



Multiple Extradural Spinal Meningiomas in a Patient with Acquired Immunodeficiency Syndrome: Case Report and Literature Review

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Key words

- Epidural
- Extradural
- Meningioma
- Spine oncology

Abbreviations and Acronyms

HIV: Human immunodeficiency virus

MRI: Magnetic resonance imaging

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INTRODUCTION

Spinal meningiomas compose 30.7% of intradural extramedullary tumors of the spine and 7.9% of all meningiomas.¹ Purely extradural spinal meningiomas are uncommon, reportedly accounting for only 2.5%–9.3% of cases.²⁻⁴ The true incidence of purely extradural meningiomas is difficult to ascertain, given that many previous studies that reported extradural meningiomas did not specify whether an intradural component was present.⁵⁻⁷ To our knowledge, this is the first case of multiple purely extradural meningiomas occurring at distinct levels of the spine. We performed a literature review of patients with extradural meningiomas. Because our patient had this unusual distribution of tumors and had a history of human immunodeficiency virus (HIV), we also conducted a literature review of articles on patients with HIV and meningiomas.

CASE PRESENTATION

A 40-year-old male presented with a 2-month history of progressive back pain

■ **BACKGROUND:** Purely extradural spinal meningiomas are uncommon. Due to their typical location in the neural foramen, they are often mistaken for schwannomas, neurofibromas, and epidural metastases. In addition, comorbid conditions such as immunodeficiency may obscure the diagnosis. We present a case of extradural spinal meningiomas in a patient with human immunodeficiency virus (HIV). This is the first reported case of multiple extradural spinal meningiomas in 2 separate regions of the spine.

■ **CASE DESCRIPTION:** A 40-year-old male with a past medical history of HIV and hepatitis B infection presented with a 2-month history of progressive back pain radiating to the left flank and thigh. Magnetic resonance imaging of the thoracic and lumbar spine with intravenous gadolinium contrast revealed 2 extramedullary masses in the left neural foramina of T6 and L1. The patient underwent laminectomy, which revealed that the 2 lesions were entirely extradural. Both lesions were resected, and the histological diagnosis for both lesions was meningioma, World Health Organization grade I.

■ **CONCLUSIONS:** Our experience with this 40-year-old male with AIDS who presented with radicular symptoms due to multiple purely extradural meningiomas underscores the importance of considering meningioma as a possible diagnosis in patients with tumors of the neural foramina. In addition, a wide differential diagnosis should be made for patients with spinal lesions and history of HIV, including illnesses that are related to immunodeficiency and those that are not.

and radiation of pain to the left flank and thigh. Clinical examination revealed generalized left lower extremity weakness with motor strength grade 4 out of 5. No sensory or myelopathic signs were present on examination. The patient's past medical history included HIV and hepatitis B, both diagnosed at 1 month before presentation when he sought initial treatment for back pain. He had a CD4 count of 59 and a hepatitis B DNA count >170 million. Magnetic resonance imaging (MRI) of the thoracic and lumbar spine with intravenous gadolinium contrast revealed 2 extramedullary masses on the left side at T6 and L1 (Figure 1). Both masses encased their respective nerve roots and extended through the neural foramina. MRI of the full neural axis was negative for any other lesions. Given the patient's medical history and presence of 2 isolated masses, infection and lymphoma were

considered as possible diagnoses. Infectious laboratory specimens and titers were sent from the patient's serum and cerebrospinal fluid (obtained with image-guided lumbar puncture to avoid the lesional level). With the exception of the known HIV and hepatitis B infections, all infectious workups were negative.

The patient was taken to the operating room and underwent laminectomy with the goal of decompression and resection of both lesions. Intraoperatively, no obvious destruction of the lamina was noted at either location. After laminectomy, the emergence of a firm and pink-colored mass was noted in the lateral recess with extension through the left neural foramina at both T6 and L1. An ultrasonic aspirator was used to debulk and resect both masses in a piecemeal fashion. An external capsule was noted with an easily dissectible plane between

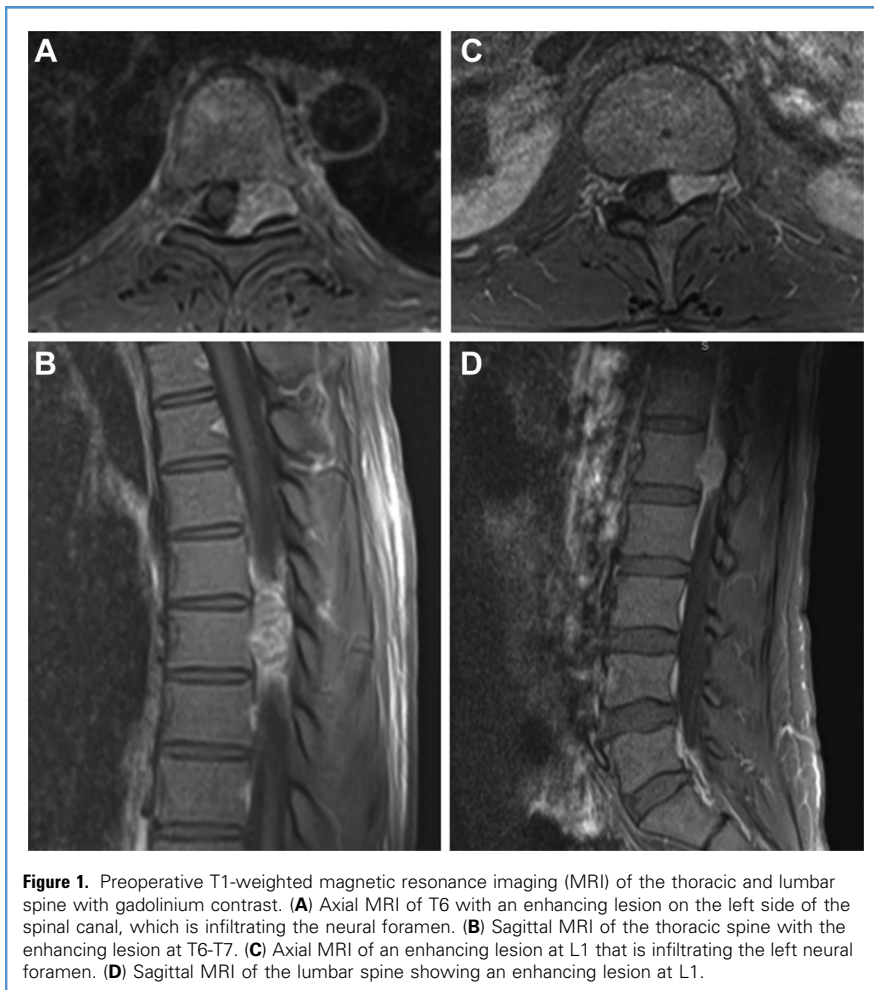


Figure 1. Preoperative T1-weighted magnetic resonance imaging (MRI) of the thoracic and lumbar spine with gadolinium contrast. (A) Axial MRI of T6 with an enhancing lesion on the left side of the spinal canal, which is infiltrating the neural foramen. (B) Sagittal MRI of the thoracic spine with the enhancing lesion at T6-T7. (C) Axial MRI of an enhancing lesion at L1 that is infiltrating the left neural foramen. (D) Sagittal MRI of the lumbar spine showing an enhancing lesion at L1.

the dura/nerve sheath and the mass. After removal of both lesions, sclerosis and bony effacement of the vertebral body was noted at each level without obvious bony destruction. Intraoperative frozen pathology revealed a spindle cell mass with a preliminary diagnosis of neurofibroma vs. schwannoma. Intraoperative infectious cultures were also obtained given the history of immunodeficiency; however, no obvious purulence was encountered.

Gross total resection of both masses was achieved without the need for intradural access. Intraoperative infectious cultures were negative. Histological analysis revealed spindle cells with spindle-shaped nuclei and prominent pink cytoplasm on hematoxylin and eosin staining. Immunohistochemical stains for both masses stained positive for epithelial membrane antigen and negative for S100, SOX10, and CD34. The final pathological

diagnosis for both lesions was meningioma, World Health Organization grade 1.

Postoperatively, the patient reported resolution of radicular symptoms and improved strength in the lower extremities. At a 6-month follow-up, the patient remained asymptomatic, and MRI of the neural axis demonstrated gross total resection with no residual or recurrent disease (Figure 2).

DISCUSSION

Extradural Spinal Meningiomas

Multiple meningiomas occur in 4%–9% of patients with meningiomas in the absence of neurofibromatosis.^{8,9} Spinal meningiomas occur most commonly in the thoracic region (73%), followed by the cervical (17%), and lumbar (7%) regions.^{10–13} Only 64 cases of purely

extradural spinal meningiomas have been reported. The proportion of spinal meningiomas located in the cervical spine is much greater in extradural meningiomas. The 44 cases in which location was disclosed included 43% (n = 19) in the cervical spine, 45% (n = 20) in the thoracic spine, 1 case was in the lumbar spine, and 2 cases were in the sacral spine (Figure 3).^{2–7,10,14–38} Other reports of extradural meningiomas included an intradural component.

In the only previous report of multiple extradural spinal meningiomas, the lesions were located at adjacent levels (T4-T5 and T5-T6), whereas in our patient the meningiomas were located in distinct locations (T6 and L1).² Levy et al.¹³ described a patient with a questionable pure extradural meningioma who was later found to have a residual intradural component requiring re-exploration, and thus did not have multiple extradural spinal meningiomas.

Both of the meningiomas in our patient were intraforaminal and involved the periradicular dural sleeves. Of the 64 previously reported cases, 22 had involvement of the nerve root and dural sleeves.^{2,5–7,18–21,24,25,27–29,31,35,37,39} There are 2 theories regarding the propensity for extradural spinal meningiomas to occur in proximity to the nerve root.³⁵ One of these theories suggests that extradural meningiomas may arise from the portion of the nerve sheath in which the arachnoid comes in contact with the dura mater. The other theory suggests that they arise from extradural embryonic arachnoid rests located in this region.^{6,21}

Meningiomas and HIV

This report is the first case of extradural spinal meningiomas in a patient with HIV. Our literature review revealed only 6 cases of meningiomas in patients with HIV, which are summarized in Table 1.^{40–44} A recently published case-control study by Samson et al.⁴⁵ noted a possible link between HIV and intracranial meningiomas; the authors found a 5-fold increased risk of developing high-grade meningioma (World Health Organization grade 2/3) for patients seropositive for HIV. A true association between meningioma and HIV has not been established, however.

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