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CASE REPORT

Spontaneous Spinal Epidural Hematoma Due to Rupture of an Arteriovenous Fistula

Shang-Wun Jhang[1], Chien-Min Chen[1], Chun-Yuan Cheng[1], Han-Chung Lee[2]

[1]Department of Surgery, Division of Neurosurgery, Changhua Christian Hospital, Changhua, [2]Department of Neurosurgery, China Medical University Hospital, Taichung City, Taiwan, Republic of China

Spontaneous spinal epidural hematoma (SSEH) is a neurosurgical emergency that requires prompt diagnosis and treatment. We report a 24-year-old woman who presented with acute onset of paralysis in both lower limbs and sensory disturbance below the fourth-thoracic dermatome. Spinal magnetic resonance image (MRI) revealed an intraspinal, extradural mass is extending from the fifth to the seventh thoracic vertebrae with compression of the spinal cord. Laminectomy of the T5 to T7 vertebrae was performed 12 h after onset. During the procedure, an epidural hematoma with hypervascularization and an abnormal vascular network were observed grossly on the dorsal dural surface. Postoperative angiography and MRI revealed complete resolution of the hematoma and no evidence of residual vascular lesion in the intra- or extra-dural region. At 6-month follow-up, the patient had regained full muscle power and sensation in the lower limbs. There was no evidence of urinary or stool incontinence. The patient had a history of remaining seated for prolonged periods of time, which may have elevated the spinal venous return pressure, resulting in spontaneous hemorrhage due to rupture of the spinal epidural arteriovenous fistula. This case report shows that patients with SSEH can have excellent neurologic outcomes if the condition is treated early with decompressive laminectomy.

Key words: Spontaneous spinal epidural hematoma, spinal epidural arteriovenous fistula, rupture

INTRODUCTION

Spontaneous spinal epidural hematoma (SSEH) is a neurosurgical emergency that requires prompt diagnosis and treatment. Sudden onset of back or neck pain that radiates to corresponding dermatomes, followed by rapid sensorimotor paralysis due to hematoma-induced compression of the spinal cord or nerve roots is a well-documented and typical presentation.[1,2] Many etiologies of SSEH have been reported, such as anticoagulant therapy, hypertension, voiding, cough, and bleeding diathesis.[2] To the best of our knowledge, spinal epidural arteriovenous fistula (AVF) is a rare cause of SSEH. We reported a case of SSEH due to rupture of a spinal epidural AVF in a 24-year-old woman presenting with acute paraplegia.

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Coressponding Author: Dr. Han-Chung Lee, Department of Neurosurgery, China Medical University Hospital, No. 2, Yude Road, Taichung 40447, Taiwan, Republic of China.
Tel: +886-4-22052121; Fax: +886-4-22053425.
E-mail: 133393@cch.org.tw

CASE REPORT

A 24-year-old woman had intermittent sharp back pain radiating to the anterior chest wall, which often manifested after sitting for long periods of time since 6 years ago, presented to the emergency department with acute onset of progressive weakness in both legs and impaired sensation below the fourth thoracic dermatome. The patient did not have a history of trauma or underlying medical conditions. The results of laboratory studies were normal. Deep tendon reflexes of the extremities were hyperactive, and Babinski's signs were present bilaterally. Anal tone was loose, and there was no peri-anal sensation. Spinal magnetic resonance imaging (MRI) showed an intrathecal mass measuring 5.4 cm × 2 cm × 1.4 cm in size, extending from the posterior T5 to T7 levels and compressing the spinal cord. There was isointensity on T1-weighted images (WI) and heterogeneous hyperintensity on T2-WI with peripheral enhancement [Figure 1a]. We performed a laminectomy on the T5 to T7 vertebrae 12 h after onset. An epidural hematoma with hypervascularization and an abnormal vascular network was observed grossly on the dorsal dural surface [Figure 1c]. The epidural hematoma had no vessel connection with the spinal cord and did not penetrate the dura. Microscopically,

the adjacent vasculature was composed of variably ectatic and hyalinized veins. Abnormal muscularized arteries and fibrous tissue were noted histopathologically [Figure 2] and spinal epidural vascular malformations with hemorrhage were seen. Postoperative angiography and MRI revealed complete resolution of the hematoma and no evidence of residual vascular lesion in the intra- or extra-dural region [Figure 2]. At 6-month follow-up, the patient had regained full muscle power and sensation in the lower limbs, and there was no evidence of urinary or stool incontinence.

DISCUSSION

Spontaneous spinal epidural hematoma is a rare cause of acute spinal cord compression. In a study of 199 cases, Groen and van Alphen[3] reported only nine cases of SSEH associated with spinal epidural AVFs. In another study at one institution, no cases of SSEH due to rupture of spinal epidural AVFs were reported in a 17-year period.[4]

Spetzler *et al*.[5] proposed a modified classification system for spinal arteriovenous lesions based on specific anatomical and pathophysiological factors. The system is divided into arterovenous malformations and spinal epidural AVFs, which are further subdivided into extradural and intradural lesions. A direct connection between an extradural artery and vein leads to the development of a high-flow fistula and engorgement of the epidural venous system. The hypothesis is congestion of vessels followed by rupture of the spinal venous system. Beatty and Winston suggest that an arterial source of bleeding originating from the extensive network of epidural arteries is a better explanation for the precipitous neurological deterioration seen clinically.[1] Such a vascular anomaly might well explain the occasional occurrence of back and radicular pain without neurologic deficit that often precedes the final attack by months or years in some patients with SSEH.

Magnetic resonance imaging is the diagnostic method of choice for patients who present with spinal emergencies because it allows for the rapid evaluation of large parts of the vertebral column and spinal cord.[4,6] On T1-WI, SSEH usually displays an isointense signal to the spinal cord within 24 h, and displays a primary hyperintensity with hypointense foci on T2-WI.[4] Studies have shown that performing preoperative angiographic evaluation of patients with MRI-confirmed SSEH is not beneficial. Early recognition and surgical decompression provide the best chance for functional recovery, and the time lost doing additional diagnostic studies may have a negative effect on functional outcome.[7]

It is accepted that surgical evacuation of a hematoma, either by laminectomy or laminotomy, must be performed immediately after radiological confirmation of SSEH to prevent further progression of neurological deficits.[3,4] The risk of permanent neurological disability has been shown to be directly related to the rapidity with which paralysis develops, the force of compression, the duration of paralysis, and the severity of the neurological deficits prior to surgical decompression.[7] In a series comprising 34 patients reported in 2004,[4] patients who underwent surgery <12 h after onset of symptoms of neurological dysfunction had better functional results than patients who underwent surgery more than 12 h after onset of symptoms ($P < 0.005$). However, nonsurgical management may be suitable for patients whose neurological deficits improve rapidly.[8]

Our patient, a 24-year-old woman with a 6-year history of intermittent sharp back pain radiating to the anterior chest wall, may have had existing spinal epidural AVFs with steal phenomenon. The patient often spent prolonged periods

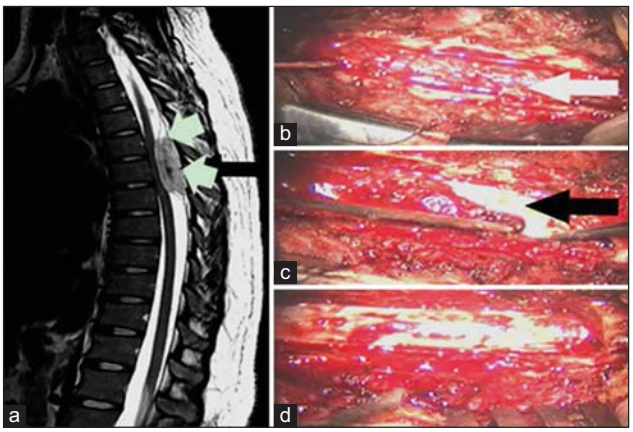

Figure 1. Preoperative magnetic resonance image of the thoracic spine shows an intrathecal mass, measuring 5.4 cm × 2 cm × 1.4 cm, extending from the posterior T5 to T7 levels and compressing the spinal cord. (a) There was isointensity on T1-weighted images (WI) and heterogeneous hyperintensity on T2-WI with peripheral enhancement. (b) Intraoperative photo shows a vascular network (white arrow) over the epidural surface. (c) The network is coagulated and separated from the dura matter (black arrow). (d) There was no obvious dural defect or vascular connection into the intradural space after the lesion was excised

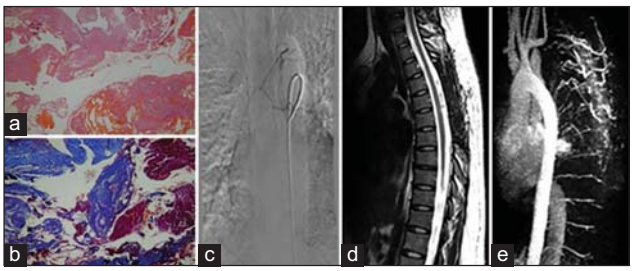

Figure 2. Arteriovenous malformations composed of abnormal muscularized arteries and dysplasic veins. (a) H and E, ×40. (b) Masson's trichrome stain ×40. (c) Postoperative conventional angiography. (d) Postoperative T1-weighted magnetic resonance (MR) image and (e) MR reconstructive angiography show no residual spinal epidural or intradural vascular lesion

of time in a seated position, which may have elevated the spinal venous return pressure and in turn, increased the risk of spinal spinal epidural AVFs rupture with spontaneous hemorrhage. Prompt diagnosis and emergency surgical decompression resulted in good neurological recovery in our patient.

DISCLOSURE

All authors declare that this study has no conflict of interest.

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