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CASE REPORT

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An unusual case of ventral spontaneous thoracic epidural hematoma

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Key Clinical Message

Spontaneous spinal epidural hematoma (SSEH) rarely occurs. Without early diagnosis, SSEH can lead to the acute onset of neurologic deficits. We report the case of a 65-year-old male with diabetes mellitus who was admitted to our emergency department with a chief complaint of sharp and severe pain in the left scapula and behind the sternum. He was misdiagnosed with cardiovascular disease until the onset of progressive bilateral paraplegia and lower limb numbness. Magnetic resonance imaging revealed a ventral thoracic SSEH. Surgical treatment to remove epidural hematoma and laminectomy for decompression were performed. Except for urine retention, bilateral lower limb paraplegia and numbness were alleviated postoperatively. Due to the high risk of poor neurological outcomes without treatment or with delayed intervention, timely surgical evacuation of the hematoma and hemostasis are recommended to ensure favorable neurological outcomes.

KEYWORDS

case report, spinal epidural hematoma (SEH), spontaneous, thoracic, ventral

1 | INTRODUCTION

Spinal epidural hematoma (SEH) is defined as blood accumulation in the spinal epidural space, creating pressure on the nerve roots and/or spinal cord, sometimes leading to disabling neurological symptoms.[1] Spontaneous SEH (SSEH) occurs without any trauma, disease, or iatrogenic procedures.[2] Its symptoms and clinical signs progress very rapidly, and diagnosis is difficult in the emergency department. We report a patient with paraplegia caused by a ventral thoracic SSEH, characterized by the early onset of chest and back pain.

2 | CASE REPORT

A 65-year-old man with diabetes was admitted to our emergency department by ambulance at 8:30 a.m., presenting with a sudden onset of sharp and severe pain in the left scapula and behind the sternum. He denied any

history of trauma, spinal cord disease, or use of medications, such as anticoagulants and herbal medicines. He denied any systemic symptoms, including fever, palpitations, shortness of breath, nausea, vomiting, blurred vision, or headache.

On examination, the patient demonstrated pain. He exhibited normal muscle strength based on the Medical Research Council grading system. Since the electrocardiogram (ECG) showed an inverted P-wave in the aVR, an intraventricular block was considered (Figure 1). Laboratory studies showed a troponin level (TNL) < 0.1 ng/mL, myoglobin level (MYO) of 13.53 ng/mL, and a creatine kinase-MB level (CK-MB) of 0.65 ng/mL. His prothrombin time and international normalized ratio were within normal limits. Computed tomography angiography (CTA) of the heart and abdominal aorta ruled out aortic dissection and a myocardial infarction.

Approximately 2 h after admission, the patient experienced sudden bilateral paraplegia and lower limb numbness, which was described as similar to the sensation after electric shock. The pain in the left scapula and behind the sternum gradually subsided. His lower limbs were paralyzed, with a muscle strength rating of 0/5. Magnetic resonance imaging (MRI) was performed 30 min after paraplegia onset. Urgent thoracic MRI showed a heterogeneous collection within the T4-to-T5 ventral epidural space. The lesion showed iso- or hypointense signal on T1WI weighted image (Figure 2) and mixed iso- and hypo-intense signals on T2WI (Figures 3 and 4), with significant spinal cord compression, causing it to shift dorsally to the left. Computed tomography (CT) of the thoracic spine revealed that the lesion was isointense in the T4-to-T5- spinal canal with unclear spinal cord boundary (Figure 5).

3 | DIFFERENTIAL DIAGNOSIS AND TREATMENT

Given the acute presentation and MRI findings, thoracic SSEH with spinal cord compression was suspected, and emergent surgical intervention was considered. The blood clot was located ventrally to the lateral right of the spinal cord, without obvious adhesion between the dura mater and posterior longitudinal ligament, of which the superficial veins were slightly dilated. No vascular malformations were detected intraoperatively. MRI demonstrated hematoma clearance and resolution of spinal cord compression at 24 h postoperatively (Figure 6). Postoperative pathological analysis revealed a blood clot (Figure 7). Immunohistochemistry showed a phenotype of SMA(−) S-100(−) CD34 (−) with lower Ki-67 labeling index (LI) expression.

4 | OUTCOME AND FOLLOW-UP

The patient's symptoms began to improve on the second day postoperatively. Within 1 week after surgery, the lower limb muscle strength increased (left: 2/5, right: 4/5). Hyperbaric oxygen (HBO) therapy and limb exercises were prescribed for rehabilitation. The patient was

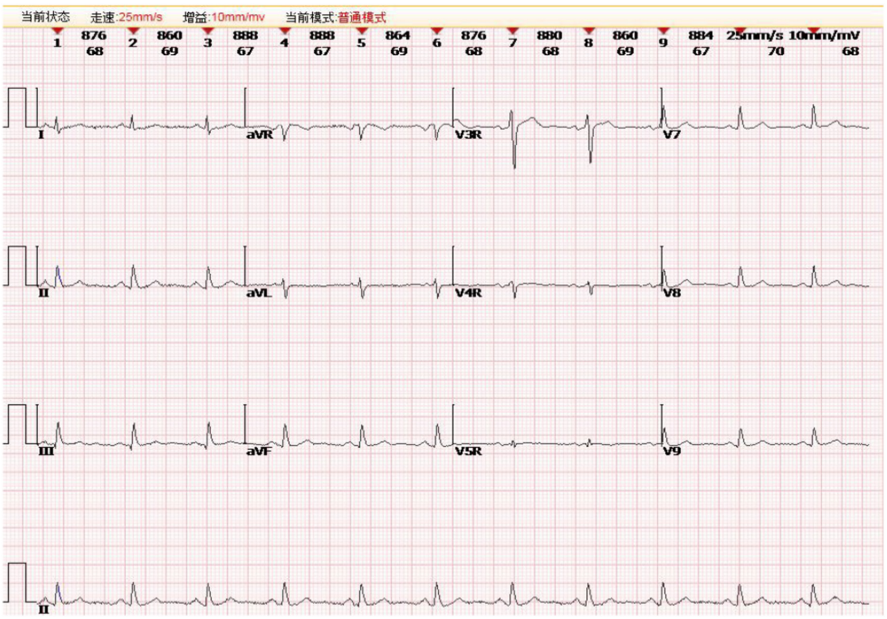

FIGURE 1 Electrocardiography revealed an intraventricular block.

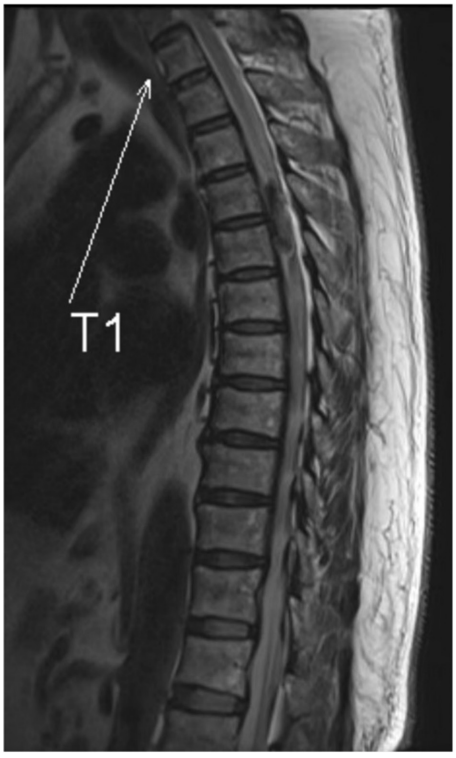

FIGURE 2 Thoracic spinal magnetic resonance imaging (MRI) at T4-5 level in sagittal T1-weighted sequence showed mixed iso- or hypo-intense signal, causing significant mass effect and compression on the spinal cord.

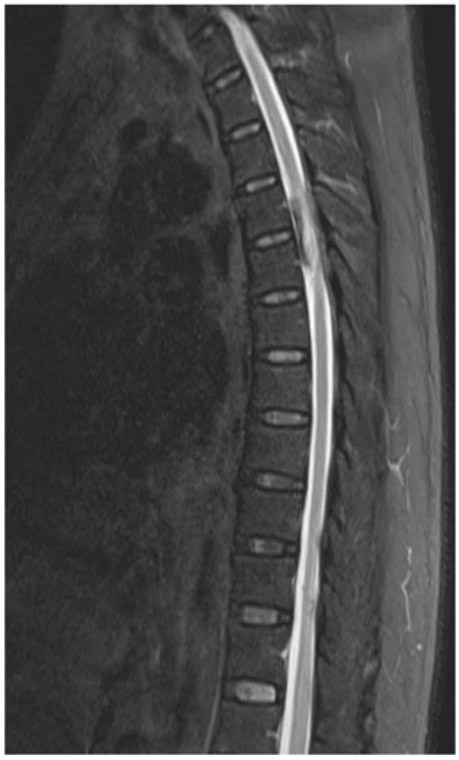

FIGURE 3 Sagittal T2-weighted sequence showed mixed hyper- and hypo-intense signal.

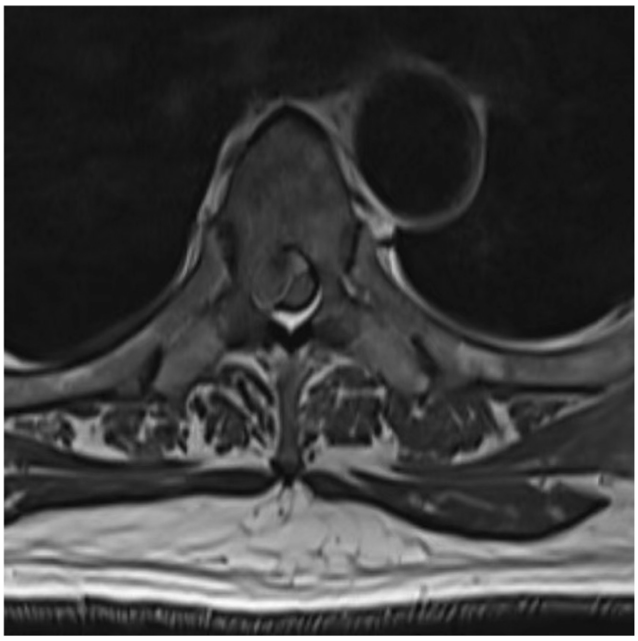

FIGURE 4 Axial T2-weighted sequence showed mixed signal.

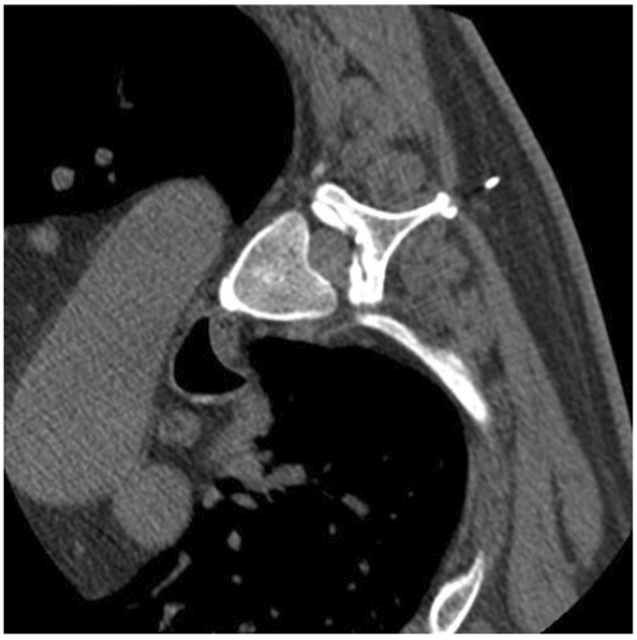

FIGURE 5 Computed tomography (CT) of the thoracic spine revealed the lesion was isointense in the intraspinal canal from T4 to T5 with unclear spinal cord boundary. The white spot on the spinous process was the anchor.

discharged on the 25th day postoperatively, with a nearly normal lower limb muscle strength (left: 4/5; right: 5/5). The patient had returned to a near-baseline condition at two-month follow-up with the only deficit being urine retention.

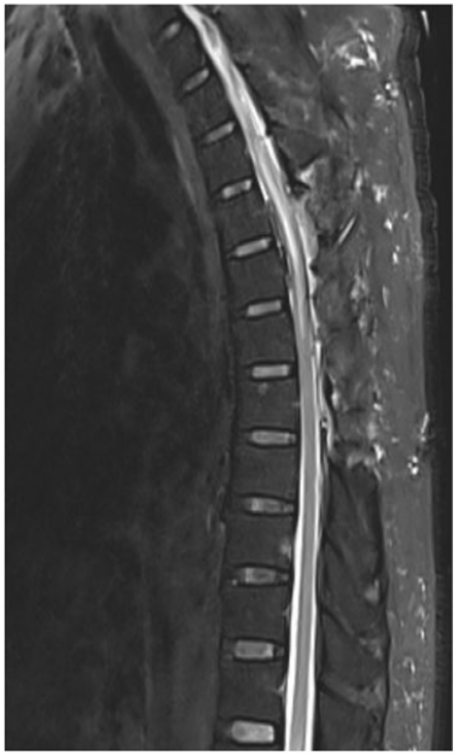

FIGURE 6 After decompression with hematoma evacuation and laminectomy, thoracic spinal MRI at T4-5 level with sagittal T2-weighted sequence showed a complete clearance of hematoma with normal ventral and dorsal epidural space and resolution of spinal cord compression.

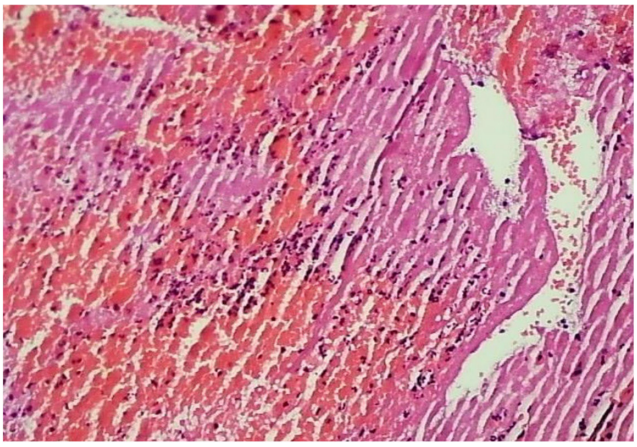

FIGURE 7 Postoperative pathological analysis revealed a blood clot.

5 | DISCUSSION

SSEH is rare, with an annual incidence of approximately 1/1,000,000 and a 1.4:1 male-to-female ratio.[3,4] Most spinal hematomas are found at the dorsal or lateral side region.[5] Ventral SSEH is rare, with only four reports to date.[6] As such, the present case is rare.

SSEH exhibits the highest prevalence around the C6 and T12 vertebral levels. These hematomas typically measure approximately 3.6 vertebral levels in length and are situated dorsally to the spinal cord.[7]

Bleeding is widely considered to be venous, since spinal epidural veins have no valves. SSEH is caused by a sharp increase in intra-abdominal or intrathoracic pressure, which is subsequently transferred to the spinal vessels and causes their rupture.[8,9] Since the dural sac is firmly attached to the posterior longitudinal ligament, anterior hematoma is unlikely to be explained by the posterior venous plexus theory of SSEH.[10,11] The ventral hematoma may be the nerve root artery. If symptoms progress rapidly with neurological decline, the cause is generally considered arterial. Otherwise, SSEH origin is likely to be venous.[12–14] In our case, the disease progressed rapidly in two stages. Severe pain caused agitation and increased thoracic and abdominal pressures. Venous bleeding was also detected intraoperatively. The posterior longitudinal ligament was not tightly adherent to the dura mater, which created conditions for a rapid increase and accumulation of venous bleeding. The hematoma is observed to be confined within two distinct levels, which serves as an indicator of limited blood vessel pressure. Therefore, ventral SSEH may also be of venous origin.

The primary risk factors for SSEH are coagulation disorders, either congenital or iatrogenic.[9,15,16] Diabetes, a recognized cause of accelerated atherosclerosis that affects the microvasculature, has not been reported as an SSEH risk factor. Since no other known risk factors were noted in our patient, the relationship between diabetes and SSEH requires further investigation. In the analysis of pathological specimens, the hematoma components allows us to preliminarily exclude the likelihood of bleeding stemming from neoplastic processes. However, to thoroughly eliminate the potential contribution of vascular malformations and alternative etiologies, confirmatory diagnostic modalities such as myelography or TRICKS MRA (Time Resolved Contrast Enhanced MR Angiography) are warranted. These modalities are based on clinical symptoms and findings from routine magnetic resonance imaging, providing a robust diagnostic framework for suspected spinal cord vascular malformations.[17]

SSEH is clinically characterized by the sudden onset of limb weakness and severe pain, which is usually radicular and radiates to the extremities. Within hours or days, due to spinal cord compression, these symptoms may cause loss of movement and sensation in various degrees, and SSEH diagnosis is difficult before the onset of neurologic deficit.[10]

Misdiagnosis of SSEH is common due to its low incidence.[18] Spinal MRI is the first choice for SSEH

diagnosis and also allows evaluation of spinal cord compression.[19,20] SSEH yields an isointense signal change on T1-weighted images within the first 24 h after bleeding and a hyperintense signal change on T2-weighted images after 24 h.[21] T2 hyperintensity in the spinal cord within 24 h signals poor clinical outcomes, because it identifies myelomalacia and radiographic spinal cord injury.[22] In most SSEH cases, sufficient findings could be provided by CT, in which hemorrhage is considered a hyperdense epidural mass.[23]

Our patient's CT and MRI findings differed from those in the literature, with mixed signals in the lesions. This made diagnosis difficult with imaging findings and required assessment of clinical manifestations.

Surgery has been the preferred definitive treatment of SSEH, and the standard operative procedure is decompressive laminectomy with hematoma evacuation.[2] Literature evidence suggests that conservative treatment should be considered under the following three conditions: (1) mild neurological symptoms (AIS grade D-E); (2) dorsolateral mass without extensive spinal cord compression and suspected venous plexus origin; and (3) spontaneous neurological recovery within 24 h.

A good functional outcome in SEH patients depends on three main factors: (1) the severity of the neurological lesion preoperatively; (2) how quickly the SEH is evacuated; (3) early postoperative rehabilitation. In patients with either complete or incomplete spinal cord lesions, recovery to the point of no/minimal neurological deficit is probable if surgical decompression is performed within 12 h.[24,25] In several experimental studies, HBO therapy has exhibited promising neuroprotective effects, which are manifested in the following aspects: (1) decreasing apoptosis through various mediators; (2) reducing oxidative stress and lipid peroxidation; (3) promoting angiogenesis; (4) mitigating spinal cord edema; and (5) enhancing autophagy.[26,27] In our case, the patient benefited from timely surgery and early HBO therapy.

Since thoracic SSEH is rare, atypical symptoms of this condition, such as chest pain, deserve clinical attention. In patients with pain and lower limb paralysis, Myelography or TRICKS MRA(time-resolved contrast-enhanced magnetic resonance angiography) is an effective test to rule out spinal cord vascular malformation causing SSEH. When the imaging diagnosis of SSEH is difficult, the risk of poor neurological outcomes without treatment is high. While decompressive laminectomy combined with surgical exploration is a reasonable treatment option, delayed intervention can be catastrophic.

AUTHOR CONTRIBUTIONS

Junge Zhou: Conceptualization; data curation; investigation; resources; writing – review and editing.

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None.

CONFLICT OF INTEREST STATEMENT

None.

DATA AVAILABILITY STATEMENT

The data used to support the findings of this study have been included in this article.

ETHICS STATEMENT

This research was approved by the ethics committee of Wuhan Brain Hospital.

CONSENT

Written informed consent was obtained from the patient to publish this report in accordance with the journal's patient consent policy.

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