INTRODUCTION

Non-traumatic spontaneous spinal epidural hematoma (SSEH) represents an extremely rare cause of spinal cord compression. Some researchers believe that venous bleeding leads to SSEH formation. However, others have observed some vascular malformations in patients with non-traumatic SSEH. Spinal arteriovenous malformation (AVM) is rare and accounts for 5% to 10% of all spinal cord lesions. An accurate diagnosis of spinal AVM is harder than that of cerebral AVM. Because of the very small size of the spinal lesion, its detection using spinal angiography is technically difficult. Moreover, the vasculature varies greatly depending on the location, type, and spinal level of the lesion. Regarding the treatment of spinal AVM, despite many reports on various analyses of various treatment modalities, including surgery, embolization, or a combination of the two, currently, no criteria for accurate diagnosis and treatment of spinal AVM exist.

Herein, we report a rare case of thoracic epidural spinal
AVM that caused SSEH and was associated with neurological impairment. This case emphasizes that accurate diagnosis and neurological recovery could be achieved in patients with SSEH through biopsy and surgical removal of the hematoma and AVM.

CASE REPORT

A 34-year-old man without underlying medical conditions presented with complaints of sudden tearing chest pain that radiated to the backside. He was admitted to the emergency room. After the patient was administered an analgesic, his chest pain gradually decreased, and he was about to be discharged. However, 3 hr after his admission to the emergency room, he complained of progressive motor weakness in the lower extremities (motor grade 4, which worsened to motor grade 1). Thoracic spine magnetic resonance imaging (MRI) showed a slightly elevated T2 signal at the T5–6 level, an epidural hematoma compressing the spinal cord, and suspicious findings of an AVM in the center of the hematoma (Fig. 1).

An emergent operation with a posterior approach was performed because of a neurologic deficit. T5 and T6 laminotomies were performed, following which the epidural hematoma was identified and removed mainly from the T5 level. An engorged vein and a small artery were identified on the dural surface at the T5 level (Fig. 2). The vessels were coagulated and ligated with clips. Laminae were attached to the detached site. Microscopic examination revealed an area of angiomatous structures containing highly packed red blood cells in the endothelial layer and collagen fibers (Fig. 3A). Immunohistochemical staining for smooth muscle

![Fig. 1. Sagittal (A) and axial (B) thoracic spine magnetic resonance images show an epidural mass with a slightly elevated T2 signal at the T5–6 level.](image)

![Fig. 2. An engorged vein (A) and a small artery (B) are shown.](image)

![Fig. 3. Histologic sections. (A) Representative area of the angiomatous structure (hematoxylin and eosin stain, ×20 magnification). (B) Immunohistochemical staining for smooth muscle actin (×20). (C) Immunohistochemical staining for elastic fibers (elastic stain, ×20).](image)
actin revealed fibrous tissue filled with red blood cells (Fig. 3B). Further, immunohistochemical staining (elastic stain) for collagen revealed collagen fibers and occasional smooth muscle fibers, consistent with AVM (Fig. 3C).

Postoperative thoracic spine MRI suggested gross total removal of hematoma (Fig. 4). After surgery, the patient was administered methylprednisolone 1000 mg (250 mg qid), which was gradually reduced for 6 days. Spinal angiography was performed 2 days after surgery, and the angiography showed no evidence of vascular malformations (Fig. 5). The patient’s neurological status improved postoperatively. Sensory disturbance disappeared, and motor weakness improved rapidly. After 7 days of operation, the patient was discharged without any special treatment other than analgesic (tridol, aceclofenac). On a postoperative day 8, the neurological status showed improvement except for weakness in the left lower extremity (motor grade 4–). At outpatient follow-up 2 years postoperatively, further improvement was observed with a motor grade of 4+.

DISCUSSION

SSEH is a rare disease, usually seen in the lower and dorsal regions of the cervical spine in children and adolescents and the thoracic or thoracic lumbar region in adults.\textsuperscript{17,20}

The usual clinical symptom of SSEH is acute severe back pain, which may be distributed in the skin, and is followed by clinical signs and symptoms of spinal cord dysfunction. Symptoms may be atypical and non-specific, which may delay the diagnosis.\textsuperscript{15,26} Moreover, SSEH is difficult to diagnose because it can be associated with anticoagulant therapy, blood disorders, vascular malformations, or neoplasia.

In the present case, the spinal epidural AVM was observed intraoperatively, and accurate diagnosis and spinal cord decompression were performed through surgery.

Most spinal AVMs are dural arteriovenous fistulas with a single intradural venous drainage from the epidural nidus, and the most common clinically epidural spinal AVM is a vertebral body (cavernous) hemangioma, which is an epidural vertebral canal, with almost no observed invasion. Symptomatic epidural spinal AVM of the spinal canal without vertebral (cavernous) hemangioma has been rarely reported.\textsuperscript{2,8,9,12} Therefore, the diagnosis and treatment were considerably difficult in the present case.

A common treatment goal for spinal AVM is to eliminate
venous congestion, blood theft, or venous hypertension, and there are three treatment options—endovascular embolization (using balloons, particles, or coils), surgical excision, or both. However, the period between symptom onset and surgical intervention is important. In particular, surgery performed 12 hr after symptom onset is less likely to be successful.

In the present case, progressively worsening neurological symptoms were present, and the hematoma was observed on MRI. During the operation, AVM was diagnosed and treated simultaneously. The intervention of neurosurgery seemed incontrovertible given that the patient’s worsening paralysis was resolved after the operation. The hemorrhagic lesion that was not accurately diagnosed even on MRI could be diagnosed during surgery, and it was confirmed that the lesion could be completely removed only with surgery.

Furthermore, complications of embolization have been observed in past studies. Berenstein and Choi reported that in patients with intracranial AVM, hemorrhagic complications occurring during embolization were associated with either iatrogenic rupture of a cerebral vessel or rupture of a blood flow-induced aneurysm or the AVM itself. Additionally, Djindjian had warned of the risk of acute bleeding after embolization. Thus, the present case indicates that surgical diagnosis and treatment are most effective in patients with spinal epidural AVM associated with bleeding, where the diagnosis is difficult and rapid neurological recovery is required. However, a study by Forbes et al. reported a neurological deficit rate of only 2%, and the neurological deficits resolved completely within 24 hr in 2 patients and within 1 week in 1 patient.

In this case, the patient’s weakness was due to spinal cord compression, and the AVM was finally diagnosed and successfully removed through surgery. However, unless it is an emergency, we should be considered that AVM should be a factor to consider before surgery, and its exact location should be assessed by angiography to prevent incomplete removal and recurrence.

**CONCLUSION**

Our case demonstrated that even when the cause of SSEH is difficult to diagnose by MRI, rapid neurological recovery is possible through accurate diagnosis and prompt surgical treatment of an AVM. Endovascular angiography and embolization can also be considered; however, in the case of severe neurological disorders due to massive hematoma, immediate surgical treatment and histopathological diagnosis of the lesion should be considered.

**ETHICS APPROVAL AND CONSENT TO PARTICIPATE**

This study was approved by the Institutional Review Board (IRB) at the participating medical center (IRB no. 2022-0836).

**CONFLICTS OF INTEREST**

No potential conflict of interest relevant to this article was reported.

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