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Case Report

A case report of haemorrhagic-aneurismal bone cyst of the mandible

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ABSTRACT

Haemorrhagic-aneurismal bone cysts (HABCs) are quite rare, benign, non-neoplastic, expansive, and vascular locally destructive lesions. They are generally considered sequelae of an earlier trauma causing an overflow of blood into the bone. HABCs are classified as pseudocysts and they should be differentiated from true cysts because their treatment is different. Since few of these cysts involve subjective symptoms, most are discovered accidentally during radiography, while a sure diagnosis is likely to be obtained only during surgery, on discovery of a non-epithelialised cavity. Here, we report a typical case of a haemorrhagic-mandibular cyst in a 13-year-old girl, which was treated by opening the cavity and scraping its walls following diagnostic arteriography and post-operative transcutaneous intralesional embolization. No further complications were recorded in the post-operative period, although the convalescence lasted for a time longer than expected, because of anemia. No further surgery was performed. She has been disease-free for two years. Evaluation of intralesional blood flow is important for HABCs because of the hemorrhagic risk in surgery. Embolization seems to be a useful procedure in the treatment of HABCs and could be tried as the treatment modality in the standard protocol for the treatment of HABCs.

Key Words: Arteriography, embolization, haemorrhagic-aneurysmal cyst, mandible

INTRODUCTION

Haemorrhagic-aneurismal bone cysts (HABCs) are quite rare, benign, non-neoplastic, expansive, and vascular locally destructive lesions. According to the 2005 World Health Organization (WHO) histological classification of odontogenic tumours, HABC is classified as a bone related lesion and is defined as 'an expanding osteolytic lesion consisting of blood-filled spaces of variable size, separated by connective tissue septa containing trabeculae of osteoid tissue and osteoclast giant cells'.[1]

HABCs are generally considered sequelae of an earlier trauma causing an overflow of blood into the bone, though a number of pathogenic theories have been put forward. HABCs 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 few of these cysts involve subjective symptoms, most are discovered accidentally during radiography, while a sure diagnosis is likely to be obtained only during surgery, on discovery of a non-epithelialised cavity.[2]

Pre-operative evaluation of the intralesional blood flow is still necessary because HABCs pose a heavy bleeding risk in surgery. Bleeding may occur during biopsy or surgery because HABCs are aneurysms with numerous pools of blood.[3] Thus, before HABC treatment or biopsy is initiated, noninvasive methods should be used for the diagnosis of lesions.[4] Most HABCs can be diagnosed using conventional radiography, computed tomography (CT) and magnetic resonance imaging (MRI). However,

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some cases may be difficult to diagnose, even if examined by contrast-enhanced CT or MRI.[3] Digital subtraction angiography (DSA) is considered the 'gold standard' in the assessment of vascularity, but not all HABCs exhibit intralesional blood flow.[5,6] Conversely, dynamic contrast-enhanced MRI (DCE-MRI) represents a less invasive method considering the risks of DSA and might be a prerequisite for DSA. It can provide hemodynamic information and measure lesion vascularity.[3]

Here, we report a typical case of haemorrhagic mandibular cyst in a 13-year-old girl, which was treated by opening the cavity and scraping its walls following diagnostic arteriography and post-operative transcutaneous intralesional embolization.

CASE REPORT

A 13-year-old girl was referred to Maxillofacial Surgery, Galeazzi Hospital, Milan, Italy, in April 2009 for examination of the radiolucency of the right mandible, which had been detected in a general dental clinic.

She had not experienced tenderness of the right mandible. On clinical examination, a normal bone without swelling at right body of the mandible, was observed. Intraoral examination revealed no percussion pain at the right premolar and molar regions and no paresthesia. There were no palpable lymph nodes, and the blood test was normal.

Her general medical and family histories were non-contributory, with no history of facial trauma. Imaging studies to evaluate this lesion included orthopantomography (OPT), CT and MRI. A panoramic radiograph revealed a large expansive, multilocular lesion with a thin bony rim in the right mandible, which was enlarged from the right, first premolar to the mandibular ramus. Pathological roots resorption of 4.6-4.7 dental elements was observed [Figure 1]. So, based on these data, a presumptive diagnosis of mandibular cyst was placed. It was decided to do surgery, in order to have a histopathological evaluation and facilitate correct diagnosis.

During the surgery, when attempting to avulsion of 4.6 dental elements, an unexpected haemorrhage occurred. The use of the haemostatic *tabotamp* only was able to stanch the haemorrhage. However, since blood tests showed a rapid decrease in haemoglobin level from 13.8 to 5.4 mg/dl, a post-operative diagnostic arteriography and a transcutaneous intralesional embolization were performed [Figure 2]. Blood transfusion was used to return to normal blood parameters.

Part of the material removed during surgery, including the extracted teeth 4.6 and 4.7, 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 spills. Based on the microscopic findings and clinical history, a diagnosis of hemorrhagic mandibular cyst was made.

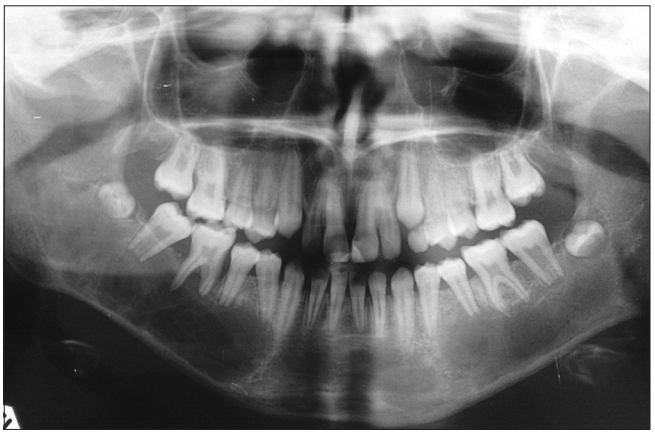

Figure 1: Pre-operative panoramic radiograph reveals a large expansive, multilocular lesion with a thin bony rim in the right mandible. It stretches from the right first premolar to the mandibular ramus; pathological roots resorption of 4.6-4.7 dental elements is present

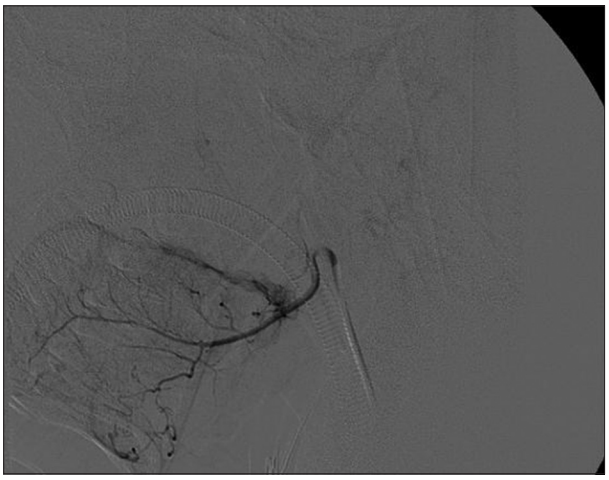

Figure 2: Post-operative transcutaneous intralesional embolization

No more complications were recorded in the post-operative period, although the convalescence lasted for a time longer than expected, because of anemia. The patient has been continually followed up closely since the surgery. No further surgery was performed. She has been disease-free for two years.

DISCUSSION

Although HABC is a benign lesion, it can behave in a locally aggressive manner, because of its rapid growth and osteolytic capacity.[6] Surgical curettage is the most common form of treatment for this lesion. The recurrence rate after curettage in the jaws ranges from zero to 53%. Many authors attribute the large recurrence rates to incomplete removal during surgery.[7] A problem that might lead to incomplete removal is the massive haemorrhage that may be encountered, which might require ligation of the external carotid artery as a precautionary measure. Although cryotherapy and radiotherapy have supplemented curettage to decrease the recurrence rate, the use of the latter is strongly discouraged as it is likely to induce sarcomatous change in the irradiated bone. The treatment modality most likely to effect a complete cure is en bloc resection, but this is restricted to large and recurrent lesions, owing to the morbidity of the procedure.[8]

Intralesional embolotherapy has been used as a definitive treatment wherein the lesions show complete involution following embolization; or as an adjunct treatment wherein embolotherapy limits blood loss from the lesion when removing it during surgery.[8] This would eliminate the need for multiple blood transfusions and result in a clearer surgical field allowing complete removal of the lesion, reducing recurrence.[9]

Arterial embolization is a technique, initially used preoperatively to decrease vascularity and intraoperative haemorrhage. It has been reported that embolotherapy occludes vascularity of the lesion without interfering with the vascularity of the surrounding tissues, which may lead to involution of the soft tissue component. The response of HABC to embolization has been involution of the soft tissue component, sclerosis and ossification. This mineralization becomes apparent two or more months after embolization.[8] However, there are few references in literature regarding use of this treatment modality for mandibular lesions. Kumar, *et al*.[6] reported that embolization of HABC interesting mandibular bone did allow safe resection in a relatively bloodless field; from which the tumour could be completely excised.

CONCLUSION

Evaluation of intralesional blood flow is important for HABCs because of the hemorrhagic risk in surgery. Embolization seems to be a useful procedure in the treatment of HABCs and could be tried as the treatment modality in the standard protocol for the treatment of HABCs.

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