Use of confirmatory imaging studies to illustrate adequate treatment of cerebrospinal fluid leak in spontaneous intracranial hypotension

Technical note

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Spontaneous intracranial hypotension (SIH) is a syndrome with serious neurological sequelae. As demonstrated by the following report, recurrent episodes of SIH can be difficult to diagnose when associated with other neurosurgical procedures, such as craniectomies. In this paper, the authors demonstrate SIH presenting as a subdural hematoma with recurrence of CSF leaks. Spontaneous intracranial hypotension was further complicated by paradoxical herniation following a craniectomy. Treatment of SIH necessitated multiple epidural blood patches for CSF leaks at different spinal levels and at different times. The efficacy of each epidural blood patch was confirmed with radionuclide imaging. Confirmation of effective blood patch placement may be useful for identifying patients at risk for a failed epidural blood patch or for patients whose neurological examination results have not fully improved. (DOI: 10.3171/2010.5.JNS091405)

KEY WORDS • spontaneous intracranial hypotension • cerebrospinal fluid leak • epidural blood patch • craniectomy • radioisotope cisternography

Spontaneous intracranial hypotension is clinically associated with intense postural headaches, meningismus, visual field obscurations, abducent nerve palsy, tinnitus, diplopia, and vertigo in patients with low CSF pressure. Spontaneous intracranial hypotension is believed to result from CSF hypovolemia or altered compliance of the craniospinal axis, commonly as a result of a spinal CSF leak. An additional theory is that SIH is the result of epidural hypotension maintained by the inferior vena cava vein outflow to the heart. If there is a delay in diagnosis, SIH can result in stupor and coma. The prevalence of SIH is estimated to be 1 per 50,000 people with an incidence of 5 per 100,000.

According to the Monro-Kellie doctrine, CSF is contained in a closed system; when this system is violated, as with the opening of the calvaria or violation of the dural lining in the spinal cord, a negative pressure gradient develops between the intracranial space and spinal canal, resulting in downward displacement of the cerebellar tonsils and midbrain. Several imaging modalities are used to diagnose SIH. Head CT may identify brainstem deformation, elongation, and obliteration of the basal cisterns. Magnetic resonance imaging, the recommended imaging modality, reveals subdural effusions, diffuse thickening of the pachymeninges, obliteration of the basilar cisterns, elongation of the brainstem, pseudosubarachnoid hemorrhage, and cerebellar tonsillar herniation. Intrathecal Gd-enhanced MR cisternography and other imaging modalities such as nuclear medicine studies and CT myelography are also used to identify a spinal CSF leak in SIH. Treatment of SIH has traditionally relied on placement of an epidural blood patch, but studies on its efficacy report failure rates ranging from 23% to 44%. This data suggests that additional confirmatory imaging studies may be warranted to identify persistent CSF leak in a subgroup of patients.

In this paper, we report a case of a patient who presented with recurrent episodes of SIH following a craniectomy for acute SDH. The episodes of SIH were separated by several months and were attributed to CSF leaks at different spinal levels. We report the use of an epidural blood patch and subsequent radionuclide imaging to confirm the adequate treatment and resolution of the CSF leaks.

Illustrative Case

Presentation and Examination. This 79-year-old man with no prior history of trauma presented to a local hospital the morning after the acute onset of headache, with a neurological examination remarkable for disori-
entation, dysarthria, and right-sided paresis with a slight right-sided pronator drift. A head CT (Fig. 1A) revealed the presence of a 1-cm thick, left-sided hyperdense lesion consistent with an acute SDH along the frontal, temporal, and parietal lobes with mass effect resulting in subfalcine herniation. Computed tomography angiography revealed no abnormality.

**Operative Course.** The constellation of clinical and imaging findings necessitated urgent surgical evacuation of the SDH with a left frontotemporoparietal craniotomy. After surgical evacuation of the SDH, the patient's confusion and right-sided pronator drift improved along with a decrease in the midline shift on head CT (Fig. 1B). Forty-eight hours postoperatively, however, he developed increased lethargy and aphasia. A head CT obtained at this time revealed edema, left-to-right midline shift, and recurrence of an extraxial fluid collection (Fig. 1C). After the patient underwent a left craniectomy, his neurological examination results slowly improved to the point at which he could follow commands, but was interrupted by intermittent periods of aphasia and stupor (Fig. 1D). A presumptive diagnosis of paradoxical herniation was made and the patient’s native bone cranioplasty was replaced. After this surgery, the patient again became progressively lethargic with right-sided hemiparesis and worsening midline shift on a head CT scan (Fig. 1E), necessitating removal of the native bone flap. His neurological examination slowly improved and several weeks later the patient underwent cranioplasty with a titanium-fashioned plate. Forty-eight hours after this surgery, a repeat head CT (Fig. 1F) revealed a recurrent extraxial fluid collection that necessitated surgical drainage. It was determined that the patient had worsening lethargy when his head was placed greater than 30° above the horizontal plane (Fig. 2).

Magnetic resonance imaging of the spine was performed to evaluate the patient’s recurrent neurological decline, but did not reveal a CSF leak. Given the concern for the patient’s persistent neurological fluctuations, a fluoroscopically guided intrathecal injection was performed with a 99mTc radiopharmaceutical. Single-photon emission CT of the lumbar and thoracic spine localized an abnormal focus of CSF leakage to the right L-2 nerve root (Fig. 3A). The CSF leak was treated with placement of an autologous lumbar epidural blood patch at the L3–4 interspace. After 10 days of monitoring, there was still no abnormality.

Follow-Up Evaluation. Four months after his discharge, the patient was readmitted for wound dehiscence and the titanium plate was removed. Forty-eight hours postoperatively, the patient developed an acute onset of lethargy and right-sided paresis after having the head of his bed elevated (Fig. 4A). The patient was again evaluated with a fluoroscopically guided intrathecal injection with a 99mTc radiopharmaceutical. Single-photon emission CT of the lumbar and thoracic spine did not reveal a CSF leak when the patient was supine (Fig. 4B), but when he was placed in an upright position, a left-sided CSF leak at the T12–L1 levels was demonstrated (Fig. 4C). The patient underwent placement of an autologous lumbar epidural blood patch followed by cranioplasty and showed improvement in his neurological examination results to the point that he could ambulate with assistance. A 99mTc radionuclide scan was performed 12 days later to exclude additional CSF leaks and confirmed the absence of a CSF leak in the cervical, thoracic, and lumbar spine regions. The patient was discharged with improving head CT results (Fig. 4D), neurological function, and ambulation.

**Discussion**

To our knowledge, the patient in this report represents the first described case of SIH with a CSF leak that was treated, and subsequently confirmed, by follow-up radionuclide imaging. Previous studies have utilized resolution of symptoms to confirm adequate treatment of a CSF leak.29 Other groups described the resolution of characteristic MR imaging changes in the brain that accompany SIH as an objective test to evaluate the success of epidural blood patch treatment.9,11,16,39 However, in 2 large series of patients with SIH treated with an epidural blood patch, approximately 44%32 and 23%6 of patients were refractory to treatment with 1 or 2 autologous epidural blood injections. This suggests that a subset of patients suffering from SIH may require tests to confirm the adequate treatment of their CSF leaks. In the patient in this report, we chose to employ radionuclide studies to verify successful blood patch treatment to ensure that a CSF leak was not responsible for residual neurological deficits observed in our patient. The patient had a complex clinical presentation with SIH and paradoxical herniation necessitating a repeat radionuclide study to confirm the elimination of the residual CSF leak. Had the patient improved with no residual neurological deficits during an observation period, a repeat study would not have been necessary.

Excluding residual CSF leakage in patients with recurrent symptoms or in those who have undergone multiple cranial procedures may be an important step in patient treatment. Delay in diagnosis of SIH is common and can lead to worsening neurological function, altered mental status, progression to coma,16,28 and ultimately, death.1,34 Spontaneous intracranial hypotension is often misdiagnosed and a patient’s neurological decline may be wrongly attributed to other causes such as hydrocephalus.26 The missed diagnosis of SIH can also prompt neurosurgical procedures such as craniectomy. Our patient’s acute SDH and recurrent extraxial fluid collections may have originated from his spinal CSF leak. He underwent a craniectomy with an undiagnosed CSF leak and was put at additional risk from the combined effects of paradoxical shift.

Paradoxical herniation can complicate craniectomies with a paradoxical shift of the brain away from the surgical site, a dilation of the ipsilateral ventricle, a skin flap cavity over the receding brain, and encephalomal-
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Acidic changes of the underlying cortex.\textsuperscript{27} The shifting of the brain in paradoxical herniation is attributed to atmospheric pressure and can compound symptoms in SIH. The definitive treatment for paradoxical herniation following craniectomy is often cranioplasty to reestablish a closed system governed by the Monro-Kellie doctrine.\textsuperscript{22,33} Performing a cranioplasty restores the CSF outflow and brain compliance.\textsuperscript{27} Cranioiasty improves cerebral blood flow to normal levels\textsuperscript{13,44} and several groups have illustrated the resolution of symptoms of SIH following cranial reconstruction of skull defects.\textsuperscript{17}

**Clinical Presentation**

Recurrent lethargy and right-sided paresis with epidural fluid accumulation noted in our patient is consistent with similar reports describing these as signs of a potential CSF leak.\textsuperscript{36,42} Intracranial pressure monitors have been used in SIH to identify low or negative pressures that normalize following treatment with an epidural blood patch.\textsuperscript{6,12,28,29,45} Other groups have reported using several empirical epidural blood patches for recurrent symptoms or when extraaxial fluid collections appear.\textsuperscript{32} In this case, we elected to confirm the presence of a CSF leak, thereby identifying the cause of the patient’s neurological fluctuations.

The CSF leaks occurred at different times and spinal levels (L-2 and T12–L1). The patient may have undergone some unrecognized minor trauma to cause his second CSF leak. Minor trauma and other conditions that weaken the dura have been described as potential causes for SIH.\textsuperscript{5,14,19} However, in our patient both CSF leaks appeared to be adequately treated by placement of a lumbar epidural blood patch. This result provides evidence that an epidural blood patch can be used in multiple CSF leaks that are separated in time and location.

**Treatment Options**

The first line of treatment for SIH involves conser-

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**Fig. 1.** Axial head CT scans obtained during the patient’s clinical course. A: Image obtained on initial presentation demonstrating left frontotemporoparietal SDH with mass effect and midline shift. B: Immediate postoperative scan illustrating evacuation of SDH and improvement in midline shift following craniotomy. C: Scan obtained 2 days postoperatively demonstrating recurrent extraaxial fluid accumulation despite surgical intervention. D: Postoperative scan revealing craniectomy and persistent midline shift despite evacuation of extraaxial fluid. E: Postoperative scan obtained after native bone cranioplasty showing continued midline shift from extraaxial fluid collection. F: Postoperative scan obtained after titanium cranioplasty illustrating persistent extraaxial fluid collection. The inset reveals presence of appropriately contoured titanium cranioplasty.

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**Fig. 2.** Axial head CT scans illustrating midline shift dependent upon the patient’s head position. Left: Scan demonstrating midline shift when patient’s head elevated 30° above horizontal plain. Right: Scan illustrating improvement of midline shift after patient remained in supine position.
Conservative measures, including bed rest, intravenous hydration, the Trendelenburg position, and caffeine administration.26,38 If these measures fail, then a variety of strategies have been attempted and include continuous epidural or intrathecal fluid infusion,7 epidural fibrin glue injections,6,21,23,29,35,45 or epidural blood patch treatment. Treatment with an autologous epidural blood patch has become an accepted therapeutic modality to treat patients based on the constellation of clinical symptoms alone.7,10,19,25,30,31,37 Some groups advocate placement of an epidural blood patch 5 to 7 days after persistent headaches if other causes have been excluded.19

The exact mechanism by which an epidural blood patch acts to alleviate the symptoms in SIH is a subject of active discussion.20 The epidural blood patch may work through a mechanism of forming a blood clot adjacent to the CSF leak, inducing dural fibrosis, increasing the epidural pressure, and tamponading the dura to prevent additional CSF egress.29 The lumbar spine is injected and the blood spreads over many spinal levels to seal the affected area.7 Alternatively, an epidural blood patch can be placed in the region of interest so as to directly target the identified defect.24

**Diagnostic Imaging Techniques**

Various imaging methods are used to diagnose SIH, including MR imaging of the spinal axis, CT myelography, and radioisotope cisternography. Intrathecal Gd-enhanced MR cisternography has recently been employed to detect the exact location of the dural leak in high- and low-flow states.3 The characteristics of the leak may be evaluated after patients are symptomatic for 1 or 2 weeks or if they respond poorly to conservative treatment.4 Cerbrospinal fluid leaks most commonly occur at the cervical spine in the thoracolumbar region.9,15,24,28,50,51,52

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**Fig. 3.** Diagnostic and confirmatory radionuclide studies demonstrating resolution of CSF leak. A: Single-photon emission CT of the lumbar and thoracic spine after fluoroscopically guided intrathecal injection with a 99mTc radiopharmaceutical. An abnormal focus of CSF leak was localized to the right L2 nerve root. B: Confirmatory SPECT imaging following placement of an autologous lumbar epidural blood patch at the L3–4 interspace to assess the resolution of the CSF leak. C: Postprocedural axial head CT scan demonstrating resolution of midline shift and presence of titanium cranioplasty.

**Fig. 4.** Images demonstrating recurrence of SIH and paradoxical herniation with CSF leakage in an upright position. A: Axial head CT scan obtained 2 days postoperatively after cranioplasty removal illustrating paradoxical herniation upon elevation of the head of the bed. B: Single-photon emission CT obtained after fluoroscopically guided intrathecal injection with a 99mTc radiopharmaceutical showing no evidence of CSF leak in the supine position. C: Single-photon emission CT revealing CSF leak at the T12–L1 levels when the patient was placed in an upright position. D: Axial head CT scan obtained after placement of an autologous lumbar epidural blood patch and cranioplasty with resolution of midline shift and paradoxical herniation.
Conclusions

In this paper, we present the use of confirmatory tests with radioisotope cisternography after placement of an epidural blood patch to verify adequate treatment of a spinal CSF leak. If this strategy is adopted on a larger scale, it may identify patients whose CSF leaks are not successfully treated by a blood patch. There have been several reports in which individuals have been refractory to multiple epidural blood patch treatments. A confirmatory test with radioisotope cisternography after placement of an epidural blood patch could potentially hasten the diagnosis of a patient’s neurological decline. There are, however, risks to performing these studies such as the possibility of increased infection, downward brainstem herniation, and creation of an iatrogenic CSF leak. These potential risks and benefits must be carefully weighed for each patient if this strategy is to be employed.

Disclosure

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

The authors contributions to the study and manuscript preparation include the following. Conception and design: Vogel, Dlouhy. Acquisition of data: Vogel, Dlouhy. Analysis and interpretation of data: all authors. Drafting the article: Vogel, Dlouhy. Critically revising the article: all authors. Reviewed final version of the manuscript and approved it for submission: all authors. Study supervision: Vogel.

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