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【 CASE REPORT 】

Spontaneous Intracranial Hypotension with a Reversible Splenial Lesion after Swimming

Hirokazu Uchigami, Tomonari Seki, Takuto Hideyama, Junko Katsumata, Risa Maekawa and Yasushi Shiio

Abstract:
Spontaneous intracranial hypotension (SIH) is an important cause of headache mainly associated with spinal cerebrospinal fluid leakage. We herein report the case of a 51-year-old man who developed SIH after swimming. Brain magnetic resonance imaging (MRI) showed a transient high-intensity lesion in the splenium of the corpus callosum (SCC), in addition to bilateral subdural hematomas (SDH) and pseudo-subarachnoid hemorrhage on brain computed tomography. The splenial lesion disappeared and SDH improved after an epidural blood patch. This case emphasizes that transient SCC lesions could coexist with SIH and that SIH should be considered in the differential diagnosis of SCC lesions.

Key words: subdural hematoma, pseudo-subarachnoid hemorrhage, swimming, transient high-intensity lesion in the splenium of the corpus callosum, intracranial hypotension

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Introduction

Spontaneous intracranial hypotension (SIH), which results from spinal cerebrospinal fluid leakage, is now being recognized as an important cause of headache (1). Although the exact cause of spontaneous cerebrospinal fluid leakage remains unknown, there are patients with SIH who have preexisting weakness of the dural sac or trivial trauma (2). The diagnosis of SIH is sometimes challenging because of atypical clinical presentations, atypical neurological imaging features, and complications.

We herein describe a case of a patient who developed intracranial hypotension after swimming, which manifested as a transient high-intensity lesion in the splenium of the corpus callosum (SCC) on brain magnetic resonance imaging (MRI) with bilateral subdural hematoma (SDH) and pseudo-subarachnoid hemorrhage (SAH).

Case Report

A 51-year-old man with no medical history and no history of traumatic injury was admitted to our hospital for sudden headache after swimming. He was not a strong swimmer and was attending a swimming school. When he was swimming, he got an intense headache suddenly and felt dizzy. After 3 days, he visited our hospital with normal physical and neurological examinations except for the presence of an orthostatic headache (on day 1). Laboratory findings from his blood sample were normal. Brain computed tomography (CT) revealed bilateral SDHs and hyperdense lesions in the basal cistern and along the tentorium cerebelli suspicious for SAH, which resulted in urgent hospitalization (Fig. 1a, b). Gadolinium-enhanced MRI showed diffuse dural enhancement and SAH was not detected (Fig. 2e-g), however, a high-intensity lesion was observed in the SCC (Fig. 2a-c).

Burr-hole evacuation was performed for SDH on day 6. Although the orthostatic headache improved, the SDH recurred postoperatively (Fig. 1c, on day 11). The radiological features and SDH recurrence suggested intracranial hypotension. A lumbar puncture was performed on day 22 and a cerebrospinal fluid (CSF) examination revealed low pressure (60 mmH_2O), normal cell counts (4/μL), and elevated protein levels (84 mg/dL). In order to identify the site of a CSF leakage, CT myelography was performed, and it revealed

Department of Neurology, Tokyo Teishin Hospital, Japan
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Correspondence to Dr. Hirokazu Uchigami, uchigamihirokazu1@yahoo.co.jp

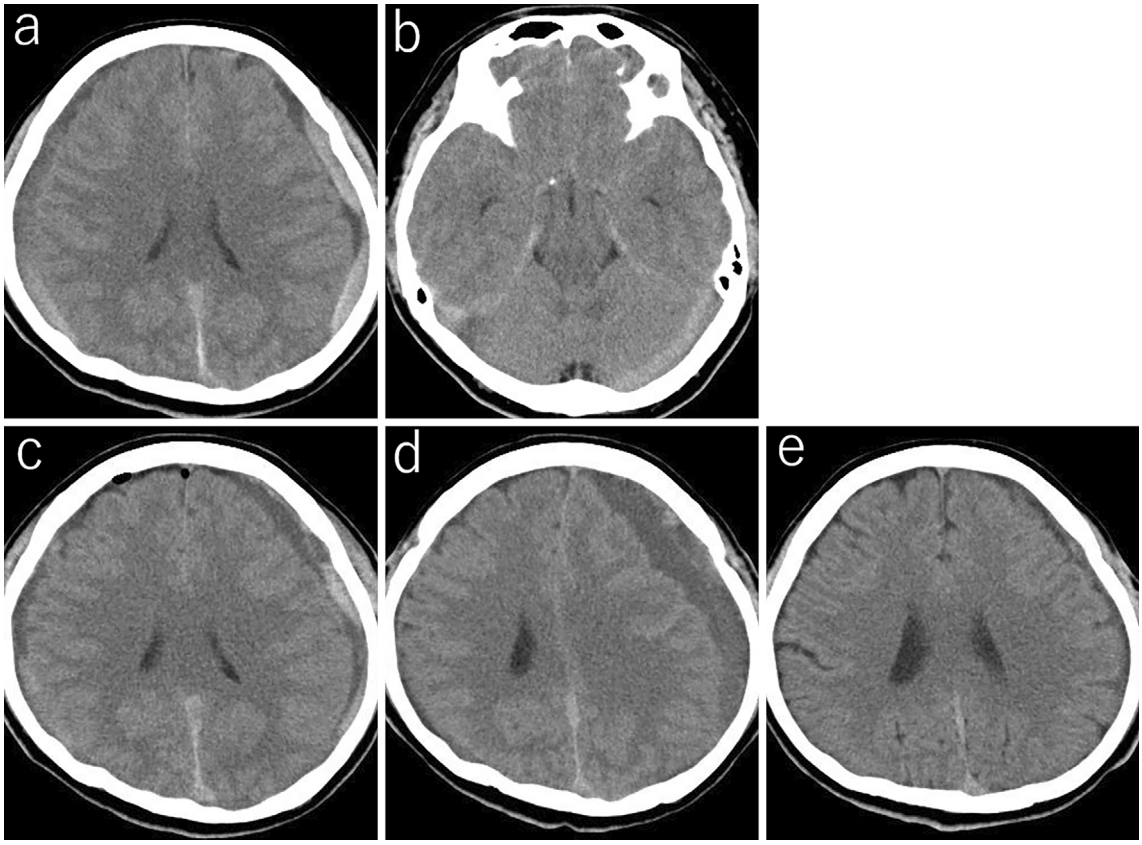

Figure 1. **Brain CT images on admission revealed bilateral subdural hematoma (SDH) (a) and hyperdense lesions in the basal cistern and along the tentorium cerebelli (b). On day 11, SDH recurred after burr-hole evacuation was performed (c). On day 34, SDH enlarged (d). Following a lumbar epidural blood patch, SDH improved on day 100 (e).**

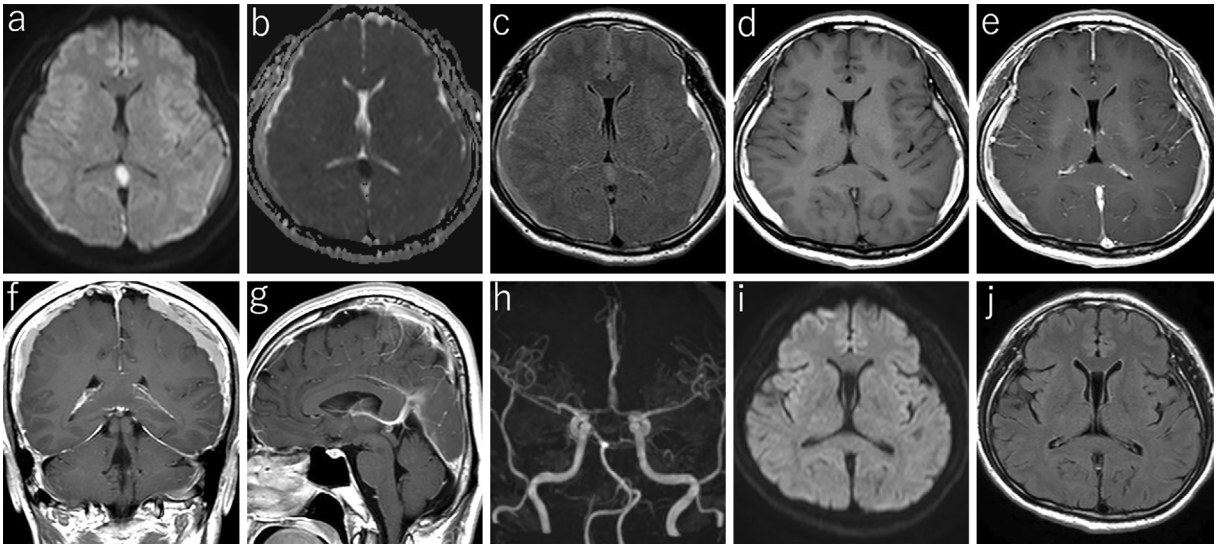

Figure 2. **On admission, axial diffusion-weighted image (DWI) (a) and apparent diffusion coefficient (ADC) map (b) demonstrated restricted diffusion in the splenium of the corpus callosum (SCC). Fluid-attenuated invasion recovery (FLAIR) image (c) showed hyperintensity in the SCC. T1-weighted image (T1WI) (d) revealed isointensity in the SCC. The SCC was not enhanced in gadolinium-enhanced (Gd-enhanced) T1WI (e). Coronal Gd-enhanced T1WI presented pachymeningeal enhancement (f). The straight sinus of our patient was standing steeply and the tentorial angle was elevated on sagittal T1-weighted images (g). MR angiography performed to rule out the possibility of a cerebral aneurysm was unremarkable (h). Following a lumbar epidural blood patch, the splenial lesion disappeared in DWI (i) and ADC map (j) on day 100.**

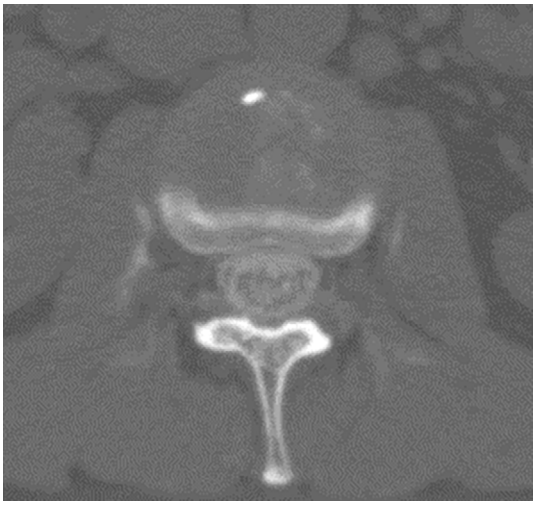

Figure 3. An axial post-myelogram CT image shows extradural contrast extravasation at the L2 level.

extradural contrast extravasation at the L2 level (Fig. 3).

Although we prescribed bed rest and adequate hydration, the patient had SDH recurrence and showed enlargement on brain CT on day 34 (Fig. 1d). We performed a lumbar epidural blood patch on day 38 and sealed the leakage site with autologous venous blood. After the procedure, SDH improved and the splenial lesion disappeared (Fig. 1e, Fig. 2i, j). In addition, the patient presented with no residual symptoms or recurrence three years after the procedure.

Discussion

We described a case of SIH that satisfies the diagnostic criteria of the International Classification of Headache Disorders, 3rd edition (3); the case showed rare clinical image findings and had a previously unreported trigger.

Intracranial hypotension is caused by a CSF volume decrease (leak) due to dural weakness. The primary intracranial hypotension is called SIH, and the secondary types are due to over-draining CSF shunts, traumatic events (major injuries or brachial plexus injuries), true hypovolemic state and herniated discs (4, 5). Contributing SIH factors include connective tissue disorders and trivial trauma (fall, sudden twist and stretch, sports activities, coughing, pulling, pushing, and lifting) (2, 6, 7). Moreover, although patients with CSF leakage at a spinal level may present with symptoms of intracranial hypotension, idiopathic intracranial hypertension is becoming more widely recognized as a cause of spontaneous CSF leakage at the skull base (8). Our patient lacked connective tissue disorder or trauma histories, but the head movement and neck hyperextension during swimming could have caused the rupture of an arachnoid membrane, leading to CSF leakage. Although no SIHs after swimming have been reported, Bai et al. reported the case of an intracranial hypotension after twisting the body when doing aerobic exercises in a swimming pool (9).

An accurate localization of primary CSF leaks is necessary for determining the targeting sites for epidural blood patch (EBP). CSF leaks most commonly occur at the cervicothoracic or thoracolumbar junctions. Cho et al. described 56 patients with SIH who received targeted or blinded EBP, and their CSF leak locations were categorized as follows: cervical, 6 patients; cervicothoracic, 15; mid-lower thoracic, 8; lumbar, 10; and unknown, 17 (10). Farb et al. showed the distribution of localized CSF leaks in 31 patients with SIH, including an L2 leak in 1 patient (11).

The characteristic point of this case is the corpus callosum lesion, which was accompanied by intracranial hypotension. Transient SCC lesions can be seen in various diseases and conditions. Garcia et al. proposed the concept of reversible splenial lesion syndrome (RESLES) associated with different etiologic categories including infection, anti-epileptic drug withdrawal, high-altitude cerebral edema, metabolic disturbances, and others (12).

Only two cases of transient SCC lesions in patients with SIH have previously been reported. In one atypical presentation case, SIH showed transient restricted SCC diffusion, which suggested that the SIH was associated with reversible diffusion restriction in the SCC because a targeted epidural blood patch resulted in a resolution of the symptoms and the imaging finding in the SCC (13). In another SIH case with a focal T2-hypersignal SCC area, the authors speculated that kinking of the vein of Galen (seen on brain MRI) may have caused a blockage or impaired flow through the vein, leading to deep cerebral venous hypertension with secondary edematous SCC changes (14).

As was the case in the latter report, the straight sinus of our patient was standing steeply on the brain MRI (Fig. 2g). Although brain sagging may have impaired the venous flow of the posterior pericallosal vein, leading to venous congestion with cytotoxic edema in the corpus callosum, the splenium of the corpus callosum was not enhanced in contrast-enhanced T1-weighted images, which thus does not support this theory. Our patient and others have shown that SCC lesions can occur with SIH, suggesting that SIH should be considered in the differential diagnosis of SCC lesions.

The patient presented with bilateral SDHs and hyperdense lesions in the basal cistern and along the tentorium cerebelli on brain CT images. Schievink et al. reported a characteristic CT finding in patients with SIH, consisting of pseudo-SAH, which was defined as an increased attenuation in the basilar cisterns, along the tentorium cerebelli or along the falx cerebri, resembling SAH despite the absence of any blood in the subarachnoid space (15). Previous reports and our case illustrate the importance of considering intracranial hypotension in cases with bilateral SDHs and increased attenuation in the subarachnoid space on brain CT.

In conclusion, we herein described an atypical presentation of SIH with a transient SCC lesion after swimming. Brain MRI revealed a transient high-intensity lesion in the SCC, in addition to bilateral SDHs and pseudo-SAH on brain CT. Transient SCC lesions could coexist with SIH. SIH should therefore be considered in the differential diag-

nosis of SCC lesions.

The authors state that they have no Conflict of Interest (COI).

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