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Case Report

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Spinal cord compression with acute para-paresis due to thoracic aneurysmal bone cyst (ABC): a case report and review of the literature

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Abstract

Aneurysmal bone cysts (ABCs) are benign, highly vascular osseous lesions characterized by cystic, blood-filled spaces surrounded by thin perimeters of expanded bone. Children and young adults are most often affected by spinal ABCs; more than 75% of patients are younger than 20 years old at the time of presentation. Although ABCs have been documented in all areas of the axial and appendicular skeleton, ABCs of the spine present unique challenges due to the risk of vertebral destabilization, pathological fracture, and vertebral body (VB) collapse with neurological compromise.

We describe here a case of a 12-year-old child who presented with cervical pain and gait disturbances starting a few weeks prior to his admission with acute paraparesis at the time of admission. Subsequently he was found to have a Thoracic ABC involving T1-T3.This was accompanied by T2 vertebral body collapse and spinal cord compression. He was investigated and treated promptly by resection of the aneurysmal bone cyst with posterior Cervical-thoracic instrumentation. There was full post-operative neurological resolution.

Keywords: Aneurysmal Bone Cyst, Paraparesis, Spinal Cord Compression.

Introduction

Aneurysmal bone cysts (ABC) are benign but maintain aggressive local invasive behavior. No precise pathophysiology has been identified, although it is considered to be a vascular malformation within the bone due to an identified fusion oncogene [1,2]. ABC are uncommon with an annual incidence between 1.4-3.2 cases per million people with a preference for long bones thus accounting for about 1.5% of primary bone tumors [3-5]. However, 10-30% of ABC affects the spine and contribute to approximately 15% of primary spinal bone tumors [3-5]. The most common proposed theory for ABC development is de-novo mutations in the absence of other bone pathologies. This theory constitutes 70% of ABC cases [6-8]. Other theories include trauma and secondary reactions due to other bony pathologies such as fibrous dysplasia, osteoblastoma, chondroblastoma, chondromyxoid fibroma or non-ossifying fibroma [6-8]. Studies have shown that ABC has a female predominance, particularly in those younger than 20 years of age, and a predilection for the lumbar and cervical spine [9-11]. ABC requires a multidisciplinary management team. There are several different treatment options available for spinal ABCs. However, surgical resection, selective arterial embolization (SAE), or a combination of the two techniques is the gold standard for treatment. There is no reported superiority of any treatment option when considering local recurrence risk or associated morbidities [12,13].

Case Presentation

Here we present a case of 12-year-old boy who was complaining

of a posterior cervical mass and local pain for few months prior to his admission. His outpatient evaluation included a soft tissue ultrasound (US) which showed hypertrophied soft tissue without any significant findings and a plastic surgery consultation. Due to his complaint of a new onset limp, radiographic lower limb imaging was performed without any skeletal findings.

A few days prior to his referral, an acute afebrile non-traumatic spastic paraparesis appeared, without sphincter involvement. Neurological examination in the emergency room revealed tender and restricted neck flexion, bilateral spastic lower limb weakness, diffuse hyperreflexia, bilateral Babinski and T4 sensory level.

He emergently underwent a total axis magnetic resonance imaging (MRI) scan. The MRI revealed normal brain anatomy, a large posterior vertebral mass with bone destruction, remodeling, and a fluid-fluid level within the lesion at the T1-T3 spinal level, vertebral collapse at T2 with significant anterior T1 translation and spinal cord compression (Figures 1A and 1B). These findings were highly suggestive for vertebral ABC. Although spinal angiography confirmed ABC, selective arterial embolization (SAE) could not be performed. For surgical planning and post-surgical follow up purposes, total axis computerized tomography (CT) scan was performed (Figures 2A and 2B). The patient underwent a T1-T2 ABC resection with C5-T5 posterior vertebral fixation under continuous intraoperative neurophysiological monitoring. During the operation, the patient required a blood transfusion and Tranexamic acid due to massive bleeding. Post-operative CT scan showed satisfactory spinal column alignment and spinal canal decompression. Histopathological examination confirmed ABC showing a cystic formation with blood hypercellular areas and osteoclast-like giant cells (Figure 3). The post-operative course was uneventful with significant neurologic improvement and full neurologic recovery at one-year follow-up and no recurrence at four years. Post-operative radiological follow up were not significant (Figure 4).

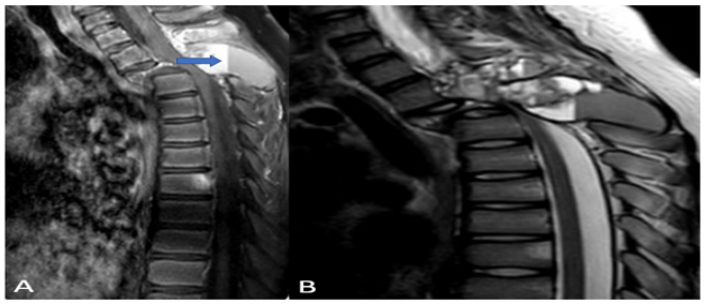

Figure 1: MRI showing large posterior vertebral mass characterized by fluid-fluid level (arrow) at spinal level T1-T3, vertebral collapse at T2 and spinal cord compression; (A) sagittal T1-weighted contrast-enhanced MRI, (B) sagittal T2-weighted MRI.

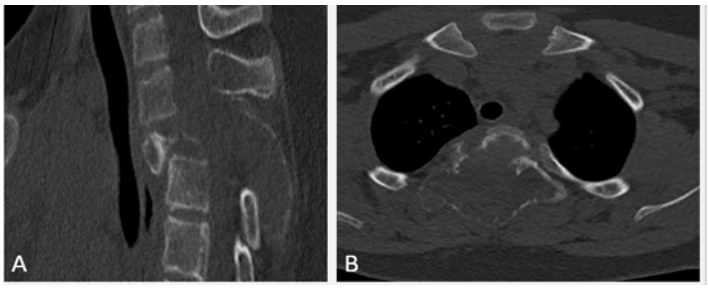

Figure 2: Preoperative CT scan Showing the lesion (A) sagittal CT of the spine (bone window), (B) axial CT scan (bone window).

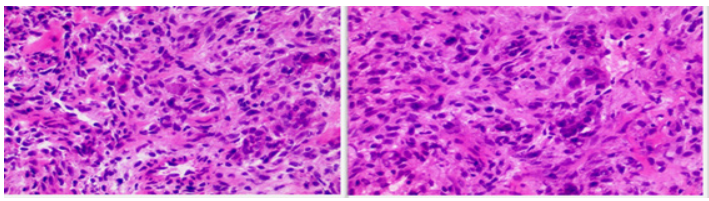

Figure 3: H&E stain showing cystic formation with blood hypercellular areas with osteoclast like Gaint cells with new bone formation bone formation confirming ABC.

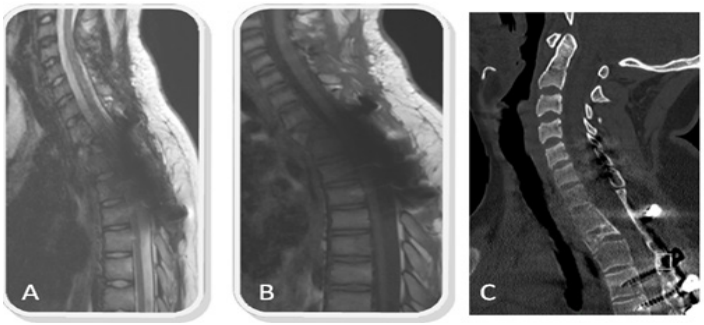

Figure 4: Post-operative radiographic findings (A) sagittal T2-weighted MRI, (B) sagittal T1-weighted MRI, and (C) sagittal spinal CT (bone window).

Discussion and Conclusion

This case illustrates that although an ABC is a benign vascular bone lesion, it can be a highly disabling disease with significant neurological sequelae that mandates urgent intervention [9,10]. Multidisciplinary management is crucial for proper diagnosis, treatment and follow up [13]. Endovascular interventions, such as selective arterial embolization (SAE), can be very useful as both diagnostic and therapeutic tools. Intraoperative massive bleeding is an issue that the treating physicians should be prepared for by many means such as, preoperative SAE and intraoperative hemodynamic and coagulation studies [12]. Here, we present a case of a rare vascular vertebral lesion-ABC with acute spinal cord compression and subsequent paraparesis. Early diagnosis

and prompt definitive treatment are mandatory for successful neurologic recovery. Treatment strategy mandate preoperative multidisciplinary approach with proper planning, aided in risks reduction such as bleeding, neurological compromise due to unintended spinal cord or spinal nerves injury and proper baseline for post-operative follow up. Post-operative intense and prolonged neurological rehabilitation and follow up are crucial as they permit early and satisfying neurological outcomes.

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Conflicts of Interest

All the authors have no conflicts of interest to declare.

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Author Contribution

Conception and design: FA, MG, MI.
Manuscript writing: All authors.
Final approval of manuscript: All authors.

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