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CASE REPORT

Spontaneous spinal epidural haematoma

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KEYWORDS
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Summary Spontaneous spinal epidural haematoma (SSEH) is a rare clinical condition with unknown aetiology. Prompt diagnosis and treatment are paramount because of the risk of permanent neurological deficits without appropriate intervention. We described a case of a 48-year-old man presenting with complete quadriplegia and hypoesthesia. Magnetic resonance imaging revealed cervical cord compression due to a haematoma posterior to the spinal cord. Surgical decompression and evacuation of the haematoma was performed within 12 hours after admission to the authors' hospital. Both the patient's motor and sensory functions recovered soon after the operation. Early surgical decompression for SSEH with neurologic impairment is therefore recommended for the recovery of this patient and also serves as a relevant reference for orthopaedic clinics.
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Introduction

Spontaneous spinal epidural haematoma (SSEH) is a rare cause of spinal cord compression that requires early diagnosis and prompt surgical intervention [1]. The definition and pathophysiology of this disease remains controversial. The underlying causes include hypertension, vascular malformation, neoplasia, and anticoagulant or antiplatelet drug usage [2]. Most SSEH patients present with acute neck pain followed by radicular pain in the extremities [3,4]. Progressive sensory loss or motor weakness of limbs can be observed below the compressed spinal cord level. Here, we report a case of spontaneous cervical epidural haematoma presenting with complete quadriplegia and hypoesthesia which was treated by surgical decompression through hemilaminectomy.

Case report

A 48-year-old male was admitted to the emergency room (ER) with a main complaint of sudden severe neck pain for the past 12 hours. This patient developed mild neck pain and upper limb weakness when he woke up in the morning. His symptoms were not relieved by rest, and even worsened. About 6 hours after the initial attack, he went to the

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local community hospital. He was diagnosed with cervical spondylosis and was treated using infrared therapy for about 2 hours. However, his symptoms continued to aggravate with general weakness and numbness. He was then transferred to the authors' hospital under the impression of acute cerebral vascular accident. He was not on any anticoagulant medication. There was no history of recent trauma. His past medical history and family history were unremarkable.

On physical examination, the patient was afebrile with a blood pressure of 128/74 mmHg. He was conscious, orientated, and cooperative. All cranial nerves were intact. Neurological examination revealed the muscle power exhibited grade 0—1/5 in both upper and lower extremities (Table 1). Impaired pin-prick sensation below C3 level was noted. All reflexes were absent. Acute urinary retention developed later and Foley catheter was indwelled. Laboratory findings including platelet count and coagulation profiles were all within normal range. Under the impression of cerebral vascular accident, both brain computed tomography (CT) and magnetic resonance imaging (MRI) were performed, while both results did not show any evidence of cerebral infarction. However, cervical MRI showed a longitudinal epidural haematoma ranging from C2 to C6, which had clearly compressed and displaced the cervical spinal cord (Figs. 1—3). Based on these findings, a final diagnosis of SSEH was made. Pulse steroid therapy was administered because of spinal cord injury. The patient underwent prompt surgical decompression and evacuation of the haematoma by hemi-laminectomy. Postoperation histopathological examination was consistent with haematoma features without evidence of vessel abnormality or neoplasm (Fig. 4). Both motor and sensory functions recovered remarkably and he regained his ability to walk 7 days later. He was discharged from hospital 2 weeks after the operation with normal cervical MRI examination (Fig. 5).

Table 1 American Spinal Injury Association evaluation.

Muscles involved	Preoperation	Postoperation (1 wk later)
Deltoid	0	3—4
Elbow flexors	1	4
Elbow extensors	1	4
Wrist extension	1	4
Finger flexion	0	4
Finger extension	0	4
Knee extension	0	4
Knee flexion	0	4
Ankle dorsiflexion	0	4
Ankle plantar-flexion	0	4

Absent = 0; trace = 1, visible or palpable contraction; poor = 2, active movement through range of motion with gravity eliminated; fair = 3, active movement through range of motion against gravity; good = 4, active movement through range of motion against resistance; normal = 5; NT = not testable.

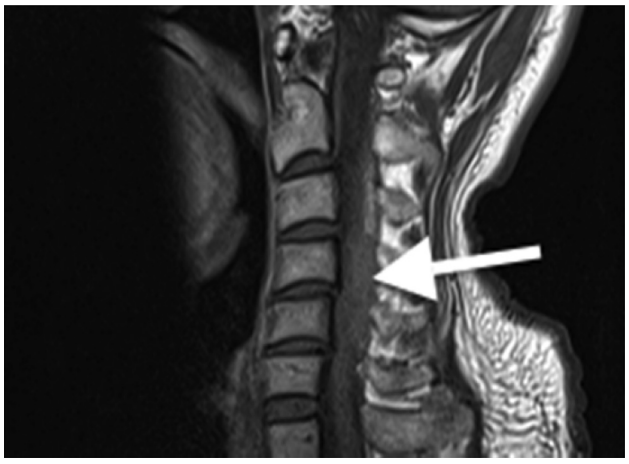

Figure 1 T1-weighted sagittal image shows isointense signal epidural haematoma of the cervical spine and compression of cervical spinal cord.

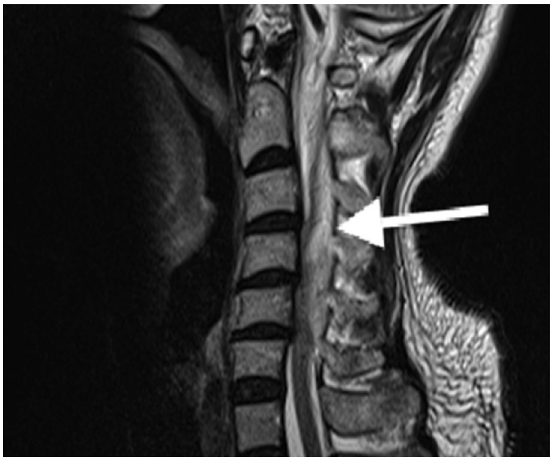

Figure 2 T2-weighted sagittal image shows a longitudinal hyperintense epidural haematoma ranging from C2 to C6.

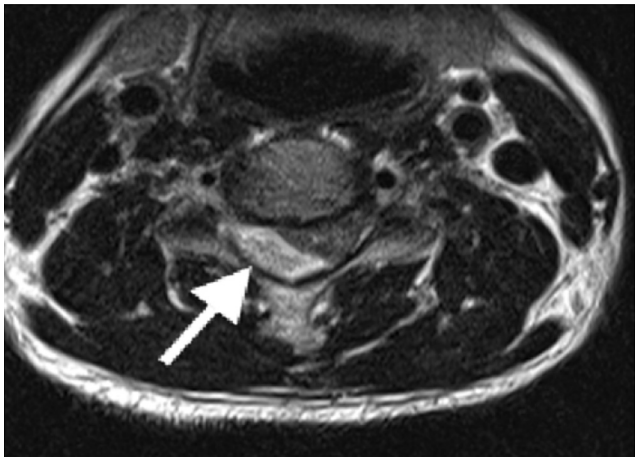

Figure 3 T2-weighted axial image shows an ovoid high-signal intensity epidural haematoma in the right postero-lateral side with spinal cord compression.

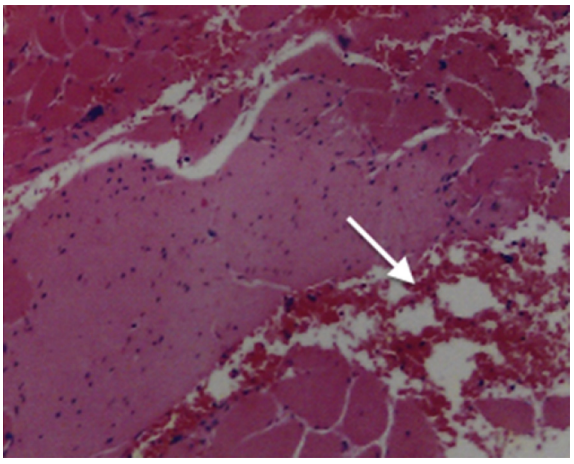

Figure 4 Histopathologic examination of the surgical specimen showing haemorrhagic material (haematoxylin and eosin staining, white arrow).

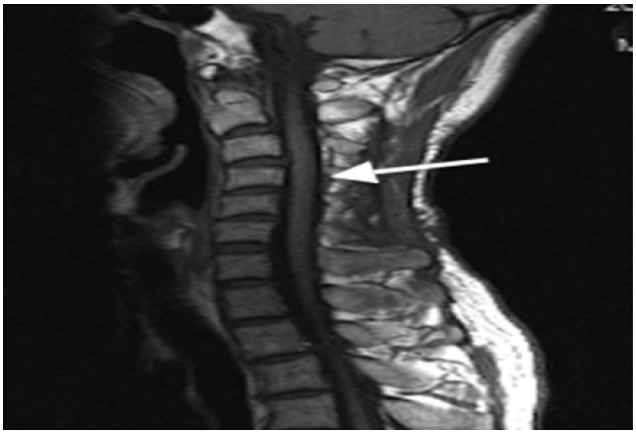

Figure 5 T1-weighted sagittal magnetic resonance image showed that the haematoma had disappeared following hemilaminectomy treatment 1 week after onset.

Discussion

SSEH is a rare clinical entity, which represents 0.3—0.9% of all epidural space-occupying lesions [5]. Jackson [6] first described SSEH in a 14-year-old patient in 1896, and Bain [7] reported the first surgically-treated case in 1897. The incidence of SSEH is estimated to occur in every.one patient per 100,000 individuals [8]. Males had a higher incidence than females (1.4:1) [9]. Up to now, there is still no agreement on the definition. Some authors include haematomas secondary to vascular malformation, hypertension, coagulopathy, and tumours, while others support that only haematomas with idiopathic origin can be labelled as spontaneous [10].

It is still not clear whether the aetiology of spontaneous epidural haematoma is arterial or venous. The most widely accepted hypothesis for the source of the haematoma is venous, because veins are valveless and walls are thin in the spinal epidural space. It is more susceptible to be damaged once there is a sudden change of the abdominal or thoracic pressure. Any Valsalva manoeuvre-like movements, such as coughing, sneezing, straining, defecation, micturition, vomiting, and coitus will lead to rupture of veins. Further study by Liao et al. [11] revealed that 54% of patients had a straining-associated event before the initial attack. However, since the pressure in the venous plexus is lower than intrathecal pressure, some authors claim epidural venous pressure is not strong enough to overcome this pressure gradient to produce a mass effect. The rapidity of the development of a cervical epidural haematoma is more likely to be arterial in origin. However, Beatty and Winston [12] investigated the arterial circulation in the epidural space. Their study showed that arteries in the epidural space were free anastomotic which connect with radicular arteries, and the most mobile segments, C6 and C7, were the most common sites of haemorrhage.

The most common initial manifestation of spontaneous cervical epidural haematoma is a sudden onset of severe neck pain with or without radicular symptoms. Without appropriate management, most cases will progress with motor and sensory deficits. Clinical symptoms vary depending on the location and the severity of compression. In our report, the initial presentation with acute right side weakness misled the ER physician to suspect a cerebral stroke. However, brain CT and magnetic resonance images did not show any significant changes. A complete neurological examination was done and all cranial nerves were intact which was not consistent with typical cerebral infarction signs. A cervical MRI was performed for the evaluation of myelopathy and it displayed a longitudinal cervical epidural haematoma ranging from C2 to C6 with a compression of spinal cord. It is a great challenge for an ER physician to distinguish SSEH from cerebral infarction in a short amount of time. Radiological data plays an important role in diagnosing SSEH. MRI is superior to CT and X-ray in defining the location, extent of the haematoma and the involvement of spinal cord. Many studies have demonstrated that it is isointense on T1-weighted images and heterogeneous or hyperintense on T2-weighted images in the first 24 hours.

Surgical decompression and haematoma evacuation through laminectomy remains the first choice of treatment. Giugno et al. [13] reported a case of a 75-year-old woman suffering from idiopathic spontaneous spinal epidural haematoma with involvement of the thoracolumbar spine that was rapidly diagnosed and successfully treated with a decompressive laminectomy. To the best of our knowledge, preoperative neurological deficits and the time interval between symptom onset and surgical decompression are two of the most important factors in determining the prognosis of SSEH. Compared with complete deficits, incomplete deficits have a better recovery. Liao et al. [11] reported that the recovery rate in incomplete deficit patients was 89% but only 37.5% in complete deficit patient at 1-year follow up. Shin et al. [14] showed that the *Japanese Orthopaedic Association* (JOA) score of patients who underwent surgery within 12 hours (84%) is better than those who underwent surgery from 12 hours to 24 hours (63.6%) after onset and those who underwent surgery more than 24 hours (46.7%) after initial attack. Also, in a clinical study conducted by Rajz et al. [15], they found that both the time interval and preoperative neurological status played

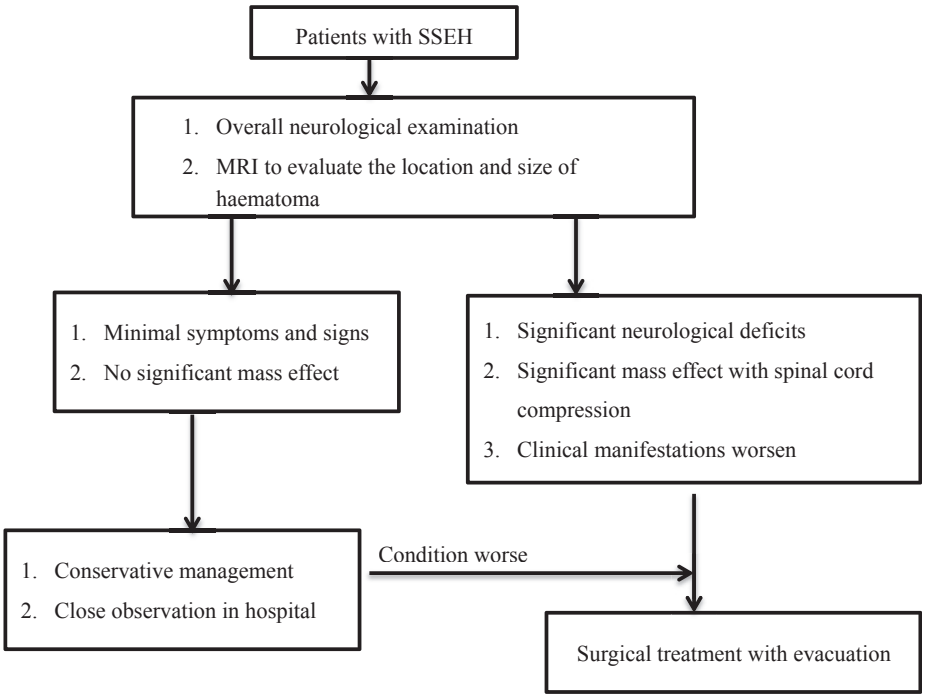

Figure 6 Treatment strategy for patients with spontaneous spinal epidural haematoma (SSEH).

important roles in the prognosis of patients with SSEH. However, their further analysis showed that the time interval from symptom onset to surgery appeared to have less impact than preoperative neurological status on the extent of recovery. There were some patients who were successfully treated through conservative management [16,17]. Further analysis revealed their signs and symptoms were mild and there was no significant mass effect. Nevertheless, close observation under an experienced doctor is necessary because of the poor outcomes of spinal cord injury without correct management. Hence, we strongly suggest overall evaluation of patients with SSEH as shown in Fig. 6. A study by Omori et al. [18] showed that pulse steroid administration accelerated the recovery of neurological deficits in a 70-year-old patient with SSEH after surgery. Steroids protect cells by reducing cellular oedema and suppressing the production of free radicals. In our report, a bolus dose of 30 mg/kg methylprednisolone was given over 15 minutes. With a 45-minute pause, a maintenance dose of 5.4 mg/kg/h was administered in 23 hours according to the National Acute Spinal Cord Injury Study II (NASCIS II) [19]. However, it is difficult to evaluate its function without a control group.

Conclusion

SSEH is an acute neurologic emergency that requires prompt recognition and treatment. The diagnosis should be suspected in the setting of acute and significant back or neck pain with an associated neurological deficit. SSEH is commonly misdiagnosed as a cerebrovascular accident. MRI plays an important role in distinguishing between these two different diseases. Rapid decompression is recommended for treating such kinds of emergent condition.

Conflicts of interest

The authors declare no conflicts of interest.

Acknowledgements

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