with thin peripheral bone [8,12,13]. Magnetic resonance imaging shows high intensity signal in the lesion and low intensity signal at the margin of the lesion. Fluid level assessment in MRI is highly suggestive of ABC and angiography can be performed to rule out hemangioma or highly vascular neoplasm. Diagnosis is verified by fine needle aspiration and incisional biopsy followed by histopathological evaluation [9,7,13]. A manometer test for ABC shows a steady increase in the column of blood oscillating with pulse when a needle is introduced into the lesion [13].

Enneking's staging of benign musculoskeletal neoplasms describes aneurysmal bone cysts stages as Latent type (stage 1) which is usually asymptomatic and incidental finding , Active type (stage 2) which has steady growth, symptomatic and may be palpable and Aggressive type (stage 3) present as visible abnormality with significant patient discomfort [14].

Biopsy followed by histopathological examination is performed to confirm the diagnosis [11]. Histologically, vascular variant of ABC shows blood filled osteolytic lesions and, separated by fibrous connective tissue septa with osteoid trabeculae. Variable level of hemosiderin and giant cells can be present. Solid variant is noncystic with foci of hemorrhage, fibroblastic, fibrohistiocytic elements with osteoclast like giant cells, osteoid and calcifying fibromyxoid tissue [9].

Management of ABC usually involves open surgical curettage and postoperatively it may cause pain, wound dehiscence, swelling, and sepsis [13]. Curettage involves removing the contents of the cavity and remaining bone space can be maintained with bone graft or cement. This process is better than en bloc excision in reducing patient morbidity and preventing greater amount of intact bone. En bloc excision involves removal of entire lesion from the bone. Due to significant morbidity, en bloc resection is mostly indicated for recurrent lesions or lesions extending to overlying tissues for which less invasive means were insufficient for management. Recurrence may occur in 20-30% of cases and it is mostly associated with incomplete removal of the lesion and is higher in postoperative first year duration [9,10,12]. Other treatments include sclerotherapy, cryotherapy or primary therapeutic embolization. Management Other than surgical treatment can be considered in very bulky lesions and adult patients [7,11]. Low recurrence is seen after cryotherapy; however it can cause nerve tissue damage [13]. Radiotherapy is not indicated for ABC as it may show failure rate and possibility of radiation induced carcinoma. Surgical curettage may cause bleeding which is considered as common complication for this vascular lesion and arterial embolization can be done preoperatively to prevent it[7]. However embolization frequently cause growth of collaterals from adjacent vessels and it may not be difficult to embolise distorted vessels. Embolization and ligation rarely may cause the loss of vision in one eye [13]. Some authors advise reconstru ction of the mandibular discontinuity with autogenous grafts for restoring aesthetics and for lesions with fracture risk [9]. Few cases showed healing even after open Biopsy. Thus wait and watch can be one option for small lesions. Some studies have shown successful management with calcitonin spray which inhibits osteoclastic action and enhances trabecular bone formation. Osteoinductive management has been recommended for ossification of the ABC. In this case stereolithographic model have been used for contouring of the mandible with satisfactory aesthetics observed by patient. Stereolithographic models assists in good contouring of the reconstruction plate with contralateral mandible contour for facial symmetry [13].

CONCLUSION

To our knowledge there is not much literature regarding the use of prefabricated implant placement post Aneurysmal bone cyst resection. This case report presented a case of ABC affecting mandible treated by surgical excision followed by stereolithographic implant placement with no evidence of recurrence. Thus for extensive aggressive lesions, resection with prefabricated implant can be an effective method to improve surgical results, as it aides in aesthetic and functional requirement after surgery. 3D-printed models provide effective simulated surgery conditions and aid in pre surgical designing. Radiographic features of these lesions can be characteristic but biopsy is required to confirm the diagnosis prior to the treatment plan.

Conflicts of Interest

The author reports no conflicts of interest.

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