

CASE REPORT

Nagoya J. Med. Sci. 79. 115 ~ 121, 2017
doi:10.18999/nagjms.79.1.115

Discrimination between spinal extradural meningioma and both intra and extradural meningioma: Case Report

Kenyu Ito, Shiro Imagama, Kei Ando, Kazuyoshi Kobayashi, Tetsuro Hida,
Mikito Tsushima, Akiyuki Matsumoto, Masayoshi Morozumi,
Satoshi Tanaka, Yoshihiro Nishida and Naoki Ishiguro

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

ABSTRACT

Meningiomas are common spinal tumor and mostly located at intradura. Recurrence rate after surgery for extradural meningioma was higher than intradural meningioma. A patient with intra and extradural spinal meningioma was treated and discussed its features and clinical management. A 41-year-old woman noted numbness of bilateral legs, gait disturbance, and mild bladder disturbance for over the two-month period. Magnetic resonance imaging revealed an epidural mass at T8-9 involving the dura mater from the left side. Intraoperatively, ultrasonography showed extradural tumor suppress the dura from the left side and no subarachnoid space. Therefore, it was difficult to diagnose the tumor was located at only extradural or both intra and extradural. The extradural tumor was resected as much as possible, then ultrasonography was performed for the second time. The spinal cord was decompressed and subarachnoid space was appeared with intradural tumor. The dura mater was opened in the midline for intradural exploration, and the intradural tumor appeared beside the spinal cord. The dura mater with attachment to the tumor was resected. To prevent a recurrence, ultrasonography after removing extradural tumor is recommended to detect intra dural tumor and invasion of tumor into dura mater. In such case, removing only extradural meningioma is not enough, and gross total resection including intradural meningioma and dura mater is required.

Key Words: extradural, meningioma, intradural, gross total resection, ultrasonography

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INTRODUCTION

Meningiomas are common spinal tumors that are primarily located at the intradura. Extradural spinal meningiomas are more invasive and aggressive than intradural meningiomas.¹⁾ The recurrence rate after surgery for extradural meningiomas is four times higher than that of intradural meningiomas.²⁾ Extradural spinal meningiomas are rare, with only a few reported cases.

A patient with intra and extradural spinal meningioma was treated and we discuss the tumor's features and clinical management.

This literature was approved by the ethical committee of Nagoya University Graduate School

Received: October 24, 2016; accepted: January 30, 2017

Corresponding author: Shiro Imagama, MD, PhD

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

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

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CLINICAL PRESENTATION

A 41-year-old woman noted numbness of bilateral legs, gait disturbance, and mild bladder disturbance for over a 2-month period. Magnetic resonance imaging (MRI) revealed an epidural mass at T8-9 involving the dura mater on the left side and extending to the left neural foramina (Fig.1 A–C). Computerized tomography (CT) showed a low density lesion. There were no signs of erosion of the mass into the vertebral body. Look back on attentively, a little bit of calcification was observed in the tumor (Fig.1 D).

Preoperatively, the radiological differential diagnosis was metastatic tumor or lymphoma. Blood test and thoracic-abdominal CT were normal and the origin of the tumor could not be determined. Therefore, an open biopsy was conducted. Under general anesthesia, T8-9 laminectomy was performed and the extradural tumor was exposed (Fig.2 A, B). Ultrasonography showed the extradural tumor suppressed the dura on the right side and there was no subarachnoid space. Therefore, it was difficult to diagnose the tumor as located only extradurally or both intra and extradurally. Pathological diagnosis during surgery revealed a psammomatous meningioma. Thus,

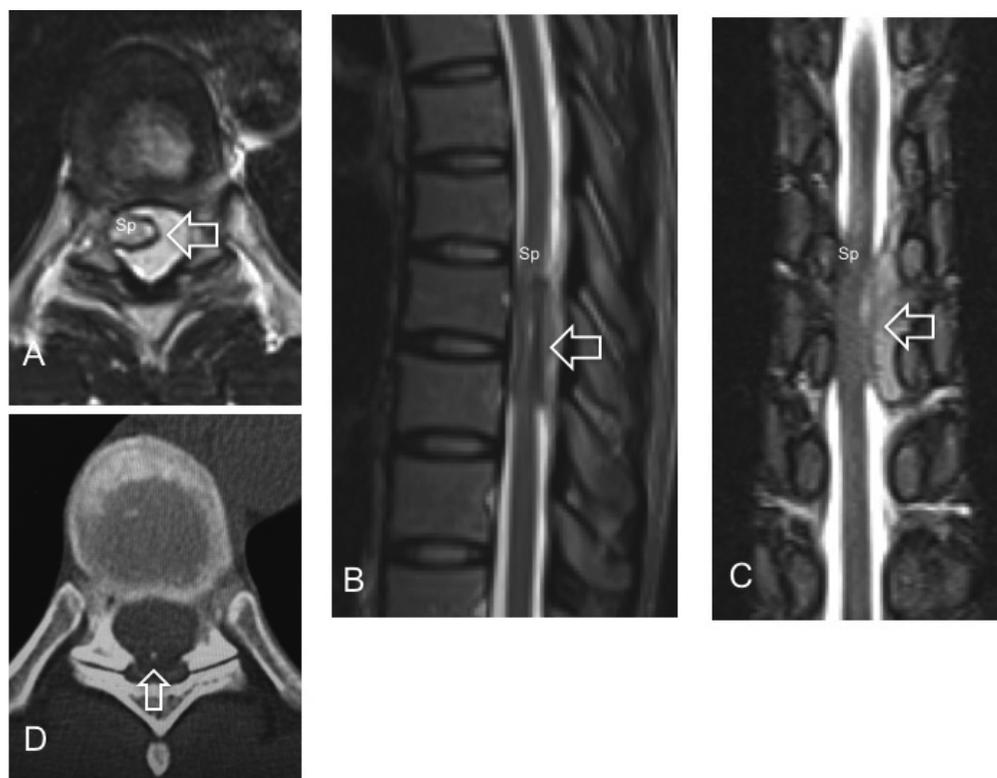


Fig. 1 Axial (A), sagittal (B), and coronal (C) T2-weighted preoperative MRI of the thoracic spine showed an iso-hyper signal intense mass (leftwards arrow) surrounding the dura mater. Preoperative CT (D) shows partial calcification (upwards arrow).

Sp: Spinal cord

a complete resection was planned. The extradural tumor was resected as much as possible and an attachment to the tumor was detected at the lateral side of the dura mater. Then ultrasonography was performed for the second time. After the spinal cord was decompressed, the subarachnoid space was revealed with an intradural tumor (Fig. 2 C, D). The dura mater was opened in the midline for intradural exploration, and the intradural tumor appeared beside the spinal cord. We removed the intradural tumor and an attachment to the tumor was detected at the lateral side of the dura mater (Fig.3). The dura mater with the attachment to the tumor was resected in a U-shape and a polytetrafluoroethylene sheet used to put in a patch. Postoperatively, numbness was limited to the toe, the gait became stable, and the mild bladder disturbance was markedly reduced. Postoperative histologic examination diagnosed a meningioma with a MIB-1 labeling index of 2% (Fig. 4). The tumor was completely resected with the dura mater and there was no recurrence during the 4 years follow-up (Fig. 5).

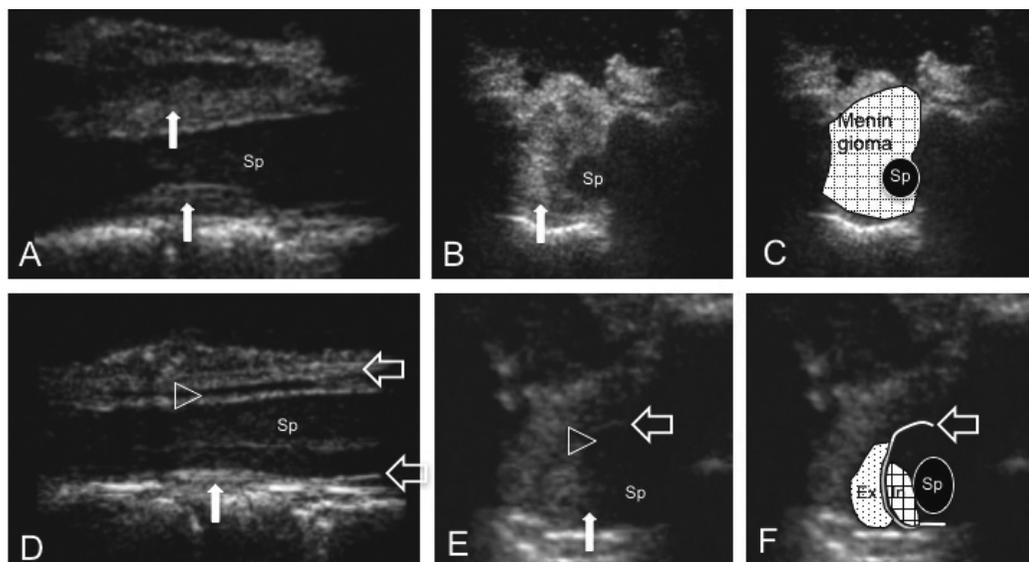


Fig. 2 Sagittal (A) and axial (B, C) intraoperative ultrasonography after laminectomy show the tumor surrounding the spinal cord; however, it could not detect the intradural tumor. Sagittal (D) and axial (E, F) ultrasonography after mass reduction of the extradural tumor reveals the dura mater (leftwards thick arrow), cerebral spinal fluid (rightwards arrow head), and intradural tumor (upwards arrow). Sp: Spinal cord, Ex: Extradural meningioma, In: Intradural meningioma

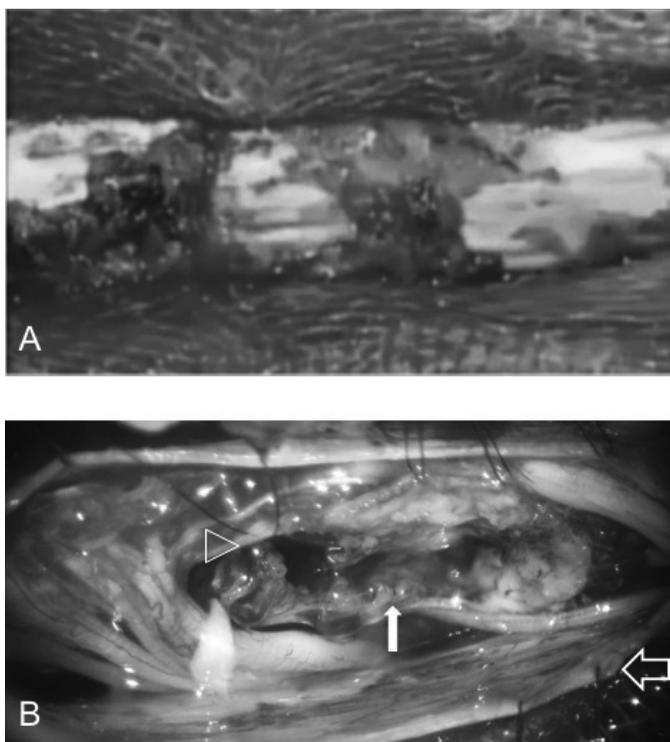


Fig. 3 Intraoperative photograph after laminectomy showed extradural tumor (A). The dura mater was bisected and pulled (leftwards thick arrow), and then the denticulate ligament was pulled to turn over the spinal cord (rightwards arrow head) (B). Finally, the intradural tumor was exposed (upwards arrow).

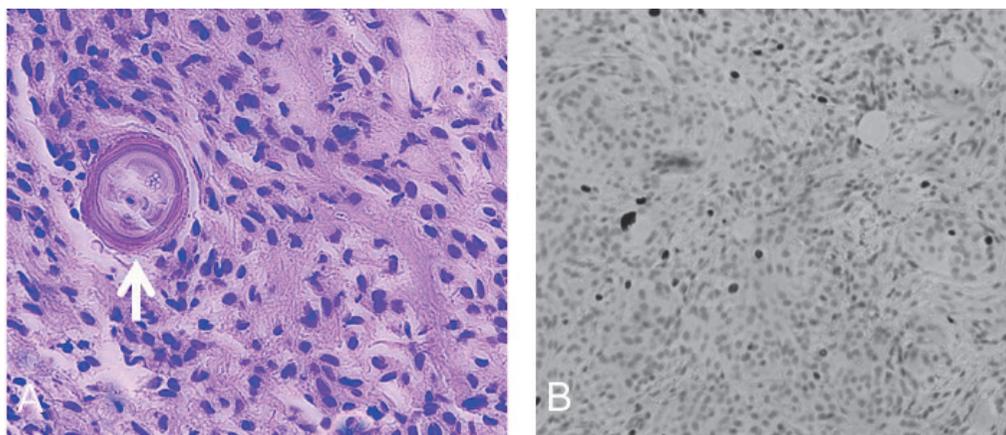


Fig. 4 Hematoxylin eosin staining of the excised lesion (A) showed spindle cells and psammoma bodies (arrow). Ki67/MIB-1 proliferation index was 2% (B).



Fig. 5 Axial (A) and sagittal (B) T2-weighted postoperative MRI of the thoracic spine shows tumor was removed. Postoperative CT (C, D) shows polytetrafluoroethylene sheet was patched.

DISCUSSION

Intradural meningioma and extradural meningioma

Intradural spinal meningioma of the thoracic spine occurs in 80% of the individuals in their 50s and 60s and affecting women four times more often than men.^{3, 4)} Extradural spinal meningioma makes up 3.3%–7.8% of all spinal meningiomas.^{5–8)} The age of diagnosis of extradural spinal meningioma is 14–75 years (mean 38 years, 47% younger than 30 years) and 64.7% of patients are women.⁹⁾ Extradural meningioma occurs most frequently in the thoracic spine and cervical spine.^{10, 11)} Based on the reported data, extradural spinal meningiomas are rare and occur at younger ages than intradural spinal meningiomas.

Imaging findings of extradural meningioma

Extradural meningiomas are mistaken as extradural spinal metastases because of their location and frequency. The primary radiological differential diagnosis of our case was a metastatic tumor or lymphoma. In other reports, primary radiological differential diagnoses were lymphoma (3 cases), cavernous angioma (2 cases), metastasis (5 cases), schwannoma (2 cases).¹⁰⁾ Lymphoma occurrence is mostly extradural and is frequently in the thoracic spine, making it difficult to distinguish from extradural meningioma.¹²⁾ Calcification in tumors observed on CT images might

support a diagnosis of extradural meningioma. However, neuroimaging shows visible calcification in only 1.0%–4.6% of all spinal meningiomas.^{1, 2, 13)} In our case, calcification in the tumor was not initially observed and hence, it was not diagnosed preoperatively.

Prevention of recurrence

The recurrence rate after surgery for extradural meningioma is four times higher than that of intradural meningioma.²⁾ The 12 case series of long-term follow up of extradural spinal meningiomas revealed that four cases with gross total resection had no recurrence and eight cases with subtotal resection had one recurrence after 88 months.¹⁰⁾ Moreover, the long-term follow up of intradural spinal meningiomas showed no recurrence even for gross total resection cases.¹⁴⁾ Based on these reports, recurrence might occur in a case of subtotal resection possibly due to a tumor invasion into the dura mater. In fact, extradural spinal meningiomas are mostly both intradural and extradural, and cases of exclusive extradural spinal meningiomas are very rare.¹⁵⁾ In our case, it was difficult to preoperatively detect that the tumor was both intra and extradural. Moreover, this was difficult to detect via ultrasonography during the surgery before removing the extradural tumor. The intradural tumor was detected via ultrasonography only after removing the extradural tumor. The surgery plan was changed to gross total resection that included removing the intradural tumor and the attachment of the dura mater. Ultrasonography is useful tool to detect intra and extradural tumors.^{16, 17)} Careful observation during surgery and gross total resection including the dura mater should reduce the recurrence rate. However, gross total resection of extradural meningioma for elderly patients, and for ventral intradural meningioma, is controversial.^{18, 19)}

CONCLUSION

Extradural meningioma mimics lymphoma on imaging. The presence of calcification can help in diagnosis. Ultrasonography after removing an extradural tumor is useful to detect any intradural tumor and invasion of the tumor into dura mater. In such a case, removing only the extradural meningioma is not enough to prevent a recurrence and gross total resection including intradural meningioma and dura mater is required.

Approved by the ethical committee of Nagoya University Graduate School of Medicine. Informed consent for the publication was obtained from the patient.

CONFLICTS OF INTEREST

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper. Approved by the ethical committee of Nagoya University Graduate School of Medicine. Informed consent for the publication was obtained from the patient.

REFERENCES

- 1) Messori A, Rychlicki F, Salvolini U. Spinal epidural en-plaque meningioma with an unusual pattern of calcification in a 14-year-old girl: case report and review of the literature. *Neuroradiology*, 2002; 44: 256–260.

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- 2) Klekamp J, Samii M. Surgical results for spinal meningiomas. *Surg Neurol*, 1999; 52: 552–562.
- 3) Solero CL, Fornari M, Giombini S, Lasio G, Oliveri G, Cimino C, *et al.* Spinal meningiomas: review of 174 operated cases. *Neurosurgery*, 1989; 25: 153–160.
- 4) Yamashita J, Handa H, Iwaki K, Abe M. Recurrence of intracranial meningiomas, with special reference to radiotherapy. *Surg Neurol*, 1980; 14: 33–40.
- 5) Arseni MC, Ionesco S. [Spinal cord compression caused by intraspinal tumors; clinico-statistical study of 362 cases]. *J Chir (Paris)*, 1958; 75: 582–594.
- 6) Fortuna A, Gambacorta D, Occhipinti EM. Spinal extradural meningiomas. *Neurochirurgia (Stuttg)*, 1969; 12: 166–180.
- 7) Bret P, Lecuire J, Lapras C, Deruty R, Dechaume JP, Assaad A. [Intraspinal meningiomas. A series of 60 cases]. *Neurochirurgie*, 1976; 22: 5–22.
- 8) Levy WJ, Jr., Bay J, Dohn D. Spinal cord meningioma. *J Neurosurg*, 1982; 57: 804–812.
- 9) Frank BL, Harrop JS, Hanna A, Ratliff J. Cervical extradural meningioma: case report and literature review. *J Spinal Cord Med*, 2008; 31: 302–305.
- 10) Wu L, Yang T, Deng X, Yang C, Zhao L, Yao N, *et al.* Spinal extradural en plaque meningiomas: clinical features and long-term outcomes of 12 cases. *J Neurosurg Spine*, 2014; 21: 892–898.
- 11) Tuli J, Drzymalski DM, Lidov H, Tuli S. Extradural en-plaque spinal meningioma with intraneural invasion. *World Neurosurg*, 2012; 77: 202.e205–213.
- 12) Cugati G, Singh M, Pande A, Ramamurthi R, Balasubramanyam M, Sethi SK, *et al.* Primary spinal epidural lymphomas. *J Craniovertebr Junction Spine*, 2011; 2: 3–11.
- 13) Yamada S, Kawai S, Yonezawa T, Masui K, Nishi N, Fujiwara K. Cervical extradural en-plaque meningioma. *Neurol Med Chir (Tokyo)*, 2007; 47: 36–39.
- 14) Nakamura M, Tsuji O, Fujiyoshi K, Hosogane N, Watanabe K, Tsuji T, *et al.* Long-term surgical outcomes of spinal meningiomas. *Spine (Phila Pa 1976)*, 2012; 37: E617–623.
- 15) Rasmussen TB, Kernohan JW, Adson AW. Pathologic classification, with surgical consideration, of intraspinal tumors. *Ann Surg*, 1940; 111: 513–530.
- 16) Imagama S, Ito Z, Wakao N, Sakai Y, Kato F, Yukawa Y, *et al.* Differentiation of localization of spinal hemangioblastomas based on imaging and pathological findings. *Eur Spine J*, 2011; 20: 1377–1384.
- 17) Imagama S, Ito Z, Ando K, Kobayashi K, Hida T, Ito K, *et al.* Rapid Worsening of Symptoms and High Cell Proliferative Activity in Intra- and Extramedullary Spinal Hemangioblastoma: A Need for Earlier Surgery. *Global Spine J*, 2016; DOI 10.1055/s-0036-1580612.
- 18) Cohen-Gadol AA, Zikel OM, Koch CA, Scheithauer BW, Krauss WE. Spinal meningiomas in patients younger than 50 years of age: a 21-year experience. *J Neurosurg*, 2003; 98: 258–263.
- 19) Ito K, Aoyama T, Miyaoka Y, Seguchi T, Horiuchi T, Hongo K. Surgery for ventral intradural thoracic spinal tumors with a posterolateral transpedicular approach. *Acta Neurochir (Wien)*, 2016; 158: 1563–1569.