

Case Report

Application of time-spatial labeling inversion pulse magnetic resonance imaging in the diagnosis of spontaneous intracranial hypotension due to high-flow cerebrospinal fluid leakage at C1-2

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Abstract

Background: Spontaneous intracranial hypotension (SIH) due to cerebrospinal fluid (CSF) leakage at C1-2 poses diagnostic and therapeutic challenges to spine surgeons. Although computed tomography (CT) myelography has been the diagnostic imaging modality of choice for identifying the CSF leakage point, extradural CSF collection at C1-2 on conventional CT myelography or magnetic resonance imaging (MRI) may often be a false localizing sign.

Case Description: The present study reports the successful application of time-spatial labeling inversion pulse (T-SLIP) MRI, which enabled the precise identification of the CSF leakage point at C1-2 in a 28-year-old woman with intractable SIH. After identifying the leakage point using both CT myelography and T-SLIP MRI, surgery was performed to seal the CSF leak. Intraoperatively, a pouch suggestive of an extradural arachnoid cyst around the left C2 nerve root was found, which was repaired by packing the pouch with muscle and fibrin glue. Clinical improvement was observed shortly after surgery, and postoperative imaging revealed the disappearance of the CSF leakage.

Conclusions: T-SLIP MRI may provide useful information on the flow dynamics of CSF in SIH patients due to high-flow leakage. However, further experience is required to assess its sensitivity and specificity as an imaging modality for identifying CSF leakage points.

Key Words: C1-2, cerebrospinal fluid leakage, CT myelography, spontaneous intracranial hypotension, time-spatial labeling inversion pulse (T-SLIP) MRI

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INTRODUCTION

Spontaneous intracranial hypotension (SIH) is a syndrome in which hypovolemia of the cerebrospinal fluid (CSF) results in various clinical symptoms.^[2,14] Identifying CSF leakage points in the spinal dura is important for treatment, particularly in patients with high-flow CSF leakage.^[6,9] Although computed tomography (CT) myelography has been the imaging modality of choice for this purpose, the importance of

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magnetic resonance imaging (MRI) is increasing owing to rapid technological advances.^[9,15] Time-spatial labeling inversion pulse (T-SLIP) is a novel noncontrast MRI technique, which uses CSF as an intrinsic visual tracer through an inversion recovery pulse that enables the tagged CSF to generate image contrast.^[17] Spine surgeons may benefit from this imaging technique by gaining a better understanding of CSF flow dynamics. The present study reports the novel application of T-SLIP MRI in the diagnosis and treatment of SIH caused by high-flow CSF leakage in the upper cervical spine.

CASE REPORT

A 28-year-old woman visited our clinic with a complaint of headache, which was aggravated by standing and relieved on lying down. She had been involved in a road traffic accident (RTA) approximately 1 year previously (she was a restrained driver at the time of a rear-end collision). MRI of the cervical spine (T2-weighted sagittal image) obtained shortly after the injury was considered within the normal range [Figure 1a]. She was alert and oriented, however, she could not maintain an upright posture due to a headache. SIH was suspected, and a brain MRI with gadolinium established the diagnosis through the identification of a diffuse dural meningeal enhancement on the axial T1-weighted image and the pituitary gland on the sagittal T1-weighted image. MRI of the cervical spine (T2-weighted sagittal image) revealed a decrease in the CSF space of the cervical spine compared with the previous MRI [Figure 1a and b]. A mild increase in the thickness of the retro-odontoid soft tissue was noted [Figure 1a and b]. There was no mechanical instability in the flexion-extension cervical spine X-rays. The accumulation of extradural CSF in the paraspinal

tissue at C1-2 was also shown in the heavily T2-weighted MRI [Figure 1c].

To identify the CSF leakage point in the spinal dura, CT myelography was performed. There was no spontaneous CSF outflow following spinal needle insertion, indicating negative CSF pressure. A CT myelogram, obtained 1 hour following the intrathecal injection of contrast material, revealed a massive CSF leakage at C1-2 [Figure 2a and b]. Although the spinal dura at C1-2 was suspected as the leakage point of CSF based on these radiographic examinations, T-SLIP MRI was performed to obtain further information on the CSF flow dynamics at that level. For T-SLIP MRI performed under an electrocardiogram-triggered procedure, 3T Vantage Titan (Toshiba Medical Systems Corporation, Tokyo, Japan) was used with the following settings: Repetition time, 19 R-R intervals; black blood time, 1800–3700 ms; interval, 100 ms; inversion pulse width, 1.4 cm. Twenty-eight consecutive images were collected and were edited using a software (Graphic Converter 9; Lemke Software GmbH, Peine, Germany) to produce a cinematic view to run in 5.6 s. With the cinematically reconstructed T-SLIP MRI [Supplementary video 1], the CSF outflow into the C1-2 epidural space on the left side was observed [Figure 3a-d]. Based on the CT myelography and T-SLIP MRI findings, the patient was diagnosed with SIH due to high-flow CSF leakage at C1-2. Following failed conservative management comprising bed rest and intravenous fluid administration for 2 weeks, two therapeutic options were suggested to the patient: Surgery or an epidural blood patch administered via a catheter inserted from the lower spine. After the patient selected the former option, she was taken to an operating theatre. Following a straight neck incision over the C1-3 laminae and partial C1 laminectomy, CSF

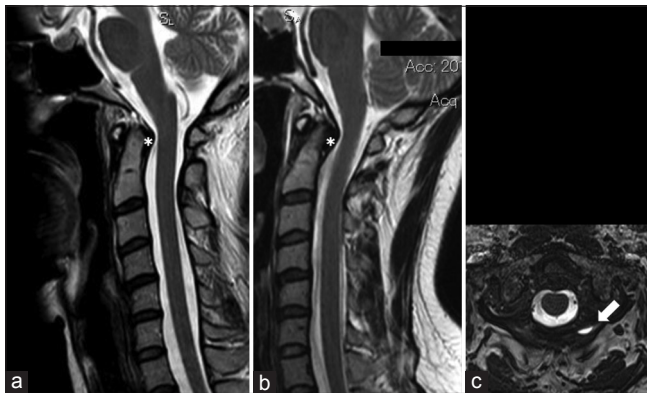


Figure 1: (a) Magnetic resonance imaging (MRI) of the cervical spine (T2-weighted sagittal image) obtained shortly following neck injury, which was considered within the normal range. (b) MRI of the cervical spine (T2-weighted sagittal image) at the time of admission, showing decreased cerebrospinal fluid (CSF) space. The increase in the thickness of the retro-odontoid tissue was also noted (asterisk). (c) Heavily T2-weighted MRI showing the accumulation of extradural CSF in the paraspinal tissue at C1-2 (arrow)

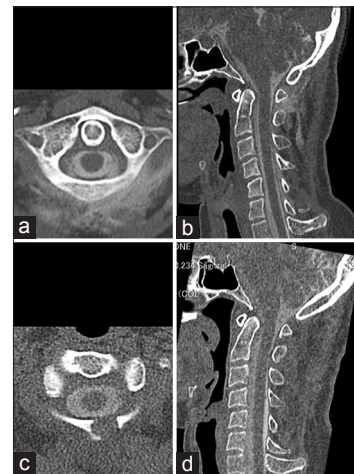


Figure 2: Computed tomography myelogram obtained following admission, showing massive leakage of contrast medium at C1-2 of the left side (a, axial view; b, sagittal view). At two weeks post-surgery, no leakage of contrast medium was observed (c, axial view; d, sagittal view)



Figure 3: Time-spatial labeling inversion pulse MRI showing the CSF flow dynamics. a/c, axial view; b/d, sagittal view. Compared with a/b, CSF leakage was observed in the C1-2 epidural space (circled) in c/d (c/d scanned 2.7 seconds after a/b)

collection was observed in the paraspinal tissue, and incessant CSF flow was observed from a pouch around the left C2 nerve root, which was considered to be an extradural spinal arachnoid cyst [Supplementary video 2]. The epidural space was packed with pieces of muscle and fibrin glue. The postoperative course was uneventful, and her headache disappeared 3 days after surgery. A repeat CT myelogram was obtained 14 days after surgery, which revealed no CSF leakage [Figure 2c and d], and she was discharged 20 days after admission. She has not sustained recurrence of SIH for more than 6 months. Permission for publication was granted by the patient.

DISCUSSION

Although SIH due to CSF leakage at C1-2 is less frequent than leakage occurring in the lower cervical or thoracolumbar spine,^[14] it poses both diagnostic and therapeutic challenges to spine surgeons. With regard to diagnosis, it has been reported that extradural CSF collection at C1-2 on conventional CT myelography or MRI may often be a false localizing sign;^[10,19] CSF leakage from the dura of the lower cervical spine may move upward. Therefore, care is required when identifying the precise leakage point in patients with extradural CSF collection at C1-2. The dynamic visualization of CSF outflow using imaging studies may be helpful and may provide evidence of CSF leakage at that particular level. Although the importance of dynamic digital subtraction myelography and CT myelography in identifying the CSF leakage point has been recognized,^[9,13,18] they present the major disadvantage of increased radiation exposure. In this context, identifying leakage points with MRI is beneficial in reducing the risk. While successful application of T-SLIP MRI has recently been reported in cases of cystic lesions of the spinal cord or

syringomyelia,^[5,12] the present report appears to be one of the first to successfully utilize T-SLIP MRI in the evaluation of patients with SIH. Using this technique, we were able to diagnose with confidence that CSF leakage occurred at the C1-2 level without performing dynamic CT or digital subtraction myelography, thus avoiding additional radiation exposure in a woman of reproductive age. The pouch observed intraoperatively may have been causally associated with the prior RTA.

With regard to therapeutics, there has been no consensus on the treatment algorithm for patients with CSF leakage at C1-2 who do not respond to conservative management. The use of conventional blood patch therapy administered from the lower lumbar spine may not be effective,^[14] considering the long distance between the lower lumbar and upper cervical spine. Furthermore, the risk of vascular and neural injury associated with percutaneous puncture at C1-2 may not be negligible, considering its anatomical complexity and proximity to the brainstem. Although the safety of blood patch therapy for patients with CSF leakage at C1-2 may have improved following the introduction of CT-guided puncture^[11] or the use of an epidural catheter inserted from the lower cervical spine,^[4,5] inadvertent damage to the engorged epidural venous plexus may be a possible complication. In this context, surgical exploration for a dural laceration or meningeal diverticula via a C1 hemilaminectomy under operative microscope may be warranted as a safe and effective treatment for high-flow CSF leakage at C1-2 refractory to medical treatment.^[3,7,16] Thus, injury to the epidural venous plexus may be minimized and dural repair can be performed on a case-by-case basis. There may be criticism of the treatment strategy used in the present case, in that it may have been better if an epidural blood patch had been attempted prior to resorting to surgery. In retrospect, however, the intraoperative finding of the incessant high-flow CSF leakage [Supplementary video 2] indicated that the leakage might not have been stopped using an epidural blood patch alone.

There are several limitations to the present case report. First, no data have been previously reported on the sensitivity/specificity of T-SLIP MRI for identifying the CSF leakage point in SIH patients. Low-flow CSF leakage may not be detectable with T-SLIP MRI; therefore, the examination may have low sensitivity and high specificity. Further, clinical experience will be required to make such a conclusion. Second, unlike dynamic CT myelography or digital subtraction myelography, T-SLIP MRI is not a real-time imaging modality, and at present, it can only be performed using MRI equipped with specific software.^[5,12] The actual speed of CSF outflow is twice that shown in Supplementary video 2. Third, it was not possible to make any conclusions regarding the pathoetiology of the pouch observed intraoperatively, as no surgical specimen was obtained. Based on her past history of trauma to the

neck, it was suggested that the pouch was a traumatic arachnoid cyst.^[1] Although a wider C1 laminectomy and meticulous inspection around the left C2 nerve root could have provided a better surgical view and allowed pouch ligation, which was not performed. Partial laminectomy was performed due to concern that a wider laminectomy and more aggressive muscle dissection might compromise the stability of the C1-2 joint in the future. Although there was no overt C1-2 instability on the flexion-extension X-rays, a causal association between the increase in the retro-odontoid mass and the development of CSF leakage cannot be ruled out.

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Conflicts of interest

On behalf of all authors, the corresponding author declares no conflict of interest.

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