

# ***A Case of Neck Pain in a Patient with Spontaneous Cerebrospinal Fluid Leak Who Showed Characteristic Findings on Cervical Spine MRI***

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## **Abstract**

We report a case of a patient with neck pain only who was diagnosed with spontaneous cerebrospinal fluid leak (SCFL) based on characteristic findings on cervical spine magnetic resonance imaging (MRI). The patient was a 47-year-old man who had been experiencing neck pain for 3 days. He experienced neck pain when he got up in the morning and could not keep standing. Cervical spine MRI showed significant dilatation of the anterolateral dural canal veins at the C2 level. Under the suspicion of SCFL, additional thoracic spine MRI was performed, which revealed epidural fluid collection in the lower thoracic spine. He underwent bed rest, but his symptoms returned. Epidural blood patch (EBP) was performed. The symptoms improved after EBP, and the venous dilatation disappeared on MRI. Venous dilatation in SCFL is considered an effect of epidural space enlargement due to dural sac shrinkage caused by cerebrospinal fluid loss. Therefore, venous dilatation in the cervical spine is an indirect finding of SCFL. It has been reported that images of epidural fluid collection and dural canal collapse on spinal MRI are useful as direct findings in the diagnosis of SCFL, and these findings are most noticeable at the thoracic level. Because SCFL with neck pain only also exists, dilated images of the epidural vein are valuable for diagnosing SCFL, and neurospine surgeons should be aware of this finding when encountering patients complaining of neck pain.

Keywords: neck pain, cervical spine MRI, spontaneous cerebrospinal fluid leak, venous dilatation

## **Introduction**

A characteristic symptom of spontaneous cerebrospinal fluid (CSF) leak (SCFL) is orthostatic headache, which is also accompanied by neck pain or ear fullness. SCFL is diagnosed by imaging findings such as diffuse dural thickening or brain sagging on cranial magnetic resonance imaging (MRI) or tracer leak on radioisotope cisternogram. However, in patients with SCFL with neck pain only, the disease may be easily overlooked because nonspecific neck pain worsens in the standing position and is relieved in the recumbent position. To evaluate patients with neck pain, MRI of the cervical spine is useful and frequently performed. There may be a hint for the correct diagnosis of SCFL. Here, we report a case in which the main complaint was neck pain, and cervical spine MRI revealed epidural venous dilatation leading to the diagnosis of SCFL.

## **Case Report**

A 47-year-old man, with no significant medical history presented with a 3-day history of orthostatic neck pain. On examination, no numbness, muscle weakness, or abnormal tendon reflexes were noted. His symptoms included orthostatic posterior neck pain that appeared within 30 min of standing and did not worsen with forward and backward bending of the cervix. Both the Jackson and Spurling tests were negative. MRI of the cervical spine revealed no spondylosis deformans or tumor, but significant dilatation of veins in the anterolateral part of the dural canal was observed at the C2 level (Fig. 1). Under the suspicion of SCFL, additional thoracic spine MRI revealed epidural fluid collection in the lower thoracic spine (Fig. 2). We instructed him to keep bed rest as much as possible at home for 2 weeks. At the time of his follow-up visit 2 weeks after the initial visit, his symptoms had improved,

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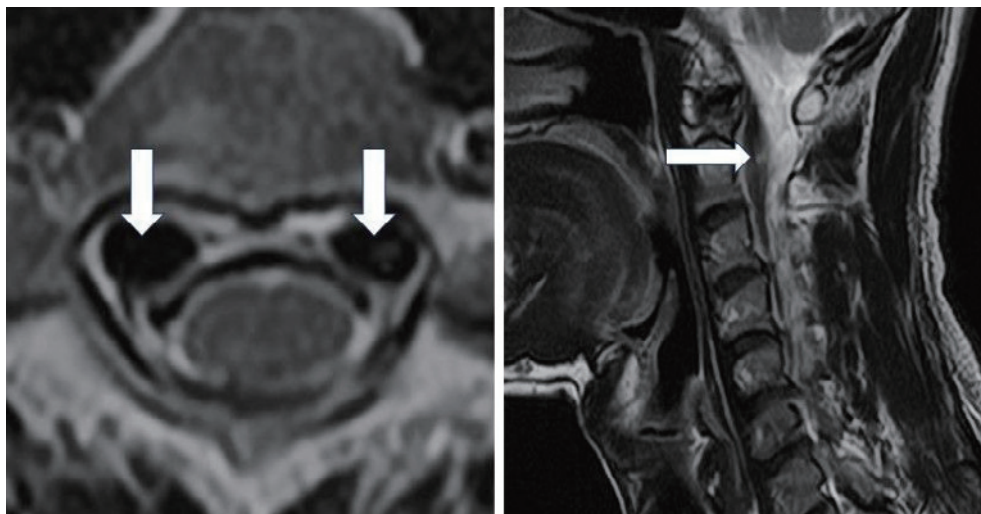


Fig. 1 T2-weighted MRI shows significant dilatation of the anterolateral dural canal veins (white arrow) at the C2 level.

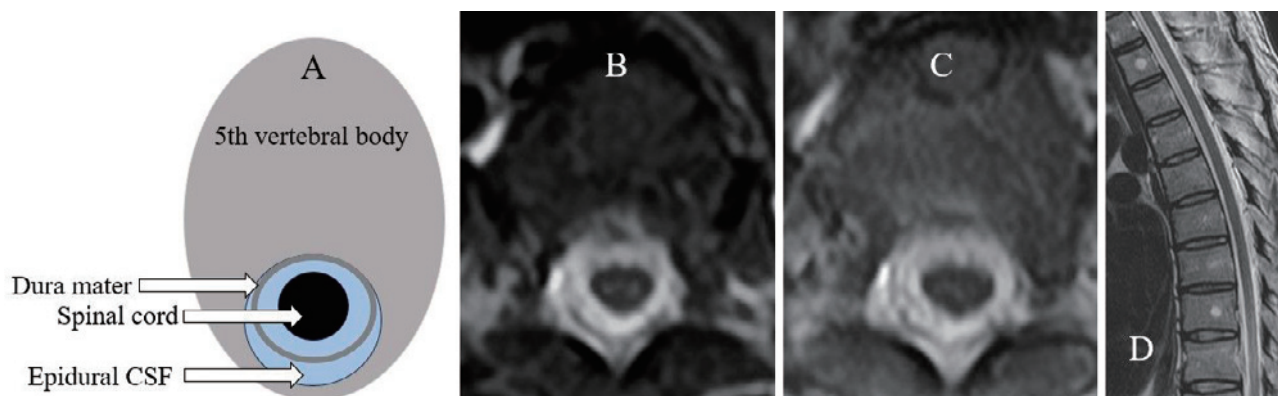


Fig. 2 Thoracic spine MRI revealed epidural CSF collection in the thoracic spine (A, pattern diagram; B, axial STIR; C, axial T2WI; D, sagittal T2WI).

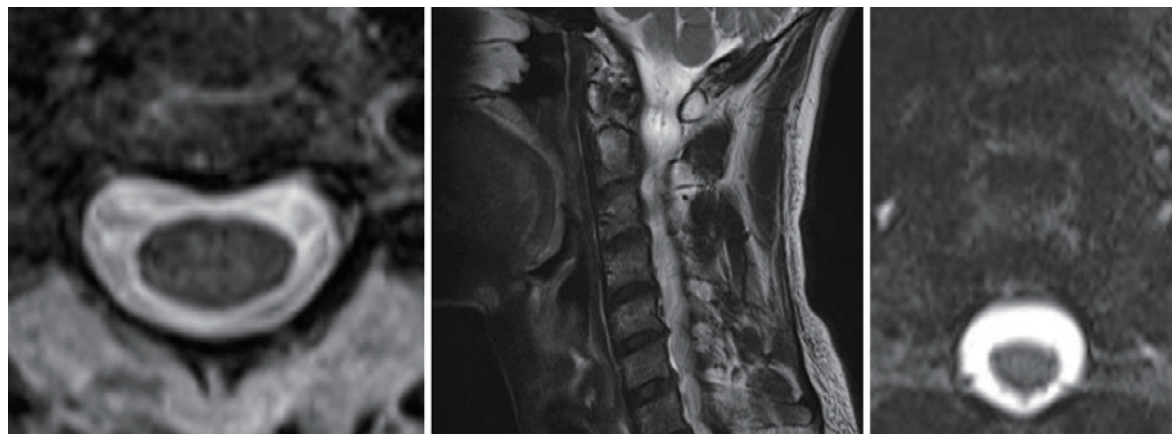
and after two additional weeks, his symptoms had almost disappeared. However, 2 weeks after his last visit, he developed orthostatic neck pain and headache and visited our hospital again. Epidural blood patch (EBP) was performed because the rest in bed was thought to be ineffective. The procedure for EBP is as follows: (1) perform epidural puncture from the L1/2 level while lying prone in the fluoroscopy room. (2) Mix 40 mL of venous blood and 10 mL of contrast medium. (3) Inject 35-45 mL of mixed liquid unless symptoms such as numbness or paralysis of the lower extremity occur. (4) Discharge the patient from the hospital after the rest in bed with bathroom privileges for 3 days. The symptoms had disappeared after the time the patient was discharged. No symptom recurrence was observed, and venous dilatation disappeared on MRI 10 weeks after onset (Fig. 3).

## Discussion

SCFL is characterized by orthostatic headache and de-

creased CSF (cerebrospinal fluid) pressure. Because of increased awareness, the disease is no longer a rare entity. CSF leakage into the epidural space is caused by spinal arachnoid membrane rupture.<sup>1)</sup>

Many reported cases are characterized by orthostatic headache.<sup>2)</sup> Thus, cases involving neck pain alone may be easily overlooked. Albayram et al.<sup>3)</sup> reported a case of SCFL with cervical radiculopathy, which is usually suggestive of spinal cord disease. In fact, two of 26 patients reported pain in the upper limbs, which appeared to be caused by radiculopathy in Mokri et al.<sup>2)</sup> The mechanism underlying orthostatic headache in SCFL remains unclear, but CSF leakage is thought to cause the brain to shift downward, causing traction pain.<sup>1)</sup> From our research, the mechanism by which CSF leakage causes neck pain is unknown, but we believe it is the same mechanism as orthostatic headache. In other words, the nerve roots are pulled because the spinal cord is deflected downward, which may cause traction pain. Hallon et al. reported the capacity for a dilated vein to occur in the central nervous system and



**Fig. 3** Venous dilatation at the C2 level (left and middle) and epidural CSF in the thoracic spine (right) disappeared on MRI after EBP.

cause neurological symptoms.<sup>4)</sup> In Hallon's report,<sup>4)</sup> a dilated epidural venous plexus was associated with either intractable radicular and back pain or severe neurological deficits, including cauda equina syndrome. In our case, venous dilatation was significant in the upper cervical vertebrae, and it cannot be denied that venous dilatation caused neck pain due to nerve compression. However, we believe that venous dilatation is a passive secondary change associated with a decrease in intradural volume due to CSF leakage and that it is unlikely that a sufficient pressure to compress the nerves would be generated. Additionally, we believe that the reason venous dilatation is significant in the upper cervical vertebrae is the large diameter of the dural canal in this area. Further pathological elucidation must confirm these findings.

In previous reports, dilation of the spinal epidural veins was detected in 75%-100% of patients in a relatively small series with variable diagnostic criteria.<sup>5-7)</sup> The differential diagnosis of epidural venous dilatation requires consideration of vascular lesions, such as arteriovenous malformations, arteriovenous fistulas, varicose veins, and compensatory neck epidural venous dilatation due to neoplasms or infections.<sup>8)</sup> The imaging characteristics of epidural venous dilatation in SCFL are that it is located symmetrically anterolateral to the dural sac, does not cross the midline, and the venous structure may expand or contract on consecutive images.<sup>3)</sup>

Shinaver et al.<sup>9)</sup> diagnosed the epidural mass on MRI as abscess and administered antibiotics. Because the patient's symptoms did not improve, he underwent surgery, and results showed epidural venous dilatation. Venous dilatation should be considered when an epidural mass is observed. For the diagnosis of SCFL, epidural fluid collection and dural canal collapse images on spinal MRI have been reported to be useful as direct findings.<sup>10)</sup> This finding is significant at the thoracic level and may be overlooked on cervical spine MRI alone. The basic treatment is rest, and

if there is no improvement, EBP should be considered.<sup>11)</sup> In this case, the symptoms improved, and the image findings improved with EBP. As in this case, focusing on venous dilatation, which is an indirect finding of SCFL, was thought to improve the diagnosis.

### Abbreviations

spontaneous cerebrospinal fluid leak (SCFL), magnetic resonance imaging (MRI), epidural blood patch (EBP), cerebrospinal fluid (CSF)

### Statements and Declarations

All procedures were performed in accordance with the ethical standards of the institutions and the 1964 Declaration of Helsinki and its later amendments.

### Consent to Publish

The participant consented to the submission of the case report to the journal.

### Conflicts of Interest Disclosure

None

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