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Aneurismal bone cyst: a conservative surgical technique. A case report treated with a small access osteotomy

ABSTRACT

Background *Aneurismal bone cysts (ABCs) are benign, non-neoplastic, expansive, and locally destructive lesions that may occur rarely. They are well defined and usually occur in the long bones, pelvis and vertebrae. According to the 2005 World Health Organization (WHO) histological classification of odontogenic tumours, ABC is classified as a bone-related lesion as ossyfing fibroma, fibrous dysplasia, osseous dysplasia, central giant cell lesion (granuloma-CGCL), cherubism and simple bone cyst (SBC). ABCs, as CGCLs and SBCs may arise as a consequence of an earlier trauma causing an overflow of blood into the bone, though a number of pathogenic theories have been put forward. The aim of this study is to present an unusual case of aneurismal bone cyst and to compare the different possibilities of surgical treatment after review of the literature.*

Case report *ABC was localised in the posterior right region of the mandible in an 11-year-old boy, and removed by a surgical procedure involving a small access osteotomy of the mandibular ramus with removal of the cortical bone in order to enucleate the whole lesion, the wisdom tooth and to preserve the healthy bone.*

Keywords *Aneurismal bone cyst; Osteolytic lesion; Small access osteotomy.*

Introduction

Aneurismal bone cysts (ABCs) are benign, non-neoplastic, expansive, and locally destructive lesions that may occur rarely. They are well defined and they usually occur in the metaphysis of the long bones, the pelvis and the posterior arch and spinous processes of vertebrae. According to the 2005 World Health Organization (WHO) histological classification of odontogenic tumours, ABS is classified as a bone-related lesion as ossifying fibroma, fibrous dysplasia, osseous dysplasia, central giant cell lesion (granuloma), cherubism and simple bone cyst [Barnes et al., 2005].

ABC is defined as "an expansive osteolytic lesion often multilocular, consisting of blood filled spaces separated by connective tissue septa containing trabeculae of osteoid tissue and osteoclast giant cells" [Schajowicz, 1993].

ABC occurs in young patients <30 years with a peak in the second decade, and has an incidence of 0.014/100,000 patients. The main localisations of ABC are long bones metaphysis as the femur (50%) and the tibia (12-30%) or the spine [Perrotti et al., 2004]. Only 1-3% of all ABCs are known to occur in the maxillofacial region. ABCs are more often found in the mandible, preferentially in the posterior region and the ascending ramus [Bernier and Bhaskar, 1958].

ABCs may arise as a consequence of an earlier trauma causing an overflow of blood into the bone, though a number of pathogenic theories have been put forward.

Simple bone cysts (SBCs), "an intraosseous pseudocyst devoid of an epithelial lining, either empty or filled with serous or sanguineous fluid" [Barnes et al., 2005], and central giant cell lesion (CGCLs), "a localized benign but sometimes aggressive osteolytic proliferation consisting of fibrous tissue with haemorrhage and haemosiderin deposits, presence of osteoclast-like giant cells and reactive bone formation" [Barnes et al., 2005] might have the same pathogenesis as ABCs. Those bone-related lesions are generally considered as sequelae of a traumatic event [Borgonovo et al., 2012].

Although the aetiopathogenesis of ABC, SBC and CGCL has not been clearly established, it has been suggested that they are the result of an exacerbated reparative process related to a previous trauma. Those lesions could

be the result of a vascular dysfunction leading to a local post-haemorrhagic ischemia, inducing an osseous aseptic necrosis and reactive granulomatous process [Harnet et al., 2008; Kauzman et al., 2004; Ustundag et al., 2002].

The hypothesis of traumatism as the aetiopathogenesis of those lesions is based on the occurrence of an intramedullary hemorrhage followed by a hematoma as a consequence of a trauma [Mayer et al., 1967]. The pressure due to hematoma in a healthy bone causes venous stasis and leads to an area of bone marrow necrosis and osteoclastic resorption [Rubin et Murphy, 1989]. This process is attributable to the decreased tissue pH [Howe, 1965]. A thrombosis or a lingering spasm of a terminal artery with ischemia and aseptic necrosis, may be caused by the trauma and thereby leading to cyst formation. All those vascular alterations are supposed to be related to the resorption phenomena. The process by which osteoclasts differentiate remains unknown. This theory could be applied to the mandible because of the numerous micro-traumas suffered by the teeth and alveolar process.

ABCs are classified as pseudocysts because they exhibit no epithelial lining. Additionally, they should be differentiated from true cysts or other pseudocysts (i.e., simple bone cysts with a static bone cavity) because their treatment is different. However, since the majority of these cysts involve subjective or no symptoms, most are discovered accidentally during radiography, while a true diagnosis is likely to be obtained only during surgery, on discovery of a non-epithelialised cavity [Asaumi et al., 2003]. ABCs can be diagnosed using conventional radiography, computed tomography (CT) and magnetic resonance imaging (MRI). However, some cases may be difficult to diagnose, even if examined by contrast-enhanced CT or MRI. Digital subtraction angiography (DSA) is considered the 'gold standard' in the assessment of vascularity, but not all ABCs exhibit intralesional blood flow [Bozbuğa et Turan Süslü, 2009; Kumar et al., 2009]. Conversely, dynamic contrast-enhanced MRI (DCE-MRI) [Yanagi et al., 2010] represents a less invasive method considering the risks of DSA and might be a prerequisite for DSA. It can provide haemodynamic information and measure lesion vascularity [Yanagi et al., 2010].

We report a typical case of aneurysmal bone cyst in the posterior right region of the mandible in a 11-year-old boy.

Case report

A 11 year-old boy was referred to the Maxillofacial Surgery Department of the Galeazzi Hospital, Milan, Italy, for examination of radiolucency in the right mandible (Fig. 1).

The osteolytic lesion, which had been detected during an orthodontic examination, was localised in the right retromandibular region associated with the roots of an unerupted wisdom tooth.

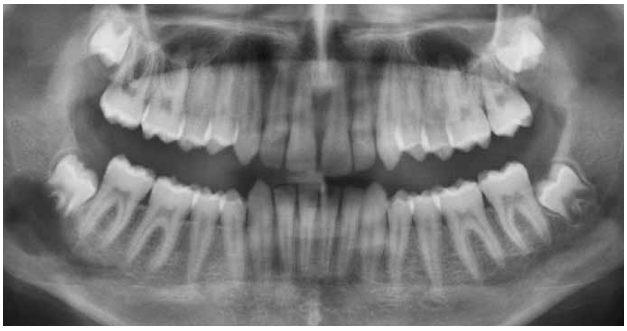

FIG. 1 Preoperative radiograph showing an osteolytic lesion of the right mandibular angle.

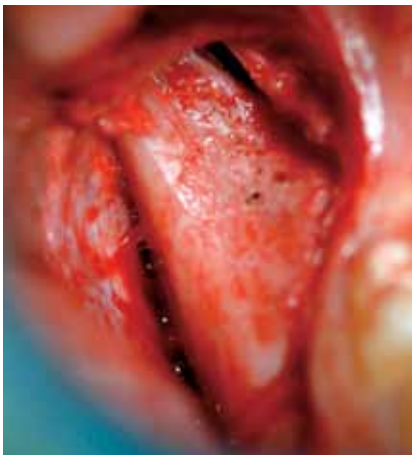

FIG. 2 Normal cortical bone was observed in the retromandibular region after the access flap.

He had not experienced pain in the mandibular region and no paresthesia was revealed. On clinical examination a normal bone was observed, without any swelling at the mandibular body. There were no palpable lymph nodes.

The general medical and family histories were non-contributory, and there was no history of facial trauma.

Imaging studies had been done in order to evaluate this lesion included orthopantomography (OPT), cone beam computed tomography and contrast-enhanced computed tomography. The imaging exams revealed a large expansive, unilocular lesion with a thin cortical residual bone in the right mandibular angle. The osteolytic lesion was in direct contiguity to the root of the right wisdom tooth and to the mandibular nerve canal.

No pathological roots resorption of tooth 4.7 and no significant contrast enhancement were observed.

Based on these data, a presumptive diagnosis of an unusual case of mandibular aneurysmal bone cyst was placed. It was decided to perform surgery, in order to have a histopathological evaluation and facilitate the correct diagnosis.

The surgery was performed under general anaesthesia. The procedure involved a small access osteotomy of the mandibular ramus with removal of the cortical bone in a single block to enucleate the whole lesion and the wisdom tooth (Fig. 2).

The residual bone cavity had no epithelial lining, the

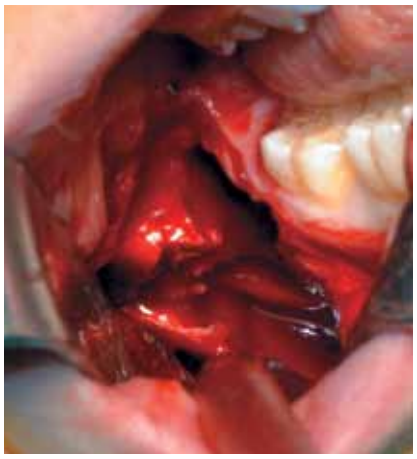

FIG. 3 The residual bone cavity after the small access osteotomy of the right mandibular ramus: the alveolar nerve is identified and preserved.

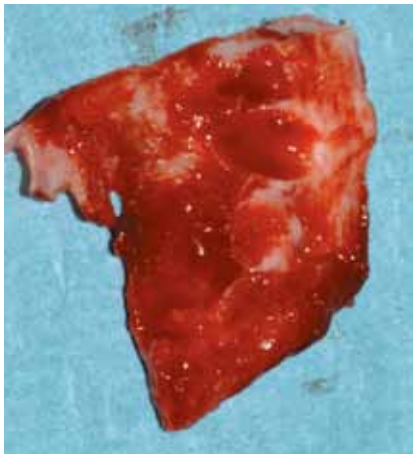

FIG. 4 The cortical bone block removed in order to enucleate the whole lesion and the wisdom tooth from the right angle of the mandible.

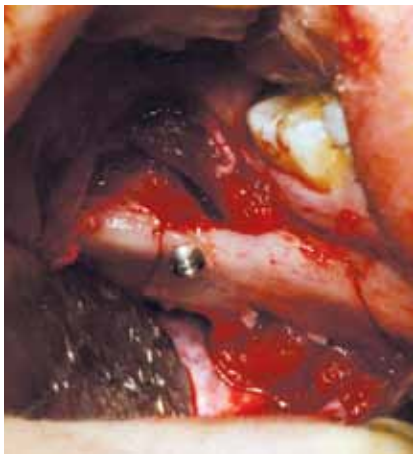

FIG. 5 The cortical bone block is replaced with micro osteosynthesis screws and adsorbable haemostatic gelatin sponge is placed into the bone cavity.

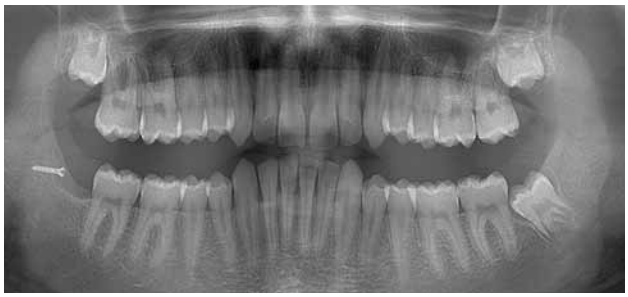

FIG. 6 Postoperative radiograph showing the cortical bone block replaced with a micro osteosynthesis screw and the optimal healing of the bone gap.

alveolar nerve was identified and preserved (Fig. 3). Absorbable haemostatic gelatin sponge was placed into the bone cavity and the cortical bone block previously removed during the small access osteotomy was replaced with a micro osteosynthesis screw (Fig. 4, 5).

The residual dental follicle removed during surgery and a specimen of the spongy bone surrounding the cyst was sent for histological examination. The microscopic examination revealed a specimen composed of strands and islands of an odontogenic epithelium, fragments of connective tissue with chronic inflammation, partially covered by fibrin, and fragments of mucosa and submucosa associated with blood extravasation. Based on the microscopic findings and clinical history, a diagnosis of aneurismatic mandibular cyst associated with the dental follicle of the wisdom tooth was made.

At the post-surgery follow-up no residual osteolytic lesion was observed, and no recurrences occurred after surgical curettage (Fig. 6).

Discussion

In 1893 ABC was firstly described as "homerus ossyfing haematoma" [Van Arsdale, 1893]. Later, in 1942 Jaffe and Lichtenstein used the scientific term aneurysmal bone cyst. These lesions were further defined by both of these authors as Jaffe-Lichtenstein disease [Jaffé et Lichtenstein, 1942].

Aneurysmal bone cyst is an uncommon lesion which has been found to occur mostly in long bones of the skeleton, and the spine.

In our case report the ABC was located in the typical site of the molar region of the mandible, involving the angle, as reported by Bernier and Bhaskar in 1958 in the first case of ABC located in the craniofacial skeleton [Bernier et Bhaskar, 1958].

Capanna et al. [1985] introduced a simple classification of ABC into three stages according to radiological and clinical aspects. The first stage presents an inactive cyst with complete periosteal and sclerotic borders, as our case report; the second stage, the active one, shows incomplete borders with defined margins. The last stage, known as aggressive, presents a uniform osteolysis with diffuse borders of the lesion. Inactive cysts usually show no proliferation, whereas active and aggressive cysts tend to recur.

ABC can behave in a locally aggressive manner, because of its rapid growth and osteolytic capacity, although it is a benign lesion. It is usually unilocular and radiographic differentiation from other osteolytic and expansive lesions, such as simple bone cyst, central giant cell lesion, giant cell tumor, non ossifying fibroma, ameloblastoma and sarcoma may be difficult because of the similarity between the radiographic appearance.

Many different diagnostic hypothesis were made in our case: the age of occurrence of the lesion was not typical,

since ABCs preferentially appear during the second decade. Also the contiguity of the lesion with the dental follicle might address the cause to a dental pathology. Only the angio-CT excluded the vascular origin of the lesion.

ABCs may develop if intramedullary clots due to trauma do not undergo lysis or resolution [Howe, 1965].

The most common form of treatment for this lesion is the surgical removal and curettage [Sun et al., 2009], unlike other cystic lesions that exhibit epitelial lining and may be approached with conservative surgical treatment based on marsupialisation [Borgonovo et al., 2011].

The extent of the bone resection depends on the size and location of the lesion, the age of the patient and the clinical aspect.

After surgical curettage the recurrence rate in the jaws ranges from 20 to 30% according to various studies [Gardre et Zubairy, 2000; Abuhassan et Shannak, 2010; Möller et al., 2011].

The large recurrence rates may be due to incomplete removal of the cyst during surgery as many authors sustain [Kalanta Motamedi, 1998].

As ABC may occur after an overflow of blood into the bone, the massive haemorrhage that may be encountered, which might require ligation of the external carotid artery as a precautionary measure, can be a problem that might lead to incomplete removal ant to frequent recurrence [Grecchi et al., 2012].

Although surgical curettage has been supplemented by cryotherapy and radiotherapy to decrease the recurrence rate, the use of the latter is strongly discouraged because it is likely to induce sarcomatous change in the irradiated bone.

In order to effect a complete cure the en bloc resection is the preferred treatment modality, but this is restricted to large and recurrent lesions, owing to the morbidity of the procedure [Cory et al., 1989].

Conclusion

The most common treatment for this lesion is the surgical removal and curettage. In this case the ABC is managed with a conservative surgical technique, avoiding a massive demolition of the involved bone. The cortical bone removed for the small access osteotomy is then repositioned in the original site. The absence of recurrence and the correct and optimal healing of the bone confirm that this surgical procedure is advantageus in order to preserve the anatomy of the mandibular bone in a young growing patient with ABC.

References

› Abuhassan FO, Shannak A. Non-vascularized fibular graft reconstruction after resection of giant aneurysmal bone cyst (ABC). Strategies Trauma Limb Reconstr 2010;5:149-54.

› Asaumi J, Konouchi H, Hisatomi M, Matsuzaki H, Shigehara H, Honda Y, et al. MR features of aneurysmal bone cyst of the mandible and characteristics distinguishing it from other lesions. Eur J Radiol 2003;45:108–12.

› Barnes L, Eveson JW, Reichart P, Sidransky D. Pathology and genetics of the head and neck tumors. World Health Organization Classification of Tumors. Lyon: IARC Press; 2005.

› Bernier J, Bhaskar S. Aneurysmal bone cyst of the mandible. Oral Surg Oral Med Oral Pathol 1958;11: 1018-1028.

› Borgonovo AE, Tafuro CM, Censi R, Poli P, Maiorana C. Minimally invasive surgical approach in a large mandibular solitary cyst: case report and review of the literature. Minerva Stomatol 2012 May;61(5):239-45.

› Borgonovo AE, Di Lascia S, Grossi G, Maiorana C. Two-stage treatment protocol of keratocystic odontogenic tumour in young patients with Gorlin-Goltz syndrome: marsupialization and later enucleation with peripheral ostectomy. A 5-year-follow-up experience. Int J Pediatr Otorhinolaryngol 2011 Dec;75(12):1565-71.

› Bozbuğa M, Turan Süslü H. Aneurysmal bone cyst of the sphenoid bone extending into the ethmoid sinus, nasal cavity and orbita in a child. Turk Neurosurg 2009;19:172–6.

› Capanna R, Betelli G, Biagini R, Ruggiere P, Bertoni F, Campanacci M. Aneurysmal bone cyst of long bones. Ital J Orthop Traumatol 1985;11:409-417.

› Cory DA, Fritsch SA, Cohen MD, Mail JT, Holden RW, Scott JA, et al. Aneurysmal bone cysts: Imaging Imaging findings and embolotherapy. AJR Am J Roentgenol 1989;153:369–73.

› Gardre KS, Zubairy RA. Aneurysmal bone cyst of the mandibular condyle: Report of a case. J Oral Maxillofac Surg 2000;58:439-43.

› Grecchi F, Zollino I, Candotto V, Gallo F, Rubino G, Bianco R, Carinci F. A case report of haemorrhagic-aneurismal bone cyst of the mandible. Dental Research J 2012;9(Suppl 2):S222-S224.

› Harnet JC, Lombardi T, Klewansky P, Rieger J, Tempe MH, Clavert JM. Solitary bone cyst of the jaws: a review of the etiopathogenic hypotheses. J Oral Maxillofac Surg 2008 Nov;66(11):2345-8.

› Howe GL. Haemorrhagic cysts of the mandible I. Br J Oral Surg 1965;3:55.

› Howe GL. Haemorrhagic cysts of the mandible II. Br J Oral Surg 1965;3:77.

› Jaffé H, Lichtenstein L. Solitary unicameral bone cyst with emphasis on the roentgen picture the pathologic appearance, and the pathogenesis. Arch Surg 1942;44:1004-1025.

› Kalantar Motamedi MH. Aneurysmal bone cysts of the jaws: Clinicopathological features, radiographic evaluation and treatment analysis of 17 cases. J Craniomaxillofac Surg 1998;26:56-62.

› Kauzman A, Li SQ, Bradley G, Bells RS, Wunder JS, Kandel R. Central giant cell granuloma of the jaws: assessment of cell cycle proteins. J Oral Pathol Med 2004;33:170-6.

› Kumar VV, Malik NA, Kumar DB. Treatment of large recurrent aneurysmal bone cysts of mandible: Transosseous intralesional embolization as an adjunct to resection. Int J Oral Maxillofac Surg 2009;38:671–676.

› Mayer R, Libotte M, Ruppol P. La lacune essentielle de la mandibule. Acta Stomatol Belg 1967;64:33.

› Möller B, Claviez A, Moritz JD, Leuschner I, Wiltfang J. Extensive aneurysmal bone cyst of the mandible. J Craniofac Surg 2011;22:841-844.

› Perrotti V, Rubin C, Fioroni M, Piattelli A. Solid aneurysmal bone cyst of the mandible. Int J Pediatr Otorhinolaryngol 2004;68:1339-1344.

› Rubin MM, Murphy FJ. Simple bone cyst of mandibular condyle. J Oral Maxillofac Surg 1989;47:1096.

› Schajowicz F. Histological typing of bone tumors. World Health Organization International Histological Classification of Tumors. Berlin: Springer Verlag; 1993.

› Sun Z, Sun H, Yang R, Zwahlen R, Zhao Y. Aneurysmal bone cyts of the jaws. Int J Surg Pathol 2009;17:311-322.

› Ustundag E, Iseri M, Keskin G, Muezzinoglu B. Central giant cell granuloma. Int J Pediatr Otorhinolaryngol 2002;65:143-6.

› Van Arsdale W. Ossifying haematoma. Ann Surg 1893;18:8-17.

› Yanagi Y, Fujita M, Hisatomi M, Matsuzaki H, Konouchi H, Katase N, et al. The utility of three-dimensional dynamic contrast-enhanced magnetic resonance imaging in delineating vessel-rich regions: A case report of an aneurysmal bone cyst of the mandible. Oral Radiol 2010;26:110-115.