

Case Report

Thoracic Extraosseous Epidural Cavernous Hemangioma

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Cavernous hemangiomas were first reported in 1929 by Globus and Doshay, and are defined as benign vascular structures developed between the neural tissues occurring in the central nervous system, consisting of a dilated vascular bed. Cavernous hemangiomas comprise nearly 5–12% of all spinal vascular malformations; however, existence in the epidural space without bone involvement is rare. Only 4% of all cavernous hemangiomas (0.22/1.000.000) are purely epidural cavernous hemangiomas. In this case report, we removed a hemorrhagic thoracic mass presenting with progressive neurological deficits in a 55-year-old male patient. We found this case to be appropriate for presentation due to the rare occurrence of this type of cavernous hemangioma.

Key Words : Cavernous hemangioma · Extraosseous-epidural · Thoracic vertebrae · Hemangioma.

INTRODUCTION

Cavernous hemangiomas (CHs) are defined as benign vascular structures inserted within the neural tissue, occurring in the central nervous system, and consisting of a dilated vascular bed⁵. They may be located in the cerebral cortex, cerebellum or spinal cord⁵. CHs create 5–12% of all spinal vascular malformations¹⁰. Existence in the epidural space without bone involvement is rare^{1,10}, and only 4% of all CHs (0.22/1.000.000) are purely epidural^{1,10}. Clinical symptoms are usually seen as slowly progressive paraparesis, myelopathy and localized pain^{1,5}. Although magnetic resonance imaging (MRI) is the best diagnostic method for this lesion, it is not a definitive one⁸. We felt it appropriate to present this case because CHs in the epidural space have rarely been encountered.

CASE REPORT

A 55-year-old male patient was admitted to our clinic with complaints of back pain beginning two years previously. Additionally, he had progressive spastic paraparesis in both lower extremities, and an inability to stand without support for the previous 3 months. Upon neurological examination, the motor power in both lower extremities was II–III/V, deep tendon reflexes were

hyperactive, and hypesthesia was present under the T10–11 level. The patient had no history of urinary or fecal incontinence, and the laboratory tests were within normal limits.

Routine X-rays were normal. However, in the thoracic, lumbar, and cervical MRIs performed for further investigation, a mass lesion was seen along the T7–8 vertebral body levels. This lesion was hypointense upon the T1-weighted images (Fig. 1A), with a slightly lower signal than the cerebrospinal fluid (CSF) on the T2-weighted images (Fig. 1B), and slight heterogeneous staining on the gadolinium-enhanced T1-weighted images (Fig. 1C). Additionally, it was observed that the spinal cord was displaced to the right side due to the mass (Fig. 2).

Under general anesthesia, a T8–9 total laminectomy was performed and an approximately 3×1.5 cm sized (Fig. 3A) dark-colored vascular mass with soft consistency was found in the epidural space without invasion of the dura (Fig. 3B), which was completely excised microsurgically.

The histopathological examination revealed a cavernous hemangioma, composed of large dilated blood-filled vessels lined with flattened endothelium (Fig. 4). Neurological deterioration was not found during the postoperative period. Ten days postoperatively, the patient could walk independently and reported reduction in the localized back pain.

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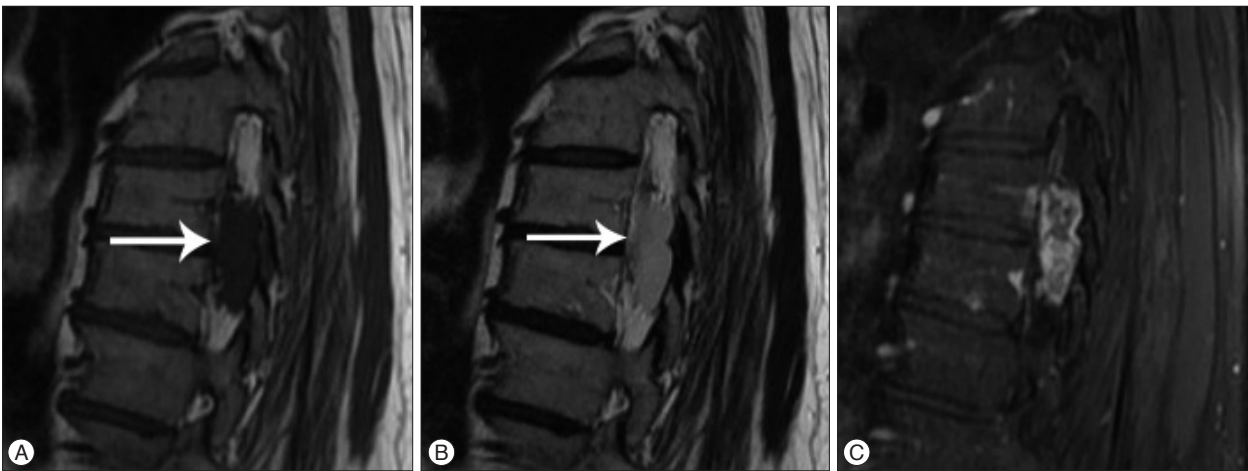


Fig. 1. Mass lesion with hypointensity in T1-weighted images (arrow) (A), with slightly lower signal than CSF in T2-weighted images (arrow) (B), and slightly heterogeneous gadolinium-enhanced T1-weighted images (C) upon sagittal thoracic spine MRI. CSF : cerebrospinal fluid.

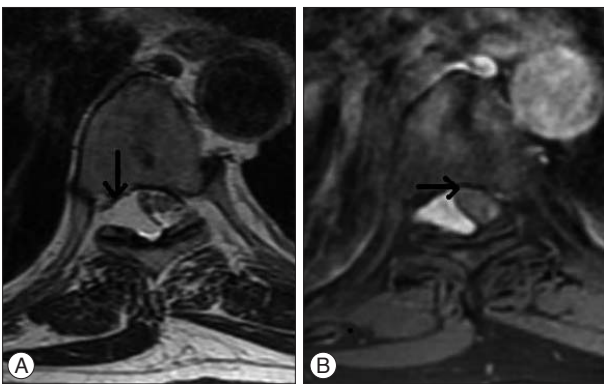


Fig. 2. Mass displacing the spinal cord to the right side in the axial plane in the T2-weighted (A) and fat suppression T1-weighted enhanced (B) images upon the MRI.

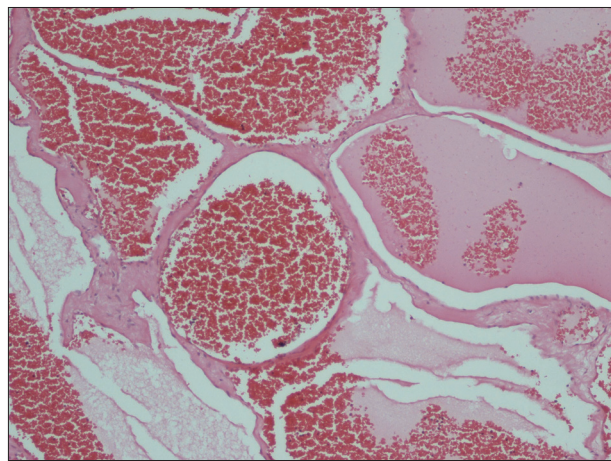


Fig. 4. Flattened endothelium-coated large dilated blood vessels (H&E, $\times 40$).

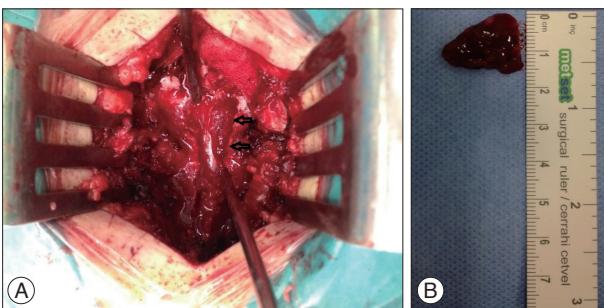


Fig. 3. Extradural hemorrhagic mass located in the operating field (arrows) (A) and its gross finding after total removal (B).

DISCUSSION

CHs were first reported in the literature in 1929 by Globus and Doshay^{4,6,8}. CHs are collections 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 channels⁶. CHs are not true neoplasms because they grow with hypertrophy without mitotic activity². How-

ever, they create clinical signs with mass effects, fluid movement within vascular structure, hemorrhages, thromboses, and the formation of cysts or caverns⁶. These lesions can occur in every region of the body; however, the most common area of occurrence in the cranial region of the central nervous system is the supratentorial area⁸. CHs have rarely been seen in the spinal canal^{1,6,10}, while only 4% of all cavernous hemangiomas (0.22/1,000,000) are purely epidural CHs^{1,10}. Spinal CHs appear at an average age of 40 (30–60) and are more common in women (70%). According to frequency, the localization in the spinal vertebrae can be listed in order of occurrence as in the thoracic (54–60%), cervical (30%), and lumbar (10%) vertebral regions^{1,7,9}; they are seen most commonly in the posterior part of the spinal canal^{1,9}. There are two currently accepted hypotheses in the literature with regard to the frequent occurrence in the thoracic vertebrae; the first is that the thoracic epidural space is wider^{3,6}, while the second is that the resistance in the posterior part of the thoracic spinal canal is lower^{3,6}. Besides the sizes are very diverse^{3,6,7} the localization of the CH in our fifty-five-year-old male patient was con-

sistent with the literature.

Although clinical symptoms may vary according to the localization, size and biological behavior, slow progressive paraparesis (71%) and radiculopathy (19%) are seen most commonly^{1,6-10}. Acute presentation may occur due to either extradural hemorrhage or thrombotic occlusion within the cavernoma, by causing rapid increase in the bulk of the lesion^{6,8}. In our case, the symptoms were presented as back pain and progressive paraparesis.

Spine MRI is the best method for the radiological diagnosis of CHs. Upon MRI, the demarcated lobular lesion appears isointense in the T1-weighted images, hyperintense in the T2-weighted images, and intensely homogeneous with contrast enhancement in the contrast-enhanced T1-weighted images^{3,9}. In case of intramedullary CHs, a hypointense ring due to peripheral hemosiderin deposition and heterogeneous intensity in the center are seen, but these are not observed in epidural hemangiomas^{3,5,7,9}. Although differential diagnosis is most commonly required with schwannoma in MRIs, it may be confused with other neurogenic tumors, metastasis, lymphoma, meningioma, multiple myeloma, extraosseous Ewing's sarcoma, disc fragments or epidural angioliipoma^{6,8}. In our case, the absence of a hemosiderin ring, as well as the displacement of the spinal cord, slightly lower signal than the CSF signal in the T2-weighted images, and slightly heterogeneous significant contrast involvement suggested epidural hemangioma⁶.

CHs have the tendency to bleed and grow⁶. Because of these behaviors, early surgical excision is recommended as soon as possible. Aim of the surgery should be total removal at first operation⁶; however, intraoperative bleeding and intramedullary placement may impede the total removal. Although hemorrhage is rare in intramedullary CHs, massive bleeding may be seen in epidural CHs^{1,2,6,8,10}. In cases of incomplete removal due to bleeding and its localization, adjuvant radiotherapy may be required^{6,8}. In our case, the bleeding in the perioperative period could be controlled surgically and adjuvant radiotherapy was not needed.

CONCLUSION

CHs are non-neoplastic vascular malformations rarely seen in the spinal region, leading to a variety of clinical symptoms. In MRIs, it is very difficult to distinguish these from schwannomas. In this case with this kind of spinal epidural CH, despite wrong diagnosis is likely to occur, the author could perform complete removal of this vascular malformation microsurgically.

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