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Letter to the Editor

A rare case of thoracic extradural thrombosed primary cavernous haemangioma in a relatively asymptomatic patient

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Sir,

Pure spinal epidural cavernous (haem) angiomas (PSECA) arising without any connection with the vertebrae are rare and few cases have been reported in the literature. They usually grow to a considerable size and are usually symptomatic in form of myelopathy with or without radiculopathy features.[2] We report an interesting case of thrombosed primary cavernous haemangioma located in the extradural space of thoracic region with only intermittent, mild paresthesia in lower limb.

A 16 year old girl presented with complaints of intermittent electric shock like sensation in left side lower limb for last 2 to 3 years. There was no history of weakness, numbness, tightness in either lower limb, bladder or bowel disturbances. On examination, there was no sensory deficit with motor power being grade 5/5 (MRCS scale) in all four limbs. All the jerks were well preserved and normal. Plantar was bilaterally downgoing. MRI thoracic region (T1 weighted) showed an elongated, spindle shaped isointense lesion in posterior aspect of spinal canal at T7-T9 level [Figure 1a]. The lesion was hyperintense in T2 image [Figure 1b] with minimal patchy enhancement [Figure 1c]. The lesion was located in extradural space compressing the cord from posterior aspect [Figure 2a and b]. Intra-operatively, after T7-T9 laminectomy, a reddish-brown, soft, well marginated, minimally vascular lesion with bluish black coloured cystic spaces found in extradural space [Figure 3]. The lesion was sessile and was not attached to any nerve roots or bony structures with clear line of cleavage between it and the underlying dura (flimsy adhesions). The lesion was removed in single piece. Histopathology showed the lesion to be packed with large numbers of cavernous spaces lined by thin endothelium [Figure 4a]. Most of the spaces were filled with organised blood clots [Figure 4b]. The final diagnosis was thrombosed cavernous haemangioma.

Cavernous hemangiomas account for 3 to 16% of spinal vascular anomalies.[6,7] Most of these lesions are extensions from primary vertebral lesions. Extradural primary cavernous haemangioma of spine not arising from bony components are rare and constitute only 4% of all spinal epidural lesions.[4,6] and 12% of spinal cavernous anomalies.[2] Only 80 such cases have been reported in the literature.[8] They are usually located in extradural location (51%)[1] at thoracic region as in our case. They are slow growing and over a period of time attain a considerable size due to repeated micro-haemorrhages thrombosis with recanalisation and organisation of tissue.[3] As they commonly occur in thoracic region where the spinal canal has minimum capacity to accommodate any further in increase in volume due to mass growth, they are usually symptomatic in form of myelopathy

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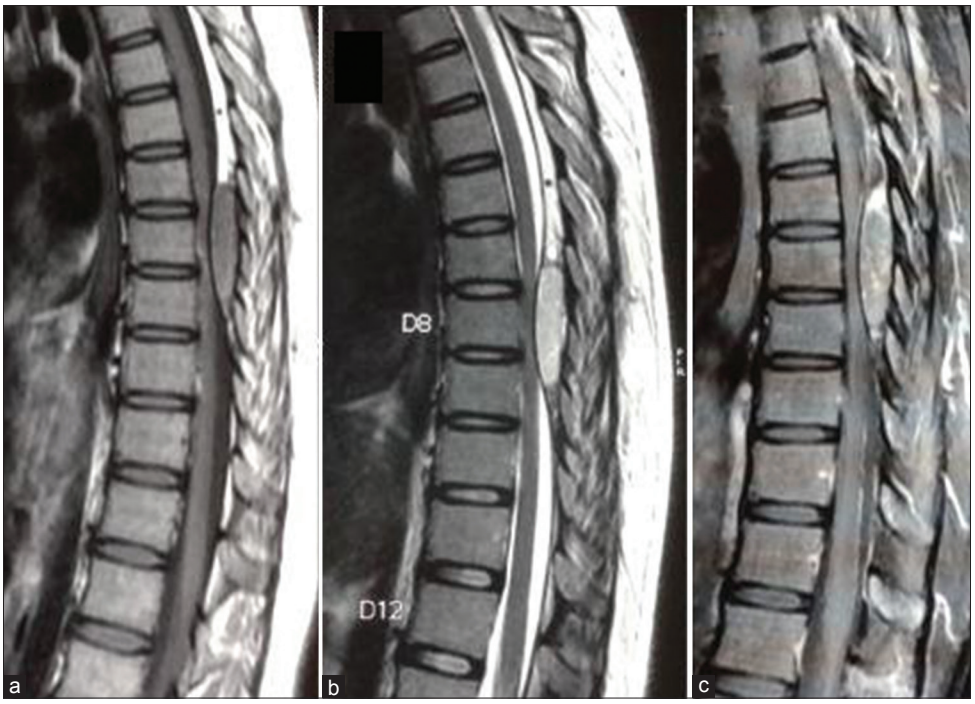

Figure 1: Sagittal image showing the spindle shaped elongated lesion in T1 weighted (a), T2 weighted (b) sequences with minimal patchy enhancement on contrast (c)

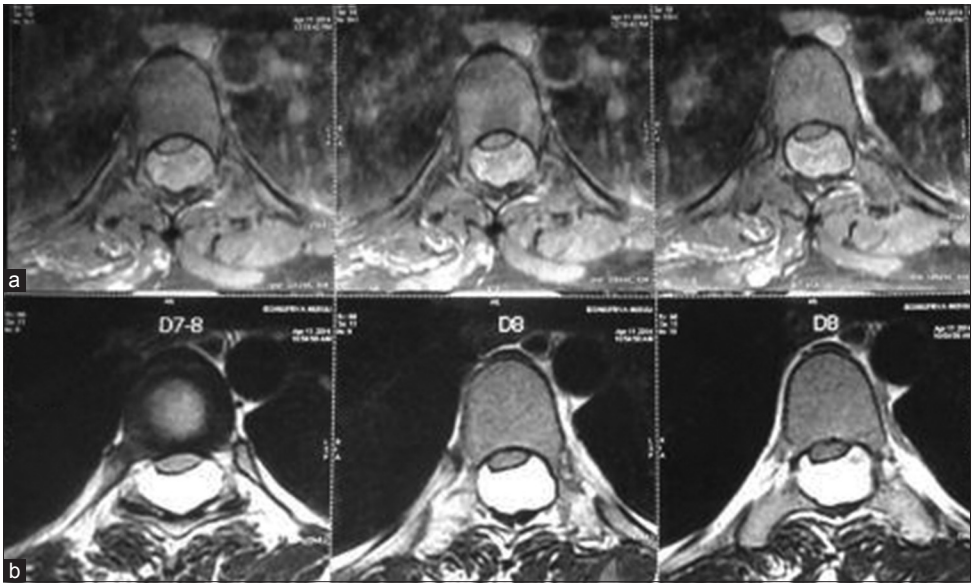

Figure2: Extradural location of the lesion with no bony or nerve root connection (a) after contrast administration, (b) T2 weighted image

features like weakness with spasticity, exaggerated jerks, sensory deficits, bladder, bowel disturbances due to cord compression. But minimal symptoms in our case was quite unusual and can be explained on the basis of widespread thrombosis which would probably had caused the lesion to soften up to exert a less compressive effect on the cord. Cavernous haemangiomas show intense contrast enhancement and resemble meningioma, schwannoma or chordoma. But this was lacking in our case and can be explained by the extensive thrombosis. Pre-operatively such radiological features make such a thrombosed lesion to mimic a more common pathology like schwannoma. Treatment of these cavernous malformations is complete excision. However, highly vascular lesions preventing complete resection can be treated with radiotherapy.[5]

Our case was exclusive in that in spite of a sizeable lesion at thoracic location, the patient was having very subtle symptoms with radiological features suggesting

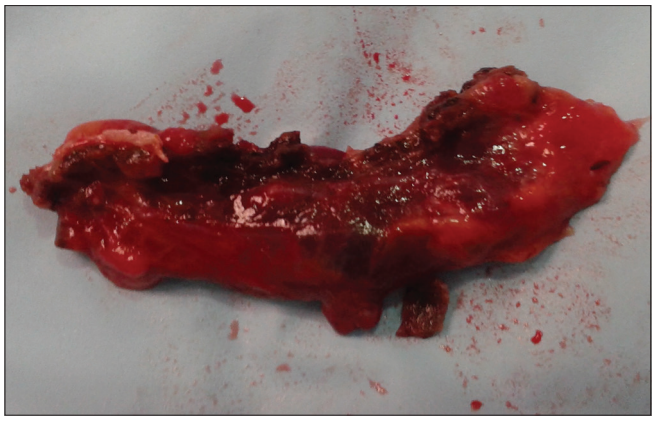

Figure 3: Reddish-brown lesion with bluish black coloured cysts within it

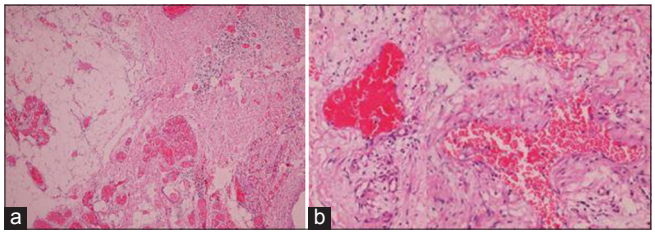

Figure 4: (a) Low magnification showing large number of cavernous spaces lined with endothelium and filled with blood along with adipocytes of epidural fat. (b) High magnification showing the typical flattened endothelial cells lining the lumina of cavernous spaces with well organised clots within it

a more common nerve sheath tumour. The cavernous haemangiomas of spine, although look benign, have a devastating outcome as these usually present with myelopathy features with bladder, bowel dysfunction and most of these do not completely recover following their excision. Fortunately our case had no preoperative neurological deficits and this could be attributed to the thrombosed nature of the lesion. Such thrombosed haemangiomas should be considered in the differential diagnosis of asymptomatic well-defined extradural lesion in a young patient with long standing symptoms.

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