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(CASE REPORT)

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Aggressive Osteoblastoma in adolescents: Radiological case report

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Abstract

In this article we represent an 18-year-old female with progressive back pain and neurologic symptoms due to a large heterogenous mass centered in posterior elements of lower thoracic vertebrae with prominent soft tissue extension into paravertebral and epidural space and subsequent central cord compression along with severe adjacent inflammatory changes.

Considering patient age, lesion location and characteristics, aggressive Osteoblastoma, eosinophilic granuloma, and with lower probability of Ewing sarcoma were included in the differential list, which aggressive Osteoblastoma was confirmed after pathologic examination.

Keywords: Osteoblastoma; Adolescents; Eosinophilic Granuloma; Ewing Sarcoma

1. Introduction

Osteoblastoma is a relatively rare bone-forming tumor that generally occurs in the spine or flat bones, when they affect the spinal column mostly originating in posterior elements. Osteoblastoma develops from non-odontogenic epithelial cells. This illness is extremely rare in children [1], with only a few occurrences of Osteoblastoma reported.

Osteoblastoma peak incidence rate is in the 2nd decade of life, may progress either slowly or aggressively, and are more common in men, although the aggressive subtype generally occurs in an older age group and is characterized histologically by the presence of a high number of epithelioid osteoblast with nuclear atypia [2, 3]

On radiographs, the lesion might look radiolucent, it might be clearly defined or it might be poorly defined, and it might include varying degrees of mineralization [4]. On imaging, they usually resent as expansile lesions with a thin cortex and may contain a mineralized matrix, and less frequently can present as entirely lytic lesions. On MRI fluid sensitive sequences ranges from low to high signal intensity with variable signal intensity and may have extensive peripheral marrow and soft tissue edema known as the flare phenomenon [5].

Generally, treatment is achieved by surgical resection or curettage followed by radiotherapy [6]. Excision of the lesion in its entirety or curettage are the two methods utilized in the treatment of Osteoblastoma [7]. There have been accounts of the Osteoblastoma shrinking after a biopsy or an incomplete excision, which lends credence to the hypothesis that it is a reactive process [8].

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Based on clinical pathological findings, it is classed as benign or aggressive. The benign variant is characterized by a well-defined, highly vascularized lesion including a moderate number of multinucleated giant cells that create irregular trabeculae of new bone [9].

The aggressive type of Osteoblastoma exhibits abnormal histological features, including epithelioid osteoblasts, a high recurrence rate, and aggressive behavior. It is relatively uncommon among children [10]. New advanced biomarkers can diagnose benign and malignant tumors, such as molecular pathologic [11]. Immunohistochemistry can be used to differentiate osteoid osteoma and Osteoblastoma because overexpression is seen in the majority of cases and is uncommon in their mimics [12].

As it mentioned, aggressive Osteoblastoma in young and adolescent is rare. In this case report, we present an 18-year-old female with aggressive Osteoblastoma and describe the radiological findings.

2. Case Presentation

In this case, an 18-year-old female with back pain and neurologic symptoms refer to our clinic. on MRI, a heterogenous lytic expansile lesion centered in posterior elements of the T11 vertebra is noted, which shows heterogeneous high T2 and heterogenous intermediate T1 signal intensity with prominent soft tissue component which has extended into epidural space from T10 to T12 level and caused severe central canal stenosis.

Areas of signal void due to the mineralized matrix are also present. On post-contrast images lesion shows avid heterogeneous enhancement, extensive bone marrow, and peripheral soft-tissue edema in fluid sensitive sequences are evidently indicative of flare phenomenon, also reactive sclerosis of T11 vertebral body is present. Figures 1 to 4, are describing the radiological findings.

After reviewing these findings, the diagnosis of aggressive Osteoblastoma has been made.

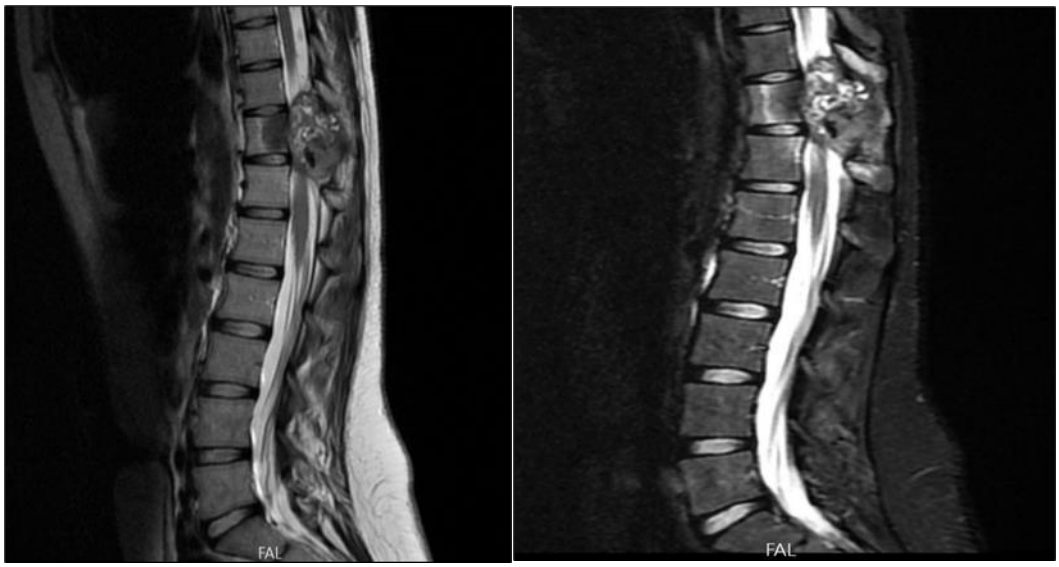

Figure 1 (A and B) Sagittal T2 image depicts large heterogenous mainly high T2 signal lesion with severe reactive bone marrow and soft tissue edema with adjacent dense reactive sclerosis extending into epidural space from T10-T12 level

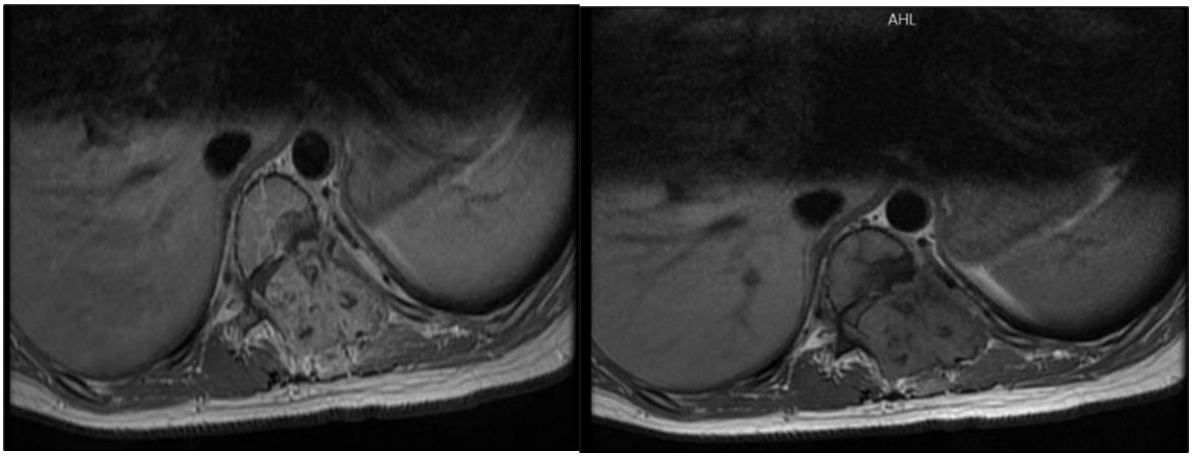

Figure 2 (A & B) Axial pre and post constrast T1 images delineate heterogenous enhancing soft tissue lesion mainly emanating from posterior elements with extension to epidural space and significant central canal stenosis, also vertebral body, paravertebral muscles and rib involvement is noted

A heterogenous lytic expansile lesion centered in the T11 vertebra shows high T2 and intermediate T1 signal intensities with a prominent soft tissue component that has extended into the epidural space from T10 to T12 and caused severe central canal stenosis.

A mineralized matrix causes signal voids. On post-contrast images, the lesion shows avid heterogeneous enhancement, extensive bone marrow and peripheral soft-tissue edema are indicative of flare phenomenon, and reactive sclerosis of T11 vertebral body is present.

3. Discussion

Osteoblastoma is a rare primary benign neoplasm that accounts for less than 1% of all bone tumors. The age of onset ranges from 3 to 70 years, with approximately 90% of patients diagnosed before the age of 30 [13]. Approximately 32% of patients develop in the spine's posterior elements, 13% in the femur, 10% in the tibia, and 9% in the foot and ankle bones. Ribs, small bones in the hands and feet, facial bones, calvarium, patella, scapula, and ilium were among the unusual locations. In some cases, the tumor will exhibit aggressive clinical and radiological behavior. Aggressive Osteoblastoma is a variant that histologically resembles osteosarcoma , so proper clinico-radiological correlation is critical to avoid misdiagnosis [14].

Imaging studies show a typical oval to round, radiolucent lesion with scattered areas of mineralization, the central density of trabecular bone, and easily discernible margins. Mineralization ranges from radiolucent to extensive. The current tumor had a distinct margin and focal mineralization. No dense cortical sclerosis surrounds the osteoid osteoma [15, 16]. The size and extent of the tumor in the cortical bones is best evaluated by CT, which aids in preoperative evaluation and surgery planning.

On MRI, the extent of the lesion within the medulla, soft tissue, areas of cystic degeneration, and hemorrhage (which may occur in a few cases) can be best perceived [16-18]. Aggressive Osteoblastoma and conventional Osteoblastoma have similar radiographic appearances. Radiological features of Osteoblastoma may resemble those of malignant bone tumors, such as cortical destruction and extraosseous soft tissue expansion [19]. Cortical destruction was observed in this case, which radiologically resembled an aggressive neoplasm. Khin et al. described a case of aggressive Osteoblastoma in the humerus with radiological features similar to osteosarcoma [16]. When consider the imaging, we should consider some bone involvement in spine can present in different orders of routines, such as neuroinflammation of vertebra in other diseases [20].

During a 17-year period, Arkader et al. reviewed the medical records and radiographs of all children diagnosed with Osteoblastoma. The criteria for inclusion were met by seventeen children. There were ten boys and seven girls, with an average age of 11 years at diagnosis (range, 20 months-15 years). The average time between symptom onset and diagnosis was 6.5 months (range, 2 months-2 years). Seven lesions were found in the lower extremity, five in the spine, four in the upper extremity, and one in the sternum. In all cases, there was pain at the tumor site. Scoliosis affected two of the five patients with spine lesions. All of the patients had open incisional biopsy with intraoperative frozen section.

In 16 cases, this was followed by a four-step procedure (extended curettage, high-speed burring, electrocauterization of cavity wall, and phenol 5 percent solution). After tumor removal, four of the five patients with spine lesions had instrumented posterior spine fusion. Two patients were referred to after recurrent lesion surgery elsewhere. Only one (6%) of the 15 children who were initially treated at our facility had a recurrence. All recurrences occurred in children under the age of six, and all were successfully treated with a four-step protocol. They proposed A four-step approach can successfully treat osteoblastomas [21]. Recurrence is more likely in children under the age of six.

For the treatment of osteoid osteomas (OO) and osteoblastomas, particularly in the appendicular skeleton, the treatment of choice is considered to be radiofrequency ablation (RFA) [22]. Several novel and trending therapeutic targets for radiofrequency ablation (RFA) have been studied [23]. There is mounting evidence to support the use of this treatment in the spine as well [24]. RF ablates and creates an electromagnetic field. In thermal RF, the patient's tissue is the therapeutic target. RF-induced interactions cause heat, necrosis, and tissue destruction, relieving pain or burning the painful nerve [25]. CT-guided percutaneous RFA is a safe and effective treatment for spinal OO, especially in lesions with no neurological deficits and intact cortical bone. Cerebrospinal fluid around the lesion is an appropriate indication for percutaneous RFA [26].

4. Conclusion

We presented a rare case of Aggressive Osteoblastoma in Adolescents on T11 vertebra. He was an 18-year-old female with progressive back pain and neurologic symptoms due to a large heterogenous mass centered in posterior elements of lower thoracic vertebrae with prominent soft tissue extension into paravertebral and epidural space and subsequent central cord compression along with severe adjacent inflammatory changes.

Considering patient age, lesion location, and characteristics, aggressive Osteoblastoma, eosinophilic granuloma, and with lower probability of Ewing sarcoma were included in the differential list, which aggressive Osteoblastoma was confirmed after pathologic examination.

Compliance with ethical standards

Acknowledgments

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Disclosure of conflict of interest

There is no conflict of interest for any of the authors.

Statement of informed consent

Informed consent was obtained from all individual participants included in the study.

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