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Case Report

Circumferential intradural meningioma of the thoracic spinal cord

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Abstract

BACKGROUND AND CONTEXT: There are very few reported cases of a meningioma circumferentially surrounding the spinal cord. To date, this entity has only been described at the conus medullaris and in the cervical cord. Herewith, the authors describe a case of an intradural extramedullary meningioma that completely encircled the *thoracic* spinal cord.

CASE REPORT: A 40-year-old woman with progressive numbness of the lower limbs and spasticity of gait following a fall presented to our hospital. Magnetic resonance imaging of the spine demonstrated an abnormality at T6–T7 completely encircling the spinal cord. The patient underwent a T6–T8 laminectomy and subtotal resection of the intradural partially calcified lesion. Resection of the anterolateral portion was not feasible. Histology revealed psammomatous meningioma (WHO Grade 1). The patient recovered well and was discharged with improved gait but some residual numbness of her feet and right hemithorax.

CONCLUSION: This is the first reported case of an intradural extramedullary meningioma completely encircling the *thoracic* spinal cord. Achieving complete resection of this circumferential meningioma was not possible via a posterior approach. The optimum management of this condition is unknown; clearly, achieving symptomatic relief with adequate cord decompression is paramount; however, the long-term outcome and risk of recurrence in these cases, given their rarity and the difficulties in achieving complete resection, is unknown.

Introduction

Meningioma is a common form of spinal neoplasia, accounting for 25%–46% of cases [1]. The majority tend to be intradural, extramedullary, and within the thoracic region [1]. Typically well-circumscribed, discrete lesions, it is rare to encounter a meningioma that completely encircles the spinal cord. There are only three cases of full circumferential spinal meningioma reported in the literature: two in the cervical region [2,3] and one at the conus-medullaris [4] (Table). Here the authors present the first case of a fully circumferential intradural, extramedullary meningioma in the *thoracic* spinal cord.

Case report

A 40-year-old woman with known history of systemic lupus erythematosus presented to our hospital following recurrent falls. She had been experiencing progressive numbness in her lower limbs for approximately 18 months, and of the pelvis for approximately 4 months. Onset of the latter symptom coincided with a few episodes of both urinary and fecal urgency. She had also experienced significant weight loss of approximately 13 kg 6 months before presentation.

On examination she was found to have a spastic gait, but normal, symmetrical power to both lower limbs, with increased tone and bilaterally brisk deep tendon reflexes, extensor plantars, and bilateral ankle clonus. She had a sensory level at approximately T10. Upper limb examination was unremarkable.

Magnetic resonance imaging (MRI) of the spine showed an enhancing lesion within the subdural space, circumferentially encasing the spinal cord at the level of T6 and T7 without involvement of the surrounding bone (Fig. 1). Differential diagnosis of the lesion was felt to include lymphoma, or other primary or (secondary) spinal tumor. Less likely differentials were felt to be a tuberculous or inflammatory process.

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Table
Cases described in the literature

Case	Age, gender	Level	Degree of cord encircling	Relation to dura	Resection	Closure	Outcome
Present case	40Y, F	T6-T8	Fully circumferential	Intradural	Partial (anterior portion remains)	Dura left open	Recovery of function
Singh and Agrawal [3]	40Y, M	C1-C4	Fully circumferential	En-plaque	Partial (anterior portion remains)	Unspecified	Reoperation caused by neurologic deterioration
Carter et al. [2]	23Y, M	C2-C7	Fully circumferential	Subdural	Partial (anterior portion remains)	Duraplasty	Improvement to function (able to ride bike)
Stechison et al. [4]	63Y, F	Conus medullaris and cauda equina	Fully circumferential	En-plaque	Partial (anterior portion remains)	Dura left open	2 years: arachnoiditis and progressive paraplegia at T11
Gamache et al. [5]	63Y, F	C3-C6	*Partially* circumferential	En-plaque: (extension to ossified PLL and LF)	Possibly complete (not specified)	Dural patch	Slow improvement in sensory and motor function
Stechison et al. [4]	76Y, F	T5-T7	*Partially* circumferential (dorsal two thirds)	En-plaque	Possibly complete (not specified)	Dura left open	9 months: T5 radiculopathy

PLL, posterior longitudinal ligament; LF, ligamentum flavum.
Note: Cases of circumferential meningioma (partial or complete) found in the literature.

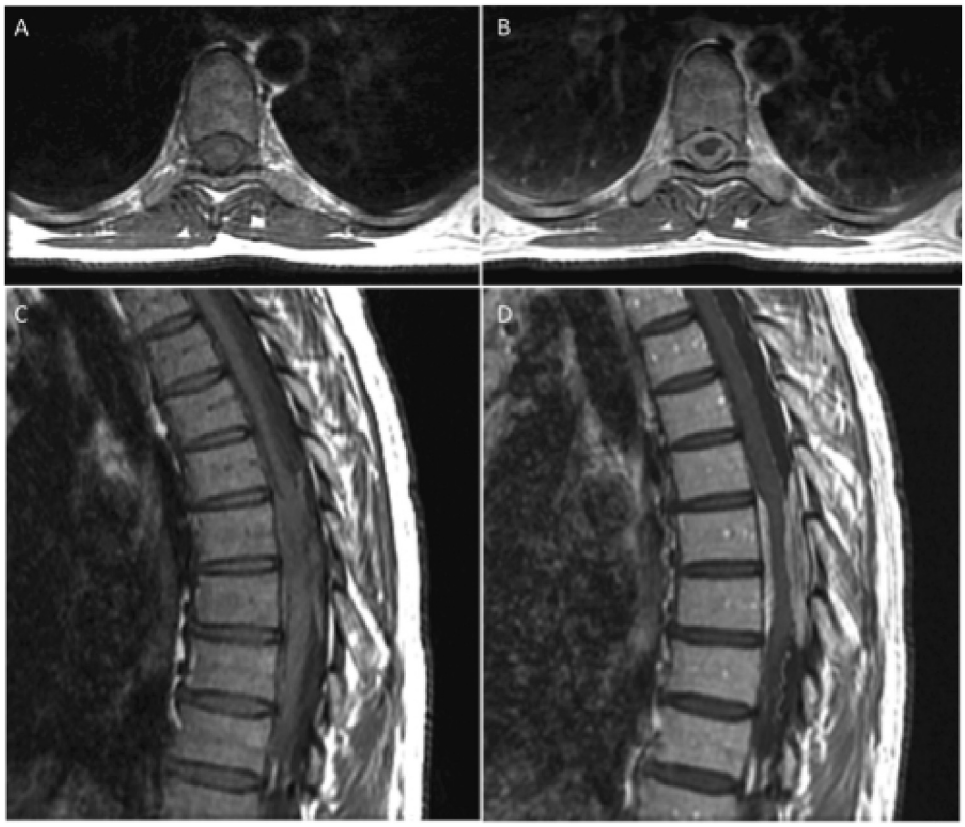

Fig. 1. Magnetic resonance imaging of the spine. Sagittal T1 pre- (A) and post-gadolinium (B) contrast. Axial T1 pre- (C) and post-gadolinium (D) contrast administration. A circumferential soft tissue abnormality at T6 and T7 vertebral levels completely surrounds the spinal cord. Signal abnormality at T7 vertebral level indicates compression. This soft tissue abnormality is in the subdural space rather than the epidural compartment. The lesion contained reasonably uniform contrast enhancement (A and B as well as C and D). There was no extension beyond the dural tube to involve the exit foramina or the adjacent vertebral marrow. There are also some prominent vessels on the dorsal aspect of the spinal cord cranial and caudal to the lumbar cord compression most likely representing congested veins. No other levels are involved.

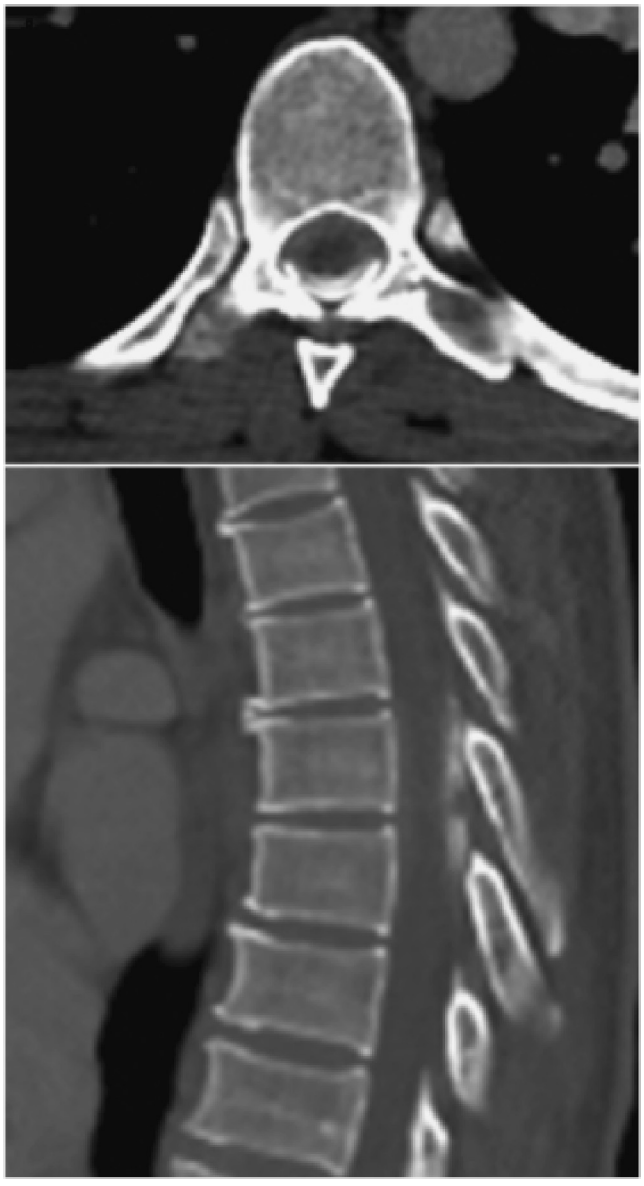

Fig. 2. Computed tomography (CT) of the chest, cropped to visualize the spine. (Top) Axial views. (Bottom) Sagittal views. Calcification is visible within the subdural space at the level of T6–T7, causing suspicion of calcified meningioma.

Given this, staging computerized tomography of the chest, abdomen, and pelvis was performed, which revealed an extensive calcification within the subdural space at the level of T6–T7, but there was no infiltration into the epidural space, or destruction of the adjacent mural arch or vertebral body (Fig. 2). There were no other distant abnormalities detected.

Given the unusual radiological appearances and clinical progression, the decision was made to proceed to urgent decompression and biopsy and debulking of the abnormality.

The patient then underwent a T6–T8 laminectomy; following a midline durotomy under microscopic vision, the dura was dissected free from a calcified layer beneath. This calcified layer was then divided longitudinally, revealing an associated extramedullary soft tissue lesion, which eventually dissected free from the spinal cord underneath. Histologic analysis of a frozen section biopsy supported the surgeon's intraoperative suspicion of meningioma. Posterolateral resection was achieved with further piecemeal dissection of heavily calcified areas with subsequent decompression of the spinal cord. However, accessing the remaining anterolateral portion of the tumor became progressively more challenging as the tumor was quite adherent to the cord and would risk unintended cord manipulation with possible serious neurologic sequelae for the patient; therefore, we elected to leave the remaining tumor intact. The dura was left open and repaired with dural substitute and fibrin sealant.

The patient recovered well postoperatively with no immediate complications. She received a short course of dexamethasone and was discharged home after physiotherapy input.

Histopathology revealed a meningothelial tumor with dense psammomatous microcalcifications. No atypical or malignant features were seen in the tumor and Ki67 nuclear labeling was very low. The diagnosis of a psammomatous meningioma (WHO Grade 1) was made (Fig. 3).

At 6 months' follow-up, the patient's gait had markedly improved; however, she was still complaining of numbness of her right hemithorax. She has returned to work and resumed fully her exercise regime. Postoperative baseline MRI confirmed reasonable decompression of the spinal cord with known residuum tumor anterior and lateral to the spinal cord as expected (Fig. 4). We aim to follow-up the patient with an annual spine MRI (if clinically stable). If there is any future growth, further treatment options will need to be explored, such as further surgery if feasible or radiotherapy to control further tumor growth.

Discussion

A review of the literature identified three cases of meningioma with complete [2–4], and two cases with partial [4,5]

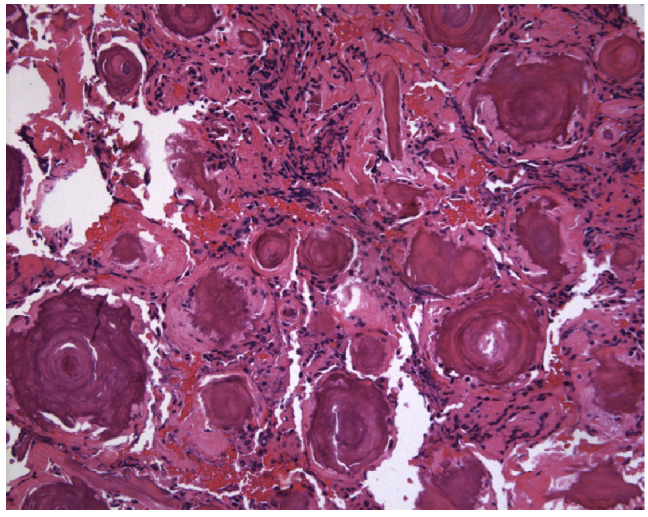

Fig. 3. Histology slides. Hematoxylin and eosin stain at 200× magnification. This reveals a meningothelial tumor with dense psammomatous microcalcifications without atypical or malignant features.

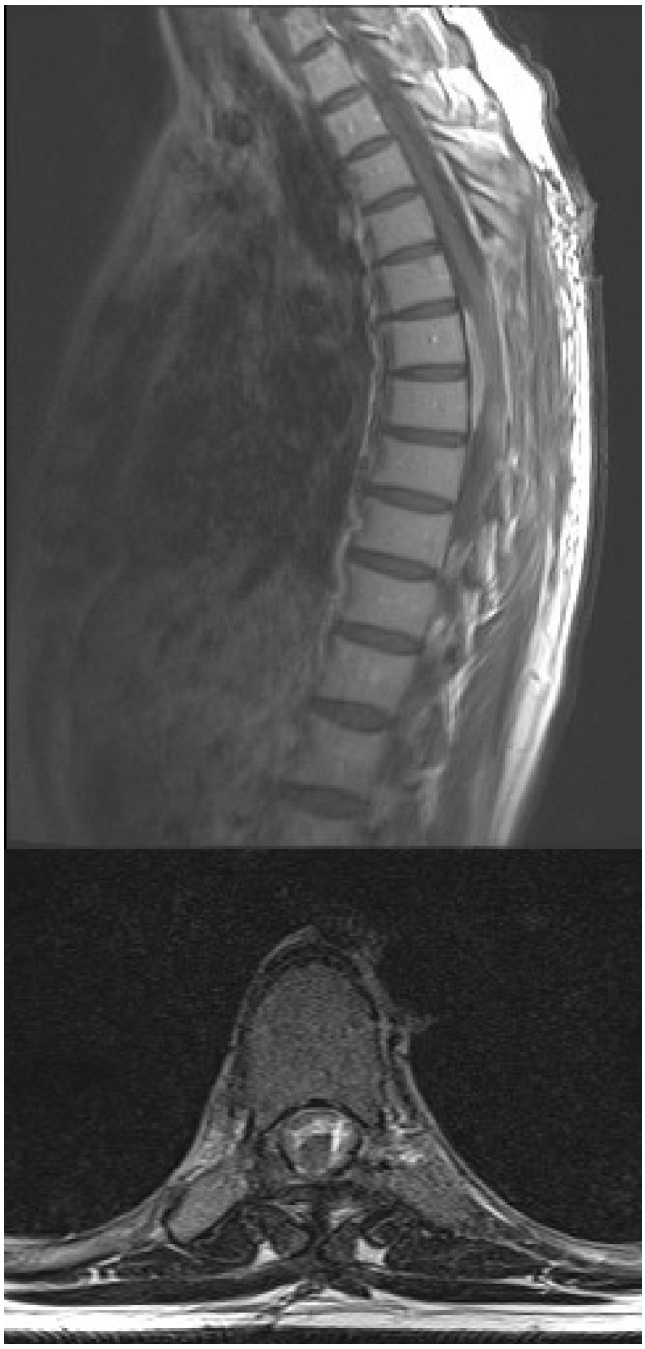

Fig. 4. Postoperative magnetic resonance imaging of the spine. (Top) Sagittal T1 post-gadolinium contrast administration. (Bottom) Axial T1 gadolinium contrast administration. There has been partial debulking of the intradural circumferential meningioma encircling the spinal cord at the T6 and T7 vertebral levels with some decompression posteriorly.

enclosure of the spinal cord (Table). Castro et al. [6] in their discussion of charged particle radiotherapy for tumors encircling the brain stem or spinal cord allude to two meningiomas of this nature. However, they do not discuss in more detail, and it is unknown at what level and how circumferential these lesions were. As such, these cases are not included in the Table. Of the reported cases, only three were fully circumferential: two in the cervical region [2,3], and one encircling the conus medullaris and cauda equina [4]. The authors believe the presented case is the first to report on a fully circumferential meningioma of the *thoracic* spinal cord.

Because of the unusual radiological appearances and the relative rarity of this disease, meningioma was initially not considered a likely primary diagnosis. Differentials of infection (such as tuberculosis) were considered because of the patient's travel history, and inflammatory disease was considered because of her history of systemic lupus erythematosus. Lymphoma was initially considered to be the most likely differential based on the diffuse soft tissue involvement and dense enhancement shown on spinal MRI. It was after computed tomography-identified calcification, being unusual for lymphoma, and initial macroscopic inspection, that meningioma was considered most likely, and confirmed with frozen section during the procedure.

A rare entity, the natural history of circumferential spinal cord meningioma condition is not known. Complete resection is optimal for spinal cord meningioma in general and is achievable in most normal circumstances (97% reported by Gezen et al. [1]). In a retrospective case series, Nakamura et al. [7] found recurrence rates of operated spinal meningiomas to be 9.7% in complete resection (Simpson grades [8] I and II; n=62), but all of the six patients with incomplete resection (Simpson III and IV) required reoperation, occurring on average after 5 years. Clearly, a complete resection is more challenging in fully circumferential lesions such as this, and given the existing literature, it is felt the risk of recurrence is high.

To achieve complete resection here would require accessing the anterior thoracic spinal cord, which typically requires a transthoracic procedure. Taghva et al. [9] describe minimally invasive vertebrectomy and instrumentation for circumferential decompression of a metastatic spinal cord lesion. However, this technical report describes only *decompression* rather than *resection* of a spinal cord tumor. In our case, a second, revision surgery by anterior approach could be considered, but because of the rarity of circumferential spinal meningioma, it is not known whether the risk of such invasive surgery is outweighed by the risk of recurrence. Furthermore, our patient had extensive calcification seen intraoperatively, a feature which proved to increase the risk of surgical morbidity according to Sandalcioglu et al. [10].

If a complete resection cannot be achieved, as in this case, symptomatic relief from cord compression is paramount. Stechison et al. [4] recommend leaving the dura open to prevent emergence of arachnoiditis. In their case it was this, rather than growth of the remaining anterior tumor that caused neurologic deterioration. In our case, the dura was left open to prevent postoperative swelling and cord compression in case of future recurrence.

None of the above-reported cases, including our own, received radiotherapy. However, Gezen et al. [1] advocate adjunctive radiotherapy after incomplete resection or recurrence.

The patient will require surveillance scans at regular intervals, and if a recurrence is identified, then further surgery

could be contemplated, if feasible, or else one would have to consider radiotherapy to prevent further tumor growth.

In view of the young age of the patient, the good neurologic recovery, the degree of decompression achieved, and the presence of heavy calcification within it, we felt that we should defer radiotherapy and only consider it in case of a recurrence.

Conclusion

To the authors' knowledge, the presented case is the first reported of a meningioma that completely encircled the thoracic spinal cord. Optimal management of this rare disease is unknown. Achieving symptomatic relief from via cord decompression is clearly an early target. Complete resection is naturally preferable in spinal meningioma but is very difficult to achieve in this circumstance; therefore, serial imaging and long-term follow-up will be required.

References

[1] Gezen F, Kahraman S, Canakci Z, Bedük A. Review of 36 cases of spinal cord meningioma. Spine 2000;25:727–31.

[2] Carter DA, Rowed DW, Lewis AJ. Subdural meningioma of spinal cord. Case report. J Neurosurg 1985;63:800–3. doi:10.3171/jns.1985.63.5.0800.

[3] Singh B, Agrawal A. En-plaque meningioma completely encircling the cervical cord: case report. Iran J Radiol 2009;6:159–61.

[4] Stechison MT, Tasker RR, Wortzman G. Spinal meningioma en plaque. Report of two cases. J Neurosurg 1987;67:452–5. doi:10.3171/jns.1987.67.3.0452.

[5] Gamache FW, Wang JC, Deck M, Heise C. Unusual appearance of an en plaque meningioma of the cervical spinal canal. A case report and literature review. Spine 2001;26:E87–9.

[6] Castro JR, Collier JM, Petti PL, et al. Charged particle radiotherapy for lesions encircling the brain stem or spinal cord. Int J Radiat Oncol Biol Phys 1989;17:477–84.

[7] Nakamura M, Tsuji O, Fujiyoshi K, et al. Long-term surgical outcomes of spinal meningiomas. Spine 2012;37:E617–23. doi:10.1097/BRS.0b013e31824167f1.

[8] Simpson D. The recurrence of intracranial meningiomas after surgical treatment. J Neurol Neurosurg Psychiatry 1957;20:22–39.

[9] Taghva A, Li KW, Liu JC, et al. Minimally invasive circumferential spinal decompression and stabilization for symptomatic metastatic spine tumor: technical case report. Neurosurgery 2010;66:E620–2. doi:10.1227/01.NEU.0000365270.23815.B1.

[10] Sandalcioglu IE, Hunold A, Müller O, et al. Spinal meningiomas: critical review of 131 surgically treated patients. Eur Spine J 2008;17:1035–41. doi:10.1007/s00586-008-0685-y.