Intralesional Excision of thoracic spine Aneurysmal Bone Cyst "ABC" with vertebral body involvement, Case Study

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ABSTRACT

The Aneurysmal Bone Cyst (ABC) is a rapidly growing tumor of an undefined neoplastic nature. It was first described by Jaffe and Lichtenstein in (1942), and it occasionally acts as an aggressive benign lesion for which the treatment of choice is a complete resection, with an increased risk of Intraoperative bleeding.

Aneurysmal Bone Cyst (ABC) is a rare localized tumor of the long bones and spinal vertebrae. This tumor can develop at early childhood and early adolescence with a slight female predominance. It constitutes 1.5% of all primary bone tumors and about 14% of all primary spine tumors.

We will discuss a case of a female patient who was referred to our hospital 20 days after the primary diagnosis of spinal tumor with progressive parasthesia and muscle weakness of lower extremities that evolved to paralysis of both lower extremities and sphincter incontinence.

Clinical approach and the investigation done for the patient along with the surgical procedure which was intralesional complete excision and the follow- up outcome will be discussed. Other non-surgical treatment options will be also be discussed.

Key words: Aneurysmal Bone Cyst (ABC), Spine Tumor, Paralysis.

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INTRODUCTION

Aneurysmal Bone Cyst (ABC) is a benign, expansile non-neoplastic relatively uncommon lesion, representing about 1.5 % of all primary bone tumors. The vertebrae are involved in 5–30 % of cases, compromising about 14% of all primary spine tumors [1,2, 3, 4, 6, 7].This lesion was first described by Jaffe and Lichtenstein(1942)as blood-filled endothelialized cavities [2, 3]. It mainly affects the pediatric age group between the age of 10 to 20 years old with a slight female predominance [3,5].

Thoracic and lumbar spines are the most affected parts of the vertebral column [3].It usually starts in the posterior part of the vertebral body, and with its expansile nature, it can extend anteriorly causing destruction of the body and spinal cord compression with neurological symptoms[3,5],so it is a benign locally aggressive bone tumor.

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Aneurysmal Bone Cyst (ABC) can be of two types; primary (de novo) ABCs, which represents about 70 % of all ABCs, and secondary ABCs with multiple hemorrhages, which develop most of the times due to cystic degeneration in a background of giant cell tumor, osteoblastoma, chondroblastoma, or fibrous dysplasia of bone [3,7].

Treatment options:

Treatment options of the spinal ABCs are still controversial ranging between surgical and non-surgical options being available according to the case [3,5].Non-surgical options include selective arterial embolization of the feeding vessels, intralesional calcitonin or steroid injection.

Radiotherapy is another option for those unresectable huge ABCs, or post incomplete surgical excision of the cyst as an adjuvant therapy, with the increased risk of radiotherapy associated osteonecrosis and myelopathy [3].

For the surgical options in treating ABCs, the best to do is a wide local resection to prevent, or to intra lesional curettage with bone grafting. If the cyst is too big, you can augment the body with Polymethyl Methacrylate (PMMA) bone cement to enhance stability. In most cases of surgical resection, instrumentation of the spine is mandatory for early rehabilitation

Case Description

A 12 years old female patient with no previous significant medical or surgical history, was referred to our hospital 20 days post admission to a public local hospital with a history of progressive lower limbs weakness and parasthesia associated with progressive urine and stool incontinence, in the form of inability to void urine voluntarily, and abdominal pain associated with constipation. She had already presented on referral with Foley's catheter in situ from the referring hospital. Taking history from the patient revealed that her symptoms were associated with mid back pain; there was no history of trauma or past history of back surgery, and no family history of neuromuscular disorders.

Upon examination of the patient, she was in pain, depressed and a little bit pale. Her systemic examination was within normal limits, with no abnormal findings apart from mildly distended abdomen with mild generalized tenderness due to constipation and gaseous bowel, and bladder was not palpable because of Foleys catheter. Conducting rectal exam for our patient showed increased tone of anal sphincter with loss of voluntary anal contraction. There were no palpable back masses or obvious deformities.

Examination of the lower limbs showed symmetrical bilateral hypertonia with difficult passive range of motion bilaterally in both ankles and knees. Muscle power was absent in both lower limbs except for toes flickering in left side. Touch and pin prick sensation was diminished up to a groin level. Knee and ankle reflexes were exaggerated and brisk bilaterally. Examination of both upper limbs was within normal with no abnormal findings.

Laboratory investigation for our patient showed normal Erythrocyte Sedimentation Rate (ESR) and Normal C-Reactive Protein (CRP), normal kidney and liver function tests, and normal hemoglobin level and normal white blood cells count.

Collecting previous data together indicated that the patient had a central cord problem, so next step was to do imaging studies for the patient. Anterior-Posterior (AP) radiograph for the whole spine and the pelvis was not informative because of gaseous abdomen , and lateral radiograph for the whole spine could not be done because the patient could not stand.

Next, we did Computerized Tomography (CT) Scan for the thoracolumbar spine of the patient that showed a destructive expansile bony lesion with egg shell bony margins involving the posterior

elements of the 10th and 11th thoracic vertebrae, as well as the pedicles and extending to the posterior two thirds on the left side of the 11th thoracic vertebral body associated with soft tissue element (**Figure 1**), so Magnetic Resonance Image (MRI) was done.

Magnetic Resonance Image (MRI) with contrast revealed an expansile multicystic lesion involving the left side body, pedicle, lamina and spinous process of the 11ththoracic vertebra (T11), and the lamina and spinous process of the 10th thoracic vertebra (T10), containing fluid-fluid levels and causing severe compression of the spinal cord pushing it anteriorly and to the right side. The lesion showed minimal septal enhancement (**Figure 2**).and by combining the data from both the CT scan and the MRI and the clinical data all together, the most likely differential diagnosis for the Radiological findings for this patient was Aneurysmal bone Cyst .

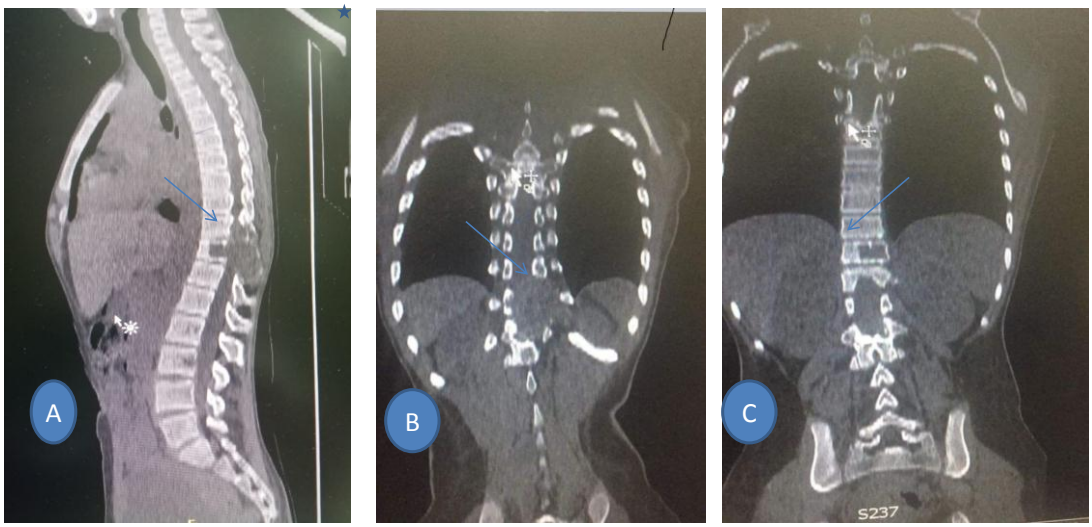


Figure 1 : computerized Tomography (CT) Scan of the patient :A:sagittal,B and C: Coronal Cuts showing destructive ostelytic lesion involving the posterior elements of the 10th and 11th thoracic vertebra and extending to the body of the 11th vertebra with soft tissue component and egg shell boundary.

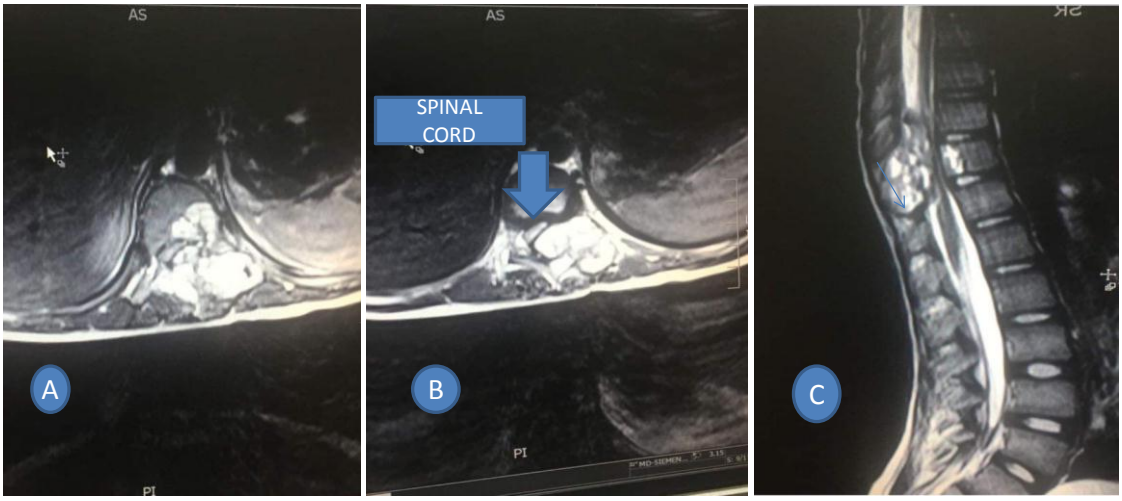


Figure 2 :**Magnetic Resonance Image (MRI) of our patient, A and B:Axial cuts , C:Sagittal cut showing sever Spinal Cord Compression with involvement of the posterior elements and posterior tow thirds of the left side of vertebral body, multicystic lesion and multiple fluid-fluid levels.**

Management

As the diagnosis of ABC was radiologically confirmed, our patient was planned for catheterization and selective embolization of the feeding vessels as a preparation for the surgery to be done. Our interventional radiologist successfully embolized the lesion following complete investigation of the patient (**Figure 3**).

Surgery was planned for the patient within the next (48) hours post embolization, especialy with the significant spinal cord compression and lower limbs paralysis. Surgery was done within (48) hours with the plan of doing long segment instrumentation, intra lesional curettage and bone grafting. This was done by a posterior approach only. Muscles were dissected from the tumor boundaries easily. Three levels above and three levels below the involved vertebrae were instrumented. The posterior elements of the 10^{th} and 11^{th} vertebrae were dissected and excised en-block and the cord was significantly decompressed. Aggressive curettage for the body of T11was done and synthetic bone graft was done for the cavity (**Figure 4**). All tissues removed were sent for histopathological examination No significant bleeding occurred intraoperatively and no blood transfusion was required. The histopathology report confirms the diagnosis of Aneurysmal Bone Cyst, Solid variant with no evidence of malignancy (**Figure 5**).

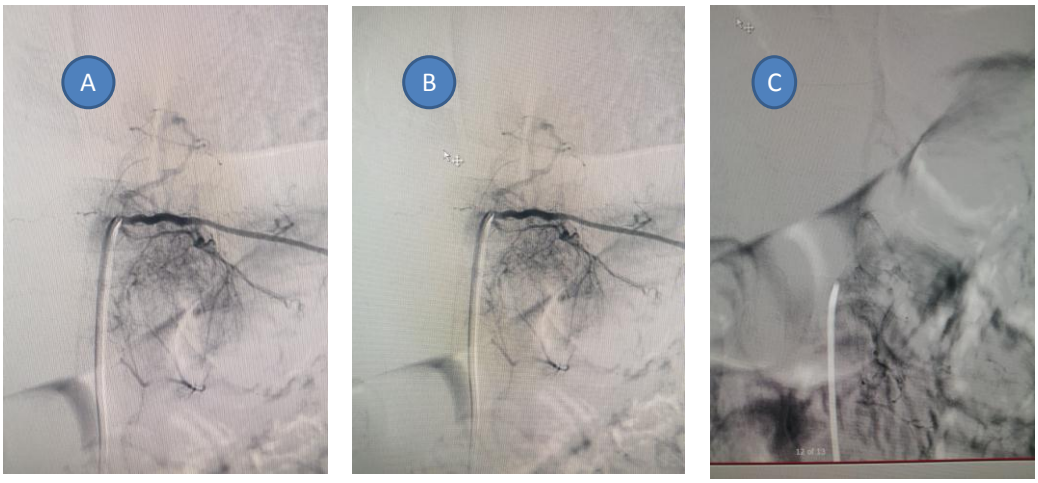
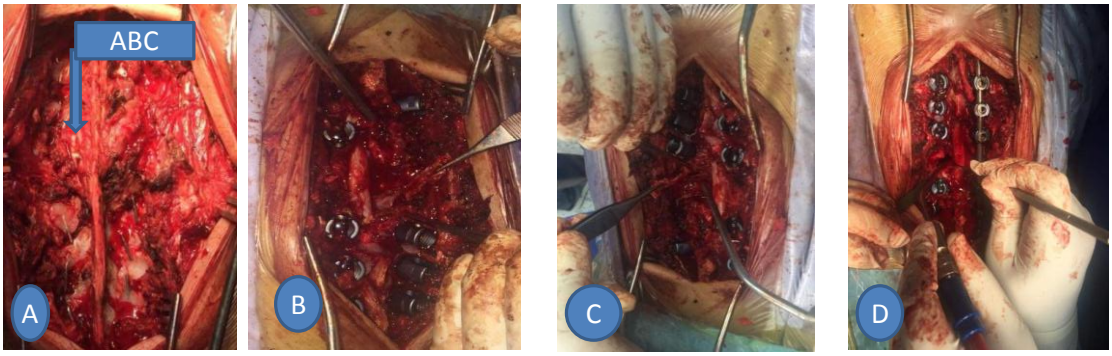


Figure 3: pre :A and B ,and post :C Angiographic Selective embolization of the feeding vessels to the lesion, done 24 -48 hours prior to our surgery.



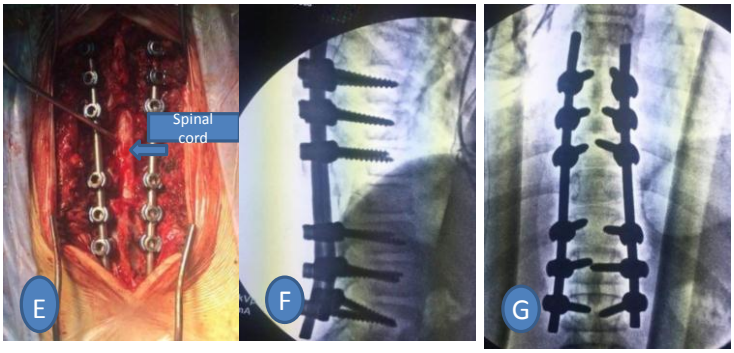


Figure 4: intraoperative technique,A:dissection of the muscles from the cyst, B and C:dissection and decompression of the spinal cord,, D:transpedicular curettage and bone grafting of the vertebral body, E:long segment fixation, F and G: lateral and anterior-posterior post-operative radiograph respectively.

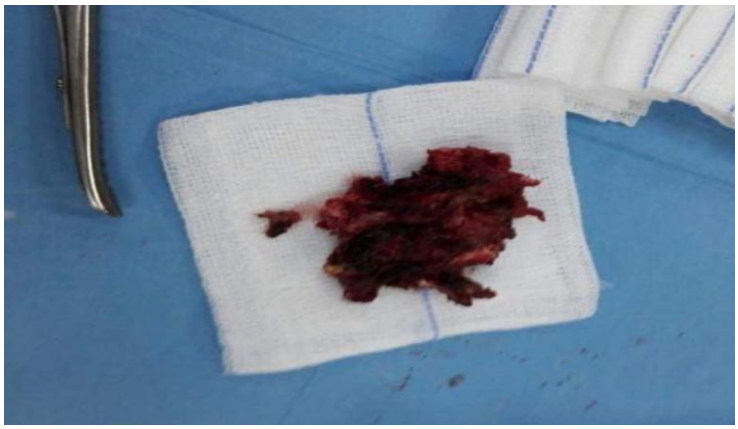


Figure 5: all en-block posterior elements and the curettaged tissue from the body sent for histopathology exam, result confirmed the diagnosis of Aneurysmal Bone Cyst, solid variant .

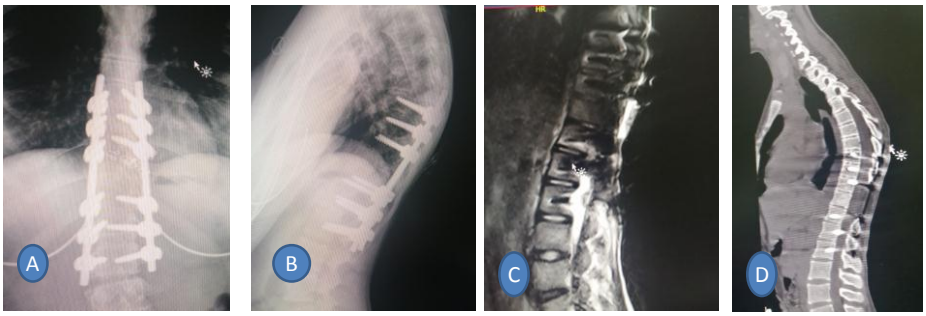
Follow up

Patient showed significant improvement within the first few days post-operatively. The lower limbs tone decreased and was back to normal within 48 hours post-surgery, and she gained sensation for touch and painful stimuli 4-5 days post-operatively, with good muscle power allowing her to stand up with the help of the physiotherapist , fourth day post-operatively. Foleys catheter was removed on the seventh day post operatively and the patient was able to control urination and defecation within two weeks post operatively. The patient was walking independently with full power and full sensation of both lower limbs, and well controlled urination, well controlled defecation, and clean dry wound 20 days post admission (**Figure 6**).

Patient was on regular follow up at our clinic for the last 3 years; she showed no alarming signs or symptoms with no any clinical or radiological evidence of recurrence.

Radiographs were done every six months and are within normal limits. Computerized Tomography (CT) Scan and Magnetic Resonance Imaging (MRI) were done at one year and two years follow up visits, which showed complete consolidation of T11 vertebral body with no evidence of recurrence (**Figure 7**).

Figure 6: clinical follow up of the patient, A: good muscle power and tone and sensation 4-5 days post operatively, B: back to normal power and sensation and sphincter control 3 weeks post operatively. Family give the permission to use the photos of the patient



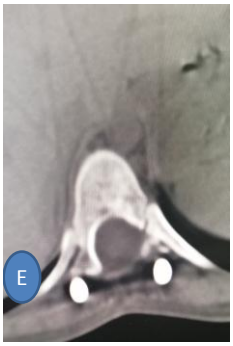


Figure 7: Three years radiological follow up for the patient by Anterior-posterior (A) and Lateral (B) Radiographs Magnetic Resonance Image MRI (C) and Computerized tomography CT scan Sagittal (D) and Axial (E) showed complete consolidation and no evidence of recurrence.

DISCUSSION

Aneurysmal Bone Cyst (ABC) can cause significant morbidity if delayed in diagnosis or inappropriately treated, even if it is a benign pathology.[3, 4, 6].

The symptoms of ABC can be present few months prior to diagnosis, and localized pain is the main presenting symptom in most of the patients which typically increases with recumbency. Some patients may come with palpable tender mass, other patients may manifest with functional deformity due to pain and muscle spasm. 60-70 % of the patients can initially show up with neurological symptoms [3],ranging from parasthesia due to root compression up to complete paralysis due to acute spinal cord compression. Accidental finding in a pathological fracture can be the initial presenting symptom.

Aneurysmal Bone Cysts ABCs are generally thought to be the result of a secondary vascular phenomenon that develop on a background of a preexisting lesion like an unnoticed fracture or preexisting subperiosteal hematoma, which presumably initiates a periosteal or intraosseous arteriovenous malformation[8].

Histopathologically, it is better described as an expansile osteolytic lesion consisting of blood-filled spaces lined by endothelial tissue and separated by connective tissue septae containing osteoid like tissue and osteoclast –like giant cells [8].

Treatment options are wide and controversial, ranging from radiological, medical, and surgical treatment option, depending on the site, size, and accessibility to the lesion, as well as the facilities and experience of the surgeon.

Some ABCs can be treated conservatively when the diagnosis is confirmed to be Aneurysmal Bone Cyst, and it is not causing any signs and symptoms and it is not increasing in size [8].When the ABC is causing signs and symptoms and is not accessible by surgery and complete surgical resection was not done, then radiotherapy or intralesional injections trials can be done [3,5].

Complete surgical excision or intralesional curettage and grafting is still the best option for symptomatic young patients, with the best short and long-term outcome and preventing recurrence.

Preoperative selective arterial embolization for the cyst will make surgery easier and decrease the rates of recurrence.

Total excision or intralesional excision or curettage must be done for the entire cyst wall, all abnormal tissues that feel spongy and unhealthy, and bone surfaces that are lined with the cyst membranes [3]. For big and destructive cysts, complete excision or extensive curettage will most likely cause iatrogenic instability, requiring instrumented fusion of some levels above and below, to maintain spinal stability in the post operative rehabilitation and recovery period,

Multidisciplinary team work showed a very rapid and good outcomes in treating this patient, starting from the caring surgical team who received ,investigated and operated upon the patient ,the diagnostic and interventional radiologist who helped a lot in diagnosis and preparing the patient for surgery, making surgery easier and less complicated, the nurses in the operating theater and caring for the patient in the ward, the rehabilitation and physiotherapy team making good and rapid recovery, and the histopathologist who confirmed the diagnosis .

CONCLUSION

Early diagnosis and appropriate surgical treatment of the Aneurysmal Bone Cysts" ABCs " in the spine remain the key factors to successful management. Complete surgical excision or intralesional

curettage and grafting of ABC remain the best method to treat symptomatic patient with or without neurological findings, and has the best short and long-term outcomes. Treating young patients with ABC and neurological signs and symptoms should be aggressive and should not be delayed for better outcome. Preoperative planning and multidisciplinary teamwork will guarantee the best outcome.

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