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Acute spontaneous thoracic epidural hematoma associated with intraspinal lymphangioma: A case report

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Abstract

BACKGROUND

Spontaneous spinal epidural hematoma is a rare neurosurgical emergency.

CASE SUMMARY

A 53-year-old healthy woman suffered from complete paraplegia in both legs and loss of all sensation below the xiphoid process. She was diagnosed as acute spontaneous thoracic epidural hematoma caused by an intraspinal lymphangioma. The primary lab survey showed all within normal limits. Presence of a posteriorly epidural space-occupying lesion at the T4-T8 level of the spinal canal was confirmed on magnetic resonance imaging. A decompressive laminectomy was performed from the T4 to T7 levels at the sixth hour following abrupt onset of complete paraplegia. The lesion was confirmed as lymphangioma. This patient recovered well within one month.

CONCLUSION

This study reports a case of acute spontaneous thoracic epidural hematoma caused by an intraspinal lymphangioma with well recovery after surgical

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Core Tip: This case suffered from acute spontaneous thoracic epidural hematoma caused by an intraspinal lymphangioma presenting with complete paraplegia in both legs and loss of all sensation below the xiphoid process. A posteriorly epidural space-occupying lesion at the T4-T8 level of the spinal canal was relieved *via* decompressive laminectomy in time and the symptoms recovered well within one month.

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INTRODUCTION

Lymphangiomas that present as spinal soft tissue tumors in the epidural space are extremely uncommon[1-5]. However, these spontaneous spinal epidural hematomas can be a rare cause of spinal cord compression, accounting for less than 1% of spinal space-occupying lesions[6]. In this report, we present probably the first reported case of an acute spontaneous thoracic epidural hematoma caused by an intraspinal lymphangioma in a 53-year-old female patient.

CASE PRESENTATION

Chief complaints

A 53-year-old healthy woman was admitted to our emergency room with complete paraplegia in both legs and loss of all sensation below the xiphoid process (T5 level).

History of present illness

One day before admission, she had taken a walk with her husband along the dam in Kaohsiung. While walking at that time, she was only wearing a backpack and did not do any specific stretching exercises. After the walk, the patient felt a slight discomfort over her back. By noon on the next day, she complained of tightness over her bilateral chest wall, around the level of the xiphoid process. She denied straining, lifting, crying, sneezing, or coughing in the past three days. On arrival at the emergency room, 27 h after the walk, the muscle power in her lower extremities was 0/5 strength bilaterally, and she experienced no sensation below the xiphoid process. She also had difficulty in voiding her bladder. Vital signs at presentation were body temperature (BT): 36.6 °C, respiratory rate (RR): 17/min, pulse rate (PR): 82/min and blood pressure (BP): 156/82 mmHg. A neurological examination revealed increased deep tendon reflexes over the right and left knees and ankles.

The patient was not receiving any anticoagulation therapy, and laboratory test results, including a complete blood count, chemistry panel, and coagulation profile, were all within normal limits.

History of past illness

The patient denied related past illness or any history of anticoagulation therapy.

Personal and family history

The patient denied related personal and family history.

Physical examination

Vital signs at presentation were BT: 36.6 °C, RR: 17/min, PR: 82/min and BP: 156/82 mmHg. A neurological examination revealed increased deep tendon reflexes over the right and left knees and ankles.

Laboratory examinations

Laboratory test results, including a complete blood count, chemistry panel, and coagulation profile, were all within normal limits (platelets: 206000/ μ L, PT: 10.1 s, INR: 0.96, and APTT: 28.2 s). Microscopic examination of the intrathoracic abnormally dilated vascularities showed a proliferation of varying sized and focally dilated vascular channels filled with blood and lymphatic substance (Figure 1). The pathologic diagnosis was lymphangioma.

Imaging examinations

Magnetic resonance imaging (MRI) of the thoracic spine, performed one hour after abrupt onset of complete paraplegia, revealed the presence of a posteriorly epidural space-occupying lesion (7.6 mm $\times$ 18 mm $\times$ 85 mm in size) at the T4-T8 level of the spinal canal. The lesion caused obliteration of the cerebrospinal fluid (CSF) signal at the T6-T7 level thecal sac, with mildly thin peripheral enhancement (Figure 2). The scanned field showed neither a definite signal intensity changes in the spinal cord nor a significant bone marrow signal change in the vertebral body.

FINAL DIAGNOSIS

Based on magnetic resonance imaging, intraoperative findings, and pathological examination, acute thoracic epidural hematoma associated with intraspinal lymphangioma was diagnosed.

TREATMENT

At the sixth hour following abrupt onset of complete paraplegia, a decompressive laminectomy was performed from the T4 to T7 levels. This procedure exposed an acute epidural hematoma in the T4-T7 region, predominantly at T6-T7 (Figure 3). During the surgery, numerous epidural vessels, with some abnormally dilated vascularities, were found at the T4 to upper T6 levels (Figure 4).

OUTCOME AND FOLLOW-UP

Postoperative spinal angiography performed at 3 d following the thoracic spinal surgery showed the artery of Adamkiewicz arising from the right T10 radiculomedullary artery (Figure 5). No gross fistula channel or aneurysmal dilatation arising from radiculopial or radiculomedullary arteries was observed at the T5 to T11 levels.

A follow-up MRI of the thoracic spine performed 7 d postoperatively revealed no residual gross epidural hematoma and no remaining spinal cord compression except for a faint T2 hyperintense signal at the T6-T8 level spinal cord (Figure 6). On the seventh postoperative day, the patient experienced substantial recovery of the muscle power in both legs (grade 4). The sensation of proprioception, pain, pin-prick, and vibration also returned to normal. At discharge (1 mo after the operation), she regained complete sphincteric control.

DISCUSSION

Nontraumatic spinal epidural hematoma was first described by Jackson in 1869[7]. Spontaneous spinal epidural hematoma (SSEH) is a rare disease entity that is usually associated with underlying hematological disorders or other predisposing conditions[8]. Notably, lymphangiomas presenting as a spinal soft tissue tumor in the epidural space are so extremely rare[1-5] that SSEH caused by intraspinal lymphangioma has never been reported previously in the literature. To our knowledge, the case described herein is probably the first reported acute spontaneous spinal epidural

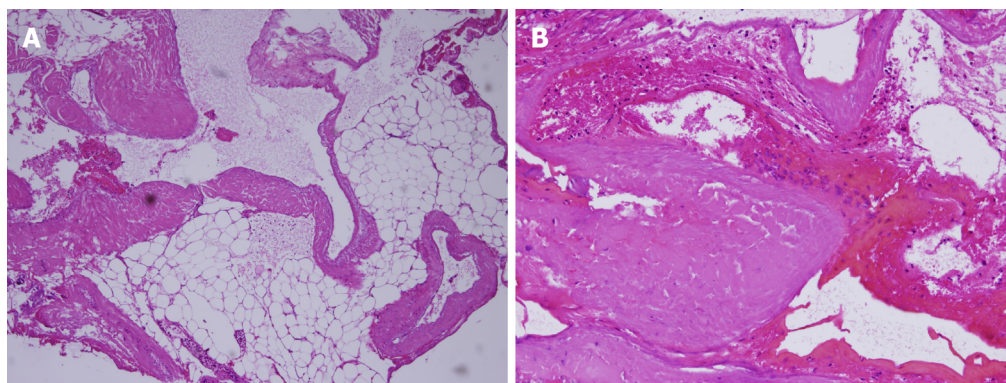


Figure 1 Hematoxylin and eosin staining. A: Proliferation of irregular thick wall vascular channels lined by flat endothelial cells with clear lymphatic fluid in the lumens [hematoxylin and eosin (H&E), 40 x]; B: Some of the proliferative vessels are filled with blood, instead of lymphatic substance (H&E, 100 x).

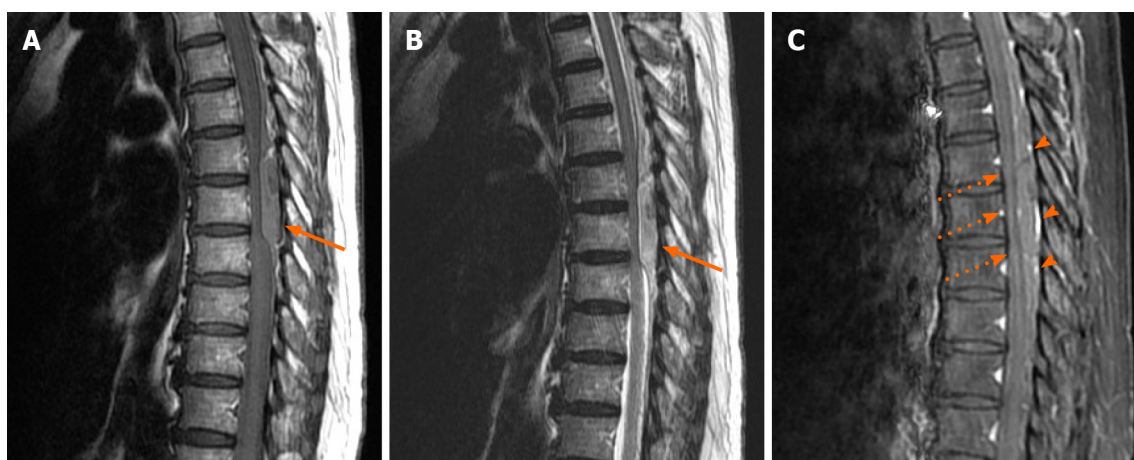


Figure 2 Preoperative spinal magnetic resonance imaging demonstrating an intraspinal epidural hematoma at the T4 to the T8 levels (arrows). A: Sagittal T1-weighted image (T1WI); B: Sagittal T2-weighted image; C: Sagittal T1WI with enhancement showing mildly thin peripheral enhancement (arrowheads) and the spinal cord is compressed and flattened (dashed thin arrows).

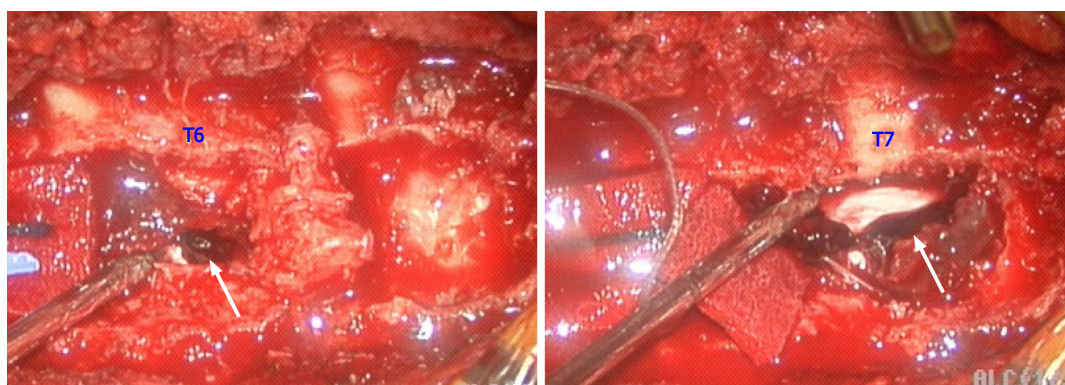


Figure 3 Dark reddish blood (arrows) in the epidural space was exposed after a laminectomy.

hemorrhage resulting from an intraspinal lymphangioma.

Lymphangiomas are congenital lymphatic malformations that are usually discovered in early childhood. They have no racial or gender predilection[9]. In up to 10% of patients, the malformations may not be discovered until adulthood, with only a few cases appearing as late as the fifth decade of life. Frequently, a lymphangioma manifests as an asymptomatic soft or semi-firm mass. Although these lesions tend to surround and sometimes invade normal anatomic structures, they have no malignant potential[10]. However, lymphangiomas presenting as a spinal soft tissue tumor in the

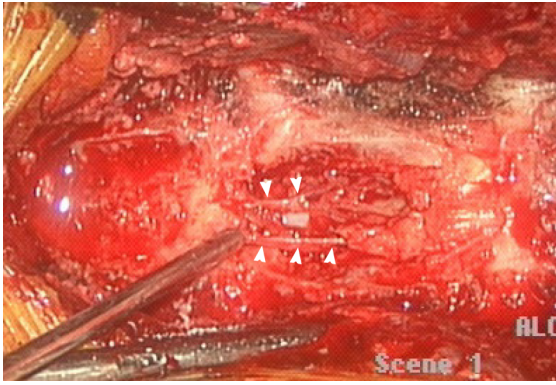


Figure 4 After a laminectomy of T5, numerous epidural vessels (arrowheads) were found.

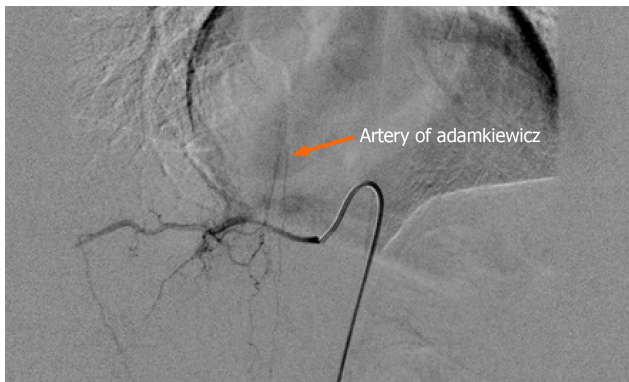


Figure 5 Postoperative spinal angiography shows the artery of Adamkiewicz arising from the right radiculomedullary artery at T10 level.

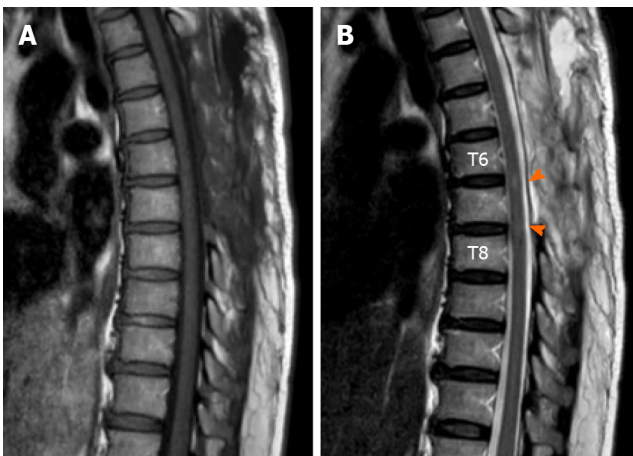


Figure 6 Postoperative spinal magnetic resonance imaging revealing a faint T2 hyperintense intramedullary signal at the T6-T8 levels (arrow heads) with no residual epidural hematoma and no remaining spinal cord compression. A: Sagittal T1-weighted image; B: Sagittal T2-weighted image.

epidural space are extremely rare[1-5]. Surgical excision is the treatment of choice, and recurrence rates as high as 15% have been reported[11].

SSEH is a rare, but disabling or even fatal, condition. The annual incidence of SSEH has been estimated at 0.1 per 100000[12]. The male/female ratio is 1.5:1, and most cases occur between the ages of 50 and 80 years[13]. The etiology is unknown, but predisposing factors can include hypertension, anticoagulant therapy, increased venous pressure (straining, sneezing, lifting, or whooping cough), pregnancy or labor, and vascular malformation[14]. The causative hematomas most frequently occur at the lower cervical and thoracolumbar spinal levels in adults. Early diagnosis and prompt

management improve the prognosis and outcome, but SSEH still remains a clinical challenge. Physicians encountering spinal soft tissue tumors should understand the typical symptoms and should consider SSEH as one of the differential diagnoses.

Currently, MRI is the main investigative tool for spinal soft tissue tumors, and it allows a prompt diagnosis of a spinal epidural hematoma. On MRI, the hematoma appears as an isointense area on the spinal cord on T1-weighted images obtained within the first 24 h. After 36 h, it appears as a hyperintense or mixed signal intensity area on the spinal cord on T2-weighted images[15]. In addition to specific signal changes, contrast enhancement pattern and morphological findings on MR images can differentiate acute SSEH from spinal epidural neoplastic mass[16]. Pan *et al*[17] reported that, from the clinical viewpoint, most patients with spinal epidural hematomas undergo some degree of irreversible cord injury by the time the hematoma is resorbed. The prognosis of SSEH correlates with the size and level of hematoma, preoperative neurological status of the patient, and the time interval between the onset of symptoms and surgery[18,19]. Therefore, immediate surgical decompression remains the primary consideration in the management of patients with progressive neurological deficits.

CONCLUSION

Acute spontaneous spinal epidural hematoma caused by an intraspinal lymphangioma is a very rare occurrence. Neurosurgeons should consider the possibility of SSEH when their patients show neurologic symptoms related to the spinal cord or root compression. In the event of a thoracic epidural hematoma, prompt surgical intervention is mandatory in order to achieve neurologic recovery.

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