rcfmc Revista Científica da Faculdade de Medicina de Campos
ISSN 2965-6575

Rev. Cient. Fac. Med. Campos, v. 19, n. 1, p. 83-86, jan./jun. 2024
doi: 10.29184/1980-7813.rcfmc.1000.vol.19.n1.2024
Faculdade de Medicina de Campos
https://revista.fmc.br/ojs/index.php/RCFMC

Case Report

Aneurysmal bone cyst in the dorsal spine: a case report

Cisto ósseo aneurismático em coluna dorsal: um relato de caso

Phatryck Lhoran Pinheiro Ferreira, Karoline de Alvarenga Leandro Lobo, Alison Chagas Ferreira, Lucas de Souza Gomes, Manoella Manhães Monteiro, Ramon Gonçalves Romano Cruz Ribeiro

1 Medical Student, Faculdade de Medicina de Campos (FMC), Campos dos Goytacazes, RJ, Brazil.
2 Professor, Faculdade de Medicina de Campos (FMC), Campos dos Goytacazes, RJ, Brazil

Corresponding Author: Phatryck Lhoran Pinheiro Ferreira
Contact: phatryck007@gmail.com

Keywords:
Bone Cysts.
Denosumab.
Spine.

Palavras-chave:
Cistos Ósseos.
Coluna Vertebral.
Denosumab.

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.

Received on:
10/01/2023

Accepted on:
04/02/2024

Published on:
06/28/2024

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 tíbia. 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 Denosumabe 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[1]. 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[1-5].

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.

This manuscript was translated with the assistance of ChatGPT, an AI language model developed by OpenAI.

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-

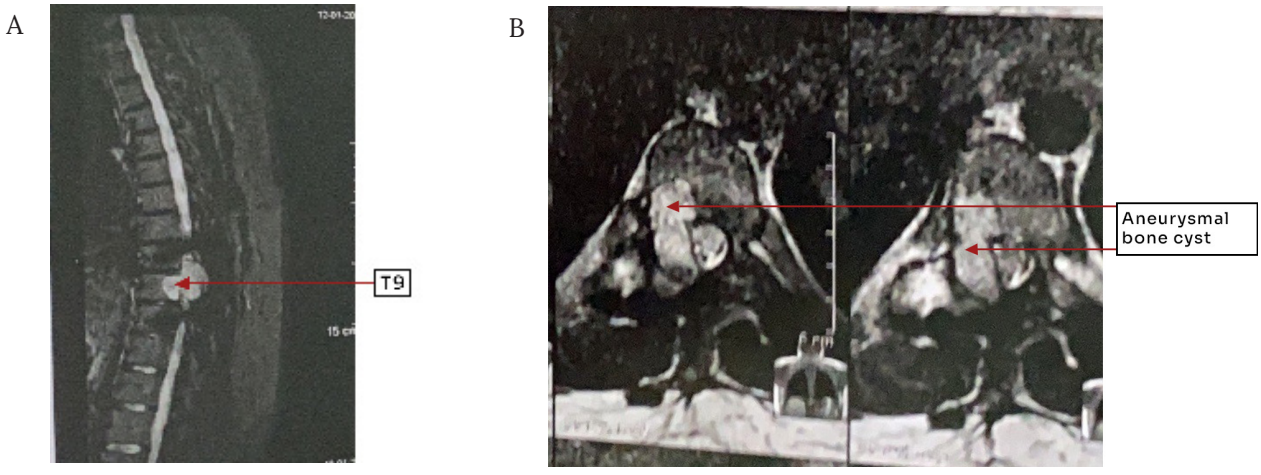

Figure 1. Magnetic Resonance Imaging of the patient's thoracic spine. A) Sagittal section of the thoracic spine showing hyperintense lesion in the pedicle region of T9 with spinal canal invasion and metallic artifacts in T8 and T10 compatible with pedicle screws. B) T2 axial sequence with a tumor in the pedicle and transverse process of T9 on the right with canal invasion.

out additional resistance) in the lower limbs (LL) ASIA C (incomplete spinal cord injury where more than half of the muscles below the affected neurological level have a strength grade less than 3). Magnetic resonance imaging (MRI) of the thoracic spine showed an expansive bone lesion in the right pedicle of T9 with invasion of the spinal canal extending to the posterior elements and posterior part of the body (**Figures 1A and 1B**). Surgical treatment with decompression of the spinal canal and arthrodesis from T8 to T10 was chosen, and subsequent histopathological analysis confirmed the diagnosis of ABC. Later, a new approach for complete lesion resection with partial corpectomy on the right and maintenance of the arthrodesis was indicated (**Figures 2A and 2B**). The patient progressed satisfactorily with partial improvement of muscle strength to grade 4 (performs movements against moderate external resistance and gravity) and complete improvement of paresthesia.

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 pathological fractures are not rare and can occur, related to localized edema and increasing pain[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[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[1, 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

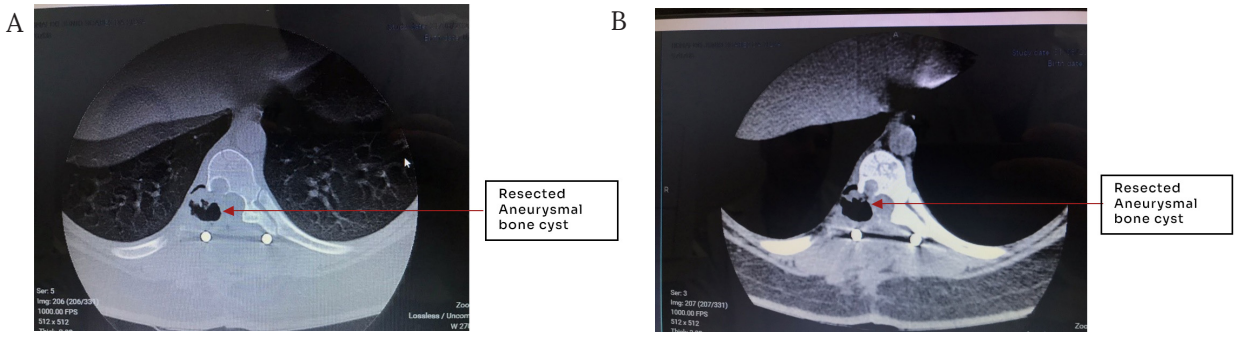

Figure 2. Computed Tomography of the Dorsal Spine reveals lesion and tumor resection. A) Significant resection of the lesion and soft tissues in T9 on the right. B) Bone window with satisfactory tumor resection on the right in D9.

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 kB (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, 9-11].

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

1. Vale BP, Alencar FJ, de Aguiar GB, de Almeida BR. [Vertebral aneurysmatic bone cyst: study of three cases]. Arq Neuropsiquiatr. 2005;63(4):1079-83.
2. Cottalorda J, Bourelle S. Current treatments of primary aneurysmal bone cysts. J Pediatr Orthop B. 2006;15(3):155-67.
3. dos Santos LAM, de Araujo GAE, Teodoro ECM, Alcântara ABA. A importância da biópsia incisional para confirmação diagnóstica de cisto ósseo aneurismático: Relato de caso. Research, Society and Development. 2023;12(8): e18112843051.
4. Gottfried JA, Welter KC, Fonseca MHd, Cezar TEF. Cisto ósseo aneurismático em costela. Rev AMRIGS. 2014;58(2):135-9.
5. Silva EDdOe, Gomes ACA, Raimundo RdC, Carvalho RWFd, Santos TdS. Cisto Ósseo Aneurismático: Relato de Caso e Revisão da Literatura. Rev Cir Traumatol Buco-Maxilo-Fac,. 2007;7(4):9-18.
6. Tierrasecaa MS, Somoza FJM. Quiste óseo aneurismático de localización vertebral. Rev Pediatr Aten Primaria. 2022;24:e157-e61.
7. Dekeuwer P, Odent T, Cadilhac C, Journeau P, Langlais J, Padovani JP, et al. [Aneurysmal bone cyst of the spine in children: a 9-year follow-up of 7 cases and review of the literature]. Rev Chir Orthop Reparatrice Appar Mot. 2003;89(2):97-106.
8. 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(2):108-12.
9. Skubitz KM, Peltola JC, Santos ER, Cheng EY. Response of Aneurysmal Bone Cyst to Denosumab. Spine (Phila Pa 1976). 2015;40(22):E1201-4.
10. Barbanti-Brodano G, Girolami M, Ghermandi R, Terzi S, Gasbarrini A, Bandiera S, et al. Aneurysmal bone cyst of the spine treated by concentrated bone marrow: clinical cases and review of the literature. Eur Spine J. 2017;26(Suppl 1):158-66.
11. Patel RS, Dhamne CA, Gopinathan A, Kumar N, Kumar N. Denosumab: a potential treatment option for aneurysmal bone cyst of the atlas. Eur Spine J. 2018;27(Suppl 3):494-500.