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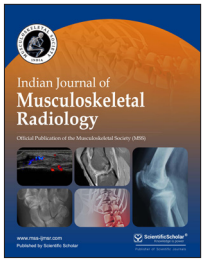

Case Report

An Unusual Long Segment Spinal Epidural Cavernous Hemangioma: A Case Report

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ABSTRACT

Spinal epidural hemangioma, mostly cavernous, is a rare lesion with many radiological mimics that has diagnostic difficulty. They can extend from one to multiple vertebral levels and may or may not be associated with vertebral hemangiomas. We are reporting a case of young adult presenting with features of compressive myelopathy. Plain and contrast-enhanced magnetic resonance imaging showed a large spinal epidural lesion extending from C7 to D10 vertebral levels with extension into adjacent neural foramina and paravertebral spaces. There were also signal changes in bodies and posterior elements of dorsal vertebrae. A provisional diagnosis of lymphoma was made. The patient was operated for decompression and histopathological diagnosis of cavernous hemangioma was made. As in our case, a review of literature shows that epidural cavernous hemangioma of spine may extend to multiple vertebral levels and difficult to diagnose on pre-operative imaging. However, such a long segment epidural cavernous hemangioma has not been reported in literature. Furthermore, we should be aware of these rare lesions to include it in our differential diagnosis the spinal epidural lesions for early diagnosis and management.

Keywords: Cavernous hemangioma, Epidural, Spine, Magnetic resonance imaging, Compressive myelopathy

INTRODUCTION

Craniospinal cavernous hemangioma is a relatively uncommon vascular malformation. They can occur in any part of nervous system but most commonly found in supratentorial compartment.[1] In spine, they predominantly affect the vertebral bodies with or without extension into epidural space, but by definition, purely epidural cavernous hemangiomas of the spinal canal are those that originate solely in the extradural space and that do not invade osseous structures.[1,2]

Pure epidural cavernous hemangiomas of the spinal canal are extremely rare.[1-3] They can extend from single to multiple vertebral levels. There are about few more than 130 case reports on spinal epidural hemangiomas. Lesions with extent up to seven vertebral segments are described.[3] We are reporting case of long-segment spinal epidural hemangioma extending from C7 to D10 vertebral levels with extension into neural foramina and paravertebral spaces presenting with compressive myelopathy.

CASE REPORT

A 32-year-old man presented with burning and tingling sensation in both lower limb followed by numbness for 1 year and asymmetrical weakness in lower limbs for the past 7 months. His

bladder and bowel control were lost for 3 months and he was bedridden from 1 month. There was no history of trauma, tuberculosis, and fever. On examination, tone and reflexes were increased, and power was 1/5 in both lower limbs. Upper limbs were not affected. The patient also had sensory loss from D6 vertebral level. There was no local spinal tenderness.

Contrast-enhanced magnetic resonance imaging (MRI) scan showed long segment altered signal intensity extramedullary lesion in anterior and posterior epidural space extending from C7 to D10 vertebral levels. The lesion was hypointense on T1-weighted images (T1WI), hyperintense on T2-weighted images (T2WI), and short tau inversion recovery (STIR) images and showed strong diffuse post-contrast enhancement [Figures 1-3]. Cranial and caudal margin of the lesion showed a dural tail. The lesion was also extending into bilateral neural foramina and adjacent paravertebral space at multiple levels [Figure 2a and b]. There was displacement and compression of thecal sac towards right [Figure 2b] with mild compression of spinal cord and subtle hyperintense signal at D6–D8 level on fluid sensitive sequences suggesting compressive myelopathy. There were also diffuse heterogeneous predominantly hyperintense signals on T1- and T2-weighted MRI in body and posterior elements of D1–D10 vertebrae with patchy hypointense areas on STIR images and showing heterogeneous enhancement on T1 post-contrast images suggesting osseous extension of the disease [Figures 1a and b, 3a]. No evidence of hemorrhage was noted within lesion. The plain computed tomography scan did not reveal any bony destruction or expansion. No

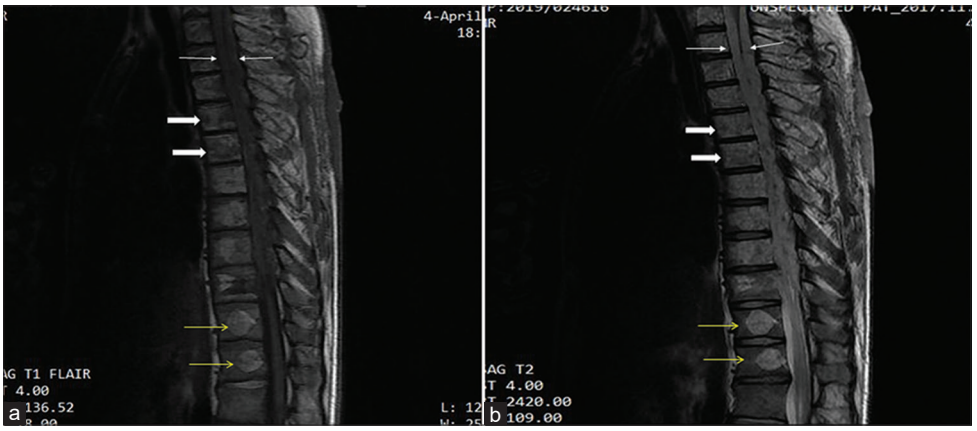

Figure 1: (a) Sagittal T1-weighted magnetic resonance image showing long segment iso- to hypointense lesion in anterior and posterior epidural space (thin white arrows). Diffuse heterogeneous hyperintense signals seen in multiple dorsal vertebrae (thick white arrows). Typical hemangiomas are noted at D11 and D12 vertebral levels (thin yellow arrows). (b) Sagittal T2-weighted magnetic resonance image showing long segment lesion in anterior and posterior epidural space (thin white arrows). Diffuse hyperintense signal seen in multiple dorsal vertebrae (thick white arrows). Typical hemangiomas are noted at D11 and D12 vertebral levels (thin yellow arrows).

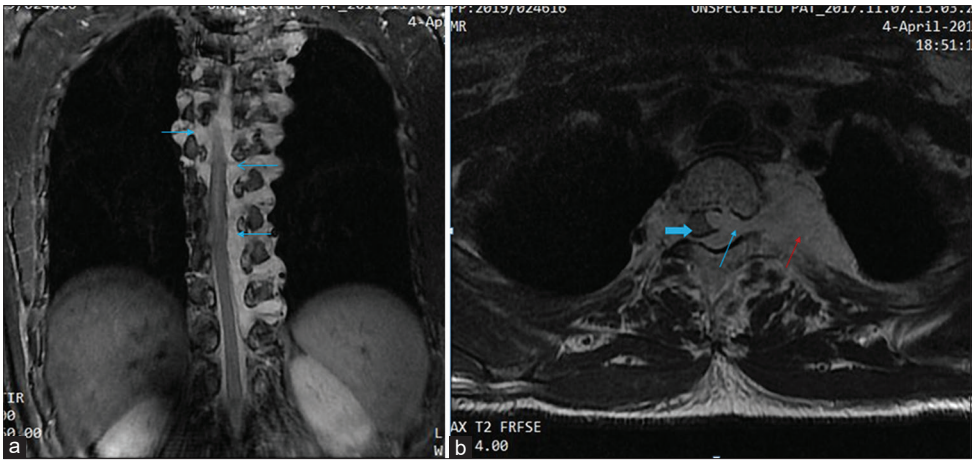

Figure 2: (a) Coronal short tau inversion recovery magnetic resonance image showing hyperintense lesion (blue arrows) in epidural space extending into adjacent neural foramina and paravertebral space (more prominent on left side). (b) Axial T2-weighted magnetic resonance image shows anterior and posterior epidural space lesion with extension into B/L neural foramina (thin blue arrow) and adjacent paravertebral space (red arrow). There is partial encasement and compression of the spinal cord with displacement towards right (thick blue arrow).

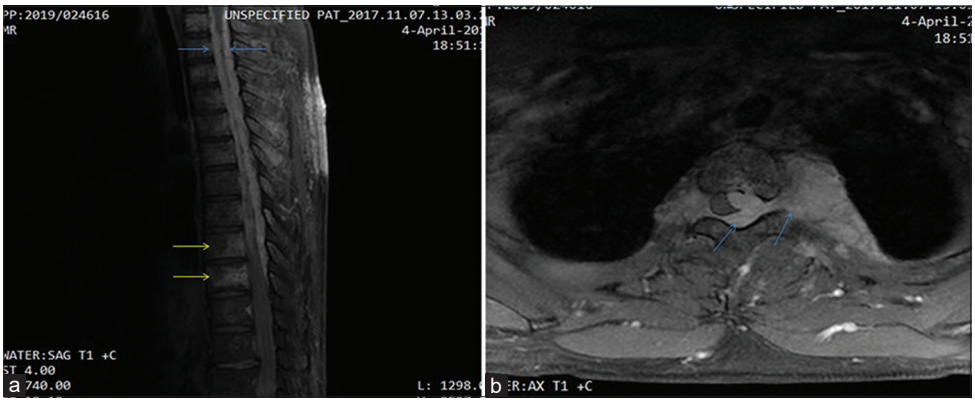

Figure 3: (a) Sagittal T1-weighted fat-suppressed post-contrast magnetic resonance image showing diffuse post-contrast enhancement of epidural lesion (blue arrows). Mild patchy enhancement of dorsal vertebrae also noted (yellow arrows). (b) Axial T1-weighted fat-suppressed post-contrast images showing diffuse post-contrast enhancement of epidural lesion (blue arrows).

calcification noted in the lesion neither was any enlargement of neural foramina. There were two well-defined altered signal lesions in bodies of D11 and D12 vertebrae with signal characteristics of hemangiomas [Figure 1a and b]. A probable radiological diagnosis of lymphoma was made due to atypical vertebral changes and large epidural lesion.

The patient underwent C7–D10 laminectomy with decompression of extradural lesion along with biopsy. Peroperatively, the lesion was red-brown, soft, highly vascular, and adherent to thecal sac with extension into bilateral neural foramina so complete excision could not be done. Bones were grossly normal, with no connection with the lesion. Histopathology revealed cavernous hemangioma [Figure 4].

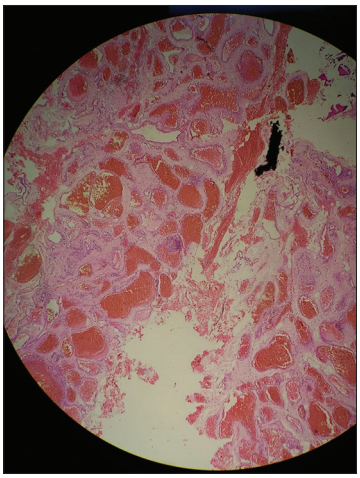

Figure 4: Microscopic histopathological image shows large cystically dilated thin walled congested vascular channels lined by single layer of endothelium. Perivascular tissue shows fibrosis and focal myxoid changes. Findings are characteristic of cavernous hemangioma.

DISCUSSION

Cavernous hemangiomas are ubiquitous in nature. They are benign proliferative vascular lesions, collection of small capillaries covered with a single layer of endothelium, characterized by lobules, separated by fibrous connective tissue septa, and composed of irregular and dilated vascular channels. Spinal epidural hemangiomas are extremely rare. In most cases, they are usually located in the vertebral bodies and comprise 5–12% of all spinal hemangiomas.[4] Pure spinal epidural cavernous hemangiomas are rare lesions. They are most commonly located in the posterior epidural space in the thoracic region and can also be seen in the foraminal and extraforaminal regions in lesser frequency.[3-5]

Hemangiomas are classified into arteriovenous, cavernous, capillary, or venous depending on the predominant type of vascular channel. Most commonly, they are of the cavernous type, which is seen as solid hypervascular mass which displays homogenous hyperintense signal on T2WI and intense post-contrast enhancement.[3,5,6] The peripheral rim of T1 and T2 hypointensity, resulting from hemosiderin deposition, is usually seen in the intramedullary cavernous hemangioma and not in extraosseous epidural cavernous hemangioma, probably due to absence of blood-brain barrier causing washing out of blood products.

The most common location is thoracic (approximately 60%) followed by cervical (30%) and lumbar (10%).[5,7] They are usually located at dorsal or dorsolateral epidural spaces of the spinal canal due to the abundance of epidural venous plexus in the region.[8]

Most patients are in the 30–60-year-old age range, with a peak of around 40 years.[4,7] The clinical presentation of spinal epidural hemangioma depends on its location, growth rate, and biological behavior. Considering its slow growth and predilection for thoracic and cervical location, the most common clinical symptom is progressive myelopathy.[4,5,8] Other symptoms include radiculopathy and local pain.

MRI is an investigation of choice for spinal cavernous hemangiomas. These are usually seen as lobulated extradural masses and show iso- to hypointense signals on T1WI, hyperintense on T2WI and show intense post-contrast enhancement similar to our case; however, these findings are also not specific to this disease.[5,9,10] The dural tail sign may be seen as thin dural enhancement near the mass with a broad angle. According to some authors, a T2 hypointense rim around spinal hemangioma may also be present due to fibrous capsule, interface between mass and adjacent posterior longitudinal ligament and dura mater, or due to chemical shift artifact between epidural fat and intralesional fluid content.[3,6] The lesion can also show its extension into the intervertebral foramen, as seen in our case.[5,10] However, bony erosion is not common and the neural foramen widening is less as compared to neurogenic tumor of same size.[8] The tumor can show dumb-bell shape on coronal images, as seen in our case. In the presence of hemorrhage lesion can show altered signals depending on the stage of blood. In very early stage of hemorrhage lesion shows very low signals on T2WI and high signals on T1WI.[3]

As in our case, review of literature shows that the diagnosis is almost always missed on pre-operative imaging. A study by Tekkok *et al*. reported on 14 surgical resections of cavernous hemangioma, none of which was diagnosed preoperatively.[2] The differential diagnosis includes lymphoma, meningioma, nerve sheath tumors, angiolipoma, granulomatous disease, and metastasis.[3,4,8]

Complete surgical removal is currently the treatment of choice for primary epidural hemangioma. Surgical treatment should be performed before worsening of the patient's neurological deficit.[2,8]

CONCLUSION

Although epidural spinal hemangiomas are rare; they can mimic many common spinal tumors clinically and radiologically. They can extend from one to more vertebral levels. Pre-operative diagnosis is very difficult. Therefore, in patient with features of compressive myelopathy if there is an epidural spinal lesion which is lobulated in shape, showing multilevel involvement, displaying hyperintense signals on T2WI with strong diffuse contrast enhancement, we should keep cavernous hemangioma in our differential diagnosis. The early pre-operative diagnosis followed by complete excision of the lesion is required as these lesions can show episodes of massive pre-/intraoperative intralesional bleeding which can cause spinal cord compression and severe neurological deficit.

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Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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