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Case Report

A Rare Case of Idiopathic Spinal Cord Herniation Treated by DuraGen® Collagen Matrix Graft

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We report a rare case of idiopathic spinal cord herniation (ISCH) with a history of cerebrospinal fluid (CSF) leakage. ISCH is a protrusion of the spinal cord through a dural defect. Thin constructive interference in steady-state (CISS) images clearly demonstrated the herniated cord in the present case. The myelopathy worsened and the patient underwent surgery for reduction of herniated spinal cord; the dural defect was filled by placing collagen matrix graft (DuraGen®) between the inner and outer dural layers. The patient's symptoms have improved without relapse for 8 months since surgery. This method may be a good surgical option for cases of spinal cord herniation.

Key words: cerebrospinal fluid leakage, constructive interference in steady state, collagen matrix graft, magnetic resonance image, spinal cord herniation

Spinal cord herniation is a protrusion of the spinal cord through a dural defect into the epidural space. Spinal cord herniations are classified as traumatic, iatrogenic, or idiopathic according to the mechanism of onset. Spinal cord herniation can result in progressive spinal cord dysfunction, for which surgical intervention is often required.

Case Report

Clinical presentation before diagnosis. The patient was a 31-year-old man. Four years before presenting at our institution, he felt a sudden strong shoulder pain, followed by a headache. CT images revealed a chronic subdural hematoma (Fig. 1A). Cerebrospinal fluid (CSF) leakage was suspected at the thoracic vertebral level and was considered the cause of the subdural hematoma. Burr hole surgery for the subdural hematoma and a blood patch for the CSF leakage were performed at the local hospital. However, the headache recurred, and the blood patch procedure was repeated. This yielded temporary relief of headache symptoms. However, two years later, the patient suffered from warm pain disorder of the lower limb extending from the right chest, and pain in the right lower limb without motor deficits or bladder and bowel disturbances. The patient was then referred to our hospital for treatment. According to his neurological symptoms and increased deep tendon reflex of the lower limbs, we suspected myelopathy caused by a thoracic lesion.

MRI image acquisition. MRI imaging was performed with a 3.0-T clinical scanner (MAGNETOM Prisma, Siemens, Erlangen, Germany). A sagittal T2-weighted image revealed ventral deviation of the spinal cord at the Th3/4 level (Fig. 1B). Axial construc-

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tive interference in steady-state (CISS) images were acquired with the following parameters: TR/TE, 5.22/2.20 msec; matrix size, 320×256; FOV, 160 mm; and slice thickness, 0.3 mm. Sagittal-plane CISS images were obtained using multi-planar reformation. On the sagittal CISS images, the spinal cord was found to be deviated and attached to the left side of the ventral dura mater. On axial CISS, part of the spinal cord was shown to be herniated through the internal dura mater (Fig. 1C, D). Thus, we made the diagnosis of a spinal cord herniation.

Surgical findings. Neurosurgeons decided to release the herniated part of the spinal cord and repair the defect in the dura mater. Transcranial motor evoked potentials (MEPs) and somatosensory evoked potentials (SEPs) were recorded to monitor spinal cord dysfunction during the procedure. The surgeons performed left hemilaminectomy of the Th3 and partial hemilaminectomy at the rostral part of the Th4 lamina, so as not to impair the articular capsule. After dural incision, the dentate ligament was cut to explore the ventral side of the spinal canal. The surgeons then observed the herniation of the ventral part of the spinal cord through the dural defect. The defect was 12 mm long in the rostro-caudal direction. First, the surgeons detached the adhesion between the cord and the edge of the dural defect, returning the cord to the spinal canal. Then, the surgeons attempted to patch the inner dural defect with a collagen matrix graft (DuraGen®: artificial dura mater, Integra LifeSciences, Princeton, NJ, US). However, the surgeons gave up on this method due to transient attenuated MEPs. Instead, the dural defect was covered with small pieces of collagen matrix graft without fibrin glue application (Fig. 2).

Clinical course. The pain in the right lower extremity was improved immediately after the operation. The warm pain disorder distal to the Th4 level was relieved on postoperative day (POD) 3. By POD 9,

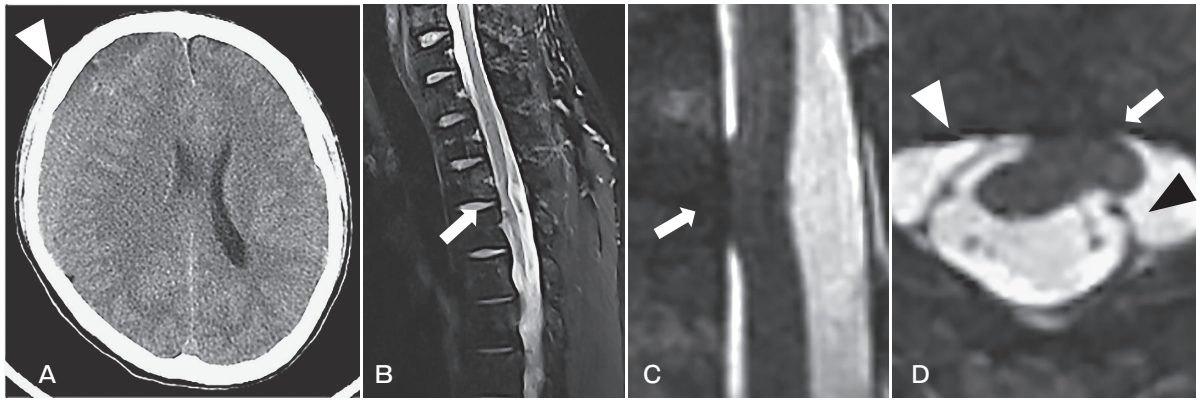

Fig. 1 Preoperative head CT and spinal MR images. **A**, A head CT image showed the right chronic subdural hematoma (arrowhead) with midline shift; **B**, A sagittal T2-weighted image revealed the ventral deviation of the spinal cord at the Th3/4 level (arrow); **C**, on a sagittal CISS image, the spinal cord was located ventrally (arrow) and no CSF was seen ventral to the cord. ; **D**, On an axial CISS image, the spinal cord was herniated (arrow) through the inner layer of the dura mater (black arrowhead) and attached to the outer layer (white arrowhead).

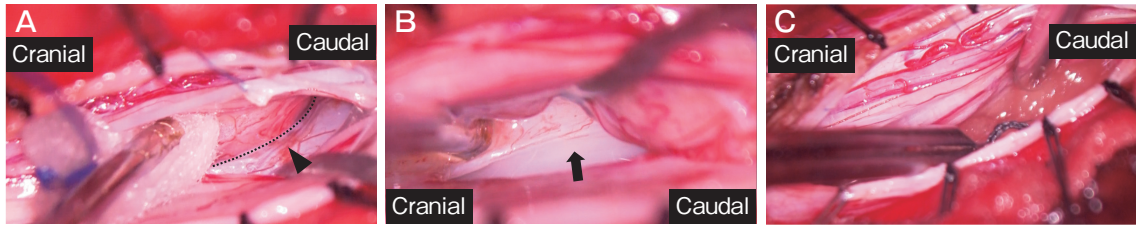

Fig. 2 Intraoperative findings. **A**, The ventral part of the spinal cord was herniated through the dural defect (arrowhead) at the Th3-Th4 level; **B**, View of the dural defect after elevating the herniated spinal cord (arrow). The dural surface could be seen behind the dural defect; **C**, Small pieces of collagen matrix graft were placed through the dural defect to fill the inter-dural space and prevent re-herniation.

abnormal sensation had almost completely resolved. The patient was discharged home on POD13 without neurological deficits, except for a slightly increased deep tendon reflex of the lower limb.

Postoperative MRI. On a postoperative MRI, the ventral deviation of the spinal cord and herniation had disappeared. At that level the spinal cord exhibited a slightly higher signal on axial CISS images (Fig. 3B, C), possibly due to the damage caused by long-term cord herniation. The inserted DuraGen® pieces could be seen as a low-signal band-like structure (Fig. 3C).

Discussion

Clinical presentation and pathogenesis of idiopathic spinal cord herniation (ISCH). First reported by Wortzman in 1974, ISCH is a rare condition in which part of the spinal cord protrudes through a ventral or lateral dural defect [1]. ISCH has been observed to occur in patients from 21 to 78 years of age (mean age, 51 years) and is relatively more common in females than males (1.8:1) [2]. In a meta-analysis by Groen in 2009, Brown-Séquard syndrome was the most common clinical manifestation (66%, 85/129) and paraplegia was the second (30%, 39/129), followed by isolated sensory (3%, 4/129) or motor (1%, 1/129) deficits [2]. ISCH can cause progressive myelopathy. Neurological symptoms are commonly associated with the degree of spinal cord herniation [3].

ISCH has become easier to recognize with the development in MRI technology, including high-resolution MRI methods such as CISS and fast imaging employing steady-state acquisition (FIESTA) [2]. The advantages of CISS are its high signal-to-noise ratio, high contrast-to-noise ratio [4], and intrinsic insensitivity to motion [5]. CISS sequences play an important role in assessing the structures surrounded by CSF. Spinal cord herniation is well visualized by CISS [6].

Various causes have been proposed to explain the occurrence of the dural defect, including minor or unrecognized trauma [1,7,8], congenital meningeal malformations [9,10], pulsation of CSF flow [9,11], and erosion of the dura mater by calcified discs or osteocytes [12,13]. In this case, the patient had no history of trauma, disc herniation, or degeneration of the vertebral body that could have caused the dural injury. Although the pathogenesis remains unclear, some kind of dural defect might be a precondition of ISCH [11]. In the present case, the inner surface of the outer layer was seen intraoperatively through the dural defect, confirming that the dura consisted of at least two layers of membrane. The dura was separated into an inner and an outer layer. In this case, the spinal cord was herniated into the space between the two layers of dura, through the defect of the inner layer. The causes of spinal cord herniation have been discussed in a previous paper. The ventral part of the spinal cord might protrude through a dural defect because of negative pressure in the epidural space in the chest and the CSF pressure gradient during the cardiac cycle [14]. We

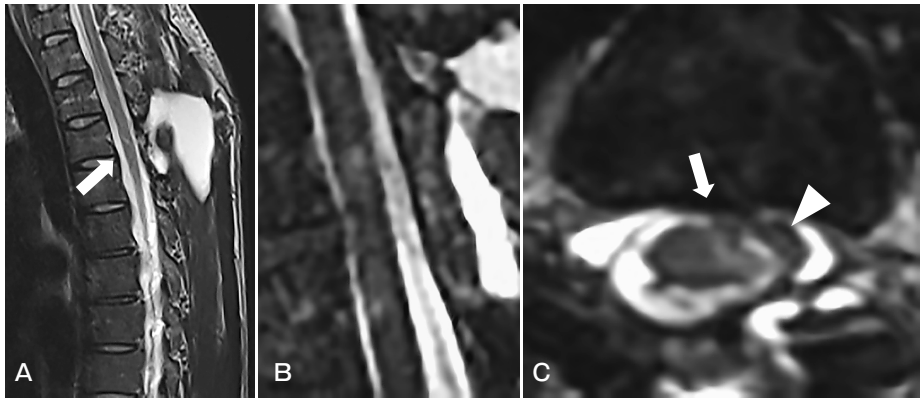

Fig. 3 Postoperative MR images. **A**, A sagittal T2-weighted image revealed the cerebrospinal fluid space ventral to the spinal cord at the Th3/4 level (arrow); **B**, On a sagittal CISS image, the ventral deviation of the spinal cord was shown to be reduced; **C**, An axial CISS image showed the release of the herniated spinal cord. A mildly hyperintense lesion (arrow) could be observed on the left side of the spinal cord. A low-signal band-like structure (arrowhead), which corresponded to the inserted DuraGen® pieces, could be seen on the outside of the inner dura mater.

believe that both layers of the dura are fragile, not just the inner layer. In our case, the defect might have existed in both layers of the dura, allowing for CSF leakage. The amount of CSF leakage might depend on the degree of the defect of the inner and outer layers of the dura mater, as well as the degree of cord herniation. Thus, the symptoms caused by CSF leakage and by myelopathy from cord herniation might change over time, although we could find no reports on this phenomenon in our literature search. In our case, the symptoms of CSF leakage were initially dominant. An earlier MRI showed ventral deviation of the cord, indicating cord herniation at that time. However, by the time of admission to our institute, the herniation volume of the spinal cord might have increased, making the symptoms of spinal cord herniation dominant while the symptoms of CSF leakage had decreased (Fig. 4). It appears that the herniated spinal cord might have reduced the amount of CSF leakage by blocking the dural defect.

Surgical intervention. Conservative treatment may be considered for asymptomatic and mild ISCH. Surgical intervention is the preferred treatment for all advanced cases of spinal cord herniation [15-20]. There have been several reports showing that even longstanding patients may experience considerable neurological improvement after appropriate treatment [2, 21].

Although various procedures have been proposed for the treatment of ISCH, there is currently no standardized procedure in terms of procedural safety and prevention of re-herniation or adhesive arachnoiditis [22]. The two goals of surgery are to release the herniated spinal cord and repair the dural defect. Several methods of repairing dural defects have been described. The two major methods are direct suturing of the dural defect and dural patching [2, 23]. Various materials have been proposed for the patch procedure, including fascia, fat, expanded polytetrafluoroethylene, dural substitute, Teflon, and bovine pericardium [1, 15, 22, 24-26]. In this case, we initially planned to patch the defect with a sheet of collagen matrix graft. However, if this proved not to be possible, we planned to simply fill the space between the inner and outer layers of dura mater with small pieces of collagen matrix graft, which is the procedure we eventually chose. There is no report of ISCH treated by this procedure as far as we can discern.

Enlargement of the hiatus involves less manipulation of the spinal cord, but it may not resolve CSF leakage. In our case, we could not choose this method because of the history of CSF leakage. Direct suturing might be better to treat CSF leakage but involves more compression to the spinal cord during suturing. In a previous report, this treatment led to a poor prognosis [27]. The patches to the hiatus might have led to a good prognosis after surgery, with good control of CSF leakage and less compression to the spinal cord than direct sutures (Fig. 5). However, the patch must be as thin as possible, and in this case, the compression to the spinal cord by the circumferentially placed watery collagen matrix graft sheets might have led to the attenuated MEPs. Furthermore, patches sometimes induce adhesive arachnoiditis over time. Based on our experience with this case, filling the inter-dural space with small pieces of collagen matrix graft appears to be a good surgical option to repair dural defects like the one described here.

Conclusion. ISCH is a rare disease, and specific

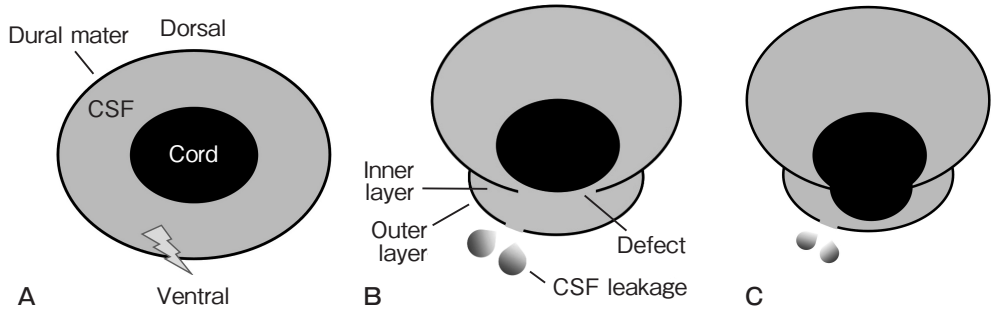

Fig. 4 Schematic drawing of spinal cord herniation pathogenesis in our case. **A**, A defect occured in the dura; **B**, The dura separated into the inner layer and the outer layer. Negative pressure in the thoracic extradural space or pulsatile flow of CSF caused spinal cord herniation. It is believed that the CSF entered the space between the inner and outer layers, and that the CSF then leaked from the defect of the outer layer, resulting in CSF leakage; **C**, Development of spinal cord herniation may have reduced CSF leakage by blocking the dural defect.

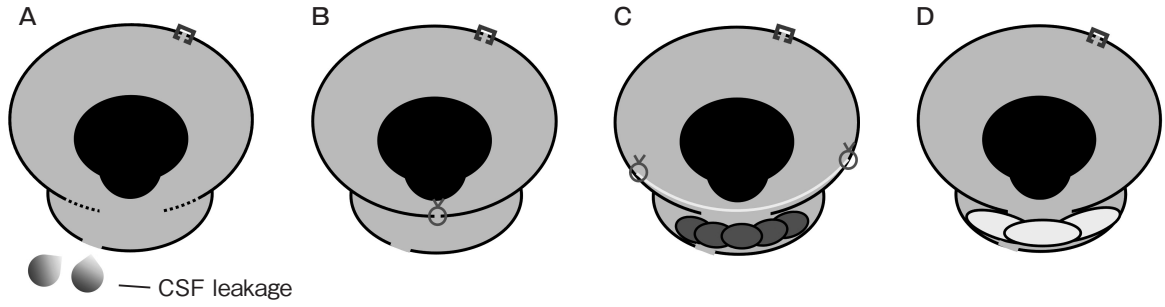

Fig. 5 Surgical strategies for spinal cord herniation. **A**, Widening the dural defect; **B**, Direct suture of the dura; **C**, Ventral dural patch; **D**, Filling the inter-dural space with small pieces of collagen matrix graft.

attention should be paid to cases with CSF leakage or myelopathy. Returning the spinal cord to the spinal canal, then filling the inter-dural space with small pieces of collagen matrix graft may be a good surgical option for repair of spinal cord herniation.

Ethical approval. All procedures performed in this study involving human participants were in accordance with the ethical standards of the institutional and/or national research committee (IRB#1911-023) and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

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