

Spontaneous Intracranial Hypotension Manifesting as a Unilateral Subdural Hematoma with a Marked Midline Shift

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Key Words

Blood patch · Midline shift · Spontaneous intracranial hypotension · Subdural hematoma, unilateral

Abstract

Spontaneous intracranial hypotension (SIH) is a syndrome in which hypovolemia of the cerebrospinal fluid (CSF) results in various symptoms. Although its prognosis is usually benign, cases with a rapid neurologic deterioration resulting in an altered mental status have been reported. One of the characteristic radiographic findings in such cases is the presence of bilateral accumulation of subdural fluid (hematoma/hygroma). When SIH-related subdural hematoma is present only unilaterally with a concomitant midline shift, making an accurate diagnosis may be challenging, and inadvertent hematoma evacuation may result in further neurologic deterioration. We report a 58-year-old woman with an altered mental status who had visited a local hospital and in whom a brain CT showed a unilateral subdural hematoma with a marked midline shift. She was referred to our department because of her neurologic deterioration after hematoma evacuation. A CT myelography revealed a massive CSF leakage in the entire thoracic epidural space. She made a full neurologic recovery following blood patch therapy. Our case is unique and educational because the suspicion for SIH as an underlying cause of subdural hematoma is warranted in nongeriatric patients not only with bilateral but also unilateral lesions. An immediate search for CSF leakage may be important in cases with failed hematoma evacuation surgery.

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Introduction

Spontaneous intracranial hypotension (SIH) is a syndrome in which hypovolemia of the cerebrospinal fluid (CSF) results in various symptoms [1–3]. The progression of symptoms is usually gradual and most patients seek medical attention complaining of chronic orthostatic headaches [1–3]. Although rare, patients may sustain a rapid neurologic deterioration and are brought to an emergency department [4–6]. On imaging studies, the great majority of those presenting with an altered mental status exhibit a bilateral accumulation of subdural fluid or hematoma, which is an important diagnostic clue [3–5]. However, when SIH-related subdural hematoma is only present unilaterally with a concomitant midline shift, diagnosing SIH may be difficult and inadvertent hematoma evacuation may result in further neurologic deterioration. We report a rare case of SIH with a unilateral subdural hematoma with a marked midline shift which posed a diagnostic challenge to neurosurgeons.

Case Report

A 58-year-old woman without a history of chronic headache suddenly experienced back pain during stretching exercises. Shortly afterwards, she began to complain of a headache which was aggravated by standing up and relieved by lying down. A local physician thought that she had a migraine and prescribed triptans. However, her headache became constant regardless of her posture, and within a month after the initial back pain, she became stuporous and was brought to a local emergency department. She was E2V3M5 on the Glasgow Coma Scale, and mild left-sided hemiparesis was noted on neurologic examination. Her pupils were reactive and isocoric. A brain CT revealed a right chronic subdural hematoma with a marked right-to-left midline shift (fig. 1a). A neurosurgeon at that hospital thought that she was suffering from an impending transtentorial herniation and performed a small craniotomy to evacuate the hematoma. No brain swelling was observed after dural opening and hematoma evacuation, and no chronological improvement in the degree of the midline shift was seen on her postoperative CT scan (fig. 1b). After a transient improvement, her consciousness level further deteriorated to E1V1M3 4 days after surgery. She was then referred to our institution; a third CT scan obtained immediately before her transfer showed a worsening of the midline shift and an increase in her subdural hematoma (fig. 1c).

A lack of neurologic and radiographic improvement after hematoma evacuation pointed to the presence of underlying intracranial hypotension, and subsequently, an MRI with gadolinium was performed. Diffuse pachymeningeal enhancement on the axial T1-weighted image (fig. 2a) together with the downward displacement of the cerebellar tonsil on the sagittal T2-weighted image (fig. 2b), verified the diagnosis of SIH. The brainstem was severely squeezed between the retro-odontoid mass and the posterior edge of the foramen magnum (fig. 2b). An MRI of the cervical spine demonstrated a mild fluid accumulation in the posterior cervical epidural space, which had been considered a nonspecific finding. A lumbar puncture was performed mainly for the purpose of infusing saline intrathecally, which has a therapeutic potential to temporarily ameliorate neurologic deficits [7]. The CSF pressure was 2 cm H₂O, and her consciousness level improved to E2V1M6 after the intrathecal saline infusion. A CT myelography revealed a massive CSF leakage over the entire thoracic epidural space (fig. 3a, b). A blood patch was urgently performed to seal the CSF leakage: 20 ml autologous blood was administered at the T3–4 and T9–10 levels (40 ml of blood volume as a total) via an 18-gauge Tuohy needle. The postprocedural course was uneventful and she became fully conscious, with a complete resolution of the left-sided hemiparesis 2 days after

blood patch therapy. A repeat CT myelography obtained 14 days after the blood patch therapy revealed no CSF leakage in the thoracic epidural space (fig. 3c, d), and she was discharged free of symptoms 20 days after the admission to our institution. A brain MRI obtained 30 days after the blood patch therapy revealed the disappearance of contrast enhancement, subdural hematoma (fig. 2c) and the return of the brainstem to the normal position (fig. 2d). She has not sustained a recurrence of SIH for more than 6 months. A written permission for this publication was granted by the patient.

Discussion

Due to the recent awareness of SIH by physicians and the public, its clinical picture has gradually come to light: symptoms are usually chronic, and most patients seek medical attention complaining of orthostatic headache, which worsens over time. Although a prognosis of SIH is generally benign, a rapid neurologic deterioration resulting in an altered mental status has been reported [4, 5]. One of the characteristic radiographic findings in those patients is the presence of the bilateral collection of subdural fluid: in a literature review by Loya et al. [4], all of the 20 SIH patients who became comatose exhibited a bilateral subdural fluid collection (10 hematomas and 10 hygromas). The presence of a rapid neurologic deterioration indicates that a large amount of CSF has leaked from the spinal dura in a short time, and subsequent pressure gradients above and below the foramen magnum may pull both cerebral hemispheres downward equally, with a resultant bilateral subdural fluid collection. In those patients, a midline shift on imaging studies is absent or mild, relative to the marked downward displacement of the brain. Beck et al. [8] found a strong causal association between the presence of bilateral subdural hematoma and SIH in the nongeriatric population (<60 years of age). Our case, a 58-year-old woman, is unique in that a differentiation from SIH-unrelated subdural hematoma was difficult because of the presence of a marked midline shift and a unilateral subdural hematoma on initial imaging studies (fig. 1), which resulted in a diagnostic delay. Therefore, the suspicion of SIH as an underlying cause of subdural hematoma may be warranted in nongeriatric patients not only with bilateral but also with unilateral lesions. In retrospect, establishing an accurate diagnosis in an earlier stage might have been possible by taking notice of a history of sudden neck pain during stretching exercises. Although it remains unclear why the hematoma was located only unilaterally in this case, an adhesion between the pial surface and the dura of the right side, secondary to prior subclinical trauma or inflammation, might have prevented the formation of a bilateral hematoma. SIH patients with subdural hematoma may complain of orthostatic headache less frequently compared to those without subdural hematoma [9, 10]. The presence of subdural hematoma may prevent orthostatic headaches from developing by correcting the abnormally low intracranial pressure or volume.

In SIH patients presenting with an altered mental status, neurosurgeons are often tempted to evacuate the subdural hematoma first. The presence of left-sided hemiparesis indicated that the patient might have sustained a concomitant transtentorial herniation in addition to a downward herniation at the foramen magnum. We should remember, however, that hematoma evacuation prior to the repair of CSF leakage may not only be ineffective but also detrimental [11, 12]: an untreated downward tractive force may lead to a further accumulation of subdural hematoma postoperatively. A deformed brain may not regain its original shape after surgery unless the CSF leakage is sealed. The depressed consciousness in our case might have been due to a severely squeezed brainstem, both anteriorly and posteriorly (fig. 2b). Therefore, in cases in which hematoma evacuation has been performed “erroneous-

ly” prior to the repair of the CSF leakage, recognizing the possibility of SIH as a cause of hematoma and the immediate search for CSF leakage with appropriate spinal imaging studies is important to avoid further deterioration. In addition, MR venography may provide further diagnostic information in SIH patients presenting with an altered mental status since distortion or occlusion of the deep cerebral veins or sinuses is occasionally responsible for the neurologic worsening [13].

Disclosure Statement

None of the authors have any financial relationships with other people or organizations that could inappropriately influence their work.

References

- 1 Inamasu J, Guiot BH: Intracranial hypotension with spinal pathology. *Spine J* 2006;6:591–599.
- 2 Inamasu J, Nakamura Y, Orii M, Saito R, Kuroshima Y, Mayanagi K, Ichikizaki K, Doi H: Treatment of spontaneous intracranial hypotension secondary to C-2 meningeal cyst by surgical packing: case report. *Neurol Med Chir (Tokyo)* 2004;44:326–330.
- 3 Urbach H: Intracranial hypotension: clinical presentation, imaging findings, and imaging-guided therapy. *Curr Opin Neurol* 2014;27:414–424.
- 4 Loya JJ, Mindea SA, Yu H, Venkatasubramanian C, Chang SD, Burns TC: Intracranial hypotension producing reversible coma: a systematic review, including three new cases. *J Neurosurg* 2012;117:615–628.
- 5 Ghavanini AA, Scott CA, Chan DK, Tang-Wai DF: Management of patients with spontaneous intracranial hypotension causing altered level of consciousness: report of two cases and review of literature. *Cephalalgia* 2013;33:43–51.
- 6 Schievink WI: Stroke and death due to spontaneous intracranial hypotension. *Neurocrit Care* 2013;18:248–251.
- 7 Binder DK, Dillon WP, Fishman RA, Schmidt MH: Intrathecal saline infusion in the treatment of obtundation associated with spontaneous intracranial hypotension: technical case report. *Neurosurgery* 2002;51:830–836.
- 8 Beck J, Gralla J, Fung C, Ulrich CT, Schucht P, Fichtner J, Anderegggen L, Gosau M, Hattingen E, Gutbrod K, Z’Graggen WJ, Reinert M, Hüsler J, Ozdoba C, Raabe A: Spinal cerebrospinal fluid leak as the cause of chronic subdural hematomas in nongeriatric patients. *J Neurosurg* 2014;121:1380–1387.
- 9 Kuramae T, Inamasu J, Nakagawa Y, Nakatsukasa M: Spontaneous intracranial hypotension presenting without orthostatic headache complicated by acute subdural hematoma after drainage for chronic subdural hematoma: case report. *Neurol Med Chir (Tokyo)* 2011;51:518–521.
- 10 Schievink WI, Maya MM, Moser FG, Tourje J: Spectrum of subdural fluid collections in spontaneous intracranial hypotension. *J Neurosurg* 2005;103:608–613.
- 11 Lai TH, Fuh JL, Lirng JF, Tsai PH, Wang SJ: Subdural haematoma in patients with spontaneous intracranial hypotension. *Cephalalgia* 2007;27:133–138.
- 12 Chotai S, Kim JH, Kim JH, Kwon TH: Brain herniation induced by drainage of subdural hematoma in spontaneous intracranial hypotension. *Asian J Neurosurg* 2013;8:112–115.
- 13 Schievink WI, Maya MM: Cerebral venous thrombosis in spontaneous intracranial hypotension. *Headache* 2008;48:1511–1519.

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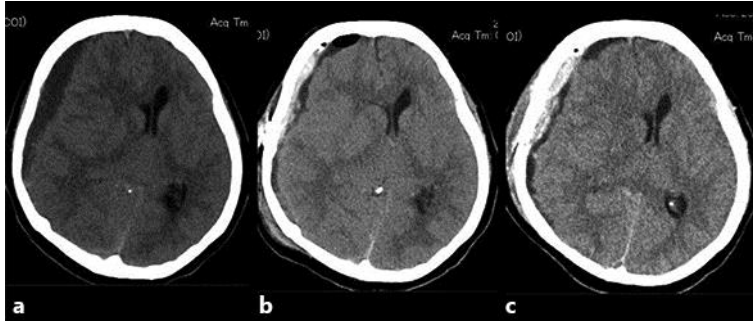


Fig. 1. CT scans of the brain. An initial scan showed a chronic subdural hematoma with a marked midline shift (a). A scan obtained a day after hematoma evacuation showed the lack of reversal of the midline shift (b). A third CT scan obtained 4 days after surgery showed a worsening of the midline shift and the accumulation of subdural hematoma (c).

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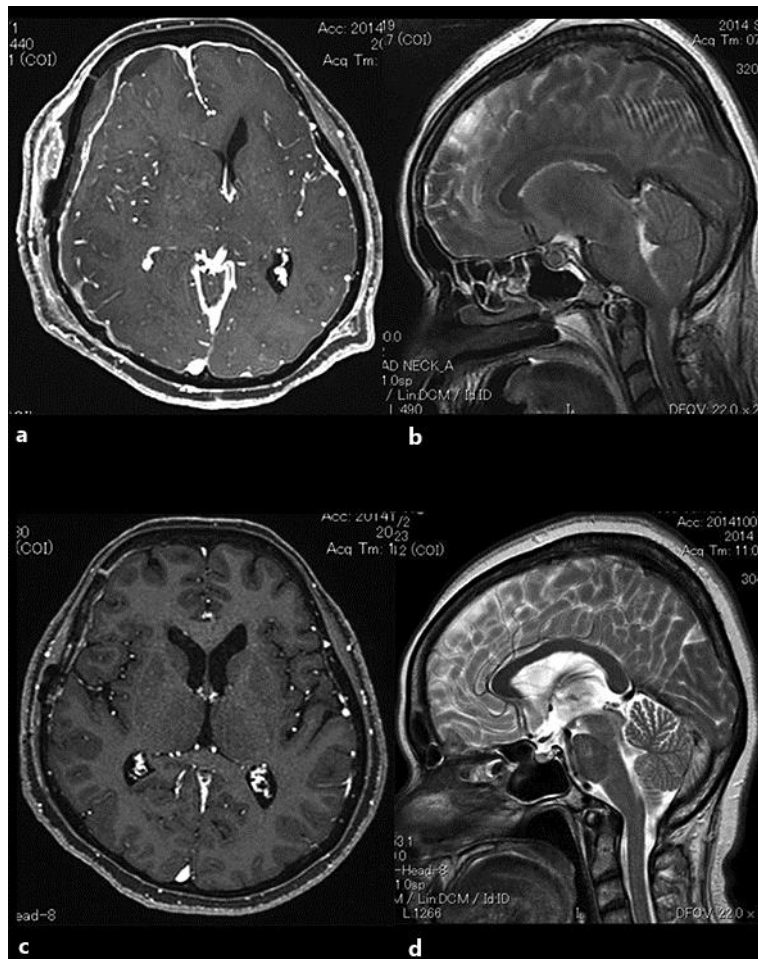


Fig. 2. An MRI of the brain on admission. An axial T1-weighted image with gadolinium showed a marked pachymeningeal enhancement, establishing the diagnosis of a spontaneous intracranial hypotension (a). A sagittal T2-weighted image showed a marked downward displacement of the cerebellar tonsils (b). At 30 days after blood patch therapy, an axial T1-weighted image with gadolinium showed a resolution of the pachymeningeal enhancement (c). A sagittal T2-weighted image showed the return of the cerebellar tonsils to the original position (d).

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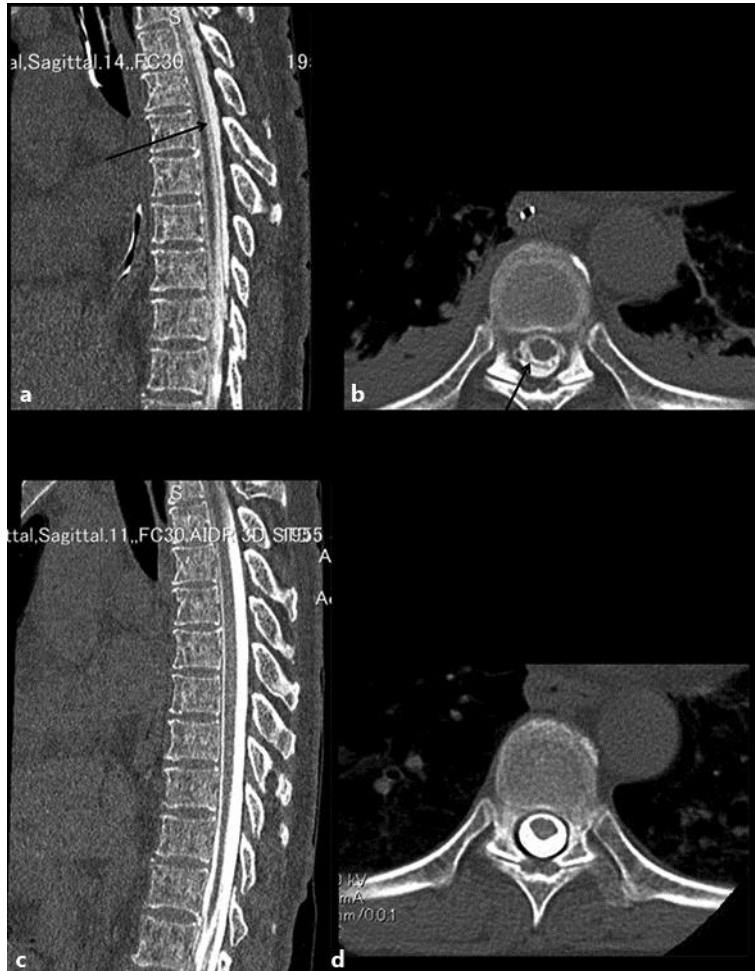


Fig. 3. A CT myelography before blood patch therapy showed massive leakage of the contrast medium in the entire thoracic epidural space on sagittal view (a). A leak of the contrast medium was also confirmed with the axial view (b, at the T9 level). An arrow shows a layer of the spinal dura. At 2 weeks after the blood patch therapy, no leakage of contrast medium in the epidural space was observed (c, sagittal view/d, axial view at T9 level).