Treatment of Spontaneous Intracranial Hypotension Secondary to C-2 Meningeal Cyst by Surgical Packing
—Case Report—

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Abstract
A 41-year-old man presented with progressive worsening of postural headache. Computed tomography (CT) showed bilateral subdural hematomas without prior history of trauma. The diagnosis was spontaneous intracranial hypotension (SIH). Conservative treatment with oral steroids failed to prevent gradual deterioration of the patient's consciousness. CT myelography revealed massive cerebrospinal fluid (CSF) leakage between the C-1 and C-2 levels. The leak was repaired surgically via a laminectomy. A cyst, thought to be a meningeal cyst, was discovered adjacent to the right C-2 nerve root, and CSF was seen seeping out from around the cyst after a Valsalva maneuver. The presumed dural defect of the cyst was sealed by packing with muscle fragments and fibrin glue. The symptoms disappeared soon after surgery. He was discharged 1 month after surgery without deficits. Most SIH cases are benign and can be managed conservatively, or by the epidural blood patch method. Surgery is more invasive than the epidural blood patch method, but should be performed in patients with a high cervical lesion and massive CSF leakage.

Key words: cerebrospinal fluid leakage, meningeal cyst, spontaneous intracranial hypotension, surgical packing, upper cervical cord

Introduction
Spontaneous intracranial hypotension (SIH) was once considered rare, but is now a common disease in neurosurgical practice,2,4,13,18) and is a fairly common cause of postural headache among adults.13,17) The etiology and pathophysiology of SIH is better understood as a result of recent advances in neuroradiological equipment including computed tomography (CT) myelography,3) radionuclide cisternography,1,10,21) and magnetic resonance (MR) myelography,4) but the treatment strategy remains undecided. Most cases of SIH are benign and can be managed conservatively,17) or by the epidural blood patch method if conservative treatment fails.1,4,6,13–15) However, surgical treatment may be indicated if the epidural blood patch fails, or is likely to be ineffective or risky.8,11) We treated a patient with SIH secondary to a suspected meningeal cyst in the upper cervical spine by surgical packing.

Case Report
A previously healthy 41-year-old man presented with a 2-week history of postural headache. The headache had initially been relieved by lying flat, but became progressive and refractory to postural change. The patient was afebrile with no meningeal signs, and he denied any recent trauma. No focal neurological signs were noted. He was lethargic and disoriented. Brain CT revealed bilateral subdural hematomas (SDHs) and descent of the brain into the posterior fossa with obliteration of the basal cisterns. Brain MR imaging revealed diffuse...
Fig. 1  T₁-weighted magnetic resonance (MR) images showing diffuse pachymeningeal contrast enhancement (A, axial view), descent of the cerebellar tonsils into the foramen magnum, and downward displacement of the optic chiasm (B, sagittal view). The patient was stuporous, so the MR imaging was affected by motion artifacts.

Fig. 2 Computed tomography myelograms showing massive cerebrospinal fluid leakage between the C-1 and C-2 levels, predominantly on the right. A: axial view, B: sagittal view.

Fig. 3 Indium-111 diethylene triamine pentaacetate radionuclide cisternograms confirming massive cerebrospinal fluid leakage at the craniocervical junction. A: anteroposterior view, B: lateral view, 1 hour post-injection.

Surgical Treatment of Intracranial Hypotension

We had planned to use the epidural blood patch method after conservative treatment failed, but the massive CSF leakage demonstrated by CT myelography and the technical uncertainty regarding insertion of an epidural needle at the craniocervical junction prompted us to choose surgical repair of the leak. C-1 and partial C-2 laminectomy was carried out with the patient in the prone position. The engorged epidural venous plexus was coagulated, and the C-2 nerve root on the right was exposed. A cystic mass was discovered adjacent to the nerve root using the operating microscope, which appeared to be a meningeal cyst originating from the C-2 nerve root (Fig. 4). A Valsalva maneuver was performed by the anesthesiologist, and escape of CSF was observed around the cyst, although no definite tear of the cyst was identified. Pieces of muscles and fibrin glue were packed epidurally around the cyst and the bilateral C-2 nerve roots to completely seal off the leak. The dura mater covering the upper cervical cord and the nerve roots appeared normal, and no intradural exploration or resection of the cyst was performed. A repeat Valsalva maneuver after the procedure caused no CSF escape. The cervical incision was closed and the bilateral chronic SDHs were evacuated by a simple drainage method.
The patient became alert and oriented the day after surgery, and the headache stopped several days after the operation. Follow-up CT myelography 2 weeks postoperatively showed no CSF leakage between the C-1 and C-2 levels (Fig. 5). MR imaging 2 weeks after surgery showed that the brain was no longer distorted (Fig. 6). He was discharged without neurological deficits 1 month after surgery.

Discussion

The etiology of SIH is varied, but dural and meningeal tears located in the spinal column and consequent CSF leakage have been implicated as a common pathophysiological mechanism in the majority of cases. CSF leakages have been observed at the cervicothoracic junction and in the upper thoracic and lumbar spinal column, but leakage in the upper cervical spinal column is rare. The causes of dural tears include trauma, disc hernia, and prior lumbar puncture, and meningeal cyst (diverticula). Meningeal cysts at the exit of a nerve root acting as the source of a CSF leakage are rarely observed macroscopically, because SIH is not often treated by surgery. Intraoperative examination in our patient found a cyst, thought to be a meningeal cyst on a morphological basis, although CT myelography and MR imaging could not detect such a small cyst, and a pseudomeningeal cyst should also be included in differential diagnosis. Resection of the cyst wall and microscopic inspection of the specimen, although not performed in our patient, would have helped establish the histological diagnosis of meningeal cyst.

Several therapeutic measures are available to treat SIH. Conservative treatment with bed rest and administration of intravenous fluids may be sufficient in mild cases, and corticosteroids may be effective, although the mechanism of action remains unknown. Epidural infusion of autologous blood via an epidural needle under fluoroscopic guidance (epidural blood patch method) is the usual treatment for SIH refractory to conservative treatment and the success rate appears to be high. However, CSF leakage may recur, probably due to inappropriate placement and/or...
insufficient blood clotting that necessitates repeating the procedure.\textsuperscript{13,17} Technical difficulties in correctly positioning the epidural needle around the nerve roots may also cause problems in patients with high cervical spine lesion, in contrast to those with thoracic and lumbar spine lesions, in whom the procedure can be performed safely. The presence of aberrant, engorged epidural veins, which may be inadvertently injured during unguided insertion of an epidural needle, is a potential source of iatrogenic morbidity in patients with high cervical spine lesions.\textsuperscript{1,3,16,20,24} In our patient, the massive CSF leakage demonstrated by CT myelography and radionuclide cisternography prompted us to skip the epidural blood patch method and to surgically repair the leak. The intraoperative CSF escape elicited by the Valsalva maneuver strongly indicated that the leak could not have been repaired by a single epidural injection of autologous blood.

Surgical treatment through a laminectomy and epidural packing of the portion responsible for the leakage with muscle fragments and fibrin glue is more invasive, but is a safer and more reliable method of repairing dural tears, particularly in high cervical spine lesions with massive CSF leakage. Surgical treatment of a patient with a refractory CSF leakage at C2–3 levels achieved immediate, complete resolution of the symptoms.\textsuperscript{11} Although surgical packing is considered the last resort, and is indicated if the epidural blood patch method fails,\textsuperscript{2,13,17} we think that direct surgical packing should be performed in patients with high cervical spine lesions with massive CSF leakage.

References

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