



Case Report

## Extradural en-plaque spinal lipomatous meningioma: A case report and literature review

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### ABSTRACT

**Background:** Extradural spinal meningiomas are rare, and few appear as lipomatous meningiomas in an “en-plaque” form. The transitional fibroblastic subtype, with lipid accumulation within the tumoral cells, is typically more aggressive and surgically challenging to resect.

**Case Description:** A 42-year-old male presented with neck pain and progressive paresthesias in both upper extremities. Based on the radiological findings, the tentative preoperative diagnosis was lymphoma. However, the biopsy confirmed a meningioma. The patient underwent a combined extradural anterior and posterior approach, resulting in full tumor resection. Histopathologically, the final report documented a lipomatous meningioma.

**Conclusions:** Extradural spinal lipomatous meningiomas (i.e., “en-plaque”) are rare and typically result in rapid clinical deterioration. The radiological diagnosis may be difficult, while ultrasonography helps to define their extradural location facilitating planning for gross total anterior/posterior excision where indicated.

**Keywords:** Lipomatous meningioma, meningioma en-plaque, spinal meningioma

### INTRODUCTION

Spinal meningiomas account for approximately 25% of primary spinal cord tumors.<sup>[5]</sup> They are usually intradural extramedullary in location and have a predilection for the thoracic spine.<sup>[1]</sup> Morphologically, they are typically round but may occasionally appear “en-plaque;” the latter histopathological diagnosis is occasionally meningotheelial/psammomatous with lipomatous variants (e.g., additional presence of adipocytes). Gross total resection is often challenging and may warrant a combined anterior/posterior approach. Here, we describe a 42-year-old male who successfully underwent gross-total resection of a cervical lipomatous extradural “en-plaque” meningioma.

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## CASE REPORT

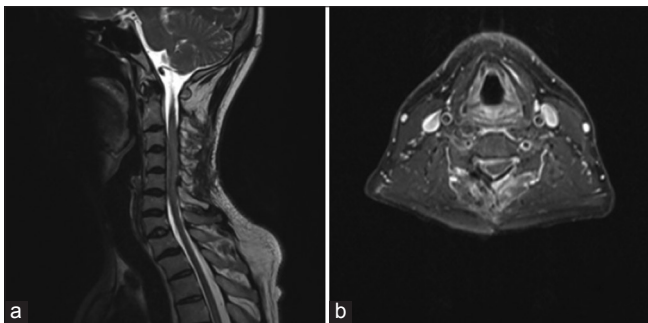
### Clinical and radiographic presentation

A 42-year-old male presented with neck pain, progressive paresthesias, tightness in both upper extremities, and 6 months of numbness involving the fourth and fifth digits of the left hand. His only neurological deficit was decreased pin appreciation in the left 4<sup>th</sup> and 5<sup>th</sup> digits. The magnetic resonance image (MRI) showed a circumferential tumor extending from C2 to C7 levels, resulting in maximal cord compression between C3 and C5. The lesion was isointense on both T1- and T2-weighted images and intensely homogeneously enhanced with gadolinium [Figure 1].

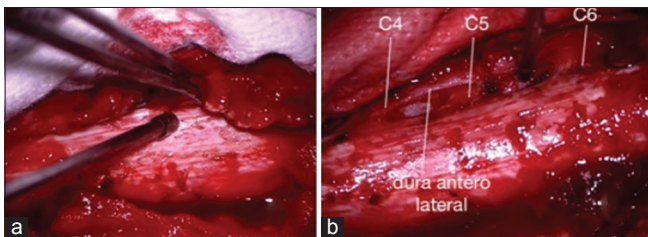
### Biopsy and surgery

After an open biopsy revealed a meningioma, the patient initially underwent a C3–C7 laminoplasty, with the removal of posterior tumor, and decompression of the dural sac. At surgery, the extradural tumor was grayish, white, non-suckable, and fibrous; it was carefully peeled off the dura [Figure 2]. The intraoperative ultrasound confirmed adequate posterior decompression of the spinal cord without the identification of any additional intradural mass [Figure 3]. Electrophysiological monitoring showed no significant changes during meticulous tumor removal.

When the postoperative MRI documented residual anterior tumor, the patient underwent a C4 and C5 anterior corpectomy/fusion to achieve gross total tumor resection.



**Figure 1:** (a) Magnetic resonance image (MRI) T2 sagittal view showing isointense round mass from C3 to C6. (b) MRI T1 with contrast axial view showing an enhancement cervical mass, encircling the dura.

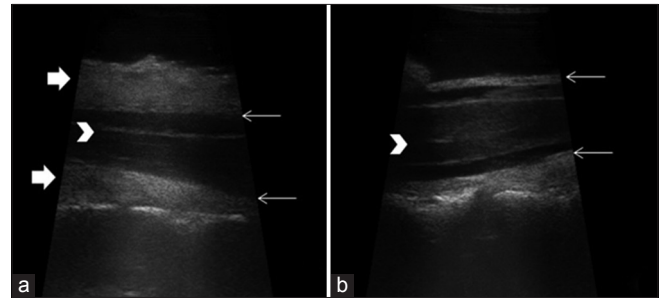


**Figure 2:** (a) Intraoperative image of posterior resection which shows a fibrous epidural mass, without dural infiltration. (b) Intraoperative image which shows cervical roots without tumor.

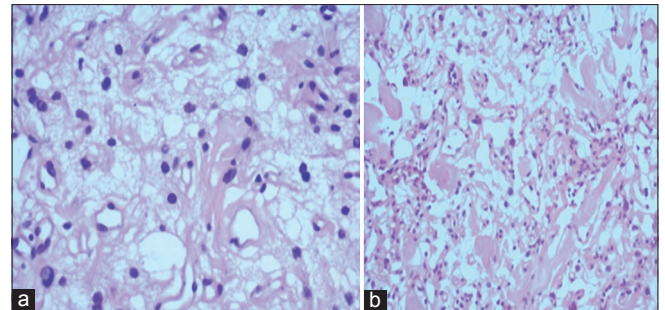
### Histopathology

Histopathological analysis revealed meningeothelial uniform cells accompanied by mature adipocyte-like cells with peripherally displaced nuclei (e.g., ovoid nuclei with stippled chromatin and eosinophilic cytoplasm with fat vacuoles in their cytoplasm). There was no necrosis, mitotic activity, or hemorrhage. These findings confirmed the diagnosis of a transitional lipomatous meningioma [Figure 4].

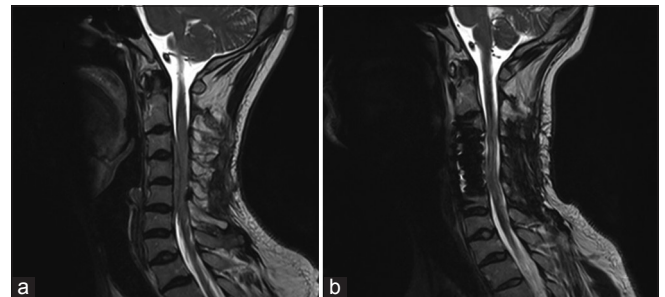
For 3 postoperative years, the patient has remained asymptomatic and follow-up MR studies showed no residual/recurrent disease [Figure 5].



**Figure 3:** (a) Intraoperative ultrasonography image showing the tumor (big arrows), the dura (thin arrows), and the epidural channel (arrowhead). (b) Ultrasonography image after posterior tumor removal, showing decompression of the spinal cord.



**Figure 4:** Hematoxylin and eosin section showing meningeothelial cell but also mature adipocyte-like cells with peripherally displaced nuclei (a) ×400, (b) ×100.



**Figure 5:** Magnetic resonance imaging T2 image sagittal view (a) after the first surgery showing anterior cervical tumor and (b) after corpectomy and removal of the anterior part of the tumor without any rest.

## DISCUSSION

### Frequency of extradural spinal meningiomas (ESMs)

Meningiomas make up 25% of primary spinal tumors.<sup>[4]</sup> They are usually round and intradural in location. ESMs are rare, occurring in just 2.7%–10% of these lesions; notably, only 3.5% are exclusively “en-plaque.”<sup>[3,8]</sup> ESM has a predilection for the thoracic spine (70%), followed by the cervical (24%) and lumbosacral regions (5%).<sup>[3]</sup> Pathologically, they are aggressive and are often surgically challenging to remove (e.g., they encircle the thecal sac), resulting in a high recurrence rate.<sup>[9]</sup> Their etiology is variously attributed to metaplasia of peripheral nerve cells or migration of arachnoid cells that invaded the dura during embryogenesis.<sup>[3,6-8,10]</sup>

### MR findings consistent with ESM

The diagnosis of ESM is established utilizing MR scans, for example, isointense on T1-weighted, isointense/slightly hyperintense on T2-weighted images with “en-plaque” encirclement of the thecal sac, with or without calcification (1%–4.6%), but with homogeneous contrast enhancement.<sup>[7]</sup>

### Histopathology of ESM

Histopathologically, most meningiomas are meningothelial, followed by the fibroblastic types. Lipomatous meningiomas are a rare variant of the latter subtype and are characterized by lipid accumulation inside the tumoral cells (i.e., metaplastic change called lipomatous or lipoblastic).<sup>[2,10]</sup>

### Recommended surgical management of ESM

Gross total excision is the procedure of choice for managing ESM. In addition, there are various adjuvant therapies that may be utilized (e.g., so as to treat atypical or malignant meningiomas).<sup>[7]</sup> Since these are thick, fibrous, and occasionally calcified lesions that often encircle/scathe the dura, complete removal frequently requires a combined anterior/posterior approach.

### Recurrence rate of ESM

Over a 5-year period, lipomatous ESMs typically show rapid clinical progression and are 4 times more likely to recur versus typical intradural meningiomas; recurrences are much more frequent following initial subtotal resection.<sup>[9,10]</sup> In this case, following a gross total resection, the patient has remained disease free over 3 postoperative years.

## CONCLUSIONS

En-plaque lipomatous ESMs are rare extradural lesions that often demonstrate rapid clinical and radiographic progression. As they are thick, fibrous, tenacious, often calcified, and may encircle the

dura, gross total surgical excision may warrant combined anterior/posterior approaches.

### Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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### Conflicts of interest

There are no conflicts of interest.

## REFERENCES

- Alafaci C, Grasso G, Granata F, Salpietro FM, Tomasello F. Ossified spinal meningiomas: Clinical and surgical features. *Clin Neurol Neurosurg* 2016;142:93-7.
- Colnat-Coulbois S, Kremer S, Weinbreck N, Pinelli C, Auque J. Lipomatous meningioma: Report of 2 cases and review of the literature. *Surg Neurol* 2008;69:398-402.
- D'Amico A, Napoli M, Cirillo M, D'Arco F, D'Anna G, Caranci F, *et al.* Imaging of cervical extradural en-plaque meningioma. A case report. *Neuroradiol J* 2012;25:598-603.
- Frank BL, Harrop JS, Hanna A, Ratliff J. Cervical extradural meningioma: Case report and literature review. *J Spinal Cord Med* 2008;31:302-5.
- Levy WJ Jr., Bay J, Dohn D. Spinal cord meningioma. *J Neurosurg* 1982;57:804-12.
- Pant I, Gautam VKS, Kumari R, Chaturvedi S. Spinal tumour: Primary cervical extradural meningioma at an unusual location. *J Spine Surg* 2017;3:509-13.
- Sivaraju L, Thakar S, Ghosal N, Hegde AS. Cervical en-plaque extradural meningioma involving brachial plexus. *World Neurosurg* 2017;108:994.e7-000.
- Tuli J, Drzymalski DM, Lidov H, Tuli S. Extradural en-plaque spinal meningioma with intraneural invasion. *World Neurosurg* 2012;77:202.e5-13.
- Yamada S, Kawai S, Yonezawa T, Masui K, Nishi N, Fujiwara K, *et al.* Cervical extradural en-plaque meningioma. *Neurol Med Chir (Tokyo)* 2007;47:36-9.
- Yüksel MO, Gürbüz MS, Tanrıverdi O, Özmen SA. Lipomatous meningioma: A rare subtype of benign metaplastic meningiomas. *J Neurosci Rural Pract* 2017;8:140-2.

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