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Solid Variant of Aneurysmal Bone Cyst Involving the Thoracic Spine: A Case Report

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ABSTRACT

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14-year-old male was first seen after several months of upper back pain and increasing weakness in lower limbs. MRI showed T2 hyper-intense lesion with fluid filled levels in T-10 vertebrae suggestive of aneurysmal bone cyst. Pre-operative radiotherapy was given to decrease the tumour mass and vascularity. Intraoperatively, the tumour was solid in consistency; Local clearance, bone grafting and fixation was done. Histological examination showed predominance of fibroblasts suggestive of a solid variant. Complete neurological recovery was seen at 1 month and no recurrence was noted at final follow-up.

Structured

Introduction

Aneurysmal bone cyst is an uncommon tumor involving the spine. However, the solid variant is comparatively even rarer and we could find only 15 cases involving the spine to date, majority in thoracic spine. They are difficult to diagnose radiologically.

Case report

We assessed a 14-year-old male patient with history of upper back pain for several months and increasing lower limb weakness since 1 month. An MRI showed T-2 hyperintense multi-cystic lesion in T10 vertebra s/o aneurysmal bone cyst. The spinal cord was significantly compressed without any evidence of myelomalacic changes. We planned to give radiation therapy pre-operatively to decrease vascularity and tumour mass. There was significant reduction in vascularity, however, cord compression was not relieved, so surgical decompression was planned through posterior approach. Intra-operatively suspected tissue was removed and cord was freed from all adhesions. The vertebral column was reconstructed with bone graft taken from PSIS and pedicle screw fixation. Histologically, the specimen showed predominance of solid fibroblasts, with abundance of osteoclastic-like giant cells, and scattered small cyst-like areas filled with erythrocytes and focal hemorrhage, consistent with a predominantly solid variant of aneurysmal bone cyst. There was complete neurological recovery at 1 month post-operatively.

Conclusions

Considering the rare nature of the lesion in this location and unclear treatment protocol, we considered this case important for reporting; as the solid variant of aneurysmal bone cyst is difficult to diagnose without biopsy or surgery. It should be included in the differential diagnosis of any lytic expansile destructive lesion of the spine.

Keywords :

Introduction

Aneurysmal bone cyst is a benign expansile, relatively uncommon lesion that represents 1.4 to 2.3% of primary tumors of bone. (1) Aneurysmal Bone Cyst is common in children and adolescents and with 80% cases occurring in patients less than 20 years of age. (2) Spine is involved in 20-30% of the cases. (3) especially posterior elements, with extension into vertebral body in 40% of cases. (4)

Solid variant of ABC was described by Sanerkin et al as a non-cystic fibroblastic bone lesion showing histology similar to the solid non-cystic parts found in the conventional aneurysmal bone cyst, with scattered osteoclastic, osteoblastic, fibromyxoid elements, without a predominant component of cavernous channels. (5) This solid variant may be easily misdiagnosed as a spindle cell tumor, especially osteosarcoma (6). However, the histological features of this tumor closely resembles to those of Giant Cell Reparative Granuloma, described by Jaffe et al. (7), occurring as a reactive lesion to intra-osseous bleeding. This indicates close relationship between these two conditions.

It is a rare lesion, accounting for 3.4% to 7.5% of all aneurysmal bone cysts (5) and only 14 cases (6,8) occurring in the spine have been previously reported. These cases were predominantly seen in children.

The treatment of choice for aneurysmal bone cysts has been complete surgical resection, but in selected cases the risks of unacceptable surgical morbidity and excessive hemorrhage in hyper vascular tumors and the challenge of maintaining spinal stability are indications for adjunctive or alternative therapy.

We report a case of the solid variant of aneurysmal bone cyst occurring in the T10 vertebra with cord compression a 14-year-old male patient, treated effectively with radiotherapy followed by surgical resection bone grafting and posterior fixation. We review the 14 prior cases that have been reported in the literature and discuss the unique features of these unusual tumor-like lesions of the vertebral column.

Case presentation

A 14-year-old male was presented to us with history of back

pain since several months and acute onset weakness in both lower limbs and difficulty in ambulation since one month. On physical examination, tenderness could be elicited on palpation of the spinous processes of the lower-thoracic spine. Neurologic examination revealed motor grade three power in both hips and knees and motor grade zero power in both ankles and feet. There was decreased light touch and pinprick sensation below the T-10 dermatome region. Bowel and bladder functions were unaffected. A rectal examination showed adequate voluntary rectal tone. There was no perianal anesthesia. The post-void residual volume of urine was negligible. X-rays showed slight collapse of T10 vertebra. (Fig 1)

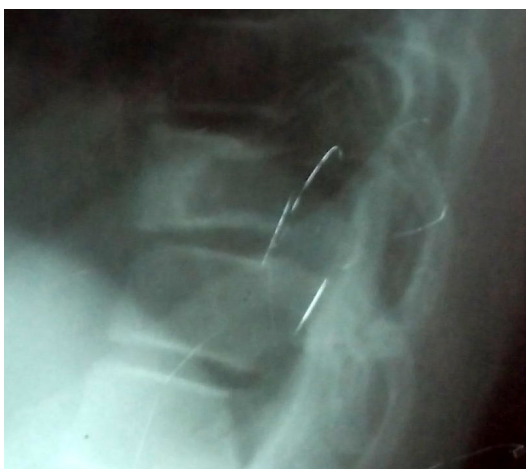

Figure 1
Pre-op xrays showing slight collapse of T10 vertebra

Computed tomography (CT) scan of the thoracic spine showed an expansile lytic lesion involving the all posterior elements and posterior half of T10 vertebral body. (Figure 2) Magnetic Resonance (Fig 3) imaging of the spine showed a multiloculated large hypointense lesion on T1-weighted images with homogenous enhancement. The lesion showed mixed areas of low signal intensity with scattered high intensity regions on T2-weighted images with fluid-fluid levels; which indicated microcyst formation. There was an epidural extension of the mass which compressed the cord. There were not any myelomalacic changes in the cord. CT-angiography was done to look for feeding vessels, which showed sharing of blood supply with the corresponding spinal segment.

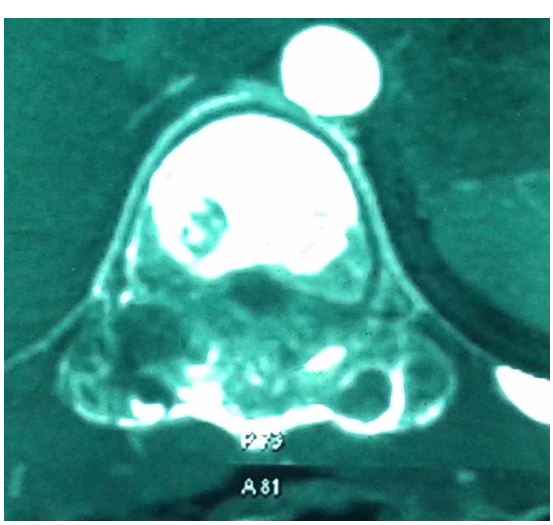

Figure 2: CT Images
Pre-operative computed tomography (CT) windowed for bone at the level of T6 shows an osteolytic and expansile lesion predominantly involving the the whole neural arch and posterior half of T10 vertebral body

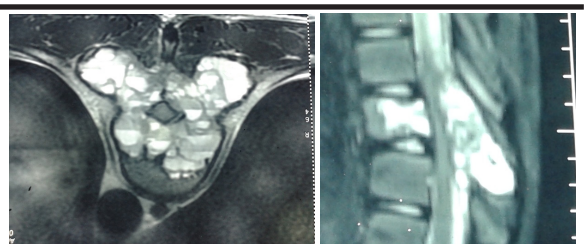

Figure 3
Pre-operative MRI demonstrate a large heterogeneous low (T1) and high (T2)signal intensity mass lesion involving T10 with fluid fluid levels.

Initially, we decided to treat the patient with Radiotherapy alone, with a hope to decrease the vascularity and tumor mass and ultimately relieve the cord compression. Radiotherapy was planned in 20 fractions of 40Gy each with a maximum radiation dose of 200 Gy per session. Patient showed mild neurological improvement post radiotherapy session in the form of regaining some power in ankles and feet; however, back pain was persistent. Post-radiotherapy MRI(Figure 4) showed decreased vascularity and involution of lesion; however, the cord compression was persistent.

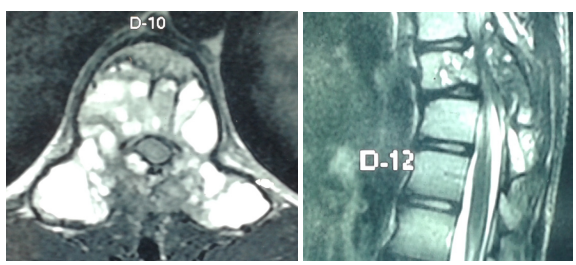

Figure 4
Post-radiotherapy MR showed decreased vascularity and mass of the tumour

Hence, to relieve the compression, surgical resection was planned. We did decompression of the cord via posterior midline approach. Surgery was performed under neuromonitoring using motor and somatosensory evoked potentials. A T9 to T11 laminectomy was performed (Figure 5). All the tumor tissue surrounding the cord was carefully removed and the cord was freed of adhesions. Pedicle screws were inserted in T9 and T11 bodies. After temporary placement of a rod on left side, involved posterior elements were removed. We approached the diseased body through right pedicle. Contrary to our belief, we encountered solid tissue instead of characteristic cystic lesions. Bleeding and adhesions were more than expected too. The tumour tissue within the diseased body was curetagged and the cord was checked again for any remaining adhesions. After ascertaining an adhesion-free cord, the defect was filled with bone graft taken from posterior superior iliac spine. Posterior stabilization was done by using pedicle screws and rods placed at T9 and T11 vertebrae.

Figure 5: Clinical Photograph
Immediate post-operatively, there was no worsening in neurology. There was no hematoma or infection. We allowed the patient to turn in bed and to sit with DLSO brace. Patient showed neurological recovery at 1 month post-operatively, the neurology was almost normal with full power in both lower limbs.

Post-operative MR showed almost total resection of the mass lesion, acceptable reconstruction of the vertebral column and adequate screw placement (Figure 6)

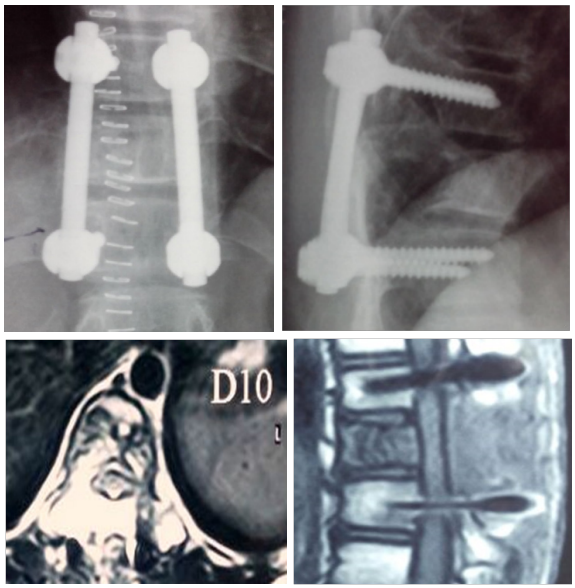

Figure 6: Post-op x-rays and MR

At 2 months after surgery our patient continues to do well and to be satisfied with the surgery, remaining pain free and neurologically intact. There is no evidence of tumour recurrence. Long term follow up is planned to watch for recurrence

Permanent sections (Figure 7) showed a predominantly solid lesion. The lesion consisted of spindle shaped stromal cells interspersed with multi-nucleated osteoclast-like giant cells. There were areas of calcification and reactive bone formation. There were foci of hemosiderin within the mass. The solid areas also showed scattered cyst-like areas filled with erythrocytes s/o areas of hemorrhage. No features of malignancy, atypia or mitosis were seen. The histopathological features were consistent with those of a predominantly solid variant of aneurysmal bone cyst.

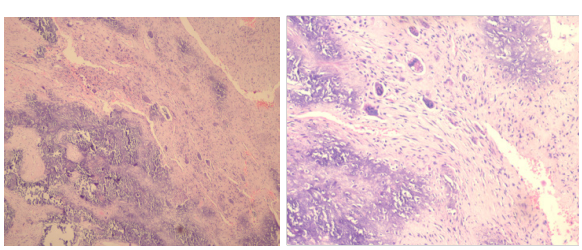

Figure 7
Photomicrographs of the specimen show the proliferating round to oval cells (Fibroblasts) mixed with randomly distributed multi-nucleated giant cells, suggestive of the solid variant.

Discussion

Aneurysmal bone cysts are rare primary bone tumors commonly affecting young patients less than 20 years age.(9) spine is involved in 20-30 percent of cases. In spine, they commonly affect the thoracic spine (34%) followed by lumbar (31%) and cervical spine (22%). (10)

Histologically, aneurysmal bone cyst is typically characterized by cavernous channels surrounded by a spindle cell stroma with osteo- clast-like giant cells and osteoid production. There is a distinct solid variant of aneurysmal bone cyst, first described by Sanerkin et al. (5) It is a rare lesion, accounting for 3.4% to 7.5% of all aneurysmal bone cysts, and only 15 cases occurring in the spine have been previously reported.

In our review of 15 cases, including our patient, of spinal involvement of the solid variant of aneurysmal bone cyst is summarized in Table 1. The age of patients ranged from six to 18 years (mean, 11.5 years), and the male: female ratio was 1:1.5. Nine cases showed involvement of thoracic spine. Pain in neck or back was most common presentation and majority of the patients had symptoms for more than one year. Neurological symptoms were seen in three cases.

Table 1
Previous reports of solid variant of aneurysmal bone cyst of the spine (Suzuki et al (11))

On plane radiograph and conventional CT, the solid variant is indistinguishable from the conventional ABC, both having anosteolytic and expansile appearance. Like conventional aneurysmal bone cysts, almost all cases showed involvement of neural arch with expansile eccentric paravertebral lesion. Vertebral body involvement, as seen in our patient, is rarely seen. It was seen in only three cases prior to our study.

On MRI, as in conventional ABC, the solid variant also shows homogeneous low-signal intensity on T1-weighted images and heterogeneous low-signal intensity with scattered high-signal intensity areas on T2-weighted images. However, with contrast-enhanced MRI, solid variant shows more homogenous high signal intensity throughout the lesion while in conventional ABC, thin, smooth septations are seen within the lesion. This is perhaps the only differentiating feature between the two variants.

There are no clear guidelines for the management of these tumours. Treatment options for aneurysmal bone cysts have included simple curettage with or without bone grafting, complete marginal excision, embolization, radiation therapy, or a combination of these modalities. Even though they are benign, prompt surgical intervention is mainstay of treatment, especially in cases with neurological involvement.

Local control may be in the form of simple curettage considering the benign nature of the lesion, however, recurrence rate may be as high as 30%. (4,10) Complete marginal excision may decrease the chances of recurrence, but it may lead to excessive vertebral body resection and spinal column instability.

There are several reports of late post-irradiation sarcomas and post-irradiation myelopathy in patients with conventional aneurysmal bone cysts. Hence, radiotherapy is indicated in selected cases; especially for patients with inoperable lesions because of location or associated medical conditions, or aggressive recurrent disease. In our review, radiation therapy was undertaken in two cases prior to our study. Intra-cystic sclerosant injections have resulted in mortality and major morbidities when used in the spine (12).

Embolization of feeding segmental arteries may be used as a pre-operative adjunct or sole treatment for aneurysmal bone cysts (13). It has many limitations, as it cannot be used alone in cases with pathological fractures or neurological compromise. Inadvertent embolization of segmental arteries to the spinal cord may result in spinal cord infarction.

The management of these tumours is controversial and one must take into account the age of the patient, the surgical accessibility of the lesion, necessity to minimize intraoperative blood loss, the presence of neurological compression, the presence of a pathological fracture and deformity, and potential postoperative instability after complete resection.

In our case, there was extensive involvement of the vertebral body with suspected high vascularity of the tumor. Owing to the fact that tumour derived its blood supply from spinal segmental artery pre-operative embolization to reduce the vascularity was considered risky. Hence, despite risk of adverse effects, we decided to give pre-operative radiotherapy. Post-radiotherapy MRI showed significant decrease in vascularity and tumour mass as well as improvement in neurologi-

cal condition; however, cord compression was persistent. So, operative decompression was carried out.

Depending on the proliferative component, the solid variant of aneurysmal bone cyst may be histologically misdiagnosed for other benign and malignant and tumor-like lesions of the bone. The pathological differential diagnosis includes solitary bone cyst, hemangioma, osteosarcoma, giant cell tumor, and chondroblastoma.

Conclusions

Because of its rarity, location, and radical treatment approach, we considered this case worthy of reporting. The solid variant of aneurysmal bone cyst is difficult to diagnose radiologically before biopsy or surgery. Our patient was treated with pre-operative radiotherapy followed by in-situ local curettage of the aneurysmal bone cyst via posterior approach and reconstruction of the vertebral column. There was complete neurological recovery at the 1 month post-operatively. Whether conservative clearance of the tumor results in a better outcome in terms of recurrence and spinal column stability than a more aggressive approach (for example, marginal excision) remains to be seen in longer-term follow-up.

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