Discovering spontaneous intracranial hypotension after failed middle meningeal artery embolization for subdural hematomas: illustrative cases

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BACKGROUND Spontaneous intracranial hypotension (SIH) is a relatively rare and underdiagnosed disease. SIH can lead to subdural hematomas (SDHs) and other complications. SDHs secondary to SIH are difficult to manage, with no consensus in management, and SDHs commonly recur if underlying SIH is not treated.

OBSERVATIONS A 46-year-old male with vague sensory and orientation symptoms presented with bilateral SDHs, which were treated with middle meningeal artery (MMA) embolization and burr hole evacuation. The patient improved initially but had recurrent encephalopathy and SDHs. The patient received 3 epidural blood patches (EBPs) over 8 days with continued improvement. A 78-year-old female presented with headaches, and imaging revealed a left chronic SDH. She underwent MMA embolization and mini-craniotomy for SDH evacuation. Her symptoms returned and imaging revealed a recurrent SDH. Pan spine computed tomography myelography showed a high thoracic cerebrospinal fluid (CSF) leak. She underwent 3 EBPs over 8 days with neurological improvement and stabilization of her SDH.

LESSONS The authors show that, if SDH recurs after initial treatment with MMA embolization, then SIH should be strongly considered and treated with EBPs. Further investigation is required to determine the role of targeted or blind EBPs and the use of imaging to find the source of occult CSF leaks causing SIH.

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KEYWORDS spontaneous intracranial hypotension; subdural hematoma; middle meningeal artery embolization; epidural blood patch

Spontaneous intracranial hypotension (SIH) is an uncommon disorder generally caused by a spinal dural breach leading to occult leakage of cerebrospinal fluid (CSF).1,2 The overall decrease in CSF volume produces downward traction on leptomeningeal and intracranial structures. Most cases of SIH are benign and patients have good clinical outcomes.3 However, SIH can lead to more serious complications, such as subdural hematomas (SDHs), due to tearing of bridging veins, and brain herniation.2,3

Patients with SIH often present with an orthostatic headache, but the wide spectrum of clinical presentation makes diagnosing SIH difficult.4 Prompt diagnosis is important because a delay in treatment can increase the chance of SDH development.3 SDH in the setting of SIH presents a treatment conundrum of whether to treat the hematoma or intracranial hypotension. Only addressing the SDH can lead to recurrence and continued symptomatology.3-6 Therefore, the differential diagnosis of SIH should be stressed when a patient presents with a nontraumatic SDH or has recurrent SDHs after initial management.

Imaging plays a key role in the diagnosis of SIH. Since SIH is caused by CSF leakage at spinal areas, computed tomography (CT) or magnetic resonance (MR) myelography can be used to detect the site of CSF leakage.3-5 Kim et al.5 showed that 80% of patients in their study with nontraumatic SDHs had spinal CSF leaks using CT myelography. The management of SDH in SIH remains controversial, with no current consensus on treatment. The management of SIH involves conservative treatment, an epidural blood patch (EBP), epidural fibrin glue injection, or surgical intervention to seal the area of dural leak.12,7 If SIH in SDH is identified at the initial presentation, then the previous treatment options can be used. However, when a patient presents with SDHs, SIH is not always
apparent and the treatment of the SDH can be emergent to prevent complications.3,6–8

Herein, we describe 2 patients who presented with SDHs. Both patients were initially treated with middle meningeal artery (MMA) embolization, but their SDHs reoccurred, increasing the suspicion for SIH. Treatment with subsequent EBPs led to clinical resolution.

Illustrative Cases

Case 1

A 46-year-old male presented to the emergency room with neck pain and headaches for several weeks, which were followed by altered sensorium. CT revealed mild bilateral mixed-density SDHs (Fig. 1A). In the absence of trauma history, a workup of SIH ensued with MR imaging (MRI) and CT spine myelography. Brain MRI revealed dural enhancement (Fig. 2A). CT myelography was negative for a CSF leak. Without a documented CSF leak, the patient was kept supine for conservative management. Despite this, his altered mental status persisted and his SDH grew minimally. A bilateral MMA embolization was performed. One week later, he grew more lethargic and a CT scan revealed enlargement of his SDHs (Fig. 1B). He was taken to the operating room for burr hole evacuation, after which consciousness improved, but he continued to have altered sensorium. Repeat MRI was performed, revealing progression of cerebellar sagging (Fig. 2B). Due to the enlargement of the patient’s SDHs after MMA embolization and progressive cerebellar sagging despite burr hole evacuation, there was high suspicion for a CSF leak. A decision was made to treat with EBPs. The patient underwent lumbar EBPs 3 times over 1 week, with rapid neurological improvement. He was discharged home and at 3-month follow was asymptomatic. CT showed resolution of his SDHs (Fig. 1C).

Case 2

A 78-year-old female presented with left frontal headache and retro-orbital pain of 1 week duration. She had no trauma history but had a prior meningioma and aneurysm treated. CT revealed a left-sided chronic SDH (Fig. 3A). She underwent preoperative left MMA embolization and subsequent mini-craniotomy for evacuation. After both the embolization and craniotomy, she had prolonged decreased consciousness, which improved after 1 day. She was discharged home. One week later, she presented again with SDH recurrence despite the embolization. Her craniotomy was reopened and the SDH evacuated (Fig. 3B). She again had prolonged decreased consciousness postoperatively. On postoperative day 4, a follow-up CT revealed re-accumulation of the SDH (Fig. 3C). CT myelography was then performed, which revealed a longitudinal extensive CSF hygroma extending from the high cervical to lower thoracic spine. A CSF leak was localized to the left T1–2 neural foramen (Fig. 4A and B). She underwent 3 targeted thoracic EBPs over 1 week and was discharged with clinical improvement. She presented again in 2 weeks with another SDH recurrence, which was redrained. A repeat thoracic EBP was performed. A post-procedure nuclear medicine myelogram was performed without radiographic evidence of active CSF leak. She was followed in the hospital for stability and discharged home. She returned to the clinic in 1 month with no symptomatology and resolved SDH (Fig. 5).

Discussion

A literature search found 3 cases describing the use of MMA embolization for SIH in SDHs.9,10 All 3 cases described patients who presented with SDHs and had SIH diagnosed before initial treatment. Two patients were initially treated with an EBP,9,10 and 1 patient was treated with continuous epidural saline injections.9 All 3 patients failed initial management and required MMA embolization. In our cases, both patients were initially treated with MMA embolization for a presumptive diagnosis of classical SDH before subsequent diagnosis and treatment of SIH.
Observations

The underdiagnosis of SIH has been well reported in the literature. It is hypothesized that this is partly due to the vague clinical presentation and unfamiliarity of the condition among physicians. In a study looking at the rate of misdiagnosis of SIH and its association with SDH formation, Kim et al. found that the diagnosis of SIH was missed in 56.3% of their patients before the correct diagnosis was made in their emergency room. They also reported a longer delay in diagnosis correlated with an increased chance of SDH formation. There is growing evidence that radiographic parameters, such as mamillopontine distance of 5.5 mm and pontomesencephalic evidence of less than 50 degrees, are sensitive and specific for intracranial hypotension. Both of our patients fit these radiographic criteria.

MMA embolization is a relatively new treatment option for SDHs. A recent review found that most studies reported a chronic SDH recurrence rate of less than 5% when MMA embolization was performed alone or as an adjunct to surgery. This is compared with treatment with surgical evacuation alone, which had recurrence rates of over 30%. Since the recurrence rate of SDHs is relatively low after MMA embolization, a recurrent SDH after MMA embolization should increase the suspicion of SIH, such as in our cases. A CT or MR myelogram is the study of choice for detection and localization of a CSF leak and should be ordered to determine the best course of treatment.

A CT myelogram revealed a CSF leakage in our second patient, and treatment with EBPs allowed for eventual resolution of her pathology and improvement in symptoms. Our first patient had a similar recurrence of his SDH after MMA embolization, but CT myelography did not reveal a CSF leakage. SIH was still suspected, so multiple EBPs were administered, which also resolved his symptoms and conditions. The use of prophylactic EBPs when CT myelography is negative despite the patient fitting the clinical picture has not been well studied.

Although EBPs were used in both of our patients, initial management with MMA embolization and surgical evacuation led to recurrent SDHs. Okuma et al. first proposed the use of MMA embolization as a therapeutic option for SIH SDH, but there is still a paucity in research describing its effectiveness and its proper role in management. They proposed using MMA embolization in the
treatment pathway of SIH in SDH, but this requires SIH to be diagnosed at the initial presentation of the patient. In our 2 patients, the SDH did not resolve with MMA alone and required EBPs for definitive treatment.

There is a lack of consensus when treating SDH secondary to SIH. The definitive treatment seems to be closure of the CSF leak; however, it is hard to justify deferring SDH evacuation if the patient is presenting with mass effect and a large collection.\(^\text{5,9,10,14,15}\) However, it is important to consider the surgical risks of SDH evacuation with concurrent SIH. Exposure to atmospheric pressure, such as after a decompressive craniectomy for hematoma evacuation, can increase caudal herniation in a CSF hypovolemic brain due to increased downward traction.\(^\text{5}\) The use of EBPs is the modality of choice for SIH if conservative treatment fails, but whether to administer EBPs before or after SDH evacuation, the effectiveness of multiple EBPs, and use of targeted EBPs all remain debated topics.\(^\text{5,9,11,14–16}\)

In both cases, the collections recurred despite the embolization, which is unusual for classical chronic SDH. The recurrence rate after embolization is estimated to be 2%–4%.\(^\text{14,15}\) Further, both patients had persistent lethargy and altered mental status despite their hematoma evacuations until they were treated via EBP. Both the recurrences and persistent symptomatology should clue the provider as to the real diagnosis if it was missed before.

**Lessons**

We show that, if an SDH recurs after MMA embolization, then SIH should be strongly considered and investigated using myelography. If CSF leakage is shown or suspicion for CSF leakage is high, then the use of EBPs is an appropriate next step in management. Overall, establishing the proper treatment of SDH secondary to SIH requires further investigation.

**References**


**Disclosures**

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

**Author Contributions**

Conception and design: all authors. Acquisition of data: Bhenderu, Wong. Analysis and interpretation of data: all authors. Drafting of the article: Bhenderu, Wong. Critically revising the article: all authors. Reviewed submitted version of the manuscript: all authors. Approved the final version of the manuscript on behalf of all authors: Bhenderu. Statistical analysis: Britz. Study supervision: Britz.

**Supplemental Information**

Previous Presentations

Portions of this work were presented as an abstract on October 8–12, 2022, at Congress of Neurological Surgeons in San Francisco, CA.

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