

Intradural Extramedullary Spinal *En-Plaque* Meningioma With Calcification: A Case Report And Literature Review

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Abstract

Introduction: Intradural extramedullary (IDEM) tumors are benign neoplasms arising in the spinal canal about two-thirds of primary spinal tumors and 15% of tumors affecting the Central Nervous System. Spinal en-plaque meningioma is a type that grows in a sheet-like or collar-like, and incidence in the literature only ranging from 0.1% to 3.1%. Pain is the most clinical symptom, weakness and sensory changes also occur frequently. Magnetic resonance imaging (MRI) is the standard modality for the radiologic diagnosis of meningioma.

Case Report: A patient, 35 years old man with a diagnosis of intradural extramedullary spinal meningioma (IDEM) en-plaque with calcification, confirmed by the symptoms, workups such as spinal MRI, and intra-operative findings. The patient was successfully treated surgically with laminectomy and total tumor resection with a posterior approach.

Discussion: Spinal en-plaque meningioma is a type that grows in a sheet-like or collar-like manner around the spinal cord can involve dura extensively with significant neurological deficits. Patient was with lower limb weakness, and had a history of back pain radiating to the right limb for the last 1 year. Spinal meningiomas are primarily found in the Intradural Extramedullary, and the tumor diagnosis is typically fairly straight forward based on radiologic findings. Meningiomas are most commonly found in the thoracic region of the spine. In this case from MRI Imaging was revealed a mass in thoracic region of the spine pressing the spinal cord anteriorly. The management of spinal en-plaque meningioma is tumor resection surgery. A retrospective study suggested a significant improvement in neurological deficit post-tumor resection on patients with spinal IDEM tumor.

Conclusion: Spinal meningioma is a reasonably frequently found case of a spinal tumor but spinal en-plaque meningiomas are rarely found. MRI scan is the radiological gold standar diagnose spinal en-plaque meningiomas. Patient was successfully treated by total tumor resection using the laminectomy method and tumor resection with a posterior approach without any postoperative complications observed.

Keywords: Spinal Tumor, Intradural Exteramedullary tumor, Spinal en-plaque meningioma

Introduction

Intradural extramedullary (IDEM) tumors are generally benign neoplasms arising in the spinal canal, accounting for about two-thirds of primary spinal tumors and 15% of tumors affecting the Central Nervous System.[1] Older population-based surveys have reported the incidence of intradural spinal tumors to be 1.1 per 100,000 person-years, whereas more recent epidemiologic studies have reported an incidence of 0.74 per 100,000 person-years, with an incidence of 0.77 per 100,000 in females and 0.70 per 100,000 in males. The most common intradural spinal tumors are meningiomas, followed in varying order by intramedullary ependymoma or NST.[2] Meningiomas arise from meningotheial cells of the arachnoid layer (not dura). Meningiomas are most commonly

found in the thoracic region of the spine.[3] Occasionally meningiomas form a diffuse sheet of tumor (meningioma *en plaque*).[4]

Spinal *en-plaque* meningioma is a type that grows in a sheet-like or collar-like manner around the spinal cord and infiltrates the pia mater. A case of spinal *en-plaque* meningioma is scarce, with a reported incidence in the literature only ranging from 0.1% to 3.1%.[5],[6]

The Clinical presentation of spinal meningiomas, depends upon their location. Pain is the most and approximately 70–90% of patients will have pain initially, with the pain being axial and/or radicular. Weakness and sensory changes also occur frequently, and more than 60% of patients who undergo surgical decompression or resection will have some degree of weakness. About 40% of patients have reflex changes, which can be hyporeflexia caused by root compression, or hyperreflexia from cord compression and myelopathy. Bowel and bladder dysfunction have been noted in 30–80% of patients with intradural tumors.[3]

Magnetic resonance imaging (MRI) is the standard modality for the radiologic diagnosis of meningioma and location is typically intradural extramedullary. On T1-weighted images (T1WI), the signal of the mass is isointense to the spinal cord on T2-weighted images (T2WI), most parts of the mass show isointense to the spinal cord. If a calcified area is included, the signal may change to low.[3],[7]

Management of spinal IDDM tumors is tumor resection surgery but biopsy is the only method to definitively confirm diagnosis through tumor resection surgery.[8],[9],[10] The pathognomonic histologic feature of a meningioma is spherical formations of meningothelial cells, called whorls, which eventually mineralize into psammoma bodies. A range of histologic patterns and findings can be seen that often mimic other soft tissue tumors, as indicated by the numerous variants just among benign grade I tumors: meningothelial, fibrous, transitional, psammomatous, angiomatous, microcystic, secretory, lymphoplasmacyte-rich and metaplastic.

In this study, we reported a case of a 35 years old man with intradural extramedullary spinal *en-plaque* meningioma with calcification that was successfully treated with total tumor resection using the laminectomy method and tumor removal with a posterior approach.

Case Report

A 35 years old man presented with complaints of lower limb weakness for 3 months, which was worsening in the last 2 weeks with the right motoric strength of 5/4 and left 5/4, accompanied by defecation and bladder disorders. The patient had a history of back pain radiating to the right limb for the last 1 year. Spinal MRI workup revealed a mass as high as TH6 pressing the spinal cord anteriorly (Figure 1).

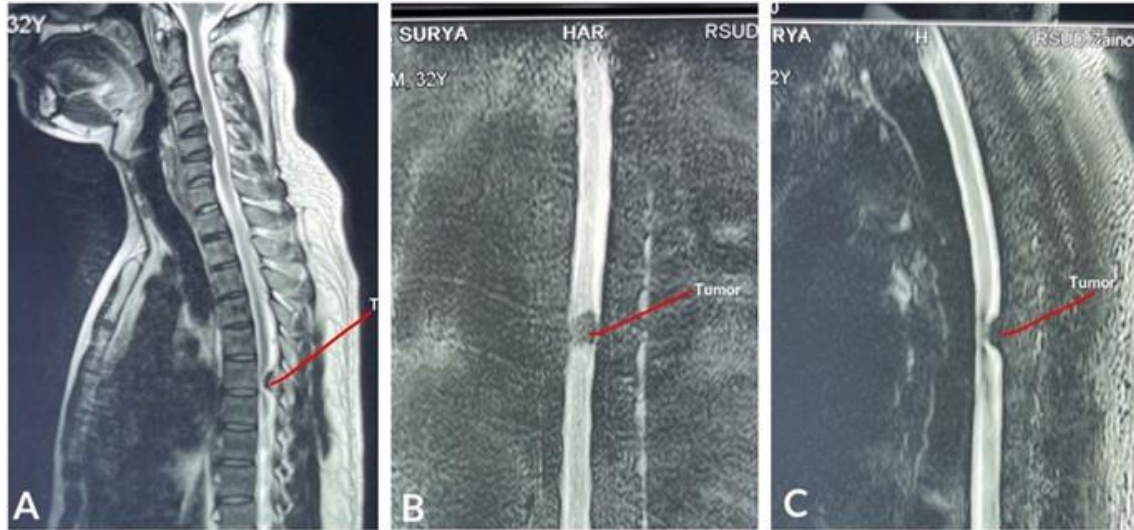
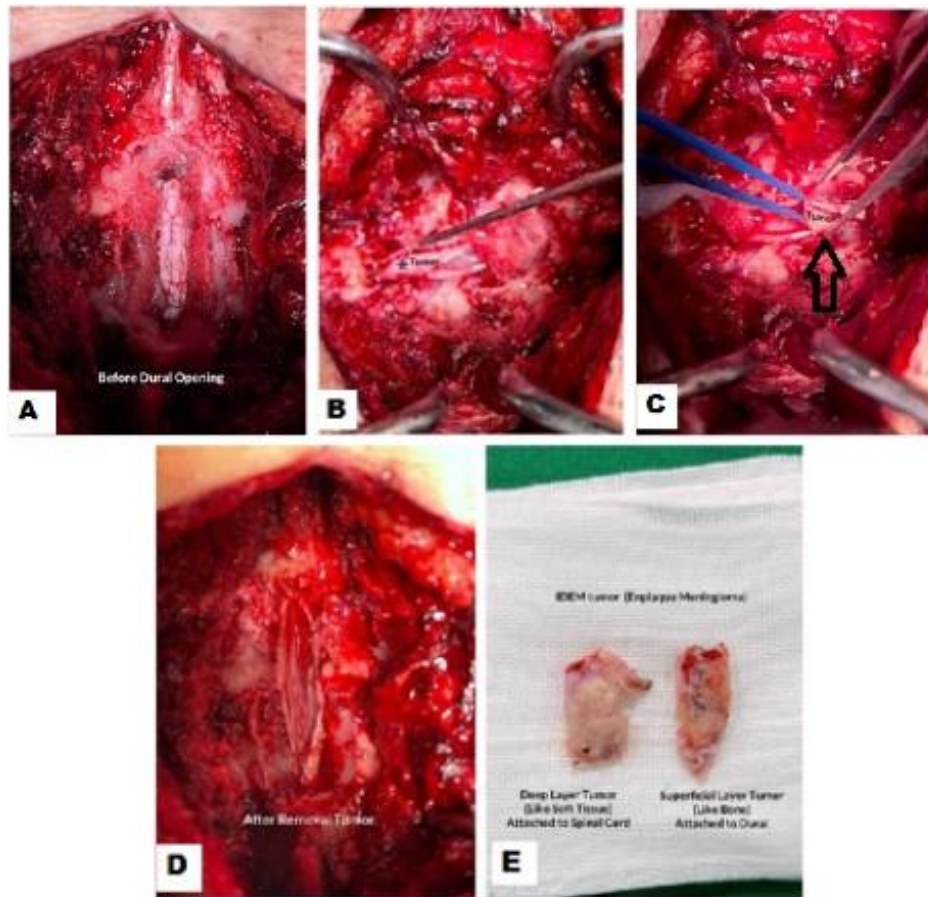


Figure 1. Sagittal plane of the spinal MRI (A,C) and coronal plane (B) a hypointense mass as high as TH6 pressing the spinal cord anteriorly (red lines) was seen.

The patient was managed by laminectomy and tumor resection with a posterior approach. During general anesthesia, the patient was placed in a prone position, followed by disinfection of the back using povidone-iodine 10%, and then covered with a sterile drape. A midline incision as high as TH6 – TH8 was performed, followed by paraspinal muscle dissection. Afterward, a laminectomy of TH7 and half of TH6 and TH8 was performed (Figure 2-D). The dura mater was opened in the midline, and a solid calcified bone-like mass was seen, adhering to the dura mater. The mass was an ellipse with a size of 20 x 10 x 2 mm (Figure 2-E). A resection of the mass was performed. The calcified mass was able to be removed from the dura mater. Under the calcified mass, there was a soft mass adhering to the spinal cord with a size of 10 x 12 x 3 mm (Figure 2-F); the mass was able to be resected with total resection (Figure 2-G, 2-H). The involved dura mater was not removed but was only coagulated. Afterward, the dura mater was sutured and patched with muscle. There was no leakage of CSF postoperative. There was an improvement in motoric strength, normal sphincter function, and less pain on the postoperative follow-up.



Histopathological finding revealed psammomatous meningioma of the spinal (WHO grade 1) with ICD-0 code 9533/0

Figure 2. Surgical view after laminectomy (A), after durotomy (B) a solid bone-like mass adhering to the dura mater (black star) was seen, after resection of the solid mass (C), a soft mass adhering to the spinal cord (black arrow) was seen. Surgical view post total tumor resection (D), and the tumor (E).

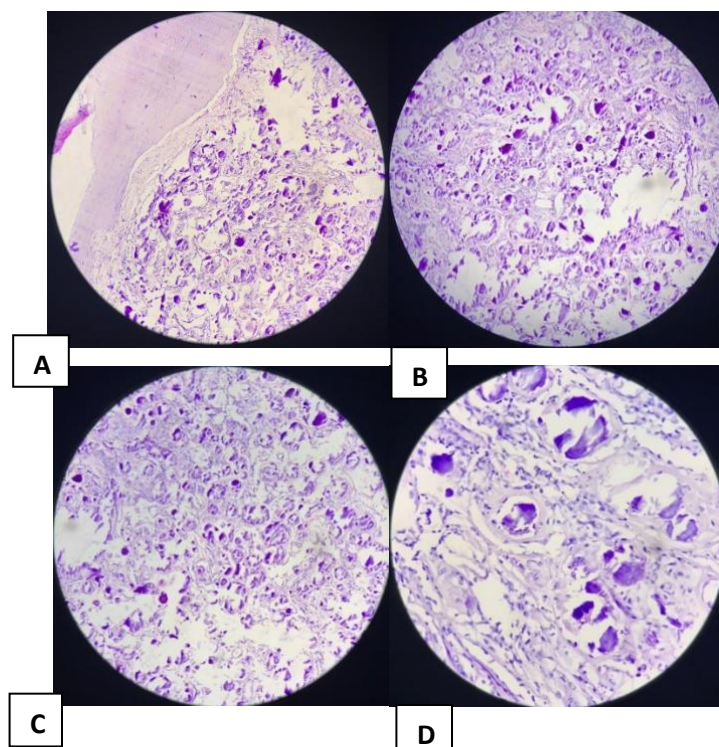


Figure 3. Histopathology Finding (A) Bone Trabeculae and psammoma bodies, (B) (C) (D) Lost meningotheelial cells are replaced by prominent psammoma bodies by concentric calcification.

Discussion

Spinal *en-plaque* meningioma is a type that grows in a sheet-like or collar-like manner around the spinal cord and infiltrates the pia mater.[5] Spinal meningioma is believed to grow from the arachnoidal cell that differentiate from neural crest cells.[6],[8] Although rare, spinal *en-plaque* meningioma can involve dura extensively with significant neurological deficits.[9] In this case patient with lower limb weakness, and had a history of back pain radiating to the right limb for the last 1 year. Pain is the most common initial symptom of spinal meningioma.[3]

Spinal meningiomas are primarily found in the Intradural Extramedullary, and the tumor diagnosis is typically fairly straight forward based on radiologic findings. [6],[22] Meningiomas are most commonly found in the thoracic region of the spine.[3],[21] However, the literature that discusses spinal *en-plaque* meningiomas located in the extramedullary intradural is still scarce. Most of the literature discusses *en-plaque* meningioma located in the extradural.[6],[8] In this case from MRI Imaging was revealed a mass in thoracic region of the spine pressing the spinal cord anteriorly.

Diagnosis of spinal IDEM tumor can be made by detailed history taking, physical examination, and MRI scan on the proximal area since the symptoms of spinal IDEM tumor might mimic that of herniated nucleus pulposus and spinal stenosis.[9] Intradural extramedullary spinal tumors are the most common type of spinal tumors that may result in serious morbidity related to progressive neurologic deficits and meningiomas and schwannomas are the two most common histologic subtypes, which comprise 55–90% of all intradural extramedullary spinal tumors. Takashima et al. reported that the ratio between T2 signal intensity of tumor and that of subcutaneous fat was significantly higher in spinal schwannomas compared to that of meningiomas. Liu et al reported the largest series to date and further categorized T2 signal of tumors with addition of fluid signal intensity. Iwata et al. and Zhai et al. analyzed the presence of cystic degeneration defined as signal intensity equal to that of the cerebrospinal fluid exhibiting low intensity on T1-weighted image and high intensity on T2-weighted image, which turned out to be a useful parameter for predicting schwannoma. The term “cystic change” using for differentiation between spinal schwannoma and meningioma in a large patient population.[7]

In accordance, the study by Caroli et al. stated that MRI is the radiological tool of choice for diagnosing spinal meningiomas *en-plaque*. CT scan is useful in detecting calcification within meningiomas.[5] *En-plaque* meningioma can also mimic IDEM

tuberculoma, which was reported by Kolakshyapati M, et al. The imaging features of IDEM tuberculoma are nonspecific and may mimic *en-plaque* meningioma. The study by Kolakshyapati M, et al. found that IDEM tuberculoma in their case derived its blood supply from the pial vessels as seen in *en-plaque* meningioma. In such cases, definitive diagnosis is nearly impossible without histopathological examination.[20],[21] In this case, the spinal IDEM meningiomas *en-plaque* was diagnosed based on history taking, preoperative MRI and intra-operative findings.

Management of spinal IDEM tumors is tumor resection surgery in generally.[10] A retrospective study suggested a significant improvement in neurological deficit post-tumor resection on patients with spinal IDEM tumor, regardless of the percentage of tumors occupying the intradural space, the degree of preoperative symptoms, and the duration of symptoms.[9] Study by Goodarzi et al. suggested that there was no significant difference in the length of operation, estimated blood loss, or in the length of hospital stays between the spinal IDEM tumor's approaches of laminectomy or hemilaminectomy. The decision for surgical technique is primarily based on the size of the volume, histopathology, and the tumor's location. [11]

The same management approach also applies to the case of spinal *en-plaque* meningiomas. A study by Caroli et al. on 5 spinal IDEM *en-plaque* meningioma patients and 2 intra-extra dura *en-plaque* meningioma patients suggested that laminectomy management and total tumor resection were successfully performed on 3 patients, whereas the other 4 patients were only managed by sub-total resection. Postoperative follow-up also showed satisfactory results; 3 patients with total resection had a significant neurological symptoms improvement in 3 months within postoperative. The patients with subtotal resection had worsened neurological symptoms both temporarily (3 patients) and permanently (1 patient). Another complication found in patients with subtotal resection was regrowing of the tumor.[5]

Regardless of the location difference (extradural) or primary entity (IDEM tuberculoma mimicking *en-plaque* meningiomas from clinical features and intra-operative findings), several cases reported similar results, including improvement of clinical symptoms with tumor resection and satisfactory postoperative condition. [6],[8], [11],[12].

Several case studies of meningioma with calcification had shown clinical improvements after tumor removal with [13],[14] or without *en bloc* resection method.[4],[15] In this case, the tumor was successfully resected with total tumor resection, both solid (calcified) tumor and soft tumor using laminectomy method and

tumor resection with a posterior approach, with a satisfactory final result in improved clinical symptoms and quality of life.

Although the previous studies had shown satisfactory final postoperative results for spinal meningiomas, several postoperative complications might occur in the short or long term. Short-term complications that might occur include leakage of cerebrospinal fluid (CSF), *meningitis*, and death.[16] Long-term complications include tumor regrowth and the need for repeat surgery, with significant factors such as aged <50 years old and Simpson grade \geq II resection in the surgery.[17],[18],[19] In this case, there were no short-term postoperative complications found. Follow-up would still be performed on the patients to observe long-term complications.

Conclusion

Spinal meningioma is a reasonably frequently found case of a spinal tumor but spinal *en-plaque* meningiomas are rarely found. MRI scan is the radiological gold standard to diagnose spinal *en-plaque* meningiomas. The management of spinal IDEM *en-plaque* meningiomas is tumor resection. In this study, we reported a case of a man with spinal IDEM *en-plaque* meningioma with calcification. The patient was successfully treated by total tumor resection using the laminectomy method and tumor resection with a posterior approach, with a satisfactory postoperative result without any postoperative complications observed. Histopathological was found *psammomatous meningioma of the spinal* (WHO grade 1). Follow-up would still be performed to observe any long-term complications that might occur.

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