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Lumbar Catheter Misplacement into the Spinal Subdural Epiarachnoid Space Causing Lumboperitoneal Shunt Malfunction: Report of Two Cases

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Abstract

Lumboperitoneal (LP) shunting is a standard treatment for idiopathic normal pressure hydrocephalus (iNPH), with equivalent efficacy to ventriculoperitoneal (VP) shunting, and it is associated with a favorable outcome in approximately 75% of patients with iNPH. Despite the advantages, LP shunting can result in problems associated with the lumbar catheter, the obstruction of which has not been well described. This report presents two cases of LP shunt malfunction caused by lumbar catheter misplacement into the spinal subdural epiarachnoid space (SSES), and by subsequent obstruction. A 67-year-old man and a 69-year-old woman with iNPH underwent LP shunt placement without intraoperative fluoroscopy. Shortly after the surgery, they experienced a temporary improvement of their symptoms which was, however, followed by recurrence within a few months. This was suggestive of shunt malfunction. Although shunt pumping tests were normal, shuntography and subsequent computed tomography (CT) revealed lumbar catheter misplacement into the SSES. Shunt revisions, in which only the lumbar catheters were exchanged, were performed with intraoperative fluoroscopy and shuntography. Their symptoms have improved again following the revisions. In the present cases, lumbar catheter misplacement into the SSES caused LP shunt malfunction, and shuntography and CT were useful to detect the abnormality. Moreover, unrecognized lumbar catheter misplacement into the SSES might potentially have occurred in some patients considered as "nonresponders" to LP shunting; hence, shuntography may be useful in those patients.

Keywords: idiopathic normal pressure hydrocephalus, lumboperitoneal shunt, shunt malfunction, shuntography, spinal subdural epiarachnoid space

Introduction

Idiopathic normal pressure hydrocephalus (iNPH) is a clinical syndrome characterized by gait disturbance, cognitive decline, and urinary incontinence.

iNPH affects patients' daily-life activities and decreases their quality of life. The prevalence of iNPH is estimated to be 1.1%-2.1% in elderly people,^{1,2)} and the number of patients is increasing in line with an increasingly aging society. Accordingly, the clinical importance of iNPH is increasing, as it represents a treatable disease in elderly patients.

Recently, lumboperitoneal (LP) shunting has become a standard treatment for iNPH alongside ventriculoperitoneal (VP) shunting. An equivalent efficacy of LP shunting compared to VP shunting for iNPH

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has been suggested by recent prospective studies (SINPHONI-2),^{3,4)} and favorable outcomes after LP shunting are reported in 75%-81% of patients.^{3,5,6)}

Despite the advantages, LP shunting can be associated with unique problems that result from placement of a lumbar catheter, such as difficulties in lumbar puncture, delayed catheter rupture, or severing of the catheter by the spinous processes. To the best of our knowledge, however, lumbar catheter obstruction following LP shunt placement has rarely been described.

We here present two cases of LP shunt malfunction caused by misplacement of the lumbar catheter into the spinal subdural epiarachnoid space (SSES), which resulted in obstruction. In these cases, shuntography and subsequent computed tomography (CT) were useful to detect the abnormality.

Case Reports

Case 1

A 67-year-old man presented with gait disturbance, cognitive decline, and urinary incontinence. The results of the Mini-Mental State Examination (MMSE) and of the timed 3 m up-and-go test (TUG) were 16 points and 26.4 seconds, respectively. The total score of the iNPH grading scale (iNPHGS)⁷⁾ was 8, and that of each domain was as follows: gait disturbance (g), 2; cognitive impairment (c), 3; and urinary disturbance (u), 3. Magnetic resonance imaging (MRI) showed ventriculomegaly with an Evans' index of 0.38, and disproportionately enlarged subarachnoid space hydrocephalus (DESH). A tap test transiently improved his symptoms and he was diagnosed as having probable iNPH.

LP shunt placement using a Strata NCS LP adjustable pressure shunt (Medtronic, Minneapolis, MN, USA) was performed without intraoperative fluoroscopy. In this procedure, a Tuohy needle was inserted by a median approach, and, at the fifth needle insertion, cerebrospinal fluid (CSF) outflow through the needle was confirmed. Subsequently, a lumbar catheter was advanced smoothly through the needle and CSF outflow through the catheter was also confirmed. His symptoms improved shortly after the surgery, namely, the results of the MMSE, the TUG, and the iNPHGS were, respectively, 21 points, 10 seconds and 4 (g 1; c 1; u 2) one week after the shunting. However, his symptoms reappeared 3 months later, the score of iNPHGS declined to 6 (g 2; c 2; u 2), and he did not respond to adjusting the pressure settings from performance level 1.5 to 1.0. Although shunt malfunction was suspected, the brain MRI and shunt device X-ray did not show any abnormalities and the shunt pumping test was normal.

We then performed shuntography. By puncturing the reservoir with a 27-gauge needle, we could not aspirate CSF. Subsequently, we injected the water-soluble nonionic iodinated contrast agent iohexol (Omnipaque 240 mg I/mL i.e., 518 mg iohexol/mL, GE Healthcare, Marlborough, MA, USA). The medium flowed through both the lumbar and the peritoneal catheter without obvious obstruction. In the spinal canal, however, it pooled without diffusion (Fig. 1A). Subsequent CT revealed both pooling of the contrast medium and presence of the tip of the lumbar catheter in the SSES (Fig. 1B and 1C). Thus, the catheter seemed to be inserted via the ventral side of the thecal sac into the SSES (Fig. 1D). We concluded that lumbar catheter misplacement caused the shunt malfunction, and we proceeded to revise the shunt system. Under fluoroscopy, we exchanged only the lumbar catheter via a paramedian approach, using the same shunt valve, and the catheter position was confirmed by intraoperative shuntography. After the revision, his

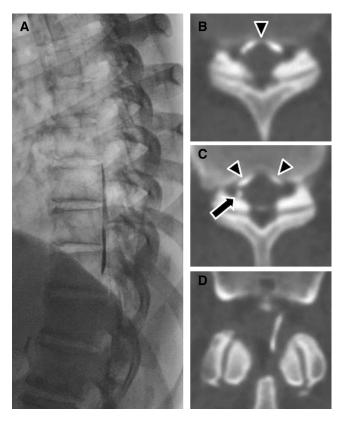


Fig. 1 Imaging studies of case 1. Shuntography shows pooled contrast medium without diffusion in the spinal canal (A). Subsequent CT shows pooled medium (arrowheads) and a misplaced lumbar catheter (arrow) in the SSES (B and C). The lumbar catheter appeared to be inserted into the SSES via the ventral side of the thecal sac because of deep puncture (D). CT: computed tomography, SSES: spinal subdural epiarachnoid space.

symptoms improved again and his iNPHGS score was 3 (g 1; c 1; u 1) two years after the revision.

Case 2

A 69-year-old woman presented with gait disturbance and urinary incontinence. The results of the TUG, the MMSE, and the iNPHGS were 15.2 seconds, 30 points, and 6 (g 2; c 1; u 3), respectively. MRI showed ventriculomegaly and the Evans' index was 0.39 without DESH. A tap test transiently improved her symptoms and she was diagnosed as having probable iNPH.

She underwent LP shunt placement using a Strata NCS LP adjustable pressure shunt. As with case 1, this surgery was performed without intraoperative fluoroscopy. A Tuohy needle was inserted via a median approach, and, at the first needle insertion, CSF outflow through the needle was confirmed. A lumbar catheter was advanced smoothly through the needle and CSF outflow through the catheter was also confirmed. Her symptoms improved shortly after the surgery, and the iNPHGS score had decreased to 4 (g 2; c 0; u 2) one month after surgery. However, her symptoms gradually worsened, leading to an

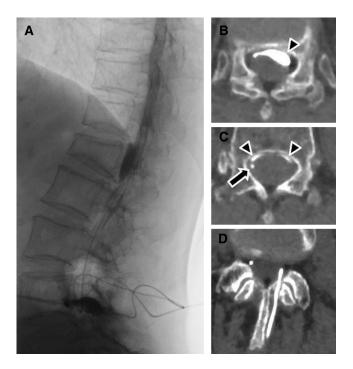


Fig. 2 Imaging studies of case 2. Shuntography shows pooled contrast medium without diffusion in the spinal canal (A). Subsequent tomography shows pooled medium (arrowheads) and a misplaced lumbar catheter (arrow) in the SSES (B and C). The lumbar catheter appeared to be inserted into the SSES via the lateral part of the thecal sac because of lateralized puncture (D). SSES: spinal subdural epiarachnoid space.

iNPHGS score of 6 (g 2; c 1; u 3) seven months after the initial surgery. The pressure setting of the shunt valve was gradually lowered from performance level 2.0 to 0.5 but her symptoms did not improve. Although shunt malfunction was suspected, cranial CT and shunt device X-ray did not show any abnormalities, and the shunt pumping test was normal.

We performed shuntography. By puncturing reservoir with a 27-gauge needle we could not aspirate CSF. Subsequently, we injected the contrast agent iohexol (Omnipaque 240 mg I/mL). The medium flowed through both the lumbar and the peritoneal catheter without obvious obstruction, and it spread partially in the spinal canal (Fig. 2A). Subsequent CT revealed both pooling of contrast medium and presence of the tip of the lumbar catheter in the SSES (Figs. 2B and 2C). Thus, the catheter appeared to be inserted into the SSES via the lateral part of the thecal sac (Fig. 2D). As with case 1, revision of the LP shunt was performed. Under fluoroscopic guidance, we exchanged only the lumbar catheter via a paramedian approach using the same shunt valve, and the catheter position was confirmed by intraoperative shuntography. After the revision, her symptoms ameliorated and the iNPHGS score was 5 (g 2; c 1; u 2) nine months after the revision.

Discussion

We describe two cases of LP shunt malfunction caused by lumbar catheter misplacement into the SSES. Shuntography and subsequent CT were useful to detect the abnormality.

Lumbar catheter misplacement into the SSES and consequent obstruction can cause LP shunt malfunction. Lumbar catheter obstruction following LP shunt insertion has rarely been reported. In a previous report of 51 LP shunts, two cases of lumbar catheter misplacement into the SSES were mentioned; however, the details were not described.⁶⁾ In other procedures involving lumbar catheterization, two cases of intrathecal drug delivery failure and a case of epidural anesthesia failure with catheter misplacement into the SSES were reported.^{8,9)}

According to anatomical reports, the SSES was usually non-existent due to adherence of the dura and arachnoid mater¹⁰; however, the lumbar puncture separated the layers and created the space.¹¹ In the present cases, the symptoms temporarily improved for a few months after the shunt was inserted despite lumbar catheter misplacement into the SSES. The improvement interval might correspond to the repair time of these membranes. Another possible explanation for the improvement interval is the occurrence of CSF leakage. CSF leakage is a pathological condition well known to occur after lumbar puncture, and it may lead to temporary improvement of clinical symptoms after LP shunting.

Shuntography and subsequent CT revealed the lumbar catheter misplacement. To provide more detail, shuntography showed shunt patency and abnormal distribution of injected contrast medium in the spinal canal, and subsequent CT showed both pooling of the contrast medium and presence of the catheter tip in the SSES. In this condition, CSF could not be aspirated via the shunt reservoir, although contrast medium could be injected. Shunt contrast studies are recommended by Japanese guidelines for iNPH in those patients with a suspicion of shunt obstruction.¹⁾ In the present cases, shunt pumping tests could not detect shunt malfunction. This is consistent with the fact that the sensitivity of the shunt pumping test to detect shunt malfunction has been reported to be only approximately 20%.12)

Intraoperative fluoroscopic guidance and shuntography were useful in the revision of the shunt. Although intraoperative shuntography was reliable to confirm the catheter tip position, we do not perform it in all patients, because the use of contrast medium presents some risks such as allergic reactions. Following the above-mentioned conditions, we would recommend using this procedure in patients with suspicion of lumbar catheter misplacement, as indicated by slow CSF outflow or resistance to lumbar catheter insertion. In most patients, we perform LP shunting without intraoperative fluoroscopy.

In the present cases, despite confirmation of CSF outflow through the lumbar catheter, misplacement of the lumbar catheter had occurred. In other words, the presence of CSF outflow does not indicate an appropriate placement of the lumbar catheter. However, smooth CSF outflow from a lumbar catheter is still regarded as a marker of an appropriate placement. Therefore, one should observe it carefully. Unfortunately, further details of CSF outflow were not recorded in these cases. In future research, it could be interesting to analyze potential differences in CSF outflow between correctly placed and misplaced catheters.

In previous research related to LP shunting with intraoperative fluoroscopy, no occurrence of lumbar catheter obstruction was reported.^{13,14} Considering the results of these reports, intraoperative fluoroscopy may reduce the risk of this complication through avoiding deep or lateralized lumbar puncture.

Although the present patients experienced improvement of their symptoms after the initial

LP shunting despite lumbar catheter misplacement into the SSES, it is unknown whether all such patients experience symptom improvement after surgery. The favorable outcome rate after LP shunting has been reported to be approximately 75%.^{3,5,6]} Unrecognized lumbar catheter misplacement into the SSES might potentially have occurred in some patients considered "non-responders" to LP shunting, Hence, shuntography may be useful for these patients. Further studies should be performed to investigate this.

In conclusion, our findings reveal that lumbar catheter misplacement into the SSES can cause LP shunt malfunction. Shuntography and subsequent CT were useful to detect the abnormality. One must pay particular attention during surgery to avoid lumbar catheter misplacement in LP shunting.

Conflicts of Interest Disclosure

The Department of Neuromodulation and Neurosurgery, Osaka University Graduate School of Medicine is a joint research department established with sponsorship from Teijin Pharma Limited. All authors declare no conflict of interest.

References

- Mori E, Ishikawa M, Kato T, et al.: Guidelines for management of idiopathic normal pressure hydrocephalus: second edition. *Neurol Med Chir (Tokyo)* 52: 775–809, 2012
- Jaraj D, Rabiei K, Marlow T, Jensen C, Skoog I, Wikkelsø C: Prevalence of idiopathic normal-pressure hydrocephalus. *Neurology* 82: 1449–1454, 2014
- Kazui H, Miyajima M, Mori E, Ishikawa M; SINPHONI-2 Investigators: Lumboperitoneal shunt surgery for idiopathic normal pressure hydrocephalus (SINPHONI-2): an open-label randomised trial. *Lancet Neurol* 14: 585–594, 2015
- 4) Miyajima M, Kazui H, Mori E, Ishikawa M; on behalf of the SINPHONI-2 Investigators: One-year outcome in patients with idiopathic normal-pressure hydrocephalus: comparison of lumboperitoneal shunt to ventriculoperitoneal shunt. *J Neurosurg* 125: 1483–1492, 2016
- 5) Giordan E, Palandri G, Lanzino G, Murad MH, Elder BD: Outcomes and complications of different surgical treatments for idiopathic normal pressure hydrocephalus: a systematic review and meta-analysis. J Neurosurg 131: 1024–1036, 2019
- 6) Nakajima M, Miyajima M, Ogino I, et al.: Use of external lumbar cerebrospinal fluid drainage and lumboperitoneal shunts with Strata NSC valves in idiopathic normal pressure hydrocephalus: a single-center experience. *World Neurosurg* 83: 387-393, 2015

- 7) Kubo Y, Kazui H, Yoshida T, et al.: Validation of grading scale for evaluating symptoms of idiopathic normal-pressure hydrocephalus. *Dement Geriatr Cogn Disord* 25: 37–45, 2008
- 8) Delhaas EM, Harhangi BS, Frankema SPG, Huygen FJPM, van der Lugt A: Catheter access port (computed tomography) myelography in intrathecal drug delivery troubleshooting: a case series of 70 procedures. *Neuromodulation* doi: 10.1111/ner.13153. Epub 2020 Apr 8.
- 9) Crosby ET, Halpern S: Failure of a lidocaine test dose to identify subdural placement of an epidural catheter. *Can J Anaesth* 36: 445–447, 1989
- 10) Reina MA, De Leon Casasola O, López A, De Andrés JA, Mora M, Fernández A: The origin of the spinal subdural space: ultrastructure findings. *Anesth Analg* 94: 991–995, table of contents, 2002
- 11) Blomberg RG: The lumbar subdural extraarachnoid space of humans: an anatomical study using spinaloscopy in autopsy cases. *Anesth Analg* 66: 177–180, 1987

- 12) Piatt JH: Physical examination of patients with cerebrospinal fluid shunts: is there useful information in pumping the shunt? *Pediatrics* 89: 470–473, 1992
- 13) Tucker A, Kajimoto Y, Ohmura T, et al: Fluoroscopic-guided paramedian approach for lumbar catheter placement in cerebrospinal fluid shunting: assessment of safety and accuracy. *Oper Neurosurg (Hagerstown)* 16: 471–477, 2019
- 14) Yang TH, Chang CS, Sung WW, Liu JT: Lumboperitoneal shunt: a new modified surgical technique and a comparison of the complications with ventriculoperitoneal shunt in a single center. *Medicina (Kaunas)* 55: 643, 2019

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