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CASE REPORT

Giant cell tumor of the eleventh thoracic vertebra in a pediatric patient: an interesting case report and comprehensive literature review

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Abstract

Introduction Giant cell tumors of the mobile spinal column are very rare tumors, especially in the pediatric age group. Although they are benign tumors, they have locally aggressive growth pattern and high risk of recurrence.

Case presentation We report a 15-year-old female patient with thoracic giant cell tumor who underwent percutaneous vertebroplasty and had cement extravasation into the spinal canal. Because of the deterioration of the patient's neurological condition, total enbloc spondylectomy and cement excision were performed. She underwent instrumentation and fusion procedures in order to prevent spinal instability.

Conclusion The main purpose of the treatment is gross total resection of the giant cell tumor. However, adjuvant methods such as denosumab should be added to the treatment protocol in patients who are older than 2 years old and can not undergo gross total resection due to tumor size and anatomic localization.

Keywords Giant cell tumor · Pediatric · Mobile spinal column · Spondylectomy · Vertebroplasty · Cement extravasation

Introduction

Giant cell tumors are benign but locally aggressive tumors of the bone. They tend to metastasize in some cases. Giant cell tumors constitute approximately 5% of all bone tumors and 20% of benign skeletal tumors. They are usually located at the metaepiphyseal ends of long bones. They are more common in female patients and in the third and fourth decades [1, 2]. Spinal giant cell tumors are very rare and they represent only 2.7–7% of all giant cell tumors [2–6]. In this report, we present a 15-year-old female patient with thoracic giant cell tumor who underwent percutaneous vertebroplasty and had cement extravasation into the spinal canal. We discuss the treatment modalities of this rare disease and present a comprehensive literature review.

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Case report

A 15-year-old female patient admitted to our clinic with complaints of severe back and back pain for 2 years. Her visual analog scale score was nine in the thoracic region and seven in the lumbar region. The patient underwent percutaneous vertebroplasty at another neurosurgical clinic 16 months ago. After the operation, progressive muscle weakness and numbness complaints developed in her lower limbs. The patient was unable to walk and was in a wheelchair. Our neurological examination revealed that she had paraparesis (left lower extremity was 1/5 and the right lower extremity was 3/5) and parahypoesthesia below the level of T10. Patellar deep tendon reflexes were exaggerated.

The thoracolumbar radiographs showed a cement appearance extending from the corpus into the spinal canal with T11 vertebra compression fracture (Fig. 1a, b). Spinal computed tomography revealed a lytic lesion in T11 vertebra corpus and cement extravasation into the spinal canal (Fig. 1c–e). Spinal magnetic resonance imaging revealed a heterogeneous mass lesion and cement extravasation extending from the T11

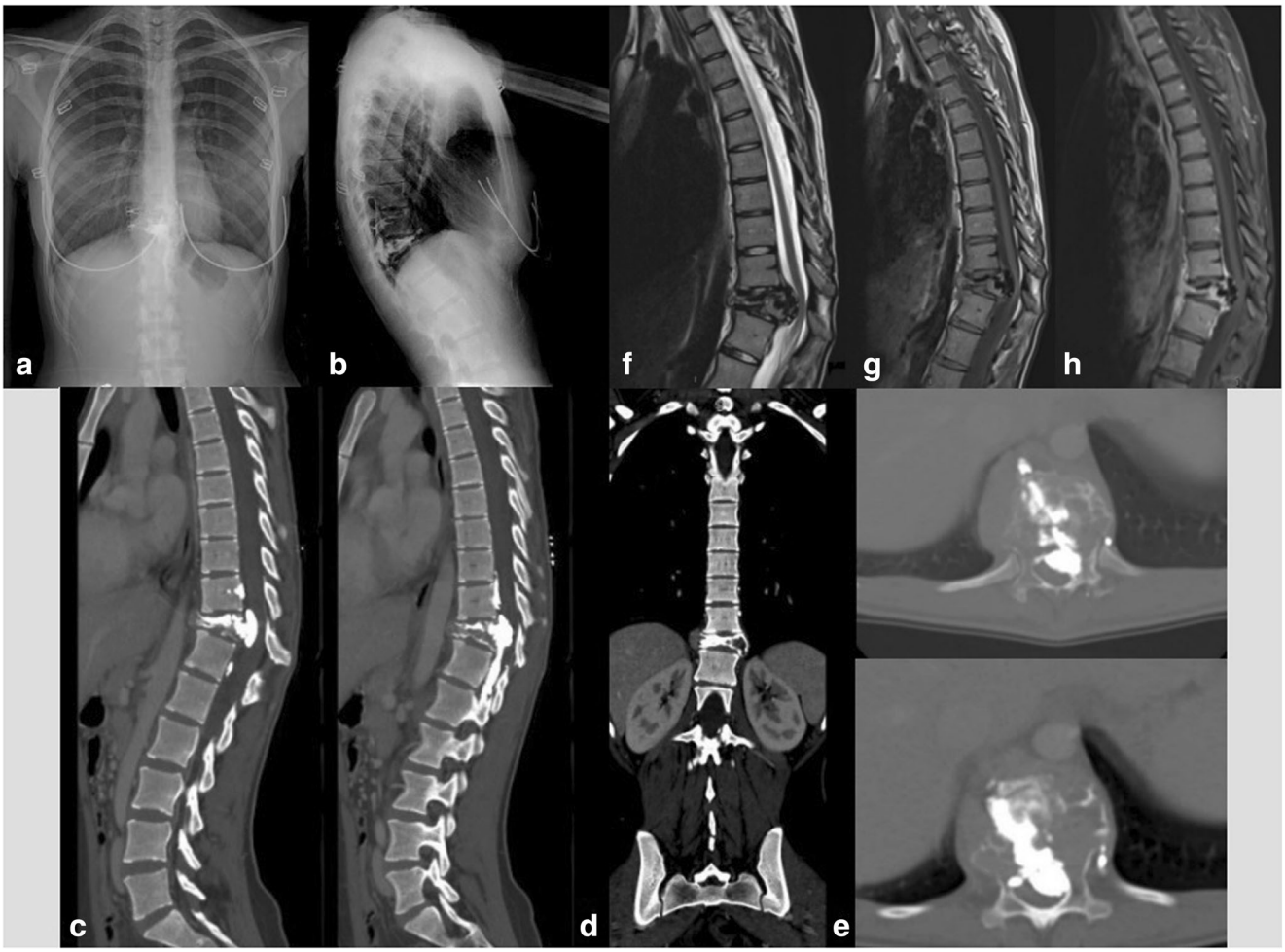

Fig. 1 Anteroposterior (**a**) and lateral (**b**) thoracolumbar radiographs showing cement appearance extending from the corpus into the spinal canal with T11 vertebra compression fracture. Sagittal (**c**), coronal (**d**), and axial (**e**) spinal computed tomography scans revealing a lytic lesion in T11 vertebra corpus and cement extravasation into the spinal canal. T1-weighted (**f**), T2-weighted (**g**), and contrast enhanced (**h**) sagittal spinal magnetic resonance imaging scans revealing a heterogeneous mass lesion and cement extravasation extending from the T11 vertebra corpus into the spinal canal. The tumor showed heterogeneous contrast enhancement

vertebra corpus into the spinal canal. The tumor showed heterogeneous contrast enhancement (Fig. 1f–h). Because the patient had neurological deterioration due to spinal canal compression, total en bloc spondylectomy, decompression of the spinal canal with cement excision, instrumentation, and fusion procedures were planned.

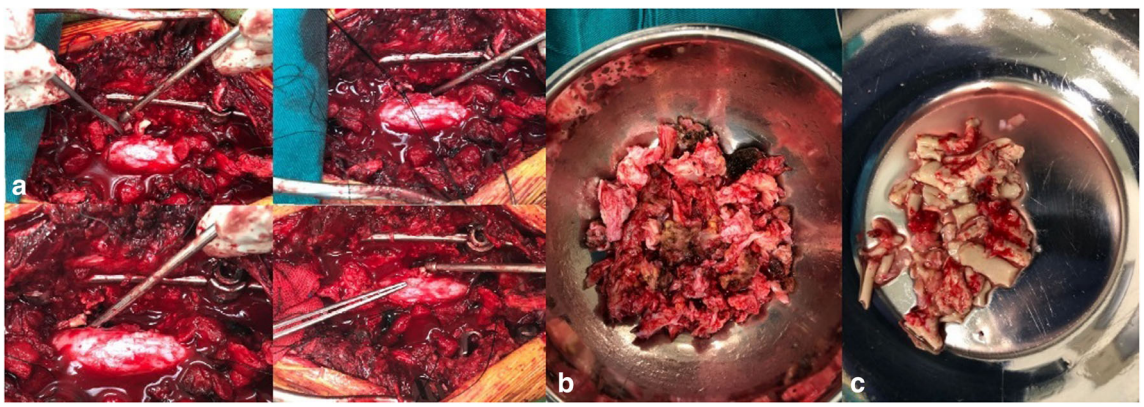

Fig. 2 **a** Excision of vertebroplasty cement compressing spinal cord and right-sided T11 root. Sacrification of right-sided T11 root. Tumor (**b**) and cement (**c**) samples which were resected in the operation

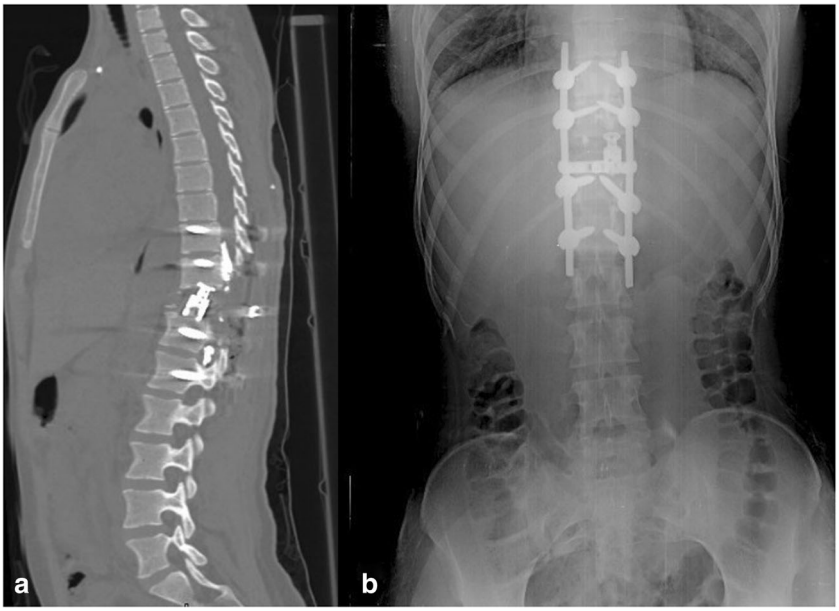

Fig. 3 Postoperative sagittal spinal computed tomography (**a**) and anteroposterior thoracolumbar radiograph (**b**) images

Total en bloc spondylectomy and gross total tumor excision were performed via posterior approach. The cement extravasation was gradually excised through the spinal canal (Fig. 2c). Decompression of the spinal canal was observed. T11 roots were sacrified bilaterally (Fig. 2a). Posterior stabilization via transpedicular screw application and anterior corpectomy cage implementation were performed (Fig. 3). Received tumoral samples underwent histopathological examination.

Histopathological examination of the tumor revealed a lesion characterized by numerous multinuclear giant cells and monocytoid histiocytes bearing 20–40 nuclei in the fibrous stroma as well as areas of connective tissue (Fig. 4). Pathological diagnosis was giant cell tumor of T11 vertebra.

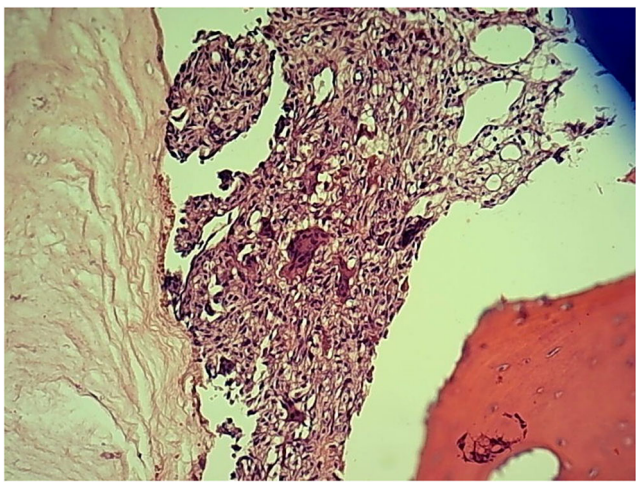

Fig. 4 Multinuclear giant cells and monocytoid histiocytes bearing 20–40 nuclei in the fibrous stroma as well as areas of connective tissue (H&E, 200 × magnification)

Postoperative course of the patient was uneventful and she began to walk without any assistance after rehabilitation with physiotherapy applications. Her postoperative follow-up continues with 3-month intervals.

Discussion

Giant cell tumors were first described by Cooper and Travers in 1818 [1, 2]. Giant cell tumors are so named because they appear histologically as nodules of osteoclast-like giant cells, from which they arise. Osteoclasts express the receptor activator of nuclear factor kappa B ligand (RANKL) which is an essential mediator for osteoclast survival. They are seen as osteolytic and destructive lesions on plain radiographs. Spinal dynamic graphs are valuable in terms of evaluating tumor-induced instability in the spinal column. Spinal computed tomography detects vertebra destruction and mass lesion infiltrating the vertebra. Spinal magnetic resonance imaging is the gold standard for imaging spinal giant cell tumors. Giant cell tumors are hypointense on T1-weighted images, hyperintense on T2-weighted images, and show heterogeneous contrast enhancement with gadolinium [7–10]. Differential diagnosis of these tumors includes brown tumors secondary to hyperparathyroidism, spinal metastases, hematological malignancies, chordoma, and aneurysmal bone cysts [2].

The main purpose of the treatment of giant cell tumors is gross total resection of the tumor. However, total resection is not always possible in spinal giant cell tumors. Adjuvant treatments are also included in these types of tumors. These treatment options include total en bloc spondylectomy (TES), total en bloc resectomy (TER), intralesional resection (IR),

Table 1 Data of the pediatric patients with giant cell tumors in the literature

Authors, year	Age, years/gender	Presenting symptoms	Location	Surgery	Adjuvant therapy	Outcome
Martin et al. [14], 2010	13, F	Pain and weakness	Sacrum	IR	RT, CT	Recurrence (9 months)
Martin et al. [14], 2010	13, F	Pain and weakness	C6	TER	RT, EMB	Recurrence (4 times)
Boriani et al. [11], 2012	15, F	Pain	T12	TER	NA	NED (48 months)
Alfawareh et al. [15], 2015	13, F	Incidental*	C2	TES	NA	NED (24 months)
Kim et al. [16], 2015	14, F	Pain	Sacrum	IR	RT	NED (92 months)
Kim et al. [16], 2015	16, F	Pain	T12	IR	RT	NED (52 months)
Ma et al. [17], 2016	15, F	Pain	Sacrum	IR (FO) TER (SO)	Denosumab	NED (86 months)**
Sigwalt et al. [5], 2016	14, F	Pain	T5	TER	NA	NED (24 months)
Present case	15, F	Pain and weakness	T11	TES	NA	NED (3 months)

*During the preoperative evaluation for idiopathic scoliosis

**No evidence of disease after second operation

IR, Intralesional resection; *TES*, total en bloc spondylectomy; *TER*, total en bloc resectomy; *FO*, first operation; *SO*, second operation; *RT*, radiotherapy; *CT*, chemotherapy; *NA*, not available; *NED*, no evidence of disease

denosumab application as an anti-RANKL monoclonal antibody, radiotherapy (RT), and embolization. In the literature, the patients who underwent TES had lower recurrence rates and better prognosis than the patients who underwent IR [7–9, 11]. However, IR and adjuvant treatments are applied especially for tumors of sacral location and larger size which may cause morbidity and mortality [9, 12].

Percutaneous vertebroplasty is an accepted procedure in patients with spinal tumors according to the literature. Successful results are obtained especially in cases with spinal metastasis [13]. However, percutaneous vertebroplasty is not an accepted procedure in the treatment of giant cell tumors. As a matter of fact, we did not also get a good result. It should be kept in mind that surgical exploration and immediate decompression of the spinal canal are required when vertebroplasty cement extravasation is detected.

According to our literature review, we found that spinal giant cell tumors are much less common in patients in the pediatric age group. Our patient is the ninth pediatric case in the literature (Table 1).

Conclusion

Giant cell tumors of the mobile spinal column are very rare tumors, especially in the pediatric age group. The main purpose of the treatment is gross total resection of the tumor with TES. In patients who underwent TES, the risk of recurrence decreases and the prognosis are much better in these patients. Adjuvant methods such as denosumab should be added to the treatment protocol in patients who cannot undergo gross total resection due to tumor size and anatomic localization. However, denosumab is not recommended in pediatric patients younger than age of 4 years because of the high rates of skeletal growth and the potential for denosumab to negatively affect long-bone growth and dentition. Treatment with denosumab may impair bone growth in children with open growth plates and may inhibit eruption of dentition.

Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interest.

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