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Case Report

Intracranial subdural hematoma coexisting with improvement in spontaneous intracranial hypotension after an epidural blood patch

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Abstract

A 36-year-old male had spontaneous intracranial hypotension (SIH) presenting with refractory headache for 4 months. Multiple epidural blood patches (EBPs) yielded relief of symptoms, but the course was complicated, with asymptomatic intracranial subdural hematoma (SDH). Except for SDH, other radiological diagnostic signs of SIH were resolved and the patient's headaches improved after EBP. Owing to a mass effect and persistent cerebrospinal fluid (CSF) leakage, surgical repair of the spinal leakage was performed, but no cranial procedures were carried out. Postoperatively, the SDH completely resolved, but there was still CSF leakage at the level where surgery was performed. The patient has remained free of headache or other events for 3 years. It was reduction rather than elimination of the spinal CSF leak that yielded remission of SIH. In summary, intracranial SDH can be a complication of inadequately treated SIH (i.e. persistent minor CSF leakage). Management of SDH should focus on correction of the underlying SIH rather than craniotomy for hematoma evacuation.

Keywords: epidural blood patch; spontaneous intracranial hypotension; spontaneous spinal cerebrospinal fluid leaks; subdural hematoma; surgical repair

1. Introduction

Spontaneous intracranial hypotension (SIH) was first described by George Schaltenbrand in the German literature in 1938 and in the English literature in 1953.[1,2] Its prevalence was estimated at 1 per 50,000 in a community-based study.[3] Women are more frequently affected than men, with a female/male ratio of approximately 2:1.[4] Onset of symptoms typically occurs in the fourth or fifth decade of life, with a peak incidence at approximately 40 years of age.[4] Single or multiple spinal cerebrospinal fluid (CSF) leaks are considered the cause, but the true etiology remains uncertain.[2,4,5] The clinical manifestations and radiological findings mainly result from intracranial fluid depletion.[2,4,6] The treatment of choice remains controversial, especially in the presence of intracranial subdural hematoma (SDH).[4,7,8]

2. Case report

A 36-year-old delivery man, who was previously in good health, presented with postural headache for 4 months. Prior to

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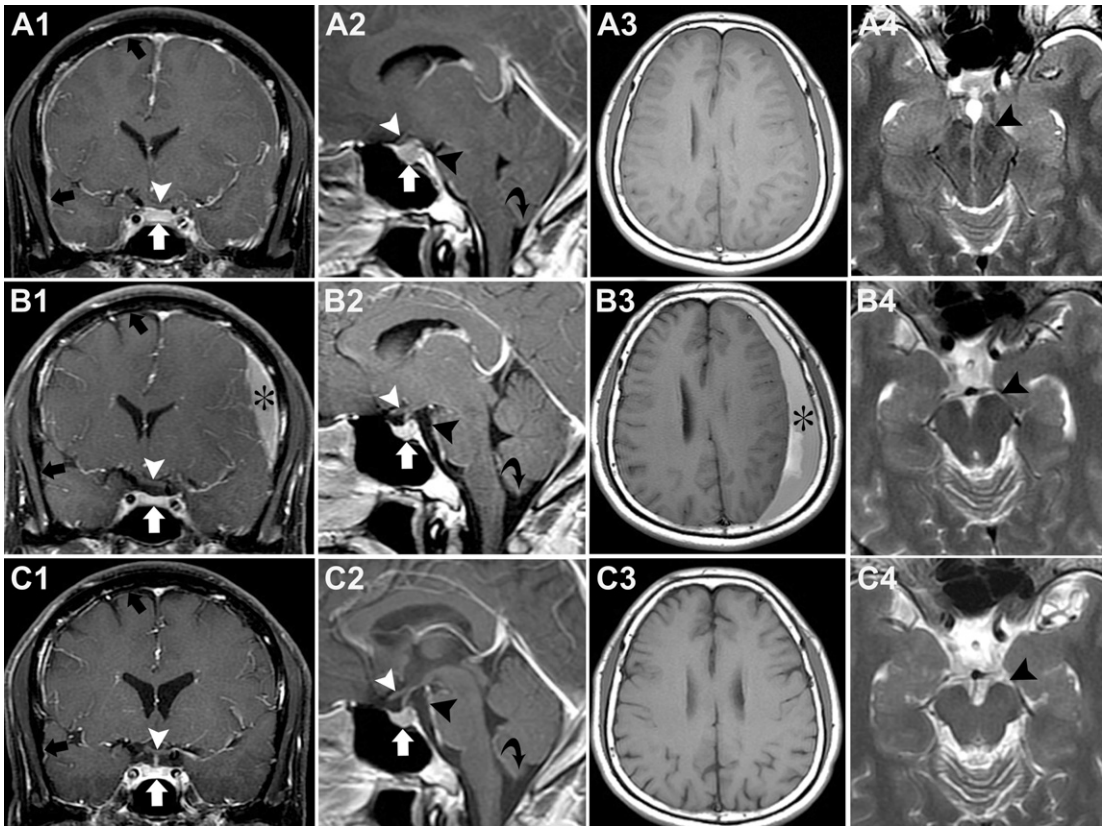

Fig. 1. Magnetic resonance imaging panel illustrating signs of spontaneous intracranial hypotension. The initial (pre-EBP), pre-operative (post-EBP), and 2-year post-operative images are shown on the top row (A1–A4), middle row (B1–B4), and bottom row (C1–C4), respectively. A1, B1, and C1 are coronal T1-weighted images (T1WI) after gadolinium contrast injection (+C). A2, B2, and C2 are sagittal T1WI+C. A3, B3, and C3 are axial T1WI without gadolinium contrast injection. A4, B4, and C4 are axial T2-weighted images. Subdural hematoma (asterisk, B1, B3) complicated after EBP treatment. However, the other signs of SIH subsided, including the pachymeningeal enhancement (arrow, A1, B1, C1), pituitary hyperemia (white arrow, A1, B1, C1, A2, B2, C2), crowded prepontine cistern and midbrain (arrowhead, A2, B2, C2, A4, B4, C4), bowing of the optic chiasm (white arrowhead, A1, B1, C1, A2, B2, C2), and downward herniation of the cerebellar tonsils (curved arrow, A2, B2, C2).

this complaint, he had no history of cranial or spinal trauma, surgery, or anesthetic procedures. Examination revealed no cranial neuropathy, cognitive disorder, or other neurological deficits. Magnetic resonance imaging (MRI) of the brain revealed signs of SIH, including diffuse pachymeningeal enhancement, bilateral subdural fluid collection, hyperemia of the pituitary gland, flattening of the pons against the clivus, bowing of the optic chiasm over the pituitary fossa, and downward herniation of the cerebellar tonsils (Fig. 1). Magnetic resonance (MR) myelography revealed epidural collection of CSF at multiple sites along the spine (Fig. 2). According to the clinical manifestations and radiological signs, a diagnosis of SIH was established. After the failure of conservative treatments, including aminophylline, nonsteroidal anti-inflammatory drugs, and intravenous hydration, we recommended an epidural blood patch (EBP) at the T6–T7 level. The patient subjectively claimed relief of postural headache immediately after the first EBP. However, the therapeutic duration lasted for only 2 weeks before the headache recurred. The patient subsequently underwent another three

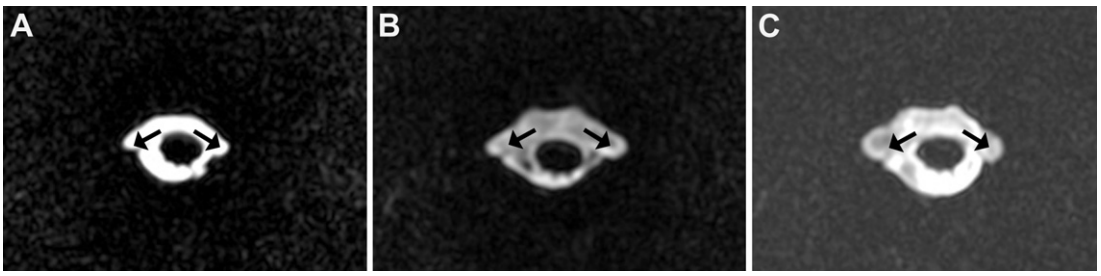

Fig. 2. Magnetic resonance myelography of CSF leakage. Heavily T2-weighted magnetic resonance myelography at the T6/7 level demonstrates spinal CSF leaks before EBP (arrow, A) and after EBP (before surgery) (arrow, B), and the CSF leaks remaining after surgery (arrow, C).

courses of EBP at different sites of spinal CSF leaks (C7–T1, T10–T11 and L2–L3, and T7–T8, respectively), and one continuous epidural saline infusion. Relief from the headaches continued for 3 months.

The patient remained free of headache after multiple EBPs. However, a follow-up brain MRI 3 months later revealed an SDH layer in the left-side cerebral hemisphere, 18 mm at the thickest point, with an evident shift of the midline structures. Apart from this SDH, all other signs of SIH on brain MRI had actually regressed (Fig. 1). The radiological improvement in SIH included subsidence of pachymeningeal enhancement, smaller size of the pituitary gland, and filling of the prepontine CSF space with CSF inside the ambient cisterns to regain a cushion appearance. In addition, the optic chiasm appeared to be free from compression and the cerebellar tonsils were restored to their normal position, above the foramen magnum. MR myelography still revealed similar epidural CSF collections at multiple sites of the spine. For this potentially critical condition, surgical intervention was recommended at the most prominent site of the CSF leak at T5–T7, which was the suspected primary site of this leakage. The patient then underwent thoracic laminectomy with direct repair of the dural defect at T5–T7. The procedures performed for repair included direct suture closure of the dural defect, coverage with a synthetic collagen matrix (DuraGen, Integra, Plainsboro, NJ, USA), and sealing with fibrin glue (Tissucol Duo Quick, Baxter AG, Vienna, Austria). The post-operative course was uneventful and the patient remained free of symptoms.

The patient was followed up for 3 years after surgery. SDH has not recurred, despite the fact that there was never direct surgical evacuation of the SDH, either by burrhole drainage or craniotomy. He has remained free of headache ever since. Serial follow-up brain MRI revealed complete resolution of the hematoma and signs of SIH (Fig. 1). However, epidural CSF collections were still observed on MR myelograms (Fig. 2).

3. Discussion

Although the exact etiology of SIH remains obscure, it has been reported that clinical manifestations and abnormal findings on neuro-images are primarily caused by CSF volume depletion, subsequently followed by two pathophysiological changes.[2,9] First, the brain sags due to loss of CSF buoyancy. Second, the intracranial blood volume increases to compensate for the CSF volume depletion according to the Monroe–Kellie doctrine.[1]

Brain sagging may lead to traction on the cranial nerves, cervical nerve roots, and pain-sensitive suspending structures, and consequently cause clinical symptoms of cranial nerve palsy, radicular pain, and orthostatic headaches. At the extreme, brain sagging might result in hindbrain herniation, compression of the optic chiasm over the pituitary fossa, and pressured pons against the clivus.

An increase in intracranial blood volume, hyperemia, may cause subdural fluid collection or venous hypervolemia. These also account for pachymeningeal enhancement, hyperemia of the pituitary gland, and vasodilation-related headache. The symptoms and signs are aggravated when the patient is upright because of exacerbation of brain sagging due to gravity. Therefore, postural headache is regarded as a hallmark of SIH.

The presence of SDH accompanying SIH has been reported.[4,7,10] It is estimated that 40% of subdural fluid collections consist of subacute to chronic SDHs associated with a significant mass effect.[6] Subdural hematoma could be a result of tearing of the neighboring bridging veins when the cerebral hemisphere shrinks due to decreased CSF volume.[5] The clinical deterioration may be due to progressive diencephalic herniation in the setting of brain sagging rather than development of SDH.[6,7] However, SDH has been observed in some patients whose symptoms actually improved while the SDH developed.[6,11] Therefore, Schievink et al. postulated that SDH development could have corrected the abnormally low intracranial pressure or volume by replacing CSF with the hematoma.[6,11] Therefore, it can be inferred that diencephalic or tonsillar herniation in the setting of brain sagging could actually be lessened by the development of SDH.

In the current case, the patient improved clinically after multiple EBPs. However, the improvement in symptoms did not correlate completely with the radiologic evidence. Although the signs of SIH on MRI showed significant remission, including the pachymeningeal enhancement, pituitary hyperemia, downward displacement of the cerebellar tonsils, flattening of the pons against the clivus, bowing of the optic chiasm against the pituitary fossa, and crowding of the ambient cistern and midbrain, a thick SDH layer was observed in the left cerebrum, with a shift of the midline structures. This phenomenon shed light on the possibility that SDH might play a positive role in the setting of intracranial hypovolume. The mass effect of SDH replenished the intracranial space, so there was no more need to increase the intracranial blood (hyperemia) as compensation. After surgical repair of the spinal dura defect, the intracranial hypovolume was corrected. Therefore, the signs of SIH completely regressed, including the SDH. Our imaging evidence not only strongly supports Schievink's postulation but also highlights the importance of identification of the primary cause, SIH.

It has been speculated that besides the decrease in spinal CSF volume, an altered distribution of craniospinal elasticity or increased compliance of the spinal CSF space can explain the mechanism of SIH.[4,12,13] On the basis of this hypothesis, Schievink et al. proposed a novel technique, called lumbar dural reduction surgery, to treat intractable SIH with an expected permanent therapeutic effect.[13] In the present case, we repaired the dura defect with direct sutures. The elasticity of the dura sac was also reduced to some degree. The patient recovered fully, in both clinical and radiological terms, after surgical repair of the dura. After 3-year follow-up, the patient remained well, although MR myelography continuously demonstrated spinal CSF leaks. The current case study illustrates that resolution of SIH does not require complete sealing of spinal CSF leaks, but perhaps a reduction in the speed of such leakage.

It is possible that headache due to SIH could be managed by multiple EBPs, but resolution of the headache could mask intracranial SDH. Although the spinal CSF leak was only partially repaired surgically, the SDH resolved and there was complete remission of SIH for 3 years. It is reduction rather than elimination of spinal CSF leaks that yields remission of SIH. In summary, intracranial SDH can be a complication of inadequately treated SIH (i.e., persistent minor CSF leakage). Therefore, management of SDH should focus on correction of the underlying SIH rather than craniotomy for hematoma evacuation.

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