Clinical Case Report

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Spontaneous spinal epidural hematoma mimicking transient ischemic attack

A case report

Chuqiao Li, MS[a,c], Rui He, BS[a], Xiaoqiang Li, MS[b], Yulan Zhong, MS[a], Li Ling, MD, PhD[c], Fangming Li, MD, PhD[c,*]

Abstract

Rationale: Spontaneous spinal epidural hematoma (SSEH) is a rare but highly disabling neurological emergency. The initial presentations are variable. Most patients of SSEH present with paraplegia or tetraplegia clinically, but recurrent hemiparesis with complete spontaneous recovery, mimicking transient ischemic attack (TIA), is a very rare initial presentation of SSEH.

Patient concerns: A 71-year-old female presented to the emergency department with 2 episodes of transient right hemiparesis in 5 hours. Two days later, above symptom reappeared and progressed to quadriplegia, dyspnea, and uroschesis quickly. The neurological examination showed tetraplegia and hypalgesia below the C2 level, but neither facial palsy nor aphasia was found.

Diagnosis: The patient was initially misdiagnosed as TIA and treated with antiplatelet therapy. But during the hospital day, the cervical magnetic resonance imaging showed a dorsal epidural hematoma extending from C2 to C6 level and she was diagnosed as SSEH.

Interventions: She underwent surgical decompression and hematoma removal 1 week later.

Outcomes: One week after operation, the sensory deficit above C6 level improved, but there was no improvement in her muscle strength and dyspnea. Unfortunately, she died 1 month later.

Lessons: Our case highlights recurrent hemiparesis with complete spontaneous recovery mimicking TIA is a rare initial presentation of SSEH. It is important to perform careful clinical assessments and neuroimaging investigations for correct diagnosis. Neck pain and hemiparesis sparing cranial nerve are important signs for distinction of SSEH from acute ischemic cerebrovascular diseases.

Abbreviations: MRI = magnetic resonance imaging, SSEH = spontaneous spinal epidural hematoma, TIA = transient ischemic attack.

Keywords: hemiparesis, spontaneous spinal epidural hematoma, transient ischemic attack

1. Introduction

Spontaneous spinal epidural hematoma (SSEH) is a rare but highly disabling neurological emergency, which accounts for <1% of all spinal epidural space-occupying lesions.[1,2] The annual incidence of SSEH was estimated to be 0.1 per 100,000 patients.[3] In SSEH, the "spontaneous" refers to atraumatic etiology and lack of iatrogenic procedure. Its etiology is related to vascular malformation, coagulopathy, anticoagulants, tumor, hypertension, and pregnancy. The classic manifestations of SSEH are sudden onset of back or neck pain followed by rapidly progressive spinal cord compression syndrome. But the initial presentations of SSEH are often miscellaneous and atypical. Recurrent hemiparesis with complete spontaneous recovery, mimicking transient ischemic attack (TIA), is a very rare initial presentation of SSEH. Here, we report a rare case of SSEH who initially presented with recurrent hemiparesis and was misdiagnosed as TIA.

2. Case report

A 71-year-old woman presented with sudden neck pain and weakness in her right upper and lower limbs while she was having a bath at home. Her symptoms resolved spontaneously within 10 minutes. After half past 4 hours later, she had the same onset of right hemiparesis and had full recovery within 15 minutes, and then was immediately admitted to our emergency department. The patient had a 30 years history of hypertension without well-control. She had no previous history of taking any antiplatelet or anticoagulant drugs. There was no recent history of head and spinal trauma or surgery.

The initial blood pressure was 210/90 mm Hg. On neurological examination, the patient had a normal cranial nervous system

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CL and RH contributed equally to this work.

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[a] Department of Neurology, The Affiliated Guangzhou Red Cross Hospital, Jinan University, Guangzhou, [b] Department of Neurology, Xiaolan Hospital of Southern Medical University, Zhongshan, [c] Department of Neurology, Shenzhen Hospital, Southern Medical University, Shenzhen, Guangdong, China.

[*] Correspondence: Fangming Li, Department of Neurology, Shenzhen Hospital, Southern Medical University, N0.1333 Xinhu Road, Shenzhen, Guangdong 518110, China (e-mail: lifly050413@163.com).

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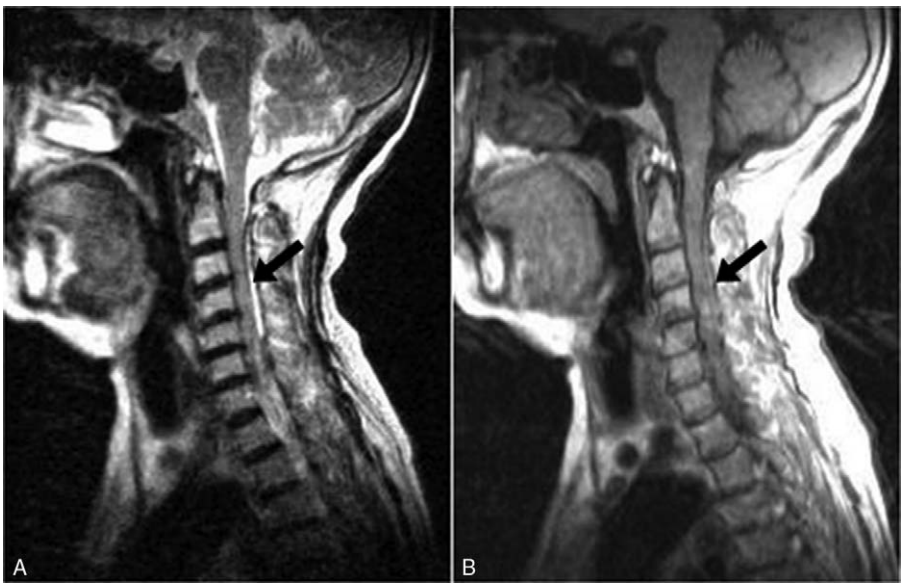

Figure 1. Cervical magnetic resonance imaging (MRI; sagittal) showed a longitudinal dorsal epidural hematoma (A: T2-weighted, B: T1-weighted) extending from C2 to C6 level.

and sensory system. Her muscle strength was grade 5/5 in bilateral upper and lower limbs, but the right knee reflex was hyperactive and Hoffman's sign was positive. Laboratory investigations showed that a complete blood cell count, prothrombin time, and partial thromboplastin time were within normal limits. An emergency brain computed tomography and cervical x-ray scans were unremarkable. A clinical diagnosis of TIA was considered. Then the patient was hospitalized in the neurological wards and treated with antiplatelet therapy (100 mg aspirin and 75 mg clopidogrel, once daily). She did not complain any other symptoms except a mild neck pain in the following 2 days after admission.

On the third hospital day, the brain magnetic resonance imaging (MRI) and cervical MRI were performed. The brain MRI showed no signs of acute cerebral infarction. The cervical MRI revealed a spinal epidural hematoma located in the posterior spinal epidural space extending from the C2 to C6 spinal vertebral level. The hematoma was isointense on T1-weighted images and hyperintense on T2-weighted images (Fig. 1). Just after returning to the wards, the patient recurred the right hemiplegia (muscle strength was grade 3/5 in right upper and lower limbs), and developed rapidly progressive quadriplegia, loss of pain and temperature sensation, dyspnea, and uroschesis. Neurological examination revealed tetraplegia (the muscle strength was grade 2/5 in left limbs and 1/5 in right limbs) and loss of pain below the C2 level bilaterally. She had neither aphasia nor abnormal cranial nerve signs. Given her progressive symptoms and imaging findings, the diagnosis of spinal cord compression was considered. The patient was transferred to the intensive care unit immediately and treated with trachea cannula and ventilator-assisted ventilation. Her family members did not consent to emergency surgical decompression and the patient became worse in the following days. After a week later, her family members finally consented to the surgical decompression. The findings of operation confirmed a hematoma arising on the dorsal spinal epidural surface and extended from the C2 to C6 spinal vertebral level, and the right was more serious.

One week after operation, the sensory deficit above C6 level improved, but there was no improvement in her muscle strength and dyspnea. Unfortunately, she died 1 month later.

3. Discussion

SSEH is a rare clinical emergency which was first described by Jackson in 1869.[4] Holtas et al[3] reported that the annual incidence of SSEH was approximately 0.1 patient per 100,000 patients. But in the last years, the number of diagnosed and reported cases of SSEH has been increased due to the progress of neuroradiological investigations and neurosurgery. Patients with SSEH usually present with sudden onset of severe back or neck pain followed by rapidly progressive symptoms and signs of nerve root or spinal cord compression. The clinical symptom severity is significantly correlated with the rate of bleeding, the extent of hematoma, and the length of time of onset. However, the initial manifestations of SSEH are sometimes atypical, even mimicking an onset of ischemic stroke. Among the initial presentations of patients with SSEH, hemiparesis is a less common symptom compared with other motor deficits such as paraplegia or tetraplegia.[5,6] But with the rapid development of MRI technology, there are increasing cases of SSEH who presented with hemiparesis initially have been reported in the last years. In clinical practice, when patients of SSEH presented with sudden onset of hemiparesis, even they complained of back pain or neck pain, they are often been suspected to have an acute cerebral infarction during the initial assessment.[7] Moreover, hemiparesis with complete spontaneous recovery, mimicking TIA, is a rare initial symptom of SSEH. Only a few patients with complete spontaneous recovery due to SSEH have been reported in the last decades.[8,9] For example, Hernandez et al[8] reported that a patient had 3 relapsing paraplegia and complete spontaneous recovery in 5 hours, and myelography and computed tomography confirmed a posterior epidural hematoma extending from T11 to T12 level. The initial presentation of our patient was 2 episodes of right hemiparesis with complete spontaneous recovery within 5 hours. The present patient

was an old woman with a previous history of uncontrolled hypertension, so she is easy to be misdiagnosed as having a TIA at the early stage and consequently administrated with antiplatelet drugs. This patient did complain of neck pain on admission, but this symptom did not be attached great importance by physicians due to the findings of cervical x-ray scans was unremarkable. Similar to the previous reported cases of SSEH with complete spontaneous recovery, the length of spinal epidural hematomas was not long in our patient. In addition, the cervical MRI of our patient showed that the shape of spinal epidural hematomas was cylindrical, which might have less pressure on the spinal cord. These may be the reasons why our patient presented with less severely neurological deficits and had complete spontaneous recovery at the early stage. In addition, we think that at the beginning of the stage, the hematoma may spread along spinal epidural space containing fat, areolar tissue, and vascular network, which would result in the subsequent decompression of internal pressure. Hence, our patient had the manifestation of recurrent hemiparesis with complete spontaneous recovery mimicking TIA. We emphasize that it is important to collect a comprehensive case history and perform neurological examination in the course of diagnosis. Radicular pain should not be ignored as an important sign of SSEH. When patients have sudden neck pain with neurological deficits, it is highly suggestive of a possible cervical spinal cord hemorrhagic disease, and SSEH should be considered as a common stroke mimic.

So far, the actual source of bleeding has not been well defined in patients with SSEH. Most authors support the venous origin hypothesis due to the anatomical characteristics of the posterior internal vertebral venous plexus and the most common location was the posterior of the thoracic region in the spinal canal.[1,10,11] They believed that it is prone to resulting in veins rupture and bleeding when patients had a suddenly increasing pressure effect from the thoracic or abdominal cavity, especially in the older patients or the patients with slow progression of neurological decline. The present patient had the onset when she was having a bath, whereas many patients of SSEH have been reported to be initially triggered by actions that suddenly increase venous pressure.[12,13] On the contrary, some authors support the arterial origin of bleeding, especially when the hematomas was located in the cervical region.[14] They think that the intrathecal pressure is higher than the venous pressure at the cervical level, which would prevent bleeding from veins in the epidural space. Considering that the hematoma of our patient was located at the cervical region, our patient had rapidly deteriorating neurological deficit after admission, as well as she had an uncontrolled hypertensive condition, we speculated that the hematoma may be of arterial origin in the present patient.

The precise etiology remains unclear. Increasing age, hypertension, anticoagulants, thrombolytics, antiplatelet agents, vascular malformation, and systemic diseases have been considered as possible predisposing factors in some patients with SSEH.[1,12,15–18] However, there are 40% to 60% of cases could not find any underlying cause.[19,20] SSEH could occur at any age, but most of the patients are in their fourth or fifth decades of life. A recent individual patient data meta-analysis revealed the median age is 58 years.[16] Hypertension has been observed in a range from 3% to 21.4% of patients with SSEH, but whether hypertension is a risk factor of SSEH or not is still controversial.[1,16] Evidences from current meta-analysis could not suggest the relationship between hypertension and the development of SSEH.[16] We thought that old age and uncontrolled hypertension may be risk factors of our case. Moreover, this patient was treated with the antiplatelet drugs due to her initial manifestation mimicking TIA, which may result in the rapid development of hematoma and poor outcome. In clinical practice, physicians tend to consider TIA as the first diagnosis for patients with acute-episodic neurological disorders. If the patient complained of persistent neck or back pain and recurred or fluctuating hemiparesis, neck or back pain should not be overlooked as an important sign for the diagnosis of SSEH, especially in the patients without cranial nerve signs.

Spinal MRI is the preferred diagnostic tool for SSEH. Spinal MRI can show the segmental and range of epidural hematoma, and the extent of spinal cord compression. However, the super acute or acute hemorrhage has atypical MR manifestations. The hematoma shows isointense on T1-weighted and hyperintense on T2-weighted images. It should combine the onset time to analyze the MR signs of hematoma. If physicians' lack of the realization of early imaging changes, it can easily lead to a misdiagnosis. For subactue hemorrhage, it shows characteristic hyperintense on T1-weighted and low signal intensity on T2-weighted images. In our patient, the findings of spinal MRI were consistent with the signs of acute hemorrhage.

Emergency surgical spinal decompression is the main treatment of SSEH, which should be performed within 12 hours and not later than 36 hours for a favorable outcome.[12,21] Conservative management can also be considered in those SSEH patients with minimal neurological deficits or who have rapidly improving neurological function.[22] Delayed surgical decompression was performed in our patients, and her condition deteriorated due to hematoma expansion.

Although our patient presented with mild and recurrent hemiparesis with complete spontaneous recovery at the initial stage, she developed rapidly progressive neurological deficits within a short time after admission. We considered that the causes of her rapid exacerbation were multifactorial, including old age, uncontrolled hypertension, the location of hematoma in the cervical area, the antiplatelet therapy, and delayed surgical spinal decompression. Among above factors, the use of antiplatelet agent may be the major cause of hematoma expansion and poor outcome.

In conclusion, our case highlights recurrent hemiparesis with complete spontaneous recovery mimicking TIA is a rare initial presentation of SSEH. It is important to perform careful clinical assessments and neuroimaging investigations for diagnosis. Neck pain and hemiparesis sparing cranial nerve are important signs for distinction of SSEH from acute ischemic cerebrovascular diseases.

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