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CASE REPORT

Clavicle aneurysmal bone cyst, case report.

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Abstract

Introduction: Aneurysmal bone cysts are a rare benign neoplasm that appear at an early age. It has a higher incidence in long bones and in the spine. Its etiology is uncertain although it is usually associated with trauma probably due to venous obstruction or the formation of fistulas that occur after contusion.

Case: This report presents the case of a 15-year-old patient with no history of trauma who presents with an aneurysmal bone cyst in the clavicle, which is an unusual location for this pathology.

Evolution: Treatment used sclerosing therapy with 3% polidocanol given eight times with a favorable response. The subject has not required surgery to date.

Conclusion: Sclerosing treatment was successful in this case report.

Key words: Aneurysmal bone cyst; Bone Cysts; Clavicle, Child; Case Report.

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Introduction

Aneurysmal bone cysts (ABC) are a benign neoplasm characterized by the formation of cystic cavities filled with blood and separated by septa of vascular connective tissue [1]. It represents 1% of all primary bone tumors [2] with an incidence of 13 -14 cases / 10 million inhabitants / year [1]. It is typical in childhood and usually affects the long bones mainly the metaphyseal and diaphyseal region [2]. It is also described in the spine in 3–30% of cases [3].

Its etiology is unknown, but it has a genetic basis via the chromosomal translocation t (16; 17) (q22; p13) that modifies the USP6-specific protease gene to produce an extracellular matrix metalloprotease. This enzyme is responsible for the marked bone expansion that can be seen in this pathology [3]. It is also often associated with trauma probably due to venous obstruction or fistula formation. Patients report pain [4] since aggressive local growth and cortical destruction are common in its evolution [5].

Clinical case

We describe a 15-year-old male patient from Esmeraldas-Ecuador.

Prenatal history: This was the fifth pregnancy with normal controls and ultrasound. Subject received iron plus folic acid from the second month, two-dose immunizations, and did not refer to prenatal complications.

Natal history: He was born by cesarean section at 39 weeks of gestation; anthropometry is not remembered but was within normal limits.

Postnatal history: Immediate crying; he was discharged with the mother at 48 hours.

Family history: Father has type II diabetes.

The patient's father reports that his son has presented pain in the right shoulder since age 12; this pain is exacerbated by movement of the joint accompanied by functional limitation. Note the presence of a mass of approximately 1x2 cm on the external clavicular border that has rapidly progressed in size over a period of 7 months (see Fig. 1). Traumatic history is denied.

Physical examination revealed a 6x6-centimeter tumor with regular edges (non-mobile and not painful on palpation) in the external third of the right clavicle that limits adduction, abduction, and rotation movements of the shoulder joint.

Diagnostic workup

The laboratory tests did not show any alteration. A postero-anterior radiograph of the right shoulder was performed in which a well-defined ovoid lesion was observed. It was trabeculated with radiolucent images and located in the distal third of the clavicle measuring 6 by 6 centimeters with a decrease in the cortical bone altering bone structure at this level (see Fig. 2). It was thickened in relation to the rest of the tissue. A right shoulder tomograph was also performed, which reported a lesion in the lateral clavicle compatible with an aneurysmal bone cyst with areas of reinforcement with typical signal characteristics for said diagnosis (see Fig. 3). At least three arterial afferents are visualized: two from the suprascapular artery and one from the axillary artery.

Evolution

Given the vascular origin, the procedure of intracyst administration of 3% polidocanol was performed. This procedure was repeated eight times on an outpatient basis (see Fig. 4), and pain was treated with analgesics (NSAIDs). Subject was discharged and is under review with external consultation. He has not yet required surgery; the size of the tumor has been reduced but there is sclerosis.

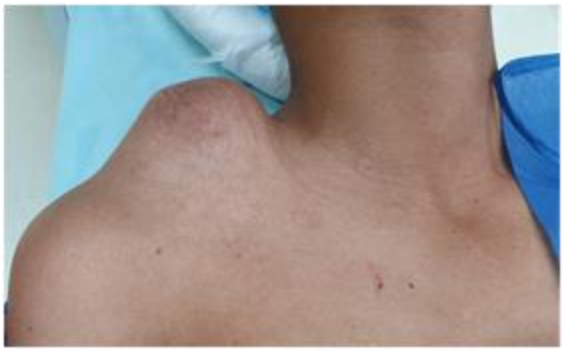

Fig. 1 Photograph of the case.
Note the presence of a mass of approximately 1x2 cm on the external clavicular border that has rapidly pro-gressed in size over a period of 7 months.

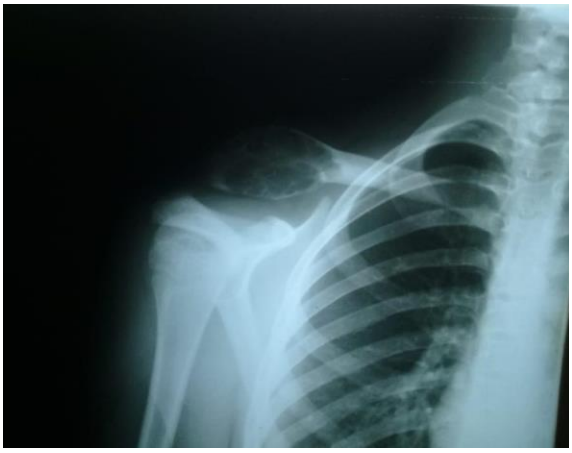

Fig. 2 Postero-anterior radiograph of the right shoulder. A well-defined ovoid lesion was observed.

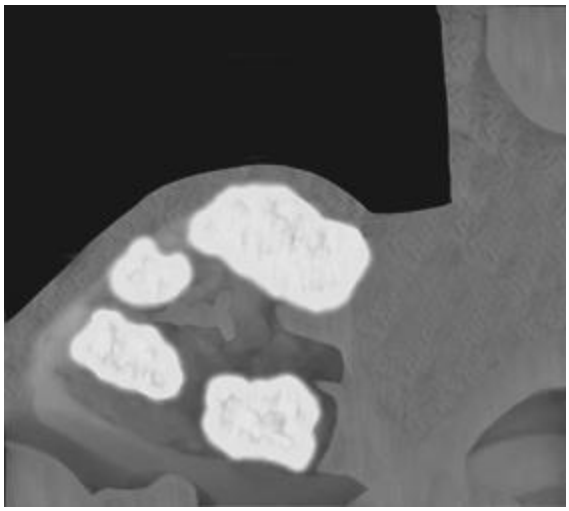

Fig. 3 Right shoulder tomography with the presence of an aneurysmal bone cyst.

There is limited aneurysmal bone cyst. Joint integrity assessments will continue to consider whether there is a need for future surgical intervention.

Discussion

ABC was first described by Jaffe and Lichtenstein in 1942 [1]. Tachdjian defined it as a benign and non-neoplastic proliferative swelling of the bone characterized by the presence of channels and spaces of different sizes surrounded by thin walls that give it an exploding appearance [6]. It occurs more frequently in patients under 20 years of age by 60–80% (rarely before 5 years and after 30 years) [7].

Within its etiology, it can arise de novo and is called primary ABC; however, but there is a high incidence of accompanying tumors in approximately 1/3 of the cases; thus, it is a secondary ABC [5]. The pathogenesis of the aneurysmal bone cyst remains controversial [8], but it is a hyperplastic process secondary to intraosseous hemorrhage leading to cyst formation.

There are theories about hemodynamic alterations in terms of venous insufficiency or arteriovenous malformations in which the cyst fills with blood and erodes the bone walls [9] for which lysis and vascular disorders occur that increase intraosseous pressure and favor growth and rapid expansion [10]. Cysts can lead to repetitive pathologic fractures; occasionally they can cause an asymptomatic union [11]. Cases generally present as a single lesion although very rare cases with multiple locations have been described [4].

This case has a wide and nonspecific variability of clinical characteristics [12]. The most frequent symptom is pain although swelling, deformity, and a palpable mass can also be detected if the location is superficial—it can even start with a pathological fracture.

For its diagnosis, the main exam is X-ray of the affected area where a maturation phase can be distinguished in which the AKA is present. The lytic phase has an eccentric or subperiosteal radiolucent lesion, and the stabilization phase involves the septa and peripheral bone ridge appears to circumscribe the cyst. The healing phase shows progressive ossification and produces a dense bone mass with an irregular structure [4]. Fluid levels can be visualized by computed tomography to assess the limits of the lesion as well as the possible destruction of the cortex. Magnetic resonance imaging showed a multilobed lesion with low signal T1-weighted sequences and high signal content in T2-weighted sequences that identify liquid-liquid levels [4].

Regarding treatment, intralesional injection with sclerosing agents is currently proposed as an alternative to surgery: This strategy is an effective, safe, and minimally invasive treatment [13]. Sometimes it is necessary to repeat the infiltration. Adverse effects include a few cases of a local reaction. When this type of treatment fails, one must resort to surgery either with en bloc excision of the lesion or by curettage of the cavity

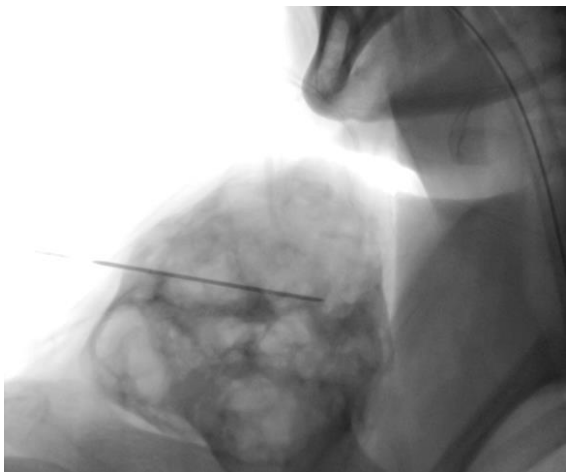

Fig. 4 Intra-cyst administration of 3% polidocanol.

and filling with autologous or heterologous bone grafts [14].

Bone cavity curettage alone has a high recurrence rate that can range from 10% to 59%. Thus, many authors recommend a joint intralesional treatment with the installation of liquid nitrogen, the use of adjuvant cryosurgery, or filling with bone cement. These procedures reduce the recurrence rate from 17% to 26% [7].

In our patient, treatment with 3% Polidocanol was used eight times complying with the common characteristics of sclerotherapy: simplicity, speed, low cost, outpatient nature, and easy administration to generate sclerosis and limit the aneurysmal bone cyst. Joint integrity assessments will continue to consider whether there is a need for future surgical intervention.

Conclusions

Aneurysmal bone cysts are benign tumors that generally respond adequately to treatment with sclerosing agents although they can sometimes evolve unfavorably requiring surgical intervention.

Abbreviations

ABC: Aneurysmal bone cysts. NSAIDs: Non-steroidal anti-inflammatory drugs

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Authors' contributions

DIRR: Research idea, article writing, critical analysis, editorial corrections.
AMPM, Data compilation, Bibliographic review.
RERM, Research Idea, Article Writing, Critical Analysis
All authors read and approved the final version of the manuscript.

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Availability of data and materials

The datasets generated and/or analysed during the current study are not publicly available due participant confidentiality but are available from the corresponding author on reasonable request.

Ethical statements

Protection of persons

The authors declare that the procedures followed were in accordance with the ethical standards of the responsible human experimentation committee and in accordance with the World Medical Association and the Declaration of Helsinki.

Confidentiality of the data

The authors declare that they have followed the protocols of their work center on the publication of patient data.

Consent for publication

The authors have obtained the informed consent from the caretakers of the patient referred to in the article. This document is in the possession of the corresponding author. The parents have signed the authorization for publication of this case.

Competing interests

The authors have no competing interests to declare.

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