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Spontaneous Intracranial Hypotension Occurring after Craniotomy for Brain Tumor Biopsy Mimicking Postoperative Bleeding

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Abstract

In this study, we report on a previously healthy 44-year-old man who underwent an open biopsy under general anesthesia for a tumorous lesion found in his left frontal lobe via a small supratentorial craniotomy. While both postoperative course and brain computed tomography (CT) scans had been considered unremarkable, the patient became stuporous on postoperative day (POD) 4. A brain CT obtained on that day showed a subdural hematoma with marked brain shift which we thought might have been due to postoperative bleeding; he was immediately brought to an operating theater for hematoma removal. However, no bleeding source was found, and the brain remained depressed after hematoma evacuation. Furthermore, the brain shift remained unchanged on postoperative CT. While spontaneous intracranial hypotension (SIH) was considered, imaging studies to search for possible cerebrospinal fluid (CSF) leakage in the spinal column were not performed as the patient's condition has improved. However, he became stuporous again on POD 8, which urged us to perform CT myelogram. The CT myelogram showed a massive CSF leakage at the L1-L2 level. Subsequent autologous blood patch has successfully terminated the CSF leakage, and he became fully oriented shortly after the blood patch therapy. Thus, it should be noted that SIH may occur during postoperative period of intracranial surgery, and it may manifest radiographically as a subdural hematoma indistinguishable from postoperative bleeding. SIH should also be included in a differential diagnosis of postoperative headache, regardless of its characteristics, because headache associated with SIH may not always be orthostatic.

Keywords: spontaneous intracranial hypotension, craniotomy, cerebrospinal fluid leakage, subdural hematoma, postoperative bleeding

Introduction

Spontaneous intracranial hypotension (SIH) may manifest either as bilateral or as unilateral subdural hematoma (SDH).[1,2] With the growing literature on the causal relationship between SIH and SDH, complications resulting from hematoma evacuation seem to have decreased.[3] Nevertheless, accurately diagnosing SIH can be difficult if a CSF leakage from the spinal dura occurs insidiously during perioperative period: SDH associated with SIH may be indistinguishable from postoperative bleeding from an index lesion. We herein report a case of SIH with a dural tear in the lumbar spine that developed in a middle-aged man who had undergone a supratentorial craniotomy for brain tumor biopsy 4 days earlier.

Case Report

A previously healthy 44-year-old man was brought to a local hospital after sustaining status epilepticus, which eventually required him to be on mechanical ventilation. His imaging scans revealed an intra-axial tumorous lesion in his left frontal lobe. He was referred to our institution for further evaluation. A brain computed tomography (CT) revealed spotty calcification within the lesion, suggesting that he has a tumor of oligodendroglial lineage. Despite the presence of mild brain edema around the lesion, no brain shift was noted (Fig. 1A). He agreed to undergo an open biopsy to establish the diagnosis. The tumorous lesion was noted to be located not only in the left frontal lobe but also in the left insular cortex as per his T2-

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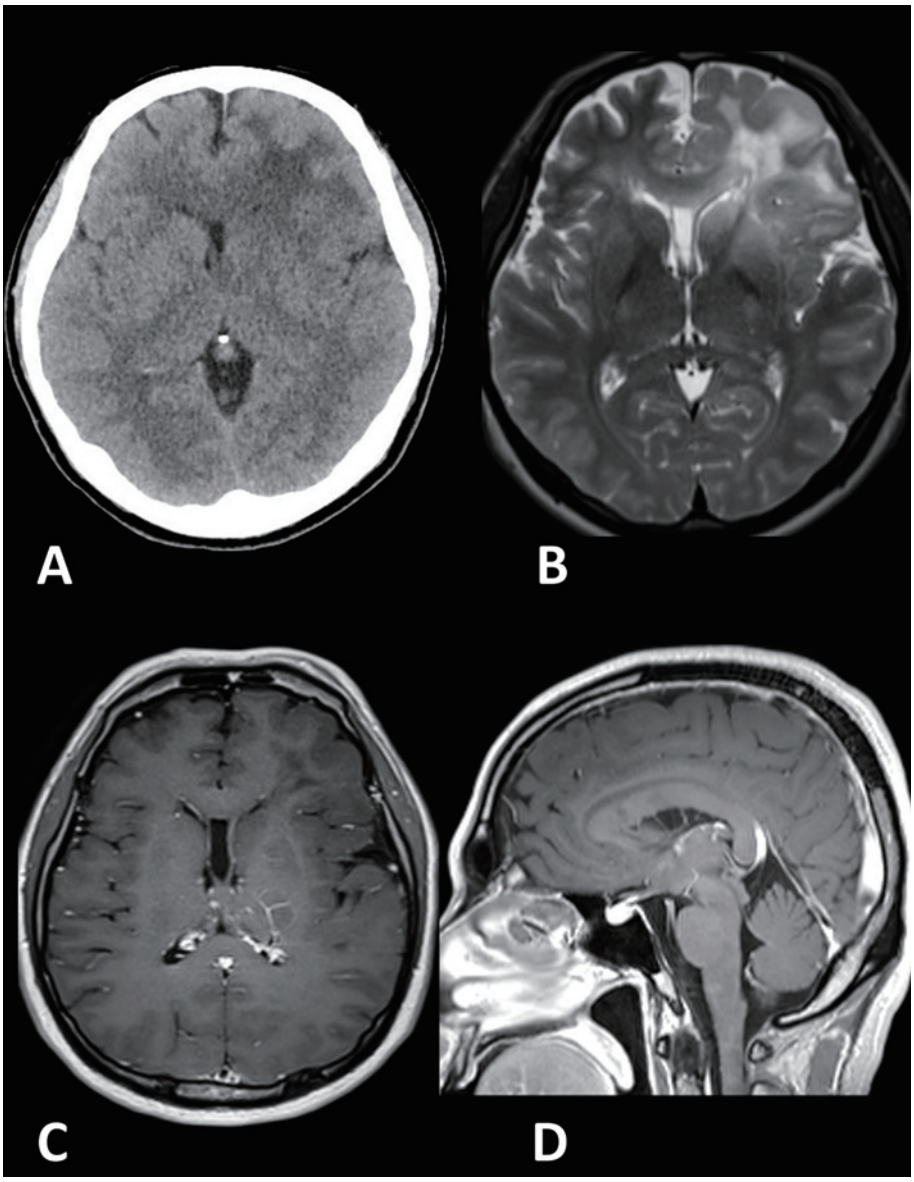

Fig. 1 (A) Preoperative brain CT showing the absence of brain shift despite mild brain edema in the left frontal lobe; (B) T2-wighted MR imaging showing that the lesion was located not only in the left frontal lobe but also in the left insular cortex; (CD) Preoperative Gd-enhanced T1-weighted MR imaging showing the absence of parenchymal and meningeal enhancement. C, axial image; D, sagittal image.

wighted magnetic resonance (MR) imaging scans (Fig. 1B). Gadolinium (Gd)-enhanced T1-weighted MR imaging showed neither parenchymal nor meningeal enhancement (Fig. 1CD). He was alert and fully oriented without any neurological deficits. His seizure had been controlled with antiepileptic medication. He had no memory of sustaining a fall at the onset of the seizure.

The open biopsy was carried out via a small left frontal craniotomy under general anesthesia with the patient in supine position. The operation consisted of excising several small pieces from the brain surface under navigation guidance. The frozen section diagnosis was a low-grade tumor of either glial or oligodendroglial lineage. During the brain excision, bleeding was inconspicuous, and it was easily controlled by electrocautery coagulation. The operation lasted 1 h 15 min without needing to place a subcutaneous drain. No Valsalva maneuver was performed intraoperatively. His postanesthesia arousal was uneventful, and he was allowed to take meal and walk out of bed on postoperative day (POD) 1. Brain CT performed on POD 1 was considered unremarkable (Fig. 2AB). On POD 3, he complained of worsening headache, which was not orthostatic. No other complaints like neck pain, back pain, or dizziness had been reported. We thought that brain edema around the biopsied lesion might have been responsible for the worsening headache; thus, intravenous glycerol was

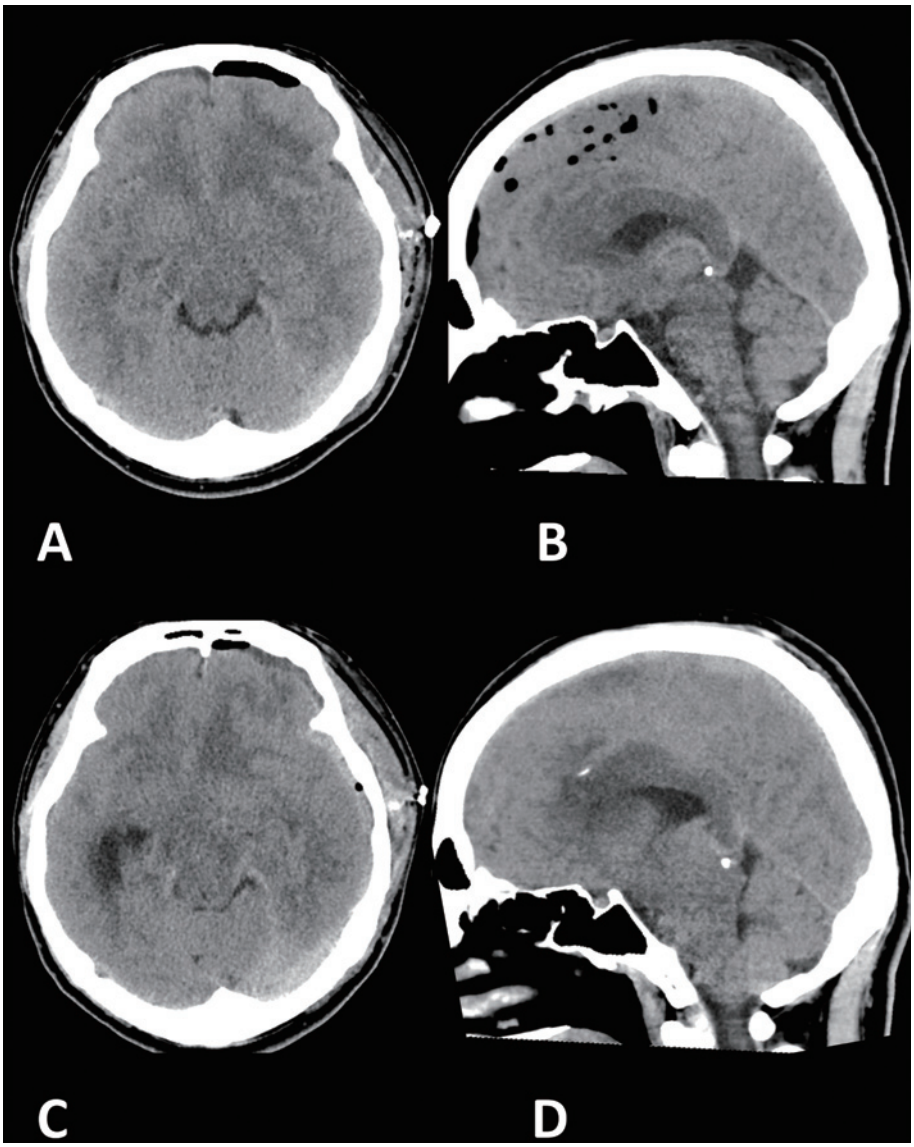

Fig. 2 (AB) Brain CT performed on POD 1, which had been considered unremarkable. A, axial image; B, sagittal image. (CD) Brain CT performed on POD 4 showing the presence of a subdural hematoma, brain shift > 10 mm, and effacement of the prepontine cistern. C, axial image; D, sagittal image.

added with a hope to lower the intracranial pressure (ICP). During that period, no episodes of seizure had been documented.

On POD 4, he was found stuporous with a Glasgow Coma Scale (GCS) score of 12 (E3V3M6). His brain CT then revealed the presence of a mixed-density SDH over the surgical site which we thought was due to postoperative bleeding, and brain shift > 10 mm as well as effacement of the prepontine cistern was also noted (Fig. 2CD). He was immediately brought to an operating room for hematoma evacuation. After reopening the dura, no bleeding sources were found. Moreover, the amount of SDH was smaller than had been expected, and the brain remained depressed after hematoma removal. During dural closure, effort was made to replenish the subdural space by infusing saline. An enhanced T1-weighted MR imaging was performed on POD 5 to rule out the presence of SIH. While intense meningeal enhancement was observed (Fig. 3AB), the pituitary gland did not seem to have enlarged (Fig. 3 C). We considered that those MRI findings were partly, but not entirely, compatible with SIH. While the brain shift remained unchanged (Fig. 3A), the prepontine CSF space was restored slightly (Fig. 3C). Because our suspicion for SIH as a cause of his deterioration had remained, intravenous fluid therapy, that is, lactated Ringer's solution 2,000 mL/day, was administered. Nevertheless, we were hesitant to perform further imaging studies to search for possible CSF leakage because his GCS score improved to 14 (E4V4

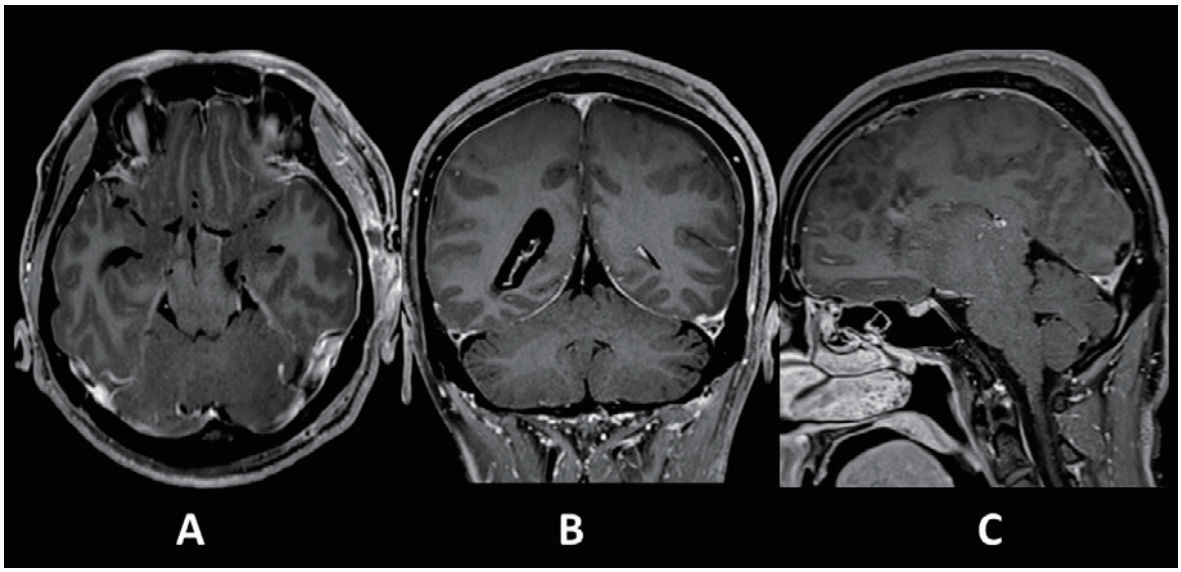

Fig. 3 Gd-enhanced T1-weighted MR imaging performed on POD 5 showing strong meningeal enhancement without marked pituitary enlargement. A, axial image; B, coronal image; C, sagittal image.

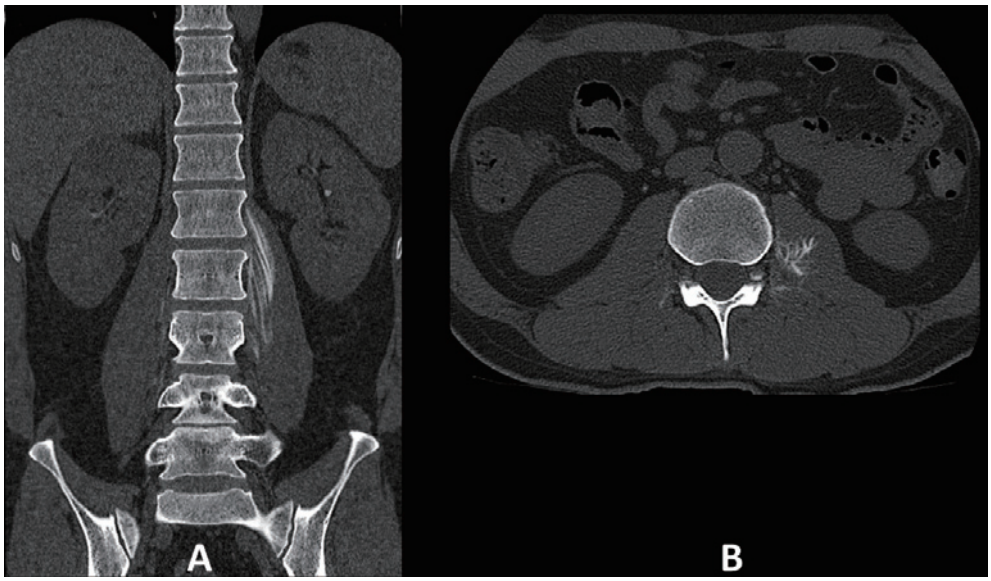

Fig. 4 CT myelogram performed on POD 8 showing marked contrast medium accumulation at the L1–L2 level, suggesting CSF leakage around the dural sleeve of the left L1 nerve root. A, coronal image; B, axial image.

M6). However, he became stuporous again with a GCS score of 11 (E3V3M5) on POD 8. Brain CT revealed worsening of the brain shift. CT myelography was subsequently performed, which then revealed marked contrast medium accumulation at the L1-L2 level, suggesting CSF leakage around the dural sleeve of the left L1 nerve root (Fig. 4AB). Shortly afterward, he was brought to an angiographic suite, and an autologous blood patch was performed: a mixture of 30 mL autologous blood and 10 mL Isovist 240 (Bayer, Leverkusen, Germany) was injected slowly into the epidural space through an 18-G Tuohy needle inserted from the L1-L2 level. The patient recovered immediately after the blood patch therapy, and his brain CT obtained 30 days after the blood patch therapy showed resolution of the brain shift and reappearance of the prepontine cistern (Fig. 5AB). The plasma level of coagulation factor XIII was within normal limits, and he was found to have no clinical features or family history of connective tissue diseases such as Marfan syndrome and Ehlers-Danlos syndrome which are known to be associated with SIH. A moleculopathological examination established the diagnosis of WHO grade II oligodendroglioma for which chemotherapy

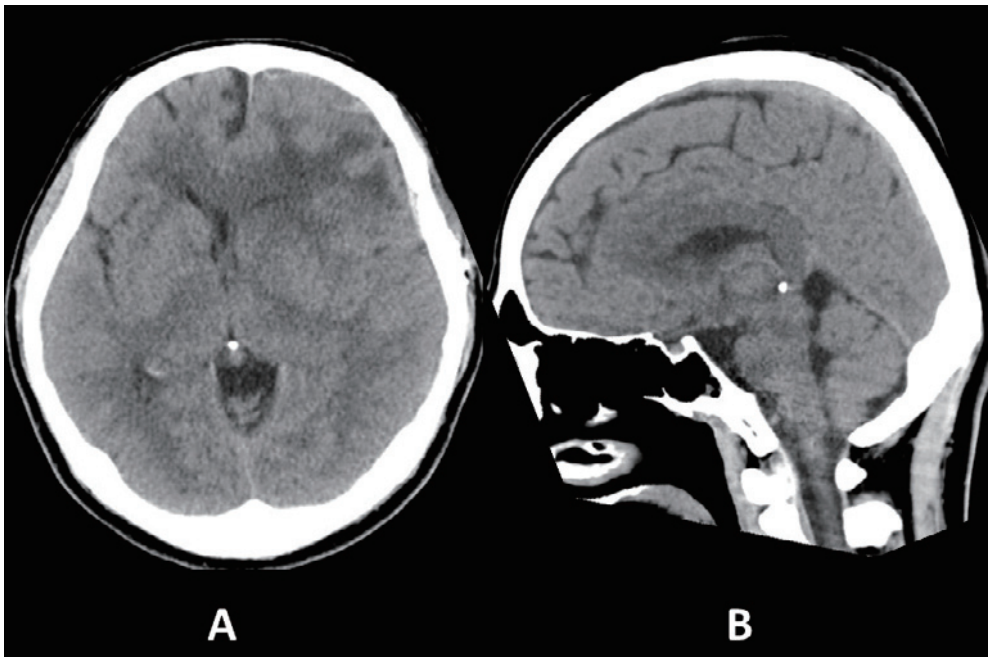

Fig. 5 Brain CT performed 30 days after the autologous blood patch therapy showing the resolution of the brain shift and reappearance of the prepontine cistern. A, axial image; B, sagittal image.

including nitrosourea, procarbazine, and vincristine was implemented. All the participants provided their consent for this report.

Discussion

Accurate and timely diagnosis of SIH may often pose several challenges. Radiographically, it may manifest either as bilateral or as unilateral SDH.[1,2] In the past when our knowledge on SIH was limited, inadvertent bilateral hematoma evacuation resulted in further ICP decrease, bridging vein disruption, and postoperative hemorrhagic complications.[4-6] But with the growing knowledge on the causal relationship between SIH and SDH nowadays, aforementioned postoperative hemorrhagic complications seem to have decreased.[3]

What was unique in this case is that SIH was not present at the time of the craniotomy, and a CSF leak had occurred in a distant location, i.e., in the lumbar dura. While spine and transsphenoidal surgery may be causally related to iatrogenic IH,[7-9] IHs occurring after a supratentorial craniotomy have rarely been reported, and they were mostly due to over drainage of the CSF via the wound suction drainage.[10] What caused a tear in the lumbar dura in our patient remains to be unelucidated: neither hematological nor connective tissue diseases are unlikely to be responsible.[11,12] Both the patient and medical staffs denied a traumatic event such as a ground-level fall in the ward. However, considering his recent history of status epilepticus, inadvertent fall with insidious strain to the lumbar spine might have occurred during the seizure. Therefore, a possibility that surgical positioning and/or bed-to-bed transfer in the operating room under general anesthesia had put additional strain to the lumbar spine,[13] resulting in the dural tear, should have been considered.

Clinical hallmark of SIH, that is, orthostatic headache, may be difficult to recognize in postoperative patients who tend to spend their time lying in bed for several days. In those patients, headache tends to be attributed preconceptually either to wound pain or to increased ICP resulting from brain edema worsening. Hence, SIH should be included in the differential diagnosis of postoperative headache. In retrospect, the use of glycerol might have been responsible for further decrease in ICP and subsequent clinical deterioration. Interestingly, severe SIH cases that manifest as coma may complain of orthostatic headache less frequently than mild SIH cases.[14] Because of the large dural tear in the former, CSF may keep leaking even in the recumbent position, which might explain why our patient had not complained of orthostatic headache and why conservative management had not been effective. Therefore, the possibility of SIH should not be excluded even if the headache is not orthostatic.

The diagnosis of SIH was not considered when we saw the POD 4 CT (Fig. 2CD), which made us to suspect bleeding from the tumor and to perform reoperation. Bleeding after brain tumor biopsy usually occurs within 24 h of surgery,[15] and timing of bleeding in our patient (between POD 1 and POD 4) might have been untypical. Nevertheless, delayed bleeding (≧3POD) was reported to have occurred in

1% of brain tumor surgery,[16] and our decision to perform reoperation might not have been mistaken. We initially suspected SIH as a cause of deterioration during the reoperation, when the brain surface remained depressed after hematoma removal. While the meningeal enhancement on the enhanced MR imaging (Fig. 3AB) suggested the presence of SIH, pituitary enlargement, which is a sensitive early MR sign for SIH,[17] was not observed (Fig. 3C), making us less confident about the diagnosis of SIH. Because of those partially compatible MR findings and partial improvement in his consciousness level, we had been hesitant in doing further imagine scans to search for possible CSF leakage.

In conclusion, it should be noted that SIH occurring after intracranial surgery may manifest as SDH indistinguishable from postoperative bleeding. SIH should be included in a differential diagnosis of postoperative headache in those patients, and it should not be excluded even if their headache is not orthostatic.

Ethical Statement

Consent was obtained from all participants (the patient and his family) in written and oral form.

Conflicts of Interest Disclosure

The authors received no external funding for the performance of this research. All authors declare that there are no conflicts of interest concerning the materials or methods used in this study or the findings specified in this article.

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