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Dural Entry Point of the Vertebral Artery: An Overlooked Route of Spinal CSF Leaks

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Abstract

Spontaneous intracranial hypotension (SIH) is no longer considered rare. Its estimated annual incidence is 5 cases per 100,000 individuals, which is half the incidence of subarachnoid hemorrhage. Epidural blood patch (EBP) is indicated for SIH patients who do not improve with conservative treatment. Accurate determination of the cerebrospinal fluid (CSF) leak site is critical for a successfully targeted EBP. We report the case of a 43-year-old woman with SIH secondary to CSF leakage at the craniovertebral junction dural entry point of the vertebral artery (VA). We treated the patient 2 months after the onset of symptoms. Fat-suppressed T2-weighted spinal magnetic resonance (MR) images revealed a massive epidural fluid collection around the upper thoracic spine. Extravasation of contrast medium through the left VA-dural entry point was clearly visible on computed tomographic myelography. A cervical EBP was injected through the C1-2 interlaminar space. The patient had a smooth recovery and was asymptomatic, with normal spinal MR findings, 6 months after treatment. The possibility of CSF leakage from the dural entry point of the VA should be considered in SIH patients. EBP targeted at the VA entry point is proposed as a safe and effective treatment.

Keywords: spontaneous intracranial hypotension, spinal cerebrospinal fluid leak, epidural blood patch, dural entry point of vertebral artery, pressure/volume enhancement

Introduction

Spontaneous intracranial hypotension (SIH) is characterized by postural headaches that are secondary to spinal cerebrospinal fluid (CSF) leaks. Additional symptoms, such as posterior neck pain or stiffness, nausea, vomiting, changes in hearing, tinnitus, and balance disturbances can also occur. With an estimated annual incidence of 5 cases per 100,000 individuals, SIH is more common than was once thought.[1)] Diagnostic imaging modalities include brain and spinal magnetic resonance imaging (MRI), computed tomographic myelography (CTM), radionuclide cisternography, and digital subtraction myelography (DSM).[1-3)] Epidural blood patch (EBP) is the recommended treatment for SIH patients who do not respond to conservative treatment with bed rest, increased fluid administration, and analgesics.[1,4)] An EBP may be targeted to the CSF leakage site or may be "blind" (distant from the leak). Several studies have shown the advantages of targeted EBP over blind EBP. Accurate determination of the CSF leakage site is critical for successfully targeted EBP treatment.[2)] Here, we describe an SIH patient with a CSF leak at the dural entry point of the vertebral artery (VA).

Case Report

A 43-year-old woman with no history of recent trauma or lumbar puncture presented to a local hospital with an orthostatic headache, nausea, and distorted vision. SIH was diagnosed based on brain MRI findings (Fig. 1A). Two months after diagnosis, the patient was referred to us when conservative measures failed. Fat-suppressed T2-weighted MRI of the spine revealed extensive epidural fluid collection around the upper thoracic spine (Fig. 1B-E). A "false localizing sign" of fluid accumulation was also detected at the C1-2 level of the spine.[5-8)] In axial views,

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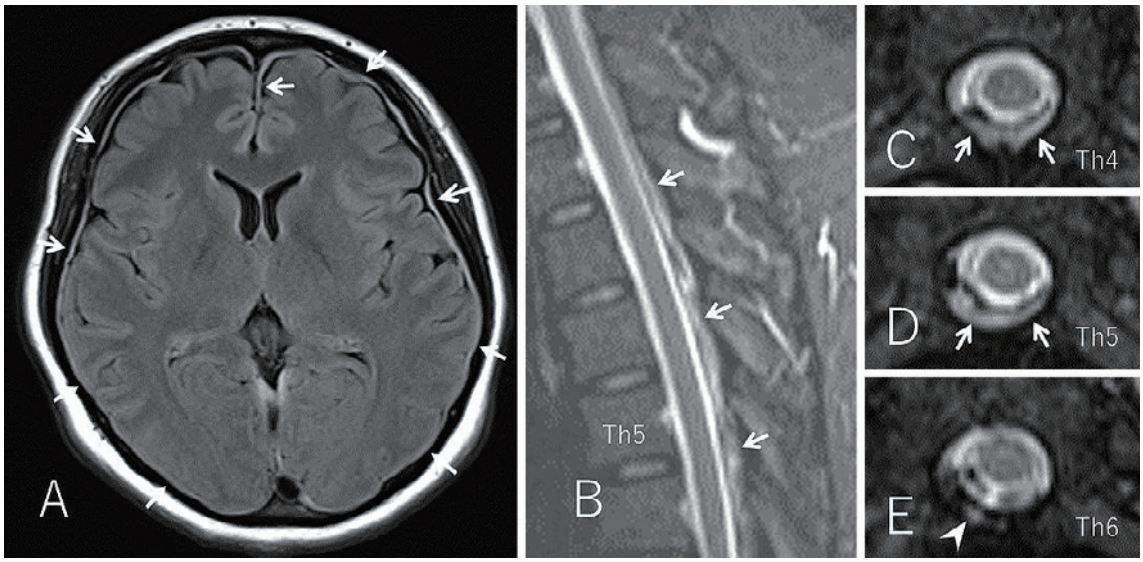

Fig. 1 Cerebral and spinal magnetic resonance images at the first visit.
[A] Fluid-attenuated inversion recovery MR image of the brain at the local hospital shows diffuse pachymeningeal hyperintensity (white arrows).
[B] Sagittal fat-saturated T2-weighted image of the upper thoracic spine illustrates abnormal spinal high signal intensities (white arrows).
[C–E] On axial images, high signal intensities are designated as floating dural sac sign (white arrows) and fringed epidural space sign (white arrowhead).

epidural fluid created the floating dural sac sign, reflecting a massive CSF leak, and the fringed epidural space sign, which signals an incomplete or milder form of floating dural sac sign.[9,10]

CTM with a 25-gauge pencil-point spinal needle yielded a CSF pressure value of 12 cm H_2O. Following this measurement, we injected 20 mL of iohexol 240 (Omnipaque 240, GE Healthcare (Japan) Inc., Tokyo, Japan). CT images obtained 30 min later showed contrast medium in the epidural space, corresponding to the spinal MRI findings (Fig. 2). In addition, extravasation of contrast medium occurred at the left VA-dural entry point. Comparison of spinal MR images before and after CTM revealed a marked increase in the volume of epidural and paraspinal fluid due to a mechanism known as "pressure/volume enhancement" (PVE) (Fig. 3).[11,12] With PVE, increased CSF pressure and postinjection fluid volume activated preexisting leaks that were undetectable with prepuncture spinal MRI. Thus, PVE may be useful for detecting known and suspected CSF leaks.

Based on these findings, we chose the craniovertebral junction as the site of EBP placement. While the patient was lying prone, 32 mL of blood was drawn aseptically from the left brachial vein and mixed with 8 mL iohexol (240 mg I/mL) to make 40 mL of autologous blood-contrast medium solution. A 16 G Tuohy needle was inserted with fluoroscopic guidance. Using the paramedian approach and loss of resistance to saline, the epidural space at the interlaminar region of C1-2 was accessed (Fig. 4A). Needle position was confirmed by injecting a small amount of contrast medium. With fluoroscopic monitoring, the blood-contrast medium solution was gradually injected into the epidural space. After injection of 30 mL of the solution, we ended the procedure when the patient reported moderate suboccipital pain. An upper spinal CT, conducted immediately following EBP placement, revealed circumferential blood distribution around the dural sac, extending the entire length of the cervical spine (Fig. 4B-D). Reflux of the injected blood into the subdural and subarachnoid spaces suggested that blood injection was adequate. The patient recovered uneventfully and remained asymptomatic with normal spinal MR image results at the 6-month follow-up appointment.

Discussion

In a recent systematic review, spinal imaging techniques identified extradural CSF leaks in only 48%-76% of SIH patients.[1] Spinal MRI with intrathecal gadolinium, DSM, and dynamic CTM proved most successful at detecting leakage sites.[1,3,13] Farb and colleagues[13] categorized CSF leakage types as 1) ventral, 2) lateral dural tear, 3) CSF-venous fistula, and 4) distal nerve root sleeve. CSF leak locations in order of decreasing frequency were the thoracic spine, cervicothoracic junction, cervical spine, and lumbar spine.[13] Multiple leaks were reported in 24% of the patients studied.[13]

To the best of our knowledge, this is the first report of a

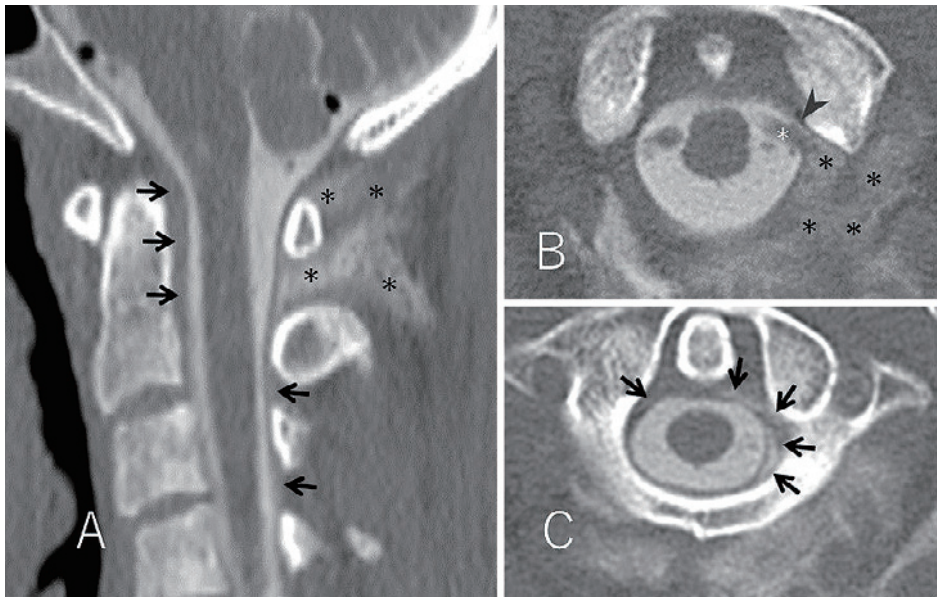

Fig. 2 Computed tomographic myelography images.
[A] Midline sagittal image of the craniovertebral junction shows epidural contrast fluid around the dural sac (black arrows). Leaked contrast medium that flew outwardly from the spinal canal is shown (black asterisk).
[B, C] Axial images at C1. Extravasation of contrast medium through the dural entry point (black arrowhead) of the left vertebral artery (white asterisk) is clearly visible. Note the caudal and circumferential distribution of contrast medium (black arrow, A, C). Spread of extravasated contrast fluid into paraspinal tissues is also evident (black asterisk, A, B).

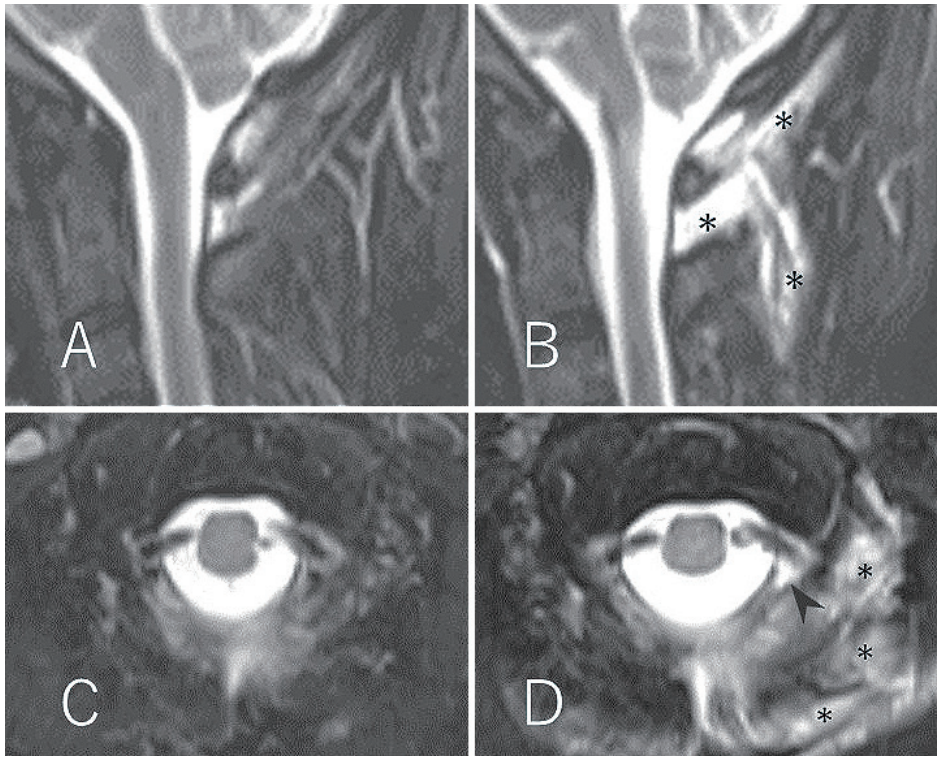

Fig. 3 Magnetic resonance images of the craniovertebral junction illustrating pressure/volume enhancement.
[A] Midline sagittal image at the first visit demonstrates retrospinal C1–2 fluid collection.
[B] The same image immediately after computed tomographic myelography shows a marked increase in fluid accumulation (black asterisk).
[C, D] Comparison of axial images depicting a marked increase in retrospinal fluid volume (asterisk). The cerebrospinal fluid leak at the dural entry point of the vertebral artery is clearly delineated (black arrowhead).

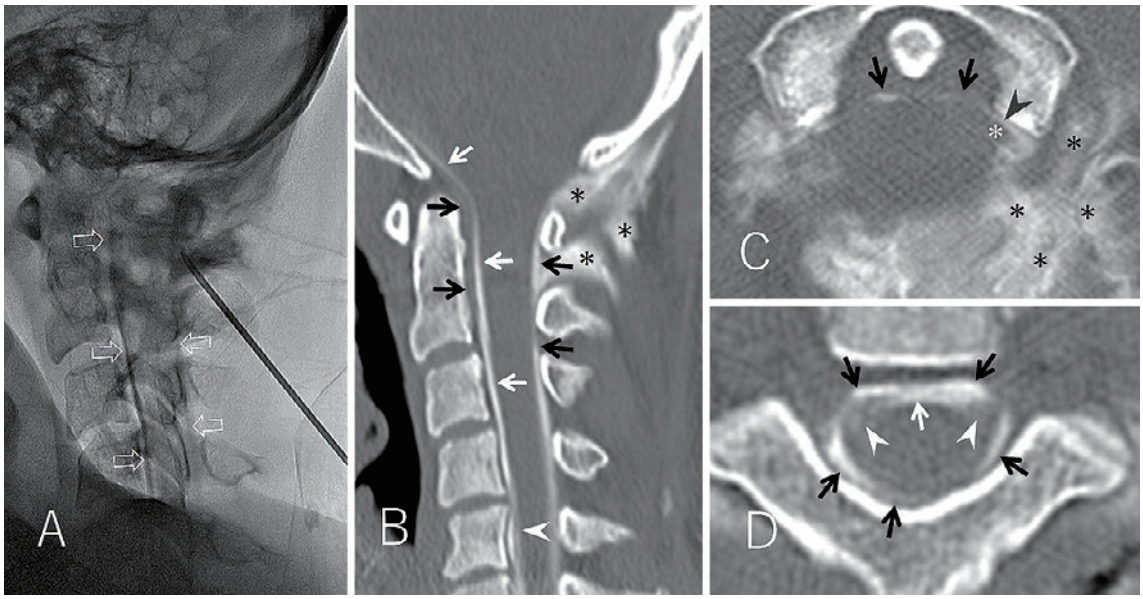

Fig. 4 Fluoroscopic image of the cervical epidural blood patch (EBP) and post-EBP computed tomography (CT) images.
[A] A lateral fluoroscopic image demonstrates the insertion of the epidural Tuohy needle and the distribution of epidurally injected blood (white open arrows).
[B] Sagittal CT image at the craniovertebral junction showing the epidural (black arrow), subdural (white arrow), subarachnoid (white arrowhead), and paraspinal (black asterisk) blood distribution.
[C, D] Axial images at the C1 and upper C3 levels. Note the circumferential epidural blood distribution (black arrow). The epidural space around the dural entry point (black arrowhead) of the left VA (white asterisk) is sufficiently filled with blood. A significant amount of blood refluxes into the subdural (white arrow) and subarachnoid spaces (white arrowhead). Patient placement in the prone position during EBP treatment and CT scans leads to gravity-assisted settling of subarachnoid blood deposits.

CSF leak through the dural entry point of the VA in a patient with SIH. We propose a new leakage type category (type 5) for this CSF leak pattern. The lack of previous reports on VA-dural entry point CSF leaks may be attributed to the difficulty in detection of these sites by conventional imaging modalities rather than to the rarity of this proposed type of CSF leak. It is likely that some SIH patients with unidentified CSF leak sites might have VA-dural entry point leaks. There are several reports of SIH patients with occult C1-2 CSF leakage site.[2,14-16] We strongly suspect type 5 CSF leaks in these patients.

Several SIH patients have histories of trivial precipitating events, including rapid positional changes, Valsalva maneuvers, and mild traumatic injuries.[17] These events would cause disruption of dural weak points, such as fragile meningeal diverticula or attenuated dura. Elevated CSF pressure or mechanical stress ought to be responsible for the final stage of dural disruption. Hence, "spontaneous" means that the event was subconscious or unmemorable. The anatomy of the extracranial VA from the C1 transverse foramen to its dural entry point (the segment known as V3) has been thoroughly investigated to establish safe operative exposure methods for this region. A detailed anatomical study of the dural entry point of VA has not yet been done.[18] Possible mechanisms of CSF leakage through the VA-dural entry point are unclear. However, some form of anatomical weakness at this point might be a predisposing factor for SIH. The possibility of a CSF leak at the VA-dural entry point should be considered in SIH patients with unidentified CSF leakage sites.

Various types of PVEs have been described in the spinal column. Sites of PVE development include the retrospinal space at C1-2, the paraspinal space of the thoracic spine, the areas adjacent to the thoracolumbar spinal nerve roots, and the lumbar epidural space. Retrospinal C1-2 fluid accumulation seen on CTM and MRI has been designated as a "false localizing sign of C1-2" since this finding does not necessarily correspond to a site of CSF leakage. There are two independent reports of retrospinal C1-2 fluid collections caused by CSF leaks from the lower cervical spine.[7,8] The authors postulated that epidural CSF ascends within the spinal canal from the site of the leak, escaping from the epidural space and extending into the soft tissue at the C1-2 level. The C1-2 area might be particularly vulnerable to soft tissue diffusion of CSF due to increased mobility of this spinal segment, along with the absence of epidural fat, lack of bone support, and presence of loose connective tissues. The authors conclude that large CSF leaks in proximity to the upper cervical spine are more likely to be associated with retrospinal C1-2 fluid collections. Our patient's findings illustrate that a CSF leak through the dural entry point of VA may present as a "true

localizing sign of C1-2."

EBP treatment for C1-2 CSF leaks has been challenging. Attempted methods include simple EBP at the lower cervical spine with the expectation of rostral blood seepage, targeted EBP using a Racz catheter, and epidural needle placement at this level under CT guidance.[2,14-16,19] We prefer direct C1-2 puncture under fluoroscopic guidance.[10] Caudal seepage of injected blood following EBP treatment makes C1-2 the optimal puncture site for patients with cervical CSF leaks. When more rostral blood distribution is desirable, the EBP is placed with an epidural catheter that is inserted into the Tuohy needle and squeezed under the C1 lamina to the level of the foramen magnum. We usually prepare 40 mL of autologous blood-contrast medium solution. Blood injection is stopped when the patient reports significant pain. Fluoroscopic monitoring and close observation of patients is essential to avoid complications due to excessive blood injection. In patients with spinal canal stenosis, we pay special attention to their neurological status to avoid missing the subtle signs of spinal cord compression.

In our experience, direct C1-2 puncture under fluoroscopic guidance is a safe and reliable procedure. However, high cervical spinal cord injuries can be devastating and life-threatening. Enough experience of ordinary EBP treatment at the lower spine is a minimum requirement for the first attempt. In addition, consultation with an expert seems to be desirable. Finally, EBP is not always curative. Recent reports suggest that intractable SIH patients can be cured surgically.[1,13,20] Operative closure of the spinal CSF leak might be indicated if the symptoms persist despite repeated EBP treatments and if the spinal CSF leak has been localized.

Conflicts of Interest Disclosure

The authors have no conflict of interest to declare.

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