



Spinal Extradural Meningioma: A Case Report and Review of the Literature

척수 경막외 뇌수막종: 증례 보고와 문헌고찰

Wonju Hong, MD¹, Eun Soo Kim, MD^{1*}, Yul Lee, MD¹, Kwansop Lee, MD¹, Sung Hye Koh, MD¹, Hwyoung Song, MD¹, Mi Jung Kwon, MD²

Departments of ¹Radiology, ²Pathology, Hallym University Sacred Heart Hospital, Hallym University College of Medicine, Anyang, Korea

Spinal meningiomas account for 12% of all the meningiomas and are usually located in the intradural extramedullary space. In some cases, they are associated with some extradural extensions. However, purely extradural spinal meningiomas are rare. Additionally, it is difficult to make an accurate preoperative diagnosis. We report a case of pathologically confirmed atypical meningioma, presented as a posterior epidural mass on the thoracic spine. We review the case, clinical symptoms, radiologic findings and the histologic features.

Index terms

Spine
Meningioma
Epidural Space
Adult

Received September 5, 2017

Revised October 25, 2017

Accepted December 23, 2017

*Corresponding author: Eun Soo Kim, MD
Department of Radiology, Hallym University Sacred Heart Hospital, Hallym University College of Medicine, 22 Gwanpyeong-ro 170beon-gil, Dongan-gu, Anyang 14068, Korea.
Tel. 82-31-380-5985 Fax. 82-31-380-3878
E-mail: silwater007@hallym.or.kr

This is an Open Access article distributed under the terms of the Creative Commons Attribution Non-Commercial License (<http://creativecommons.org/licenses/by-nc/4.0>) which permits unrestricted non-commercial use, distribution, and reproduction in any medium, provided the original work is properly cited.

INTRODUCTION

The diagnosis of the spinal meningiomas is not very difficult based on radiologic findings and typical locations. However, diagnostic difficulties occur when the meningioma has atypical imaging characteristics or is in an unfamiliar location. Spinal meningiomas, which accounts for approximately 12% of all meningioma, are mostly intradural extramedullary in location (1). Some cases of intradural spinal meningiomas are associated with extradural extensions; however, it is uncommon to have pure epidural location. Extradural spinal meningiomas account for only 2.5% to 3.5% of all spinal meningioma (2). We report a case of pathologically confirmed spinal epidural meningioma. Additionally, we review the literature and the radiographic features of meningioma in unusual locations.

CASE REPORT

A 58-year-old woman presented with progressive weakness and paraesthesia of both lower legs for four months. The woman showed paraparesis of motor Grade IV on both legs and had difficulty maintaining her balance due to weakness. She had hyperesthesia and dysesthesia on light touch, pain, and temperature sense below the T9 dermatome level. The position and vibration senses were intact. She did not have symptoms related to sphincter dysfunction.

A spine MRI demonstrated a well-defined mass approximately 3.0 × 1.0 cm in diameter in the posterior epidural space of the seventh thoracic vertebral level on sagittal T1-weighted (Fig. 1A) and T2-weighted images (Fig. 1B). The mass caused compression and signal change of the spinal cord, suggesting compressive myelopathy. However, adjacent bone marrow was intact with no evidence of osseous infiltration. T1-weighted im-

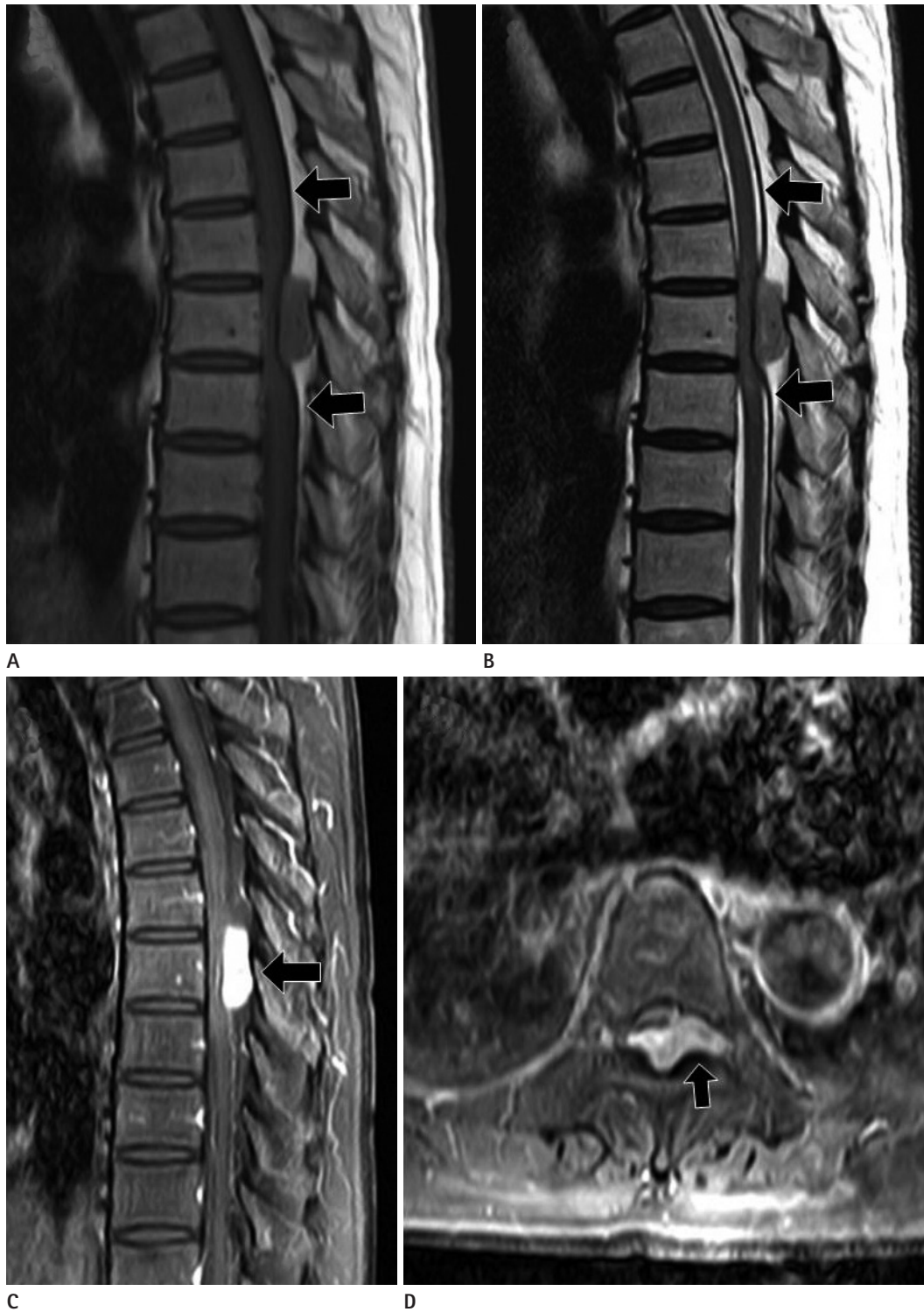


Fig. 1. MRI and pathologic findings of spinal extradural meningioma in a 58-year-old woman who has progressive weakness of both lower legs for 4 months.

A, B. The T1-weighted image (**A**) and the T2-weighted image (**B**) display the posterior epidural mass with a similar signal intensity to the spinal cord. The subarachnoid space is obliterated at the level of the mass and the spinal cord is compressed. The dura mater is seen as a dark line (arrows) separating the extradural mass from the intradural structures. Elevated signal change, within the compressed spinal cord, represents compressive myelopathy on the T2-weighted image (**B**).

C, D. Contrast-enhanced sagittal (**C**) and axial (**D**) T1-weighted images depict strong and homogeneous contrast enhancement of the posterior epidural mass (arrows). Note the complete filling of the posterior epidural space and the extension to the left neural foramina of the vertebrae (**D**).

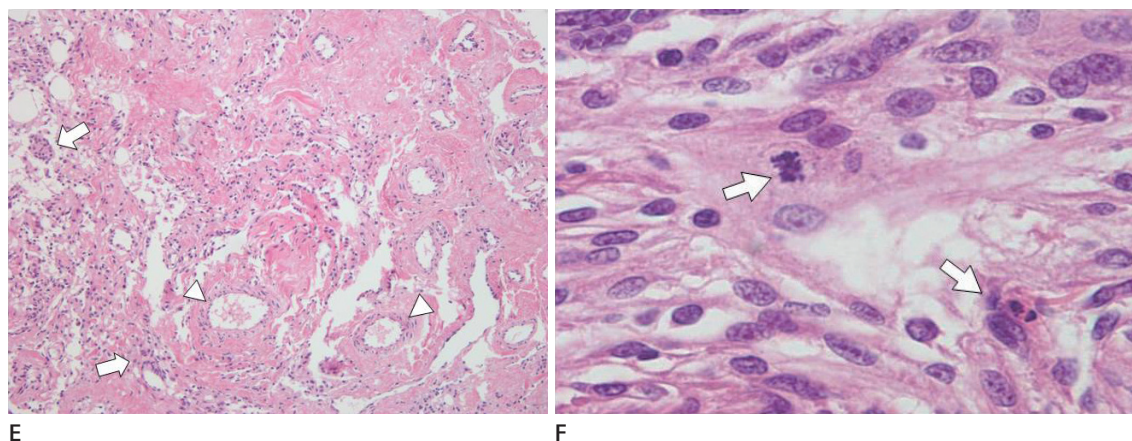


Fig. 1. MRI and pathologic findings of spinal extradural meningioma in a 58-year-old woman who has progressive weakness of both lower legs for 4 months.

E. Histologically, the tumor was dominated by multiple vessels (arrowheads) interspersed with small meningotheelial tumor cells (arrows) and focal necrosis (hematoxylin and eosin, $\times 100$).

F. Mitotic figures (arrows) are frequently identified, despite bland cytologic features of the tumor cells (hematoxylin and eosin, $\times 400$).

ages with contrast enhancement showed strong enhancement of the mass with a left neural foraminal extension of the T7 vertebrae (Fig. 1C, D). Preoperatively, the radiologic differential diagnosis was lymphoma or metastatic tumor, but no primary lesion was found in the quick preoperative radiologic evaluation.

The patient received excision with partial laminectomy of T7. Microscopic examination of the epidural space mass revealed an atypical meningioma. Histologically, the tumor was dominated by multiple vessels interspersed with small meningotheelial tumor cells. Focal necrosis was also seen (Fig. 1E). The tumor cells showed increased mitoses up to 10 per 10 high-power fields despite bland cytological features (Fig. 1F). Tumor cells were also positive for epithelial membrane antigen (not shown). The final histological diagnosis was an atypical (WHO Grade II) meningioma.

After the surgery, the patient did not undergo further treatment such as post-operative radiotherapy. Nonetheless, the neurologic symptoms were relieved, and no evidence of tumor recurrence was shown on three month follow up CT.

DISCUSSION

Spinal meningiomas represent approximately 12% of all meningioma (1). Most meningiomas are intradural tumors that arise at the thoracic level. In the spine, meningiomas are thought to arise from the denticulate ligaments and more than 95% are classified as WHO Grade I (3). Extradural component is thought to

be extended from an intradural mass. It is uncommon for a spinal meningioma to have an epidural location; in fact, exclusively extradural spinal meningiomas are infrequent and account for only 2.5% to 3.5% of all spinal meningioma (2).

Spinal extradural meningiomas show the same histology, peak incidence in the fifth and sixth decades, the same frequent location in the thoracic spine and the same well-known sex preponderance of female patients compared with intradural meningiomas (2).

The clinical findings of epidural meningiomas are not significantly different from those of intradural located ones. Patients may present with back pain, sensory and motor changes, and finally sphincter disturbances in late phases (2).

MRI is the best imaging modality to diagnose spinal meningiomas. It delineates the location and extent of the tumor, guiding the plan of the surgery. However, extradural spinal meningiomas are uncommon, and they can be mistaken as others. It affects the extent of the surgery and patient's outcome, so the diagnosis of the extradural spinal mass needs to be carefully done.

Spinal meningiomas have typical features on MRI. On T1-weighted images, the mass often has signal intensity similar to that of the spinal cord and exhibits mildly increased signal intensity on T2-weighted images (1). In addition, the pattern of contrast enhancement is strong and homogeneous. Foraminal extension does not favor diagnosis of a meningioma over diagnosis of a schwannoma or neurofibroma (1). The latter two lesions usually demonstrate high signal intensity on T2-weighted

Table 1. Review of the Previously Reported Cases of Spinal Extradural Meningioma

Author Year	Patient No.	Age/ Sex	Location (Level)	Clinical Presentation	CT, MRI Findings	Pathology	Treatment	Outcome
Suzuki 2002	1	58/M	T10–T11–T2	Normal exam, Abnormal mass on the chest radiograph		Fibroblastic meningioma	T11–12 hemilaminectomy + thoracoscope	Complete excision
Restrepo 2006	1	57/F	C7–T2	Loss of strength in lower extremity, increase in reflex	MRI: A homogeneous enhancing soft tissue from an infiltrative process at C7–T2 area involving the epidural space, predominantly on the right side, compressing the cord	Psammomatous meningioma, infiltrations	Total laminectomy + adjuvant RT	No recurrence
Frank et al. 2008 (4)	1	45/F	C5–C7	Chronic posterior cervicothoracic pain, mild bladder disturbance	MRI: An enhancing epidural mass with spinal cord displacement and compression	Psammomatous meningioma	C5–7 laminectomy and left sided foraminotomies with C5–T1 instrumentation	No recurrence, no symptoms
Santiago 2009	1	42/M	T2–T3	Paraparesis	MRI: Posterior and left lateral epidural mass at T2–T3 with foraminal extension and bone remodeling. Isointense on T1WI and T2WI with intense and homogeneous enhancement	Psammomatous meningioma	T2–T3 laminectomy	Complete excision
Kim 2011	1	50/M	T6–T7	Paresthesia at T6	MRI: An intraspinal extradural tumor located from the T6 vertebral body to the upper margin of the 7th vertebral body, dumbbell-like through the intervertebral foramen	Meningothelial meningioma	Hemilaminectomy of T6	Recovery of the symptom
Tuli 2012	1	42/F	T5–T6	Progressive lower leg weakness and numbness below T3 for 4 months	MRI: An extradural lesion compressing the cord from T4 through T6, homogeneous enhancement	WHO Grade I meningioma	T4–6 laminectomy	Full recovery of motor function
Yaldiz 2014	1	48/F	C7–T2	Hypoesthesia under T1	CT: A mass originating from the spinal canal and extending to the hemithorax from the right paravertebral area at the level of the T1 vertebra and extending toward the T1 bony spinal canal MRI: Dumbbell appearance mass, hyperintense mass on T2WI, hypointense on T1WI with homogeneous enhancement	Psammomatous meningioma	T1 total laminectomy	No recurrence

Table 1. Review of the Previously Reported Cases of Spinal Extradural Meningioma (continued)

Author Year	Patient No.	Age/ Sex	Location (Level)	Clinical Presentation	CT, MRI Findings	Pathology	Treatment	Outcome
Jeong et al. 2014 (5)	1	49/F	T12	Low back and left leg pain for 5 years, hypesthesia of the left leg without motor deficit	MRI: A 1.8cm extradural mass with rim enhancement in the spinal canal CT: Extension to the midline and the left T12-L1 foramen, dense calcification within the mass	Psammomatous meningioma	T12 total laminectomy, linear durotomy at the midline	N/A
Yang et al. 2016 (6)	1	55/F	T6-T8	2-year progressive back pain and weakness in both legs	MRI: A well-circumscribed oval lesion in dorsal-lateral space (isointense on T1WI, hyperintense on T2WI, homogeneous enhancement with dural tail sign)	Angiomatous meningiomas	T6-8 laminectomy	Motor weakness improved, no recurrence
Bettaswamy et al. 2016 (7)	2	50/M	C2-C4	Neck pain with tingling and burning paresthesia of all the four limbs for 8 months, progressive spastic quadripareiss with bladder disturbance for 1 week	MRI: An extradural en plaque lesion from C1 to C4, transverse through the right C2/3 intervertebral neural foramen, encasing the vertebral artery, iso-to hypointense on T1WI, isointense on T2WI with intense enhancement	Meningothelial meningioma	C2-4 laminectomy	Completely symptom free, no recurrence
Khayal et al. 2017 (8)	1	9/F	T5-T7	Tingling sensation in the right hand with progressively increasing spastic quadriparesis for 4 months, neck movement restriction and strain during micturition for 2 months Sudden progressive spastic paraparesis of lower limbs with bilateral feet drop, unable to walk	MRI: An extradural en plaque lesion extending from C3 to C7 on the left side, hypointense on T1WI and T2WI with intense enhancement, no transforaminal extension or widening MRI: A 40 x 18mm homogenous enhancing mass with isointense on T1WI, hyperintense on T2WI at anterior extradural space on left side causing compression and displacement of spinal cord, with posterior wall vertebral body erosion and destruction of the posterior element	Meningothelial meningioma Choroid meningioma	Laminectomy T5-T7 laminectomy	Significant alleviation in spasticity and improvement in motor power at 6 months follow up, no recurrence Recovery of muscular strength to power Grade 3

N/A = not available, RT = radiotherapy, T1WI = T1-weighted images, T2WI = T2-weighted images

images with cystic change and inhomogeneous enhancement (1). Therefore, the two tumors are easy to exclude. In addition, lymphoma, a malignant epidural mass, is the other misdiagnosis of meningioma due to homogeneous and strong enhancement and low signal intensity on T2-weighted images due to its dense cellularity (2, 3, 5). Moreover, hyperostosis is observed less frequently in patients with epidural spinal meningiomas than in patients with cranial meningiomas (9). Hyperostosis was not clear in our case.

We have reviewed other 12 cases of spinal extradural meningiomas previously reported since 2002 in Table 1. They were most commonly located in the thoracic spine followed by the cervical spine, and patients showed predominance of female. On imaging studies, cases appeared as intraspinal extradural masses with mostly homogeneous enhancement. In most cases, the mass showed extradural en plaque lesion suggesting the dural based origin of the mass, also known as dural tail sign, making it dumbbell shape appearance. Nevertheless, our case did not present any signs that clarified that the mass originated from the dura. Compared to other reported cases, our case showed more well-delineated margin. These findings made it more difficult to make the accurate diagnosis and it could easily be mistaken as other extradural tumor such as lymphoma.

Pathologically, spinal meningiomas tend to be well-defined discrete lesions with a dural attachment. Microscopically, the common patterns are meningothelial, fibroblastic, transitional, and psammomatous and most case reports describe features of a meningothelial or psammomatous meningioma (2, 7). The WHO classification of meningiomas are divided into three grades. Among many subtypes, meningothelial, fibrous and transitional are the most common, and belong to Grade I. Our patient's pathology report suggested atypical meningioma (WHO Grade II) with increased mitotic activity, focal necrosis and frequent vascular tumor emboli. Among reviewed 12 cases in Table 1, there is no case confirmed as atypical meningioma. Apart from one case of choroid meningioma (WHO Grade II) occurred in a 9-year-old girl, all of the other cases were Grade I meningiomas.

In literature review of major spinal meningioma series, including both intra- and extradural subtypes, recurrence rates of surgical excision are reported as 3% to 7% (2). Although, there

was no case which showed recurrence of the tumor among the reviewed 12 cases, the recurrence rate after surgery for extradural meningioma is four times higher than that of intradural meningioma (4, 10). Our patient yet did not show any tumor recurrence for three months, but careful observation and follow up is still necessary.

In conclusion, we describe here a rare case of spinal extradural meningioma in the thoracic level with atypical one (WHO Grade II). However, it must be not forgotten that meningiomas may occur in rare locations and with variable imaging characteristics.

REFERENCES

1. Haranhalli N, Nakhla JP, Yassari R, Kinon MD. Radiographic pearls in the evaluation of an extradural thoracic meningioma: a case report. *Cureus* 2017;9:e1031
2. Ben Nsir A, Boughamouira M, Mahmoudi H, Kilani M, Hattab N. Uncommon progression of an extradural spinal meningioma. *Case Rep Surg* 2014;2014:630876
3. Plank C, Koller A, Mueller-Mang C, Bammer R, Thurnher MM. Diffusion-weighted MR imaging (DWI) in the evaluation of epidural spinal lesions. *Neuroradiology* 2007;49:977-985
4. Frank BL, Harrop JS, Hanna A, Ratliff J. Cervical extradural meningioma: case report and literature review. *J Spinal Cord Med* 2008;31:302-305
5. Jeong SK, Seong HY, Roh SW. Extra-intradural spinal meningioma: a case report. *Korean J Spine* 2014;11:202-204
6. Yang T, Wu L, Yang C, Xu Y. Epidural angiomatous meningioma of the thoracic spine: a case report. *Oncol Lett* 2016;11:458-460
7. Bettaswamy G, Ambesh P, Das KK, Sahu R, Srivastava A, Mehrotra A, et al. Extradural spinal meningioma: revisiting a rare entity. *J Craniovertebr Junction Spine* 2016;7:65-68
8. Khayal HB, Abograra A, Iashhab M. Extradural spinal meningioma in a nine-year-old girl: a case report and review of the literature. *Open Access J Neurol Neurosurg* 2017;5:555659
9. El Khamary SM, Alorainy IA. Case 100: spinal epidural meningioma. *Radiology* 2006;241:614-617
10. Klekamp J, Samii M. Surgical results for spinal meningiomas. *Surg Neurol* 1999;52:552-562

척수 경막외 뇌수막종: 증례 보고와 문헌고찰

홍원주¹ · 김은수^{1*} · 이 열¹ · 이관섭¹ · 고성혜¹ · 송화영¹ · 권미정²

척수 수막종은 전체 수막종의 12% 정도를 차지하며 주로 경막내 수외 공간에 위치한다. 어떤 경우에는 경막 외로 연장되기도 한다. 그러나 온전하게 경막외에 위치한 수막종은 드물며, 수술 전 정확한 진단을 내리기 어려운 경우가 있다. 우리는 흉추의 후방 경막외 종양으로 나타난, 병리학적으로 확진된 비정형 수막종 증례를 보고하고 그 임상적 증상과 영상학적 소견, 조직학적 특징에 대해 고찰하고자 한다.

한림대학교 의과대학 한림대학교성심병원 ¹영상의학과, ²병리과