Delayed Gadolinium Enhancement in Epidural Space of the Cervicothoracic Spine in a Patient with Spontaneous Intracranial Hypotension

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Spontaneous intracranial hypotension (SIH) due to a spinal cerebrospinal fluid leak is a rare but increasingly recognized cause of postural headache. Its extravasated epidural fluid collection tends to be non-enhanced or mildly enhanced on enhanced magnetic resonance (MR) imaging. The mild enhancement of the epidural fluid in SIH is usually attributed to fenestrated neovascularization provoked by an inflammatory component, such as blood, of the fluid collection. In this report, we present a case of SIH with a prominent delayed enhancement of the spinal epidural fluid collection on MR imaging. Subsequent vertebral angiography revealed that this delayed enhancement was related to contrast extravasation from a torn anterior meningeal branch of the right vertebral artery. Therefore, we suggest that contrast extravasation from a torn meningeal vessel may be a possible cause of the enhancement in the spinal epidural fluid of SIH. (Chang Gung Med J 2002;25:854-9)

Key words: spontaneous intracranial hypotension, disc herniation, contrast enhancement, magnetic resonance imaging.

CASE REPORT

A 46-year-old woman experienced an abrupt onset of severe occipito-cervical and upper back pain while eating dinner. The pain aggravated while standing and alleviated while lying flat. Tracing her medical history, she had bronchiectasis with regular treatment for 6 years, but vigorous cough with blood-tinged sputum occurred unremittingly.

Physical and neurological examinations were unremarkable. The neck was supple. Cerebrospinal fluid examination revealed an opening pressure of 0 mm H₂O. A little amount of reddish CSF was drained out on coughing. No CSF analysis was done because of inadequate amount of the sample.
Magnetic resonance (MR) imaging of the brain revealed characteristic findings of SIH, including small, bilateral subdural fluid collections, diffuse pachymeningeal enhancement, and prominent dural sinuses (Fig. 1). Spinal MR imaging showed multiple disc herniations, collapsed thecal sac, prominent epidural venous plexus, and diffuse pachymeningeal enhancement. A ventral epidural fluid collection, followed by thecal sac collapse and prominent epidural venous plexus were also observed (Fig. 2).

Fig 1. (A) Post-gadolinium T1-weighted MR image of the brain shows diffuse, intense enhancement at the pachymeningeal space of the brain (arrow) due to intracranial hypotension. (B) Follow-up post-gadolinium T1-weighted MR image of the brain after relief of symptoms shows no abnormal findings on the pachymeningeal enhancement.

Fig 2. (A) Cervical spine MR image immediately after gadolinium administration shows mildly enhanced ventral epidural CSF collection at the cervicothoracic junction (arrows). (B) 10 minutes after the first MR image, on thoracic study there shows a strong contrast enhancement in the cervical and thoracic epidural CSF collection (arrows). The enhanced extradural space is obvious at the dorsal aspect of the thoracic spine and ventral aspect of the cervical spine. This is related to the physiological curvature of the spine. At the cervical spine, the lordotic curve makes it more easily to expand in its ventral aspect; however, at the thoracic spine the kyphotic curve allows an easy expansion on its dorsal aspect. (C) Axial T1-weighted image, (D) T2*-weighted image, and (E) post-gadolinium T1-weighted image show enhancement of the epidural CSF fluid collection (isointense on T1-weighted image and hyperintense on T2-weighted image). The arrows in C and D indicate inward collapsed dura mater.
**Fig 3.** (A) AP view and (B) lateral view of the right vertebral arteriogram. There is contrast medium (arrows) extravasation ventrally from the anterior meningeal branch of the vertebral artery at the C2 level.

**Fig 4.** Axial gradient-echo MR imaging at the C2/3 discal level. (A) On admission, there is a central herniated disc. Part of the posterior annulus and posterior longitudinal ligament complex (arrow), adhering to the ventral dural sac, is retracted backward by the collapsed thecal sac owing to SIIH. Engorged epidural venous plexuses compensating the shrunken thecal sac is noted (arrowheads). (B) 1 month after admission, restoration of the volume of the thecal sac is noted. The complex is no longer pulled backward.
isointense to CSF on all sequences was noted at the cervicothoracic junction. This fluid collection was mildly enhanced on the initial cervical study, but homogeneously and strongly enhanced 10 minutes after administration of contrast material (Fig. 2). To investigate the unusual enhancement, angiography was performed. On common carotid injections, no obvious abnormalities were seen, but on the right vertebral injection there were tiny spots of contrast leaking from the anterior meningeal branch (Fig. 3). Repeated vertebral angiograms obtained 5 days later showed a decrease of the contrast leakage. Therefore, no neurosurgical intervention was planned and the patient was treated with bed rest and analgesics. One week later, patient's symptoms had improved. A follow-up MR imaging showed resolution of the spinal epidural CSF collection and cerebral and spinal pachymeningeal enhancement (Figs. 1 and 4).

DISCUSSION

Postural headache associated with low CSF pressure is a common clinical syndrome following lumbar puncture. In 1938, Schaltenbrand(1) described a spontaneous form of this syndrome and called it "aliquorrhea". Recently, it has been termed as spontaneous intracranial hypotension and thought to be caused by the rupture of an arachnoid membrane in the spine that allows egress of CSF into the subdural and epidural spaces.(2,8) The imaging findings related to SIH reflect the decreased CSF volume throughout the brain and spine. On computed tomography (CT) of the brain, the findings are usually unremarkable. However, on brain MR imaging, diffuse dural enhancement, prominent dural sinuses, enlarged epidural venous plexus, enlarged pituitary glands, subdural effusions or hematomas, and downward displacement of the cerebral structures are revealed.(2,3,6) Most of the spinal findings of SIH have been studies of radionuclide cisternography and CT myelography.(2,3,8) There have been only a few reports mentioning the associated spinal MR findings.(7,9) Among these findings, collapsed thecal sac and epidural fluid collections are important findings. The spinal epidural fluid collection is believed to be caused by CSF leakage and accumulation.(7) After administration of contrast material, the epidural fluid collection is usually not enhanced or only mildly enhanced.(7,9) Rabin et al(7) attributed the mild epidural fluid enhancement to leakage of contrast material into the collection because of fenestrated neovascularization provoked by an inflammatory component, such as blood, of the fluid collection. In the present case, we saw another cause of enhancement - the contrast extravasation from a torn meningeal vessel.

Reviewing the MR imaging of this case, a herniated disc was seen abutting upon the location of the torn meningeal artery (Fig. 4). A scenario for the arterial tear may begin with a typical disc herniation perforating the complex of the posterior annulus and posterior longitudinal ligament. A focal chemical inflammatory reaction leads to adhesions between the complex and the ventral dural membrane. The tiny meningeal vessels within the adhesive dura may be susceptible to rupture after minor external force, such as cough or retraction force of the collapsed thecal sac owing to SIH. If a tiny vascular tear occurs, the extravasated contrast agent may disseminate slowly into the epidural space. Therefore, we can see epidural enhancement on the delayed study.

For a dural leak of SIH, the treatment options are similar to those for post-puncture headache, including bed rest, analgesics, sedatives, oral caffeine, and intravenous hydration.(2,4) For most patients, the symptoms will resolve over a period of weeks to months.(2,4) More aggressive treatments such as epidural blood patches, saline infusions or neurosurgical interventions may be necessary when the headache persists or is incapacitating.(2,6-8) However, in the literature there has been no mention about the treatment of SIH associated with a torn meningeal vessel. Thus, the question of whether the patients need aggressive treatment arises. According to the angiographic follow-up and clinical course of our case, conservative treatment is suggested first because the injured vessel can heal gradually. Invasive intervention may be reserved until the symptoms become worse.

In conclusion, there have been only few reports on SIH that mentioned the associated spinal MR findings as well as the epidural fluid enhancement.(7,9) However, the enhancement of the epidural fluid was usually attributed to fenestrated neovascularization provoked by an inflammatory component, such as blood, of the fluid collection. In our case, we presented another cause for the enhancement - the contrast extravasation from a torn meningeal vessel.
REFERENCES

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一自発性顱內低壓病人之頸胸脊硬膜外腦脊液堆積之
不正常延遲顯影

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本篇報告一自発性顱內低壓併脊硬膜外腦脊液堆積及不正常延遲顯影的病例。隨後的血
管攝影發現，這個不正常的延遲顯影是由於椎動脈的一條小硬膜分枝破裂所致。自発性顱內
低壓併脊硬膜外腦脊液堆積的不正常顯影，雖然很少被報告，大部份是歸因於發炎反應所形
成的成窗血管新生 (fenestrated neovascularization) 所致。本文報告另一種因小的硬膜動脈破裂所
造成不正常顯影的例子。(長庚醫誌 2002;25:854-9)

關鍵字：自発性顱內低壓，椎間盤突出，顯影，磁振造影。