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Unusual presentation of a spontaneous spinal epidural haematoma

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Abstract

Spontaneous spinal epidural haematoma (SSEH) is a rare clinical entity that generally requires an urgent surgical evacuation. The combination of Brown-Séquard syndrome (BSS) and Horner's syndrome (HS) as the presenting symptoms of a traumatic spinal epidural haematoma is very unusual, but it has never been observed in cases of spontaneous haematoma.

We herein describe a case of SSEH presenting with simultaneous BSS and HS. The possibility of a conservative management in similar cases is discussed.

Key words: Spontaneous spinal epidural haematoma; Brown-Séquard syndrome; Horner's syndrome; conservative management.

Introduction

Spinal epidural haematoma (SEH) is a possible cause of cord compression. It occurs predominantly in patients who are anticoagulated, thrombocytopenic or have a bleeding diathesis or vascular lesions (Braun *et al.*, 2007).

SEH can be either post-traumatic, i.e. following lumbar puncture, epidural anaesthesia (Tekkok *et al.*, 1991) and spinal surgery, or spontaneous (SSEH) (Packer and Cummins, 1978).

The clinical presentation is usually characterized by a severe back pain with a radicular component followed by a rapidly progressive cord compression syndrome. Immediate decompressive laminectomy is generally indicated (Liao *et al.*, 2004), although cases of spontaneous resolution have been reported.

We describe an unusual presentation of a SSEH with simultaneous Brown-Séquard syndrome (BSS) and Horner's syndrome (HS) that happened to resolve spontaneously both clinically and radiologically.

Case report

A 52-year-old male without previous history of trauma, chiropractical treatment, coagulopathy or anticoagulant therapy, developed a sudden severe occipital and neck pain, with radiation to the shoulders. A few minutes later, a left hemiparesis (initially with the exclusion of the face) set in and quickly worsened. On admission the neurological examination also showed: left abnormal light touch and proprioception; absent pinprick sensation on the right side; normal reflexes; palpebral ptosis and miosis in the left eye. Cervical plain x-ray and cerebral CT did not show any significant pathology. Neurological symptoms spontaneously greatly regressed within a few hours. Some persisting pain plus left ptosis and miosis improved two days later. The day after, a cervical MRI showed a C2-C5 left epidural mass which displaced the cord and reduced its diameter. The lesion was isointense on T1 and hyperintense on T2-weighted images with a peripheral rim of hypointensity (Fig. 1 A-B-C-D). A cervical CT scan confirmed the haemorrhagic nature of the lesion. Angiography was negative for vascular lesions. The patient remained asymptomatic on the following days. Two weeks later, a cervical MRI showed the complete resolution of the haematoma (Fig. 1 E-F).

Discussion

SSEH is a rare cause of spinal cord compression. Its annual incidence is about 0,1 per 100.000 individuals (Holtas *et al.,*1996), with a male/female ratio of 1,5:1 (Groen, 2004). Hypertension, anticoagulant therapy, increased venous pressure, pregnancy and vascular malformations have been described as predisposing factors (Liao *et al.*, 2004), but the exact pathogenesis remains unknown. The posterior internal vertebral venous plexus seems to play an important role (Groen, 2004); a reversal of blood flow due to fluctuations of intra-thoracic and intra-abdominal pressures may induce a rupture of a vein in the valveless epidural plexus (Patel *et al.,* 1998).

Symptoms usually begin with sudden-onset severe back or neck pain, radiating to the corresponding

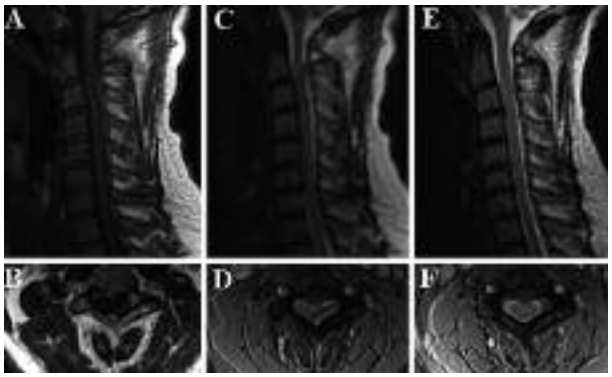

FIG. 1. — T1-weighted sagittal (A) and axial (B) and T2-weighted sagittal (C) and axial (D) MR images one day after the onset of the symptomatology: a left epidural posterolateral haematoma is shown displacing the spinal cord.

Two weeks later T2-weighted sagittal (E) and axial (F) MR images showing the complete resolution of the haematoma.

dermatomes; afterwards, symptoms and signs of spinal cord compression appear (Liao *et al.*, 2004).

In our report, we observed the rare occurrence of a BSS associated with a HS.

BSS is characterized by ipsilateral weakness and loss of proprioceptive sensitivity with contralateral loss of pain and temperature sensitivity below the level of spinal cord hemisection. The most common cause is represented by penetrating trauma; other causes include syringomyelia, extramedullary spinal neoplasms and blunt injury (Miranda *et al.*, 2007). The prognosis is usually favourable, particularly for blunt rather than penetrating injury, with motor recovery within 6 months (Roth *et al.*, 1991). BSS was associated with SEH in twenty cases in literature (Shen *et al.*, 1995; Narberhaus *et al.*, 2002) and a significant trauma history was found only in three cases. All SEHs were located in the posterolateral region of the cervical spine. A transient cord ischemia due to local mass effect was hypothesized as the cause of BSS (Shen *et al.*, 1995). Three patients received a conservative management, whereas the others underwent a laminectomy. A complete neurological recovery was observed in all cases.

HS results from interruption of the ipsilateral sympathetic pathway at any level along its course between the hypothalamus and the eye. In our case, HS was likely caused by a compression of the sympathetic pathway along the lateral column at C2-C4 levels.

Crabbe *et al.* (1992) and Shen *et al.* (1995) described cases of post-traumatic SEH, with the simultaneous presence of BSS and HS. Garcia-Manzanares *et al.* (2000) and Edwards and Andrews (2001) observed the same clinical association following, respectively, penetrating trauma and blunt injury to the cervical spine.

To our knowledge, the present appears to be the first case in which both syndromes are due to a non-traumatic SEH (Table 1).

In cases of acute SSEH associated with neurological deficits, an emergent surgical evacuation is considered the standard of care (Packer and Cummins, 1978; Shen *et al.*, 1995; Groen, 2004). The neurological outcome is determined by the preoperative status and time to surgery (Liao *et al.*, 2004). However, several cases of spontaneous clinical and radiological complete recovery in conservatively treated SSEH patients are reported in literature. Groen (2004) collected 64 cases and suggested that spontaneous resolution of neurological symptoms may be due to an elution of the haematoma along the spinal epidural space. This hypothesis appears to be consistent with the significantly longer extension of the SEH in conservatively treated cases.

Non-surgical management could be a reasonable alternative in patients presenting with minimal neurological involvement and a rapid recovery (Groen, 2004). Narberhaus *et al.* (2002) suggested a conservative treatment in SEH presenting as BSS.

Table 1

Spinal epidural haematomas presenting with Brown-Séquard syndrome and Horner's syndrome

Authors - Years	Sex - Age	Etiology	Diagnosis	Location	Treatment	Results
Crabbe 1992	M - 20	trauma	CT	C4 - C6	Conservative	Complete recovery
Shen 1994	M - 58	trauma	MR	C2 - C6	Right hemilaminectomy	Complete recovery
Manzanares 2000	M - 21	trauma	MR	cervico-medullary junction	Conservative	Complete recovery
Edwards 2001	M - 26	trauma	MR	C6	Decompression and stabilization	Complete recovery
Present case	M - 52	spontaneous	MR	C2 - C5	Conservative	Complete recovery

Moreover, the risk of spinal cord infarction after decompressive laminectomy for SSEH should be considered (Packer and Cummins, 1978). MRI appears to be the tool of choice to follow the evolution of the clot in non-operated patients.

Conclusion

SSEH is a rare clinical entity, even more rare with a BSS and HS presentation. Surgery is the first-choice treatment, although a conservative therapy remains an option especially in patients without neurological deficits or showing only a minimal clinical impairment but in rapid improvement.

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