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Giant aneurysmal bone cyst of the mandible with unusual presentation

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Abstract

Aneurysmal bone cysts are rare benign lesions of bone tissue, infrequent in craneofacial 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 29-year-old Caucasian male with a big swelling in the left mandible associated to pain and rapid growth. He referred previous extraction of the left inferior third molar. On the X-ray study, an expansive multilocular and high vascularized bony lesion within the mandibular angle was observed. It produced expansion and destruction of lingual and buccal cortex. An incisional biopsy was performed showing a fibrous tissue with blood-filled spaces lesion suggestive of an aneurysmal bone cyst. After selective embolization of the tumour, surgical resection was done with curettage and immediate reconstruction of the defect with an anterior iliac crest graft.

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.

Key words: Aneurysmal bone cyst, mandibular tumours, benign bone lesions, mandibular pseudocysts.

Introduction

Aneurysmal bone cysts consist on 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 craneofacial 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 from (4). Occasionally, its presentation can be as a rapid expansive growing mass locally destructive that may be misdiagnosed 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 a 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 Reports

A 29-year-old caucasian male was referred to our department with a left hemifacial swelling two months of evolution, associated to pain and rapid growth within the left mandibular angle. He had extraction of the left inferior third molar just before the beginning of clinical symptoms. A previous panoramic radiography carried by the patient showed a small radiolucency of less than 5 mm in the left mandibular angle but it was not considered at that moment (Fig. 1A). Absence of symptoms like fever or suppuration excluded an infectious process.

In a recent panoramic radiography, a litic and expansive lesion in the left mandibular angle was identified showing a “honeycomb” and “soap-bubble”-like appearance

with undefined moth-eaten margins. The basilar cortical plate of the mandible was also desestructurated (Fig. 1B). A big litic and multilocular mass of 7x5 cm within the mandibular angle with destruction of the lingual and buccal cortex was confirmed in the CT scan. Radiographic examination was completed with a magnetic resonance imaging (MRI) in which this expansive mass showed signs of hipervascularization and fluid-fluid levels after contrast administration (Fig. 1C).

Under the suspected diagnosis of a high vascularized neoplasm, a supraseductive angiography was requested in which a big vascularized tumour was identified with blood supply from the left internal maxillary and the left facial arteries and with arterio-venous shunts. Because of the aggressive pattern of the lesion and the suspicious of a high grade neoplasm, an incisional biopsy under general anesthesia was performed through an external submandibular approach. In this biopsy a fibrotic lesion with blood-filled spaces was observed. The histopathological examination showed the presence of a fibromuscular tissue with organized fibrohematic content that suggested the diagnosis of a giant aneurysmal bone cyst.

Surgical treatment with conservative resection of the lesion and immediate reconstruction of the defect was performed. Due to the hipervascularized aspect of the mass, in order to decrease intraoperative blood loss a previous selective embolization with polyvinyl alcohol microparticles was done two days before surgery by canalization of internal maxillary and facial arteries. The post-embolization images showed significant decrease

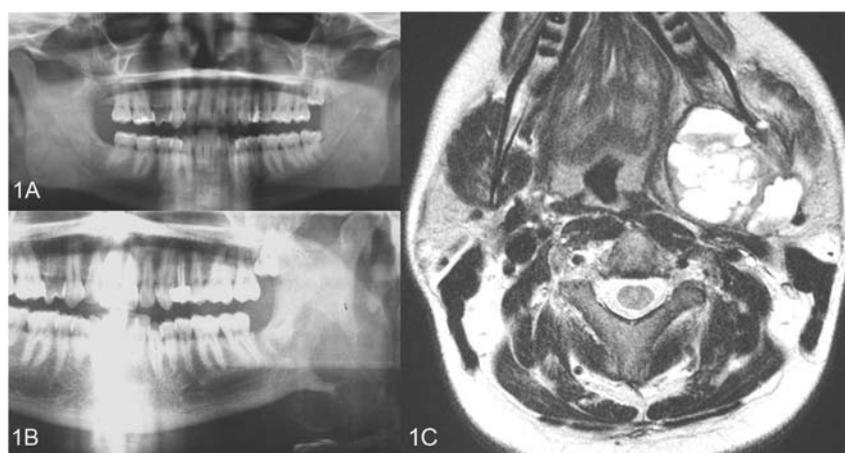


Fig. 1A. Initial panoramic radiography before left third molar extraction.

Fig. 1B. Panoramic radiography in which an expansive multilocular lesion within the left mandibular angle is observed.

Fig. 1C. Contrast MRI showing a cystic lesion divided by coarse septa is observed, with deformed “moth-eaten” bony cortex and fluid-fluid levels.

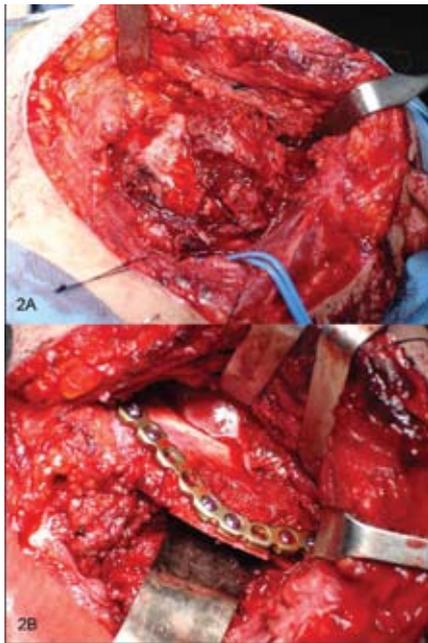


Fig. 2A. Blood-filled cystic lesion within the mandibular angle, with desestructured and "egg-shell" like appearance cortex.

Fig. 2B. Reconstruction of mandibular defect with a corticospongiosa anterior iliac crest graft fixed by a titanium reconstruction plate.



Fig. 3. Histopathological imaging of the lesion in which fibrous septa and blood-filled sinusoidal spaces are identified (H-E x10).

of lesion's blood flow. The patient underwent conservative resection of the tumour located at the left mandibular angle tumour through an external submandibular approach, with previous dissection of the external carotid artery. 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. 2A). Primary reconstruction of the defect was done with a corticospongiosa anterior iliac crest non vascularized graft fixed with a titanium reconstruction plate (Fig. 2B). 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 (Fig. 3). These cavities were separated by fibrous septa with osteoid tissue, giant cells and macrophages with occasional mitoses, no atypias and hemosiderin foci. 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 twelve months after surgery, with a good healing of the autogenous graft in the postoperative CT-scan and without signs of local recurrence.

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 coexist 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). Another pathologic mechanism could be bone trauma that may facilitate these hemodynamic alterations (10). In that way, trauma associated to dental extraction might have acted as a mechanism of rapid growth in the reported case. 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 micros-

copious signs of other pre-existing lesions were found. So we have to assume the primary origin of the lesion, in spite of the small radiolucency near the mandibular angle described in the initial 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 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 or "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 resorptions 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 not pathognomonic "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 haemangio-

ma or a high grade neoplasm is suspected when MRI shows hypervascularization (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).

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