

Spinal Epidural Cavernous Hemangioma of the Thoracic Spine: A Case Report

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Abstract

Cavernous hemangiomas can arise in any region of the body, including the central nervous system. Spinal cavernous hemangiomas account for 5% - 12% of all cases of vertebral vascular malformation. Most of these are of vertebral origin, and cases that are non-vertebral in origin are rare. We encountered a patient with a relatively rare spinal epidural cavernous hemangioma of the thoracic spine that was non-vertebral in origin. The patient was a 63-year-old man. He had become aware of bilateral leg pain and numbness about 2 months earlier, and gait disturbance appeared gradually thereafter. On MRI, a lesion showing iso-intensity on T1-weighted imaging and high intensity on T2-weighted imaging was detected at the 7th thoracic vertebra. On gadolinium contrast-imaging, the lesion was found to be a homogenously-enhanced dumbbell-shaped extradural spinal neoplasm protruding from the left 7th/8th thoracic intervertebral foramen. A neurogenic tumor was suspected based on myelography and MRI findings, and complete tumorectomy was performed, which improved the lower limb symptoms and gait disturbance. The histopathological diagnosis was cavernous hemangioma. Epidural hemangiomas arise from the vertebra in many cases, and pure spinal epidural cavernous hemangiomas are rare. It is difficult to make a preoperative diagnosis because there are no specific imaging findings that can differentiate these tumors. It may be important to consider this disease before surgery in the differential diagnosis of epidural tumors.

Keywords

Epidural Cavernous Hemangioma, Thoracic Spine, Myelopathy

1. Introduction

Cavernous hemangiomas may develop in any region in the human body, and many cases of spinal epidural ca-

vernous hemangiomas arising from the vertebrae have been reported [1]. While expansion of cavernous hemangiomas of vertebral origin into the epidural space has also been reported occasionally, cavernous hemangiomas of purely spinal epidural origin are rare [2]. With respect to the differential diagnosis, there are no characteristic physical or imaging findings for such epidural tumors. Thus, it is difficult to make a definite diagnosis before surgery, due to the low incidence and the absence of characteristic imaging findings. We encountered a patient who developed spinal cord symptoms associated with spinal epidural cavernous hemangioma arising from outside the vertebra.

We report the case with a review of the literature.

2. Case Presentation

The patient was a 63-year-old man who had developed bilateral leg pain and numbness about 2 months earlier, with the subsequent gradual onset of gait disturbance. When he visited a physician, MRI findings suggested a spinal cord tumor at the 7th thoracic vertebral level. Thus, the patient was referred to our department. During the first examination, left-side dominant pain was noted in the bilateral thighs over the medial crura, but there was no tenderness or knocking pain in the dorsal region. No sensory disturbance was observed, and the bilateral lower limb muscle strength was normal. The patellar and Achilles tendon reflexes were enhanced on the left side, but no morbid reflex was observed, nor were there any bladder or rectal disturbances.

On chest plain radiography, there were no abnormal findings. Plain MRI demonstrated a dumbbell-shaped tumor protruding from the left 7th/8th thoracic intervertebral foramen, and a spinal epidural lesion with intensity equivalent to that of the spinal cord on T1-weighted imaging and high intensity on T2-weighted imaging. On gadolinium-contrast imaging, the lesion was mostly homogeneously enhanced (Figure 1). On myelography, the dural canal was excluded by an extramedullary tumor at the 7th thoracic vertebra (Figure 2). Schwannoma was suspected based on these findings, and complete tumorectomy was planned. When laminectomy was applied to the 7th thoracic vertebra, a lesion that occupied about half the circumferential space was noted in the left dorsal dura mater over the lateral side, and a dark reddish elastic soft mass with abundant blood vessels was present. To observe the entire mass, the left T7 inferior articular process was resected and the dumbbell-shaped mass was confirmed. Since the mass and nerve root were strongly adherent, the nerve root was ligated and transected and the mass was excised. After excision of the mass, bulging and pulsation of the dural canal appeared favorable (Figure 3). Histopathological examination showed many blood vessels of various sizes and a thin fibrous or smooth muscular wall. Some of these blood vessels were accompanied by organized thrombi in the lumen, and the mass was diagnosed as a cavernous hemangioma (Figure 4).

As of one year after surgery, no neurological findings have been observed, and the gait disturbance has improved. No recurrence has been observed on MRI, and the course has remained favorable (Figure 5).

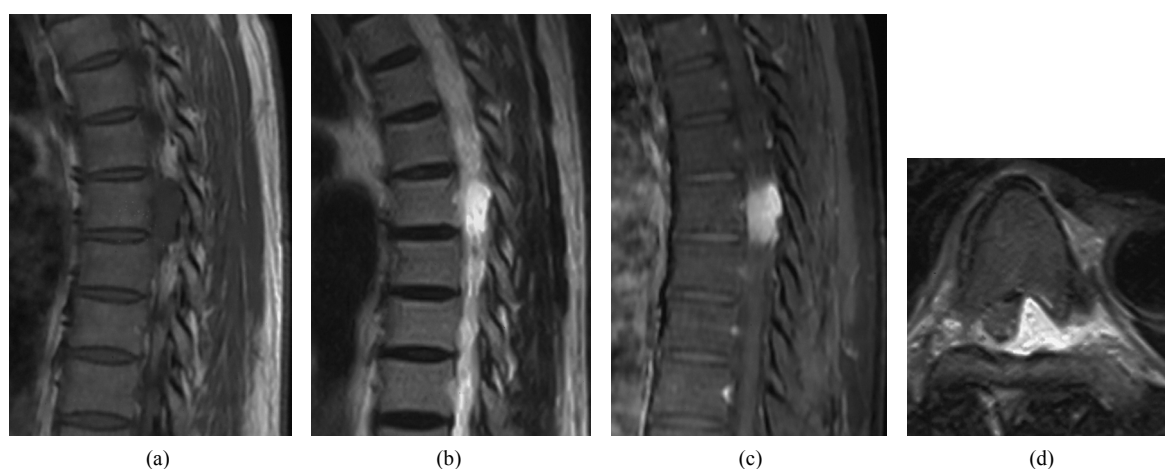


Figure 1. Thoracic spine MRI before operation. A space occupying lesion was evident showing iso-intensity on the T1 weighted image and high intensity on the T2 weighted image that was enhanced with gadolinium-contrast imaging. The tumor was dumbbell-shaped, protruding from the left 7th/8th thoracic intervertebral foramen. (a) Sagittal T1 WI; (b) Sagittal T2 WI; (c) Sagittal Gd-DTPA; (d) Axial T2 WI.

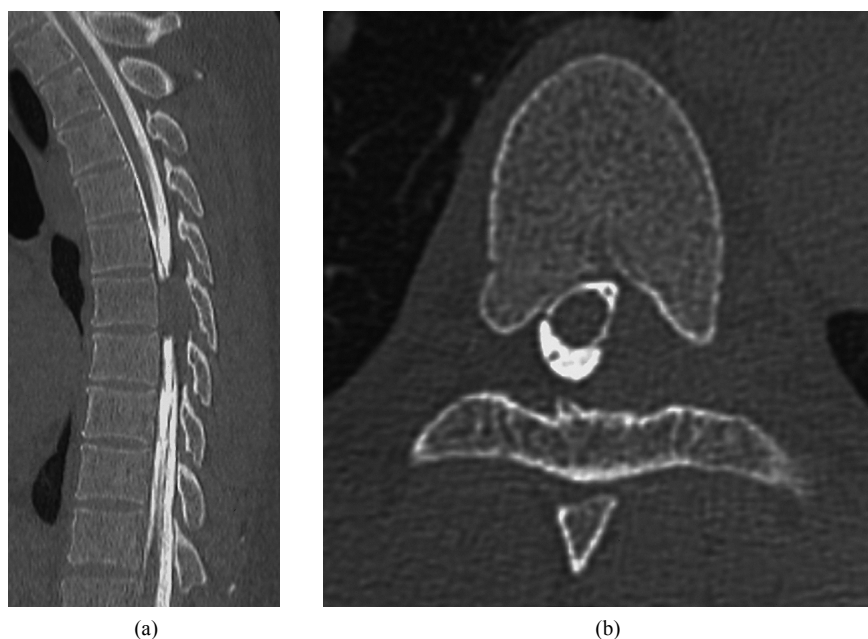


Figure 2. CT myelogram. (a) Sagittal reconstruction CT after myelogram; (b) Axial CT after myelogram.

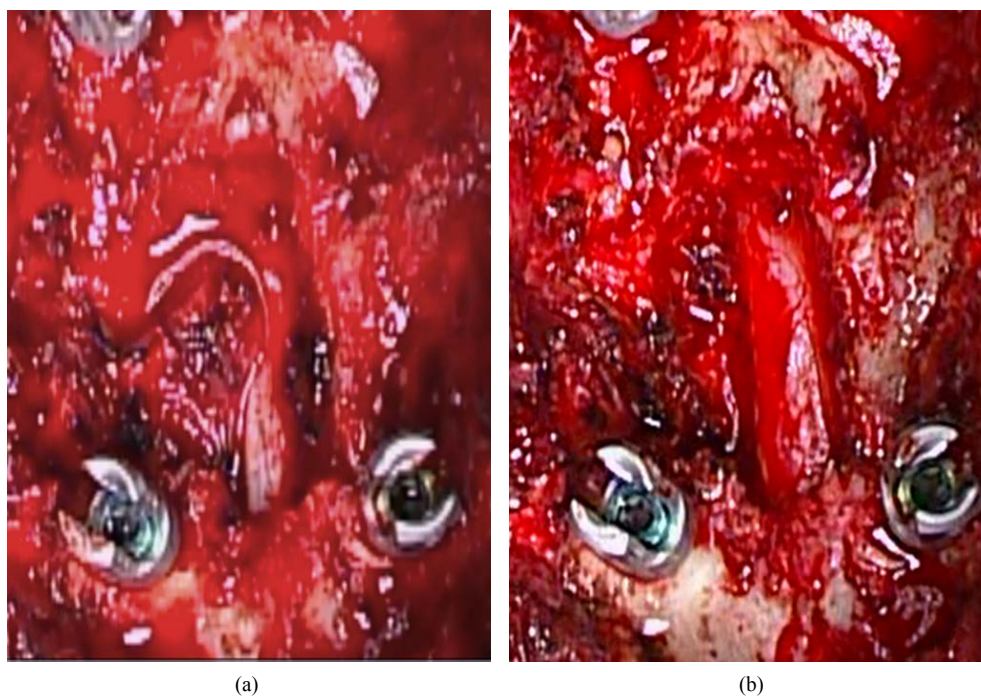


Figure 3. Intraoperative findings. (a) A dark reddish elastic soft mass with abundant blood vessels was present in the dorsal dura mater over the lateral side; (b) After excision of the mass, bulging and pulsation of the dural canal appeared favorable.

3. Discussion

Cavernous hemangiomas can arise in any region of the body, including the central nervous system. Epidemiologically, according to Hillman *et al.* [1], about 220,000 people worldwide develop cavernous hemangiomas annually, and cases of vertebral origin are relatively rare. Spinal cavernous hemangiomas account for 5% - 12% of all cases of vertebral vascular malformation. While most cases are of vertebral origin, cases of extravertebral origin account for only 0.4% [2]-[4]. Epidural cavernous hemangiomas account for about 4% of all spinal epidural

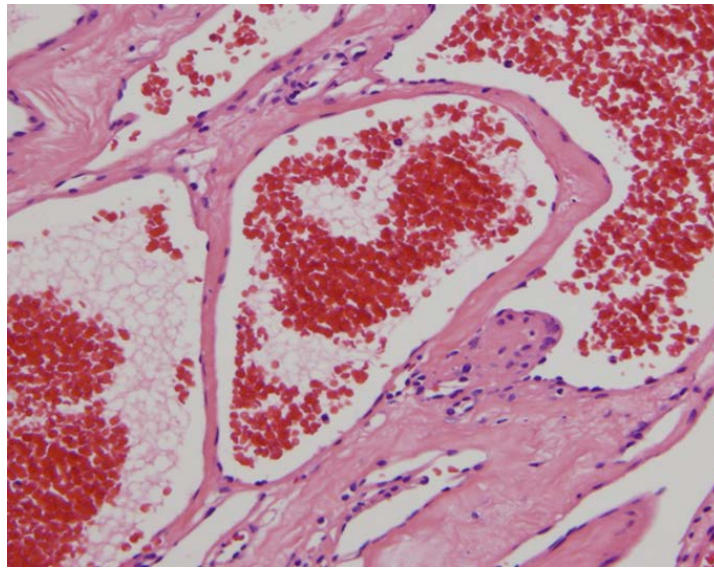


Figure 4. Histological examination showed many vessels of various sizes and a thin fibrous or smooth muscular wall. Some of these blood vessels were accompanied by organized thrombi within the lumen (Hematoxylin and eosin 100 \times).

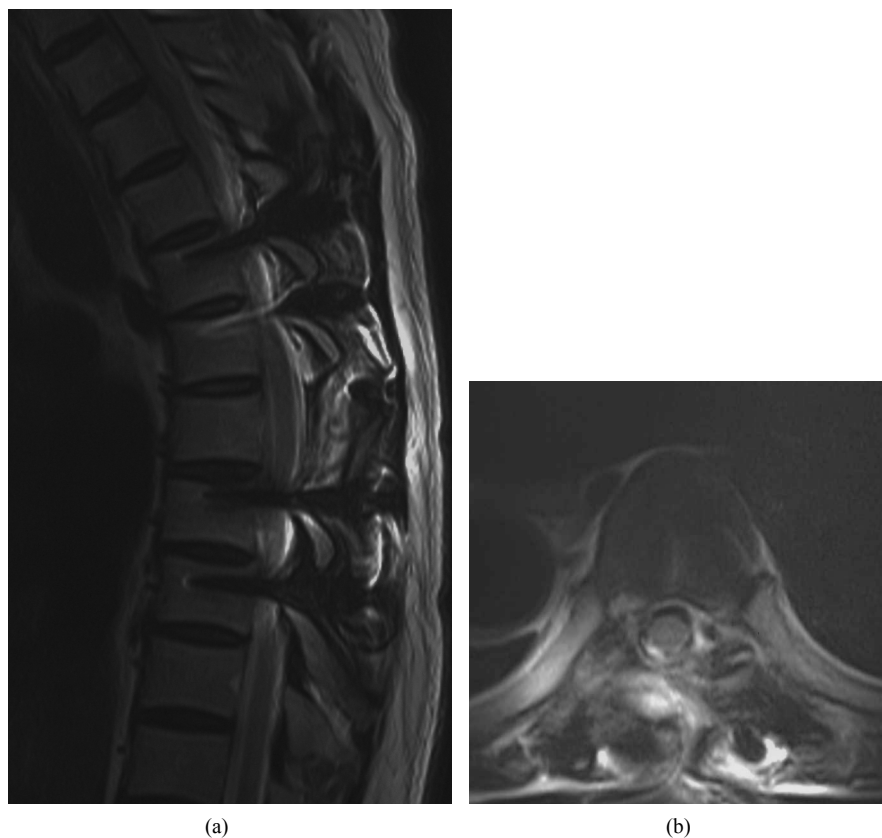


Figure 5. Thoracic spine MRI after operation. As of one year after surgery, no recurrence has been observed on MRI. (a) Sagittal T2 WI; (b) Axial T2 WI.

lesions [2]. However, purely epidural cavernous hemangiomas are rare, and, to our knowledge, only 24 cases have been reported in Japan [3]-[21]. In other countries, 99 cases have been reported [1] [2] [22]-[64], and 55 [3]-[29] [32] [35]-[38] [41] [45] [46] [52] [55] [60] were reported from Asian countries. When the Japanese cases

were investigated, the mean age was 53.7 years old (7 - 80 years old), and the male: female ratio was 7:4; showing that the incidence tends to be higher in men. The development site was the cervical spinal cord in 2 (8%), the cervicothoracic spinal cord in 3 (13%), the thoracic spinal cord in 13 (54%), the thoracolumbar spinal cord in 2 (8%), the lumbar spinal cord in 2 (8%), and unclear in 2 (8%); thus, development from the thoracic spinal cord accounted for more than half of such cases (54%). The initial symptoms have been diverse, including myelopathy, radiculopathy, and local pain [1], in 12 (50%), 8 (33%), 3 (13%) of the Japanese cases, respectively, and unclear in 1 (4%). In our patient, the initial symptom was myelopathy. With respect to the pattern of development, when the time to surgery after onset was divided into acute (<1 month), subacute (1 - 6 months), and chronic (6 months or later), the development was acute, subacute, and chronic in 4 (17%), 3 (13%), and 11 (46%), respectively, and was unclear in 6 (25%). Three chronic cases that became acutely aggravated were treated with emergency surgery.

Among the imaging findings for this disease, Padovani *et al.* [51] and Kurose *et al.* [18] have reported dilation of the intervertebral foramen on plain radiography; however, this was not observed in our patient. Furthermore, dilation of the intervertebral foramen on plain radiography has been described as not being characteristic of epidural cavernous hemangioma in many reports.

On myelography, exclusion of the dural canal was observed, but this is also a general finding of epidural tumors that is not specific to this disease.

Iso-low- and high-intensity regions are generally observed on T1- and T2-weighted imaging, respectively, and enhancement is observed on gadolinium contrast imaging [3] [4]; however, these findings are also not specific to this disease.

With respect to the clinical diagnosis, there are no characteristic physical or imaging findings, and the tumor could be diagnosed as schwannoma, epidural hematoma, and epidural abscess, excluding cases that were pathologically diagnosed before surgery using VATS. Thus, it is difficult to make a definitive diagnosis before surgery due to the low incidence and the absence of characteristic imaging findings.

For treatment, surgery is generally selected [3] [7] [51]. While one conservatively-treated case has been reported in another country, the outcome was poor [3]. All Japanese cases were treated with tumor resection under decompression. In our patient, since the tumor was not diagnosed before surgery, we selected tumorectomy combining facetectomy and fixation to safely allow observation of the entire tumor. The postoperative course was favorable, with disappearance of the spinal cord symptoms and improvement of the gait disturbance.

It is difficult to preoperatively diagnose epidural cavernous hemangiomas, because there are no characteristic clinical symptoms or imaging findings and the incidence low. When a tumor lesion is observed, it is important to suspect epidural cavernous hemangioma in the differential diagnosis of epidural tumors before surgery.

4. Conclusion

We encountered a patient with spinal epidural cavernous hemangioma of the thoracic spine. There are very few reported cases of this disease, and its preoperative diagnosis is difficult. The disease could not be diagnosed before surgery in our patient. However, complete excision was performed and a favorable outcome was achieved.

Consent

Informed consent was obtained from the patient for publication of this case report.

Conflicts of Interest

The authors declare that they have no conflicts of interest with respect to the publication of this case report.

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