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Solid Variant of an Aneurysmal Bone Cyst of the Thoracic Spine

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Disclosures can be found in Additional Information at the end of the article

Abstract

The solid variant of an aneurysmal bone cyst (ABC) has been observed very rarely, especially those involving the spine. In this case report, we present a very unusual tumour of the thoracic spine which was managed by 360° decompression via posterior-only approach and stabilization.

A 16-year-old boy presented to us with a sudden onset of weakness in both lower limbs leading to paraplegia. He also had a history of back and chest pain over the past one year. A collapse of the T5 vertebrae on plain radiograph was observed. The patient was immediately shifted to the operation theatre with an initial plan of a total en bloc spondylectomy of the T5. However, intraoperatively, histology favored a solid-ABC variant rather than a spindle cell tumour or giant cell tumour. Thus, the initial plan was revised to a 360° decompression without resecting the body en bloc via a posterolateral approach.

After surgery, complete resolution of his sensory and motor dysfunction was achieved. His chest and back pain also resolved. The diseased vertebral body gradually healed and new bone formation was seen at 18 months postoperatively.

This case report concludes that a solid variant of an ABC should be considered as a differential diagnosis for tumours involving the spine. An intraoperative frozen section procedure should be undertaken, especially during emergency situations. Early diagnosis and appropriate surgical management play an important role in the successful management of a solid variant of ABC.

Categories: Internal Medicine, Pathology, Orthopedics

Keywords: abc, osteolytic lesion, bone tumor

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Introduction

The solid variant of an aneurysmal bone cyst (ABC) is a very rare case which generally accounts for about 3.4% to 7.5% of all ABCs [1]. It is a cystic lesion that can occur and expand in any part of the bone, often affecting individuals in the second decade of their lives [2]. A definitive diagnosis is impossible but there are radiographic characteristics that are highly suggestive of ABCs on computed tomography (CT) and magnetic resonance imaging (MRI) scans.

Jaffe and Lichtenstein first described ABC in 1942, when they noted, "a peculiar blood-containing cyst of large size" [3]. As many as 69% of primary ABCs demonstrate a characteristic clonal t(16;17) genetic translocation which can lead to an upregulation of the TRE17/USP6 oncogene [4].

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Although benign, an ABC has the potential to grow acutely. Its expansile nature can cause a lot of swelling and pain while disrupting the growth plates. The changes in the bone can also lead to deformity and pathological fractures. It can also cause some neurological manifestations depending upon its location [2]. A distinct solid variant of ABC was first reported by Sanerkin, et al. in 1983 [5]. This type of solid variant could be easily misdiagnosed as a spindle cell tumour, especially osteosarcoma [1].

Symptomatic ABCs are generally treated with surgery. However, asymptomatic ABCs (characterized by clinically insignificant destruction of the bone) are generally left alone with just close monitoring for any abnormal changes [6].

Case Presentation

A 16-year-old boy was presented to us with a sudden onset of weakness in both lower limbs leading to paraplegia. He also had a history of back and chest pain over the last one year. A good rectal tone without any perineal anesthesia was noted in the rectal examination and the post-void residual urine volume was negligible. All laboratory findings were within normal range. Plain x-ray showed the collapse of the T5 vertebra (Figure 1).

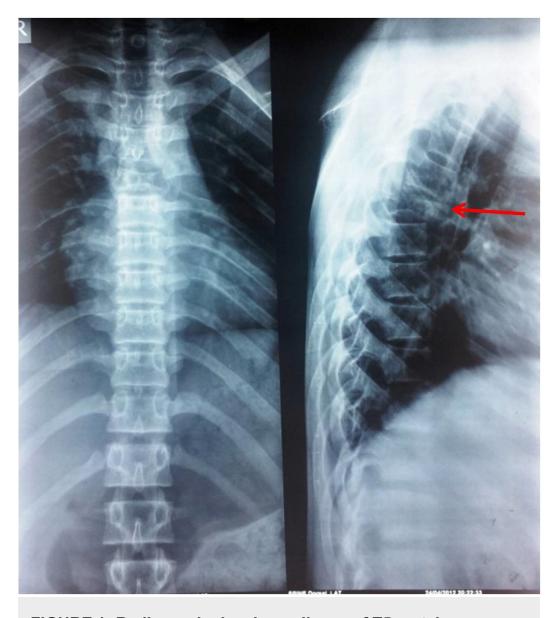
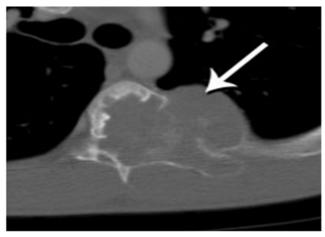


FIGURE 1: Radiograph showing collapse of T5 vertebra

Anteroposterior and lateral view x-ray of the thoracic spine showing pathological fracture and collapse of T5.

CT axial images showed an expansile and lytic lesion in the vertebral body, left pedicle, and transverse process. T2-weighted sagittal and axial MRI T2-weighted images of the thoracic spine showed hyperintense signals in the T5 vertebral body, left pedicle, and transverse process with a pathological fracture (Figure 2).





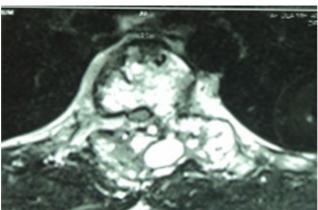


FIGURE 2: CT (top left) and MRI (bottom left and right)

Computed tomography (CT), magnetic resonance imaging (MRI).

A preoperative biopsy was not successful because of the urgent requirement of decompression. The patient was taken to the operating room on an emergency basis with an initial plan of a total en bloc spondylectomy of the T5.

Intraoperatively, histology favored a solid-ABC variant rather than a giant cell tumour. Thus, the initial plan was revised to palliative surgery with a 360° decompression (without resecting the body) en bloc via a posterolateral approach as shown in Figure 3. The vertebral column was reconstructed with an expandable titanium cage (inserted via a costotransversectomy approach) and pedicle screw fixation in a 360° manner (Figure 3).

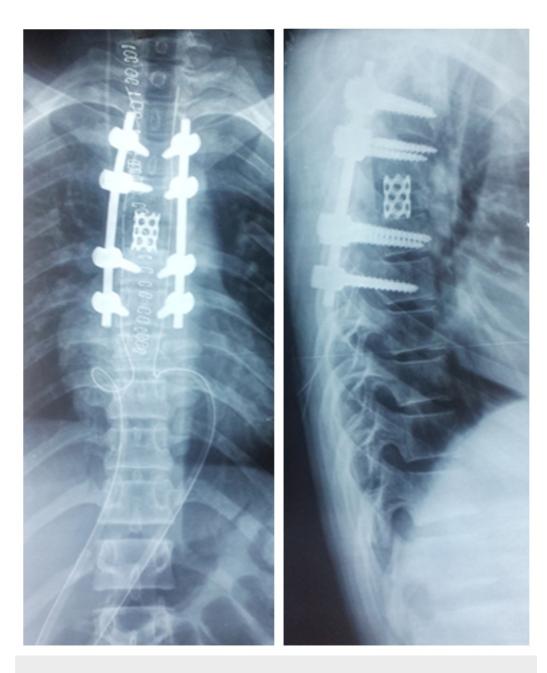


FIGURE 3: Intraoperative anteroposterior and lateral view x-ray of the thoracic spine showing cage and pedicle screw fixation

Results

After surgery, a complete resolution of sensory and motor functions was achieved. The pain in his chest and back also resolved gradually within a few days. Also, the diseased vertebral body gradually healed and new bone formation was seen at 18 months postoperatively (Figure 4).

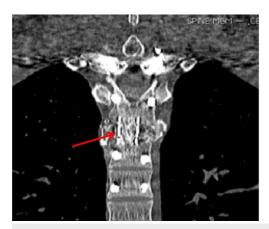




FIGURE 4: Postoperative MRI showing new bone formation at 18 months

Magnetic resonance imaging (MRI).

Discussion

ABCs, although benign, are locally very aggressive and highly vascularised tumours. Generally, post-surgery, it takes a long time to obtain a healing state and new bone formation with a recurrence rate of about 50% [7]. However, Malghem, et al. concluded that spontaneous healing is possible [8].

ABCs are found to have a predilection for the lumbar spine, which was reported by Boriani, et al. [9]. Although CT and MRI scans are the preferred diagnostic methods, a biopsy is the utmost necessary prerequisite for confirming the diagnosis, due to its similarity in appearance with many other bone lesions. The biopsy generally shows proliferating round or oval cells, generally mixed with multinucleated giant cells distributed randomly. It also contains regions of reactive fibroblastic proliferation and microcystic components filled with blood [10].

Depending on the proliferative component, the solid variant of an ABC is often misdiagnosed histologically for other benign or malignant tumour-like bone lesions [1]. The pathological differential diagnosis (solitary bone cyst, giant cell tumour, hemangioma, osteosarcoma, and chondroblastoma) should always be kept in mind while thinking of ABC [10].

The treatment of ABC is also very controversial which includes arterial embolization, curettage with or without bone grafting, complete excision, intralesional drug injections (steroid and calcitonin), and radiation. However, early diagnosis and appropriate surgery play the most important role in the successful management of ABC [10].

Whether surgical management results in a better outcome and lower recurrence rate than other methods (conservative or palliative; for example, curettage alone) remains controversial. Hence, more studies comparing these methods should be conducted.

Conclusions

This case report concludes that an ABC should be kept as a differential diagnosis for tumours involving the spine. Intraoperative frozen sections should always be performed when there is a doubt in mind and in cases of emergencies. An effective spinal decompression and stabilization of the ABC can be achieved by partial or subtotal excision. However, for the successful

management of ABC, early diagnosis and appropriate surgical management should be considered.

Additional Information

Disclosures

Human subjects: Consent was obtained by all participants in this study. Conflicts of interest: In compliance with the ICMJE uniform disclosure form, all authors declare the following: Payment/services info: All authors have declared that no financial support was received from any organization for the submitted work. Financial relationships: All authors have declared that they have no financial relationships at present or within the previous three years with any organizations that might have an interest in the submitted work. Other relationships: All authors have declared that there are no other relationships or activities that could appear to have influenced the submitted work.

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