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Case Report

Posttraumatic thoracic epidural capillary hemangioma – A rare case report

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ABSTRACT

Background: Capillary hemangiomas are benign vascular lesions commonly seen in subcutaneous tissues. The most common site of origin is from the vertebral body, and only a few cases of isolated lesions in thoracic epidural space, especially after trauma, have been reported in the literature.

Case Description: We report a case of 63-year-old male with progressive bilateral lower limb weakness and exaggerated lower limb deep tendon reflexes without bowel and bladder involvement. His history revealed T7 fracture with paraparesis which was treated surgically, and implants were removed a year later. MRI showed an epidural lesion from T6-T8 extending into the right T7-8 foramen which showed hypointensity on T1, hyperintensity on T2, and homogenous enhancement in contrast images with severe cord compression. Laminectomy was done and the lesion was removed en bloc. Histopathological examination revealed it to be capillary hemangioma. The neurology came back to normal after 3 months.

Conclusion: Although capillary hemangiomas are rare lesions, it has to be considered in the differential diagnosis of epidural space-occupying lesions which require early surgical removal to prevent a progressive and permanent neurological deficit.

Keywords: Epidural capillary hemangioma, Posttraumatic, Thoracic hemangioma

INTRODUCTION

Capillary hemangiomas are benign tumors of endothelial cell origin arising most commonly from the soft tissues of head and neck.[13] It occurs due to improper differentiation of primitive mesoderm at the embryonic mesodermal plate during the initial stages of angioblastic differentiation.[17] It consists of an aggregation of abnormal blood vessels that used to be better known by the misnomer pyogenic granuloma. Capillary hemangiomas are usually seen arising from the vertebral body and extend into the epidural space. Most of the reported epidural hemangiomas are of cavernous type. Isolated capillary hemangiomas in the epidural space, especially in the thoracic level, are extremely rare. [3] Although posttraumatic capillary hemangiomas are common in mucous membranes and subcutaneous tissues, its occurrence in the epidural space following trauma is extremely rare with only two cases reported in the literature. Proper evaluation is needed in these patients to differentiate it from other epidural

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space-occupying lesions. We report a rare case of thoracic epidural capillary hemangioma following trauma with neurological involvement.

CASE DESCRIPTION

A 63-year-old South Asian male presented with pain over the upper and mid-back with progressive weakness of bilateral lower limbs for 2 months. The pain was insidious in onset and gradually progressive which aggravated on walking and relieved on rest. He had a history of T7 fracture with paraparesis 9 years back, for which he underwent posterior stabilization and instrumentation followed by a complete recovery of his neurological deficit and implant removal a year later. Neurological examination revealed spastic paraparesis with Grade 2 power (MRC grading) and exaggerated knee and ankle reflexes in both lower limbs. His bowel and bladder functions were normal. MRI showed an extradural lesion measuring $4.2 \times 1.6 \times 1.9$ cm (CC × AP × Trans), which was mildly hypointense on T1- and hyperintense on T2-weighted images with strong relatively homogenous enhancement following contrast administration within the posterior aspect of the spinal canal at the T6-T8 vertebral level, causing severe compression and anterior displacement of the cord [Figures 1-4]. It was also protruding into the proximal aspect of the right T7/T8 neural foramen. Altered signals with abnormal enhancement were also seen in the adjacent right pedicle, proximal transverse process, and right posterolateral aspect of the T7 vertebra.

In view of neurological deficit, the patient underwent T6-T8 wide laminectomy and facetectomy. The lesion appeared as a well-circumscribed spongy, purple hemorrhagic epidural mass over the dura extending into the T7/T8 neural foramen, which was removed in toto by carefully detaching it from

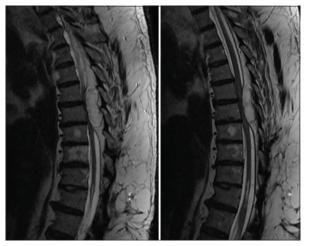


Figure 1: T2 Sagittal images showing hyperintense lesion in thoracic epidural space with compression of spinal cord.

dura. Histopathology examination revealed a highly vascular tumor with innumerable thin-walled capillary vessels of varying sizes, lined by flattened endothelium, and embedded within a fibro-fatty tissue with a total lack of smooth muscular elements suggestive of capillary hemangioma [Figure 5]. Unlike cavernous hemangiomas, the lesion was devoid of abnormally hyalinized vascular channels, thromboses,

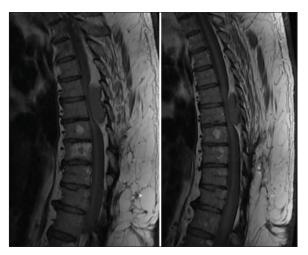


Figure 2: T1 Sagittal images showing hypointense lesion in thoracic epidural space with compression of spinal cord.

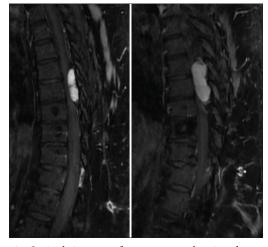


Figure 3: Sagittal images after contrast showing homogenous enhancement of the lesion.

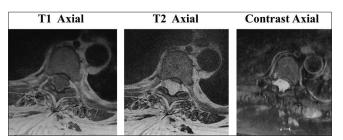


Figure 4: Axial images showing the lesion compressing the cord.

calcifications, or surrounding deposition of hemosiderin pigment. No mitotic activity or cellular atypia was seen.

Postoperative period was uneventful and the neurology improved significantly to Grade 4 within 2 weeks and to Grade 5 at the end of 3 months.

DISCUSSION

Capillary hemangiomas are benign vascular lesions commonly seen in the skin and subcutaneous tissues.^[13,17] Most of the epidural hemangiomas reported in the literature are of a cavernous type. [7,15] Very few cases of spinal epidural capillary hemangiomas have been reported in the literature, [8,13] the most common site being intradural

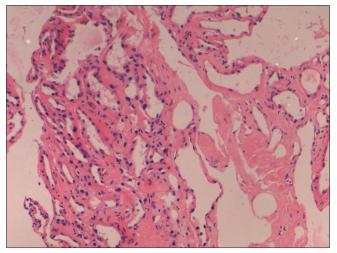


Figure 5: Histopathological features suggestive of capillary hemangioma.

location.[13] extramedullary Intramedullary capillary hemangiomas usually occur due to dysembryogenetic process.^[12] Epidural capillary hemangiomas of the thoracic spinal cord are quite rare, and very few cases have been reported in the literature [Table 1]. They frequently present with extension into the neural foramen.[3,13,24] In our case, the tumor was extending into the right T6-7 foramen. The common clinical presentation is back pain or radicular pain with features of myelopathy due to the mass effect of the tumor unlike cavernous hemangiomas which present with an acute profound neurological deficit due to bleeding.[19] Our patient had progressive weakness of both lower limbs with exaggerated reflexes. They appear iso- to hypointense in T1- and hyperintense in T2-weighted images with strong enhancement after contrast administration similar to the images of our patient. Radiologically, they mimic schwannoma, neurofibroma, meningioma, cavernous hemangiomas, and lymphomas due to their close resemblance in the MRI. [9,11,16-19,22] However, the presence of draining veins in MRI is a differentiating feature from other epidural lesions.[1] Histologically, capillary hemangiomas should be differentiated from cavernous hemangiomas, capillary telangiectasias, and arteriovenous malformations. Cavernous hemangiomas are characterized by dilated hyaline vessels with thrombosis, calcification, and hemosiderin deposition. The presence of neural parenchyma between vessels helps to identify capillary telangiectasia and arteriovenous malformations.

Posttraumatic capillary hemangiomas, also known as pyogenic granulomas, have been reported in skin and mucous membranes.[4,14] However, posttraumatic epidural capillary hemangiomas are very rare, and only two cases have been reported in the literature [Table 1]. One patient

Table 1: Summary of spinal epidural capillary hemangiomas reported in literature.				
Study	Age/sex	Location	Treatment	Recurrence
Badinand et al.[3]	40/F	T2-T4 with extension into left T3-4 foramen	Complete excision	No
Hasan et al.[10]	57/M	T10-12 with extension into right T11-12 and T12-L1 foramen	Complete excision	No
Vassal et al.[24]	59/F	T5-T7 with extension into right T6-7 foramen	Complete excision	No
Tekin et al.[23]	56/F	L3-4 with no foraminal extension	Complete excision	No
Kang et al.[13]	56/M	T3-T4 with extension into right T3-4 foramen	Complete excision	No
Gupta et al.[8]	50/M	T8-T10 with extension into left T8-9 foramen	Complete excision	No
Seferi et al.[20]	58/M	T2-T4 with extension into bilateral foramen	Complete excision	No
Shilton <i>et al</i> . ^[21]	43/F	T7 with no foraminal extension	Complete excision	No
		Past history of trauma 10 years back with compression fractures of T7, T8		
Abe et al. ^[2]	59/M	T11 with no foraminal extension	Complete excision	No
		History of injury to lumbar level	-	
Sudhir <i>et al</i> .	63/M	T6-8 with extension into right T7-8 foramen	Complete excision	No
(Present study)		Past history of trauma 9 years back with T7 burst fracture		
Posttraumatic capillary hemangiomas have been reported only in two cases				

presented with an epidural capillary hemangioma at T7 level with the neurological deficit with a history of compression fractures of T7 and T8.[21] The other patients had an intradural extramedullary capillary hemangioma at T11 after an injury to the lumbar spine.[2] The pathogenesis of the formation of these hemangiomas after trauma may be due to the release of angiogenic growth factors by the inflammatory cells which result in increased vascularity, [5] or it may be due to the chronic repetitive irritation due to microinstability of the posterior elements after trauma. [6]

Although few authors have suggested a conservative approach considering the benign nature of these lesions, [15,19] surgical treatment in the form complete resection of the tumor tissue remains the treatment of choice.[20,24] Since most of them are well-circumscribed lesions without significant adherence to the dura, en bloc resection should be performed whenever possible. Preoperative angiography and selective embolization of the feeding vessels are another option to prevent intraoperative bleeding which tends to occur with piecemeal removal of the tumor.[17,23] Intraforaminal extension of the tumor poses a challenge for complete removal of the tumor.[3,10] Furthermore, routine follow-up of these patients is recommended as rapid progression of the lesion after complete resection within 6 months has been reported.

CONCLUSION

Epidural capillary hemangiomas are rare tumors that have to be differentiated from other vascular lesions of the spine. Although posttraumatic capillary hemangiomas are common in subcutaneous tissues, it is not uncommon for its occurrence in the spine. Early and prompt diagnosis by MRI and histopathological examination is vital. Surgical excision after one careful preoperative workup is the treatment of choice to prevent a progressive and permanent neurological deficit.

Declaration of patient consent

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

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Conflicts of interest

There are no conflicts of interest.

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