

CASE REPORT

Dumbbell-Shaped Epidural Capillary Hemangioma Presenting as a Lung Mass

Case Report and Review of the Literature

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Study Design. A case report and literature review.

Objective. We present the fourth case of a spinal epidural capillary hemangioma with a dumbbell-shaped appearance in the magnetic resonance image reported in the literature and the second presented as a lung mass.

Summary of Background Data. Hemangiomas are congenital vascular malformations that pathologists frequently consider to be hamartomatous malformations. Hemangiomas of the spine are usually lesions of the vertebral bodies, but they can sit in other locations such as the intramedullary or epidural space. Purely epidural hemangiomas are rare and most of them are of cavernous type.

Methods. We present a 67-year-old female with a thoracic dumbbell-shaped capillary hemangioma with both foraminal and intrathoracic extensions, whose presentation was pleural effusion associated with mediastinal mass suggestive of pulmonary neoplasia. Surgical treatment consisted of total removal *en bloc* of the lesion.

Results. Microscopic evaluation showed a fibrofatty tissue with a proliferation of vascular structures that were generally of a small size, with areas of myxoid appearance. To date, there have been 8 epidural capillary hemangiomas of the thoracic and lumbar spine reported in the literature, and only 3 of them were dumbbell-shaped with extraforaminal extension.

Conclusion. It is important to consider the diagnosis of hemangiomas in the differential diagnosis of epidural lesions with dumbbell-shaped appearance in the magnetic resonance image, especially at the thoracic level. It is a benign and potentially

curable disease and the most appropriate surgical treatment is *en bloc* resection of the entire lesion. They are usually presented as a progressive myelopathy, so early treatment may prevent permanent neurological deficits.

Key words: capillary hemangioma, cavernous hemangioma, dumbbell-shaped, epidural mass, review, spinal cord.

Level of Evidence: 5

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Purely epidural hemangiomas of the spine are extremely rare. Most of them are of the cavernous type and, to date, there have been only 8 reported cases of epidural capillary hemangiomas. We present a female with a thoracic dumbbell-shaped capillary hemangioma with both foraminal and intrathoracic extensions, whose presentation was pleural effusion associated with a mediastinal mass suggestive of pulmonary neoplasia.

CASE REPORT

A 67-year-old female, without a significant medical history, presented with a 20-day history of sudden dyspnea. In her chest radiograph, an increased density in the right hemithorax with an ipsilateral pleural effusion was observed (Figure 1). The patient was admitted in hospital with a suspicion of lung cancer.

A chest computed tomographic scan demonstrated an extensive right pleural effusion with mediastinal shift to the left and injury of the T4 spinal segment with a predominantly lytic component, which affected the right posterior arch and the adjacent vertebral body.

Magnetic resonance imaging (MRI) showed a right paravertebral heterogeneous rounded mass extending from T4 to T5, with intrathoracic extension. It was isointense to the spinal cord on T1-weighted images, strongly hyperintense on T2-weighted images, and enhanced homogeneously upon intravenous gadolinium administration (T1-weighted images). The mass extended through ipsilateral conjunction

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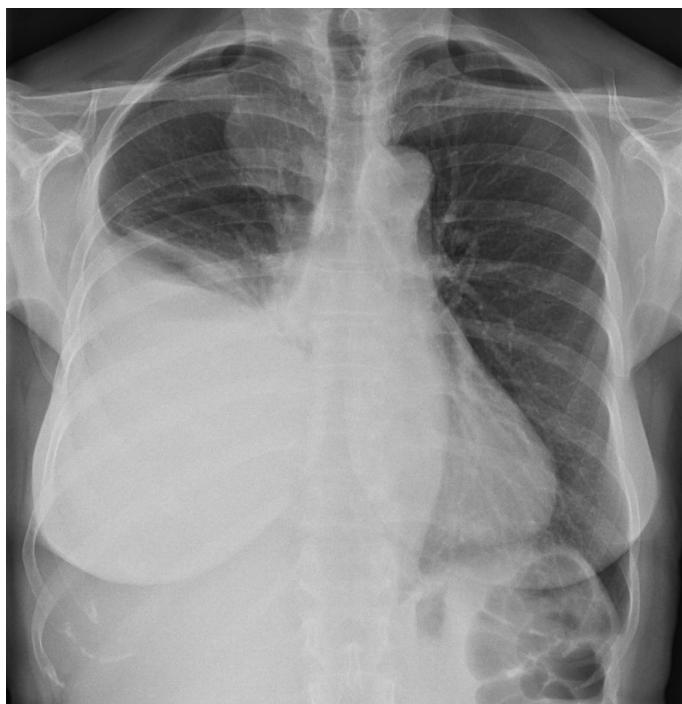


Figure 1. Chest radiograph shows an increased density in the right hemithorax with an ipsilateral pleural effusion.

foramen, widening and obliterating the epidural fat and displacing and compressing the contralateral spinal cord. No medullary hyperintensity indicating compressive myelopathy was observed (Figures 2 and 3 a, b, c).

Under neuronavigator guidance, a posterolateral approach was performed with a T4-T5 hemilaminectomy with removal of the right T4-T5 facet joint and the corresponding

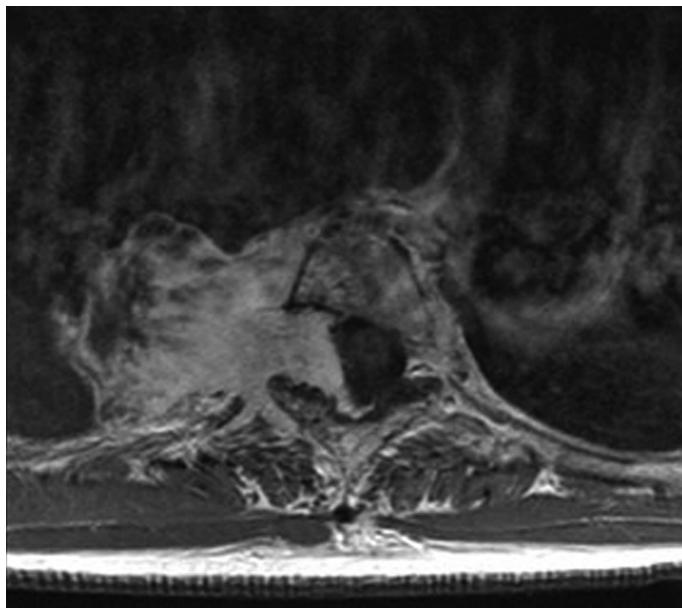


Figure 2. Axial magnetic resonance image shows a right epidural heterogeneous mass extending into the right intervertebral foramen with intrathoracic extension. It enhances homogeneously upon intravenous gadolinium administration on T1-weighted imaging.

costotransverse joint to expose the lesion. A red hemorrhagic epidural mass was observed, which was removed *en bloc* after circumferential dissection.

Microscopic evaluation showed a fibrofatty tissue with a proliferation of vascular structures, which were generally of small size, seating areas with myxoid appearance. No cellular atypia was seen (Figure 4). The diagnosis was a capillary hemangioma.

The patient had a good postoperative course, with full expansion of the right lung after an evacuate thoracentesis. A postoperative MRI test showed the usual postoperative changes and complete resection of the lesion (Figure 5).

DISCUSSION

Hemangiomas are congenital vascular malformations that pathologists frequently consider to be hamartomatous malformations.¹

Hemangiomas of the spine are usually lesions of the vertebral bodies, but they can seat in other locations.² The most frequent site of involvement in spinal cord is the intradural extramedullary compartment.³ Purely epidural hemangiomas are rare; only 8 cases in the English literature have been described.²⁻⁹ Seven of these cases involved thoracic spinal levels and 1 case involved a lumbar level.⁸ Three cases presented with an extraforaminal extension causing a dumbbell-shaped appearance in spinal MRI studies.^{3,5,9} Our patient is the fourth case with a dumbbell-shaped appearance reported in the literature and the second presented as a lung mass³ (Table 1).

The tumors may be iso- or hypointense on T1-weighted images, hyperintense on T2-weighted images, and enhanced relatively homogeneously on T1-weighted images with gadolinium.^{3,6,10} These radiological aspects are nonspecific, well-circumscribed tumors and the dumbbell shapes on magnetic resonance images are also commonly found in other tumors such as schwannoma, neurofibroma, or meningioma.

Histologically, hemangiomas are divided into capillary and cavernous types, depending on the dominant vessel size at microscopy.^{3,11} The cavernous type histologically presents a large number of sinusoidal channels in collagenous tissue,^{12,13} whereas the capillary hemangioma shows capsulated lesions characterized by lobules of thin irregular capillary-sized vessels lined by endothelial cells, which are separated by septa of fibrous connective tissue.^{2,11,12}

Capillary hemangiomas usually present in a chronic progressive manner because of mass effect and nerve root irritation.³ Cavernous hemangiomas, on the contrary, often present with an acute symptomatology, mainly related to intratumoral bleeding.^{7,14}

Complete surgical removal is the treatment of choice for epidural capillary hemangioma.^{15,16} *En bloc* removal of the tumor is preferred in appropriate cases. In our case, we planned a wide exposure of the lesion (including intrathoracic extension) through a posterolateral approach, a T4-T5 hemilaminectomy and removal of the right T4-T5 costotransverse joint. We performed a true *en bloc* removal.

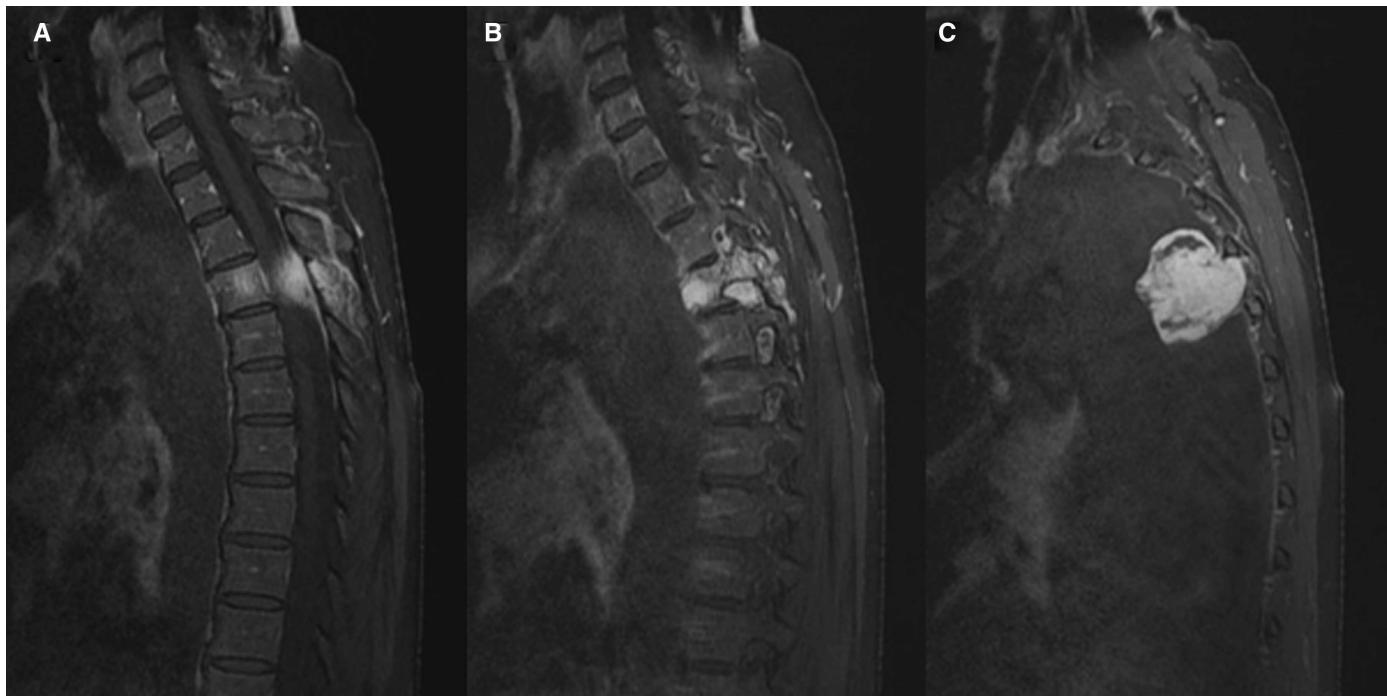


Figure 3. Sagittal gadolinium-enhanced T1-weighted magnetic resonance images showing an epidural lesion from T4 to T5 (**A**), extending through ipsilateral conjunction foramen (**B**) and with intrathoracic extension (**C**).

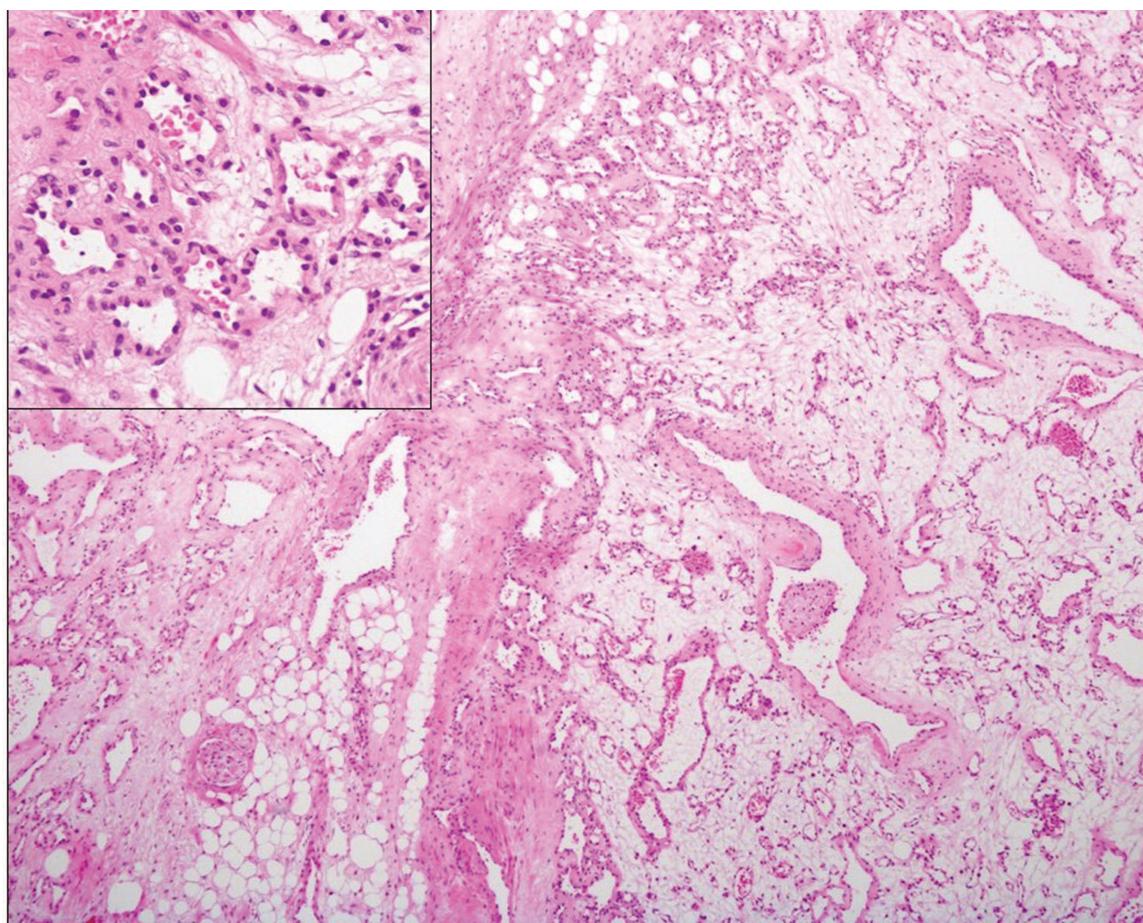


Figure 4. Photomicrographs obtained from tumor specimens. H&E-stained sections of capillary hemangioma consisting of small vessels proliferation (original magnification $\times 100$). Inset: small vessels lined by flattened mature endothelium (original magnification $\times 400$).

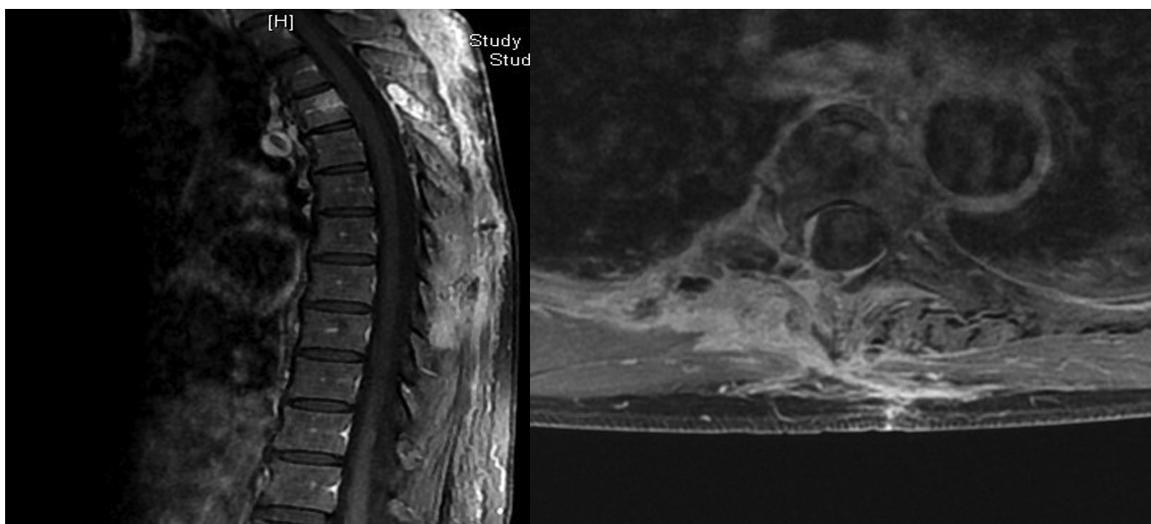


Figure 5. Postoperative sagittal (left) and axial (right) gadolinium-enhanced T1-weighted magnetic resonance images showing the usual postoperative changes and complete resection of the lesion.

TABLE 1. Epidural Capillary Hemangiomas Reported in the Literature

Authors	Age (yr)/Sex	Location	Presenting Symptoms	Extraforaminal Extension	Complete Removal
Gupta <i>et al</i> (1996) ⁶	50/M	T8–T10	Back pain, burning sensation of both limbs and weakness of the left lower limb	No	Yes
Badinand <i>et al</i> (2003) ⁵	40/F	T2–T4	Dorsal pain and muscle contracture, weakness in the lower limbs, and sphincter weakness	Yes	No
Kang <i>et al</i> (2000) ³	56/M	T2–T4	Chest wall pain	Yes	No
Tekin <i>et al</i> (2008) ⁸	56/F	L3–L4	Back pain radiating through right anterior thigh and hypoesthesia at medial foot.	No	Yes
Akhaddar <i>et al</i> (2010) ⁴	19/F	T5–T6	Paraplegia and urinary retention	No	Yes
Hasan <i>et al</i> (2011) ²	57/M	T10–T12	Dorsal pain, weakness in the right leg and sphincter weakness	No	Yes
Vassal <i>et al</i> (2011) ⁹	59/F	T5–T7	Back pain and right intercostal neuralgia	Yes	Yes
Seferi <i>et al</i> (2014) ⁷	58/M	T2–T4	Pain and weakness in both legs	No	Yes
Present case	67/F	T4–T5	Pleural effusion	Yes	Yes

F indicates female; M, male; L, lumbar; T, thoracic.

Recurrences have not been described in the literature and in most cases, as in our case, the pain improved considerably, and sensory and motor deficits were reversed.

CONCLUSION

It is important to consider the diagnosis of hemangiomas in the differential diagnosis of epidural lesions with dumbbell-shaped appearance in the magnetic resonance image, especially at the thoracic level. It is a benign and potentially curable disease and the most appropriate surgical treatment is *en bloc* resection of the entire lesion. They are usually presented as a progressive myelopathy, so early treatment may prevent permanent neurological deficit.

➤ Key Points

- Purely epidural hemangiomas of the spine are rare and most of them are of a cavernous type.
- The radiological aspects are nonspecific, well-circumscribed tumors and the dumbbell shapes on magnetic resonance image are also commonly found in other neurogenic tumors.
- Capillary hemangiomas usually present in a chronic progressive manner and cavernous type and often present with an acute symptomatology.

- It is a benign and potentially curable disease, and the most appropriate surgical treatment is *en bloc* resection of the entire lesion.

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