Targeted Epidural Blood Patch Treatment for Refractory Spontaneous Intracranial Hypotension: A Case Report

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Abstract

Purpose of Review Spontaneous intracranial hypotension (SIH) is recognized far more commonly than before, and it is well known that SIH is sometimes complicated by chronic subdural hematoma (SDH). We reported a patient who was treated with epidural blood patch (EBP) five times for refractory SIH and SDH surgery.

Recent Findings We experienced that targeted EBP was useful in refractory SIH, and also can be performed safely prior to drainage of the hematoma.

Summary We report the case of SIH patient with a bilateral SDH who came to our hospital and was discharged 2 weeks later with no neurologic deficit after trephination and five times EBP treatment. It shows that targeted EBP was useful in refractory SIH, and also can be performed safely prior to drainage of the hematoma.

Keywords
- spontaneous intracranial hypotension
- chronic subdural hematoma
- epidural blood patch
- burr hole drainage

Introduction

Spontaneous intracranial hypotension (SIH) is typically characterized by postural headache, low cerebrospinal fluid (CSF) pressure, and no previous history of head trauma or dural puncture.[1] For SIH, conservative treatment is usually attempted first. If conservative management is not fully relieved, an autologous epidural blood patch (EBP) may be considered.[2]

Subdural hematoma (SDH) is a complication of SIH, which is also treated with EBP. Cases of large subdural hemorrhage require surgical drainage and treatment of the underlying cause of SIH.[3]

We report the case of SIH patient with a bilateral SDH who came to our hospital and was discharged 2 weeks later with no neurologic deficit after trephination and five times EBP treatment.

Case Presentation

A 28-year-old man presented to our hospital with headache that had been occurring for more than 1 month, without history of trauma. The headache worsened when standing and improved after lying down. He was diagnosed with SIH and treated conservatively in a local hospital, but his symptom did not improve, so he was transferred to our hospital. Neurological examinations, such as consciousness, papillary light reflex, and muscle force of limbs, were intact, excepted for tendon reflex 2(+). Brain computed tomography (CT) showed bilateral SDH at the top of the frontotemporal region (►**Fig. 1**). Whole spine magnetic resonance myelography (MRM) revealed multiple CSF leakages in cervical–thoracic and upper thoracic levels (►**Fig. 2**). The SIH patient was diagnosed with bilateral SDH. As conservative treatments were ineffective, EBP was performed under strict aseptic

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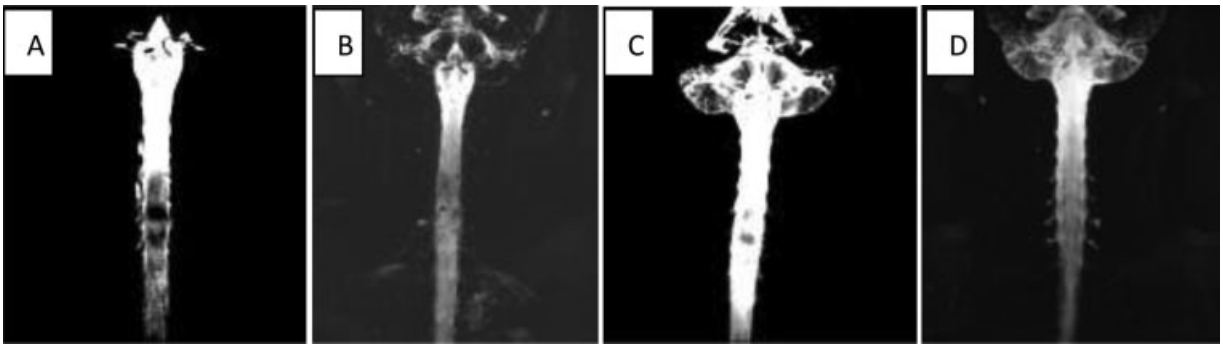

Fig. 1 Magnetic resonance myelogram. (**A**) Multiple cerebrospinal fluid (CSF) leakages in cervical–thoracic and upper thoracic regions: myelogram performed on day 2 of hospital admission. (**B**) CSF leakage at the right cervical–thoracic region: myelogram performed on day 3 after the second epidural blood patch (EBP). (**C**) Blurry fluid extends along the nerve roots on both sides of the spinal cord at cervical–thoracic region: myelogram performed on day 6 after the third EBP. (**D**) No significant CSF leakage is observed; nerve root sheath cyst is shown in cervical–thoracic region: myelogram performed on day 8 after the fifth EBP.

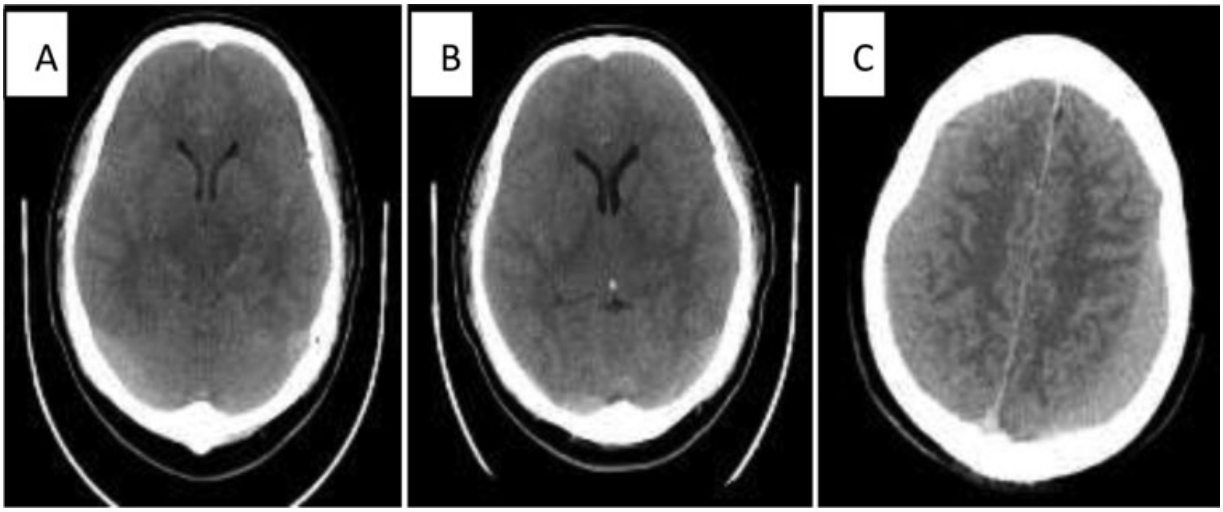

Fig. 2 Computed tomography images of the brain. (**A**) Bilateral subdural hematoma: imaging performed on day 1 of hospital admission. (**B**) Hematoma absorption than before: imaging performed on day 2 after the first epidural blood patch (EBP). (**C**) Hematoma progress: imaging performed on day 4 after the third EBP.

conditions. The patient was awake during the procedure, in a prone position. An 18-gauge needle was inserted at the T3–4/T2–3 level intervertebral space using a midline approach. The epidural space was identified using the saline loss-of-resistance. We performed the blood patch by an injection of autologous peripheral unclotted vein. The volume of injected blood was $<$15 mL including contrast agent. The patient was continued in the prone position for 30 minutes and placed in a supine position with strict bed rest for the next 48 hours, to ensure sufficient spread. In the period of performing EBP five times, other symptoms appeared, such as dizziness and vomiting. On the third day after the second EBP, MRM showed CSF leakages only in cervical–thoracic regions (►**Fig. 2**); compared with previous result, thoracic CSF had vanished. After the fourth EBP later, CT showed wide hemorrhage (►**Fig. 1**). After the fifth EBP later, his CSF leakages had vanished, and magnetic resonance imaging showed hematoma absorbed (►**Fig. 3**). Because of his persistent headache, our clinicians decided to burr hole trephination hematoma evacuation. Finally, his headache had improved.

Discussion

SIH is caused by spinal leakage of CSF and it is characterized by an orthostatic headache without a history of trauma or dural puncture.[2] Spontaneous CSF leaks, for sure, due to dural weakness ranging from simple dural tears to multilevel complex meningeal diverticula, allow CSF to leak into the epidural. It is usually seen in the cervical or thoracic region, and is also associated with connective tissue disorders such as Marfan syndrome, neurofibromatosis, polycystic kidney disease, and Ehlers–Danlos syndrome.[4] Our patient denied any underlying disorder or notable medical history. Nerve root sheath cyst rupture may have led to CSF leakage, which was the reason of this posture headache.

Measures such as bed rest, adequate hydration, caffeine intake, and abdominal binders are conservative approaches for SIH. A study showed that only 37% of patients received headache relief at 6 months and 63% at 2 years. Due to long periods of bed rest, without certainty of a cure, many patients will therefore pursue other therapies.[5]

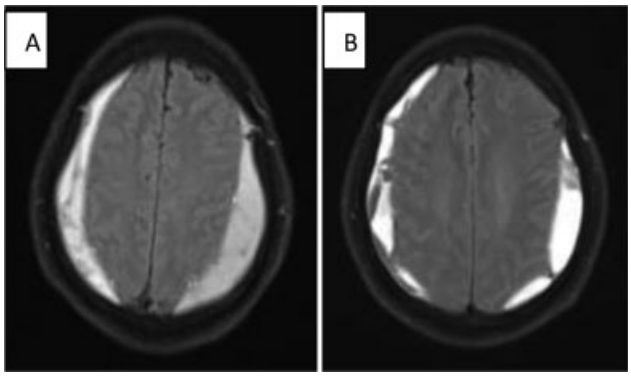

Fig. 3 Magnetic resonance image of brain. (**A**) Wide hematoma: imaging performed on day 1 after the fourth epidural blood patch (EBP). (**B**) Hematoma absorption than previous: imaging performed on day 6 after the fifth EBP.

When initial conservative measures are ineffective, antilogous EBP should be considered. It is primary method of treatment for SIH, especially when there are explicit leakages. EBP acts as a mass lesion compressing the dura and raising intracranial pressure by increasing CSF volume; the blood clot that forms seals off the dural hole and finally the CSF leak stops.[4] EBP can be targeted to the CSF leakage site or nontargeted. Some studies have shown that targeted EBP is more effective than nontargeted ones.[2,6–8] No protocol has yet been developed. In our institution, we recommend placing an EBP at least 2 weeks after conservative approaches. To perform targeted EBP, it is important to know the location of CSF leakage. In our patient, MRM spinal imaging technique was considered first, because we believe that the site of CSF leakage can efficiently and safely be identified, and the location of the tear can be identified accurately.[9] Therefore, we opine that EBP procedures should be performed as near to the leakage site as possible to improve the efficacy and success rate. This patient's puncture point below the MRM demonstrated CSF leakage. The volume of blood to be injected while performing EBP remains uncertain. Typical volumes range from 10 to 40 mL for nontargeted patches via the interlaminar approach. Less volume is required when the injection is targeted to a known CSF leak.[5] Wu et al[10] found that a larger blood volume for EBP was associated with a higher success rate. But with higher volumes, caution must be exercised not to compress the spinal cord. In this patient, the volume of injected blood was $\leq$15 mL (13 mL, 14 mL, 12 mL, 12 mL, 12 mL, respectively). We stopped the injection when the blood was once through the leaks, or the patient complained of radicular pain, numbness, or headache. In this case, after the third EBP, his SDH progressed. On day 20 after five times EBP, MRM imaging of the entire spinal revealed no abnormality.

However, in recent years, SIH is often reported to be associated with chronic SDH development. Although the pathophysiology of SDH in patients with SIH remains unknown, studies have proposed several mechanisms, such as: a brain downward shift due to low CSF pressure may tear the bridging veins, or as subdural CSF collections gradually enlarge the subdural space, it causes these veins to stretch and rupture.[11]

Moreover, the initial procedure therapies for SDH in SIH remain controversial. SDH, which is limited in volume, usually resolves after normal CSF volume has been restored. In slow enlargement of the hematoma, the period of spatial compensation is possibly enough to neutralize the intracranial pressure; evacuation of the hematoma is not needed and may lead to worsened outcomes.[12] The downward displacement of the brain and precipitation by craniotomy for evacuation of SDH caused stroke leading to death, as reported in a study.[13] Wu et al[10] indicate that delay in treatment for CSF leakage with EBP perhaps was the primary cause of SDH complication occurring with SIH. In accordance with the previous reports, EBP is peculiarly required to precede surgical drainage of chronic SDH to stop CSF leakage.[10] Takahashi et al consider that even in SIH cases with large SDH, EBP should and can be performed safely prior to drainage of the SDH.[14] Our study[9] also expressed that most SIH patients with SDH should give priority to EBP rather than surgical drainage of the hematoma, especially when the epidural hematoma is not obvious, and the site of CSF leak is identified. In case of large subdural hemorrhage or symptomatic one, surgical drainage of the hematoma should be considered.

In contrast, Ikeda et al reported that after a burr hole irrigation of hematoma, not only chronic SDH but also the symptoms of SIH were completely resolved and there was no recurrence.[15]

Conclusions

The present case shows that targeted EBP was useful in refractory SIH, and also can be performed safely prior to drainage of the hematoma.

Conflict of Interest
None.

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