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Cerebral Infarction and Remote Cerebellar Hemorrhage in Patients with Intracranial Hypotension

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Abstract:

Cerebrospinal fluid leakage is largely associated with spontaneous intracranial hypotension and iatrogenic events, such as complications associated with spinal tapping or durotomy. Intracranial hypotension causes a positional headache, neck stiffness, nausea, dizziness, or tinnitus. Although rare, intracranial hypotension can lead to serious complications, including subdural hematomas and cerebral infarction. Given its rarity, there is no consensus protocol for treating cerebrovascular complications after intracranial hypotension. We report two cases of intracranial hypotension with cerebrovascular complications, including acute cerebral infarction and cerebellar hemorrhage.

Key Words:

Acute cerebral infarction, cerebellar hemorrhage, intracranial hypotension

Key Message:

Intracranial hypotension can cause serious cerebrovascular complications, such as subdural hemorrhage, cerebellar hemorrhage, or cerebral infarction. Cerebral infarction can be precipitated after a craniotomy to address a subdural hemorrhage associated with spontaneous intracranial hypotension.

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Cerebrospinal fluid (CSF) leakage is largely associated with spontaneous intracranial hypotension (SIH) and iatrogenic events, including complications following spinal tapping or durotomy. Although rare, intracranial hypotension can lead to subdural hematomas and hygromas, diencephalic herniation, cerebellar hemorrhage, and cerebral infarction[1]Currently, there are no consensus protocols for addressing cerebrovascular complications after intracranial hypotension.

Here, we report two cases, including a patient with SIH who developed a postcraniotomy acute infarction in the territory of the posterior cerebral artery and a patient with cerebellar hemorrhage, which was associated with iatrogenic intracranial hypotension.

Case Report

Case 1

A 38-year-old female presented with a positional headache, which she had been experiencing for the last 45 days. A brain computed tomography (CT) revealed a subdural hematoma (SDH) [Figure 1a]. Brain magnetic resonance imaging (MRI) revealed an SDH [Figure 1b], a reduced mamilo-pontine distance, and inferior descent of cerebellar tonsils [Figure 1c], collectively suggestive of an aggravated SIH. Cervical spine MRI also showed diffuse dorsal extradural fluid at the C4-T3 level, suggesting a CSF leak [Figure 1d]. We performed an epidural blood patch (EBP) using the C7-T1 interlaminar approach. Her postural headache improved immediately. Thereafter, her consciousness deteriorated (Glasgow Coma Scale: E2V2M4). We determined that the altered mental status was associated with downward displacement of the brain following an SDH. The patient's response to a second EBP was also temporary. Consequently, we performed an evacuation surgery; however, despite the evacuation of the SDH, her consciousness deteriorated further, and she became comatose (E1V1M1). A third EBP was ineffective. A follow-up MRI examination revealed acute infarctions in both posterior cerebral artery territories [Figure 1e].

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By conservatively managing her acute infarctions, her consciousness improved, and on postoperative day 45, she was able to obey one-step commands. However, a CT myelogram showed that CSF leakage persisted in the ventral epidural space at T1-T9 [Figure 1f]. A fourth EBP was performed. Six months after the operation, she was able to walk independently, despite mild limb ataxia.

Case 2

A 54-year-old female presented with pain in her left flank and anterior thigh. A spinal MRI examination revealed an intradural extramedullary mass at the T10-T11 level [Figure 2a]. The mass was removed via an excision surgery through a durotomy. A biopsy indicated the mass to be a meningioma. On postoperative day 1, she complained of a severe headache, and by postoperative day 2, she developed dysarthria. A brain CT revealed pneumocephalus in the basal cistern, acute intracranial hemorrhage in the superior cerebellar vermis, a subarachnoid hemorrhage in both cerebellar folia, and intraventricular hemorrhage in the left lateral ventricle [Figure 2b, 2c, and 2d]. Brain MRI revealed diffuse pachymeningeal enhancement in both cerebral and distended transverse sinuses, which indicated intracranial hypotension [Figure 2e]. She improved with conservative treatment, and at 1-year follow-up, she showed a complete recovery without any complications.

Discussion

Most intracranial hypotension events are spontaneous, or associated with iatrogenic CSF leakage, and are generally considered to be benign.[1] Intracranial SDH and cerebral infarction have been reported to be associated with SIH and can cause a deterioration in consciousness.[2] A decreased level of consciousness has been reported to be associated with diencephalic herniation rather than the SDH itself.[3] Cases of acute cerebral infarction following SIH are extremely rare. To the best of our knowledge, only five cases have been reported in the literature.[2,4-6] Remote cerebellar hemorrhage (RCH) refers to a cerebellar hemorrhage, typically localized within the rostral part of the cerebellum, which occurs following spinal surgery. RCHs are closely associated with CSF leakage and have commonly occurred in patients receiving surgical durotomy.[7] Previously published cases of cerebrovascular complications with intracranial hypotension are summarized in Table 1.[2,4-9] Our cases illustrate the serious and unusual complications associated with intracranial hypotension.

SDH which is associated with SIH may require neurosurgical drainage. However, a review of the literature indicates that in some patients with an SDH associated with SIH, consciousness deteriorated after a craniotomy. Indeed the four cases of acute cerebral infarctions in SIH patients, which were reported previously, were detected after a craniotomy to address an SDH. Kelley described the sinking brain syndrome, which refers to brainstem herniation after a craniotomy in patients with CSF hypovolemia, and suggested that in cases of SIH, performing drainage surgery was usually not necessary.[10] Schievink reported that performing a craniotomy for treating CSF hypovolemia may result in a vicious cycle, which generates new spontaneous sources of CSF leakage and recurrent SDH.[4] Given this possibility, when a patient with SIH shows deteriorating consciousness, physicians may struggle with a treatment course. Currently, there is no consensus on when a craniotomy should be performed for treating SDH in patients with SIH. Further discussion is needed to determine a treatment in patients with SIH when consciousness declines.

As the volume of spinal CSF decreases, the volume of the vascular compartment within the cranium increases by the way of the Monroe–Kellie doctrine.[10] Both SDH and RCH are thought to be the consequence of intracranial hypotension

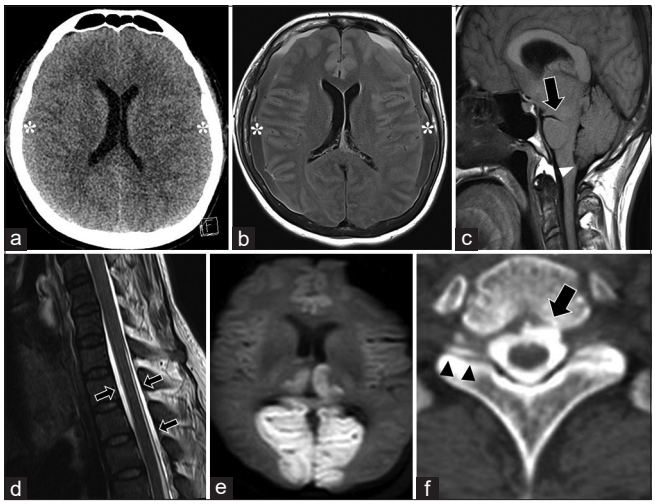

Figure 1: Case 1. (a, b) A brain computed tomography and magnetic resonance imaging showed a subdural hemorrhage (asterisks). (c) A T1-weighted brain magnetic resonance imaging showed descended cerebellar tonsils (arrowhead) with a decreased mamillo-pontine distance (arrow). (d) T2-weighted cervical magnetic resonance imaging showed diffuse dorsal extradural fluid (arrows). (e) Diffusion-weighted magnetic resonance imaging demonstrated acute infarctions in both posterior cerebellar artery territories. (f) Postmyelography computed tomography showed cerebrospinal fluid leakage into the ventral epidural space (arrow) and contrast leakage through the nerve root (arrowheads)

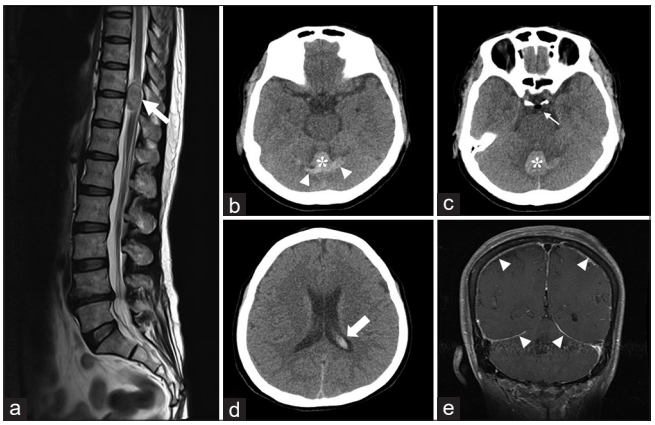

Figure 2: Case 2. (a) A T2-weighted sagittal spine magnetic resonance imaging revealed a 13 × 15 × 27 mm-sized intradural extramedullary mass at the T10-T11 level (arrow). (b-d) A brain computed tomography revealed an acute intracranial hemorrhage in the superior cerebellar vermis (asterisk), a subarachnoid hemorrhage in both cerebellar folia (arrowheads), pneumocephalus in the basal cistern (thin arrow), and intraventricular hemorrhage in the left lateral ventricle (thick arrow). (e) A brain magnetic resonance imaging revealed diffuse pachymeningeal enhancement (arrowheads), suggestive of intracranial hypotension

Table 1: A summary of published cases of cerebrovascular complications with spontaneous intracranial hypotension

Cerebrovascular complications	Author	Age/ sex	Detection after operation	Territory	Outcome
Cerebral infarction associated with SIH[a]	Whiteley, *et al*. (2003)[5]	62/M[b]	Craniotomy	Midbrain infarction	Minimally impaired executive function
	Chi, *et al*. (2007)[2]	43/M	Craniotomy	PCA[c] infarction with Duret hemorrhage	Residual weakness and oculomotor palsy
	Schievink (2013)[4]	45/M	Craniotomy	PCA infarction	Death
	Schievink (2013)[4]	42/M	Craniotomy	Pontine and midbrain infarction	Residual quadriparesis
	Stephen, *et al*. (2016)[6]	57/F[d]	Craniotomy	PCA infarction	Residual weakness
RCH[e]	Friedman, *et al*. (2002)[7]	43/M	T9-T10 discectomy	Vermis and right cerebellar hemisphere hemorrhage	Residual dysarthria and gait ataxia
	Friedman, *et al*. (2002)[7]	56/F	L3-S1 laminectomy, fixation	Superior folia of the cerebellar vermis and bilateral cerebellar hemisphere hemorrhage	Residual dysarthria and gait ataxia
	Suzuki, *et al*. (2015)[8]	57/F	Intradural extramedullary tumor resection, T1-T5 fixation	Superior folia of the cerebellar hemisphere	Residual gait ataxia
	Hofler, *et al*. (2019)[9]	58/F	L2-S1 laminectomy, fixation	Superior folia of the cerebellar hemisphere	Residual gait ataxia

[a]SIH=spontaneous intracranial hypotension, [b]M=male, [c]PCA=posterior cerebral artery, [d]F=female, [e]RCH=remote cerebellar hemorrhage

caused by CSF leakage. While cerebellar hemorrhage has not been described in patients with SIH, it is closely related to postduratomy CSF leakage. The development of SDH, or RCH, can depend on the speed of CSF leakage. In cases where the loss of CSF is rapid, such as after a durotomy, sagging of the cerebellum caudally causes venous bleeding, which, in turn, leads to RCH localized within the upper vermis. In cases of chronic CSF loss, such as SIH, a collateral cortical vein could be developed.[7] With the progression of SIH, the cortical veins crossing the subdural space would be ruptured, leading to the development of an SDH.[1]

The prognosis of RCH after spine surgery is relatively good, with a reported good outcome in more than 75% of patients. The occurrence of RCH is associated with prolonged postoperative CSF leakage rather than intraoperative deliquoration.[11] The management of RCH depends on the volume of hemorrhage and the presence of mass effect in the posterior fossa. Small hemorrhage without brainstem compression can be managed medically while confirming the resolution of the hemorrhage with following CT. On the other hand, a large hemorrhage with a mass effect in the posterior fossa may require surgical intervention, such as hematoma evacuation and suboccipital craniectomy.[12,13]

Depending on the speed of CSF loss, intracranial hypotension can cause serious cerebrovascular complications, such as SDH, or cerebellar hemorrhage. Cerebral infarction also can be associated with SIH. Cerebral infarction can be precipitated after a craniotomy to address an SDH associated with SIH. In patients with SIH with deteriorating consciousness, it is more important to seal the site of CSF leakage, preferentially, using the EBP or intrathecal saline infusion than to evacuate the associated SDH. Early recognition of RCH and prompt imaging investigation is mandatory to establish a good outcome in patients with neurological deterioration after spine surgery and prompt investigation of the image is necessary.

Declaration of patient consent

Written informed consent was obtained from the patients for publication of this case report and accompanying images. In the form, the patients have given their consent for images and other clinical information to be reported in the journal. The patients understand that their name and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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