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Case Report

Cervical Epidural Blood Patch for Spontaneous Intracranial Hypotension-A Case Report

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Abstract: Spontaneous intracranial hypotension (SIH) is an uncommon disorder caused by cerebrospinal fluid (CSF) leaks in the spine. Treatment is directed at sealing the site of leak, which is often difficult to localize. We present a case of near fatal SIH that was treated with cervical epidural blood patch (EBP) and burr hole drainage of the hematoma. A 41-year old male presented with severe headache after drinking alcohol over several weeks. Brain magnetic resonance imaging (MRI) showed features typical of SIH. But, further imaging with MR myelography and radionuclide cisternography failed to identify a precise site of leak. Despite lumbar EBP, the patient's orthostatic headache recurred and developed acute subdural hematoma(SDH) to require burr hole drainage for hematoma evacuation. After surgery, the patient's mental state improved, and the amount of SDH decreased, but he still complained of severe orthostatic headache. We performed EBP at cervico-thoracic junction level. The headache was rapidly relieved and the SDH was completely absorbed. In this case, we aim to remind clinicians that cervical EBP should be considered management for SIH as an alternative to the repeated lumbar blood patch.
Keywords: CSF, epidural blood patch, headache, spontaneous intracranial hypotension.

INTRODUCTION

Spontaneous intracranial hypotension (SIH) is a debilitating condition induced by spinal CSF leaks [1, 2]. It is distinguished by an orthostatic headache that has no history of trauma or dural puncture [1]. Subdural hematoma (SDH), a major consequence of SIH, can cause neurological impairments and possibly death [3]. However, the cause of SDH in SIH patients is unknown [4].

Initial management of SIH may involve conservative measures such as hydration and bed rest. If these measures fail, epidural blood patch (EBP) is the typical treatment of choice [5, 6].

We describe the successful treatment of a near-fatal SIH case in which the exact location of the leak was not detected using EBP at cervical levels.

CASE PRESENTATION

A 41-year-old male, previously healthy and not on any medications, presented to the neurology department with the sudden onset of a severe headache that occurred after drinking alcohol.

There was no history of head trauma. Physical examination revealed no sensory or motor deficits. Brain computed tomography (CT) and magnetic resonance imaging (MRI) were organized to find any abnormal brain lesions after admission. The brain MRI showed shortening of the pontomamillary distance (about 4.6mm) on sagittal T1-weighted imaging and enhancement of the pachymeninges on gadolinium-enhanced MRI with no other demonstration of diffusion-restricted lesions or acute intracranial hemorrhage (Fig. 1). These MRI findings were consistent with intracranial hypotension.

Initially, patient is treated with conservative medical therapies, which include bed rest, oral hydration, caffeine, and steroids. But, the patient did not respond to these management.

Therefore a blind L3-L4 lumbar EBP was performed using 20 ml of autologous blood. Following the EBP, the patient reported improvement of his headaches and was subsequently discharged.

He returned one month later with a recurrence of his headaches. The headaches were worst when standing or sitting and lessened when lying down. He also complained of nausea and vomiting. A brain CT was performed, which revealed the development of acute SDH. He was referred to the department of neurosurgery. Further workup, including an MR myelography of the whole spine and radionuclide cisternography, revealed no definitive site of CSF leakage.

On the fourth day of admission, obstructive hydrocephalus and increased SDH were found on follow-up CT (Fig 2). Therefore, he was urgently taken to the operating room for burr hole drainage of the hematoma.

Postoperatively on the first day, he was in a similar clinical condition, and experienced a change in mental status to drowsy. Postoperatively on the fifth day, the patient maintained an alert mental status; however, he still mentioned severe headaches. Decreased SDH was found on follow-up CT. A C-arm-guided EBP at the cervicothoracic junction level was performed using 13ml of autologous blood by the anesthesiologist. Following the procedure, he experienced relief of his headaches.

Finally, on the twenty-second day postoperatively, he remained free of headaches with reduced SDH observed on follow-up brain CT. He was confirmed for discharge. During the one-year follow-up period, no recurrences were identified.

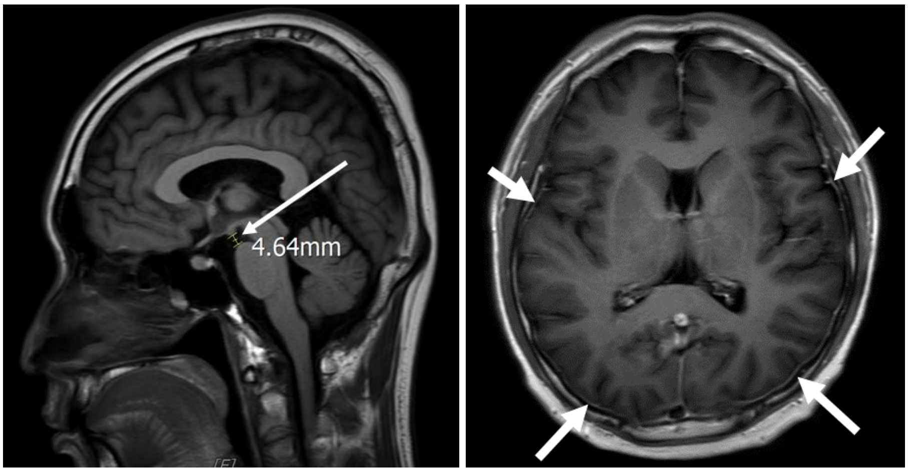

Figure 1: Sagital (left) and axial (right) views of a T1 post-contrast MRI of the brain showing shorting of the pontomamillary distance(White arrow, left), pachymeningeal enhancement(White arrow, right), suggestive of SIH

Figure 2: CT scan of the brain showing increased SDH (White arrow) and obstructive hydrocephalus (arrow head)

DISCUSSION

SIH is characterized by orthostatic headache, low CSF pressure, and diffuse pachymeningeal enhancement on MRI, all in the absence of antecedent trauma or lumbar puncture [7].

The precise prevalence of SIH is unknown, although estimates range from one in 50,000 to five in 100,000 [8]. SIH is caused by CSF leakage, which results in a decrease in total CSF volume. The pathogenesis is mainly unclear, however various theories suggest that an underlying weakening of the spinal meninges is likely [7]. Certain connective tissue disorders, such as Marfan's and Ehler's Danlos, are risk factors for SIH [9]. Another idea contends that negative pressure within the inferior vena cava is important in pathophysiology [6]. Negative pressure in the inferior vena cava may promote over-drainage of the epidural spinal veins. The ensuing decrease in spinal epidural vein pressure affects the CSF-blood gradient, producing CSF aspiration into the epidural space and potentially triggering CSF leak [10].

Although the exact cause of SDH development in the presence of SIH is unknown, bridging vein rupture is most likely to blame [10]. Diffuse pachymeningeal enhancement, evidence of brain sagging, subdural fluid collections, and engorged cerebral venous sinuses are all characteristic brain MRI findings of SIH [11]. Myelography is considered the study of choice for localization of the CSF leak, while radionuclide cisternography can be useful for localization when myelography results are normal [1]. In this case, pachymeningeal enhancement in the brain MRI was confirmed, but MR myelography and radionuclide cisternography were unable to identify the exact location of the leak.

The goal of SIH treatment is to stop the CSF leak and restore the CSF volume [12]. Patients are initially treated with conservative therapy such as bed rest, oral water, caffeine, and steroids [5, 6]. An EBP is the basis of treatment for patients who fail these conservative management [5]. EBP may function via a two-part mechanism whereby the EBP initially functions by replacing lost CSF volume and tamponading the leak. It is speculated that the delayed effect of EBP is due to the creation of a hemostatic clot, which eventually forms a more permanent seal [13]. EBP is often providing instantaneous relief of symptoms in 90% of cases [7].

It is traditionally conducted in the lumbar region and involves the injection of autologous blood into the epidural space [14]. Many people advocate for administering EBP without first locating the source of the CSF leak [15]. Therefore we attempted lumbar EBP with 20 ml of blood because of the uncertain leaking site. The success rate of EBP increases with the volume of blood used, with quotes of 80% success rate with 10-15 ml and >95% success rate with 20 ml of blood [14]. However, in this case, the patient had a recurrence of SIH with SDH one month later. This time, considering the possibility that the site of leakage was not the lumbar region, EBP was performed in the cervico-thoracic junction. Prolonged clinical course has been identified as a risk factor for the development of SDH in the setting of SIH [16]. Additionally, the first lumbar EBP may have not been effective in treating the higher cervical/thoracic source of CSF leak [17, 18]. Despite this, it is highly likely that the cervical EBP ultimately did provide control of the CSF leak in our patient, as during the burr hole procedure the brain appropriately rose to fill the intracranial space following drainage of the hematoma.

In summary, directed EBP in the cervicothoracic spine should be considered for SIH when lumbar EBP fails. Cervical EBP, however, do carry a higher risk of complications.

CONCLUSION

We describe a case of SIH complicated by SDH where a lumbar EBP approach was unsuccessful and a cervical approach led to control of the CSF leak in SIH and ultimate drainage of the SDH was required.

Conflict of Interest: No conflict of interest was declared by the authors.

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Consent for Publication

Written informed consent was obtained from the patient for publication of this case report and the accompanying image.

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