CASE REPORT

Nagoya J. Med. Sci. **81**. 701–705, 2019 doi:10.18999/nagjms.81.4.701

Natural reduction in acute intratumoral hemorrhage of spinal schwannoma in the cauda equina

Kenyu Ito, Kei Ando, Kazuyoshi Kobayashi, Mikito Tsushima, Masaaki Machino, Kyotaro Ota, Masayoshi Morozumi, Satoshi Tanaka, Naoki Ishiguro, and Shiro Imagama

Department of Orthopaedic Surgery, Nagoya University Graduate School of Medicine, Nagoya, Japan

ABSTRACT

A 58-year-old woman presented with acute pain in her back and her left leg. Magnetic resonance imaging (MRI) revealed an intradural schwannoma with an intratumoral hemorrhage between the lower L4 vertebra and L5/S1 disk level. A follow-up MRI after one month revealed that the tumor had regressed from the middle of L5 to the L5/S1 disk level. The tumor was totally resected. There have been several reports of intratumoral hemorrhage of spinal schwannomas occurring at the cervical, thoracic, and conus levels; however, there has been only one previous report of it being seen at the L2-3 middle lumbar level and none at all at lower lumbar levels. This is the first report of an intratumoral hemorrhage of a schwannoma in the lower lumbar area. Furthermore, natural regression of an intratumoral hemorrhage of spinal schwannoma at lower lumbar levels has not previously been reported.

Keywords: schwannoma, intratumoral, hemorrhage, regression, shrinking

This is an Open Access article distributed under the Creative Commons Attribution-NonCommercial-NoDerivatives 4.0 International License. To view the details of this license, please visit (http://creativecommons.org/licenses/by-nc-nd/4.0/).

INTRODUCTION

Spinal schwannomas are slow-growing benign tumors of the spinal nerve sheath and present with gradually progressive symptoms.¹ They rarely present with subarachnoid hemorrhage,² and intratumoral hemorrhage of a spinal schwannoma is extremely rare. There are several reports of spinal schwannomas occurring at the cervical, thoracic, and conus levels.³⁻⁵ However, there has been only one report of a schwannoma manifesting at the L2-3 middle lumbar level, and no reports of these tumors occurring at lower lumbar levels.⁶ Natural regression of an intratumoral hemorrhage of spinal schwannoma has not previously been reported in the literature. We report a case of natural regression of acute intratumoral hemorrhage of a spinal schwannoma in the lower lumbar area.

Received: January 18, 2019; accepted: March 13, 2019

Corresponding Author: Shiro Imagama, MD, PhD

Department of Orthopaedic Surgery, Nagoya University Graduate School of Medicine, 65 Tsurumai-cho, Showa-ku, Nagoya, 466-8550, Japan

Tel: +81-52-741-2111, Fax: +81-52-741-2260, E-mail: imagama@med.nagoya-u.ac.jp

CASE REPORT

A 58-year-old woman presented with acute pain in her back and left leg. The patient had no history of disease or drug use, and her bowel and bladder functions were normal. Physical examination revealed grade 4/5 weakness upon dorsal flexion of the left ankle, with decreased sensation of touch on the left leg. Magnetic resonance imaging (MRI) revealed an intradural schwannoma between the lower L4 vertebra and the L5/S1 disk level. T2-weighted images demonstrated the fluid-fluid level as hyperintense in the ventral area and hypointense in the dorsal area on the left upper side of the tumor. The left sagittal view of the initial T1-weighted MRI demonstrated an isointense to high-intense area in the tumor. The radiological diagnosis was schwannoma with intratumoral hemorrhage (Figure 1).

A follow-up MRI after one month revealed that the tumor had regressed from the middle of the L5 vertebra to the L5/S1 disk level (Figure 2). Laminectomy of the L5 vertebra was performed under general anesthesia. After opening the dura under microscopic view, a tumor with a cystic lesion containing intratumoral hemorrhage on the dorsal side was seen below the cauda equina.

The tumor was totally resected (Figure 3). Postoperatively, the patient's pain resolved and her sensitivity to touch improved, with no motor deficit remaining. Hematoxylin and eosin (H&E) staining of the specimen showed a typical schwannoma consisting of spindle cells with a hemorrhagic lesion within the mass (Figure 4).



Fig. 1 Initial MRI

- Fig. 1A: Midsagittal view of the initial T2-weighted MRI demonstrates an intradural spinal tumor from the lower L4 vertebra to L5/S1 disk level.
- Fig. 1B: Left sagittal view of the initial T2-weighted MRI demonstrates a fluid-fluid level (arrow).
- Fig. 1C: Left sagittal view of the initial T1-weighted MRI demonstrates a T1 iso-high area in the tumor (arrow).
- Fig. 1D: Axial view of the initial T2-weighted MRI demonstrates a fluid-fluid level at the left side of the tumor.



Fig. 2 Follow-up MRI

- Fig. 2A: By one month after the initial MRI the tumor had regressed from the middle of the L5 vertebra to the L5/S1 disk level.
- Fig. 2B: One month after the initial MRI the fluid-fluid level had decreased in both the hyperintense and hypointense areas.



Fig. 3 Intraoperative image

Fig. 3A: Intraoperative image showing the tumor located on the ventral side of the cauda equina. Fig. 3B: The dorsal intratumoral hemorrhage (arrow) was visible after removing the cauda equina from the tumor.



Fig. 4 Hematoxylin and eosin staining Fig. 4A: H&E, ×4: Hemorrhagic lesion in the mass along with spindle cells. Fig. 4B: H&E, ×20: Cellular area (Antoni A pattern) consisting of spindle cells.

DISCUSSION

The natural history of intratumoral hemorrhage of spinal schwannomas is unknown. Ando et al reported the natural history of spinal schwannomas in general, which did not include intratumoral hemorrhage.⁷ However, a discussion of the natural history of vestibular schwannomas included intratumoral hemorrhage, with reasons given for the growth of these schwannomas reported to be necrosis, rupture of microcysts, and repeated episodes of intratumoral hemorrhage.⁸ Shrinking of tumors after intratumoral hemorrhage was not defined.⁸ In the case that we report here shrinkage of the tumor may have been due to a rupture of the tumor or natural regression thereof, but there was no direct evidence to prove this.

Intratumoral hemorrhage of spinal schwannomas is rare. Hdeib et al reviewed the literature and reported 11 cases of hemorrhagic schwannoma. Of these 11, five patients presented with subdural hemorrhage and six with intratumoral hemorrhage.⁵ None of the hemorrhagic schwannomas were located at the lower lumbar junction. A reason for this could be that symptoms associated with moderate compression in the lower lumbar junction and cauda equina will not always present owing to the volume of cerebrospinal fluid. In the case which we report here, the patient experienced radiculopathy and an intratumoral hemorrhage was observed on the left side of the tumor, compressing the left L5 root.

The cause of acute intratumoral hemorrhage is unknown. One hypothesis suggests that the hyalinized ectatic vessels inside schwannomas undergo spontaneous thrombosis, resulting in distal necrosis and hemorrhage, while another theory supports the occurrence of traction of the tumor vasculature during movement.^{5,9} Trauma or a history of anticoagulant therapy may cause intratumoral hemorrhage, but our patient had no such medical history.

In conclusion, this is the first report of intratumoral hemorrhage of a schwannoma in the lower lumbar area. Other cases may have occurred, but they are difficult to detect because they are usually asymptomatic.

CONFLICTS OF INTEREST

The authors declare that there are no conflicts of interest.

REFERENCES

- Conti P, Pansini G, Mouchaty H, Capuano C, Conti R. Spinal neurinomas: retrospective analysis and longterm outcome of 179 consecutively operated cases and review of the literature. *Surg Neurol.* 2004;61:34-43, discussion 44.
- Chalif DJ, Black K, Rosenstein D. Intradural spinal cord tumor presenting as a subarachnoid hemorrhage: magnetic resonance imaging diagnosis. *Neurosurgery*. 1990;27:631-634.
- 3) Sahoo RK, Das PB, Sarangi GS, Mohanty S. Acute hemorrhage within intradural extramedullary schwannoma in cervical spine presenting with quadriparesis. *J Craniovertebr Junction Spine*. 2015;6:83-85.
- 4) Kukreja S, Ambekar S, Sharma M, Nanda A. Cauda equina schwannoma presenting with intratumoral hemorrhage and intracranial subarachnoid hemorrhage. *J Neurosurg Spine*. 2014;21:357-360.
- 5) Hdeib A, Goodwin CR, Sciubba D, et al. Hemorrhagic thoracic schwannoma presenting with intradural hematoma and acute paraplegia after spinal manipulation therapy. *Int J Spine Surg.* 2016;10:42.
- 6) Jenkins AL 3rd, Ahuja A, Oliff AH, Sobotka S. Spinal Schwannoma presenting due to torsion and hemorrhage: case report and review of literature. *Spine J.* 2015;15:e1-4.
- Ando K, Imagama S, Ito Z, et al. How do spinal schwannomas progress? The natural progression of spinal schwannomas on MRI. J Neurosurg Spine. 2016;24:155-159.
- Park CK, Kim DC, Park SH, et al. Microhemorrhage, a possible mechanism for cyst formation in vestibular schwannomas. J Neurosurg. 2006;105:576-580.
- Ng PY. Schwannoma of the cervical spine presenting with acute haemorrhage. J Clin Neurosci. 2001;8:277-278.