

Aneurysmal bone cyst of the mandible: Case presentation and review of the literature

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ABSTRACT

The aneurysmatic osseus cyst is a very infrequent bone lesion which in some occasions can be found at the craneofacial skeleton. Among all the cystic lesions that can be found at the mandible or the maxilla it is very rare. On the other side it is at the same time very interesting in terms of its differential diagnose with other types of maxillary bone lesions. We present the case of a Caucasian male with an aneurysmatic cyst located at the right angle of the mandible and a review of the literature concerning the case. We have focused on the differential diagnose, mainly with the malignancies that can be found at this location. We also comment the therapeutic options classically described for these kind of pathologies. In our patient, the surgical excision allowed a complete removal of the lesion and a posterior bone healing which made possible a implant-supported rehabilitation of the edentulous segment.

Key words: *Anurysmatic, bone cyst, mandible cyst, ameloblastoma.*

RESUMEN

El quiste óseo aneurismático es una lesión ósea infrecuente que raramente se localiza a nivel del esqueleto craneofacial. Es una lesión de naturaleza benigna pero de comportamiento localmente agresivo. Del conjunto de lesiones quísticas de los maxilares, constituye una variedad muy poco frecuente, que presenta sin embargo características diferenciales, respecto a otro tipo de patologías con carácter de malignidad. Presentamos un caso clínico de un varón caucásico que presentaba dicha lesión a nivel mandibular y revisamos la literatura, centrándonos en el diagnóstico diferencial con lesiones neoplásicas como el ameloblastoma o el tumor de células gigantes. Así mismo, comentamos las diferentes posibilidades terapéuticas clásicamente descritas para el tratamiento de este tipo de patologías. En el caso que nos ocupa, el tratamiento fue quirúrgico, con cureteado exhaustivo de la lesión. Dicho tratamiento consiguió una extirpación completa y una posterior regeneración ósea local, que ha permitido una rehabilitación implantosoportada del segmento dentario sobre el que asentaba la lesión inicial.

Palabras clave: *Quiste mandibular, quiste óseo maxilar, quiste aneurismático, ameloblastoma.*

INTRODUCTION

The Aneurysmal Bone Cyst (ABC) is an infrequent bone lesion. Only 60-70 cases have been described in the Head and Neck region. Although ABC is a benign lesion, it can behave locally in an aggressive manner because of its rapid growth and osteolytic capacity. This lesion represents less than 1% of all the bone cysts biopsied. There are many theories about its etiology, mostly referring to alterations of the homeostatic-vascular equilibrium of the bone (1). What seems clear is that it is not a tumour, which is one of the most differential characteristics (2) between the ABC and two other similar entities like the Giant Cell Tumour or the Ameloblastoma (3). They can usually be found in the metaphysis of the long bones, mainly in the "tibia and the femur". Between 2-12% of the ABC is located at the head and neck region (4-5). From this last group almost 90% of the cases affect the posterior mandible (body of the mandible) (40%), ramus (30%), angle (19%), symphysis (9%) and condyles (2%). (6-9). The median age at diagnosis is 13 years old. 80 % of the patients are under 20 years old. There is a slight greater incidence in women (62%).

Histology:

- Conventional (95%): Osteolytic lesion. Expansive with vascular lagoons of variable size, separated by connective tissue that includes bone trabecula, osteoid tissue and multinucleated giant cells.
- Solid (5%): Solid Mass without a cystic compound of irregular density. Contains multiple hemorrhagic foci. It has a great fibroblastic, osteoblastic and osteoclastic compound.

DESCRIPTION OF THE CASE

In the case, the subject is a 29 year old Caucasian male who comes to our office for dullness and pain at the right angle of the mandible. In the Clinical Report, the only remarkable data was the previous excision of another cystic lesion at the same location, two years before. The physical examination showed pain and an expansion of the outer cortex at the 45-47. In the Panorex, we noted a unilocular radiolucent lesion 3 cm in diameter between the 45-47. The Denta-Scan report: Cystic lesion at the right body of the mandible of 3.4× 1.8 cm between the 45 and the 47. Bone erosion is not observed. (Fig 1-3)



Fig. 1. PANorex image showing an osteolytic lesion in the region of the 45-47.



Fig. 2. Axial CT-scan image showing the cystic lesion that involves all the mandible width.



Fig. 3. Sagittal CT-scan image showing in greater detail the lesion and its relationship with the first molar roots and the inferior alveolar nerve canal.

The extirpation of the cyst was performed under general anaesthesia. After raising a mucoperiosteal flap with a discharge incision distally to the 44, a complete excision of the lesion was performed followed by an extensive curettage. Finally the flap was repositioned and sutured with reabsorbable Vicryl 3/0. Prophylactic antibiotic was administered as well as corticoids on the following days. (Fig 4)



Fig. 4. Panorex image showing no recurrence 6 months after the surgery.



Fig. 5. Panorex image showing implant rehabilitation and no recurrence of the ABC.

In the follow-up, a Panorex was taken after one month, three months and 6 months. Approximately one year after the surgery, an implant-prosthetic rehabilitation was accomplished after placement of two 3I implants (13mm) at the 46-47 position. One year later no further controls have showed a recurrence of the lesion. (Fig 5)

DISCUSSION

The majority of the ABCs found in the head and neck region measure between 1 to 10 cm in its maximum diameter. In the Macroscopic examination ABCs have spongelike appearance, consisting of blood-filled cavities separated by thin, fibrous septa.

The main symptoms include dull pain and/or oedema. They usually have rapid growth. Depending on its location, other signs/symptoms can be found like headache, diplopia, loss of vision, proptosis, tooth mobility, hearing loss, etc.

The Radiological features include: Cystic bone expansion. Honey-comb or soap bubble like inner structure. Sometimes we can also see a destruction of the bone cortex and periosteal reaction (2,4,5).

All of these characteristics are not exclusive to the ABC. They can be found in different grades in many other types of lesions: unichameral cyst, ossifying fibroma, ameloblastoma, giant cell granuloma or even in fractures.

Besides, concerning its histological similarity with the brown tumours, it is recommended to study the seric levels of calcium, phosphate and alkaline phosphatase.

One of the most controversial issues of the ABC refers to whether its origin is a primary or secondary lesion.

Some authors like Struthers and Shears (1), consider it as a secondary lesion to a trauma followed by the development of a subperiosteal haematoma.

Swing (3) suggested that the ABC is derived from another type of benign lesion like a giant cell tumour in which some changes have made possible the communication between the stroma and the medullaris vessels. When this communication is maintained, an Aneurysmal Bone Cyst should be established. If the connection is interrupted, a giant cell granulome is constituted.

Lichenstein(2) added that maybe some alterations of local circulation may enhance the venous pressure and therefore result in a subsequent dilatation and resorption of the bone adjacent to the vessels.

In summary there is no consensus about the pathogenesis of the ABCs.

The vascular variety behaves more aggressively, including local invasion of the adjacent tissues in contrast with the solid type.

In a review published in 2003 (10) about the radiological characteristics of the ABCs, they concluded that the most differential ones were the following:

- Multiple anomalous branches in the angiogram.
- Multi-locular cystic structure in the CT with bone window.
- Presence of liquid both in the CT or MR
- Accumulative pattern in the scintigraphy and in the angiography with radionucleids.
- Bubble like structure in the MR T2W1

Concerning the treatment, although many options have been performed, the gold standard is still the surgical excision and curettage of the cavity (11,12). The use of Radiotherapy is not recommended because of the probability of radio-induced tumours.

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