Giant Invasive Spinal Schwannoma With Vertebral Body Collapse In The Cervical Spine: A Case Report

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Research article

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Abstract

Study Design: A case report.

Objective: To describe the first case of GISS with VBC in the cervical spine, analyze the clinical features and surgical outcomes, and discuss diagnosis and treatment strategies.

Summary of Background Data: The giant invasive spinal schwannoma (GISS) extends more than two vertebral levels, erodes the vertebral body and extends posteriorly and laterally into the myofascial plane. To the best of our knowledge, never before has there been a report of GISS with vertebral body collapse (VBC).

Methods: A 44-year-old man was admitted as a result of a 1-year history of limb weakness and defecation difficulties. Power were reduced in all limbs: right upper limb middle finger muscle strength was 2/5, both lower limb hip flexion and knee flexion muscle strength was 3/5, and the muscle strength of the remaining limbs was 0/5. Preoperative MRI revealed that an extradural tumor, localized at the C3 to C5 levels, invaded the C4 vertebral body and attachment. A two-stage operation by posterior and anterolateral approaches was performed. One year after surgery, the patient could walk and write unassisted, and strength was regained to 4/5 in the upper extremities and 5/5 in the lower extremities. MRI showed no recurrence of the tumor. CT showed good fusion of the vertebral bodies, and X-ray films confirmed stable fixation materials without loosening or displacement. The bowel function has improved though patient remains only able to defecate in bed.

Conclusions: We herein describe the case of GISS with VBC in the cervical spine. Paired with imaging for malignant features, MRIs meeting the criteria for GISS diagnosis may reveal similar cases that could be preoperatively evaluated by biopsy for optimal surgical planning.

Introduction

Spinal schwannoma is a benign, typically encapsulated nerve sheath tumor composed entirely of well-differentiated Schwann cells, comprising of about 20-30% of primary intraspinal tumours.[1, 2] In the vast majority of cases, schwannomas are solitary and sporadic, following a benign clinical course. According to the research of Sridhar et al.[3], the giant invasive spinal schwannomas (GISS) is defined as a tumor that extends more than two vertebral levels, erodes the vertebral body and extends posteriorly and laterally into the myofascial plane. At present, there are few reports on the clinical features and treatment methods of GISSs.[4, 5] Moreover, to the best of our knowledge, never before has GISS with vertebral body collapse (VBC) been reported. We herein described the first case of GISS with VBC in the cervical spine, analyzed the clinical features and surgical outcomes, and discussed diagnosis and treatment strategies.

Case Presentation
A 44-year-old man without hypertension, diabetes mellitus or congenital disease was referred from an outside hospital as a result of a 1-year history of limb weakness and defecation difficulties. He initially complained of progressive right limb weakness of unknown origin and became progressively quadriplegic with difficulty defecating after a fall. He was diagnosed with a metastatic spinal tumor after magnetic resonance imaging (MRI) was performed at a Grade A hospital. He was told that the survival time was about 6-12 months, and the specialist recommended that further visits to the radiotherapy department be available. However, the patient abandoned further treatment. After one year bedridden at home, his symptoms persisted with no new discomforts. The patient had not received any treatment before referral.

Upon neurological examination, the results revealed that muscle power had been reduced in all limbs: right upper limb middle finger muscle strength was 2/5, both lower limb hip flexion and knee flexion muscle strength was 3/5, and remaining limb muscle strength was 0/5. His biceps reflexes, triceps reflexes and knee-jerk reflexes were exaggerated bilaterally. The patient exhibited serious hypoesthesia to light touch sensations and thermal nociception on his chest, abdomen and both lower extremities. Myelopathy was assessed with the modified Japanese Orthopedic Association (mJOA) scale [6]; the patient scored 4 of 15 points preoperatively.

MRI (Figure 1) of the cervical spine revealed that an extradural tumor, localized at the C3 to C5 levels, compressed the spinal cord to the left side and invaded the C4 vertebral body and attachment. The tumor penetrated the C4 vertebral body at the anterior edge. In addition, T2-weighted images (T2WI) revealed the lesion was hyperintense compared with the spinal cord. After the administration of intravenous gadolinium, the tumor demonstrated significant enhancement. Computed tomography (CT, Figure 2) subsequently delineated the VBC of the C4 and soft tissue density in the cancellous substance of C4. Finally, a comparison of CT (Figure 2C) and MRI (Figure 1C and 1D) images was performed at the external hospital in 2017. There was no significant progress in the range of the lesion.

A two-stage operation by posterior and anterolateral approaches was planned due to the large extradural component and eroded nature of the entire C4 with VBC. After obtaining written informed consent, the patient underwent microsurgically-assisted extradural exploration and intraoperative tumor biopsy through a posterior approach. The pathological results of intraoperative tumor biopsy indicated a "neurogenic tumor". Therefore, the tumor was removed except for the portions remaining in C4 and lateral mass plating was added for stability. During the second surgery, the patient underwent C4 vertebrectomy and intervertebral bone graft fusion with instrumentation for stability. The histological diagnosis of tumor tissue from both operations was consistent with schwannoma (Figure 3).

The postoperative course was uneventful, and the patient’s limb weakness gradually resolved. One year after surgery, the patient could walk and write on their own, strength was regained to 4/5 in the upper extremities and 5/5 in the lower extremities, and the mJOA of the patient was 15 of 15 points at that time. A review of cervical MRI (Figure 4A and 4B) one year after surgery showed no recurrence of the tumor, CT (Figure 4C) showed good fusion of the vertebral bodies, and X-ray films (Figure 4D) showed stable
fixation materials without loosening or displacement. At present, the bowel function of the patient are improved, however the patient remains with the need to defecate in bed.

**Discussion**

Spinal schwannoma is usually a single and benign lesion that is straightforward to remove and is associated with a uneventful course after surgery.[7] GISS is rare, and has the following two characteristics:[3] 1) In imaging, GISS extends beyond two vertebral levels, erodes the vertebral body and extends posteriorly and laterally into the myofascial plane. 2) Histologically, GISS has the typical pathological manifestations of schwannoma and remains benign.

Metastatic spinal cord compression (MSCC) is the most serious complication of spinal metastasis.[8] MSCC can also meet the imaging standards of GISS (ie. giant epidural tumors that erode the vertebral bodies and extend into the myofascial planes).[9] In this case, the CT revealed C4 vertebral body collapse caused by GISS like spinal metastasis. Therefore, this tumor was not distinguished by clinical presentations, CT, or MRI alone. With respect to giant spinal tumors when first discovered, biopsy of the tumor is more useful and is recommended to identify the source of the unknown tumor.[10] The CT-guided needle biopsy is safe and a reliable method in these cases.[11]

With respect to spinal schwannoma, total resection is recommended because inadequate removal may be followed by regrowth and repeated surgery introduces risks of complications.[4] The giant invasive spinal schwannomas may span a wide range of areas, coexisting with nerve compression and bone destruction, making the operation difficult. Yu et al.[4] and Ozdemir et al.[5] summarized and reported the surgical methods used for the treatment of GISS in their hospitals, in which the majority of GISS were completely resected through the posterior approach for tumors, and spinal internal fixation was not routinely selected. When the tumor invades more than 25% of the vertebral body, additional internal fixation surgery is usually required.[3] In this case, the GISS invaded the whole C4 vertebral body and caused C4 VBC, resulting in spinal instability. Therefore, a two-stage operation by posterior and anterolateral approaches was planned, including removal the tumor except the portion in the C4, followed by C4 vertebrectomy and intervertebral bone graft fusion with instrumentation for stability.

In the present study, the common symptom of GISS was pain, motor weakness and urinary symptoms.[3, 5, 4] In this case, the patient was referred from outside hospital as a result of a 1-year history of limb weakness and defecation difficulties after a fall. Muscle strength of the limbs and the excretion function improved significantly one year after surgery. Therefore, the patient's quadriplegia was mainly attributed to acute spinal cord compression with incomplete ischemia of cervical spinal cord caused by GISS.

**Conclusions**

We diagnosed and treated a rare case of cervical GISS with malignant imaging findings, which resulted in an unidentified diagnosis due to atypical symptoms and radiography. Therefore, for giant spinal tumors,
MRI consistent with the characteristics of GISS with accompanied by malignant imaging findings, we cannot exclude the consideration of GISS, and preoperative biopsy is recommended.

**Abbreviations**

C-cervical

CT-Computed tomography

GISS-giant invasive spinal schwannoma

MRI-magnetic resonance imaging

mJOA-modified Japanese Orthopedic Association

MSCC-Metastatic spinal cord compression

T-thoracic

VBC-vertebral body collapse

**Declarations**

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This study does not have funding resources.

**Conflict of Interest**

The authors have declared that no competing interests exist.

**Ethics approval and consent to participate**

This case report was approved by the ethics committee of Fujian Medical University Union Hospital, and written consent was obtained from the patients for the publication of this case report and any accompanying images.

**Consent to participate**

Not applicable.

**Consent for publication**
I, Chun Mei Chen, certify that this manuscript is a unique submission and is not being considered for publication, in part or in full, with any other source in any medium. All authors have contributed to this study and approved this document and its submission to the journal.

**Availability of data and materials:**

The data will be available upon the reasonable request.

**Code availability**

Not applicable.

**Authors’ Contributions**

Ze-Yan Liang, Zhe-Wei Wang and Chun-Mei Chen designed this study, collected the data and wrote the manuscript. Xin-Yao Chen collected the data and approved the manuscript.

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**Figures**

**Figure 1**

Preoperative cervical magnetic resonance image (MRI). Preoperative sagittal enhanced T1-weighted MRI (A) showed a giant invasive spinal tumor at the C3-5 level. Axial enhanced T1-weighted MRI (B) revealed the tumor with spinal cord compression extended bilaterally across the C3-4 extraforaminal region, surrounded the spinal cord in a “U”-like shape and invaded the C4 vertebral body (B, arrow a) and pedicles (B, arrow b). Compared to the sagittal and axial T2-weighted MRI (C, D) performed in the outside hospital a year ago, the tumor was not enlarged.
Figure 2

Preoperative cervical computed tomography (CT). Preoperative sagittal CT (A) showed C4 vertebral body collapse and soft tissue density in the cancellous substance of C4. Axial CT (B) revealed the tumor eroded bilateral pedicles (B, arrow). Compared to the sagittal CT (C) performed in the outside hospital, it was determined that the C4 vertebral body collapse existed a year ago.
Figure 3

A histopathological image after hematoxylin-eosin staining (40×) The tumor tissue was removed A during the first operation and B the second operation. Histopathological images demonstrate the proliferation of
spindle-shaped cells (A and B), which are compatible with a schwannoma.

**Figure 4**

Follow-up MRI, CT, and X-ray 12 months after surgery. The sagittal T2WI (A) and enhanced T1WI (B) MRI revealed that the tumor was removed totally, and there was no recurrent tumor. CT (C) demonstrated good fusion of the vertebral bodies, and X-ray films (D) showed stable fixation of materials without loosening or displacement.