CASE REPORT

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# Spinal cord herniation with characteristic bone change: a case report

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## ABSTRACT

Spinal cord herniation (SCH) is a rare disease characterized by herniation of the thoracic spinal cord through an anterior dural defect, presenting with progressive myelopathy. A case of a 69-year-old woman who presented with Brown-Sequard syndrome and a bone defect, in which an osteophyte created a hemisphere-like cavity with spinal cord herniation, is presented. The strangled spinal cord was released, and the defect was closed microsurgically using an artificial dural patch to prevent re-herniation. Postoperatively, the patient experienced gradual improvement in neurologic function. The SCH mechanism and surgical strategy are discussed.

Key Words: spinal cord herniation, Brown-Sequard syndrome, bone defect, dural patch

## INTRODUCTION

Spinal cord herniation (SCH) is an unusual and possibly under-recognized condition that is being diagnosed more frequently with the widespread use of magnetic resonance imaging (MRI). It refers to herniation of the spinal cord through a ventral dural defect, typically in the mid-thoracic spinal column. The main symptom of this disease is a gradually progressive sensorimotor deficit, such as Brown-Sequard syndrome, and the cause of the symptom is tethering of the thoracic spinal cord, which is herniated into the ventral dural defect. Although more than one hundred cases of SCH have been previously reported since its first report in 1974[1)], and many hypotheses regarding the pathogenesis have been propounded to explain the mechanism of SCH, few reports mentioned a bone defect related to SCH with intraoperative photographs of dural laceration and the bone defect. In this report, a case of SCH with characteristic bone change, which might suggest a mechanism, is presented.

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## CASE REPORT

A 69-year-old woman presented with progressive weakness of the right leg and limping over a period of three years. She reported an episode of falling from a stepladder, but described the trauma as mild and noted that it occurred 10 years ago. She had no history of spinal surgery. Neurological examination revealed spastic monoparesis of the right leg with acceleration of ankle clonus and an exaggerated deep tendon reflex. She could not walk more than 20 meters. She had loss of vibratory and position sense below the T4 level on the right side, combined with contralateral loss of pain and temperature sensation, typical of Brown-Sequard syndrome below the level of T4. Her bowel and bladder functions were normal. MRI showed focal thoracic cord pulling out from the epidural space and tethering ventrally through the dural defect at the edge of the T4 vertebral body level (Figure 1). CT myelography demonstrated an osteophyte that made a hemisphere-like cavity, and the spinal cord was herniated into the cavity. The cerebrospinal fluid (CSF) signal disappeared on the ventral side of the herniated lesion (Figure 2). Laminectomy was performed, along with intradural exploration of the spinal cord. The cord was herniated into the depressed region of the vertebral bone. The herniated part of the spinal cord was released microsurgically and redressed intradurally. The bone defect and laceration of the dura mater were seen, and duplication of the dura mater could not be identified (Figure 3). The laceration length was approximately 1.2 cm longitudinally. Additionally, a dural patch (polytetrafluoroethylene 10 mm×15 mm) was inserted to cover the dural defect, ventral to the spinal cord, to prevent re-herniation. The patch was held in place with stay sutures and 3 stitches each on both sides. A transient reduction in the voltage of motor evoked potentials during surgery was observed while the tethering of the cord was released, but it returned to normal baseline by the end of the operation. The patient's motor weakness improved gradually, and she was able to walk with a cane over 50 meters one week after the operation. Temperature sensation of her left leg returned to normal two weeks after operation. A postoperative MRI showed improvement of the ventral displacement of the spinal cord (Figure 4).

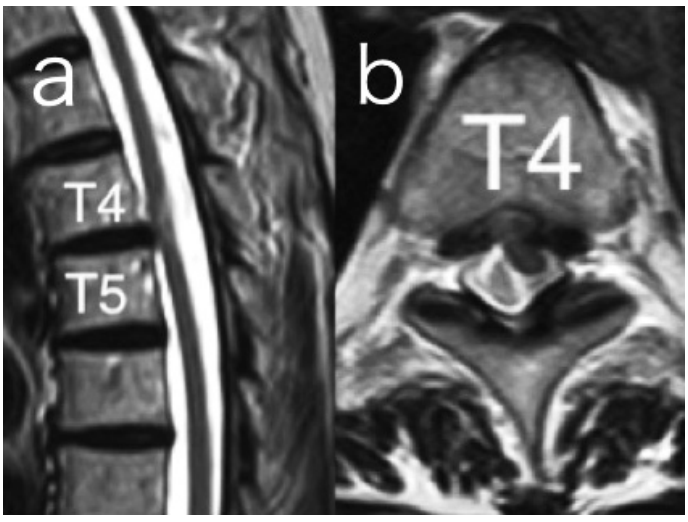


**Fig. 1 (a and b)** Preoperative MR (1.5-T) T2-weighted image
Ventral displacement of the spinal cord at the inferior edge of the T4vertebral body in sagittal section (a), and herniation of the spinal cord into the right anterolateral aspect through the dural defect in axial section (b).

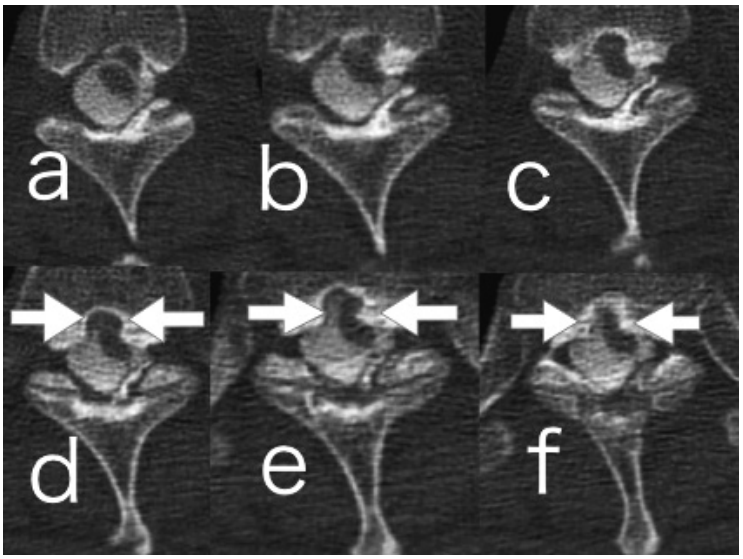


**Fig. 2 (a, b, c, d, e and f)** Preoperative CT myelography (axial section), rostral (a) to caudal (f) view
An osteophyte has formed a hemisphere-like cavity (arrow), and the spinal cord in the right anterolateral side has herniated into the cavity. The CSF signal has disappeared into the ventral side of the cord.

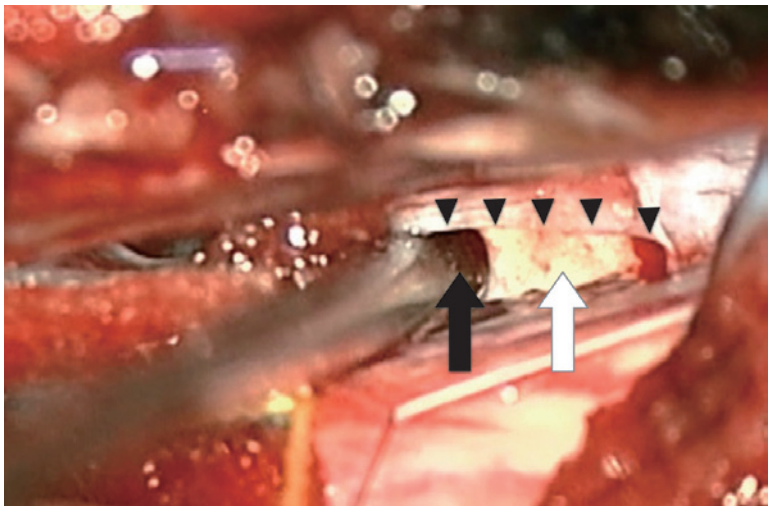

**Fig. 3** Intraoperative photograph with laceration of the dura mater and bone defect
Vertebral bone (white arrow) and bone defect (black arrow) with suction tube, and laceration of the dura mater (arrow heads).

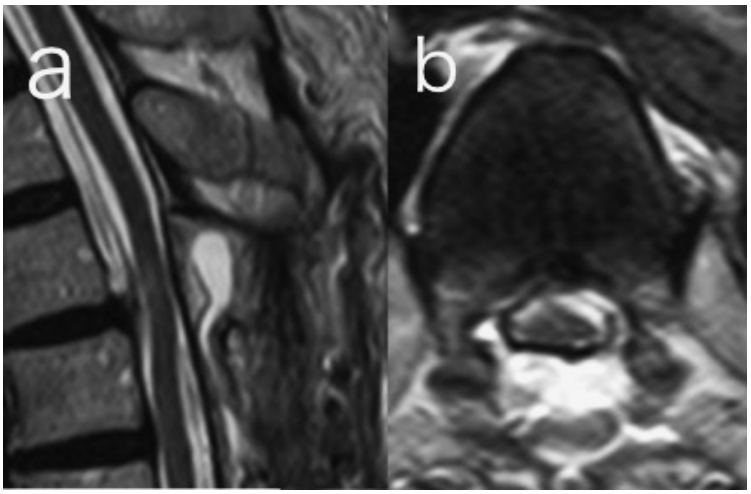


**Fig. 4 (a and b)** Postoperative sagittal (a) and axial (b) T2-weighted MR images showing that the ventral displacement of the spinal cord has improved.

## DISCUSSION

SCH is a rare condition in which the spinal cord herniates through a ventral defect, mostly occurring between T3 and T7 (80%) during middle age (mean age 51 years), and the main clinical presentations are Brown-Sequard syndrome (66%) and paraparesis (30%).[2] Since most cases of SCH have occurred at the thoracic level, an important factor might be the anatomical characteristics of the thoracic spinal cord. Thoracic vertebrae are formed in a kyphotic curve, and the thoracic spinal cord lies more ventrally here than the cervical and lumbar regions because of the characteristic physiological curvature. The cause of SCH remains speculative, but abnormalities of the dura mater seem essential in almost all previously reported cases. A defect or duplication of the ventral or ventrolateral dura mater has been noted. While the spinal cord herniates through the defect, the pathogenesis of the dural defect is unclear. Several theories to explain the occurrence of a ventral thoracic dural defect have been postulated, such as a history of trauma,[3-5] thoracic disc herniation,[4-7] congenital disorder,[1, 8] a duplication of the ventral dura mater,[9-11] arachnoid cyst,[12] or an inflammatory process.[13] Analysis of 70 published cases with SCH revealed that the herniations occurred at the disc level in 67.1% of cases, a herniated nucleus pulposus was present in 30.2%, and osteophytes were present in 29.8%,[14] which might explain why SCH occurs in middle age.

In the present case, the intraoperative findings revealed a 1.2-cm longitudinal laceration of the ventral dura mater. In addition, CT myelography demonstrated the characteristic changes of the vertebral bone including two protuberances that formed a hemisphere-like cavity (subsidence). The cord had herniated through the dura mater directly into the depressed lesion of the vertebral bone. Although it is unclear how a physical force such as repetitive trivial trauma or the disc herniation can induce the dural defect, the characteristic bone changes might have played an important role in the induction of the dural laceration and defect in the present case. The ventral dura mater of the thoracic spine might have been stretched and injured because of unrecognized trauma[3,5] or daily activities involving greater than average flexion and extension of the thoracic spine.[4] The rugged bone changes of the vertebral bone adjacent to the dura mater could be one of the causes of dural laceration.

The spinal cord develops adhesion to the dural orifice and becomes strangled, resulting in compression of the neural elements. The constant pounding of CSF pulsation, cardiac pulsation, respiratory movement, and negative pressure in the extradural space[15] affect the spinal herniation, move the cord further into the dural hole, and the hole is slowly eroded. Neurological deficits may be due to thoracic spinal cord compression and ischemic change, tethering, and distortion. The majority of patients with SCH, especially symptomatic cases, need operation.[2,16] There are 3 main surgical treatment strategies: (1) primary direct sutures to close the dural defect[4,17,18]; (2) use of a dural graft to close the defect[2,16,19-21]; and (3) enlargement of the dural defect.[10,22,15] Groen *et al.* reviewed 129 cases of anterior thoracic spinal cord herniation,[2] and they speculated that spinal cord release and subsequent widening of the dural defect might contribute to favorable results in motor function improvement. In the present case, the bony defect and protuberance of T4 vertebral bone seemed to play an important role not only for dural laceration, but also in lowering the pressure in the thoracic epidural space.

There have been a few previous reports of spinal cord herniation with a bone defect.[23-25] Imagama reported that 5 of 12 SCH patients had a bone defect, and although a bone defect in itself was a poor prognostic factor for postoperative recovery, it was a characteristic imaging feature that predicted severe preoperative symptoms.[25] While the patch method was adopted in the present case, in addition to the use of an artificial dura mater, it may be one of the useful techniques to prevent re-herniation of bone fragments packed into the defect.

Patient consent: The patient has consented to the submission of this case report to the journal.
Conflicts of interest: None.

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