Case Report

Journal of INTERNATIONAL MEDICAL RESEARCH

Journal of International Medical Research
2019, Vol. 47(6) 2746–2753

DOI: 10.1177/0300060519847340
journals.sagepub.com/home/imr
SAGE

Minimally invasive resection of a glomus tumor of the thoracic spine: a case report and literature review

Zhiwei Liao*, Chao Chen*, Bingjin Wang and Cao Yang

Abstract

Objective: Spinal involvement of glomus tumors is extremely rare. We herein present a case of a spinal glomus tumor and reviewed the literature to identify the most effective surgical treatment of spinal glomus tumors.

Methods: A 48-year-old man presented with a huge paravertebral space-occupying lesion. In this report, we present the diagnostic process and surgical procedure in this case and review the literature of glomus tumors with spine involvement.

Results: We suspected a primary diagnosis of neurilemmoma based on the imaging results; however, the postoperative pathologic examination confirmed a glomus tumor. Considering the size of the tumor and involvement of surrounding areas, we performed complete tumor resection and unilateral fusion with pedicle screws at the T2 to T4 level. This unilateral approach with fixation was less invasive than the standard open posterior approaches that are used when one side of the spinal canal is intact without bony destruction.

Conclusions: Surgical resection is a suitable treatment for most symptomatic glomus tumors. For most glomus tumors with spine involvement, total tumor resection with suitable internal fixation and fusion is recommended.

Keywords

Glomus tumor, thoracic spine, minimally invasive, surgery, internal fixation, fusion

Date received: 1 October 2018; accepted: 8 April 2019

Department of Orthopaedics, Union Hospital, Tongji Medical College, Huazhong University of Science and Technology, Wuhan, China

*These authors contributed equally to this work.

Corresponding author:
Cao Yang, Department of Orthopaedics, Union Hospital, Tongji Medical College, Huazhong University of Science and Technology, No. 1277 Jiefang Avenue, Wuhan 430022, China.
Email: caoyangunion@hust.edu.cn

Introduction

Glomus tumors (GTs) are rare lesions that are usually located in the distal extremities, such as the subungual area and other soft tissue sites. The incidence of GTs is reportedly <2% among all soft tissue tumors.[1] The incidence peaks in the third through fifth decades of life.[2] GTs with spine involvement are extremely rare and can cause neurological symptoms when the spinal cord or nerve roots are compromised.[1–3] These tumors are generally benign, and clinical observation may be reasonable. However, complete surgical excision is the most common treatment when neurological symptoms arise.[4] We herein report a rare case of a GT in the thoracic spine and introduce a minimally invasive surgical approach. This report could provide a clinical choice for treatment of spinal GTs.

Case report

A 48-year-old man with a space-occupying lesion on a chest radiograph presented to our hospital. The patient complained of chest discomfort with intermittent back pain. Physical examination revealed normal limb strength and muscular tension and negative pathological reflexes. Computed tomography (CT) revealed a hemispheric soft tissue mass of $5.6 \times 3.8 \times 3.5$ cm in the third thoracic vertebra and paravertebral region near the thoracic cavity (Figure 1 (a), (b)). The intervertebral foramen and T3 vertebral body were obviously compromised (Figure 1(c)). The tumor protruded into the spinal canal through the left intervertebral foramen, while the right side of the vertebra was intact. Further examination by thoracic magnetic resonance imaging (MRI) showed a hemispheric large space-occupying

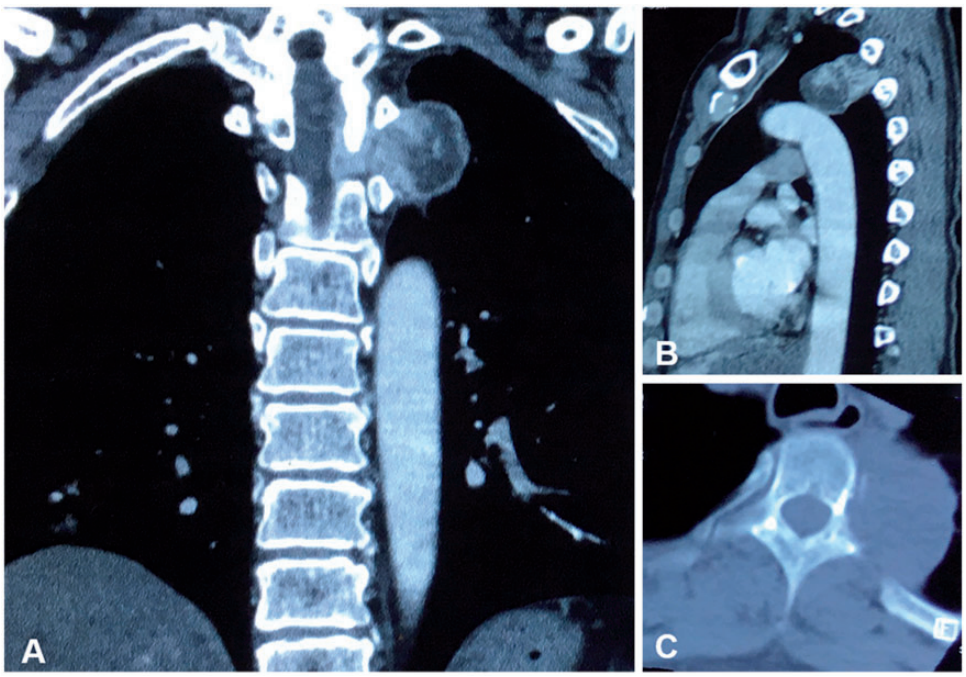

Figure 1. Computed tomography in the (a) coronal plane and (b) sagittal plane depicted a hemispheric soft tissue mass compromising the intervertebral foramen and vertebral body. (c) The cross-sectional computed tomography image showed that the tumor was accompanied by bone erosion.

lesion that was isointense on T1-weighted imaging and hyperintense on T2-weighted imaging, located at the T3 to T4 level of the spinal cord and paravertebral tissues (Figure 2(a)–(c)).

Based on the CT and MRI results, the tumor was most likely a benign lesion. The primary diagnosis was suspected to be a neurilemmoma in the thoracic spine. The patient refused a CT-guided biopsy because of concerns regarding the biopsy risk and hospitalization costs and instead preferred to undergo surgical resection. After the patient had provided informed consent and undergone preoperative preparation, surgery was performed to remove the lesion. After induction of anesthesia, the left-side paraspinal muscles were dissected along the left side of the spinous processes until the left lamina and lateral costotransverse joints from T2 to T4 were adequately exposed. The left lamina was then removed to open the spinal canal, and the left costotransverse joint of T3 was resected to expose the tumor both inside and outside of the spinal canal. The tumor body was completely excised by a posterior and anterior lateral approach, and unilateral pedicle screws and rods were then instrumented at the T2 and T4 levels.

Hematoxylin and eosin staining of the postoperative specimen showed that the tumor was composed of diverse-appearing rounded cells with regular, round to ovoid

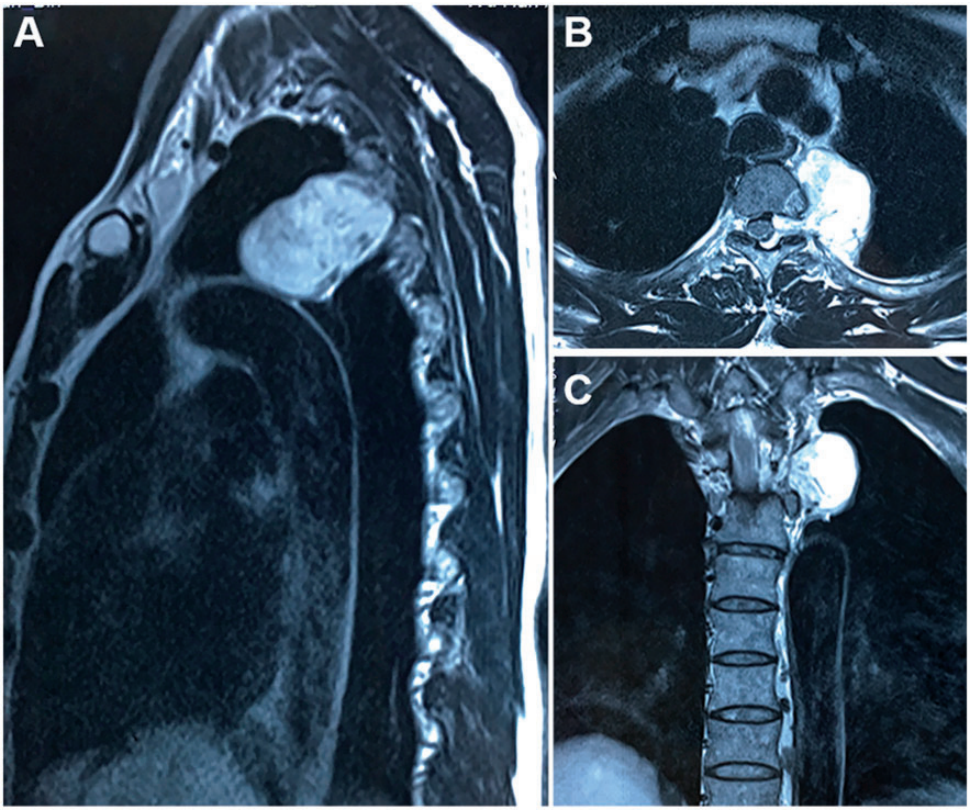

Figure 2. Thoracic magnetic resonance imaging indicated a dumbbell-shaped space-occupying lesion in the (a) sagittal plane, (b) cross-sectional plane, and (c) coronal plane.

individual nuclei and no significant nuclear pleomorphism (Figure 3(a), (b)). The immunohistology results showed that the tumor was positive for smooth muscle actin, CD34, and CD31; negative for S100, CD68, and CD57; and had a Ki67-positive rate of <5%. The final pathologic diagnosis of a GT was confirmed. The patient underwent 1 week of hospitalization after surgery, and postoperative radiographic examination showed stable internal fixation (Figure 4(a), (b)). During the 18-month follow-up, the patient showed no sign of recrudescence and was satisfied with the treatment.

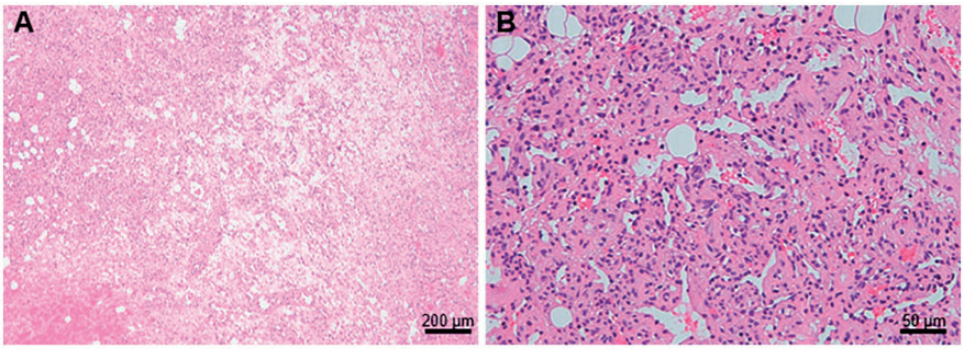

Figure 3. Representative pathological images of the surgically excised lesion. No mitotic figures were captured in hematoxylin and eosin staining. (a) 40×. (b) 200×.

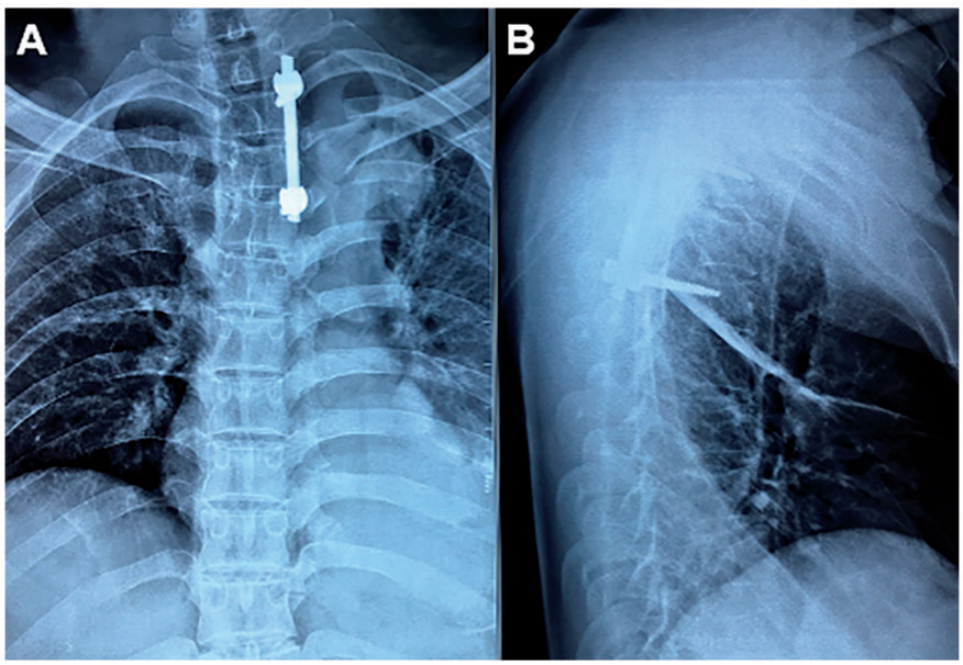

Figure 4. Postoperative thoracic radiographs showed unilateral pedicle screw fixation by (a) frontal and (b) lateral projections. The right side of the thoracic spine was intact without destruction or instability.

The study was endorsed by the Ethics Committee of Union Hospital, Tongji Medical College, Huazhong University of Science and Technology. The patient provided informed consent to publish this case report.

Discussion

GTs arise from glomus bodies, which are commonly found in arterioles, anastomotic vessels, and efferent veins.[3] Generally, GTs are small lesion in the distal extremities, especially in subungual areas.[5] GTs are usually derived from soft tissue, and spine involvement is particularly rare. The intervertebral foramen and spinal cord are compromised in some patients.[4] When bony structures are compromised, erosion and lytic lesions may be observed.[3,6] GTs may be difficult to diagnose in deep sites and are easily confused with neurilemmomas or paragangliomas based on imaging findings.[1,7] We reviewed the English-language literature of GTs with spine involvement (Table 1).

GTs were regarded as benign lesions in most previously reported cases, and patients without obvious symptoms underwent clinical observation. However, some reports described malignant GTs.[8–11] Folpe et al.[9] studied 52 unusual GTs that were considered atypical or malignant and statistically analyzed the risk factors for such GTs. The authors proposed that malignant GTs may be associated with a deep location, size of >2 cm, or atypical mitotic figures.[9] In the present case, a huge tumor (>2 cm) was found in a paravertebral location near the thoracic cavity. Even if the tumor had been considered benign according to the imaging examination results, the tumor shared some features with atypical GTs. Moreover, the left half of the T3 body was damaged, and the tumor extended into the spinal canal through an expanded intervertebral foramen. The patient had a history of intermittent back pain, and the spinal cord was slightly compressed as shown by MRI. These findings suggest that more severe clinical symptoms might arise with continual tumor growth.

To date, the most effective therapeutic method for GTs is surgical resection. A preoperative biopsy is highly recommended for tumors of undetermined nature. In the present case, the tumor was located on the left side of the thoracic spine, the other side of which was intact. The medial border of the intracanal portion of the tumor was not over the midline of the canal according to the MRI results, and the pedicles of the right-side and adjacent vertebrae were intact. Minimally invasive surgery was able to be performed through a posterior and anterior lateral approach. Unilateral fixation and fusion using pedicle screws at the T2 to T4 level was then applied to maintain spinal stability. The use of unilateral versus bilateral pedicle screws for surgical fixation is debated. A previous study has compared unilateral and bilateral pedicle screw fixation in lumbar fusion.[12] Both approaches showed similar outcomes and complications, while unilateral fixation caused less blood loss and saved operative time.[12] In the present case, surgical procedures from one side were adequate for complete and minimally invasive tumor resection. Because the right side of the spinal canal including the capsules and facet joints was intact and stable, unilateral pedicle screws were applied on the left side to maintain stability.

In most patients with GTs, the clinical symptoms disappear after removal of the tumors. Kobayashi et al.[13] described a 22-year-old woman with sacral involvement and severe sacral pain, and Kuo et al.[14] described a 45-year-old woman with lumbar GTs causing low back pain. These two patients underwent complete tumor curettage, and the symptoms rapidly disappeared. However, the most appropriate surgical approach to GTs should be based on

Table 1. Reported cases of glomus tumors with spine involvement.

Cases	Age (y), sex	Symptoms	Location, size	Treatment	Recrudescence
Axmann et al.[1]	50, male	Chronic low back pain	L1–2 vertebrae, 3 × 2 cm	Hemilaminectomy of lumbar vertebrae	No
Liu T. et al.[2]	45, female	Compressive myelopathy	T2–4 vertebral body, 5 × 6 cm	Angiographic embolization, lateral posterior resection, and bilateral spinal fixation	No
Bambakidis et al.[3]	44, male	Radicular symptoms	L3 vertebral body with epidural and extraperitoneal lesion, 11 × 11 × 12 cm	Posterior decompression and bilateral spinal fixation	Not mentioned
Payer et al.[5]	55, female	Compressive myelopathy	T4 vertebral body, 5 cm	Transthoracic T3–5 corpectomy	No
Zhou et al.[7]	39, male	Chronic low back pain	T12–L1 vertebral body, 5.0 × 4.0 × 3.3 cm	Incomplete tumor excision	Asymptomatic residual tumor
Kuo et al.[14]	26, male	Compressive myelopathy	T3 vertebral body with epidural intraspinal lesion	T7 total resection, spinal reconstruction and fusion	Not mentioned
Becce et al.[15]	73, female	Upper and lower back pain	T11 right pedicle, 1.5 × 0.6 × 1.0 cm	Percutaneous computed tomography-guided radiofrequency ablation	No
Current case	48, male	Intermittent back pain	T3 vertebrae, 5.6 × 3.8 × 3.5 cm	Unilateral resection and spinal fixation	No

the size and location of the tumors. Becce et al.[15] described a 73-year-old woman with a small tumor measuring $1.5 \times 0.6 \times 1.0$ cm in the right pedicle of T11, and radiofrequency ablation was administered. This minimally invasive treatment resulted in a good prognosis.[15] In some patients, the GTs grew large enough to complicate the surgical resection. Bambakidis et al.[3] described a 44-year-old man with a huge tumor in the lumbar region and abdominal cavity. He underwent tumor resection and spinal canal decompression involving a large volume of intraoperative hemorrhage.[3] In the present case, we did not consider radiofrequency ablation because the left pedicle was compromised.[15] Compared with open approaches for thoracic spinal tumors, unilateral tumor resection and fixation causes fewer destruction in a minimally invasive manner. However, unilateral resection is limited in some cases and is unsuitable in the lumbar spine without the support of the thoracic cavity. The surgical indications for unilateral resection and fixation require further confirmation. More research is expected to improve the surgical methods of GTs and clarify the long-term prognosis.

Conclusions

GTs with spine involvement are extremely rare. GTs can grow large enough to cause bone destruction and spinal cord compression. Suitable surgical approaches should be based on the tumor size and location. Unilateral resection and fixation can serve as a minimally invasive technique for tumors confined to one side of the spinal canal without excessive bone destruction.

Declaration of conflicting interest

The authors declare that there is no conflict of interest.

Funding

This study was supported by the National Key Research and Development Program of China (2018YFB1105700), the National Natural Science Foundation of China (U1603121, 81772401, and 81541056), and Natural Science Foundation of Hubei Province (WJ2017Z016).

ORCID iD

Cao Yang https://orcid.org/0000-0002-0058-614X

References

1. Axmann C, Feiden W, Moeller V, et al. Paravertebral glomus tumors. *Skeletal Radiol* 2005; 34: 112–115. 2004/12/09. DOI: 10.1007/s00256-004-0868-5.
2. Liu T, Zou W, Kong J, et al. Embolization in the treatment of an intraosseous glomus tumor in the upper thoracic spine complicating compression myelopathy: a case report and a literature review. *Turk Neurosurg* 2015; 25: 479–484. 2015/06/04. DOI: 10.5137/1019-5149.jtn.7854-12.1.
3. Bambakidis NC, Gore P, Eschbacher J, et al. Intraosseous spinal glomus tumors: case report. *Neurosurgery* 2007; 60: E1152–E1153; discussion E1153. 2007/06/01. DOI: 10.1227/01.neu.0000255463.37634.f2.
4. Ogboli MI, Ilchyshyn A, Walker RS, et al. Glomus tumor as a cause of chronic low back pain: case report. *Spine* 2003; 28: E146–E147. 2003/04/17.
5. Payer M, Grob D, Benini A, et al. Intraosseous glomus tumor of the thoracic spine. Case illustration. *J Neurosurg* 2002; 96: 137. 2002/01/25.
6. Maggiani F, Kashima T, Ostlere SJ, et al. Immunophenotypic analysis of glomus coccygeum associated with coccygodynia. *Skeletal Radiol* 2011; 40: 1455–1459. 2011/03/02. DOI: 10.1007/s00256-011-1128-0.
7. Zhou P, Zhang H, Bu H, et al. Paravertebral glomangiomatosis. Case report. *J Neurosurg* 2009; 111: 272–277. 2009/03/10. DOI: 10.3171/2009.2.jns081276.
8. Chung DH, Kim NR, Kim T, et al. Malignant glomus tumor of the thyroid

gland where is heretofore an unreported organ: a case report and literature review. *Endocr Pathol* 2015; 26: 37–44. 2014/12/30. DOI: 10.1007/s12022-014-9352-5.
9. Folpe AL, Fanburg-Smith JC, Miettinen M, et al. Atypical and malignant glomus tumors: analysis of 52 cases, with a proposal for the reclassification of glomus tumors. *Am J Surg Pathol* 2001; 25: 1–12. 2001/01/06.
10. Ugras N, Yerc IO, Yalcinkaya U, et al. Malignant glomus tumor with oncocytic features: an unusual presentation of dysphagia. *APMIS* 2015; 123: 613–617. 2015/04/25. DOI: 10.1111/apm.12394.
11. Nagata K, Hashizume H, Yamada H, et al. Long-term survival case of malignant glomus tumor mimicking "dumbbell-shaped" neurogenic tumor. *Eur Spine J* 2017; 26: 42–46. journal article. DOI: 10.1007/s00586-016-4703-1.
12. Liu H, Xu Y, Yang SD, et al. Unilateral versus bilateral pedicle screw fixation with posterior lumbar interbody fusion for lumbar degenerative diseases: a meta-analysis. *Medicine (Baltimore)* 2017; 96: e6882. DOI: 10.1097/MD.0000000000006882.
13. Kobayashi Y, Kawaguchi T, Imoto K, et al. Intraosseous glomus tumor in the sacrum. A case report. *Acta Pathol Jpn* 1990; 40: 856–859. 1990/11/01.
14. Kuo CH, Huang WC and Wu JC. Unusual imaging presentation of spinal glomus tumor: case report. *J Spine Surg* 2017; 3: 715–718. 2018/01/23. DOI: 10.21037/jss.2017.11.01.
15. Becce F, Richarme D, Letovanec I, et al. Percutaneous radiofrequency ablation of primary intraosseous spinal glomus tumor. *Skeletal Radiol* 2012; 41: 467–472. 2011/11/11. DOI: 10.1007/s00256-011-1308-y.