Clinical Case Report

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Primary Ewing's sarcoma of the vertebral body

A case report

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Abstract

Rational: The occurrence of Ewing's sarcoma in the vertebral body of elderly women is extremely rare, and the case of Ewing's sarcoma in the spine with secondary surgical repair after wrong diagnosis and treatment has not been reported. We report a case involving primary Ewing's sarcoma of the vertebral body in an elderly female. Owing to its rarity and controversial issues, we report a case report to discuss its clinical features, treatments, radiological, and histological characteristics.

Patient concerns: The elderly female patient came to see us with the manifestation of total paralysis of both lower limbs. The patient with a vertebral compression fracture as the primary manifestation was misdiagnosed in another hospital. The patient underwent inappropriate surgical treatment and was transferred to our hospital for diagnosis and second-stage surgery.

Diagnoses: The postoperative pathological examination and immunohistochemical examination in our hospital confirmed: Ewing's sarcoma; Surgical history at other hospitals suggests: after Bone cement injection.

Interventions: The patient underwent a T6 and T8 laminectomy and T5/6-T9 pedicle screw fixation.

Outcomes: Reexamination 1 month after the surgery showed that the tumor had been partially resected, the spinal cord compression was relieved, the tumor did not grow further, and the patient's lower limb physical ability, tactile sense, algesia and temperature sense recovered slightly.

Lessons: For patients with ewing's tumor in the spinal canal with symptoms of spinal cord compression, even if the patients with poor results after a unadvisable operation, it is still necessary to be actively in spinal cord compression by surgery. The differential diagnosis of Ewing's sarcoma and compression fractures is very important. For patients with vertebral tumors, special attention should be taken during vertebroplasty for bone cement leakage caused by excessive bone cement injection and increased local pressure. And some experience with imaging and laboratory findings.

Abbreviations: ALT = glutamic-pyruvic transaminase, AST = glutamic oxalacetic transaminase, CRP = C-reaction protein, CT = computed tomography, DD = D-Dimer, ES = Ewing's sarcoma, ESR = erythrocyte sedimentation rate, GGT = gamma-glutamyl transpeptidase, LDH = lactate dehydrogenase, MRI = magnetic resonance imaging, SF = transferrin, T6 = the sixth thoracic, T7 = the seventh thoracic, T8 = the eighth thoracic, T9 = the ninth thoracic, WBC = white blood cell.

Keywords: bone tumor, elderly female, Ewing's sarcoma, vertebral body

1. Introduction

Ewing's sarcoma is a neuroectodermal tumor that mostly occurs in children and adolescents and is characterized by rapid growth and development. Ewing's sarcoma is more common in males than females. The long bones are most often affected, but Ewing's sarcoma are also known to occur in the humerus, scapula, pelvis, and other flat bones.[1] Shoubash et al[2] reported a case of primary Ewing's sarcoma in an elderly woman in June 2018, but such cases with tumor originating in the vertebral body with further enlargement and involvement of the spinal canal in an elderly woman is very rare. Referring to existing literature, there are no similar reports of primary tumors in the lower thoracic spine. The case of Ewing's sarcoma in the spine with secondary surgical repair after wrong diagnosis and treatment has not been reported. In this paper, we introduce the experience of treating such an atypical patient.

2. Case report

The patient is a 62-year-old female with a compression fracture of the 7th thoracic vertebra that was diagnosed in March 2017 (Fig. 1. The diseased T7 vertebral body can be clearly seen). The patient underwent a T7 percutaneous vertebroplasty in another hospital in September 2017. The bone cement overflowed from the posterior edge of the vertebral body after surgery, followed by T7 laminectomy, and T6-T8 pedicle screw fixation (Fig. 2. Nine months after surgery in another hospital, we can see the vertebral

Editor: N/A.

Duo-Yi Zhao and Wei-Lin Zhang have contributed equally to this work.

The Science and Technology Program of Liaoning Province, No. 20170541058.

The authors have no conflicts of interest to disclose.

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Medicine (2019) 98:24(e15905)

Received: 16 January 2019 / Received in final form: 20 April 2019 / Accepted: 9 May 2019

http://dx.doi.org/10.1097/MD.0000000000015905

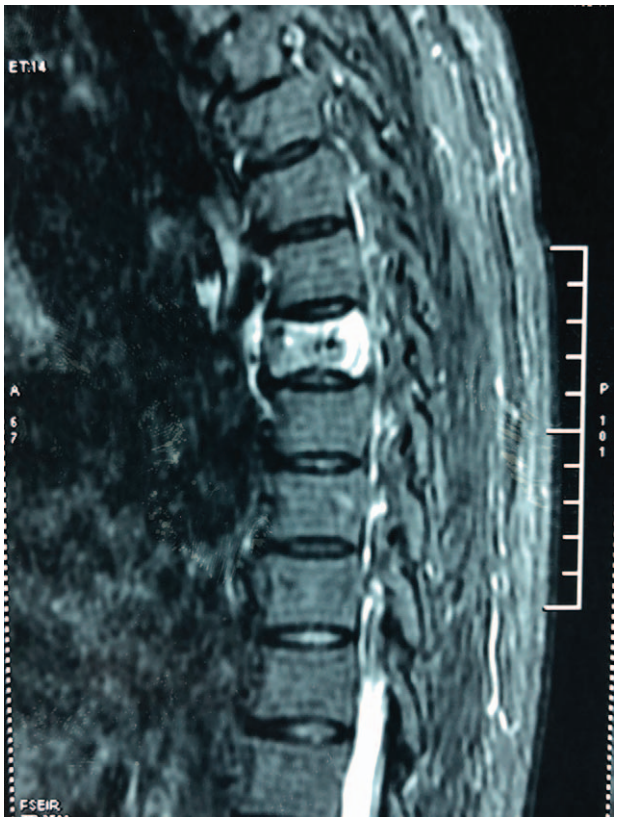

Figure 1. Pre-operative thoracic spinal MRI shows compression fractures of T7, with central cavity of the vertebral body and high signal of local soft tissue.

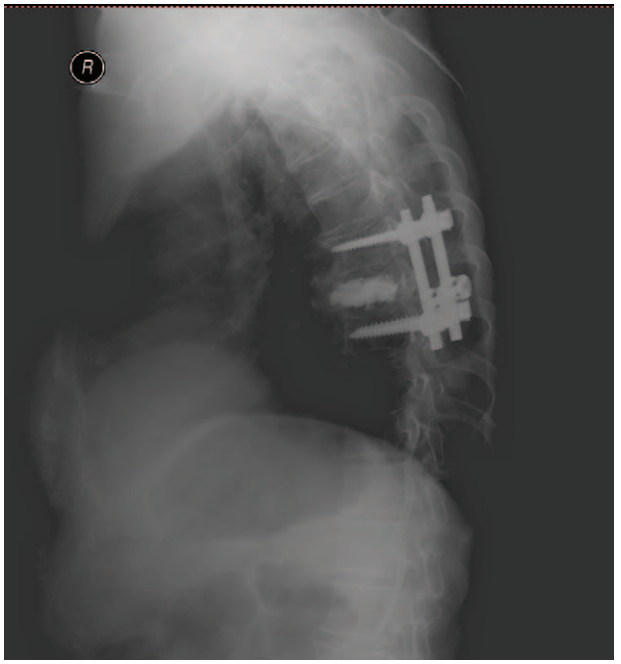

Figure 2. Chest lateral DR, 9 months after surgery in another hospital shows visible T7 vertebral bone destruction and bulging to the surrounding.

Figure 3. Pre-operative 3D-CT of thoracic vertebrae in our hospital shows T7 flattening and visible hyperplasia of surrounding soft tissues and bone.

bone expands destroy, surrounding soft tissue infiltrates, and internal fixation treatment). In April 2018, the patient developed numbness and pain in the chest and back. In June 2018, the patient developed lower extremity paralysis and incontinence, and was referred to our hospital. The specialist examination revealed the following: pain and temperature sensation below the 12th rib and the abdominal wall reflex were absent; lower limb muscle strength was 0; the knee palpebral and anus reflexes were absent; positive Babinski sign; and sputum test was positive. The laboratory findings were as follows: WBC, $12.4*10^9$/L; ESR, 39 mm/h; CRP, 15.6 mg/L; DD, 611 ng/mL; ALT, 52 U/L; AST, 51 U/L; GGT, 86 U/L; LDH, 332 U/L; and SF, 519 ng/mL. Imaging was significant for the following: X-ray (T7 vertebral bone destruction with bulging); 3D-CT (Fig. 3, Pre-operative 3D-CT in our hospital, the relationship between bone, fixation and local tissue was defined); MR (Fig. 4, The destruction of the vertebral body, the tumor protrudes into the posterior spinal canal, causing compression on the spinal cord can be clearly seen); Enhanced MR (irregular enhancement signal of T7 vertebral body and accessory with involvement of the spinal cord); head MRI (lacunar cerebral infarction); and total abdominal CT (no abnormal density image of the liver with splenomegaly). The patient underwent a T6 and T8 laminectomy and T5/6-T9 pedicle screw fixation. Tumor-like tissue was noted in the spinal canal of T6-T8 with spinal cord compression intra-operatively. Cone plate decompression showed that the T6 and T8 cone plates were sparse and brittle. The pathologic results suggested Ewing's sarcoma (Fig. 5. Post-operative pathology: Ewing's sarcoma). The immunohistochemistry results were as follows: CgA(−); Syn (−); CD56(−); CD99(++); EMA(−); GFAP(−); IDH1(−); p53 (+5%); SOX-10(−); CK(++); Vimentin(+); Ki-67(+20%); CD34

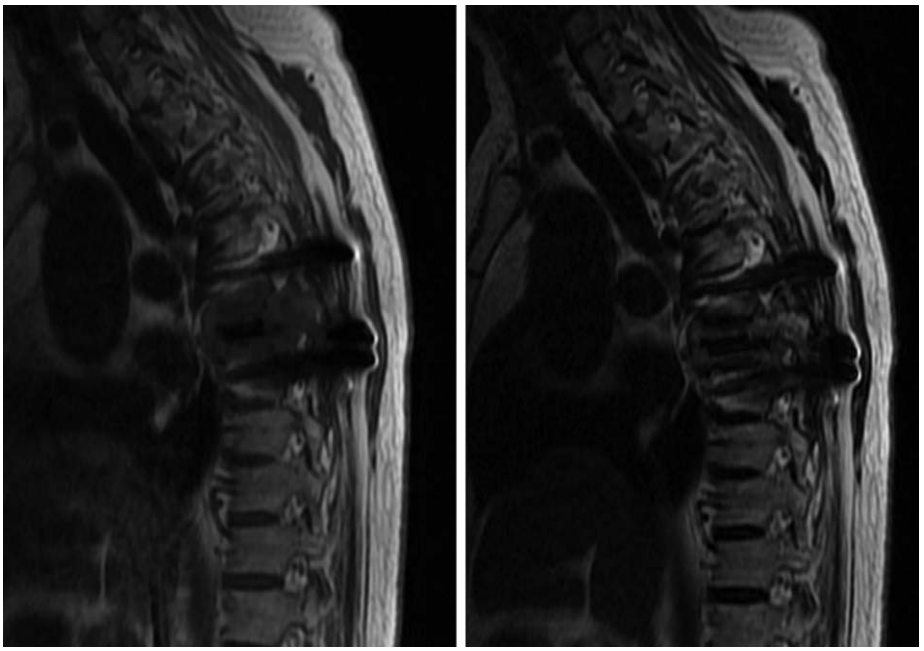

Figure 4. Pre-operative thoracic MRI in the lateral position, T1W1 low-signal shadow, T2W1 high-signal shadow, showing T7 vertebral destruction, bulging to the periphery, patchy mixed signal in and around the vertebral body, spinal cord distortion, and an intramedullary abnormal signal.

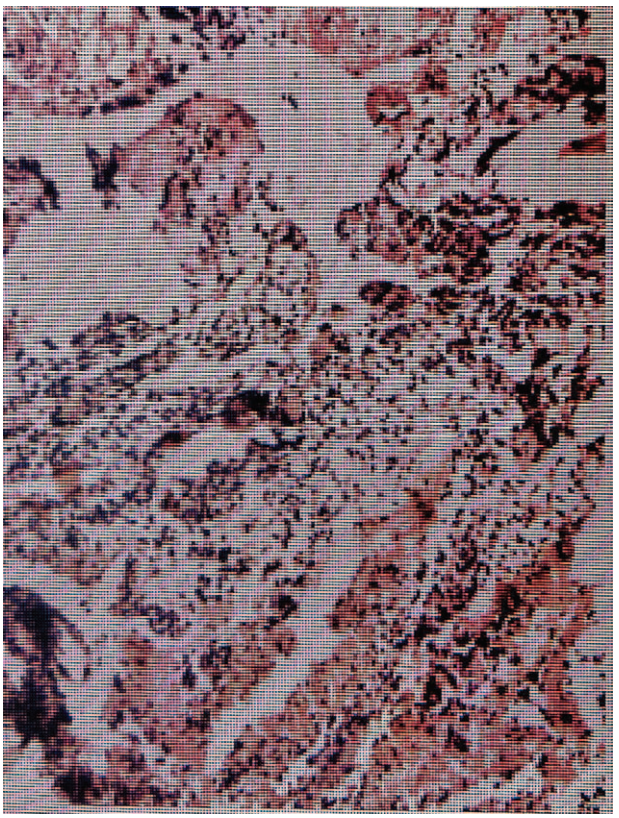

Figure 5. Microscopically, a large number of single, diffuse, dense small round cells were observed. Cytoplasm is few, nucleus is round, and staining is deep.

(−); S-100(−); and LCA(−). Partial skin sensation of both lower limbs was restored 3 days after the surgery, and partial muscle strength of lower limbs was restored 1 month after the surgery. MRI images 1 month after surgery are shown in the figure (Fig. 6. The tumors that compressed the spinal canal in the rear have been cleared, the spinal cord compression has been relieved, and the tumor scope in the front has not been further expanded). At 3 months after the operation, the muscle strength of both lower limbs increased further, and some of them could move autonomously on the bed.

Ethics approval was not required for this paper as it is a case report. Informed consent was obtained from the patient for publication of this case report and accompanying images.

3. Discussion

The case reported by Shoubash et al[2] was a primary treatment case. The patient underwent first-stage laminectomy, post-operative radiotherapy and chemotherapy, and second-stage vertebral resection. In our case, the thoracic vertebral compression fracture was the primary manifestation. After misdiagnosis and inappropriate treatment at another hospital, the tumor continued to expand and involved the affected spine segment, which limited treatment options and increased the difficulty of second-stage surgery in our hospital. Due to local post-operative changes of soft tissue, the separation of tumor tissue requires great careful. However, even if the operation is very difficult, and the patient had pre-operative symptoms of paralysis of both lower limbs for 1 month, we still believe that surgical treatment is very necessary. The results of post-operative follow-up showed that patients benefited from this positive surgical choice.

Belkoff et al[3–6] believe that local injection of bone cement can kill tumor cells in the vertebral body via local high temperature, cytotoxicity, and blood supply destruction, but in the current case

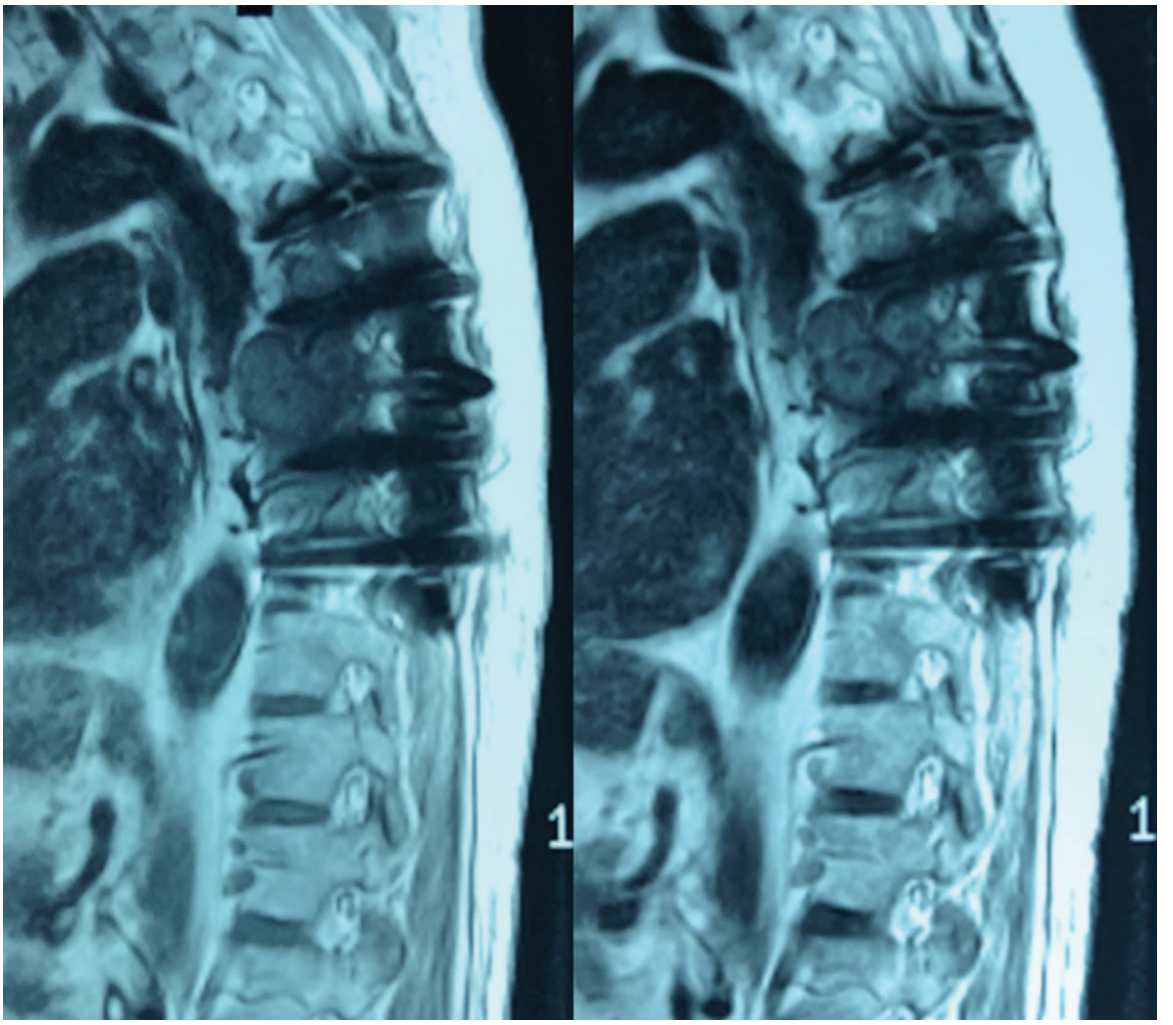

Figure 6. Post-operative thoracic MRI, showing the tumors that compressed the spinal canal in the rear have been cleared, the spinal cord compression has been relieved, and the tumor scope in the front has not been further expanded, internal fixation was performed again for stabilization.

vertebral injection of bone cement in the first stage failed to effectively inhibit the growth of the primary Ewing's sarcoma. In contrast, increased local pressure in the vertebral body led to passive expansion of the primary tumor and bone cement leakage at the posterior margin of the vertebral body. Such a result reminds us of the importance of choosing bone cement injection during vertebroplasty. For patients with vertebral tumors, special attention should be taken during vertebroplasty for bone cement leakage caused by excessive bone cement injection and increased local pressure.

This case further reminds us of the importance of the differential diagnosis of Ewing's sarcoma and compression fractures. For compression fractures of the spine, we must consider the cause with thorough local soft tissue examination before surgery and pay attention to eliminate the possibility of a tumor. In this case, the tumor marker (SF) level was elevated, which is suggestive of a Ewing's sarcoma. The patient had abnormal liver function, with no abnormal liver areas on total abdominal CT or hepatitis virus infection, as suggested by hepatitis screening. Statistical analysis of more cases is needed to explore the relationship between liver function abnormalities and the occurrence of Ewing's sarcoma.

Author contributions

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Software: Duoyi Zhao, Shenghui Ni.
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Visualization: Duoyi Zhao, Yan Cui.
Writing – original draft: Duoyi Zhao, Weilin Zhang, Chengbin Ma.

Writing – review & editing: Duoyi Zhao, Weilin Zhang, Zhiyu Zhang.

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