

ANEURYSMAL BONE CYST OF MANDIBLE - A CASE REPORT

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ABSTRACT

Aneurysmal bone cysts are rare benign lesions of bone tissue, infrequent in craniofacial skeleton with regard to other structures like long bones or the spine. They are composed of sinusoidal and vascular spaces blood-filled and surrounded by fibrous tissue septa. We present a case of a 42-year-old female with a big swelling in the left mandible associated to pain and rapid growth. On the X-ray study, an expansive multilocular and high vascularized bony lesion within the mandibular parasymphysis region was observed. It produced expansion and destruction of buccal cortex. An incisional biopsy was performed showing a fibrous tissue with blood-filled spaces lesion suggestive of an aneurysmal bone cyst. Surgical enucleation was done with curettage. Aneurysmal bone cysts are non-neoplastic but locally aggressive tumours with occasional rapid growth that may be differentiated from other multilocular process like ameloblastoma, ossifying fibroma, epithelial cyst, giant cell granuloma and sarcomas. Treatment of choice consists on conservative surgical excision of the mass with curettage or enucleation. When resection creates a big defect, primary surgical reconstruction is recommended.

KEYWORDS: Aneurysmal Bone Cyst (ABC), Mandibular Tumours, Benign Bone Lesion

INTRODUCTION

Aneurysmal bone cyst is a benign cystic lesion of bone composed of blood filled spaces separated by connective tissue septa containing fibroblasts, osteoclast type giant cells and reactive woven bone 50% in long bones, 20% in the vertebral column. Mandible is a usual site of involvement in the craniofacial skeleton. Accounts for 1.5% of the non odontogenic, non epithelial cysts of the mandible, aneurysmal bone cysts consist of non-neoplastic benign bony lesions with multilocular appearance. They are considered as pseudocyst because of the lack of an epithelial lining.^[1] They are principally located in long bone metaphysis like the femur and the tibia (more than 50% of aneurysmal bone cysts) and spine (12-30%).^[2] The presence of these tumours in facial bones is infrequent, with a 2-12% of all the aneurysmal

bone cysts of the body.^[2-4] In case of craniofacial location, the lower jaw is more frequently affected than the maxilla with a proportion from 2:1 to 11:9.^[4-6] The body and the mandibular ramus are the main location with rare case reports in the coronoid process and the mandibular condyle.^[5-7] Age of presentation of aneurysmal bone cysts is the first two decades of life, being infrequent in patients up to 20 years.^[1,2,8] There is a slight sex preponderance in females.^[2,9] The clinical signs and symptoms of these lesions are nonspecific and may lead to further difficulty in diagnosis.^[4] Occasionally, its presentation can be as a rapid expansive growing mass locally destructive that may be miss diagnosed as a high-aggressive or a malignant neoplasm.^[1-3,5] This rapid growth may be as a result of the erosion of one of the cortical plates of an asymptomatic slow

growth lesion that becomes then symptomatic.^[1,3] However, other authors consider a traumatic pathogenesis and local vascular alterations within a latent lesion as an explanation of this rapid progression in some aneurysmal bone cysts.^[2,10] Although they are non-neoplastic lesions with possible local aggressiveness, a differential diagnosis with other multicystic processes like ameloblastoma, ossifying fibroma, epithelial cyst, giant cell granuloma and sarcomas should be established.^[3,5,8]

CASE REPORT

A 42 year old female was referred to our department with a left mandibular swelling two months of evolution, associated to pain and rapid growth within the left mandibular parasymphysis. A panoramic radiography showed presence of a well-defined mixed radiolucent-radio-opaque lesion with cortical border between 33 and 34 measuring approx. 2.5x3 cm extending mesio-distally from the distal aspect of 33 to the mesial aspect of 34. Mesial displacement of 33 and lateral displacement of 34 also could be appreciated in the left mandibular parasymphysis region (Fig. 1). Absence of symptoms like fever or suppuration excluded an infectious process. Panoramic radiography shows a lytic and expansive lesion in the left mandibular parasymphysis showing a “honeycomb” and “soap-bubble” like appearance with undefined moth-eaten margins. Occlusal radiograph shows buccal cortical plate expansion with scanty trabeculae within it, the lesion measures 2x3cm extending from the distal aspect of 32 to the distal aspect of 35 (Fig. 1). Because of the aggressive pattern of the lesion and the suspicious of a high grade neoplasm, an incisional biopsy under local anesthesia was performed through an intraoral approach. In this biopsy a fibrotic lesion with blood-filled spaces was observed. The histopathological examination showed numerous small and large vascular spaces lined by endothelial cells. Abundant pools of RBCs were seen. Hemosiderin pigment was seen at places along with giant cells, which was suggestive of aneurysmal bone cyst (Fig. 3). Surgical treatment with conservative resection of the lesion and enucleation with curettage was performed under general anaesthesia through intraoral approach. The jaw cortex was ballooned out and thinned by the mass with an “egg-shell” like appearance but

without loss of continuity. The lesion was formed of solid spaces and blood-filled cavities (Fig. 2). There was no significative blood loss during surgery. The histopathological examination of the surgical sample revealed a cystic lesion with many dilated blood-filled spaces. The diagnosis was of an aneurysmal bone cyst. No other associated lesions were confirmed in the histological diagnosis. The patient had a good evolution, showing a correct functional and aesthetic appearance four months after surgery, with a good healing of oral tissues in the postoperative photographs without signs of local recurrence (Fig. 4).

DISCUSSION

Aneurysmal bone cysts were first described in the literature by Jaffé et al.^[11] The term “aneurysmatic” refers to the “blow-out” effect or expansion of the affected bone that appears in this type of lesions. This fact provides a radiolucent expansive imaging, frequently multilocular, in the X-ray studies.^[3,12] Nowadays, etiopathogenesis of these pseudocysts is still controversial. There are some theories that try to explain its origin and classify these cysts in primary or secondary lesions. Most of these cysts are considered as congenital primary lesions that may co-exist with other osseous pathologies.^[6] Other authors suggest a vascular origin, in which local hemodynamic disturbances, like arterio-venous shunts or malformations, would lead to increased venous pressure and subsequent bony resorption and destruction of the vascular bed that would form these lesions.^[5,11] Other different theory considers that aneurysmal bone cysts are secondary lesions related to degeneration of a pre-existing bone lesion such as the central giant cell granuloma, fibrous dysplasia or ossifying and cementifying fibromas. However, this origin has not been demonstrated in histopathological examinations.^[3] This occurs in the histological analysis of the present case, in which no clear microscopic signs of other pre-existing lesions were found. So we have to assume the primary origin of the lesion, in despite of the small radiolucency near the mandibular parasymphysis described in the panoramic radiography. Histologically, these cysts are described as osteolytic lesions with blood-filled cavities and sinusoidal spaces, separated by fibrous connective tissue septa with osteoid trabeculae. Variable

Fig. 1: Pre-Operative Photographs and X-Rays

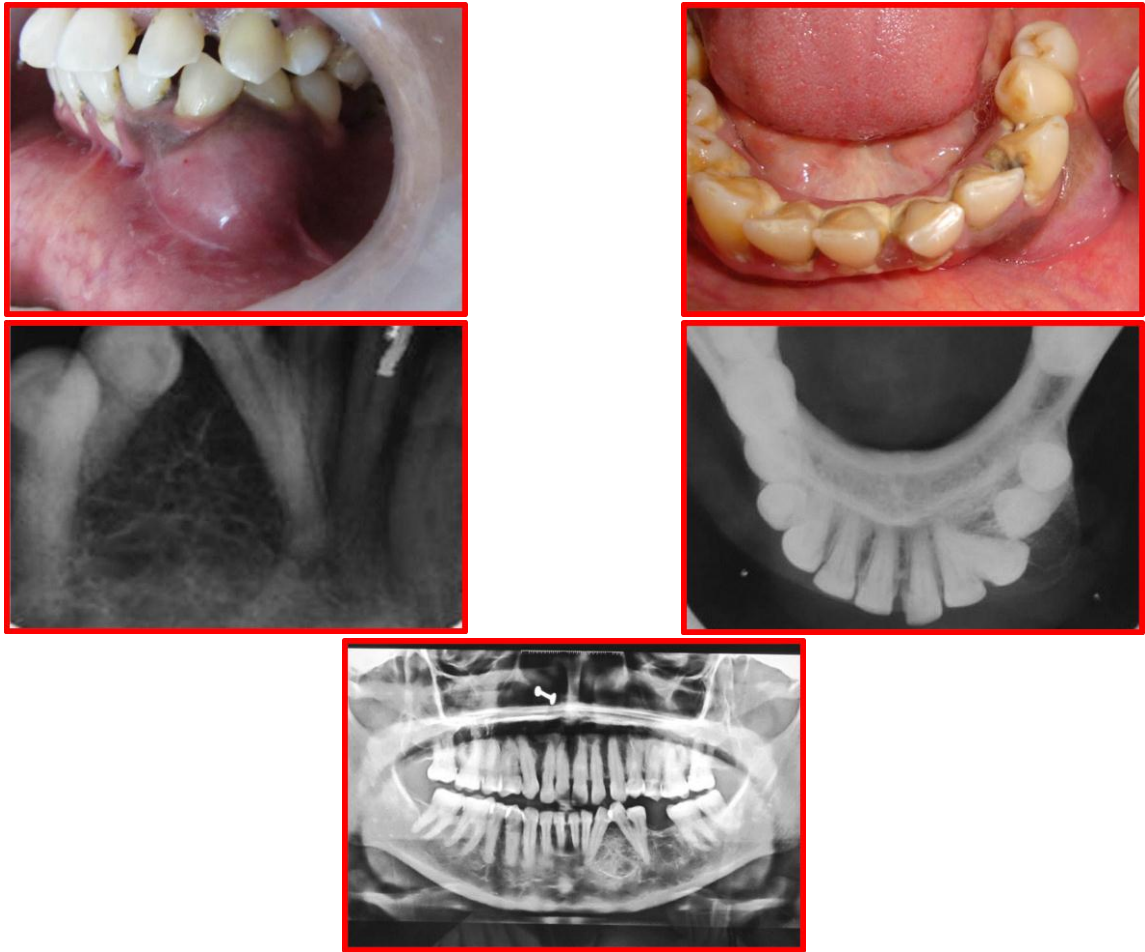


Fig. 2: Intra-Operative Photographs

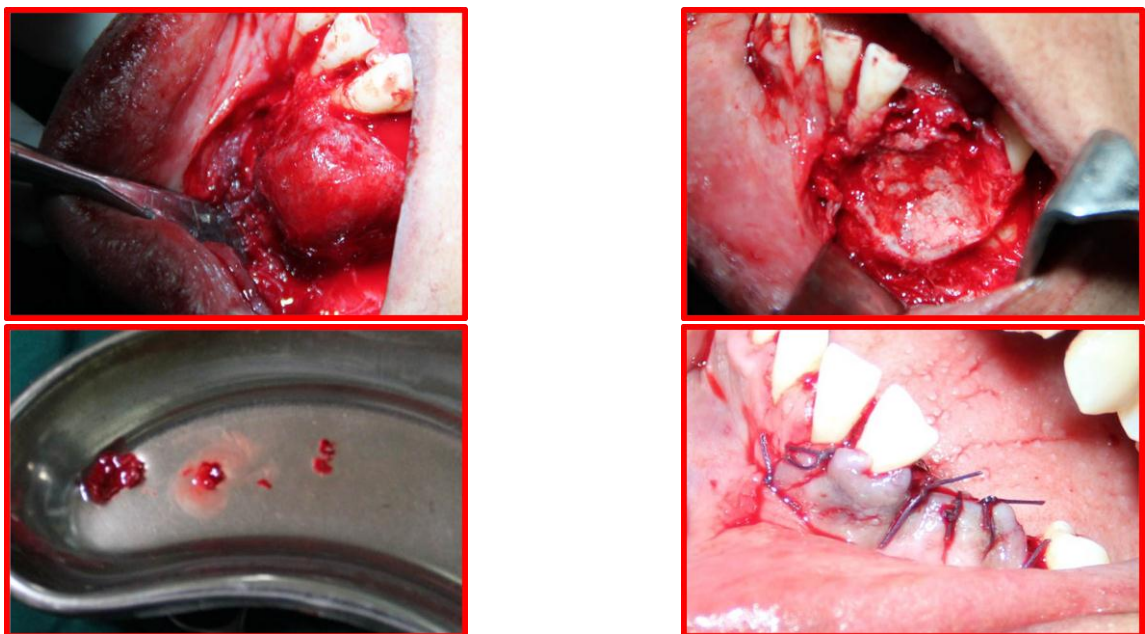


Fig. 3: Histopathological Sections

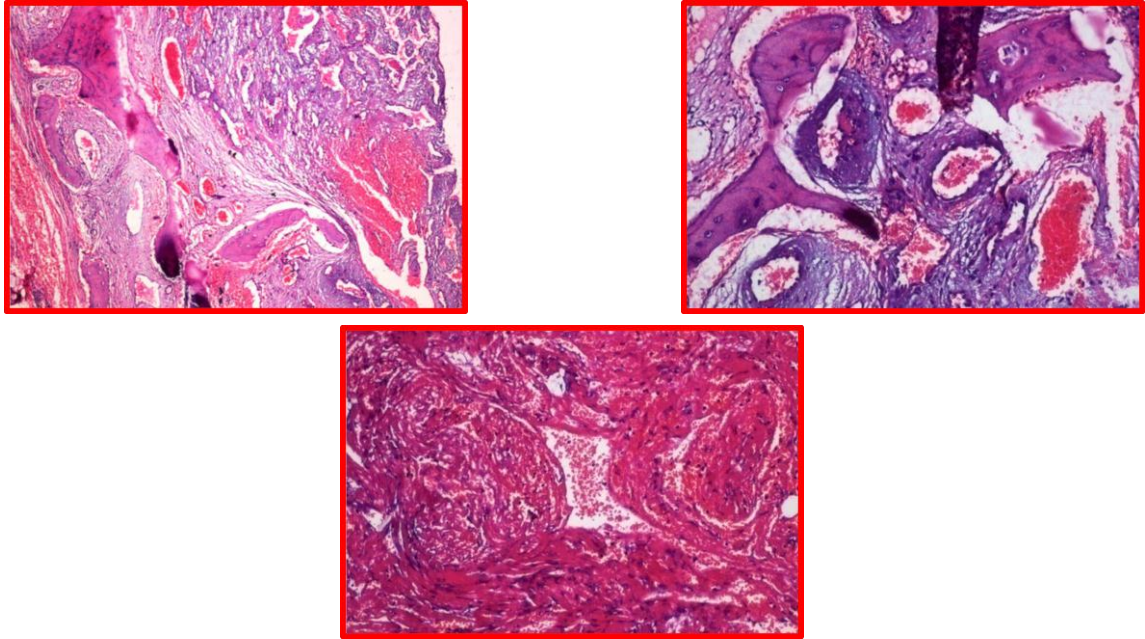
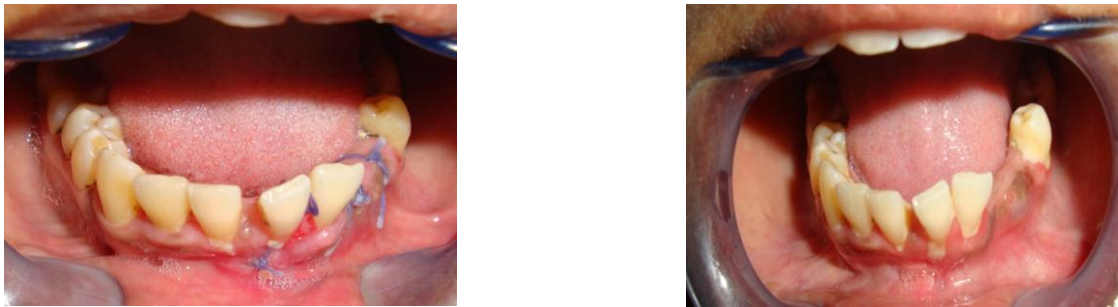


Fig. 4: Post-Operative Healing After 1 Week and Four Months



amount of hemosiderin and giant cells can be found.^[1,2,13] This description is characteristic of the “classic or vascular” form, which is the most frequent. “Solid” form is the other histological type that represents only a 5% of all the cases. This form is a noncystic variant with solid gray-white tissue, hemorrhagic foci and abundant fibroblastic and fibrohistiocytic elements with osteoclast-like giant cells. Osteoblastic differentiation areas with osteoid and calcifying fibromyxoid tissue complete the picture.^[2,5] A third variant “mixed” form demonstrates elements of both vascular and solid types.^[5] The most typical clinical presentation of these lesions is a well-defined swelling of soft tissues due to expansion of the adjacent bone, causing noticeable facial asymmetry.^[3,14,15] They usually present a slow progressive growth until cortical plates are eroded at any point and then show a rapid growth. Malocclusion can be a consequence of facial deformity.^[10] Pain is an infrequent

symptom except of rapid growth cases as the present case.^[4] Other less common clinical presentations could be root resorption of teeth, disesthesias, proptosis, diplopia and progressive nasal obstruction in maxillary lesions.^[4,8] Solid aneurysmal bone cyst is usually asymptomatic whereas vascular form usually presents an invasive rapid growing evolution with extension to overlying tissues.^[5,13,14] MRI is mandatory in complex cases such as the reported case, in order to improve plain radiographic examination and CT-scan as it is more accurate in soft-tissue contrast.^[12] Panoramic radiography frequently shows the presence of a cystic radiolucent imaging, usually multilocular, with a cystic meshwork divided by coarse septa.^[12] Bony cortex can also be expanded. The multilocular effect gives this cyst the characteristic but no pathognomonal “honeycomb” and “soap-bubble”- like appearance seen in other lesions such as giant cell granuloma, myxoma,

desmoplastic fibroma, haemangioma, keratocyst, ameloblastoma and other tumours.^[3,8,12] Occasionally, destruction of bony cortex may be identified, displaying a periosteal reaction imaging or “ray-sun” effect that is characteristic of osteosarcomas, with which differential diagnosis should be done.^[5,12] CT-scan accurately identifies tissue septa. MRI findings of fluid-fluid levels inside the lesion are high specific of aneurysmal bone cysts.^[3,12] Angiography is only occasionally used in the diagnosis of these pseudocysts, however it may be necessary if an haemangioma or a high grade neoplasm is suspected when MRI shows hyper vascularization.^[4] Fine needle aspiration and incisional biopsy may be also performed when high grade tumours are suspected.^[3] Nowadays, treatment of choice is conservative surgical resection of the lesion. This must be limited to careful enucleation or curettage of the mass as it is a benign process. Segmentary resection must be done only in case of multiple recurrences or extension to overlying tissues.^[1,3,4,6] Recurrence rates range from 20% to 30% according to different series and seems to occur most frequently within the first year after surgery.^[3,4,6] This is usually attributed to incomplete removal of the lesion especially in soft tissue invasive cases.^[3,5,10,13] Several authors recommend immediate reconstruction of the defect with autogenous grafts in cases of aesthetic deformity and in cases with high risk of fractures and loss of mandibular continuity.^[1,3,6,15]

CONCLUSION

As the radiological features of ABC are varied, resembling many lesions, histopathological analysis is a must for the diagnosis.

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