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CASE REPORT

Giant cell tumor of the rib with direct invasion into the thoracic spine

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Abstract Giant cell tumors of bone are relatively rare, benign, but locally aggressive osteolytic skeletal neoplasms of young adults. They usually affect the epiphyses of long bones, especially around the knee joint, and are rarely seen in the ribs. The mainstay of therapy is surgical resection. Herein, we report a case of successful resection in a patient who presented with primary giant cell tumor of the rib, directly invading the thoracic spine. Computed tomography and magnetic resonance imaging were helpful for assessing the depth of tumor invasion. Radical resection of the tumor and reconstruction of the vertebrae with preserved allograft bone were performed. No respiratory or neurological problems occurred, and the patient remained well 2 years after surgery.

Keywords Chest wall · Giant cell tumor · Reconstruction

Introduction

Giant cell tumors (GCTs) of bone account for 5 % of all primary bone tumors [1]. Most occur in patients aged 20–45 years [2], and are located mainly in the proximal tibia, humerus, distal radius, and pelvis, with only 0.3–0.6 % of primary tumors occurring in the ribs [1–3]. There have been some reports of surgical resection of primary or recurrent chest wall GCTs [1–4], but successful resection of a primary GCT invading the thoracic spine is very rare. Herein, we report a case of successful resection and disease-free survival in a patient who presented with GCT of the rib directly invading the thoracic spine.

Case

A 38-year-old man with a 2-month history of back pain and cough was referred to our department. There was no significant medical history, previous radiotherapy or chemotherapy, or Paget's disease. Chest computed tomography (CT) and magnetic resonance imaging (MRI) showed a heterogeneous mass arising from the fourth rib, directly invading the third and fourth thoracic vertebrae, and extending into the spinal canal through the intervertebral foramina (Fig. 1). ^{18}F-fluorodeoxyglucose (FDG) positron emission tomography (PET) demonstrated uniform, strong FDG accumulation (max standardized uptake value 8.39) at the primary tumor site. There was no other FDG accumulation suggestive of metastases.

Histological examination of a core needle biopsy specimen was suggestive of osteosarcoma. Based on the imaging results, the tumor was preoperatively diagnosed as a primary osteosarcoma of the rib, which was considered to be resectable together with the thoracic cage and vertebral body, under posterior spinal fusion. An adequate surgical margin was considered preoperatively to be at the level of the fourth rib angle (about 5 cm from the tumor), with the surrounding intercostal muscles and the entire bodies of the third and fourth vertebrae. We decided to perform radical

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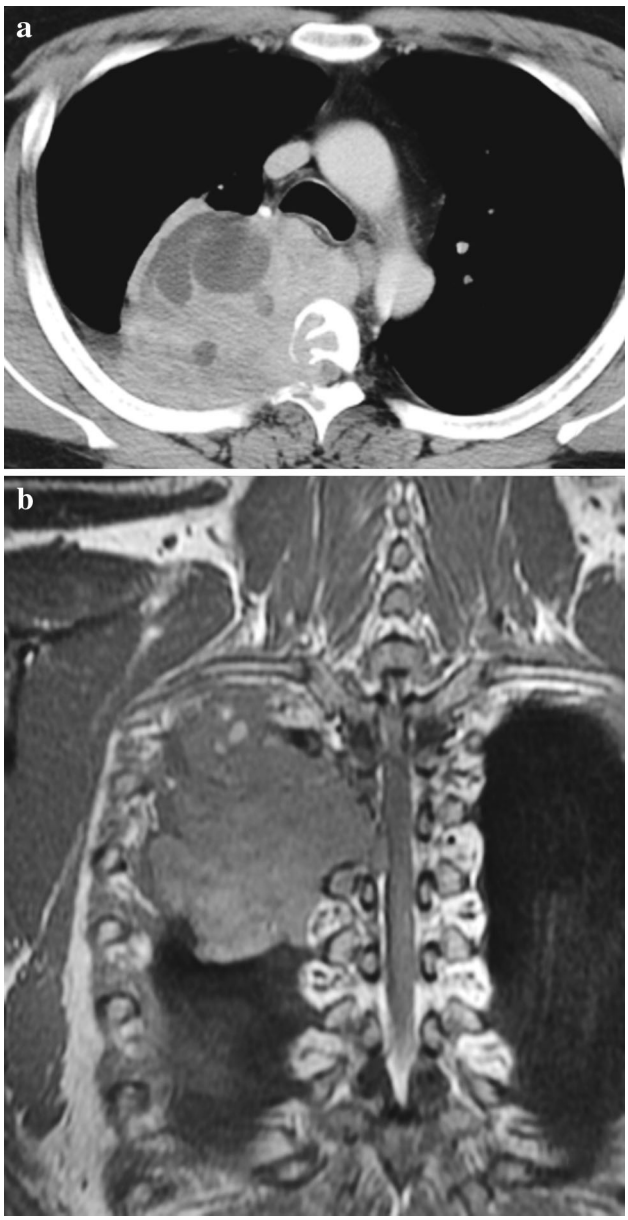

Fig. 1 **a** CT scan showing a heterogeneous structure in the fourth rib invading the third and fourth thoracic vertebrae. **b** Axial MRI demonstrating extension of the mass into the spinal canal through the intervertebral foramina

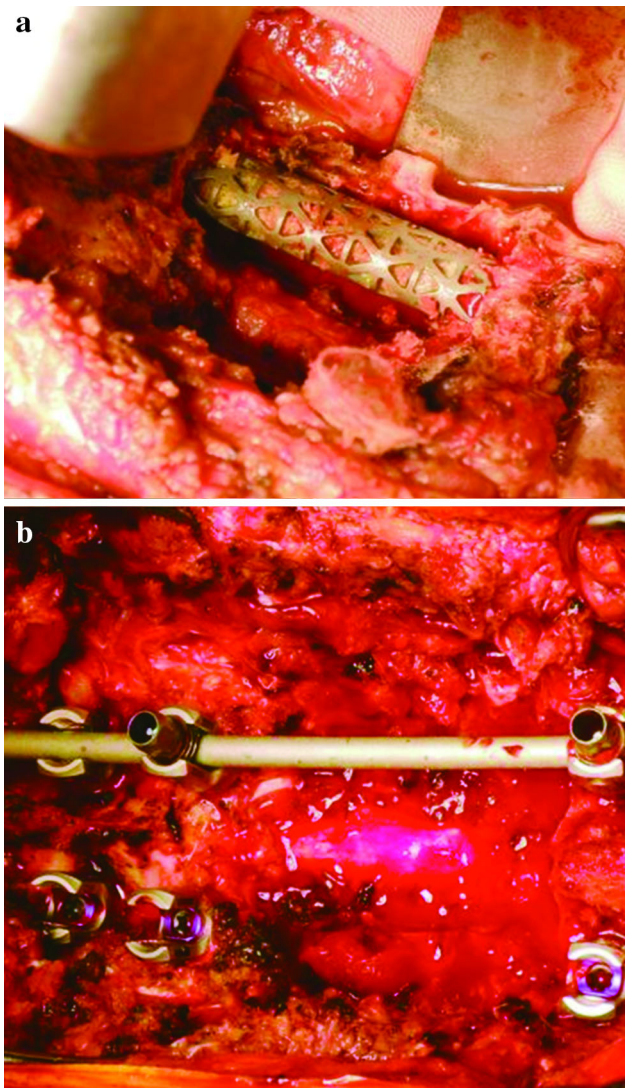

Fig. 2 **a** Surgical titanium cages filled with preserved allograft bone were placed for vertebral reconstruction. **b** Posterior thoracic spinal fusion was performed using surgical titanium rods

resection immediately, because of the increasing risk of spinal nerve compression due to tumor enlargement.

The surgery was performed in two stages. The first stage involved insertion of pedicle screws to Th1, 2, 5, and 6, and laminectomies at lower Th2 to upper Th5 via a posterior approach in the prone position, followed by resection of the transverse processes at levels Th3 and Th4. This procedure resulted in decompression of the spinal nerve. After dissection of the bilateral third and fourth intercostal nerves, fixation was performed by inserting screws and titanium rods (Fig. 2).

The second stage of the procedure involved radical resection of the Th3 and Th4 vertebral bodies and the fourth rib, together with the adjacent intercostal muscles at the level of the rib angle and part of the right upper lung lobe via a posterolateral approach in the lateral position, guided by the preoperative CT and MRI findings. To obtain as wide surgical margins as possible, most of the Th3 and Th4 vertebral bodies were removed, except for the left side periosteum and the spinal dura mater with surgical airtome. The fifth rib was dissected and reconstructed using absorbable costal coaptation pins. The Th3 and Th4 vertebrae were then reconstructed using cages filled with preserved allograft bone (Fig. 2) previously collected from patients who had undergone other orthopedic surgeries in our hospital. The total operation time was 1175 min and blood loss was 3724 ml.

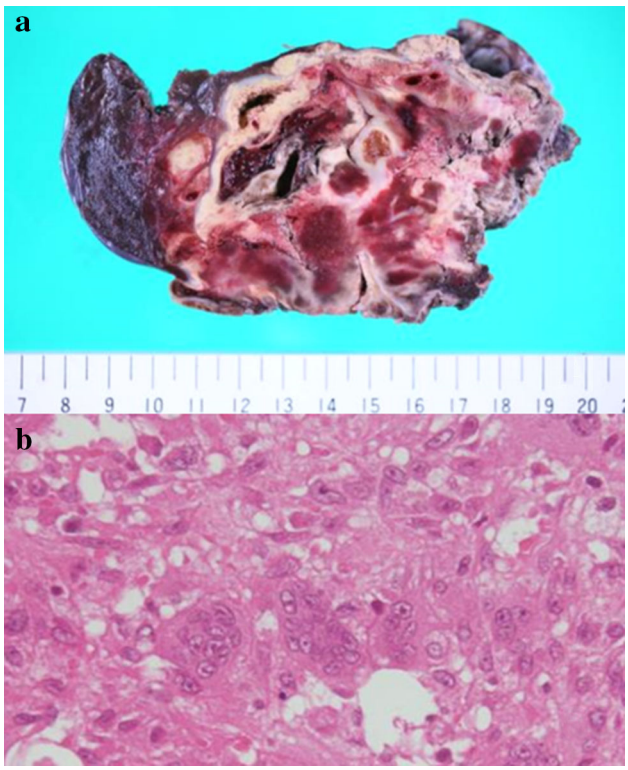

Fig. 3 **a** The photograph shows the cut-surface of the resected tumor. The surface was composed of white tumor, yellowish-white necrotic tissue and hemorrhage area. **b** Microscopic findings revealed diffuse sheets of mononuclear or multinuclear giant cells with no prominent nuclear atypia

Transfusions of 1680 ml concentrated red blood cells and 480 ml fresh frozen plasma were needed during surgery.

No signs of allograft rejection were observed after surgery, but the patient required opioids for severe postoperative pain. The patient was discharged at postoperative day 25 after rehabilitation and physiotherapy, with no respiratory or neurological problems. The resected tumor measured 12 × 11.5 cm. Histological examination revealed destructive and infiltrative growth of diffuse sheets of mononuclear or multinuclear giant cells with no significant nuclear atypia or definitive osteoid formation in the resected specimens (Fig. 3). According to these findings, we revised the diagnosis as GCT originating from the rib. The surgical margins of the fourth rib and right upper lobe were negative, but there was a risk of tumor cell contamination around the spine. The patient remained well 2 years after surgery, with no recurrence.

Discussion

GCTs of bone are generally regarded as benign (despite exceptional metastasis), but locally infiltrative tumors involving ligaments, synovia, or adjacent bone [2, 3]. Although the epiphyses of long bones are the most common site of involvement, GCTs may also occur in the metaphysis and in membranous bone [3]. GCTs in a rib usually develop in the epiphysis of the head or tubercle [1]. Radiographic evaluation with chest radiographs and CT scans are essential for identifying and characterizing chest-wall masses, while MRI is useful for assessing the depth of tumor invasion into the spine [5]. In our case, preoperative CT and FDG-PET were useful for ascertaining the absence of apparent metastases.

Some previous reports have reported on the surgical resection of primary or recurrent chest wall GCTs [1–4], but successful resection of a primary GCT invading the thoracic spine is very rare. Although curative resection is a valid and feasible strategy for the treatment of GCTs, the ability to achieve an optimal outcome and functional preservation depends on the location and extent of the tumor [4]. Axial GCTs, particularly in the sacrum, are associated with a poor prognosis because the tumors tend to be larger at diagnosis and more difficult to excise completely [5, 6]. In our case, the primary tumor was located in the fourth rib and directly invaded into the third and fourth thoracic vertebrae. We performed radical resection of the tumor and reconstruction of the vertebrae with surgical titanium mesh filled with preserved allograft bone and titanium rods, with no respiratory or neurological sequelae. In the case of osteoid tumors, it is generally difficult to decide on a safe surgical margin based on intraoperative frozen sections, and extensive resection should be performed, with as wide margins as possible, based on preoperative imaging findings. A previous report showed that total excision of tumors, including the capsule, should be performed en bloc or intralesionally [7, 8]. The current case suggests that surgical resection and reconstruction can be successful and should be attempted for GCTs involving the thoracic cage, and that achieving tumor-free margins can lead to a good prognosis.

Conclusion

Intensive resection should be considered for the treatment of GCTs in the ribs, even if extensive reconstruction is required. The current case suggests that successful surgical resection with tumor-free margins and reconstruction can lead to a good prognosis.

Compliance with ethical standards

Conflict of interest The authors have declared that no conflict of interest exists.

Informed consent Written informed consent was obtained from the patient for publication of this case report and accompanying images.

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