Received: 19.02.2013 / Accepted: 30.05.2013
DOI: 10.5137/1019-5149.JTN.8096-13.1

Case Report

Thoracic Epidural Blood Patch for Spontaneous Intracranial Hypotension: Case Report and Review of the Literature

Spontan İntrakraniyal Hipotansiyon için Torasik Epidural Kan Yaması: Olgu Raporu ve Literatür Derlemesi

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ABSTRACT

Spontaneous intracranial hypotension (SIH) is caused by spinal leakage of cerebrospinal fluid (CSF). Treatment is directed at sealing the site of leak, which is often difficult to localize. We present a case of near fatal SIH that was treated with thoracic epidural blood patching. A 47-year-old male presented with orthostatic headache and bilateral cranial nerve VI palsies progressing over several weeks. Brain magnetic resonance (MR) imaging showed features typical of SIH and identified an epidural collection stretching from spinal levels C6 to T4, but further imaging with MR myelography and radionuclide cisternography failed to identify a precise site of leak. The patient worsened in the hospital requiring craniotomy for evacuation of an evolving subdural hematoma (SDH). Epidural blood patch was performed at the T1-2 level, the presumed location of the leak due to presence of a bone spur on computed tomography and the large corresponding CSF collection. This quickly led to resolution of the headache and cranial nerve palsies, and later to the complete resolution of his SDH. Through this case and review of the literature, we aim to demonstrate that directed cervical or thoracic blood patching should be considered for SIH as an alternative to the conventional lumbar blood patch.

KEYWORDS: Epidural blood patch, Intracranial hypotension, Thoracic

ÖZ

Spontan intrakraniyal hipotansiyonun (SIH) nedeni beyin omurilik sıvısının (BOS) spinal sızıntısıdır. Tedavi, sızıntı bölgesinin kapatılmasını hedefler ama bu bölgenin bulunması genellikle zordur. Torasik epidural kan yaması konmasıyla tedavi edilen neredeyse ölümcül bir SIH olgusu sunuyoruz. 47 yaşında bir erkek hasta birkaç haftadır ilerleyen ortostatik başağrısı ve bilateral VI. kraniyal sinir paralizisiyle geldi. Beyin manyetik rezonans (MR) görüntülemesi SIH için tipik özellikler gösterdi ve spinal C6 ila T4 seviyeleri arasında bir epidural sıvı toplanmasına işaret etti ama MR miyelografi ve radyonüklid sisternografi ile yapılan ek incelemeler tam olarak sızıntı yerini gösteremedi. Hastanın durumu hastanede kötüleşti ve giderek büyüyen bir subdural hematomun (SDH) boşaltılması için kraniyotomi gerekti. Bilgisayarlı tomografide bir kemik çıkıntısı bulunduğundan ve karşılık gelen büyük bir BOS birikimi olduğundan sızıntının varsayılan konumu olan T1-2 seviyesine epidural kan yaması uygulandı. Bunun sonucunda başağrısı ve kraniyal sinir paralizileri hızla iyileşti ve sonrasında SDH tamamen geçti. Bu olgu ve literatür derlemesi yoluyla servikal ve torasik kan yaması yerleştirmenin SIH durumunda konvansiyonel lumber kan yaması yerleştirmeye bir alternatif olarak düşünülmesi gerektiğini belirtiyoruz.

ANAHTAR SÖZCÜKLER: Epidural kan yaması, İntrakraniyal hipotansiyon, Torasik

INTRODUCTION

Spontaneous intracranial hypotension (SIH) is often a benign condition that is managed with conservative measures. When more aggressive treatment is warranted, an epidural blood patch (EBP) is the modality of choice. Historically, lumbar EBP has been used regardless of the site of leak because of its low risk and the ability of blood to travel many levels from the site of injection. An alternative is to direct the EBP at the site of CSF leak, when a location has been determined by imaging. We present a case of near fatal SIH where no definite site of leak was identified. The general location of spinal leak was hypothesized based on imaging to be in the upper thoracic spine, so an EBP was placed at T1-2 level, resulting in the patient's full recovery. We have reviewed the literature to present the reported cases of directed EBP in the cervical and thoracic spine and discuss its implications.

CASE REPORT

A 47-year-old male, previously healthy on no medications, presented to the emergency department with sudden onset upper neck and mid back pain that occurred while watching television. There was no history of trauma. He was given a prescription for an analgesic and sent home without

investigations. He presented again ten days later with similar complaints, now mentioning that the headaches were worse with standing, and improved with recumbency. This time he also reported a new onset of double vision. On exam he had a left lateral rectus palsy. Computed tomography (CT) and computed tomography angiography (CTA) were normal, and the patient was referred to the urgent neurology clinic two days later. The neurologist confirmed the known left lateral rectus palsy, but found no other abnormalities on neurological testing. The patient was told to patch his eye, and magnetic resonance (MR) imaging was organized for two months later (Figure 1).

MR showed bilateral chronic subdural hematomas with pachymeningeal enhancement, with no decent of the cerebellar tonsils into the foramen magnum. At this time the patient described persistent headache in the suboccipital region, again worse with standing and sitting, and immediately resolving with lying down. He had one recent episode of vomiting. Neurological exam now revealed bilateral cranial nerve VI palsies. The patient was admitted to the hospital (day 1) with a diagnosis of SIH. He was restricted to complete bed rest with head of bed flat.

On day 2 of admission, MR myelography of the cervical (C) and thoracic (T) spine (Figure 2) were arranged to look for overt CSF leak, because his pain symptoms localized to those regions of the spine. This showed a thin layer of epidural CSF fluid collection from C6 to T4, with no abnormal CSF out-pouches along any of the nerve roots. As the results were inconclusive, radionuclide cisternography (RNC) was arranged on day 5. The radiologist was unable to access the thecal sac to inject the contrast, and neurosurgery was consulted for insertion of a lumbar catheter. After several unsuccessful attempts, the patient was taken to the operating room (OR) on day 10 where he underwent L5 and SI laminectomies for insertion of a lumbar catheter under direct visualization. The thecal sac was found to be 'floppy' intraoperatively, but the procedure was successful.

On day 11 the patient underwent the RNC study (Figure 3) using the newly inserted lumbar catheter. This did not show any prominent nerve root sleeves nor was there any extravasation of radiotracer. Tracer was, however, identified in the kidneys and bladder 90 minutes after the scan, signifying that a CSF leak was present. On day 13, a computed tomography (CT) myelogram was performed showing effacement of the contrast column ventral to cord from C5 to T3, corresponding to the previously demonstrated epidural collection. Again, no obvious site of leak was identified. Clinically on day 14, the headache was mostly resolved but the bilateral cranial nerve VI palsies persisted. The lumbar intrathecal catheter was removed and the patient began mobilizing.

On the morning of day 16, the patient was found drowsy and aphasic, with an urgent CT head showing enlargement of the left-sided subdural hematoma with 6 mm of midline shift (Figure 4). Urgent burr hole drainage only produced a small amount of blood with no resolution of symptoms, so the patient was taken urgently to the OR for a craniotomy.

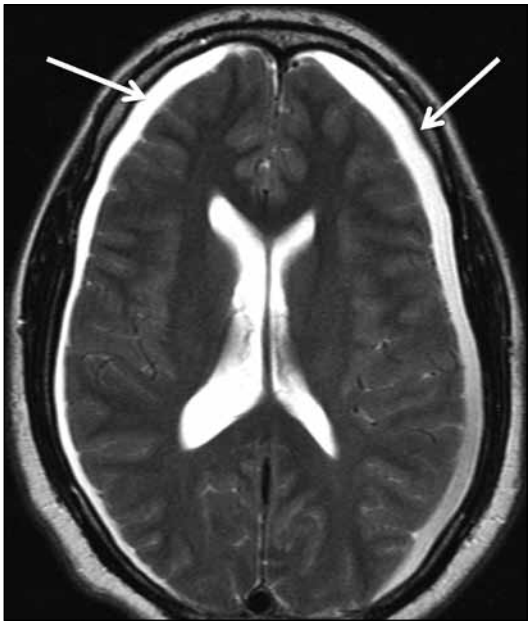

Figure 1: Axial T2-weighted MR showing bilateral chronic subdural hematomas (white arrows). Performed 2 months after onset of symptoms.

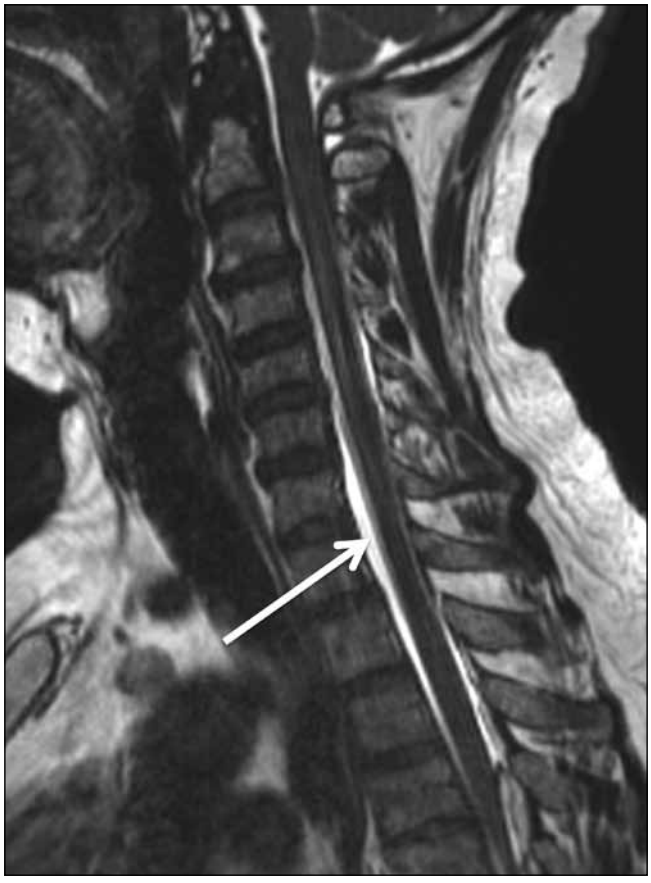

Figure 2: Sagittal MR myelography showing a thin layer of epidural CSF fluid collection from C6 to T4 (white arrow), with no abnormal CSF out-pouches along any of the nerve roots. Performed day 2 of hospital admission.

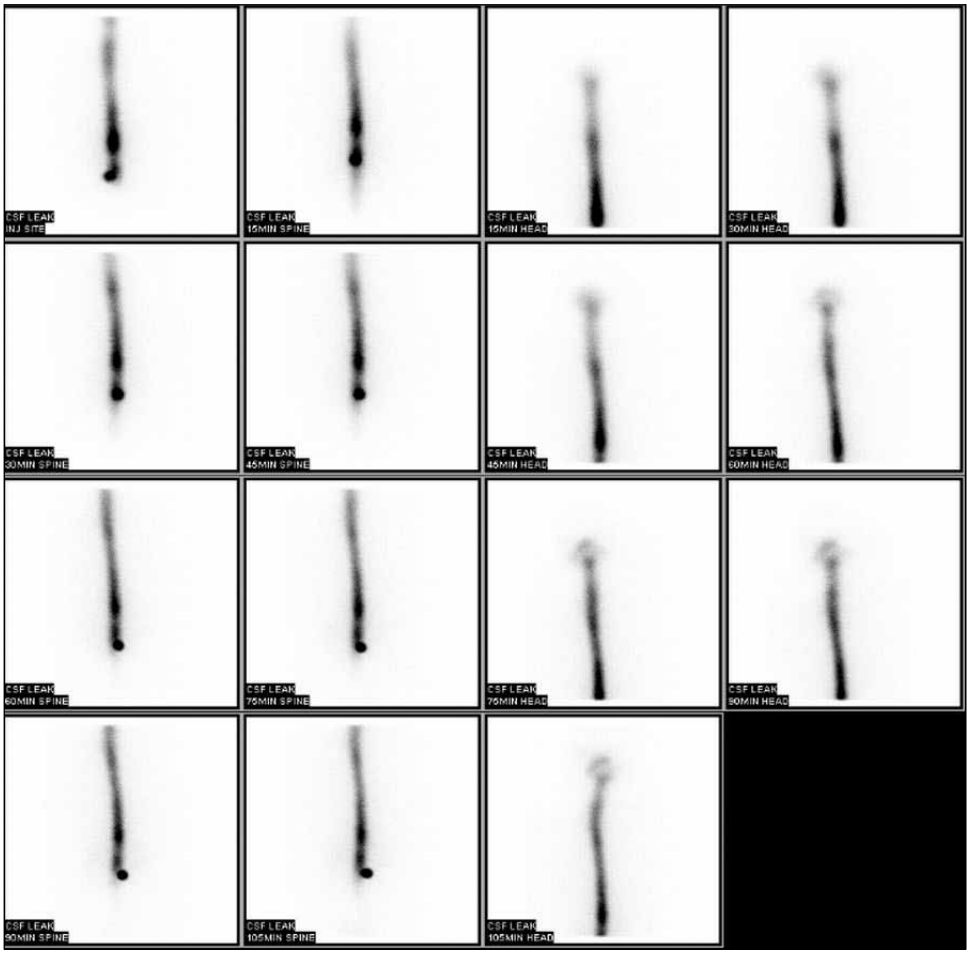

Figure 3: Radionuclide cisternography using Technetium-99 did not show any prominent nerve root sleeves nor was there any extravasation of radiotracer. Tracer was, however, identified in the kidneys and bladder 90 minutes after the scan (not shown in figure), signifying that a CSF leak was present. Performed on day 11 of hospital admission.

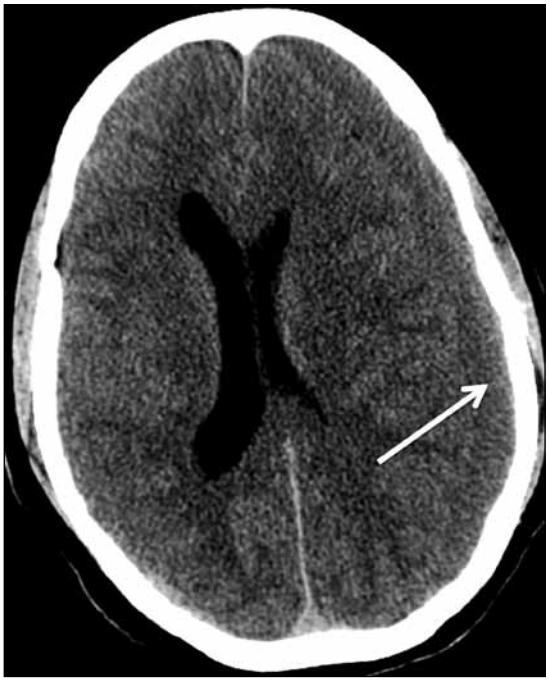

Figure 4: Axial CT showing enlargement of the left-sided subdural hematoma (white arrow) with 6 mm of midline shift. Performed on day 16 of hospital admission.

Post-operatively on day 17 the patient was in similar clinical condition, drowsy and dysphasic with minimal speech, so a lumbar drain was re-inserted in the OR. He was then placed in the Trendelenburg position at 20 degrees, with normal saline infusing into the lumbar catheter at a pressure of 15 cm H2O (equal to 200-400 cc per 24 hours). The patient quickly awoke, his speech normalized, he regained full extra-ocular movements, and reported complete resolution of his headache.

It was clear at this point that the patient had a spinal CSF leak, but we were still uncertain as to the exact location of the leak. We again extensively reviewed the imaging with our neuroradiologists to discern the site of leakage so that a focused EBP could be performed. Re-examination of the day 13 CT myelogram (Figure 5) showed a high index of suspicion that the leak arose from the anterior region of the spinal canal at approximately the T1 level due to the presence of a bony spur on the posterior aspect of this vertebral body and the fairly large CSF collection in the adjacent high thoracic area.

Now that we had a reasonable idea as to the site of leak, the patient underwent an epidural blood patch at the T1-2 spinal level by the anesthesiologist on day 22. 18 cc of autologous blood was used, drawn in a sterile fashion once the epidural

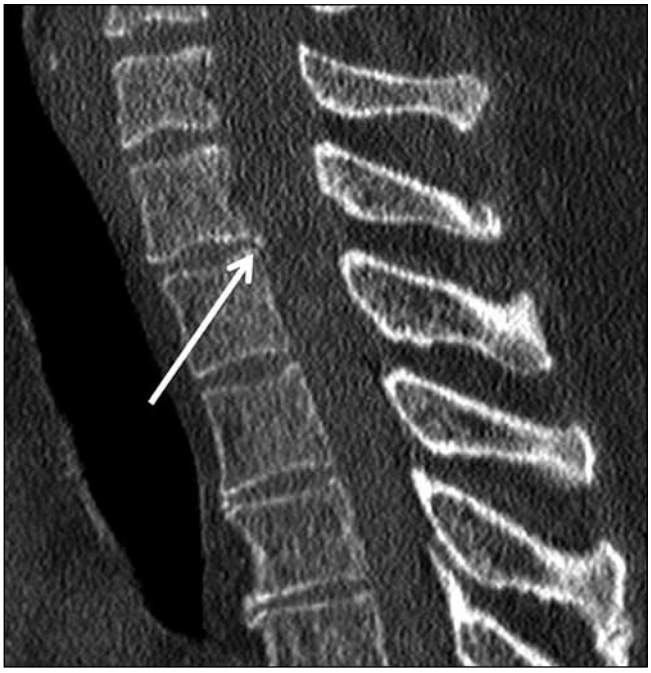

Figure 5: Sagittal CT myelogram showing presence of a bone spur on the posterior aspect of the T1 vertebral body (white arrow), corresponding to the large CSF collection in the high thoracic area. Performed on day 13 of hospital admission.

space was entered. This volume of blood was mixed with 2 cc of Optiray 320 to allow visualization of the spread of the EBP on the cervico-thoracic CT scan. The EBP was done with the patient in right lateral decubitus position. Immediately after the completion of the EBP, the patient was placed prone for about 30 minutes to ensure the spread of blood to the anterior aspect of the canal. This was subsequently confirmed by post-procedure CT scan (Figure 6). The procedure was well tolerated with only mild complaints of neck stiffness afterwards. The drain remained clamped until it was removed the following day.

From day 23 until discharge on day 30 the patient gradually began mobilizing. He remained headache free with no recurrence of diplopia. A follow-up CT head done as an outpatient on day 45 was essentially normal showing complete resolution of the bilateral subdural hematomas.

DISCUSSION

Spontaneous Intracranial Hypotension (SIH), also known as essential aliquorrhea, spontaneous hypoliquorrhea, and Schaltenbrand syndrome, is characterized by orthostatic headache, low CSF pressure, and diffuse pachymeningeal enhancement on MR, all in the absence of antecedent trauma or lumbar puncture (5). The syndrome has an annual incidence of 5 per 100000 (7). Peak incidence occurs at approximately 40 years of age, and is more common in females than in males (15).

SIH occurs due to spinal CSF leak, but the exact cause of the leak is unknown. The leak often occurs at the root exit site, and

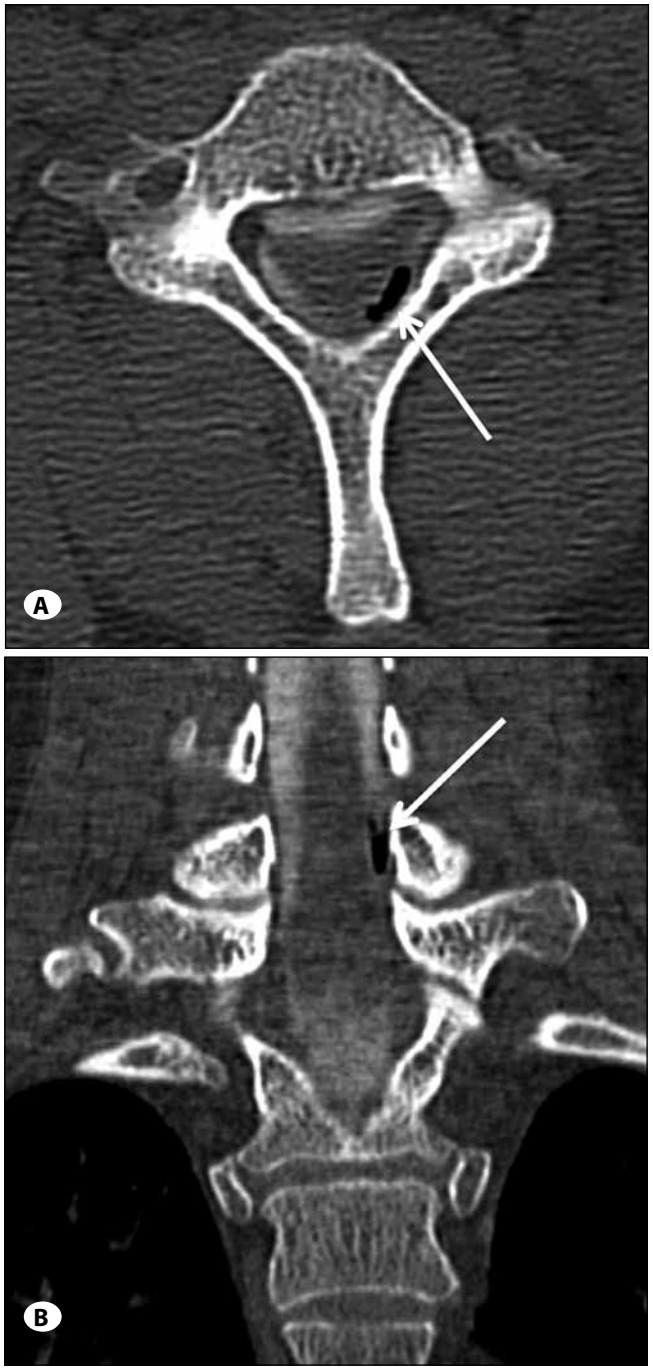

Figure 6: A) Axial and **B)** coronal CT of thoracic spine post-epidural blood patching at T1-2 showing blood in the epidural space (bright signal surrounding cord). Note presence of air at injection site (white arrows). Performed on day 22 of hospital admission.

can be after trivial spinal trauma (13). The pathophysiology is largely unknown, but different theories state there is likely an underlying weakness of the spinal meninges (5). One theory argues that CSF volume loss is responsible for the symptoms of SIH (3). This is supported by certain connective tissue disorders, such as Marfan's and Ehler's Danlos, being risk

factors for SIH (7). Another theory argues that the disease is due to epidural hypotension maintained by the inferior cava vein outflow to the heart. The EBP would then work not by sealing the CSF leak, but rather to help in reversing the CSF-blood gradient within the epidural space along the entire cord (6).

The hallmark feature of SIH is orthostatic headache (7, 14). Other symptoms include neck pain and stiffness, nausea, diplopia due to unilateral or bilateral cranial nerve 6 palsy, dizziness, hearing change, tinnitus, confusion, blurred vision, and photophobia (10).

Different imaging modalities can be used to localize the site of CSF leak. Of these, radionuclide cisternography (RNC) is likely the most accurate, followed by MR myelography (19). Standard enhanced MR can show signs indicative of brain sag, including diffuse pachymeningeal enhancement, descent of cerebellar tonsils, obliteration of basal cisterns, subdural fluid collections, enlarged pituitary, engorged cerebral venous sinuses, and decreased size of ventricles (10). Features in spine MR include extra-arachnoid fluid collections, extradural fluid extravasation extending to paraspinal soft tissues, diverticulae, pachymeningeal enhancement, and engorgement of epidural venous plexuses. Despite advanced imaging, the site of leak remains elusive in up to 50% of cases (17).

The goal in treatment of SIH is to stop the CSF leak and to restore CSF volume and brain buoyancy (1). SIH often will resolve spontaneously or with conservative treatment, such as bed rest (2), hydration, or an abdominal binder (10). Several medical treatments have been attempted, such as steroids, intravenous caffeine, acetazolamide, and oral theophylline, but these are controversial (10). Other techniques include percutaneous placement of a fibrin sealant, but in this case the site of leak must be known (7). Intrathecal infusion of saline is a good temporizing method (11), especially if there is a decreased level of consciousness, as was the case with our patient.

EBP is the mainstay of therapy, often providing instantaneous relief of symptoms in 90% of cases (5). It involves the injection of autologous blood into the epidural space, and has classically been performed in the lumbar region (14). Many advocate for using EBP without identifying a site of CSF leak (18). The success rate of EBP increases with the volume of blood used, with quotes of 80% success rate with 10-15 cc and >95% success rate with 20 cc of blood (14). The ideal volume of blood to inject remains unknown, but it should likely be titrated to relieve the patient's symptoms. Relief of symptoms is often instantaneous, likely due to acute compression of thecal sac causing an increase in CSF pressure. Long-term relief is thought to be due to formation of dural tamponade that seals the leak (14). Epidural blood patches have been shown to spread within the epidural space over many spinal levels (18), as was shown in the post-procedure CT for our patient.

The initial epidural blood patch generally is placed in the lumbar spine regardless of the level of the CSF leak as this has the least risk (4). If lumbar epidural blood patching fails to provide relief, it can be repeated (12) or the placement of an epidural blood patch directed at the exact site of the CSF leak can be tried (20). Thoracic or cervical epidural blood patches are associated with higher risk, including spinal cord and nerve root compression, cord puncture, chemical meningitis, intrathecal injection of blood, blindness, seizures and neck stiffness (1).

In our patient, epidural blood patching was delayed, as the exact site of leak was not identified. In retrospect, we could have avoided the serious complications of worsening brain sag by performing an EBP as soon as SIH was suspected. It is not currently known whether lumbar or directed cervical/thoracic EBP is more effective, but we decided to place the patch at the suspected site of leak. Although we were successful, as were other reports in the literature, further investigation is needed to determine which EBP location is preferred. It is not currently known whether lumbar EBP should be tried first, or whether the risk is low enough to proceed directly to cervical or thoracic EBP.

If repeated epidural blood patching fails, surgical repair is the only remaining option. Surgical treatment is very effective, with one study showing 100% resolution of headache after surgery (16). Surgery should be considered when patients are sufficiently symptomatic to warrant the risks of surgery, when site or sites of the leak have been clearly demonstrated preoperatively, and when symptoms have been refractory to less invasive measures (8). Techniques include suturing dural defects, ligating diverticula including the sacrifice of non-essential nerve roots, plicating dura surrounding essential nerve roots, and packing the epidural space with gelfoam with or without fibrin glue (9).

In summary, directed epidural blood patching in the thoracic or cervical spine should be considered for SIH when lumbar blood patching fails. Cervical and thoracic epidural blood patch, however, do carry a higher risk of complications.

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