The authors describe two occurrences of idiopathic spinal cord herniation, an entity that has been reported previously in only 11 cases. The patients described in this report presented in midlife with Brown-Séquard syndrome. Computerized tomography myelography and magnetic resonance (MR) imaging showed ventral displacement of the spinal cord with no apparent dorsal mass. Intraspinal cerebrospinal fluid (CSF) flow studies in which phase-contrast pulse sequence cine MR imaging was used displayed a normal pattern dorsal to the spinal cord. Percutaneous intrathecal endoscopic observation did not disclose dorsal intradural cysts, but ventral adhesions were seen between the spinal cord and the dura. Microsurgical intradural exploration revealed ventrolateral herniation of the cord and a ventral root through a dural defect into an epidural arachnoid cyst. The adhesions were released, the cord was repositioned intradurally, and the dural defect was patched. The patients showed gradual improvement postsurgery, as did most of the patients in the previously reported cases. The CSF flow and endoscopic studies were found to be particularly informative, and dural patching with surgical membrane is recommended.

**Key Words** * idiopathic spinal cord herniation * extradural arachnoid cyst * cerebrospinal fluid flow study * endoscopy * Brown-Séquard syndrome

Idiopathic spinal cord herniation is rare. We have report two such cases, in which the patients presented with Brown-Séquard syndrome. We found computerized tomography (CT) myelography, magnetic resonance (MR) imaging of the thoracic spine, and especially cerebrospinal fluid (CSF) flow studies, obtained by means of phase-contrast pulse sequence cine MR imaging and endoscopic inspection to be useful in the preoperative diagnosis. We also review the literature and report the clinical, neuroradiological, endoscopic, and operative findings in idiopathic spinal cord herniation.

**CASE REPORTS**

**Case 1**

**History.** This 45-year-old woman noted decreased temperature sensation in her left foot in June 1992,
which slowly progressed until 1994, when she experienced loss of temperature sensation and noted numbness and a tight feeling below the left L-2 dermatome. In the spring of 1995, she noticed weakness in her right lower extremity and a burning sensation on the left side that extended over the T-10 and L-1 dermatomes. In July 1996 she was admitted to Kobe University Hospital, complaining of a severe burning sensation over the right lateral trunk. She denied any history of trauma or surgery involving the spine.

**Examination.** Neurological examination revealed a right-sided Brown-Séquard syndrome below the level of T-6. Her right lower extremity was moderately weak. The right patellar and Achilles tendon reflexes were slightly hyperactive and a Babinski sign was present on the right side. She complained of numbness, tightness, and loss of temperature sensation below T-6 on the left side, and a burning sensation from the T-6 to L-1 dermatomes. Somatosensory evoked potentials showed a low-amplitude P36 wave on right-sided tibial nerve stimulation. No significant findings were observed on thermography.

**Neuroradiological and Endoscopic Studies.** Preoperative x-ray films of the thoracic spine were unremarkable. The CT myelography and MR studies of the thoracic spine showed ventral displacement of the spinal cord with no apparent dorsal mass (Fig. 1). Because a communicating arachnoid cyst could not be excluded, endoscopic inspection of the thoracic spinal dural sleeve was performed. After percutaneous lumbar puncture, a No. 4 French straight catheter normally used for cerebral angiography was inserted into the spinal subarachnoid space in the same manner as a spinal drainage tube, and then a flexible fiberscope with an outside diameter of 0.5 mm was introduced into the catheter (OES Angioscopy System AF-5; Olympus Optical, Tokyo, Japan). While we monitored the position of the catheter tip with the aid of fluoroscopy, both the catheter and the endoscope were advanced slowly to the T-3 and T-4 levels in the subarachnoid space. The spinal cord was seen to be displaced ventrally, with no dorsal arachnoid cyst. The endoscope was maneuvered ventrally to visualize the spinal cord where it was attached to the ventral dura by arachnoid adhesion. The endoscopically confirmed diagnosis was spinal cord herniation.
Fig. 1. Case 1. Preoperative T2-weighted MR image showing ventral spinal cord displacement with no apparent dorsal mass.

**Operation.** On September 26, 1996, general anesthesia was induced in the patient and a laminoplastic laminotomy was performed from T-3 through T-5. The dura and arachnoid membrane were opened in the midline, and the spinal cord was seen to be rotated to the right and displaced anteriorly. The dentate ligament on the right side was divided and the spinal cord was turned slightly to reveal its point of adhesion to the ventral dura. After turning the spinal cord gently by lifting the dentate ligament, we released the cord from the dura by using blunt and sharp dissection. Herniation of the right anterolateral funiculus of the spinal cord and right T-5 ventral nerve root was reduced. The herniated portion of the spinal cord appeared yellowish and slightly softened. The dural defect was approximately 10 mm in diameter (Fig. 2 upper). Intraoperative oblique-view endoscopy (NU-6530; Machida Endoscope, Tokyo, Japan) revealed that the cyst was an epidural arachnoid cyst rather than a dural diverticulum. A sheet of polytetrafluoroethylene surgical membrane (Gore-Tex, Tokyo, Japan) was positioned over the dural defect and affixed to the dura with two stay sutures to prevent recurrence of herniation (Fig. 2 lower).
Postoperative Course. The patient's postoperative course was uneventful. Within 3 days she noted improvement in the numbness and tight sensation below the left T-6 dermatome. By 20 days postoperatively she had completely recovered from the weakness in her right lower extremity, deep tendon reflexes had returned to normal, and the Babinski sign had disappeared. Temperature sensation below the left T-6 dermatome and burning sensations from T-6 to L-1 dermatomes improved gradually. Somatosensory evoked potentials showed improved amplitude of the P36 wave on stimulation of the right tibial nerve. Postoperative MR imaging of the thoracic spine demonstrated a normal intradural location of the spinal cord (Fig. 3). The patient was discharged 1 month postsurgery.
Case 2

History. In 1990 this 53-year-old man noted decreased temperature sensation in his right foot and mild weakness of his left foot, which slowly progressed until August 1995, when he noticed that his gait was disturbed by progressive left-sided lower-extremity weakness and muscle atrophy. In October 1996 he was admitted to Kobe University Hospital. He had no history of trauma or surgery involving the spine.

Examination. Neurological examination revealed a left-sided Brown-Séquard syndrome below T-6. The patient's left lower extremity was moderately weak. Numbness and a girdlelike sensation were noted below the right T-9 dermatome and loss of temperature sensation was observed below the right T-3 dermatome.

Neuroradiological and Endoscopic Studies. Preoperative CT myelography and MR studies of the thoracic spine demonstrated ventrolateral displacement of the spinal cord at the level of T2-3 with no dorsal intradural cyst (Fig. 4). A CSF flow study disclosed a normal flow pattern dorsal to the spinal cord. On endoscopic examination, the spinal cord was seen to be displaced ventrally. No arachnoid cyst was present dorsal to the spinal cord and the dorsal subarachnoid space was wide at T2-3. The spinal cord was attached to the ventrolateral dura by arachnoid adhesions. The diagnosis was therefore confirmed as spinal cord herniation.
Fig. 4. Case 2. Preoperative T₂-weighted MR image showing ventrolateral displacement of the spinal cord at T2-3 with no dorsal intradural cyst.

**Operation.** On December 9, 1996, after induction of general anesthesia in the patient, a laminoplastic laminotomy was performed from T-1 through T-3. The spinal cord was displaced to the left and ventrolaterally. We lifted the dentate ligament with gentle rotation, and the spinal cord was completely released from the dura. A yellowish tongue-shaped projection from the spinal cord was repositioned intradurally. The dural defect was approximately 12 mm in diameter (Fig. 5). A sheet of polytetrafluoroethylene was placed over the dural defect and affixed to the dura with two-stay sutures.

Fig. 5. Case 2. Intraoperative photograph showing the left anterolateral funiculus of the spinal cord, which was herniated into an extradural arachnoid cyst through a dural defect.
Postoperative Course. The patient’s postoperative course was uneventful. Within 2 days we noted improvement of weakness in his right lower extremity. An MR image of the thoracic spine showed the spinal cord in a normal intradural location (Fig. 6). The patient was discharged 1 month postsurgery.

Fig. 6. Case 2. Postoperative T<sub>2</sub>-weighted MR image of the thoracic spine showing normal intradural location of the spinal cord.

DISCUSSION

Idiopathic spinal cord herniation is an unusual, reversible cause of spinal cord dysfunction that had been reported previously in only 11 cases, which are summarized in Table 1.[1-8] All patients in published reports were adults (age range 36-69 years), and there was no gender preponderance. In all cases, herniation occurred at the thoracic level, either ventrally or ventrolaterally. Twelve of 13 patients presented with a Brown-Séquard syndrome, and in most patients the dural defect occurred in the left ventrolateral dura. With a single exception all patients improved postoperatively, especially in motor function. Borges, et al.,[1] have stated that spinal cord herniation typically presents as a Brown-Séquard syndrome that progresses unless treated, but improves on surgical reduction of the herniated spinal cord, even in patients with longstanding deficits.
The neuroradiological features of these cases are very similar. The CT myelography studies demonstrate no filling defect dorsal to the spinal cord or retention of contrast medium along the ventral aspect of the dural sac. The MR images of the thoracic spine show ventral or ventrolateral displacement of the spinal cord, with no apparent dorsal intradural mass. Intraspinal CSF flow dorsal to the spinal cord was normal according to phase-contrast pulse sequence cine MR imaging, essentially ruling out a dorsal cystic lesion. Spinal cord herniation may coexist with intradural spinal arachnoid cyst, as Isu, et al.,[2] reported in two cases and Oe, et al.,[6] reported in one. Borges, et al.,[1] also reported on a patient with a dorsal intradural arachnoid cyst in whom the preoperative diagnosis, based on imaging studies, was spinal cord herniation. We found CSF flow studies conducted by means of cine MR imaging and endoscopy to be useful in ruling out a cyst.

With respect to the pathophysiological mechanisms of spinal cord herniation, Nakazawa, et al.,[5] and Oe, et al.,[6] have postulated a duplication of the dura mater. Intraoperatively, these authors found an oval defect in the ventral dura and a cavity beyond it. However, histological examination in our second case indicated that the resected dural edge around the defect was true dura mater. Kumar, et al.,[3] have proposed a mechanism of spinal cord herniation based on abnormal positioning of the cord in the dural sleeve and anteroposterior movement of the cord occurring with cardiac and respiratory pulsations or vertebral movement. As adhesions develop between the cord and the edges of a preexisting dural defect, CSF pulsations could propel the cord into the cyst. We postulate that the initial event is herniation of arachnoid membrane through a preexisting congenital dural defect, followed by adherence of the spinal cord to the edge of the dural defect, aided by CSF pulsation and an anterior location of the thoracic spinal cord. The final step could be herniation of the spinal cord caused by negative epidural pressure. After that, continuing mechanical stress would progressively compromise spinal cord function.

The goals of surgery for spinal cord herniation are restoration of the spinal cord to its normal intradural location and prevention of recurrent herniation. We released adhesions between the spinal cord and the dura to return the spinal cord to an intradural position. Although Kumar, et al.,[3] sutured the dural defect in their cases, we believed that we had insufficient space to accomplish this repair safely. Instead we placed a patch of polytetrafluoroethylene between the spinal cord and the dural defect and affixed the patch to the dura with two stay sutures to prevent recurrent herniation. A further advantage of dural closure with a graft patch is that it maintains sufficient subarachnoid space around the spinal cord.
CONCLUSIONS

All neurosurgeons and neurologists managing spinal disorders need to be aware of idiopathic spinal cord herniation, a rare but treatable cause of Brown-Séquard syndrome, that may be more common than is currently recognized. Endoscopic inspection is diagnostically useful and early surgical treatment is recommended to optimize outcome.

References


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