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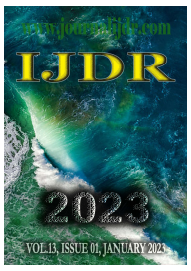

RESEARCH ARTICLE OPEN ACCESS

PARAPLEGIA SECONDARY TO ANEURYSMATIC BONE CYST IN THORACIC VERTEBRA: CASE STUDY

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ABSTRACT

Introduction*:* Aneurysmal bone cyst (ABC) is a benign, osteolytic, and locally aggressive bone lesion, and is difficult to treat, with a recurrence rate of up to 30%. When occurring in the spine it can cause compression of neural structures, with eventual neurological deficit. If relapse occurs after initial unsuccessful treatment, ABC becomes more difficult to treat and may result in significant and irreversible neurological impairment. Therefore, selecting the best treatment quickly is critical. ***Objective****:* to describe the case of a teenager with COA in the spine and severe neurological deficit. ***Methods****:* We report the case of a 17-year-old male who presented spinal cord compression with complete paraplegia of the lower limbs, due to an ABC in the third thoracic vertebra (T3). ***Results****:* He underwent posterior decompression, resection, T1–T5 vertebral fusion, selective embolization, and T3 corpectomy with complementary T2–T4 arthrodesis with anterior approach. The patient was discharged from hospital with improvement in pain and full recovery of sensitivity. ***Conclusion****:* After 1 year, he was walking independently. Despite being a single case, our study may help other surgeons with the proper management of ABC in the spine in patients with severe neurological deficits.

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INTRODUCTION

Aneurysmal bone cyst (ABC) is a benign, osteolytic, and locally aggressive bone lesion.[1] It occurs between 5 and 20 years of age[2,3,5,6] and is difficult to treat, with a recurrence rate of up to 30%.[3,4,6] The vertebral location is atypical (3% to 30%) and rarely symptomatic.[1,2,4] However, lesion growth can cause nerve compression, pain, paresthesias, and paraplegia.[2] There are several treatment options for spinal ABC. Current paradigms prefer surgical resection combined or not with selective arterial embolization (SAE).[4,5] We describe the treatment for a serious complication of an ABC in the thoracic spine.

Case Study: A previously healthy 17-year-old male came to our service in October 2012, with a history of back pain and weakness in the lower limbs for 4 months. On physical examination, he had residual motor function in the lower limbs, preserved sensitivity (Frankel-C), no sphincter impairment, a negative Babinski sign, and patellar and calcaneal hyperreflexia. Radiographs showed T3 destruction reaching the spinous process and pedicles, with vertebral collapse [Figure 1]. Computed tomography showed a T3 lesion involving the body, apophyses, and pedicles, obliteration of the spinal canal, and spinal cord compression [Figure 2].

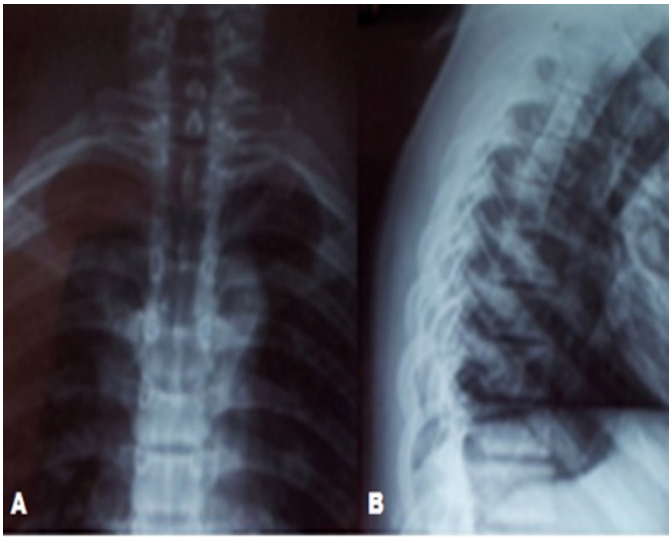

Figure 1. Radiograph of the thoracic column in anteroposterior (a) and lateral (b) views, respectively

It is observed fracture with body destruction of T3, especially extending into the pedicles and posterior elements, and vertebral body collapse with consequent marked thoracic kyphosis.

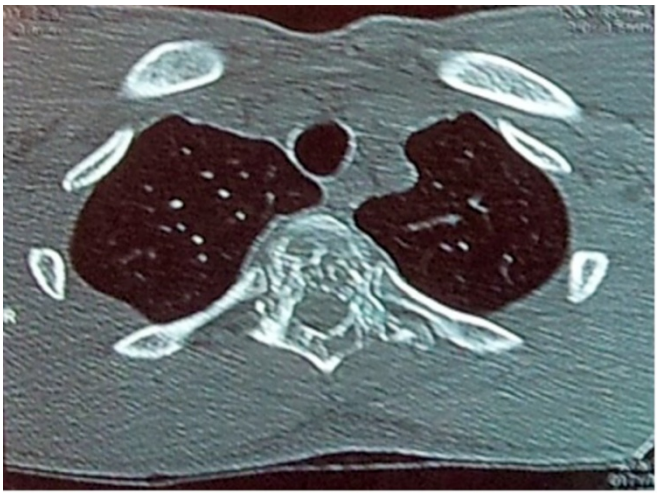

Figure 2. Axial CT of the spine – typical ABC lesion on T3, with expansive, lytic and aggressive characteristics, thin cortical layer involving the vertebral body, apophyses and posterior arch, with posterior epidural extension and projection over the spinal canal

Magnetic resonance imaging revealed T3 fracture and collapse, posterior bulging, kyphotic angulation, reduced canal amplitude and spinal cord compression. Hypersignal on T2 indicated compressive myelopathy [Figure 3A and 3B].

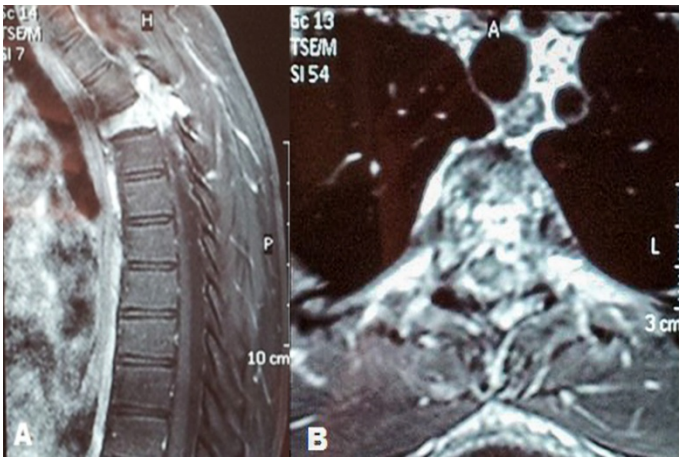

Figure 3. (a) T2-weighted MRI of the sagittal spine – presence of hypersignal suggesting compressive myelopathy, severe kyphotic deformity. (b) T2-weighted axial spine MRI – large, expansive lesion with multiple fluid levels and secondary spinal cord compression

We performed emergency spinal decompression with laminectomy. In the biopsy, bone trabeculae and fibro-connective tissue with inflammatory infiltrate, without atypia, were observed. The histopathological report showed ABC [Figure 4]. Microphotography shows fibroblast proliferation, with osteoclast-like multinucleated giant cells (H&E x40). There was no cytological atypia. A moderate amount of hemorrhage was evident. In November, the patient underwent surgical treatment. At the time, he had complete motor paralysis, residual sensitivity of the lower limbs (Frankel B) and patellar hyperreflexia. We performed decompression with dorsal approach and found T3 bone destruction and exacerbated vascular bed formation [Figure 5]. Resection of the lesion and posterior arthrodesis T1–T5 were performed [Figure 6]. Profuse bleeding made posterior visualization difficult. Pedicle screws were placed between T1 and T5, then we placed vertical rods connecting them. On the 1st postoperative day, the patient developed complete paraplegia of the lower limbs (Frankel A) due to spinal cord compression by the haematoma and intense bleeding in the surgical wound. He was re-operated for haemostasis.

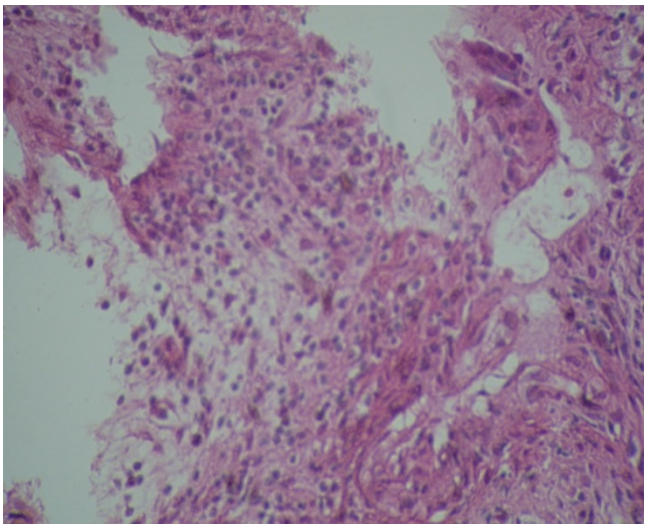

Figure 4. Histopathological examination revealed the cyst cavity filled with hemorrhage and surrounded by bone trabeculae.

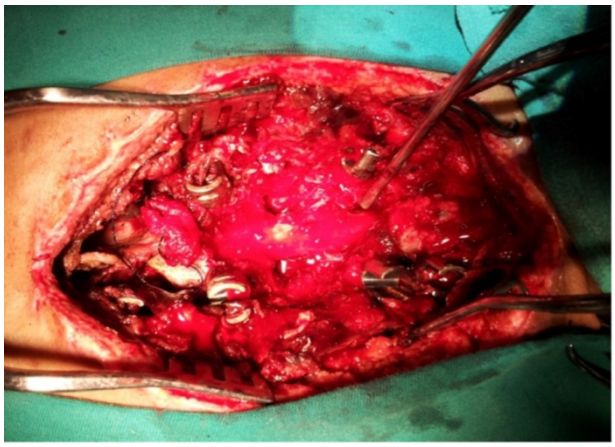

Figure 5. Intraoperative photograph – T3 posterior corpectomy performed at the level of the lesion, exposing the spinal cord.

On the 4th day, arteriography was performed, which revealed a hypervascular tumour nourished by the 3rd left intercostal artery and branches of the deep cervical artery. SAE of these vessels was performed [Figure 7A and 7B]. There was a reduction in bleeding. The following day, thoracotomy was performed with T3 corpectomy, vertebral body replacement with Cage of Harms and complementary arthrodesis T2–T4 with anterior approach [Figure 8].

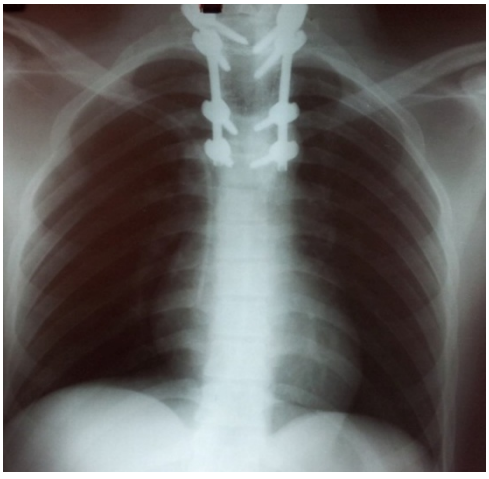

Figure 6. Radiography anteroposterior (AP) of the thoracic column, showing adequate postoperative coronal alignment and good positioning of lateral mass screws after laminectomy, en bloc resection of the T3 lesion and posterior stabilization of T1 to T5 with instrumented fusion

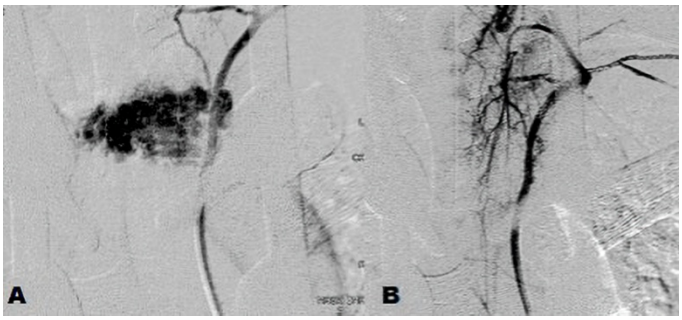

Figure 7. (a) Preoperative embolization, acquisition of panoramic angiography with selective embolization of the 3rd left intercostal artery and branches of the deep cervical artery with contrast enhancement and (b) post-embolization angiographic study, showing absence of opacification of the arteries

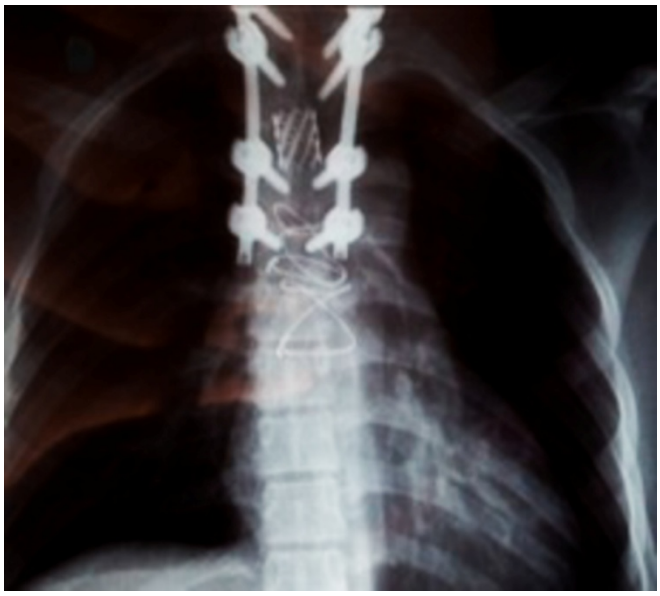

Figure 8. Anteroposterior radiograph of the thoracic spine after the second surgery, showing T3 total corpectomy, vertebral fusion and positioning of the Cage of Harms longitudinally supporting the proximal and distal vertebral plateaus, restoring the anterior column

The patient showed progressive neurological recovery over 10 days. He was discharged from the hospital with improvement in pain, total sensitivity, and partial recovery of the lower limbs, calcaneal and patellar normoreflexia. After 1 year, he presented with progressive physical-motor recovery and was walking independently, with full muscle strength.

DISCUSSION

Due to the proximity to neurovascular structures, spinal ABCs are a therapeutic challenge.[3,4] The neurological damage caused by these lesions ranges from radiculopathies to complete paraplegia.[2,5,6] The therapeutic approach for spinal ABC is a matter of debate. Different factors influence any decision, such as location, size, fracture and/or neurological injury.[1,4,6] If relapse occurs after unsuccessful initial treatment, ABC becomes more difficult to treat and may result in significant neurological impairment. Therefore, selecting the best treatment is critical.[1,4,6] Available therapeutic options include curettage, total resection, radiotherapy, embolization, steroid injection, and a combination of these. Radiotherapy, however, has been associated with growth plate injury, bone marrow necrosis, and sarcomatous degeneration.[3,5,6] The total resection of the ABC presents superior results, with recurrence of 8% to 12%.[3,4,5,6] However, this option can cause great morbidity in the spine and requires experience on the part of the surgeon.[3,4,6] Subtotal resection can be used for lesions close to neurovascular structures, but has high recurrence rates (21% to 50%).[1,5,6,9] In a review of 790 patients Hauschild *et al*.[6] found recurrence after curettage of 12% to 59%, compared to recurrence after total resection + curettage (5.4%); curettage + radiotherapy (6.6%); and embolization + intralesional steroid (26.5%).[6] Non-surgical treatment for ABC includes SAE, intralesional steroids, calcitonin, doxycycline, and *Ethibloc*.[1,3,5,6] However, we believe that treatment in the spine should be aggressive, and the neurological injury requires immediate decompression.[1,3,6] In our case, non-invasive treatment was not indicated, as the patient developed spinal cord compression and, therefore, complete excision of the lesion with surgical fusion was recommended.[1,7,8] SAE should be considered an adjunct to surgery in large and hyper-vascularized tumours, minimizing blood loss.[1,4,5,6] Cruz *et al*.[9] listed 13 studies that used SAE exclusively for primary ABC and found recurrence in 19% of cases, compared to 11% by Addisu *et al.*[8] who performed en bloc excision, surgical fusion and adjuvant SAE.[8]

In another review with 272 patients, Parker *et al*.[5] found the following rates of recurrence: complete excision + SAE showed recurrence of 7.7%; partial resection (35.7%); isolated curettage (25.0%); and curettage + SAE (16.7%).[5] Our case report is unique because we describe a serious and rare complication, a vertebral collapse with thoracic kyphosis and spinal cord compression, which catastrophically progressed to an *"A" lesion* according to the *American Spinal Injury Association*. We obtained an excellent result, by performing spinal cord decompression, en bloc resection, instrumented fusion, and adjuvant SAE. There is a need for further studies to discuss the optimal treatment for the association between spinal ABC and paraplegia. Peyton *et al*.[10] treated a T4 ABC with secondary paraplegia. They performed T4 corpectomy and T1–T7 fusion. The patient made a complete recovery. In addition, they researched spinal ABC-associated spinal cord compression documented over the past 55 years and found only 12 patients with the complication of paraplegia.[10] Despite being a single case, our study may help other surgeons with the proper management of ABC in the spine in patients with severe neurological deficits. The therapy in these cases is controversial, but there is a tendency to choose spinal decompression, total resection with spinal stabilization and preoperative embolization.

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