Spontaneous intracranial hypotension with bilateral subdural hemorrhage: Is conservative management adequate?

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Abstract

The aim of this study is to report a case of spontaneous intracranial hypotension complicated by bilateral subdural hemorrhage that resolved with conservative management. A young male presented with severe orthostatic headache associated with dizziness, neck pain and diplopia. Brain imaging revealed characteristic pachymeningeal enhancement and bilateral subdural hemorrhage. Radionuclide cisternography confirmed the Cerebrospinal fluid leak at the cervical 5 and cervical 6 vertebral level. He had clinical and radiological resolution with bed rest, hydration and analgesics and has remained symptom free since then. Spontaneous intracranial hypotension may be complicated by bilateral subdural hemorrhage. A conservative treatment approach is a viable option, as it may help improve the clinical and radiological outcome, especially when interventional facilities are not available.

Key Words

Intracranial hypotension, pachymeningeal enhancement, subdural hemorrhage

Introduction

The syndrome of spontaneous intracranial hypotension (SIH) is a single pathophysiological entity of diverse origin. Usually, it is characterized by an orthostatic headache that occurs or worsens with upright posture and is relieved by recumbency, although patients with chronic headaches or even no headache have been described.[1] In addition, patients with SIH may also experience dizziness, nuchal pain, nausea, vomiting, diplopia or blurring of vision.[2-4] This entity was first described in 1938 by George Schaltenbrand.[5] In the 90s, Magnetic resonance imaging (MRI) brain made a major breakthrough in the diagnosis of SIH with characteristic diffuse, non-nodular T1-weighted pachymeningeal thickening and enhancement. We report a patient who had clinical and neuroimaging features that were characteristic of SIH, and later developed bilateral subdural haemorrhage; however, improved with conservative management.

Case Report

A 35 year old Chinese man experienced severe generalized headache over a couple of days. The headache was throbbing in nature and maximum in the posterior parietal regions. In addition, he also complained of double vision, neck pain and dizziness; however, denied having nausea or vomiting. The headaches were aggravated when he assumed an upright posture (sitting or standing) and alleviated when lying down. He did not have preceding history of strenuous physical activities such as heavy weight lifting, trauma or a lumbar puncture procedure. Neurological examination revealed bilateral mild restriction of lateral gaze (likely due to VI nerve involvement), which resulted in binocular diplopia on extreme horizontal gaze. His brain computed tomography with contrast was normal. MRI brain showed prominence of cortical veins and significant distension of dural venous sinuses raising the possibility of pachymeningitis without venous sinus thrombosis [Figure 1 a,b]. In addition, cervical spine MRI revealed engorged anterior epidural venous plexus at cranio cervical junction and upper cervical spine, with mild effacement of the subarachnoid space, raising the possibility of spontaneous intracranial hypotension secondary to cerebrospinal fluid (CSF) leak; however, no nerve root diverticula was noted. Repeated lumbar puncture (LP) failed to extract any CSF. Subsequently, LP under fluoroscopic guidance confirmed a low CSF pressure (5 cm of water). The routine CSF investigations for infective and inflammatory markers were all negative.
He was treated conservatively with strict bed rest, adequate hydration and analgesia. A radionuclide cisternography showed CSF leak at the junction of cervical 5 and cervical 6 (C6/C7) vertebrae [Figure 2]. Treatment options including targeted epidural blood patch (EBP) were discussed; however, the patient opted for conservative management. He was discharged after a week. We had advised him to rest at home for another 2 weeks; however, he resumed normal activities on the very next day. Three days later, he presented again with severe headaches and worsening of diplopia. A repeat MRI brain showed bilateral subdural hematoma (SDH) [Figure 3]. Once again options of treatment were discussed with the patient and he decided for conservative management. He was continued on intravenous hydration, tablet caffeine and strict bed rest. After 2 weeks his headaches resolved completely and he was discharged well. He had further rest at home and resumed his routine duties 6 weeks after hospital discharge. He remained well and a follow-up CT scan done 3 months after discharge showed complete resolution of SDH. There has been no recurrence of symptoms since the last 14 months of follow-ups.

**Discussion**

Intracranial hypotension is a relatively uncommon but important cause of headache. Improvements in neuroimaging techniques have led to an increase in the identification rate. However, some patients may be misdiagnosed as migraine, tension headache, and viral meningitis. The diagnostic criteria for headache due to SIH, established by the International Headache Society in 2004, include diffuse or dull headache that worsens within 15 minutes of sitting or standing, and is associated with symptoms of neck stiffness, tinnitus, photophobia, nausea and hyperacusia. A trivial trauma may precede the onset of symptoms. More recently, the diagnostic criteria have been revised to include the broad spectrum of clinical presentations. The revised criteria include four diagnostic components [Table 1]. The typical MRI findings in these patients are diffuse pachymeningeal T2-weighted hyperintensity and gadolinium enhancement as well as evidence of descent of the brain.

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**Figure 1:** (a) Post gadolinium contrast enhanced axial T1-weighted Magnetic resonance imaging of the brain shows diffuse non-nodular enhancement and thickening of the pachymeninges. (b) Post gadolinium contrast enhanced coronal T1-weighted Magnetic resonance imaging of the brain shows diffuse non-nodular enhancement and thickening of the pachymeninges

**Figure 2:** Radionuclide cisternography reveals cerebrospinal fluid leak at C6/7 level on right side paucity of activity in cerebral convexity, slow ascent of radionuclide along the spinal axis and early accumulation of the radionuclide in kidneys and urinary bladder

**Figure 3:** Follow up Magnetic resonance imaging of the brain shows acute subdural haemorrhage bilaterally susceptibility weighted images
Table 1: Diagnostic criteria for spontaneous spinal CSF leak and intracranial hypotension

| Criterion A | Demonstration of a spinal CSF leak (i.e. presence of extradural CSF), or if Criterion A not met, no sign of intracranial hypotension. |
|------------|----------------------------------------------------------------------------------------------------------------------------------|
| Criterion B | Cranial MR imaging changes of intracranial hypotension (i.e. presence of subdural fluid collection, enhancement of the pachymeninges or sagging of the brain), or in the absence of Criterion A. |

and the presence of at least one of the following:

1. Low opening pressure (< 60 mm H₂O)
2. Spinal meningeal diverticulum;
3. Improvement of symptoms after epidural blood patching;

or if criteria A and B not met:

Criteron C; the presence of all of the following or at least two of the following if typical orthostatic headaches are present:

1. Low opening pressure (< 60 mm H₂O)
2. Spinal meningeal diverticulum;
3. Improvement of symptoms after epidural blood patching;

Our patient had orthostatic headache along with neck pain, blurred vision, diplopia and was relieved by lying down. The brain MRI showed diffuse pachymeningeal hyperintensities on Fluid attenuated inversion recovery (FLAIR) images and enhancement on post gadolinium studies, indicative of SIH. Cervical MRI showed extradural collection of fluid on the ventral side of the cord as well as dural enhancement which was suggestive of CSF leak. A radionuclide cisternography was carried out and it confirmed the lateral CSF leak at C6/7, C7 vertebral level. Radionuclide cisternography is useful for better diagnosis while planning the management of SIH, and it may reveal the direct signs of CSF leakage in up to 80% of the patients. The management of SIH includes conservative treatment by hydration and bed rest as it is believed that the supine position reduces CSF pressure at the site of leakage, therefore allowing healing of the underlying meningeal defects. Epidural Blood Patch (EBP) remains the treatment of choice for complicated SIH. An EBP has to be carried out close to the region of the CSF leak for it to be effective (also called targeted EBP); however, a remote EBP distal to the CSF leak also has been shown to cause remission of headaches. EBP is a relatively safe procedure, and the contraindications include systemic sepsis, coagulopathy and local infection at the puncture site.

In our patient, EBP was not performed as he did not consent. Headache recurred after several days. Although the exact mechanism for the recurrence of SIH symptoms is debatable, we postulate that it is likely because of inadequate rest and early return to normal activities by our patient. This may have triggered a repeat damage and CSF leakage through the dura mater at the same site, which was likely not healed completely, and thus not firmly sealed. During the second admission, the SIH was complicated by SDH, which is a known complication in about 50% of cases. In general, SDH has high mortality (around 55%) and functional recovery after surgical drainage has been reported to be around 33%. SIH complicated by SDH is commonly treated with EBP; however, there are case reports which have described the successful burr hole drainage of hematomas. In our patient, a more lengthy conservative approach was adopted which resulted in complete resolution of symptoms and radiological findings. From our literature review, we believe that this is the first case of SIH complicated by bilateral subdural hemorrhage, which improved without any intervention. Moreover, the recurrence rate of SIH has been reported to be 28% in all patients and 38% in surgically repaired cases; however, our patient experienced no recurrence of low pressure headache in the last 14 months of follow-up.

In conclusion, we suggest that SIH complicated by SDH may be managed conservatively, especially if interventional facilities are lacking or refused by the patient. If a conservative approach is adopted, the treatment period would become relatively longer, which would buy time for the effective spontaneous closure of the CSF leakage.

References

1. Mokri B. Spontaneous cerebrospinal fluid leaks: From intracranial hypotension to cerebrospinal fluid hypovolemia—evolution of a concept. Mayo Clin Proc 1999;74:1113-23.
2. Mokri B, Piepgras DG, Miller GM. Syndrome of orthostatic headache and diffuse pachymeningeal gadolinium enhancement. Mayo Clin Proc 1997;72:400-13.
3. Su CS, Lan MY, Chang YY, Lin WC, Liu KT. Clinical features, neuroimaging and treatment of spontaneous intracranial hypotension and epidural blood patch. Eur Neurol 2009;61:301-7.
4. Turgut N, Ulu E, Hamamcioglu MK, Guldenik B, Albayram S. Postural tremor as a manifestation of spontaneous intracranial hypotension. J Clin Neurosci 2010;17:255-7.
5. Schalltenbrand G. Neuere anschauungen zur pathophysiologie der liquozirkulation. Zentralbl Neurochir 1938;3:290-300.
6. Schievink WI, Maya MM, Louy C, Moser FG, Tourje J. Diagnostic criteria for spontaneous spinal CSF leaks and intracranial hypotension. AJNR Am J Neuroradiol 2008;29:853-6.
7. Hyun SH, Lee KH, Lee SJ, Cho YS, Lee EJ, Choi JY, et al. Potential value of radionuclide cisternography in diagnosis and management planning of spontaneous intracranial hypotension. Clin Neurol Neurosurg 2008;110:657-61.
8. Schievink WI, Maya MM, Moser FG, Tourje J. Spectrum of subdural fluid collections in spontaneous intracranial hypotension. J Neurosurg 2005;103:608-13.
9. Hataashita S, Koga N, Hosaka Y, Takagi S. Acute subdural hematoma: Severity of injury, surgical intervention, and mortality. Neurol Med Chir 1993;33:13.
10. Yamakawa H, Murase S, Tanigawara T. Spontaneous intracranial hypotension associated with chronic subdural hematoma successfully treated with burr hole irrigation alone. Neurosurg Q 2010;20:273-6.
11. Schievink WI, Maya MM, Riedinger M. Recurrent spontaneous spinal cerebrospinal fluid leaks and intracranial hypotension: A prospective study. J Neurosurg 2003;99:840-2.

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