

A New Mechanism of Cerebrospinal Fluid Leakage after Lumboperitoneal Shunt: A Theory of Shunt Side Hole—Case Report

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Abstract

Cerebrospinal fluid (CSF) overdrainage after lumboperitoneal (LP) shunt placement for the patients with idiopathic normal pressure hydrocephalus (iNPH) is mainly caused by insufficient management of pressure settings of the shunt valve and/or siphon effect of shunt systems induced by the patient's postural changes. We here report a unique case of intracranial hypotension (IH) due to CSF leakage after LP shunt placement in which another mechanism leads to the CSF leakage. A 67-year-old man suffered from persistent headache worsening with postural change 2 months after LP shunt reconstruction for iNPH. Brain computed tomography scan showed bilateral chronic subdural hematomas (CSDH). Lumbar images including shuntography and magnetic resonance imaging showed the tip of the lumbar catheter was spontaneously pulled out close to the dura mater with expansion of the epidural space due to CSF leakage from a shunt side hole of the lumbar catheter to the epidural space. Shunt removal and subsequent irrigation of CSDH improved his headache. CSF leakage in our case differs from those in previous reports, because early and enormous CSF leakage into the epidural space can be explained only by a different mechanism through a side hole just located in the epidural space in our case. We must pay attention to the possibility of this rare cause of IH due to CSF leakage in patients suffering from postural headache after LP shunt placement.

Key words: cerebrospinal fluid leakage, intracranial hypotension, lumboperitoneal shunt, subdural hematoma, spontaneous migration

Introduction

Headache after lumboperitoneal (LP) shunt placement for the patients with idiopathic normal pressure hydrocephalus (iNPH) is commonly attributed to intracranial hypotension (IH) due to cerebrospinal fluid (CSF) overdrainage.¹⁾ The IH is mainly caused by insufficient management of pressure settings of the shunt valve and/or siphon effect of the shunt system induced by the patient's postural changes.^{1,2)} But recently some cases of IH due to CSF leakage with other mechanisms have been reported.^{3,4)} We here report a unique case of IH after LP shunt placement in which another mechanism leads to the CSF leakage, from the side hole of the shunt catheter to the lumbar epidural space. Therapeutic strategies for patients with

chronic subdural hematoma (CSDH) associated with IH are also discussed.

Case Report

A previously healthy 67-year-old man was brought to our hospital because of gait and memory disturbances and urinary incontinence. He was diagnosed with iNPH. The lumbar puncture revealed CSF pressure of 140 mmH₂O. LP shunt placement was performed using a Codman® Hakim® programmable valve with SIPHONGUARD® (Johnson & Johnson Co., Raynham, Massachusetts, USA). In this procedure, the lumbar catheter was inserted through the dura mater with a Tuohy needle at the level between the 2nd and 3rd lumbar lamina (L2/3). His symptoms diminished after the surgery with a decrease of final valve opening pressure to 120 mmH₂O. Two months after the

surgery, his symptom of gait disturbance worsened soon after twisting his body at the waist while working in the yard. Further examination revealed shunt malfunction due to disconnection of the caudal shunt tube from the shunt valve and consequential obstruction of the lumbar catheter. The revision of the LP shunt system was done using the same valve system after insertion of a new lumbar catheter, 13 cm from the fascia at L3/4 after multiple dural punctures with a Tuohy needle (Fig. 1). Because of his postural headache, the valve opening pressure was changed from 120 mmH₂O of initial setting pressure to 200 mmH₂O in a stepwise fashion.

Two months after the second surgery, worsening of headache and appearance of nausea and anorexia, without any traumatic episode, brought him to our hospital. His consciousness level was alert with no focal neurological deficits and with persistent headache worsening with postural change. Computed tomography (CT) of the brain showed bilateral CSDH compressing the adjacent brain and slit like ventricles (Fig. 2B). Sagittal view images of lumbar magnetic resonance imaging (MRI) and CT scan showed the tip of the shunt catheter was spontaneously pulled out and located close to the aforementioned dura mater in the ventral side of the subdural space (Fig. 3A, B)

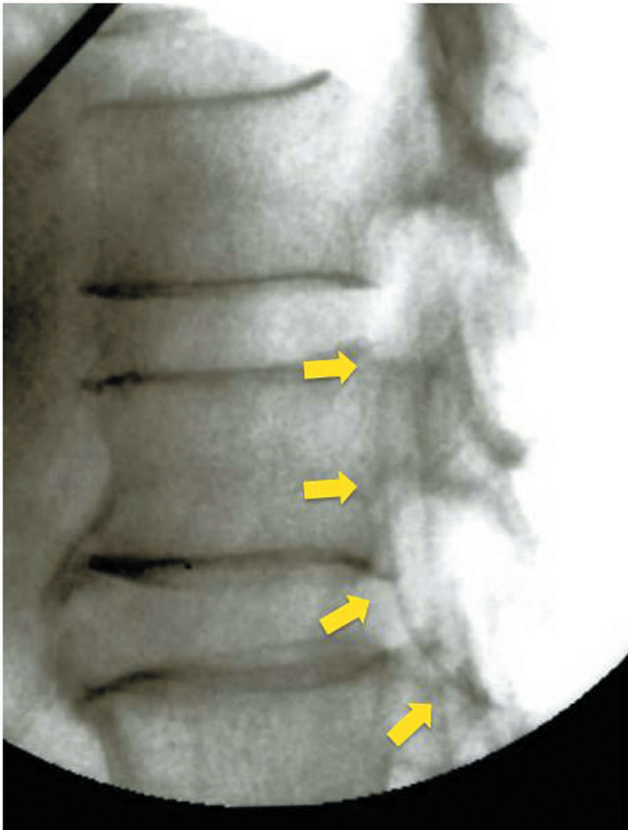


Fig. 1 A: An X-ray image taken during the second operation of lumboperitoneal (LP) shunt placement shows that more than 7 cm of the shunt catheter locates in the subdural space (arrows).

CT-myelography, punctured at the level between L4/5 to prove the leak point, revealed that contrast medium was spread along with the nerve root at the levels from Th12/L1 to L3/4 (Fig. 3C, D), indicating CSF leakage spreading into epidural space existed although the leak point was not detected clearly. Strict bed rest, analgesia for headache, and hydration failed to improve the symptoms. Next, the shunt catheter ligation, at just proximal of the reservoir, also did not improve his symptoms. For further evaluation, shuntography after releasing the ligation and subsequent CT scan revealed the epidural spread of the contrast medium, corresponding to the epidural space on thin slice T₂-weighted images on MRI (Fig. 4). Thin slice T₂-weighted fat suppression images showed the epidural space corresponded to CSF intensity. This phenomenon suggested the CSF leakage related to the shunt system. Considering the possibility of the CSF leakage from the side hole to the epidural space as well as minor leakage from the tip of the lumbar catheter (Fig. 5), shunt removal was done.

Soon after the shunt removal, his postural headache diminished, but the headache itself worsened. Two weeks later, his cognitive function had been deteriorated and CT revealed the worsening of the bilateral CSDH (Fig. 2C). On the other hand, lumbar MRI showed disappearance of expansion of the epidural space (Fig. 4D). The patient underwent bilateral burr-hole drainage of the CSDH (Fig. 6A). The opening pressure of the subdural space was positive (80 mmH₂O [right] and 30 mmH₂O [left] from the outer plate). His cognitive dysfunction and persistent headache resolved immediately. Ventriculoperitoneal (VP) shunt placement was done (Fig. 6B, C) 3 months after the drainage because of gradual regression of the gait and memory disturbances. The patient had no neurological symptoms 1 year after the first LP shunt placement.

Discussion

We report a unique case of IH due to CSF leakage after LP shunt; the main route of CSF leakage was thought to be from the side hole of the shunt catheter to the epidural space caused by slip-out of the lumbar catheter. Such migration of LP shunt catheter is not so rare event. Previous studies show that 12–23% of LP shunt patients had shunt malfunctions resulting from migration (or fracture) of the lumbar or peritoneal catheter.^{1,5,6)} On the other hand, symptomatic IH after LP shunt is commonly attributed to improper pressure settings of the shunt valve and/or siphon effects of shunt systems, which is observed in 15–20% after LP shunt.^{1,2)} However, IH due to other reasons including catheter migrations is very rare. In a previous report 2 out of 93 patients (2.1%) had IH owing to CSF leakage from a penetration hole of the inserted spinal catheter.³⁾ This phenomenon is similar to CSF leakage into the paraspinal area after a routine lumbar puncture.⁷⁾ However, our case differs

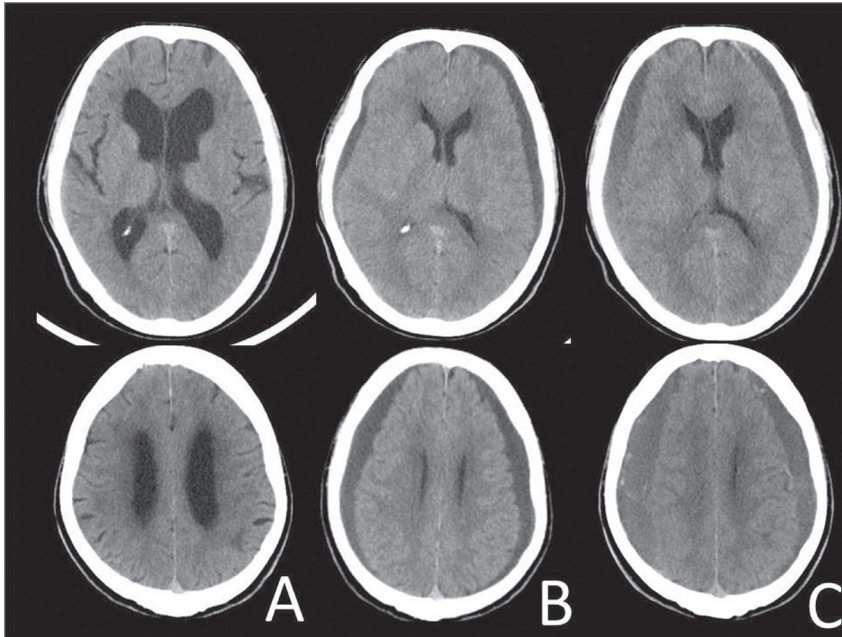


Fig. 2 A: Computed tomography (CT) scans before lumboperitoneal shunt surgery show ventricular enlargement, compatible with idiopathic normal pressure hydrocephalus. B: CT scans taken on admission show bilateral chronic subdural hematoma (CSDH) compressing the adjacent brain with slit like ventricles. C: CT scans 2 weeks after the shunt removal reveal the worsening of the bilateral CSDH.

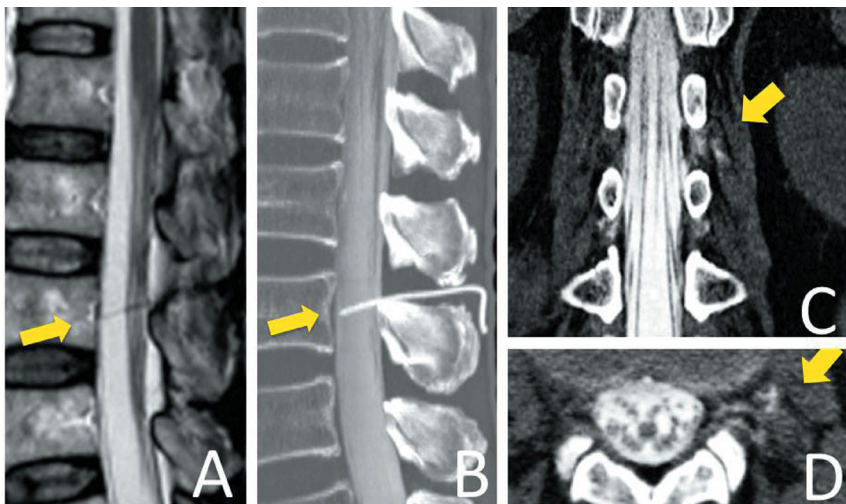


Fig. 3 A, B: Sagittal view images of lumbar computed tomography (CT) scan and magnetic resonance imaging showing that the tip of the shunt catheter is spontaneously pulled out and located between the 3rd and 4th lumbar levels and close to the dura mater in the ventral side of the subdural space (arrow). C, D: Coronal (C) and axial (D) views of CT-myelography before the shunt removal reveal spreading of the contrast medium along with the nerve root at the upper lumbar and the lower thoracic levels (arrows).

from the previous reports, because early and enormous CSF leakage into the epidural space can be explained only by a different mechanism, through a side hole of the lumbar catheter just adjusted to the epidural space, but not simply through the penetration hole of the catheter (see Fig. 5). The previously reported CSF leakage from the ventral dural sac was probably caused by inadvertent puncture or erosion of the dura mater after LP shunt placement.⁴⁾ Similarly, there may be small holes or erosion in the ventral dura mater in the present case. However, we consider that it was not the main cause, as the shunt removal was effective in this patient. If the main route of CSF leakage was from the small dura holes around the tip of the shunt catheter, the shunt removal would not lead to the symptoms improvement. Thus we

concluded that the CSF leaked was from the side hole of the lumbar catheter.

The IH due to the CSF overdrainage or leakage causes several symptoms including headache via meningeal traction and subdural effusions associated with a venous dilatation (Monro-Kellie hypothesis),⁸⁾ and subdural hematomas via the rupture of bridging veins.⁹⁾ Therapeutic measures for the IH include bed rest, increased volume intake, infusions of caffeine, epidural injection of venous blood, and direct surgical repair of the dural tear.¹⁰⁾ But the therapeutic strategies for CSDH associated with IH are not yet confirmed. The incidence of CSDH among patients with spontaneous IH patients ranges from 20% to 40%.¹¹⁾ Bed rest, increased volume intake as well as shunt ligation, and high pressure setting of the shunt valve are effective

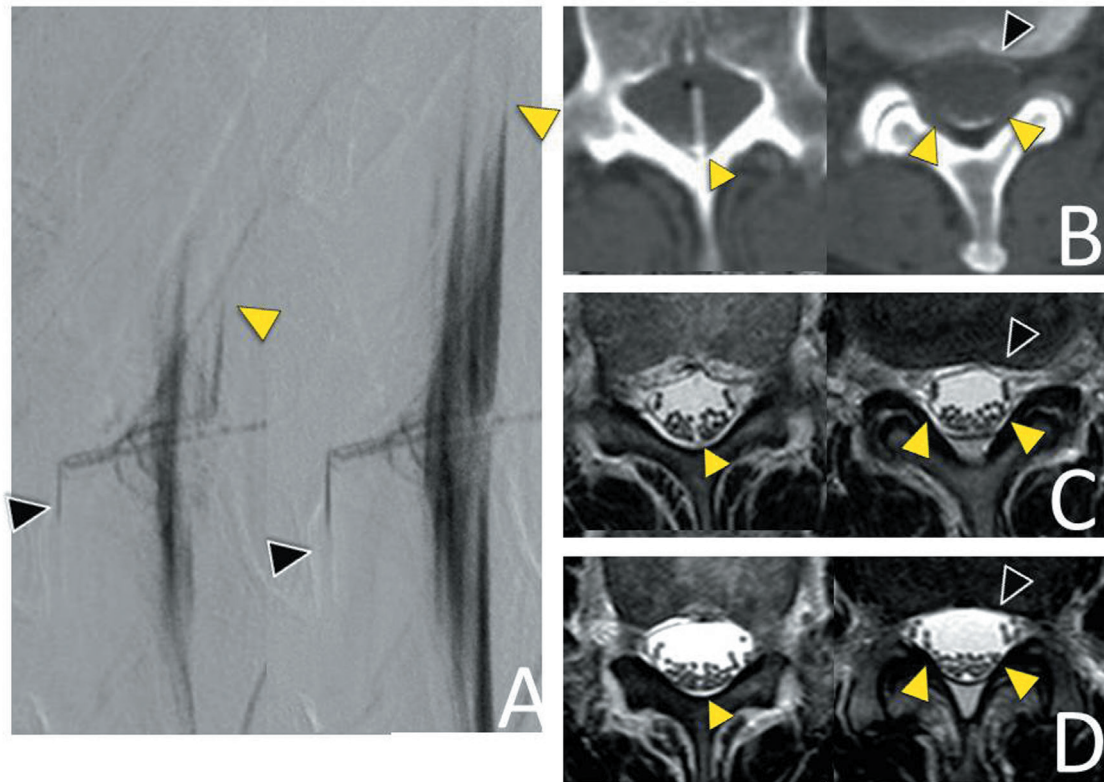


Fig. 4 A: In the early phase of shuntography (*left*) contrast medium is pushed into the subdural space through the shunt catheter. Early spread of the contrast medium into the epidural space is shown (*yellow arrowhead*), coincident with spread into the subdural space. In the subsequent phase (*right*) contrast medium emanates from the upper lumbar to the lower thoracic level of the epidural space with a negative pressure gradient (*yellow arrowheads*). In addition, there is a minor cerebrospinal fluid (CSF) leakage from the tip of the shunt catheter (*black arrowhead*) probably due to multiple dural punctures. B: Computed tomography (CT) scans soon after the shuntography reveal the contrast medium leakage around the inserted lumbar catheter and spreading into the epidural space (*black and yellow arrowheads*). C: T₂-weighted image (T₂WI) on magnetic resonance imaging at the same level as the CT scans before shunt removal shows the epidural space is expanded at the lumbar level (*black and yellow arrowheads*). D: T₂WI after the shunt removal shows disappearance of expansion of the epidural space.

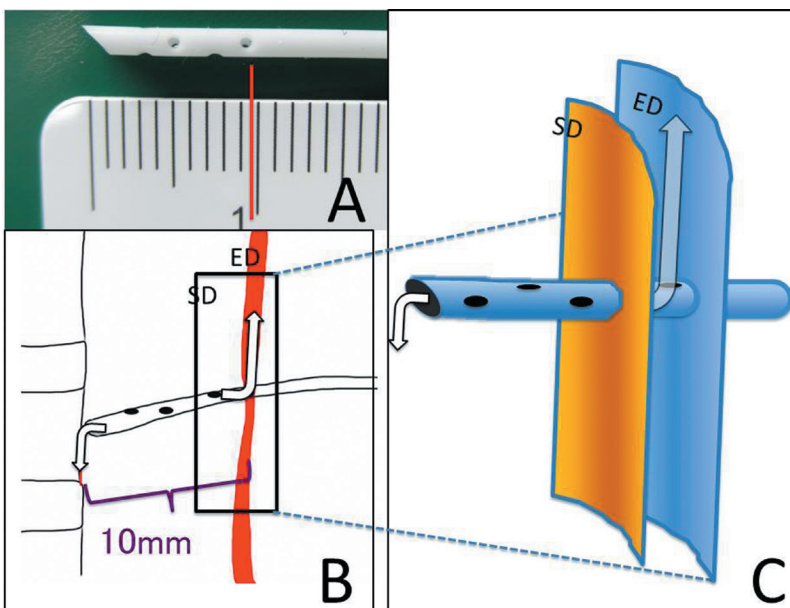


Fig. 5 An actual figure of the inserted catheter (A) and schemes of the cerebrospinal fluid (CSF) leakage in the present case (B and C) revealed by several images shown in Figs. 3 and 4. The red area means the epidural space. As shown in A and B, actual length between the tip of the shunt catheter and the epidural space corresponds to one of side holes setting 9–10 mm from the tip. The schemes describe two routes of CSF leakage; the main route from the shunt side hole, and another from a small dura hole around the tip of the shunt catheter. ED: epidural space, SD: subdural space.

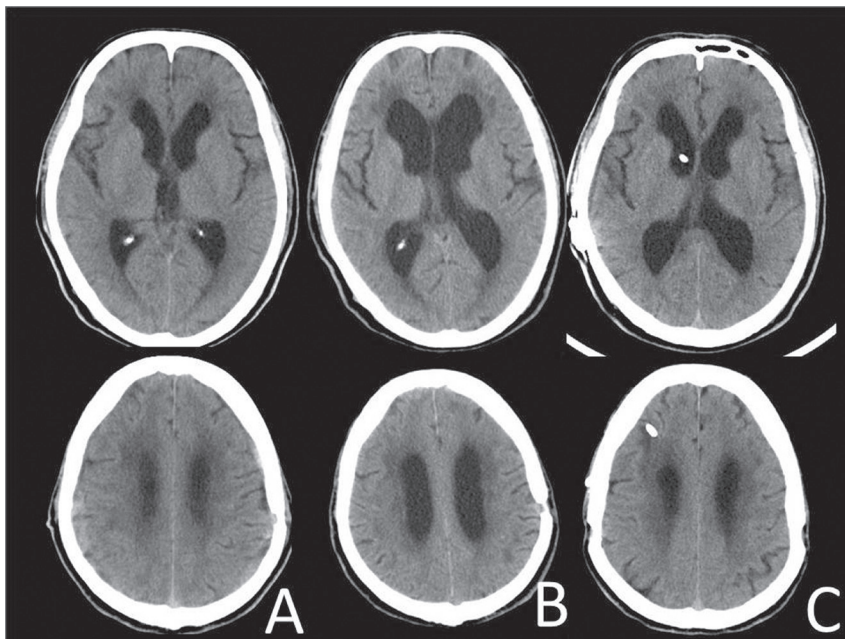


Fig. 6 A: Computed tomography (CT) scans taken soon after the burr-hole drainage show disappearance of the hematoma. B: CT scans before ventriculoperitoneal (VP) shunt shows the ventricle size is quite large with Evans index (0.44). C: CT scans just after VP shunt shows the ventricle size is improving.

for both of IH and CSDH. But the efficacy of hematoma removal before treatment for IH is controversial. There is a case report of two patients with CSDH associated with spontaneous IH for which hematoma removal alone was effective.¹² In contrast, hematoma removal without treatment for IH resulted in a hematoma recurrence in some cases of CSDH^{9,13,14} as well as acute subdural hematoma (ASDH) cases.¹⁵ ASDH could also develop after drainage for CSDH associated with IH, where one patient fell into coma after the drainage.^{11,16} We emphasize that hematoma removal should be taken into account after the management of CSF leakage is made.

In conclusion, here we described the first case of CSDH associated with CSF leakage caused by a unique mechanism after LP shunt placement. The main CSF leakage to the epidural space occurred from the shunt side hole of the lumbar catheter. We should pay attention to the possibility of such rare cause of CSF leakage in patients suffering from postural headache after LP shunt placement.

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Conflicts of Interest Disclosure

The authors have no personal, financial, or institutional interest in any of the drugs, materials, or devices in the article. All authors who are members of The Japan Neurosurgical Society (JNS) have registered online Self-

reported COI Disclosure Statement Forms through the website for JNS members.

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