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Case Report

Aneurysmal Bone Cysts of the Spine: Report of a Case Simulating Giant cell tumor and Review of the Literature

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Abstract: Aneurysmal bone cyst (ABC) is a benign bone lesion, it is now recognized as a highly vascular bony lesion of unknown origin. ABC is a relatively rare lesion, representing 1.4% of primary bone tumors, which can involve any bone in the body, Spinal involvement accounts for 3% to 20% of all skeletal ABC cases. Sometimes, it is difficult to distinguish ABC from other benign bone lesions clinically and radiologically. We report a 19-year-old female complained of persistent thoracic pain for 2 months. She was diagnosed of Giant Cell Tumor at the local hospital. Pre-operative CT and MRI scans demonstrated the lytic nature of the tumor without fluid- fluid sign of ABC at the T10 vertebral body. However, after a successful preoperative embolization, a total resection with internal fixation was performed and complete pain relief without any complication was experienced.

Keywords: Aneurysmal bone cyst, thoracic spine, surgical treatment

INTRODUCTION

Aneurysmal bone cysts (ABCs) are infrequently occurring bone tumors. Most of the ABC lesions involved cervical and thoracic spine, female patients is seen more often than male (JAFFE, H. L., & Lichtenstein, L. 1942). Back pain and neurologic compression are the main symptoms. Radiologically, Eggshell-like periosteal calcification is a typical feature of a spinal ABC, which could be used as a criterion for differential diagnosis. A typical MRI of an ABC depicts tumor masses with smooth margins and osseous destruction. Inside the tumor, there are multiple cysts, commonly with fluid levels and varying signal intensity in T1- and T2-weighted images. However, in rare cases, the cystic components can be completely absent, simulating a solid benign bone, making it impossible to differentiate such as giant cell tumors (Jaffe, H.L.1950). We report a ABC case which is hard to differentiate and offer the treatment strategy.

CASE REPORT

History

A 19-year-old female with history of 3 months moderate back pain and 2 weeks lower limb neurological deficits admitted in our hospital. The pain started without an inducement and somehow worse at night. The pain could be aggravated by motion, especially standing, walking, forward bending movement and could not be relieved by bed rest. Her pain was initially 3/10 in severity but worsened to 6/10 later. The pain localized to the thoracic spine area without radiation. 2 months later there was numbness at both lower limb which was more server of the right side. 2 weeks later, she developed lower limb weakness and complained difficulty of standing. There was no sphincter disturbance and no history of trauma recently. Laboratory examinations consisted of complete blood count, electrolytes, liver, renal function tests, and urinalysis. They were all normal, except remarkable evaluation of C-reaction protein level. Before admission, she admitted at local hospital and discovered a neoplasm located at the T10 and diagnosed as giant cell tumor.

Examinations

At neurologic examination, spinal movements were painful and restricted. Physical examination revealed decreased sensory function at lower abdomen and lower limbs. Increase of tendon reflex was noted. The manual muscle strength test demonstrated 3/5 strength on the left and 2/5 on the right. Babinski sign was positive bilaterally. Muscle tonus of lower

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extremities was increased. X-ray findings indicated a compressive pathological fracture at the T10 level. Para-spinal tissue mass located at right of T10 was not noticed (Fig. 1) Computed tomography (CT) revealed a density increase of the T10 vertebra and show marked thinning of the cortex over the site, with only minimal bone formation. The right transverse processes and laminas were engaged. (Fig. 2) Magnetic resonance imaging (MRI) confirmed the presence of lesions at the T10 level. The lesion had a low signal on T1-weighted images and high signal on T2-weighted images. Intravenous infusion of gadolinium increased the intensity of the signal of the lesion non-harmoniously. There was no para-spinal soft-tissue mass, and the tumor is significantly invading the spine canal and compressing the spinal cord. (Fig.3)

Pathological findings and Pre-operative course

As the radiological findings were similar with a giant cell tumor. She was misdiagnosed and given Denosumab. However, the tumor did not give a positive response to the medicine. A misdiagnose was suspected and biopsy was performed. During biopsy, an obvious bleeding was obtained when the needle was inserted into the tumor. The surgery was stopped and there was no positive pathological result. The significant bleeding suggested the diagnose of Aneurysmal Bone Cyst. During preoperative embolization, the firework appearance of the tumor was observed in digital subtraction angiography.

Surgical treatment

The differential diagnosis included osteoblastoma, giant cell tumor, fibrous dysplasia, and chronic infection. As the neoplasm of lumbar spine produced neurological symptoms. Internal fixation of the spine and marginal resection surgery were performed. Histological examination of the specimen revealed fibrous components, macrophages, and islands of bone consistent with the diagnosis of an aneurysmal bone cyst. The patient recovered completely without complications. After an uncomplicated hospital course, she was discharged home 4 weeks later. After follow-up for 6 months, there was no recurrence of back pain, neurologic examination was normal, the VAS was 0. Now this patient is completely symptom free. (Fig.4)

The design and performance of this study conformed to ethical standards of Helsinki Declaration and our national legislation. It was approved by Medical Ethical Committee of our institution. The patient was enquired whether or not willing to take part in a scientific research and informed consent forms were signed by her self.

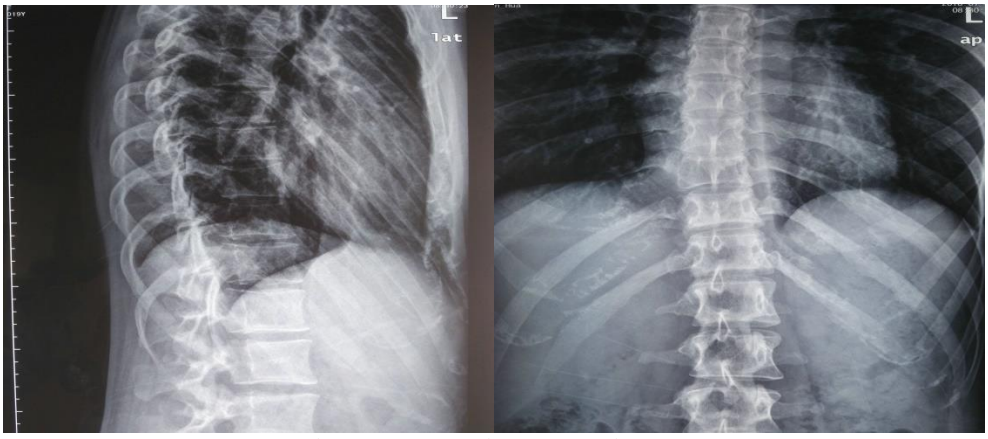

Fig.1 Pre-operative X-ray pictures

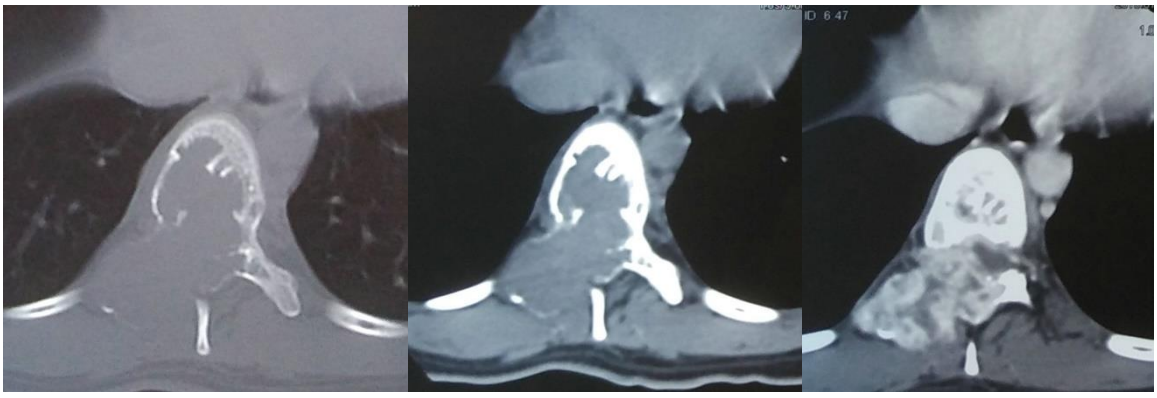

Fig. 2 Pre-operative CT scans

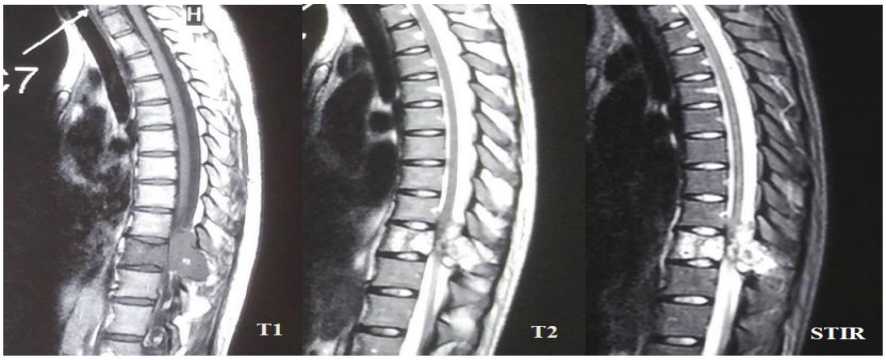

Fig.3 Pre-operative MR images

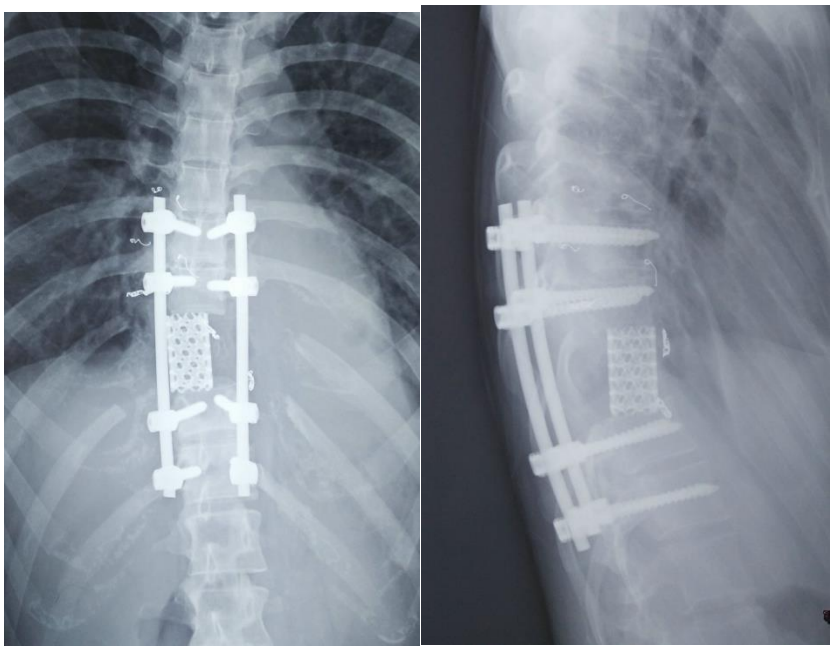

Fig. 4 Post-operative X-ray pictures

DISCUSSION

Aneurysmal bone cysts (ABCs) are infrequently occurring bone tumors. According to an epidemiological study, the annual incidence of ABCs amounts to 0.14 per 105. Overall, women are more often affected than men, with a ratio of 1.04:1 (Lichtenstein, L. 1957). The mean age of the patients affected lies in the 2nd decade of life. The long bones of the lower extremities are afflicted most often, with the tibia and femur being respectively affected in 24.7% and 17.3% of all cases, followed by the upper extremities (10%) and the pelvis (9%). About 14% of all ABCs are encountered in the spine (Jaffe, H.L.1950; Lichtenstein, L. 1957). The involvement of the thoracic spine by an ABC is therefore quite rare. We present a case of ABC with pronounced bone destruction of T10 segments. The images of the lesion were similar to giant cell tumor which induced a misdiagnosis. These diseases are totally different from the therapeutic options. The giant cell tumor should have a positive response to Denosumab, and pre-operative administration is suggested. However, an ABC dose not response to the medicine and should be embolized before operation.

Despite attempts on the part of investigators to establish a relationship of the disorder to other entities, the term aneurysmal bone cyst remains purely descriptive. It does not provide any concept of pathogenesis or causation mechanisms, and efforts on the part of a number of investigators to discover a genetic or neoplastic cause have failed (Aho, H. J. *et al.*, 1982; Bonakdarpour, A. *et al.*, 1978). Examination of the tissue at the time of surgery has, in the past, demonstrated a blood-filled cavity within an expanded region of the bone, and the cells that line the cyst wall show fibrous components, macrophages, giant cells, and islands of bone (Campanacci, M. 1999; Dorfman, H. D., & Czerniak, B. 1998). The term aneurysmal seems to relate to the blowout distension, and the word cyst reflects the fact that the tumor often presents as a blood-filled cavity (De Silva, M. *et al.*, 2003; Feigenberg, S. J. *et al.*, 2001). Occasionally in prior studies, there have been findings suggesting the possibility that the aneurysmal cyst is actually a result of hemorrhagic degradative events occurring in patients with other lesions including giant cell tumor, hemangioma, chondroblastoma, osteoblastoma, non-

ossifying fibroma, fibrousdysplasia, chondromyxoid fibroma, eosinophilic granuloma, and other tumors (Dick, H. M. *et al.*, 1979; Yildirim, E. *et al.*, 2007). Of greater concern is the possibility that the lesion is not an aneurysmal cyst but a partially necrotic and extremely vascular telangiectatic osteosarcoma, which has a high rate of metastasis (DeRosa, G. P. *et al.*, 1990).

Of some importance is the difficulty that can occur in diagnosing these lesions. The imaging studies, even CTs and MRIs, sometimes do not provide clearly diagnostic criteria for the diagnosis of aneurysmal bone cyst, and aneurysmal bone cyst is sometimes added on to a list of diagnoses including eosinophilic granuloma, giant cell tumor, non-ossifying fibroma, unicameral bone cyst, fibrous dysplasia, chondroblastoma, chondrosarcoma, chondromyxoid fibroma, Ewing's tumor, and, in older patients, metastatic carcinoma or myeloma. According to reports, the lesions are often eccentric and irregular in structure and sometimes show calcification in the central areas. As a rule, the cortex is thin, but there is rarely a cortical defect or a soft tissue mass. However, in our case, the cortex of T10 is destroyed and the cortical defect is obvious. It is not easy to identify the diagnosis on first impression. CT and MRI are often helpful in defining the extent of the lesion. A biopsy is often helpful, and many of our patients underwent a needle biopsy before definitive treatment. Needle biopsies are sometimes a problem because the material obtained may consist of mostly blood elements. However, an open biopsy and frozen section have to be carried out carefully to avoid significant intra-operative bleeding.

There are a variety of treatments for ABC, included surgical en bloc excision, complete curettage, adjuvant selective arterial embolization, radiotherapy, and intralesional injection of a sclerosing agent (De Cristofaro, R. *et al.*, 1992). When the lesion is accessible, surgical removal of the tumor is preferred. Although en bloc removal of the tumor has the lowest rate of local recurrence, it can be technically challenging or impossible depending on the location of the tumor and the age of the patient. In a recent case series, It is described a most effective surgical approach involving intralesional removal of the tumor via: intralesional curettage with a high-speed bur, electrocauterization of the lesion, and bone grafting. Selective arterial embolization (SAE) has been used successfully as a sole treatment modality and is also often recommended before surgery to reduce potential bleeding during surgery. Additionally, some authors advocate SAE as a preferred primary treatment option for ABC. However, SAE has some significant disadvantages. In cervical and thoracic spine cases, embolization can cause spinal cord ischemia or embolic events to the brain. Additionally, for SAE to be successful an adequate feeder vessel must be present and multiple procedures are frequently required (Peraud, A. *et al.*, 2004).

CONCLUSIONS

ABCs are benign lesions with a spectrum of behavior ranging from relative indolence to locally aggressive growth. The pathogenesis of aneurysmal bone cyst is still debated, and the origin of this peculiar lesion is not yet clearly known. It has considered it to be a possible result of hemorrhage into either a giant-cell tumor or a chondroblastoma. Therefore, it is difficult to distinguish the tumor from other lesions. Pre-operative biopsy is important to avoid life threatening bleeding. Surgery treatment is preferred when the tumor is accessible. Ultimately, decisions concerning treatment must be carefully weighed by considering a patient's unique clinical picture, size and location of the spinal ABC, and the surgeon's familiarity with these techniques.

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