Case Report

Aneurysmal bone cyst in the dorsal spine: a case report

Cisto ósseo aneurismático em coluna dorsal: um relato de caso

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ABSTRACT

The present study aims primarily to report a case of aneurysmal bone cyst in the dorsal spine, which was diagnosed, treated, and is currently under follow-up. The secondary objective is to understand how the aneurysmal bone cyst (ABC) can be identified and effectively treated, avoiding its consequences. ABC is a rare benign disease, potentially destructive due to its high osteoclastic activity. It mainly affects young women and occurs primarily in the metaphysis of long bones, the spine, femur, and tibia. This is a case report of a patient diagnosed with ABC. Symptoms are nonspecific but include pain, paresthesia, difficulty in walking, and local edema. Diagnosis begins with computed tomography and/or magnetic resonance imaging, which is definitively confirmed through biopsy and histopathological study. Treatment varies depending on the location of the ABC, its aggressiveness, and the risk of bleeding. In cases with a risk of recurrence or inoperable cases, the use of the monoclonal antibody Denosumab is an excellent alternative. The prognosis is favorable if the diagnosis and treatment are initiated early.

Keywords: Bone Cysts. Denosumab. Spine.


RESUMO

O presente estudo tem o objetivo primário de relatar um caso de cisto ósseo aneurismático em coluna dorsal, que foi diagnosticado, tratado e segue em acompanhamento. O objetivo secundário é compreender como o cisto ósseo aneurismático (COA) pode ser identificado e tratado de forma efetiva, evitando suas consequências. O COA é uma doença benigna rara, potencialmente destrutiva devido a sua alta atividade osteoclástica. Acomete majoritariamente mulheres jovens. Ocorre principalmente na metáfise dos ossos longos, coluna vertebral, fêmur e tibia. Este é um relato de caso de paciente diagnosticado com COA. Os sintomas são inespecíficos, mas incluem: dor, parestesia, dificuldade de deambulação e edema local. O diagnóstico é iniciado através de tomografia computadorizada e/ou ressonância magnética, o que é confirmado, definitivamente, por meio de biopsia e estudo histopatológico. O tratamento é variável e depende da localização do COA, da agressividade e do risco de sangramento. Em casos com risco de recidiva ou casos inoperáveis, o uso do anticorpo monoclonal Denosumab é uma ótima alternativa. O prognóstico se mostra favorável caso o diagnóstico e o tratamento sejam instituídos de maneira precoce.
INTRODUCTION

Aneurysmal bone cyst (ABC) is defined as a rare benign lesion with high destructive potential due to its excessive growth, which can even lead to compression of surrounding structures, characterized by hypervascularization in the bone tissue. Its etiology is still undefined, but the most accepted hypothesis describes it as a hemodynamic disorder associated with primary malformations of the venous system and secondary to bone tissue.

In general, ABC predominantly affects young people, with a significant incidence in women from the age of 20 and is uncommon in the elderly. It represents approximately 91% of all bone tumors and 14% of primary bone tumors. Despite being found in any bone, the locations where this pathology usually presents are: metaphysis of long bones, especially in the posterior elements of the spine, femur, and tibia.

This case report study on ABC was conducted through the analysis of the patient’s medical records, highlighting its rarity. The literature review was carried out through searches on PubMed, Scientific Electronic Library Online (SciELO), and Latin American and Caribbean Health Sciences Literature (LILACS). The study was conducted in accordance with resolutions for research involving human beings of the National Health Council. Privacy and confidentiality of data were ensured at all stages of the study, and this work was approved by the local Ethics and Research Committee with CAAE number 74799423.6.0000.5244.

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CASE REPORT

A 21-year-old white male from Campos dos Goytacazes, Rio de Janeiro State, Brazil, was admitted to the emergency unit with paraparesis associated with paresthesia in both lower limbs and progressively worsening dorsalgia for about 30 days. He used common analgesic medications and did not seek medical help during this period. He denies comorbidities and does not present sphincter changes. Neurological examination: awake, lucid, and oriented with bilateral patellar and Achilles hyperreflexia, muscle strength grade 3 (performs movements against gravity but with-

Figure 1. Magnetic Resonance Imaging of the patient’s thoracic spine. A) Sagittal section of the thoracic spine showing hyperintense lesion in the pedicle region of T9 with spinal canal invasion and metallic artifacts in T8 and T10 compatible with pedicle screws. B) T2 axial sequence with a tumor in the pedicle and transverse process of T9 on the right with canal invasion.
logical fractures are not rare and can occur, related to localized edema and increasing pain\textsuperscript{1,3,6}.

Diagnosis begins with imaging studies. The exams of choice are computed tomography (CT) and/or MRI. In CT, the cyst presents a characteristic image showing a partially cystic area divided by septa. In MRI, the lesion is classically bordered by a complete or incomplete low-intensity signal rim. In T1 with contrast use, ABC acquires a "honeycomb" appearance with polycystic borders showing cortical expansion, cystic spaces, marginal areas with contrast enhancement, internal septations, and diverticulum-like projections from the cyst wall. In T2, the cyst appears as a homogeneous structure with high signal intensity resembling a bubble\textsuperscript{1,2,7}.

Due to the nonspecific clinical and radiological findings, biopsy through surgical procedure is the most indicated path for these patients. Because of an increased bleeding risk, outpatient biopsy through preoperative procedure is not indicated. With the biopsy, the histological type involved in the lesion is confirmed, thus providing definitive diagnosis\textsuperscript{7,8}.

The therapeutic approach varies depending on clinical factors such as lesion topography, aggressiveness, and bleeding risk. Surgical resection of the lesion is currently the treatment of choice for spinal lesions, which alleviates symptoms of radicular and/or spinal cord compression and prevents possible irreversible deformities. Among the techniques used are simple

DISCUSSION

Patients with ABC seek health services primarily due to a nonspecific clinical picture involving symptoms such as pain due to nerve compression and possible paresthesias for the same reason or even the possibility of spinal cord injuries that can lead to difficulty and inability to walk, as in the reported case. Associated patho-

Figure 2. Computed Tomography of the Dorsal Spine reveals lesion and tumor resection. A) Significant resection of the lesion and soft tissues in T9 on the right. B) Bone window with satisfactory tumor resection on the right in D9.
curettage or bone grafting, where in most cases it is necessary to fix the spine to preserve stability. For patients with high bleeding risk, pre-surgical selective arterial embolization is ideal to reduce cyst size and bleeding during surgery and postoperatively\(^1\), \(^2\), \(^9\).

For cases where ABCs are located in hard-to-reach regions, the use of the human monoclonal antibody Denosumab, which binds to the receptor activator of nuclear factor \(\kappa B\) (RANKL), is used due to its ability to inhibit activation, differentiation, and migration of osteoclasts and osteoclast precursors, thus preventing bone resorption. The literature consensus is that administering 120 mg subcutaneously once a month for twelve months is the ideal way to use the drug. The main indications are tumor recurrences in case of incomplete resection and can even be done lifelong in inoperable and disabling ABC cases. Additionally, with prolonged use, adverse effects such as hypocalcemia, dyspnea, musculoskeletal pain, hypophosphatemia, decreased appetite, jaw osteonecrosis, arthralgia, bone pain, myalgia, fever, nausea, diarrhea, constipation, and vomiting are expected. Nevertheless, despite all these adverse effects, the prolonged use of this drug proves beneficial and is not contraindicated\(^2\), \(^9\), \(^10\).

This study concludes that for primary spinal tumors like ABC, imaging exams such as CT and/or MRI with specific characteristics, along with the patient’s clinical presentation, indicate incisional or excisional biopsy for definitive diagnosis and thus better treatment, which is often surgical resection with a safety margin to avoid recurrence. We also conclude that for inoperable cases or with a high chance of recurrence, the use of Denosumab is the best option to control bone destruction.

**REFERENCES**