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*Case Report*

# Thoracic spine aneurysmal bone cyst causing paraplegia in a child: A case report

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## ABSTRACT

A primary aneurysmal bone cyst (ABC) of the spine is a benign lesion with an aggressive nature that can lead to bone destruction and the potential of recurrence after surgical resection. This report focuses on a rare case of the primary ABC in the proximal thoracic spine that is associated with paraplegia in a 7-year-old patient. Nonetheless, the previous research has confirmed that the best therapy for thoracic lesions is challenging and the paucity of evidence-based practice remains an impediment to addressing the issue. The main attribute that leads to these difficulties is the position of the lesions that occur close to the spine and the documented relationship to deformity. Surgical resection decompression can be used alone or in combination with fixation, curettage, selective arterial embolization, and radiotherapy. This report aimed to discuss the clinical, radiological, and therapeutic features of the primary spinal ABC and stress the need for complete surgical excision and decompression if complete spinal cord injury is present. We report this rare case, which was treated surgically with excellent neurological recovery and no recurrence of the ABC at 7 months of follow-up.

**Keywords:** Aneurysmal, Bone cyst, Thoracic spine, Paraplegia, Pediatric, Spinal instrumentation, Decompression

## INTRODUCTION

An aneurysmal bone cyst (ABC) is a benign, reactive, and non-neoplastic lesion that is mostly linked to young demographics. ABC has a reported incidence of 0.14/100,000 individuals. It commonly arises from the long bones, but approximately 12–30% of lesions involve the spine.[1-4] The lumbar spine is the most frequent site, followed by the thoracic spine (34% and 32%, respectively), and only 2% of ABCs occur in the cervical spine.[1,3,4] ABC is a benign tumor formed by blood-filled cavities separated by connective tissue and encircled by a thin cortical bone that may expand. The diagnosis is usually made 3–7 months after the onset of variable symptoms, such as back pain, malaise, fatigue, local tenderness, muscle spasm, and rapid deterioration to complete paraplegia, which may occur if lesions are left untreated.[1,3,5] A spinal ABC generally arises in the posterior element and the pedicle is affected first, which can be expanded and extended into the vertebral body (VB) in 60–70% of cases and causes VB collapse.[3] ABC treatments include surgery, selective arterial embolization (SAE), and radiotherapy, which

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can be used alone or combined with either one of the first two modalities.[4] Due to the proximity of the lesion to the spinal cord and nerve root, finding an optimal treatment can be challenging and is frequently associated with a deformity. Despite positive results being reported, the intricacies of each operation are different. Moreover, there are no clear indications for any of the treatment modalities.[1-3] Due to the rarity of ABC of the thoracic spine that is associated with a complete or near-complete spinal cord injury, only a few cases were reported in the literature and they were treated with different modalities.[6] The report aims to discuss the clinical, radiological, and therapeutic aspects of a primary spinal ABC and report excellent outcomes after complete surgical excision and decompression.

## CASE REPORT

A 7-year-old boy was referred to our institution because of incomplete paraplegia due to a T3 destructive lesion compressing the spinal cord. The symptoms started gradually with back pain for 2 months until he developed a sudden deterioration of neurological status with the lower limb paralysis for 2 days before presenting to our institution. There was no history of trauma, constitutional symptoms, or recent infection. Physical examination at presentation revealed stable vital signs. He had pain with swelling and tenderness over the upper thoracic region with kyphotic deformity. Neurological examination of the upper limbs was normal; however, the lower limbs showed bilateral spasticity, power of zero out of five in all myotomes bilaterally, and decreased sensation of pain and temperature below the level of the T3 dermatome, according to the American Spinal Cord Injury Association (ASIA); this was consistent with ASIA B. Deep tendon reflexes were exaggerated and associated with clonus and a positive Babinski sign. Anal and bladder functions were spared. Laboratory results were within the normal range and included complete blood count, erythrocyte sedimentation rate, C-reactive protein level, and Brucella titer. Computed tomography (CT) and magnetic resonance imaging (MRI) images are shown in [Figures 1 and 2], respectively. Pre-operative biopsy was not performed because the result would take a long time, and we preferred to take the patient to the operative room as soon as possible to minimize delay and give him a better chance for spinal cord recovery. The patient was shifted to the operative room 8 h after the presentation for urgent spinal cord decompression, posterior spinal instrumentation, and complete tumor resection. The patient was placed on the Jackson table, and standard prep and draping were done in an aseptic manner. A posterior spinal midline approach was utilized after determining the appropriate levels using fluoroscopy. The paraspinal muscles were dissected subperiosteally, the lamina of the T3 was completely exposed on each side, and

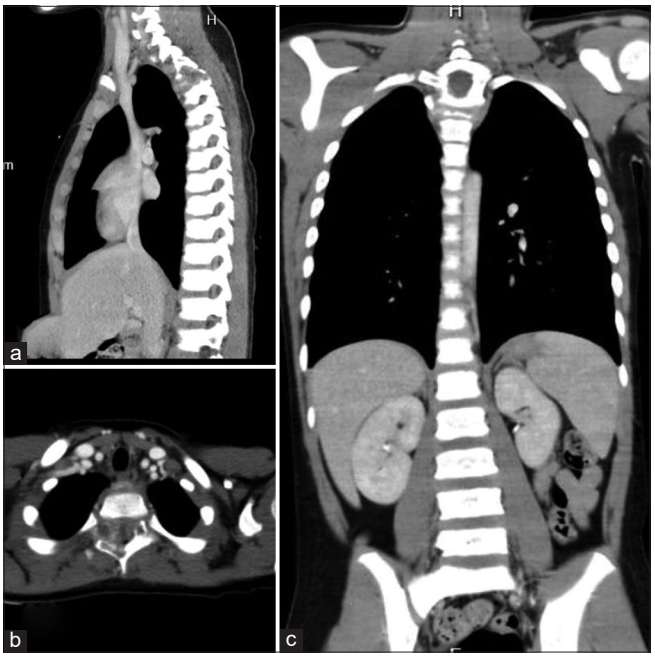


**Figure 1:** Pre-operative computed tomography; (a-c) sagittal, coronal, and axial views of an expansile osteolytic lesion involving the T3 right lamina, pedicle with severe vertebral body collapse, and loss of normal kyphosis.

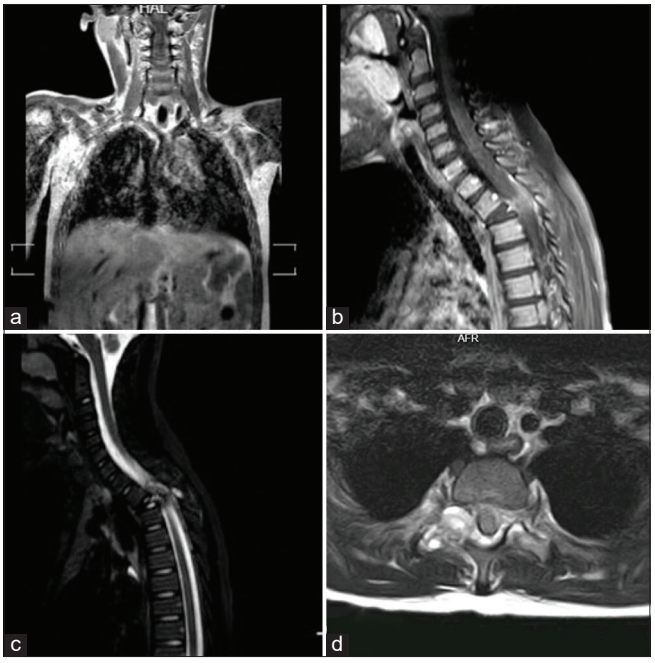


**Figure 2:** Pre-operative magnetic resonance imaging; (a) T2 coronal view, (b) T1 sagittal view, (c) STIR sagittal view, and (d) T2 axial view showing an expansile lesion with high signal intensity involving T3 right lamina, pedicle with a severe collapse of the T3 vertebral body associated with significant spinal cord compression and kyphotic deformity.

we found a grayish rubbery mass that destroyed the right lamina and pedicle with more than 50% of the left lamina affected as well as a severe T3 VB collapse with a very narrow intervertebral space caused by vertebrae above and below. Complete T3 laminectomy and wide mass excision were achieved successfully through the transpedicle approach with posterior spinal instrumentation from T1 to T5 using pedicle screws [Figure 3]. Appropriate screw and rod position was confirmed under fluoroscopy. The excised lesion was sent for histopathology and culture, which confirmed the ABC diagnosis with no evidence of malignancy. After that, the patient was followed up daily until he was discharged to the rehabilitation center with no change in the lower limb power and sensation. Anterior support was planned as a second stage, and the parents were informed about it if instability was encountered during follow-up. The patient was followed in the clinic regularly for neurological assessment, instability, and recurrence. At the 2-month follow-up, radiographs and MRI were ordered that showed no recurrence of ABC or instability, with significant improvement at the level of the motor function and sensation [Figures 3 and 4]. At the 7-month follow-up, the patient showed full recovery in terms of sensation as well as full power recovery with normal walking, which was consistent with ASIA E, without signs of instability [Figure 5].

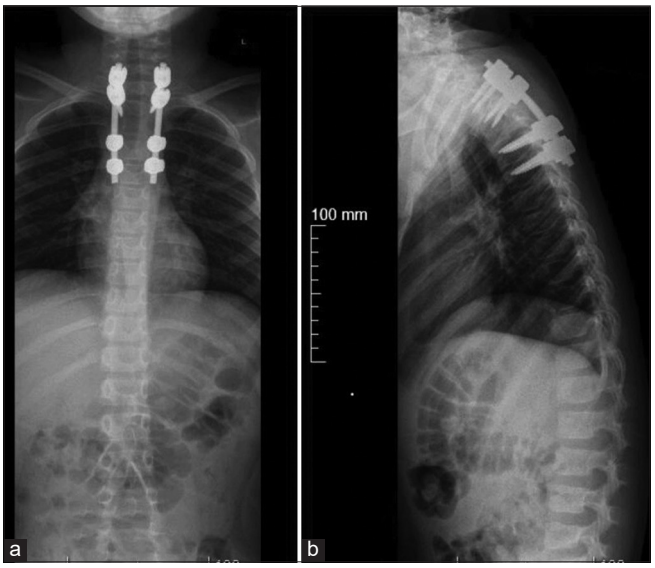


**Figure 3:** Immediate post-operative radiograph; (a) anteroposterior and (b) lateral radiographs showing posterior spinal instrumentation from T1 to T5 with excellent coronal and sagittal alignment.

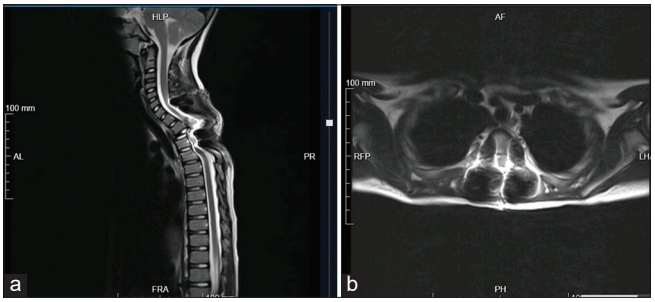


**Figure 4:** Two-month post-operative magnetic resonance imaging: (a) T2 sagittal and (b) T2 axial views showing sagittal alignment maintained without aneurysmal bone cyst mass recurrence.

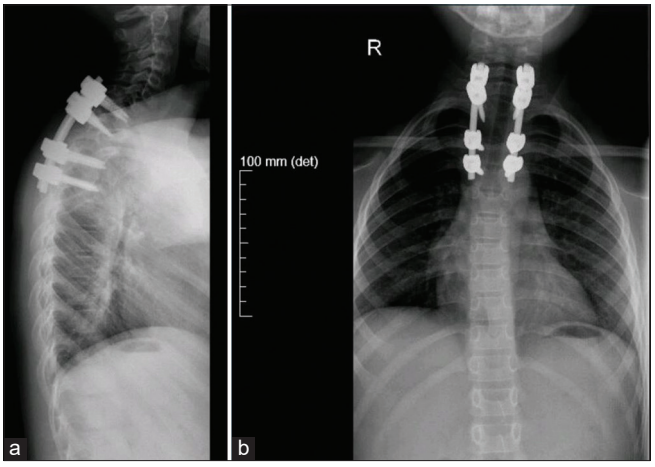


**Figure 5:** Seven-month post-operative radiographs: (a) Lateral and (b) anteroposterior view showing posterior spinal instrumentation maintained from T1 to T5 without signs of instability or sagittal and coronal malalignment.

## DISCUSSION

The specific origin of ABCs remains uncertain. Nonetheless, despite being benign, the current discussions on the potential neoplasticity of such tumors are gaining attention. Research has shown that cytogenetically, a significant number of lesions occur alongside the development of a translocation chromosome that has the projected attributes of a neoplasm.[4] Indeed, such developments help in understanding the condition, especially in young patients, such as that presented in the current report. This novel phenomenon may provide new prompts necessary for understanding the nature of tumors in patients with ABCs to identify those suffering from a benign or malignant neoplasm. The lower back pain in pediatric patients varies from 10% to 33% of the total demographic, suggesting that vigilant action should be taken to prevent exacerbation.[3,5] Atypical back pain accompanied by postural change, fatigue, and weakness creates the basis for paraplegia occurring due to the extensive destruction of the spine caused by ABC lesions.[6,7] Back pain is a common presentation that is usually associated with a tender palpable mass.[1,3] An ABC of the spine can be associated with an epidural compression arising from VB collapse, kyphotic deformity, and instability, which may result in myelopathy or acute paresis.[3] Despite ABC being first reported in the 19th century, little insight has been gained on the prognosis of ABC with paraplegia.[6,7]

Venous obstruction in the bone can cause a reduction in venous outflow, which contributes to the accumulation of interstitial fluid, leading to a cyst formation.[6] However, recent discussions have provided varied conclusions regarding the nature of ABCs and the different approaches to treatment. The dominant conventional treatment for such conditions includes complete surgical removal of part of the whole lesion.[6-9] The difference in therapy depends on the size and position of the lesions, along with the potential for recurrence. Back *et al.* reported two cases of thoracic ABC similar to our previously discarded case: The first case had ASIA B and the second case had ASIA A, both of which were treated with tumor excision, posterior spinal instrumentation, and complete decompression. They achieved complete neurological recovery at 6–9 months and at the final follow-up.[7] Boriani *et al.* reported that serial embolization of cysts could guarantee favorable results in such cases. Therefore, this case study expounds on the occurrence of ABCs and the potential avenues for addressing such conditions when they present with paraplegia. Furthermore, intralesional injections have been recorded as a convenient and effective option for using calcitonin to address the ABCs. Recovery after such procedures is likely to be better when the entire lesion is removed than when a part of the condition is addressed. Therefore, the outcome of any treatment approach should be focused on removing the ABC lesion while ensuring recovery from paraplegia. ABCs may occur as solitary lesions or in association with other tumors, such as giant cell tumors, osteoblastomas, chondroblastomas, and fibrous dysplasia.[5] Although a diagnosis of ABC is only possible by biopsy, other critical procedures, such as pre-operative radiological evaluation with MRI and CT, can be useful in the diagnostic process.[2] The optimum management of thoracic lesions is still unclear due to the different approaches that have been developed based on the location of the lesion on the spine. Boriani *et al.* reported that using an SAE has not been linked to negative patient outcomes, but full recovery is observed in 88% of spinal ABCs requiring at least two embolization procedures.[4,9] Therefore, we agree with the suggestion of Eun *et al.* for the conventional use of SAE during the pre-operative phase to reduce the potential for heavy bleeding during the process.[10] However, the function of radiotherapy in the overall treatment is still controversial due to potential risks such as malignancy, growth resistance, and post-radiotherapy myelopathy. Moreover, significant research does not recommend using the latter approach during the preliminary stages, particularly when dealing with neonates and children.[4,9,11,12] However, surgical resection is frequently considered the treatment of choice. Total excision of the lesion, including the entire cyst wall and spongy tissue, is critical to obviate the chance of recurrence, which is related to the degree of excision.[7,12,13] Finally, managing such cases requires thorough examinations and investigations for planning non-operative and operative management to avoid any potential complications, such as paraplegia, deformity, and recurrence.

## CONCLUSION

An ABC is a rare entity that can be associated with severe VB collapse and spinal cord injury. Complete neurological recovery can be achieved after surgical decompression, even in severe cases. Total tumor excision is mandatory to reduce the risk of recurrence. Finally, meticulous pre-operative planning and regular follow-up are very important to avoid complications such as recurrence and instability.

## AUTHORS' CONTRIBUTIONS

OA involved in writing, designing the report, and reviewing the literature; AA concept, draft, and revising; AFB concept, reviewing the literature, drafting, and revising; and MB final reversion. All authors have critically reviewed and approved the final draft and are responsible for the manuscript's content and similarity index.

## DECLARATION OF THE PATIENT CONSENT

The author certifies that they have obtained all appropriate patient consent forms. In the form, the patient's parent has given his consent for the patient's images and other clinical information to be reported in the journal. The parent understands that the patient's name and initials will not be published, and due efforts will be made to conceal his identity, but anonymity cannot be guaranteed.

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## CONFLICTS OF INTEREST

There are no conflicts of interest.

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