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Management of an intracranial hypotension patient with diplopia as the primary symptom: A case report

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Abstract

BACKGROUND

Intracranial hypotension (IH) is a disorder involving cerebrospinal fluid (CSF) hypovolemia due to spontaneous or traumatic spinal CSF leakage and is easily being misdiagnosed or missed, especially in these patients without the prototypical manifestation of an orthostatic headache. At present, the management of IH with both cranial nerve VI palsy and bilateral subdural hematomas (SDHs) is still unclear.

CASE SUMMARY

A 67-year-old male Chinese patient complained of diplopia on the left side for one and a half mo. Computed tomography revealed bilateral SDHs and a midline shift. However, neurotrophic drugs were not effective, and 3 d after admission, he developed a non-orthostatic headache and neck stiffness. Enhanced magnetic resonance imaging revealed dural enhancement as an additional feature, and IH was suspected. Magnetic resonance myelography was then adopted and showed CSF leakage at multiple sites in the spine, confirming the diagnosis of having IH. The patient fully recovered following multiple targeted epidural blood patch (EBP) procedures.

CONCLUSION

IH is a rare disease, and to the best of our knowledge, IH with diplopia as its initial and primary symptom has never been reported. In this study, we also elucidated that it could be safe and effective to treat IH patients with associated cranial nerve VI palsy and bilateral SDHs using repeated EBP therapy.

Key Words: Cranial nerve VI palsy; Epidural blood patch; Intracranial hypotension; Subdural hematoma; Case report

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Core Tip: Intracranial hypotension (IH) is an uncommon disorder of cerebrospinal fluid (CSF) hypovolemia due to spontaneous or iatrogenic spinal CSF leakage, and is easily being misdiagnosed or missed as it can be associated with a large diversity of clinical signs. To the best of our knowledge, this is the first case in the literature describing a patient of having IH with diplopia as his initial and primary complaint, and demonstrating the efficacy of repeated epidural blood patch therapy in managing a patient with multiple sites of CSF leakage in the spine complicated with cranial nerve VI injury and subdural hematomas.

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INTRODUCTION

Intracranial hypotension (IH) is a disease being recognized with increasing frequency and is caused by decreased intracranial pressure (ICP), usually due to cerebral spinal fluid (CSF) leakage[1]. This case report describes the case of a 67-year-old male with diplopia as his primary symptom; the diagnosis of IH was missed until CSF leakage in the spine was found on magnetic resonance myelography (MRM), and the patient was successfully managed by multiple epidural blood patch (EBP) procedures. This is the first case to describe a patient with IH with diplopia as his initial and chief complaint and discuss the efficacy of using repeated EBP therapy to treat a patient with IH complicated with cranial nerve VI palsy and subdural hematomas (SDHs).

CASE PRESENTATION

Chief complaints

A 67-year-old Asian man complained of diplopia on the left side.

History of present illness

The patient's symptom started one and a half mo prior to presentation. However, after half a mo of conservative treatment for paralytic strabismus, his symptom did not improve.

History of past illness

The patient had a history of type 2 diabetes for 15 years, with well-controlled glucose. He suffered a stroke 10 years ago without sequelae and took sitagliptin regularly afterwards.

Personal and family history

No smoking or drinking history, and no similar family history were noted.

Physical examination

Physical examination revealed left lateral gaze palsy and horizontal diplopia, consisting of left abducens nerve paralysis, as well as weaker strength in the left upper limb (motor strength score of 4) and a positive Babinski sign.

Laboratory examinations

Blood analysis revealed an elevated total cholesterol of 5.57 mmol/L and a very low density lipoprotein of 0.96 mmol/L. The electrocardiogram showed a first-degree atrioventricular block and a conduction block in the left forearm. The microprotein level in CSF obtained from a lumbar puncture performed 2 wk after the third EBP

treatment was 950 mg/L. The Pandy test was positive. The ICP was normal.

Imaging examinations

The initial plain computed tomography (CT) scan revealed SDH, which resulted in referral of the patient to the Neurosurgery Department (Figure 1A). The patient suffered a sudden severe non-orthostatic headache with neck stiffness on day 3 after admission. Enhanced magnetic resonance imaging (MRI) of his head revealed dural enhancement as an additional feature (Figure 1B and C). He was then suspected of having IH. MRM was utilized and showed multiple sites of CSF leakage in the spine, confirming the diagnosis of IH (Figure 2).

MULTIDISCIPLINARY EXPERT CONSULTATION

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Since CSF leakage in the spine was confirmed and the patient had a stable condition, EBP therapy could be performed first.

FINAL DIAGNOSIS

The final diagnosis of this case was IH complicated with cranial nerve VI palsy and SDHs.

TREATMENT

He was treated with bed rest, hydration, neurotrophic drugs for treating cranial nerve VI palsy, cholesterol lowering and glycemic control medication after the confirmation of having IH. On day 15 after his admission, the patient underwent the first EBP procedure. An 18-G needle was inserted into his epidural space at the T2/3 level, and 21 mL of autologous peripheral venous blood was infused. The second EBP treatment was performed on day 22 after considering the progression of the SDHs, with 21 mL of blood being infused at the T8/9 level. According to the MRM results on day 30, the third EBP procedure was performed on day 32 using 22 mL of blood at the T5/6 level. He accepted the same drug treatment after each EBP procedure.

OUTCOME AND FOLLOW-UP

After the first EBP therapy, his headache and neck stiffness were markedly alleviated. On day 21, the follow-up CT revealed bilateral SDHs with greater density volume than the previous SDHs (Figure 3A). However, he had no complaints other than diplopia. This was followed by the second EBP therapy on day 22. The MRM of the spinal column on day 30 revealed a similar scan with no obvious decrease in CSF leakage (Figure 3B and C). Accordingly, the third EBP procedure was performed on day 32. To evaluate the curative efficacy of EBP therapy, CT myelography was employed on day 57 and showed no evident CSF leakage (Figure 3D). His symptom of diplopia was relieved 3 wk after the third EBP therapy. Over a follow-up period of 6 mo, none of the symptoms recurred, suggesting a favorable recovery.

Figure 4 summarizes the key events of this case report chronologically.

DISCUSSION

IH is a disorder of CSF hypovolemia due to spontaneous or traumatic spinal CSF leakage. The most common symptom of IH is a headache. The characteristic headache is orthostatic, aggravating in the upright posture and alleviating in the supine position [2]. In this patient, diplopia presented as the initial and primary complaint; it should be noted that this is the first reported case with this feature, which delayed making the

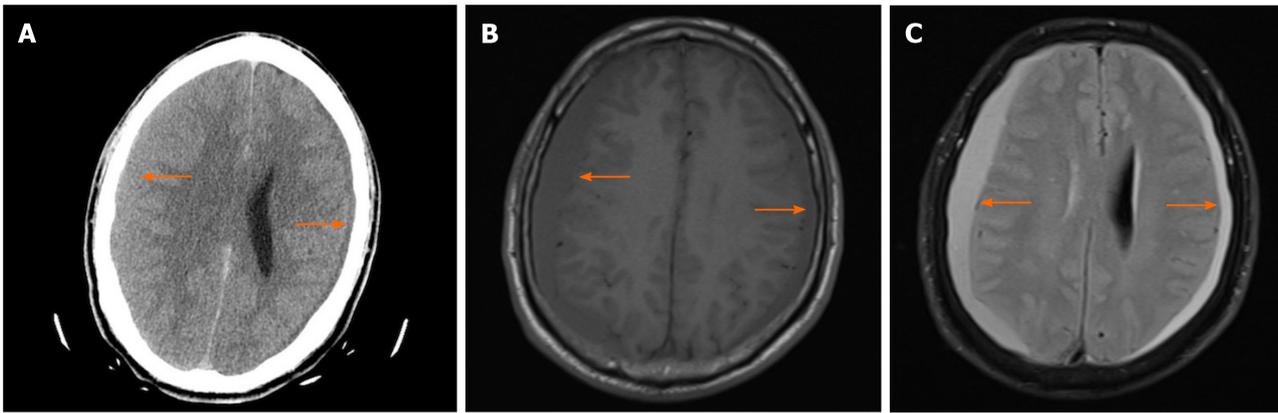


Figure 1 Axial brain image acquired before suspicion of having intracranial hypotension. A: Axial computed tomography image shows left-sided subdural effusion, right-sided subdural hematoma (orange arrows), compression of the right ventricle and midline shift on day 1; B: Axial T1-weighted magnetic resonance imaging (MRI) with gadolinium enhancement reveals diffuse pachymeningeal thickening and subdural hematomas (SDHs) on both sides on day 3 (orange arrows); C: Axial T2-weighted MRI with gadolinium enhancement reveals SDHs with high signal in the same area on day 3 (orange arrows).

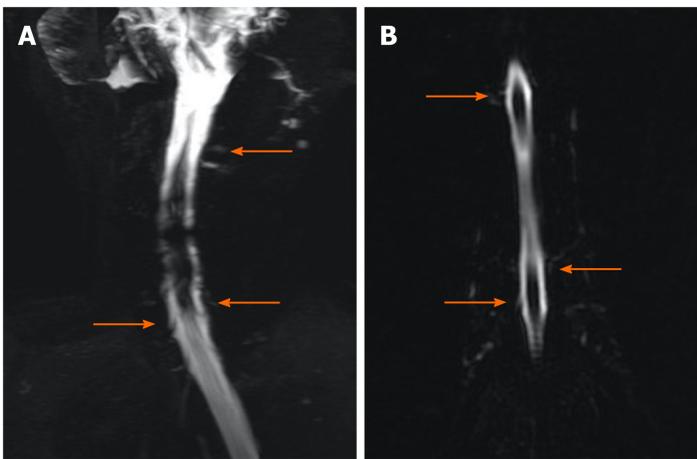


Figure 2 Magnetic resonance myelography imaging of the spine performed before epidural blood patch therapy. A: Magnetic resonance myelography (MRM) shows multiple sites of cerebrospinal fluid (CSF) leakage at cervicothoracic junctions on day 9 (orange arrows); B: MRM shows multiple sites of CSF leakage in the thoracic region on day 9 (orange arrows).

correct diagnosis and increased the difficulty of proper therapy. In this case, horizontal diplopia, which was worse in one direction of lateral gaze, occurred with an abduction deficit and presented as esodeviation. Possible etiologies of abduction deficits include conditions affecting the pons, cranial nerve VI, neuromuscular junctions, and extraocular muscles[3]. Specifically, this patient's diplopia was attributed to cranial nerve VI palsy derived from continuous CSF leakage. The sixth cranial nerve is the most frequently injured because of its long intracranial path. MRI has shown that continuous CSF leakage leads to IH with descent of the brain, causing traction of the sixth nerve and subsequent symptoms[4].

The presence of SDH was another characteristic feature in this case. SDH is a severe complication of IH that may lead to neurological deficits and can be life-threatening [5]. SDH associated with IH is considered to be secondary to the collapse of the intrathecal space due to CSF leakage[6]. The reported incidence of SDH among spontaneous IH patients ranges from 16% to 57%, prevailingly in males[7], whereas spontaneous IH without SDH is predominant in females and the average onset age of spontaneous IH with SDH is 44.9 years[8,9].

The association between SDH and abducens nerve palsy is rarely mentioned in the literature. These two complications were related to descent of the brain, causing traction of cranial nerves with a long intracranial course and on intracranial vessels. This traction leads to the avulsion of dural veins and subsequent subdural bleeding [10].

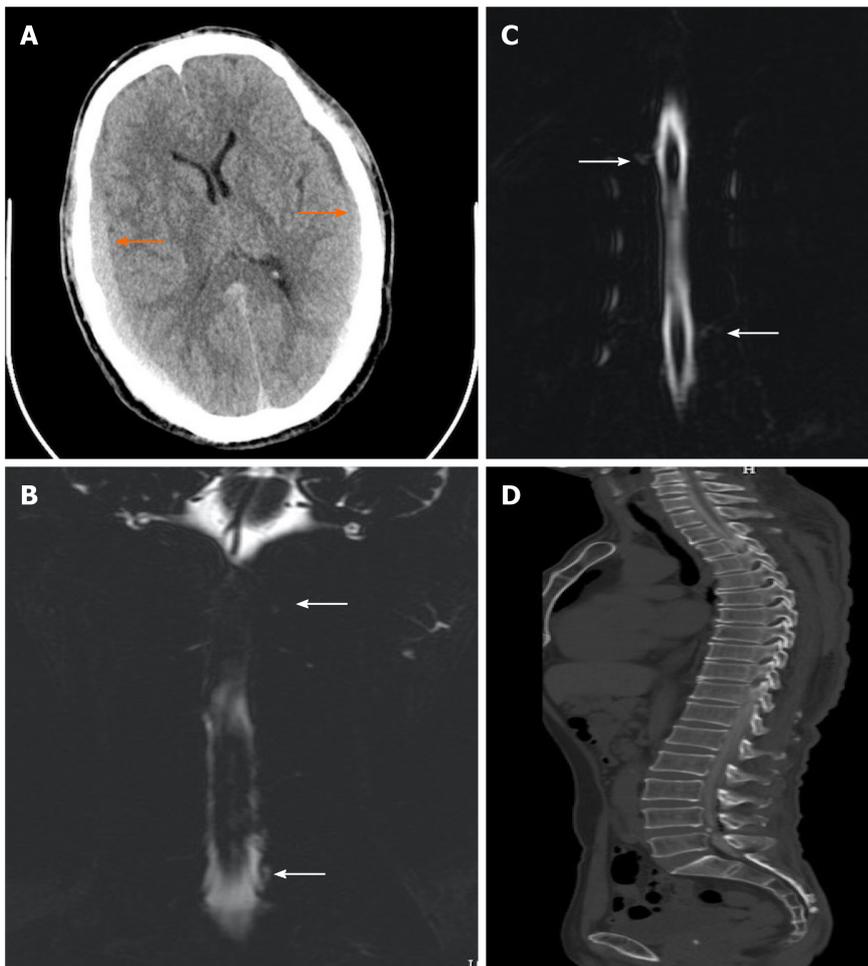


Figure 3 Images of the brain and spine acquired after the epidural blood patch therapy. A: Axial computed tomography (CT) image shows bilateral subdural hematomas with greater density and volume than the previous hematomas on day 21 after the first epidural blood patch (EBP) therapy (orange arrows); B: Magnetic resonance myelography (MRM) shows multiple sites of cerebrospinal fluid (CSF) leakage at cervicothoracic junctions on day 30 after the second EBP therapy (white arrows); C: MRM shows multiple sites of CSF leakage in the thoracic region on day 30 after the second EBP therapy (white arrows); D: Sagittal CT myelography imaging of the entire spine reveals no obvious CSF leakage on day 57 after the third EBP therapy.

MRI has facilitated the recognition of IH. The typical imaging features of IH on MRI include subdural fluid collections, engorgement of venous structures, pachymeningeal enhancement, pituitary hyperemia and sagging of the brain. Although not as conclusive as MRI, CT can be of significant diagnostic value in detecting SDH, particularly in the emergency department setting[11]. In this case, repeated CT was used to detect SDH and monitor the intracranial lesions, facilitating the diagnosis and treatment of IH. If there is a high clinical suspicion of IH but normal MRI or CT findings, radioisotope cisternography or CT myelography may be the next modality of choice to pinpoint sites of CSF leakage. Furthermore, MRM is a developing cutting-edge technology that was used in our case, serving as an alternative to CT myelography. This male patient presented with diplopia and a non-orthostatic headache. MRM of the entire spinal column revealed multiple sites of CSF leakage, and brain CT showed bilateral SDHs. Hence, the diagnosis of IH complicated with cranial nerve VI paralysis and SDHs was clear in this patient.

The treatment strategy for IH consists of conservative schemes such as bed rest, intravenous hydration, caffeine intake and invasive therapies, including EBP therapy and surgery. However, there is a lack of systematic studies on therapeutic methods and outcomes in IH patients with both cranial nerve VI palsy and SDH. We provide a review of published reports describing IH as an explicit cause of SDH and abducens nerve paralysis (Table 1). As shown in the chart, conservative treatment always fails to yield persistent improvement in such patients, and currently, whether to perform surgical SDH evacuation or EBP therapy as the initial procedure is still controversial [12,13]. Through the review of those reports above and other published reports, we believe that IH patients with both SDH and cranial nerve injury require emergency surgery under the following conditions: Moderate to large hematoma causing brain

Table 1 Summary of reported cases of intracranial hypotension with subdural hematoma and diplopia

Case No.	Ref.	Age in yr/sex	Presenting symptoms			Positive imaging findings	Subdural hematoma eva. and outcome	IH treatment	Outcome
			Postural headache	Diplopia	Consciousness disturbance				
1	Takahashi <i>et al</i> [6], 2016	49/M	√	√	√	BSDH, DME, DOCT, NOC	√ × 2, trans imp	EBP × 2	Near FR
2	Veeravagu <i>et al</i> [17], 2013	36/M	√	√	√	Spur, DME, DOOC, SOT, Large BSDH, MS	√ + osteophyte repair	EBP, trans imp	Near FR
3	Fiala <i>et al</i> [12], 2012	28 /F	√	√		BSDH, thrombosis after EBP ¹		EBP × 1	FR
4	Slowinski <i>et al</i> [20], 2003	38 /F	√	√		Thick BSDH	√		FR
5	Whiteley <i>et al</i> [13], 2003	62/M	√	√	√	Large BSDH, DME	√ × 2, trans imp	EBP × 1	FR
6	Velarde <i>et al</i> [10], 2000	58/M	√	√		Thin BSDH		EBP × 1	FR
7	Welch <i>et al</i> [22], 1959	69/M	NP H/A	√	√	NA	√ ²		Dead

¹Low molecular heparin therapy after epidural blood patch.

²Bilateral subdural blood was found during the operation.

BSDH: Bilateral subdural hematomas; DME: Diffuse meningeal enhancement; DOCT: Descent of the cerebellar tonsil; DOOC: Draping of the optic chiasm; EBP: Epidural blood patch; Eva: Evacuation; F: Female; FR: Full recovery; IH: Intracranial hypotension; M: Male; MS: Midline shift; NA: Not available; NOC: Narrowing of the cistern; NP H/A: Non-positional headache; SDH: Subdural hematoma; SOT: Sagging of the tons; trans imp: Transiently improved.

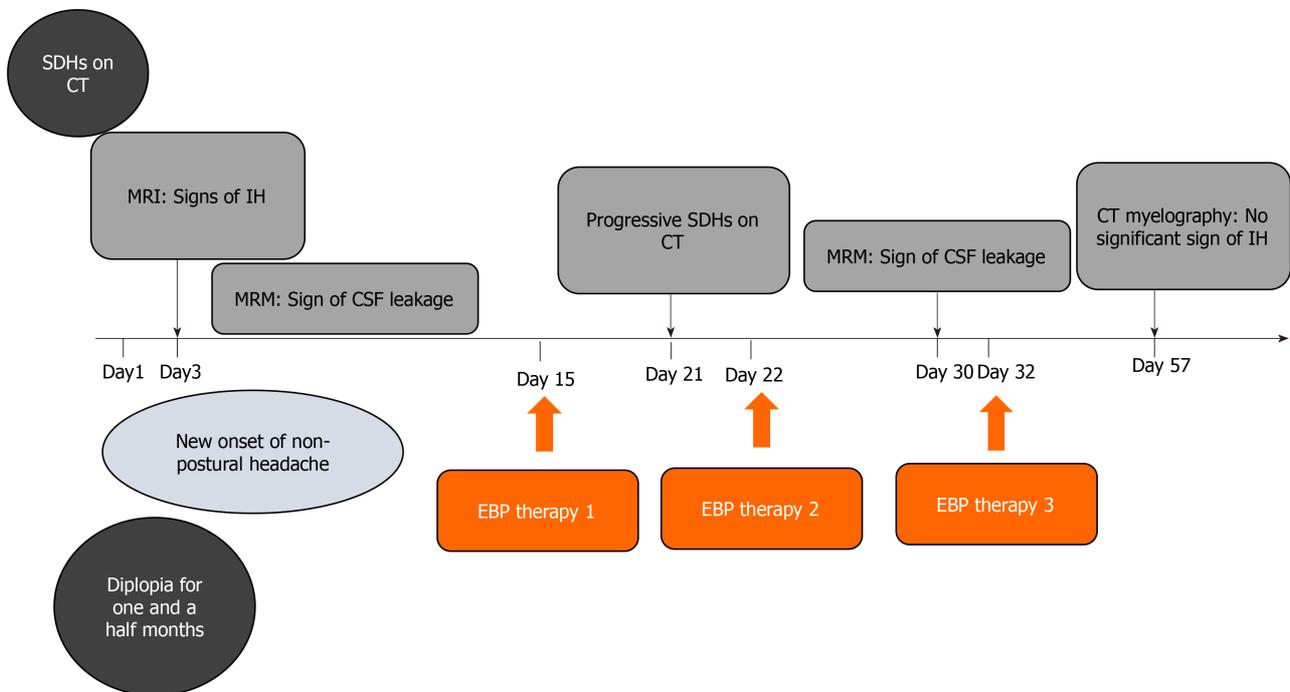


Figure 4 shows the medical history chronologically. CSF: Cerebrospinal fluid; CT: Computed tomography; EBP: Epidural blood patch; IH: Intracranial hypotension; MRI: Magnetic resonance imaging; MRM: Magnetic resonance myelography; SDH: Subdural hematoma.

hernia and neurological deterioration[14], failure to improve after applying the Trendelenburg position in comatose SDH patients with a mass effect and pupil dilation[6], and progressive SDH or rapid cognitive deterioration after EBP therapy[15, 16]. Furthermore, surgical repair is indispensable in patients with evidence of cranial

or spinal anatomic abnormalities (*i.e.* osteophytes, arachnoid cyst congenital abnormalities, *etc.*) to prevent the recurrence of IH[9,17]. In terms of this case, we gave preference to repeated EBP therapy due to these reasons. First, EBP therapy is proven to be effective in relieving symptoms in 90% of cases, and if symptoms can be ameliorated after EBP therapy, even a thick SDH could resolve spontaneously[18,19]. Second, conservative treatment failed in our patient, and although the follow-up CT image showed SDH progression after the first EBP procedure, his symptoms did not worsen. We decided to perform a second EBP procedure in consideration of his stable conditions and multiple sites of CSF leakage. In addition, although we found a case of recovery after surgical evacuation without EBP therapy[20], many authors believe that empirical EBP therapy after surgical evacuation could treat the underlying cause of SDH and therefore minimize the risk of relapse[5,6,21]. Moreover, our literature search yielded a report of mortality after surgery and mentioned the possibility of pneumonia and cerebral infarction as postoperative complications[22]. Loya *et al*[5] summarized that in most cases, surgical correction is not inevitable and may bring about deteriorated outcomes[5]. Thus, EBP therapy may be a priority since it is much safer, with less adverse reactions. Last, compared with injecting blood into multiple targeted sites of CSF leakage at one time, putting the patient at a risk of deterioration after EBP therapy due to ICP elevation[16], it is safer to perform repeated targeted EBP procedures after some interval of time with careful follow-up.

CONCLUSION

When a patient presents with diplopia and SDH on CT, the clinician should perform a comprehensive examination to search for even subtle evidence of IH. Once IH is confirmed, EBP therapy may be a priority in such patients, except under emergency conditions necessitating surgical evacuation.

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