Case Report

Epidural blood patch for spontaneous intracranial hypotension with chronic subdural haematoma: A case report and literature review

Jian Zhang, Dan Jin and Kong-Han Pan

Abstract
Spinal leakage of cerebrospinal fluid (CSF) is considered to be the primary cause of spontaneous intracranial hypotension (SIH). Subdural haematoma (SDH) is a serious complication of SIH. This current report presents a case of bilateral SDH with SIH that was treated with epidural blood patching (EBP). A 43-year-old male complained of experiencing orthostatic headaches for 2 months without neurological signs. The patient worsened in a local hospital and was transferred to the Sir Run Run Hospital. Brain computed tomography showed bilateral SDH with a midline shift. The patient underwent emergency trephination in the left frontal temporal region. Postoperative magnetic resonance myelography showed a CSF leak originating at the T11–L2 level. As a consequence of clinical deterioration of the patient, EBP was subsequently performed at the T12–L1 level. The headache was rapidly relieved and later the SDH was completely absorbed. This case report and literature review aims to remind clinicians that SIH can cause SDH and that EBP is a viable treatment option.

Keywords
Spontaneous intracranial hypotension, subdural haematoma, epidural blood patching

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Introduction
Spontaneous intracranial hypotension (SIH) is caused by spinal leakage of cerebrospinal fluid (CSF) and it is characterized by an orthostatic headache without a history of trauma or dural puncture.1 Subdural haematoma (SDH), a serious complication of SIH, may lead to neurological deficits and it can even be life-threatening.2 However, the aetiology of SDH in SIH patients

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remains unclear.³ Epidural blood patching (EBP) has been regarded as the mainstay of therapy for SIH, often providing instantaneous relief of symptoms in 90% of cases regardless of the site of the leak.⁴ Lumbar EBP is considered to be a viable treatment choice because of its low risk of recurrence.⁵ This report presents a case of bilateral SDH with SIH that was successfully treated with lumbar EBP. In addition, the current literature has been reviewed to provide some suggestions to clinicians treating SDH with SIH in clinical practice.

Case report

A 43-year-old male patient presented to the Department of Neurosurgery, Jiangsu Provincial People’s Hospital, Nanjing, Jiangsu Province, China on 27 June 2015 with orthostatic headaches that had been occurring for 2 months. There was no history of trauma. Brain computed tomography (CT) and computed tomography angiography (CTA) were normal. Lumbar puncture test showed that the CSF pressure was 0 mmH₂O. The patient was given a prescription for volume expansion therapy and sent home without further investigations. On 8 July 2015, the patient presented with similar complaints that the headaches were worse when standing, but improved when he was recumbent. On 23 July 2015, the patient was referred to the emergency department of Sir Run Run Hospital, School of Medicine, Zhejiang University, Hangzhou, Zhejiang Province, China with progressive symptoms. Neurological examinations, such as consciousness, pupillary light reflex, and the muscle force of limbs, were intact. Brain CT imaging revealed bilateral SDH in the fronto-parietal region and a midline shift (Figure 1a). This patient was diagnosed with a chronic SDH and cerebral hernia. The patient underwent a trephination of the left SDH because it was the larger SDH. Postoperative magnetic resonance myelography of the entire spinal column revealed a thin layer of epidural CSF fluid collection from T11 to L2, with no abnormal CSF out-pouches along any of the nerve roots (Figure 2). The patient was restricted to complete bed rest with the head of the bed flat.

Figure 1. Axial computed tomography images of the brain of a 43-year-old male patient who presented with orthostatic headaches: (a) showing enlargement of the left-sided subdural haematoma (arrow) with 10 mm of midline shift. Performed on day 1 of hospital admission; (b) showing effusion pneumatosis in the operative region (arrow), midline shift, and compression of the posterior fossa. Performed on day 7 of hospital admission; and (c) showing haematoma absorption without midline shift. Performed on postoperative day 15.
At 5 days after trephination, the patient had clouding of consciousness with a Glasgow coma scale (GCS) score of 8. Brain CT on day 7 of hospital admission revealed effusion pneumatosis in the operative region, midline shift, and compression of the posterior fossa (Figure 1b). With the patient resting in a supine position under general anaesthesia, a 22-gauge needle was inserted into the middle epidural compartment at the T12–L1 level. An autologous blood patch was collected from the patient’s right brachial vein. A total of 18 ml of autologous blood was injected into the epidural space via an extension pipe. On postoperative day 1, the patient had clear consciousness with a GCS score of 14. On postoperative day 14, his headache was almost completely dissipated. Brain CT on postoperative day 15 revealed haematoma absorption without midline shift (Figure 1c). The Sir Run Run Hospital does not require ethical approval for reporting individual cases. Written informed consent was obtained from the patient for publication of this case report and its accompanying images.

**Discussion**

Spontaneous intracranial hypotension is characterized by an orthostatic headache and low CSF pressure in the absence of a history of trauma or dural puncture. It has an annual incidence of 5 per 100 000. Spontaneous CSF leakage at the level of the spine is regarded as the main cause of SIH with a high incidence. CSF leakage mainly occurs in weak areas around nerve roots.
root sheaths and the dura mater, and as a result of small defects due to slight traumas caused by severe exercise or a cough.\(^8\) Although the pathophysiology of SIH remains unclear, several theories have been proposed to account for the cause of the CSF leakage.\(^9\) One theory states that CSF volume loss might be the cause of SIH,\(^10\) which is supported by the fact that certain connective tissue disorders, such as Ehler’s Danlos and Marfan’s, are risk factors for SIH.\(^11\) Another theory considered epidural hypotension as the main cause of SIH, because EBP works by increasing the epidural space pressure, rather than sealing the CSF leak.\(^12\)

Orthostatic headache is regarded as the most common presenting symptom of SIH,\(^1\) although other symptoms such as nausea, diplopia, neck pain and stiffness, dizziness, tinnitus, photophobia, and blurred vision, were also reported in a previous report.\(^13\) Orthostatic headache is defined by the International Headache Society as a headache that develops in temporal relation to low CSF pressure or CSF leakage, or led to its discovery.\(^14\) However, the exact pathophysiological mechanism involved in orthostatic headaches in SIH remains unclear. The Monro-Kellie hypothesis states that the sum of the volumes of brain tissue, CSF, and intracranial blood is constant.\(^15\) CSF volume loss might cause engorgement of cerebral pain-sensitive venous sinuses that leads to headaches.\(^15\) SDH is a serious complication of SIH that may lead to neurological deficits and it can even be life-threatening.\(^2\) Although the pathophysiology of SDH in patients with SIH remains unknown, several mechanisms have been proposed. For example, a brain downward shift due to CSF volume loss may tear the bridging veins, causing these veins to rupture.\(^1\) In this present case, the patient presented with orthostatic headaches that had been occurring for 2 months and he subsequently suffered SDH.

The diagnosis of headaches due to SIH has been described by ICHD-3,\(^14\) and the diagnostic criteria have been reported.\(^16\) Headaches due to SIH are diagnosed when all the following items are present: (A) orthostatic headache; (B) the presence of at least one of the following: low opening pressure (≤ 60 mmH\(_2\)O), sustained improvement of symptoms after epidural blood patching, demonstration of an active spinal cerebrospinal fluid leak, cranial magnetic resonance imaging changes of intracranial hypotension (e.g. brain sagging or pachymeningeal enhancement); (C) no recent history of dural puncture; and (D) not attributable to another disorder.\(^16\) Radionuclide cisternography is considered to be the most accurate method for localizing the site of a CSF leak.\(^17\) MR myelography is the most common noninvasive technology for detecting the site of a CSF leak, which can show pachymeningeal enhancement, extradural fluid extravasation extending to the paraspinal soft tissues, and engorgement of epidural venous plexuses.\(^18\) Enhanced MRI can also show subdural fluid collections, diffuse pachymeningeal enhancement, obliteration of basal cisterns, descent of cerebellar tonsils, engorged cerebral venous sinuses, enlarged pituitary, and decreased size of the ventricles.\(^19\) However, despite advanced imaging, the site of the CSF leak can only be detected in up to 50% of cases.\(^18\) The present case presented with an orthostatic headache, a low CSF pressure (0 mmH\(_2\)O), and MR myelography of the entire spinal column revealed a thin layer of epidural CSF fluid collection from T11 to L2. Brain CT showed bilateral SDH in the fronto-parietal region. Hence, the diagnosis of SIH and SDH were clear in the present case.

The primary goal of SIH therapy is to stop the CSF leak and increase CSF volume.\(^1\) Conservative treatment, such as complete bed rest with the head of the bed flat, hydration, or the application of an
abdominal binder, are viable choices. Medical treatments, including steroids, acetazolamide, intravenous caffeine, and oral theophylline, have been reported in previous studies with inconsistent conclusions. After the failure of conservative and/or medical treatments, EBP is the modality of choice that should relieve symptoms in 90% of cases. EBP can also be used in SIH patients without identifying the site of the CSF leak. The EBP (10-20 ml) from the patient's brachial vein is performed in the lumbar region. Previous research has reported that the success rate of EBP was high (80%–95%). If lumbar EBP fails to provide relief, it can be repeated because of its low risk of severe complications. However, whether to perform surgical intervention of SDH remains controversial. SDH can be managed by safely sealing the CSF leak without the evacuation of the SDH. If symptoms can be improved using EBP treatment, then even a thick SDH can be resolved spontaneously. However, large SDHs might cause uncal herniation and lead to neurological deterioration. Surgical evacuation of SDH is necessary for those patients with SIH who have acute changes in consciousness. Another controversial issue is whether to perform EBP or surgical intervention of the SDH as the initial procedure. Previous research has indicated that drainage of the SDH before EBP might cause a rapid decline in intracranial pressure that disrupts bridging veins, which might lead to an acute haemorrhage. However, if uncal herniation is initiated, it might lead to an irreversible negative outcome even after drainage of the SDH. The present case showed clouding of consciousness on the 5th day after surgical drainage of the SDH (i.e. trephination). A 22-gauge needle was immediately inserted into the middle epidural compartment at the T12–L1 level. A total 18 ml of autologous blood from the patient’s right brachial vein was injected into the epidural space via an extension pipe. The orthostatic headache was relieved and the SDH disappeared. If an EBP had been performed shortly after drainage of the SDH, it is probable that the deterioration would have been prevented. This experience provided a valuable lesson for the treatment of SDH associated with SIH. In addition, surgical repair can be performed if repeated EBP fails, which resulted in very effective headache resolution in a single case report. However, surgical treatment is more appropriate for patients with SIH and a clear site of the CSF leak.

In conclusion, this report describes the case of a bilateral SDH, which was a severe complication of SIH with a CSF leak originating at the T11–L2 level. EBP was performed at the site of the leak to restore CSF volume and relieve the orthostatic headache.

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Declaration of conflicting interests
The authors declare that there are no conflicts of interest.

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