**CASE REPORT**

**Bilateral Subdural Hematomas Caused by Spontaneous Intracranial Hypotension**

Hsin-Hung Chen1,4*, Chun-I Huang1,4, Shu-Shya Hseu2,4, Jiing-Feng Lirng3,4

1Department of Neurosurgery, The Neurological Institute, and Departments of Anesthesiology and Radiology, Taipei Veterans General Hospital, and 4National Yang-Ming University School of Medicine, Taipei, Taiwan, R.O.C.

Patients with both spontaneous intracranial hypotension (SIH) and subdural hematomas (SDH) are frequently undiagnosed. SIH may recur very often over a short interval or result in disastrous consequences if only the SDH is dealt with. We report a young adult with severe posterior nuchal pain; brain computed tomography showed bilateral SDH. He was discharged smoothly without any neurologic deficit after epidural blood patches were applied after proper and timely diagnosis.

Patients with SIH complicated by SDH should not be overlooked. When patients complain of typical orthostatic headache without any history of trauma, SIH should be highly suspected. The therapeutic strategy for this type of SDH is different from those without SIH. We review the literature on the disease. [J Chin Med Assoc 2008;71(3):147–151]

Key Words: intracranial hypotension, orthostatic headache, subdural hematoma, surgical intervention

**Introduction**

Spontaneous intracranial hypotension (SIH) is caused by spontaneous cerebral spinal fluid (CSF) leakage of unknown etiology at the level of the spine. Patients present with a new headache that occurs shortly after assuming an upright position and is relieved by lying down (orthostatic headache). Although such a positional headache pattern is well-known following a lumbar puncture, SIH is not well recognized, and the patient may be diagnosed with migraine, tension headache, viral meningitis, or malingering. The spontaneous form of intracranial hypotension was first described in 1938, and much has been learned about this syndrome, particularly since the early 1990s, but frequent initial misdiagnosis remains.1 Unfamiliarity with SIH among physicians in general and the unusually varied spectrum of clinical and radiographic manifestations may all contribute to a delay in diagnosis for months or even years.

In most cases, SIH patients have typical orthostatic headaches that generally occur or worsen within 15 minutes of assuming the upright position. The associated symptoms are neck stiffness, tinnitus, hypacusia, photophobia, and nausea. The CSF pressure is low (< 60 mmH2O or even negative). Magnetic resonance imaging (MRI) findings are diffuse pachymeningeal enhancement, subdural fluid collections, engorgement of venous structures, pituitary hyperemia, and sagging of the brain.1

Most SIH patients recover after hydration or application of epidural blood patches (EBP). If left untreated, some SIH patients may encounter the complication of subdural hematoma (SDH) and may easily develop neurologic deficits such as cranial nerve palsy,2,3 frontotemporal dementia,4 parkinsonism,5 or cerebral sinus venous thrombosis.6 If only the SDH is treated without diagnosis of SIH, it may result in severe consequences such as coma.7

We report a SIH patient with bilateral SDH who came to our hospital and was discharged 3 weeks later without any neurologic deficit after EBP treatment; there was also no residual or recurrence at the 1-year follow-up. The therapeutic timing, modality, and role of surgical intervention are discussed and the literature is reviewed.
Case Report

A 27-year-old male interior designer was well until he developed sudden onset of posterior neck pain radiating to bilateral shoulders after waking up on the morning of July 15, 2006. He denied any history of recent trauma. The pain became intolerable at noon after he had been sitting for several hours during work, with the pain extending to the occipital and forehead region. He discovered that the symptoms were relieved within 1–2 minutes after lying down. He went to a hospital’s emergency department for help and lumbar puncture was suggested there, but he refused. Brain computed tomography (CT) was performed instead, which showed a normal result. After intravenous hydration and medication, he was discharged without a definite diagnosis. He noticed, however, that if he maintained an upright position for more than an hour, the headache returned.

The symptoms fluctuated thereafter. The posterior nuchal pain was exacerbated 1 week before the patient visited our outpatient department on August 21, 2006; even lying down did not relieve the pain. Physical and neurologic examinations were normal. Brain CT showed bilateral subacute SDH (1 cm in thickness) with obliteration of CSF space (Figure 1). He was admitted on the same day under the impression of SIH. Brain MRI with gadolinium contrast demonstrated diffuse pachymeningeal enhancement, SDH (high signal intensity in both T1- and T2-weighted images), and brain descent with obliteration of basal cisterns, but absence of midline shift (Figure 2). Therefore, a diagnosis of SIH was made.

Conservative treatment of intravenous hydration and bed rest were given initially. Whole-spine heavily T2-weighted MR myelography revealed CSF along the left nerve root at the level of T2 and epidural fluid collection from C2 to the lumbar region (Figure 3A). Two applications of targeted EBPs of 20 mL autologous blood via right side T2 level and 30 mL autologous blood via left side T4 level were performed on the 8th and 10th days of hospitalization, respectively. The previous symptoms of neck pain and headache subsided after the 11th day, and the patient felt even better when he was in the upright position. Follow-up 2 weeks later with MR myelography demonstrated disappearance of fluid budding from epidural space of T2–T4, left side (Figure 3B). The patient was discharged without any neurologic deficit on the 20th day of hospitalization. The SDH subsided completely, and there was no recurrence detected on brain CT 4 months later (Figure 4).

![Figure 1](image1.png)  ![Figure 2](image2.png)  ![Figure 3A](image3A.png)  ![Figure 3B](image3B.png)

**Figure 1.** Serial axial computed tomography images of the brain show bilateral subdural hematomas, small size of the ventricular system and obliteration of the basal cistern. The latter 2 findings of spontaneous intracranial hypotension mimic those of increased intracranial pressure.
Discussion

With physician awareness of the disease and improvement of diagnostic tools, SIH is half as common as spontaneous subarachnoid hemorrhage, for an estimated annual incidence of 5 per 100,000.8 SIH was probably more frequently underdiagnosed previously than it is now, and it is unlikely that there has been an actual increase in its incidence.

SIH usually affects women more frequently than men, with a female-to-male ratio of approximately 2:1.1 Compared to patients with chronic SDH or even recurrent chronic SDH, it has a reversed epidemiologic female-to-male ratio of 1:2.9 Onset of symptoms typically occurs in the 4th or 5th decade of life, with a peak incidence around 40 years of age, but children and elderly persons may also be affected. We think that perhaps some cases of recurrent chronic SDH are misdiagnosed.
SIH is caused by spontaneous spinal CSF leaks. The precise cause of spontaneous spinal CSF leaks remains largely unknown, but an underlying structural weakness of the spinal meninges is generally suspected. A history of a more or less trivial traumatic event (e.g., chronic cough, lifting heavy objects) preceding the onset of symptoms can be elicited in about a third of patients, indicating a role of mechanical factors. There is some good evidence to suggest that a generalized connective tissue disorder plays a crucial role in the development of spontaneous spinal CSF leaks, such as Marfan syndrome, Ehlers-Danlos syndrome type II, and autosomal dominant polycystic kidney disease. Based on physical examination alone, evidence of an underlying generalized connective tissue disorder is found in about 2 thirds of patients.

As in our patient, brain CT alone cannot be used to confirm the diagnosis of SIH since the shrinkage of ventricle size and obliteration of basal cisterns cannot be differentiated from signs of increased intracranial pressure. Along with the typical symptoms of SIH, it alerted us for admission and further study. Nonetheless, brain CT is useful for detection of complicated SDH.

The characteristic MRI features of SIH make it the imaging modality of choice for early definite diagnosis, which prevents the risk of cerebral herniation caused by invasive procedures, such as lumbar puncture for myelogram or intracranial pressure monitoring. Further spinal MRI myelogram may be used to show the ongoing leakage sites and the extent of the CSF leaks, which make targeted EBP or percutaneous placement of fibrin sealant possible to perform. However, CT myelography is still considered the study of choice if CSF leakage is to be located. Radionuclide cisternography is usually helpful when diagnosis of SIH is in doubt and myelography results are normal.

SIH may sometimes resolve spontaneously, and conservative treatment of bed rest, oral hydration, a generous caffeine intake, and use of an abdominal binder is also effective in many patients, but it is time-consuming and the recurrence rate is high. Administration of steroids, intravenous caffeine, or theophylline have all been advocated as specific treatments for SIH, but their effectiveness is limited. Osmotic or loop diuretics are not advocated as specific treatments for SIH, but their effectiveness is limited. Osmotic or loop diuretics are not effective in such cases and may deteriorate the condition.

The mainstay of treatment is the injection of autologous blood (10–20 mL) into the spinal epidural space, the so-called EBP. Relief of symptoms is often dramatic after EBP. If EBP fails the first time, it can be repeated. Sometimes, nonsurgical measures do not help at all; surgical repair of CSF leak is safe and often succeeds when a structural abnormality (such as meningeal diverticula) or focal CSF leak is identified. Intrathecal infusion of saline or artificial CSF may be required as an effective temporizing measure to restore CSF volume until the leak can be permanently repaired in patients who require urgent treatment.

There is no consensus regarding the management of SDH caused by SIH, but we still have to emphasize the difference between SDH and hygroma. The subdural hygroma itself seldom has a mass effect, and surgical evacuation is potentially harmful. But it predisposes to rupture the cortical veins crossing the subdural space of SIH patients, which may predispose to the development of SDH. In 1 case series, half of the 40 SIH patients had subdural collections. All of them recovered well after the CSF leaks were secured, including 3 who underwent surgical drainage of SDH at first. None of their SDHs resolved until the underlying CSF leak was treated by either EBP or surgical repair. Another case series supported the surgical drainage of SDH as being necessary and indicated in select cases. We suggest following the surgical indications of chronic SDH (focal neurologic deficits, decreased level of consciousness, or SDH with maximum thickness > 1 cm) after the diagnosis of SIH is established. In our patient, the thickness of SDH was 1 cm without any neurologic deficit, so there was no emergent surgical indication for drainage. It is important to treat CSF leakage and restore CSF volume earlier in patients with SIH and SDH to prevent irreversible sequelae. A straightforward case such as one with bilateral SDH can turn into a nightmare if presurgical history and radiologic examination are not reviewed thoroughly.

The diagnosis of SIH should be considered in young patients presenting with SDH in the absence of trauma and with normal clotting, particularly in those who may experience recurrence following drainage.

References


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