

Giant cell tumor expanded into the thoracic cavity with spinal involvement

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Title

A case report of a giant cell tumor that had expanded into the thoracic cavity and had spinal involvement

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Abstract

We report on a case of giant cell tumor (GCT) that had expanded into the thoracic cavity as well as through the spinal canal into the vertebra. A 36-year-old male presented with a six-month history of back pain and dyspnea. Plain chest radiographs showed a huge mass accompanied by right pleural effusion. The mass also involved the 12th thoracic spine and the spinal cord was severely compressed. To resect the tumor, we carried out a two-stage procedure. As a first stage, to separate the tumor from the anterior vital structures under direct vision, a right thoracotomy with chest wall reconstruction, from the 8th to the 11th rib, was carried out by thoracic surgeons. The right lung and inferior vena cava were gently retracted, and the tumor was carefully detached from these structures. We were not able to separate the tumor from the right diaphragm because of severe invasion; therefore a partial resection of the right diaphragm with the tumor was performed. After excision of the anterior part of the tumor, the thoracic wall was reconstructed with the right 8th rib and Marlex mesh. After improvement of the patient's general condition two weeks later the second stage of the procedure was carried out: spondylectomy by posterior approach was performed.

In conclusion, we achieved excision of a GCT that had expanded into the thoracic cavity as well as through the spinal canal into the vertebra. The patient achieved full rehabilitation with no neurological or respiratory abnormalities at 7 years after the surgery.

Introduction

Giant cell tumor (GCT) is a benign primary bone tumor that usually occurs in the epiphysis of the long bones but rarely in the axial skeleton. It is found most commonly after skeletal maturity, and shows a slight female predominance.^{2,4,10} Histologically, GCT consists of multinucleated osteoclastic giant cells in the stroma of spindle-shaped cells.¹⁷ Because it is typical for a GCT to be aggressive locally, curettage alone can lead to a high rate of local recurrence.^{1,2} In the extremities, it has been reported that the combination of adjuvant therapy reduces the rate of local recurrence.^{1,5,8,9} When the spine is involved, these adjuvant therapies present the risk of postoperative complications because of the presence of major blood vessels and the spinal cord. Therefore, complete excision of GCT is generally considered to be the best treatment option. However, due to the difficulties of removing the spinal tumor, a complete resection is often not a feasible option for GCT especially when it has expanded into the thoracic cage. We report on a case of a GCT that had expanded into the thoracic cavity and had spinal involvement.

Case report

A 36-year-old male presented with a six-month history of back pain and dyspnea. A week before our work-up, the patient had worsening of dyspnea and sought medical attention at his primary hospital. After examination, he was referred to our institute due to an abnormal shadow on chest radiographs. On admission, he complained of dyspnea at rest and physical examination showed diminished right breath sounds. An arterial blood gas revealed pH 7.41, pCO₂ 46 mmHg, pO₂ 66 mmHg, and an oxygen saturation of 93% on room air. There were no neurological deficits in his lower limbs. Plain-chest radiographs showed an atelectatic region in the right thoracic cavity (Figure 1A). In addition, disappearance of the right 12th rib as well as absence of the right pedicle shadow at T12 was observed. Computed tomography (CT) revealed a large mass in the right hemithorax accompanied by pleural effusion. Due to the mass, the right lung was severely compressed, and the mediastinum was shifted to the left. The mass also involved the 12th thoracic vertebral body, right pedicle and lamina (Figure 1B). A coronal MRI showed a large mass (25 cm x 12 cm) that enhanced heterogeneously by gadolinium (Figure 2A). Invasion of the tumor mass into the spinal canal through the vertebra was also

observed (Figure 2B). Further investigation revealed that there were no other tumor lesions. A biopsy was performed, and the histological diagnosis was giant cell tumor.

To resect the tumor, we carried out a two-stage procedure. As a first stage, to separate the tumor from the anterior vital structures under direct vision, a right thoracotomy with chest wall reconstruction, from the 8th to the 11th rib, was carried out by thoracic surgeons. There were adhesions between the tumor and the right lung, inferior vena cava, and right hemidiaphragm. The right lung and inferior vena cava were gently retracted, and the tumor was carefully detached from these structures. We were not able to separate the tumor from the right diaphragm because of severe invasion; therefore a partial resection of the right diaphragm with the tumor was performed. After excision of the anterior part of the tumor (Figure 3A), the thoracic wall was reconstructed with the right 8th rib and Marlex mesh. The operative time was 460 minutes with a blood loss of 8700 ml during the first surgery. After the first surgery, intensive respiratory and circulatory care was performed. After excision of the anterior part of the tumor, the arterial blood gas improved to pO₂ 91 mmHg, and oxygen saturation to 98% on room air.

After improvement of the patient's general condition two weeks later the second stage of the procedure was carried out: spondylectomy by posterior approach was performed. After insertion of pedicle screws from T9 to L3, laminectomies at T11 and T12 were performed. The tumor compressed the dura matter, so the dura matter was carefully detached from the tumor to preserve neurological function. Total spondylectomy was carried out after cutting at the inferior border of the T12/L1 disc and the superior border of the T10/T11 disc using a threadwire saw (Figure 3B). The vertebral body was then reconstructed using of titanium mesh cage filled with autogenous iliac crest bone. Pathologic examination showed GCT with proliferation of multinucleate giant cells and mononuclear cells, without significant cellular atypia. A year after surgery, the patient was found to have developed a subcutaneous recurrence of the tumor at the site of the surgical wound, likely due to seeding of the site during a prior surgery. This new growth was successfully excised. Four years after the surgery, he received revision surgery with anterior reconstruction using a titanium cage and fibula strut graft due to pseudoarthrosis. Seven years after the surgery, follow-up imaging showed no signs of tumor recurrence and complete bony fusion at the reconstruction site; one of the

pedicle screws at right T9 was seen to be placed laterally (Figures 4A, 4B, 4C).

The patient achieved full rehabilitation with no neurological or respiratory abnormalities.

Discussion

GCT is a histologically benign tumor; however it often takes an aggressive course clinically. Local recurrence rates after intralesional curettage have been reported to be high, and there are some options for adjuvant treatment: these include methyl methacrylate implantation,⁸ cryosurgery,⁹ and phenolization⁵ in the extremities. However, in the spine, physical adjuvant treatments carry the risk of damaging the spinal cord, nerve roots, and/or major blood vessels. Therefore, thorough excision during the initial surgery is the best course of action for GCT in the spine in order to limit the chance of recurrence.^{1, 14-16}

There have been a few cases involving GCT occupying the thoracic cavity, (similar to our case) reported in the literature.^{3,11} In any case, the tumor was not detected until it had expanded into the thoracic cavity. Even if a large tumor is formed and lung involvement or compression occurs, it can be difficult to detect the tumor in the thoracic cavity due to the mild subjective symptoms.

Furthermore, invasion of the tumor into the vertebral body, pedicle, and lamina, made it difficult for us to excise the tumor completely. Some reports have recommended that surgical resection followed by radiotherapy may be effective since complete excision of GCT in vertebrae is difficult.^{4,11} However, several authors do not recommend radiotherapy because of the risk of sarcomatous change, which occurs in 27% of patients.² Therefore, radiotherapy for GCT might be limited as a postoperative treatment to cases of palliative treatment of postoperative recurrences or unresectable large tumors.

In our case, as our first-stage procedure we removed the tumor from the vital structures (including the lung, and inferior vena cava) by anterior approach. We did this to improve both respiratory and circulatory functions. It is recommended that a thoracic surgeon be consulted to dissect the lung from the tumor capsule and to perform chest-wall reconstruction. After stabilizing the patient's general condition, we performed total spondylectomy without damaging the spinal cord.

It is feasible to do en bloc excision for an aggressive tumor such as GCT. Harrop et al., in a review of aggressive benign tumors including GCT, recommended en bloc resection in the thoracic and lumbar spine.⁶ In our

case, we excised the tumor in two stages. The patient had a subcutaneous recurrence in the surgical wound a year after the surgery, possibly because of contamination during the surgery. However there was no recurrence at the primary site and there was no evidence of disease for six years. This good result was probably due to capsule excision of the tumor. In addition, GCT is not a malignant tumor, so preserving neurological function could be considered as a higher priority. Junming et al. discussed 22 cases of cervical GCT in which local recurrence was detected in 5 of 7 cases that underwent subtotal resection. However in only 1 of 13 cases was total excision (even intralesional) performed.⁷ Tomita et al. consider it mandatory to carry out total tumor excision including the tumor capsule, in en bloc or piecemeal fashion, for benign tumors such as the GCT.¹⁶ We believe that the total excision of the tumor including the capsule should be carried out, en bloc or intralesionally.

In conclusion, we achieved excision of a GCT that had expanded into the thoracic cavity as well as through the spinal canal into the vertebra. The patient achieved full rehabilitation with no neurological or respiratory abnormalities at 7 years after the surgery.

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Figure legends

Figure 1

- (A) Chest radiograph showing an atelectatic legion in the right thoracic cavity.
- (B) Reconstructed sagittal computed tomography. The mass involved the vertebral body, pedicle and posterior elements of T12.

Figure 2

- (A) Coronal magnetic resonance image enhanced by gadolinium showing a large mass (25 cm x 12 cm) in the thoracic cavity.
- (B) Axial T2-weighted magnetic resonance image showing invasion of the tumor mass into the spinal canal through the vertebra.

Figure 3

- (A) Resected specimen from the thoracic cavity.
- (B) Specimen from vertebral body.

Figure 4

- (A) Chest radiograph 7 years after the surgery. Atelectatic legion in the right thoracic cavity almost recovered.
- (B) Lateral radiographs of the thoracic spine.
- (C) Reconstructed sagittal computed tomography. Complete bony fusion at

reconstruction site was observed.

Figure 1A



Figure 1B



Figure 2A



Figure 2B

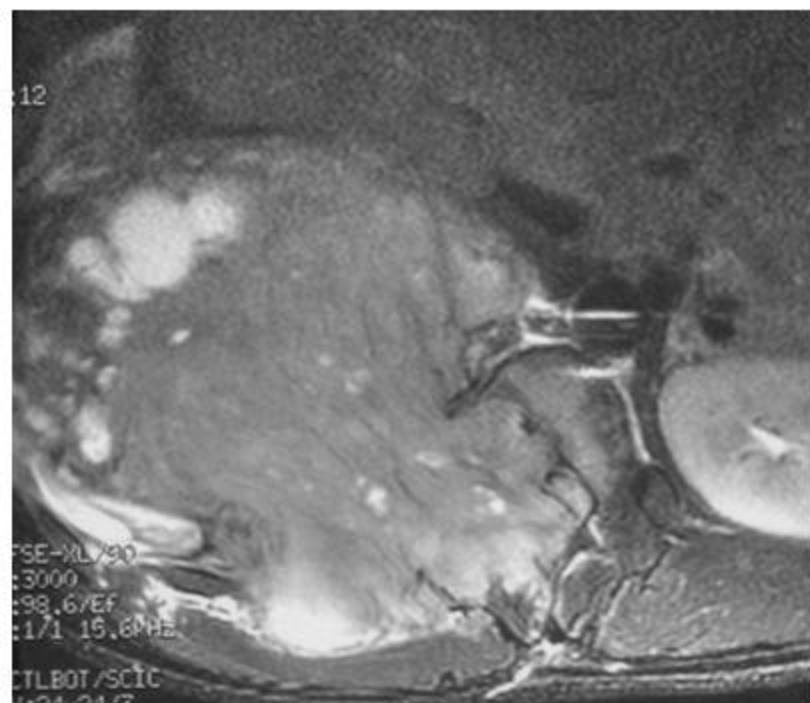


Figure 3A



Figure 3B

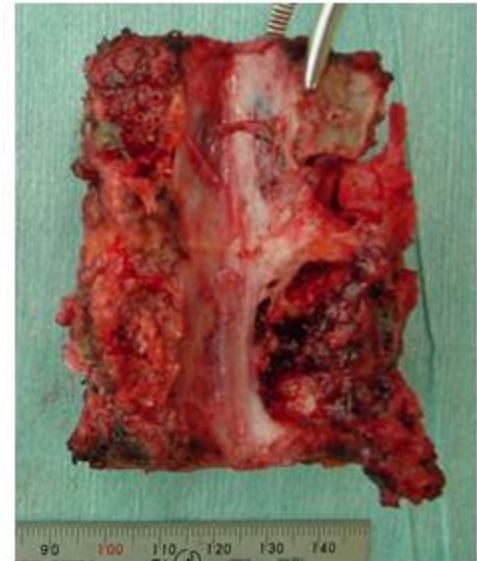


Figure 4A

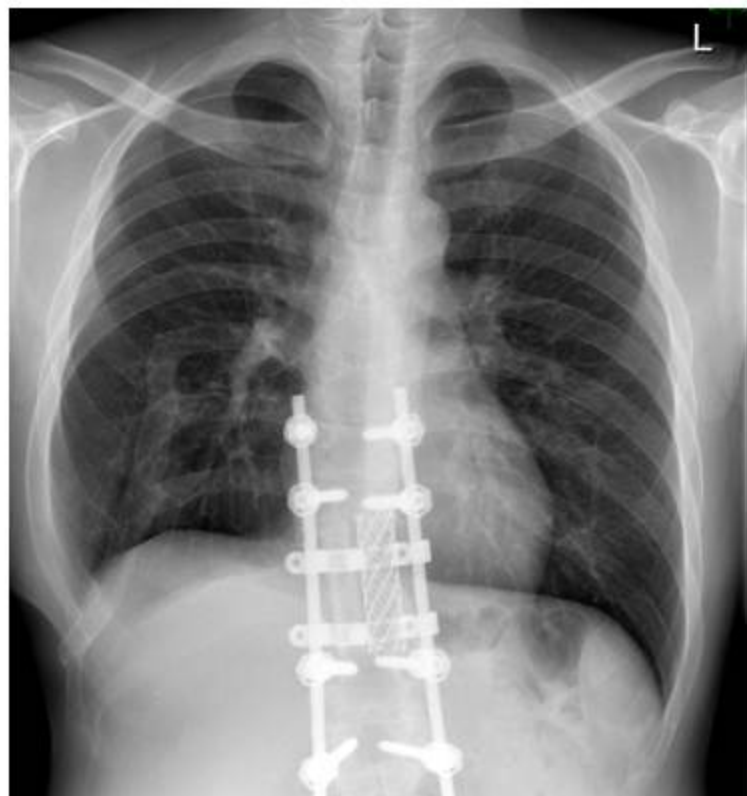


Figure 4B

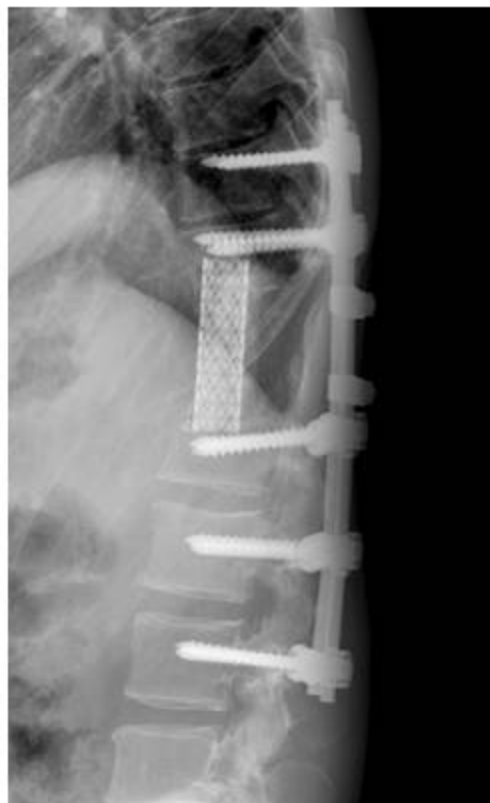


Figure 4C

