CASE REPORT

Aneurysmal bone cyst: Case report and literature review

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Abstract

The aneurysmatic bone cysts are uncommon bone injuries, but well-recognized occurring a more frequent in long bones and vertebrae of patients with <30 years. As a distinct entity, it was first described by Lichtenstein, they rarely occur in the maxillary sinuses, which may occur concomitantly with other injuries such as brous dysplasia, and central injury of giant cells. The etiology and pathogenesis of this injury are still not completely elucidated, and there is controversy whether injuries arise anew or represent some form of vascular accident in a preexisting injury. Their clinical characteristics such as fast growth, expansion, cortical destruction and osteoid formation can easily confuse the clinician about malignant injuries. This is a case report of a pediatric patient treated of an aneurysmal bone cyst (ABC) associated with juvenile ossifying broma in Buccal Maxillofacial Surgery of Service of Pedro Ernesto Hospital.

Introduction

The ABC is a bone benign injury, recognized as a solitary clinical-pathological entity distinguished by Jaffé and Lichtenstein.[1,2] The first report in the craniofacial skeleton seems to be Bernier and Baskhar (1958)'s.[3]

The aneurysmal term is used to describe the balloon-shaped distension of part of the affected bone that results in characteristic radiographic appearance frequently viewed.[4] This injury is rare in the mandible, representing around 1% of pseudocysts, being that the mandible is affected twice more than the maxilla.[5] It occurs preferably in young individuals, usually in the second and third decade of life and there are no gender preference.[6-11] In a review carried out by Struthers (1980) APUD Shear (1998) in the analysis of 45 cases, all except three patients were in the first three decades of life, with a peak in the 2nd decade. 29 patients were below 20-year-old and there was a preference for the female gender. The molar region, angle, and branch of the mandible are the most affected areas.[12,13,4]

Shear *et al.*[5] mentions a case where an injury was observed near the orbit's floor and another one near the zygomatic arch. Clinically, it is common the occurrence of an increased volume of firm consistency, sometimes soft, with the evolving time that can range from several weeks to 3 or more years, which increasingly increases piercing through the bone cortical and moving the teeth, which are still healthy. The coverage mucosa remains with clinical aspect of normality. Pain is not a common report. In some cases, when there is bone rupture, and the injury remains covered only by the periosteum or a thin layer of bone, the cyst presents a non-pulsatile crackling without vascular noises. The radiograph presents a uni or multilocular radiolucent injury, cortical bone expansion is described as a balloon-shaped stretching of the affected bone. When multilocular a honeycomb or bubble shapes are observed.[14,15] In some cases, there is cortical destruction and the periosteal reaction can be evident. The teeth can be found displaced, and their roots might have resorption. In the computerized tomography, an image compatible with uni or multilocular cyst

with cortical expansion is observed, with periosteal reaction or the appearance of moth-eaten shape.[16] The diagnosis based only on image features is not required because there are several pathologic identities that can produce the same image, like the ameloblastoma, the myxoma, the central giant cells granuloma, the odontogenic cysts, and the central bone hemangioma.[16]

The pathogenesis of the ABC is controversial, and several theories were postulated to explain it.[4,5] It has been proposed that the trauma, a malformation or a neoplasia could disorganize the local bone microvasculature resulting in an abnormal vascular condition that is the ABC.[10] Although the trauma was suggested as an etiologic factor, there are few evidences to sustain it as such. Several researches have agreed with the perspective that the cyst results from a vascular disturb in the manner of a sudden venous occlusion or arteriovenous detour. The concept that the ABC is a secondary phenomenon and originate from pre-existing bone injury is receiving considerable support, and undoubtedly, there are good evidence to endorse it.[4] Clough and Price (1968) APUD Shear (1998) describe two cases, one of which had histological areas that resembled fibrous dysplasia and the other resembled chondromyxoidal fibroma. The same suggested that the cyst could be either a primary injury or a secondary phenomenon on benign or malign bone injuries.

On their review, Struthers (1980) APUD Shear (1999) it was observed that an associated injury could be identified on 33 from the 303 analyzed cases. Two cases were ossifying fibromas, two cement-ossifying fibromas, four fibrous dysplasias, 24 central granulomas of giant cells and one osteosarcoma. Working with the hypothesis that the ABC is a secondary phenomenon originated from derangement and degeneration of a pre-existing bone injury, Struthers (1980) APUD Shear (1999) studied histologically 303 bone injuries, of variable nature, searching for evidences of early changes that could indicate a potential for the development of an ABC. The same author suggested that the first injury alteration seems to be a microcyst. The central granuloma of giant cells has an enormous tendency to form microcysts, due to their stroma made of loose, edematous and fibrinous connective tissue, which has several blood vessels of thin walls and leaking erythrocytes.[17,18]

Macroscopically during the operation, it is common to note an intact periosteum and a very thin layer of bone covering the cyst. When this is removed, several hidden blood vessels can be seen. The bleeding can be profuse and hard to control until the pseudocyst is removed. The ABC has several variable quantities of soft tissue, composed by crumbly vascular tissue, which subdivides the cavity into several stores filled with blood. No direct communication any vases can be shown during the operation.

Histopathologically, the injuries are composed by several capillaries and spaces with variable size filled with blood, covered by fusiform flattened cells and divided by fibrinous tissue of loose texture. Most of the injuries have small-loosened interwoven multinucleated cells and trabeculae of osteoid/bone tissue. In some firm areas, the stripes of vascular tissue that contains a great number of giant multinucleated cells, fibroblasts, hemorrhage, and hemosiderin are similar of those found in the central injury of giant cells.[19]

The treatment of the ABCs is determined by the nature of some associated injuries. According to El Deeb *et al.*[6] the most frequent treatment in the reported cases of ABC are the curettage, however, Struther and Shear (1983) APUD Shear (1999) refer high recurrence rate and report that the ABC is harder to eradicate compared to the central injury of giant cells. Because of that, some authors recommend a resection of the block as form of treatment.

Case Report

Patient L.E refereed to the Buccal-Maxillo-Facial Surgery Service of Pedro Ernesto University Hospital by the general Surgery Service for the treatment of a mass in the region of the right mandible body and angle [Figure 1]. The refereed injury was byopsed, 6 months before, by another service and the histological diagnostics was of juvenile ossifying fibroma.

At the exam, it was observed an increase of volume on the right mandible, which extended from the parasymphysisal to the mandible branch region, firm to the touch, painless, not associated to lynphadenomegaly, and with approximately 1 year of evolution. At the intra, oral exam [Figure 2] it was verified a small volume increase in the vestibular region of right premolars and molars, firm to the touch, not observing bad occlusion or trismus, but there was paresthesia in the right inferior labial region.

In the radiographic evaluation [Figure 3] - panoramic view of the face - A radiolucent injury was observed, expansive multilocular with well-defined and irregular margins which extended from the region of lower incisors to the region of the right mandibular branch.

A computed tomography scan was requested and three-dimensional reconstruction where it was evident an expansive

Figure 1: Frontal, profile, submentonian pictures, which we can observe the volume increase at the right mandible body and angle region

injury on the right mandible body, angle and branch of approximately 8 cm × 7 cm. For better planning of the event, it was prepared a prototype where the injury could be resected and the reconstruction plate modeled [Figure 4].

The patient was subsequently taken to surgery under general anesthesia, by extra-oral, a low cervical incision

Figure 2: Intra oral pictures showing volume increase at the right vestibular

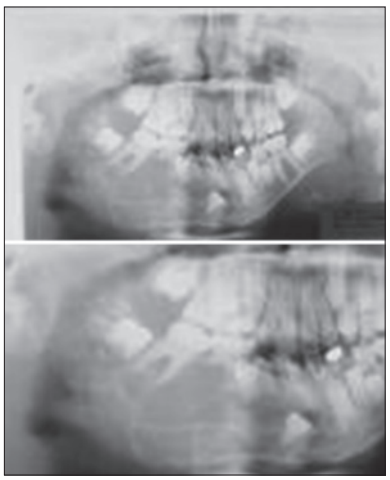

Figure 3: Panoramic radiography showing a radiolucent multilocular injury extending from the right mandible notch to the canines region in the left side

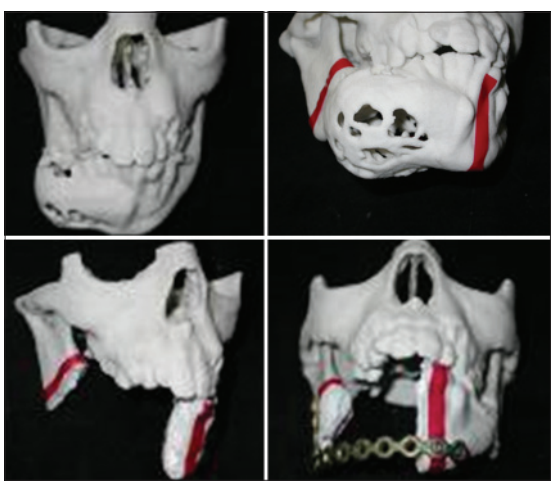

Figure 4: Sterelithographic model where the injury was resected and the reconstruction plate was shaped

was performed and dissection of the anatomic planes until the injury in question [Figure 5], being that during the manipulation it was observed intense bleeding in sheets coming from the same.

Before the resection, the patient was placed in maxillomandibular blockage through skeletal fixation. After resection, the reconstruction plate was installed [Figure 6] and the operative wounds were sutured without intercurrences.

The patient recovered well in the 1st week, but starting from the 2nd week of the post-operative, the state evolved for drainage of purulent secretion from the cervical wound. The same was irrigated, and the stitches were removed, so the healing after this procedure occurred without any new intercurrence. The analysis of the sectioned matter [Figure 7] showed the microscopic diagnosis of ABC associated with juvenile ossifying fibroma.

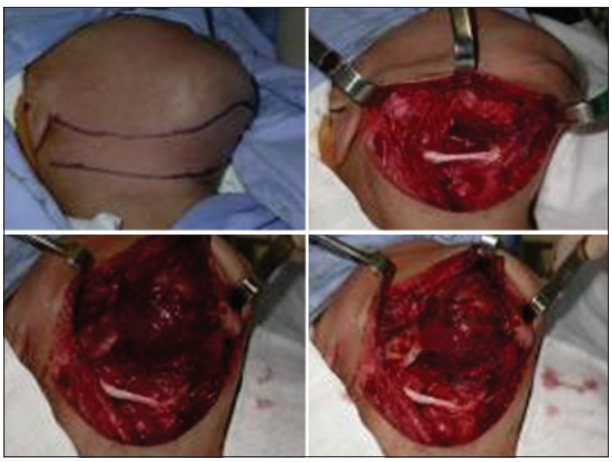

Figure 5: Surgical pictures showing incision and dissection by planes until the mandibular injury

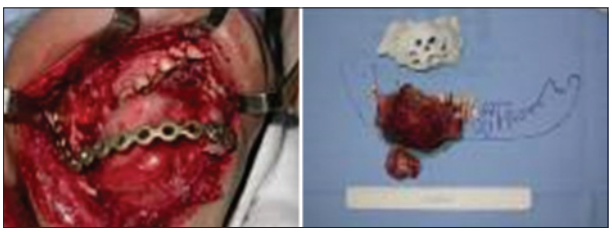

Figure 6: Surgical pictures showing the 2.4 mm plate installed and resected injury compared to the model injury

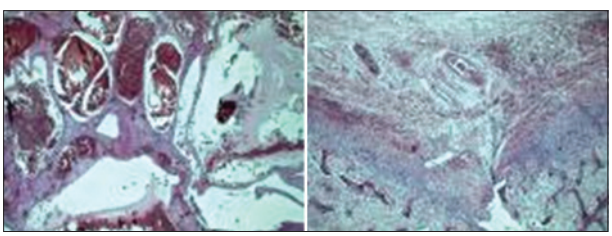

Figure 7: Microscopic aspect of the injury

After a period of 10-month, the patient was sent to the National Institute of Orthopedics-Trauma (NIOT) to be submitted to a new surgical reconstructive procedure. The grafting surgery of a microvascular fibula flap [Figure 8] was performed, and the patient had good post-operative evolution. Currently, he is in follow-up at the NIOT, already being a period of 18-month after the first intervention.

Discussion

In general, the juvenile ossifying fibroma if compared with the ossifying fibroma has more aggressive behavior and presents itself sooner, being more commonly found between the 3rd and 4th decades of life.[20]

The juvenile ossifying fibroma is truly a fibro-osseous neoplasia, being that for which we will complete the diagnosis we should analyze the result of the histopathological examination associated with the age of the patient and his sudden and aggressive clinical presentation. Although they are solid tumors, some studies demonstrated series in which 88% of the injuries developed micro and microscopical cysts. These injuries can also be present as cystic cavities filled with mucoid material and covered by relevant tissue of the juvenile ossifying fibroma. Such cystic spaces are called "areas with aneurysmal cyst appearance," and they contain blood and giant cells. The presence of giant cells is an unspecific finding and the same have, possibly, reactive nature that can be found in numerous osseous injuries including the aneurysmatic bone cysts.[20]

There is a consensus in the literature[3-10] that the ABCs may present themselves associated to other injuries such as non-ossifying fibroma, cement-ossifying fibroma, chondroblastoma, central injury of giant cells, osteosarcoma and fibrous dysplasia. The incidence and possible mechanisms in the pathogenesis of the ABC already were previously reviewed.[5] It is believed that the fundamental step in the formation of an ABC is vascular rupture and hemodynamic change leading to a rapid increase in volume by the fulfillment of the bone cavities with extravasated blood. Beside the preexisting bone injury, trauma is also proposed as the etiological agent for the development of an isolated ABC (not associated to other osseous injuries).[20] In our case, the injury could have developed due to vascular injury and hemodynamic change, because of the negative report about local trauma. Despite this fact is purely of academic importance, because the treatment of ABC associated with another injury should be based in the treatment of primary bone injury, in this case, the juvenile ossifying fibroma.

Under the histopathological point of view, two forms of juvenile ossifying fibroma were previously described: Trabeculae and psammomatoid.[21] Although it is believed to be variants of the same tumor, histopathological patterns were described as separate entities with different characteristics regarding to the demographic data, clinical presentation and places of occurrence.

El-Mofty[21] in their review described the psammomatoid form as the most frequent in naso-orbital complex and the trabeculae form predominantly occurring in maxillaries. The same author also says that the aggressive behavior can be displayed by the two forms of juvenile ossifying fibroma and is more associated with younger patients and the coexistence of an ABC being what we reported in our case.

Radiographically, the injury may present as radiolucent and with well-defined margins, radiopaque or mixed depending on the degree of calcification presented. A radiographic pattern used to distinguish between ossifying fibroma and fibrous dysplasia is the presence of a separation line between the tumor and the healthy bone, in the case of the ossifying fibromas.[4,16,20]

The treatment of juvenile ossifying fibroma remains a controversy. The controversy exists in when indicate an initial resection in opposition to the enucleation and curettage with the premise to treat possible recurrences with additional local excisions. Although the curettage is generally recommended for the initial treatment of juvenile ossifying fibroma and conventional ossifying fibroma, the recurrence rates may reach 56%. In another reported series, of 31 patients, there were 90% of recurrence occurred in children aged <10 years.

It seems there is a consensus in the literature,[5] that there is no absolute histological criterion that anticipates the biological behavior of this injury as well as its recurrence rate, being the surgical resection the treatment that seems to determine a lower recurrence rate.

In summary, the treatment should take into consideration the location, extension and biological behavior of the tumor and the possible existence of an ABC.

The block resection was chosen as a form of treatment for this case considering: The tumor presented aggressive behavior, with rapid growth and dental elements reabsorption. The second motive is that the tumor extension was such that an enucleation with curettage would have been impractical

Figure 8: Trans-operative aspects of reconstructive surgery using microvascular fibular graft

and very unpredictable. The block resection with immediate reconstruction, using a reconstruction plate anatomically modeled from a stereolithographic model, was the most indicated treatment as it is more predictable and offers a good aesthetic result.

Conclusions

After the completion of this work, we can conclude that:

- There is no consensus in the literature regarding the etiopathogenesis of ABC
- Treatment should take into consideration the location, extension and the biological behavior of associated injury.

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