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CASE REPORT

Solitary mandibular bone cyst. Case report and literature review

Quiste óseo solitario mandibular. Reporte de un caso y revisión de la literatura

Gabriela Wong-Romo,* Erick Carrillo-Terán,* Esther Ángeles-Varela§

ABSTRACT

Solitary bone cyst of the mandible is an intra-osseous cavity lacking epithelial lining considered a pseudocyst. Due to its uncertain etiology and pathogenesis, it has received several names such as traumatic bone cyst or idiopathic bone cyst. From a clinical perspective, it is oftentimes an asymptomatic lesion, with festooned borders when located between dental roots. It is an empty cavity but might contain blood, serous or serous-hematic fluid and can be perceived in routine X-ray examinations. The present article describes the case of a solitary bone cyst located in the body of the mandible of a 17-year old female patient. Afflicted with Fallot's tetralogy. Clinical, diagnostic and radiologic aspects as well as treatment are described.

RESUMEN

El quiste óseo solitario de la mandíbula, es una cavidad intraósea sin recubrimiento epitelial, considerado un pseudoquiste, ha recibido diversas denominaciones debido a su etiología y patogenia inciertas como quiste óseo traumático, quiste óseo solitario y quiste óseo idiopático. Clínicamente suele ser una lesión asintomática, muestra bordes festoneados cuando está localizado entre las raíces dentales, es una cavidad vacía pero puede contener sangre, fluido seroso o serohemático y es descubierta en exámenes radiológicos de rutina. En este artículo se presenta un caso de quiste óseo solitario localizado en el cuerpo mandibular que acomete a un paciente femenino de 17 años de edad con tetralogía de Fallot, revelando aspectos clínicos, diagnósticos e imagenológicos y tratamiento.

Key words: Simple bone cyst, solitary bone cyst, traumatic bone cyst, haemorrhagic bone cyst, single chamber bone cyst, extravasation bone cyst, idiopathic bone cavity, progressive bone cavity.
Palabras clave: Quiste óseo simple, quiste óseo solitario, quiste óseo traumático, quiste óseo hemorrágico, quiste óseo unicameral, quiste de extravasación, cavidad ósea idiopática, cavidad ósea progresiva.

INTRODUCTION

Lucas and Blum first described solitary bone cysts of the mandible in 1929. Nevertheless, it was not until 1949 that Rushton established the diagnostic criterion of this condition. It was defined as a simple cyst lacking epithelial lining with intact bone wall, filled with fluid and lacking evidence of chronic or acute inflammation.[1,2]

Donkor and Punnia-Moorthy suggested a possible sub-classification of solitary bone cyst based upon its content: empty cysts could be named idiopathic cysts, cysts with solid filling could be named according to the histological aspect of its content (fibrous or with granulation tissue), cysts with liquid content with a biochemical profile similar to blood plasma could be called extravasation cysts.[3]

These cysts have been described with different terms such as: solitary bone disease, solitary bone cyst, haemorrhagic cyst, extravasation cyst, single chamber cyst, simple and idiopathic cysts. This diversity in terms only reflects the uncertain origins of this lesion.[3-5]

The World Health Organization's (WHO) International Tumor Histological Classification accepted the term «simple bone cyst» in 1971, and the «term solitary bone cyst» in 1992 so as to differentiate this lesion from other cystic lesions of the jaws.[6,7] In the 1997 WHO classification, solitary bone cyst is included in the group of bone-related non-neoplastic diseases along with aneurismatic cysts, ossifying fibroma, fibrous dysplasia, bone dysplasia, giant cell central granuloma and cherubism.[1,4,8]

* Maxillofacial surgeon at the Specialty Regional Hospital Bicentenario de la Independencia, ISSSTE (National Institute of Social Services for Government Employees), State of Mexico.
§ Graduate, School of Dentistry, Faculty of Graduate Studies, Iztacala National University of Mexico (UNAM).

Maxillofacial Surgery Service Social Security Institute for Government Worlcers (ISSSTE) Specialized Hospital «Bicentenario de la Independencia».

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LITERATURE REVIEW

Epidemiology

Solitary bone cysts are also found in other parts of the skeleton, more commonly in long bones (90-95%) with predominance in the metaphysis region of the humerus proximal ends (65%) as well as femoral shaft (diaphysis) (25%).[6,9]

According to scientific literature, solitary cysts occur frequently in young subjects, especially males. This could be due to the fact that males are more exposed to traumatic lesions than females. This is particularly the case of extra-facial solitary cyst variants[9,10.] Nevertheless, it has also been reported that solitary bone cysts have their onset in equal proportion in males and females in their second decade of life.[4,11]

In contrast, authors such as Cortell-Ballester and Peñarrocha referred having observed that solitary bone cysts were more frequently present when subjects were under 20 years of age; they exhibited mild predilection for females.[1,12]

Saito et al concluded that solitary bone cysts in young subjects were characterized as being asymptomatic, radio-lucid lesions with minimal expansion in the mandible, whereas solitary bone cysts in older groups were radio-opaque with cement hyperplasia or dysplasia and with loss of tooth-related *lamina dura*.[3,8]

Horner and Forman describe a different sub-group of solitary bone cysts characterized for appearing with fibrous-bone lesions seemingly developing in older patients.[8]

Incidence of these cysts in the upper jaws is not common, it represents 1% of all maxillary cysts. Most solitary bone cysts have been located in the body of the mandible, in the premolar and molar region (75%), with possible and sometimes severe extension towards the back. The second most common site is the mandibular symphisis.[12,13] Few cases are reported in the mandibular ramus, condyle and upper jaw.[11] It has been proposed that the maxillary sinus might impede visualization of maxillary lesions.[2] This condition can appear as multi-lobular,[13] multiple[2,3,14] and bilateral[14,15] lesions.

Symptomatology

In most cases, solitary cysts of the mandible are asymptomatic, lacking inflammation or other functional signs; teeth adjacent to the lesion respond to vitality tests. These lesions are frequently accidentally discovered in x-ray studies.[12,16,17]

Some patients report pain, inflammation and/or tooth sensitivity.[3] These cysts can uncommonly present fistula, root resorption, paresthesia and/or pathological fractures.[7]

These lesions rarely cause complications, nevertheless, the possibility of a pathological fracture in extended lesions cannot be overlooked.[4]

When solitary bone cysts of the mandible are associated to bone cement dysplasia (florid bone dysplasia),[3,6,18] cementoma, odontoma[5,18] and mesodermal tumor,[10] patients have reported pain and inflammation.

Radiographic findings

In most cases, solitary bone cysts appear as a radio-lucid lesion with irregular but well defined borders, of normal appearance and mixed with cancellous bone, they might be partially sclerotic.[3,8,12,17,19,20] The main characteristic of solitary bone cysts is their «festooned effect» when they extend towards dental roots, nevertheless, this festooned line has equally been present even in edentulous zones.[8,11,19,21] Matsumura et al describe a «double line» circumscribing the lesions in 19 cases.[3]

Greater recurrence has been reported in long bones where solitary cysts appear with a festooned border when compared to solitary cysts with smooth borders; the aforementioned has not been reported in the lower jaw. Therefore, a festooned margin is a sign of possible recurrence, nevertheless it must not be confused with inter-dental festoon associated to intact *lamina dura*.[4]

Loss of *lamina dura* is mainly observed in patients over 30 years of age. In younger subjects it is minimal.[22]

Another radiographic characteristic is the widespread extension of the lesion along the mandibular body without bone expansion, cortical bone becomes thinned down due to endosteal erosion. In most cases, there is no displacement of adjacent teeth, dental root divergence or loss of *lamina dura*.[3,20,23] Root resorption is rare (0-8.7%) and can cause *lamina dura* disappearance in 16-62% of all cases.[8] In some studied cases, the mandibular canal partially disappears without any displacement, and in others displacement of said canal can be suspected.[3,22,23]

Mathew et al concluded that canal displacement could be the result of local increase of osmotic pressure resulting from the decomposition of hemolytic products, or the fact that the canal could be pushed in a lingual direction by the formation of an intra-bone hematoma as result of a given traumatic event. Both aforementioned mechanisms could support the etiology of solitary bone cyst due to trauma.[24]