



Idiopathic Spinal Cord Herniation Incarcerated into a Bone Hole: A Case Report and Review of the Literature

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Abstract

Background: Idiopathic spinal cord herniation (ISCH) was first reported in 1974 and is defined as a segment of spinal cord herniated through a ventral defect in the dura. Several reports have suggested the pathogenesis, but it remains unclear. We report a very rare case of ISCH incarcerated in a bone hole and suggest the pathogenesis was related to adhesions that resulted from a pre-existing continuous leakage of cerebrospinal fluid (CSF) created by the bone defect or that the dura was incarcerated in the pre-existing bone defect and adhesion between the dura and vertebra resulted in the secondary dural defect.

A 77-year-old man presented with Brown- Séquardsyndrome below the T6 level. Magnetic resonance imaging (MRI) showed that the thoracic spinal cord was displaced ventrally and partially incarcerated into the vertebral body. There was no septum between the spinal cord and the dura mater.

Intraoperatively, the dorsal dura mater was absent, and herniated spinal cord was identified after durotomy. After releasing the cord herniation, the hiatus and the spinal cord were reduced.

Conclusions: The present case indicates the spinal cord herniation may have (1) been manufactured by incarceration into a bone defect created by the flow of CSF through a pre-existing anterior dural defect, (2) taken place after the incarceration of dura into the pre-existing bone defect, or (3) resulted from adhesion between the dura and vertebra because the dura secondarily could be the underlying pathogenesis of ISCH.

Keywords: Idiopathic spinal cord herniation (ISCH); Lumbar spinal cord stenosis (LCS)

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Introduction

Wortzman et al. [1] first reported a case of idiopathic spinal cord herniation (ISCH) in 1974. In the years since magnetic resonance imaging (MRI) came into widespread use, the concept of ISCH has gradually become more frequently appreciated, and the number of reported cases has increased since 1990. This herniation exclusively occurs in the thoracic spine, typically between T4-T7 causing progressive myelopathy. Diagnosis is based on ventral displacement of the spinal cord observed on MRI and computed tomographic (CT) myelography. Nevertheless, the pathogenesis of ISCH has yet to be established. We report a case of ISCH with incarceration into a vertebral body and suggest two hypotheses.

Case Presentation

History and examination

A 77-year-old man presented with muscle weakness in his lower legs and difficulty walking, which he began to notice at the age of 70. He consulted a physician and underwent lumbar surgery at the age of 72 after being diagnosed with lumbar spinal cord stenosis (LCS). His symptoms did not change. His lower leg muscles became weaker, and walking became increasingly difficult. Neurological examination showed Brown-Séquard syndrome below the T8 level. Dyskinesia, decreased sensation to pressure in the right lower leg and thermal nociception in the left lower leg were seen. Muscle strength was reduced to 2/5 (unable to move joint against gravity) in both lower legs. Deep tendon reflexes were exaggerated bilaterally at the knees and the ankles. His JOA score, excluding the upper limb, was 4/11. Vesicorectal functions were preserved. No abnormalities were seen on blood tests, biochemical tests, or chest X-rays.

Radiological findings

T2-weighted MRI showed that the spinal cord was displaced ventrally at T4/5 (Figure 1). CT



Figure 1: T2-weighted MRI showed that the spinal cord was displaced ventrally at T4/5. CT myelography showed a hollow in the dorsal side of the T5 vertebra, and we suspected the presence of a bone defect and incarceration of the spinal cord into the bone defect.

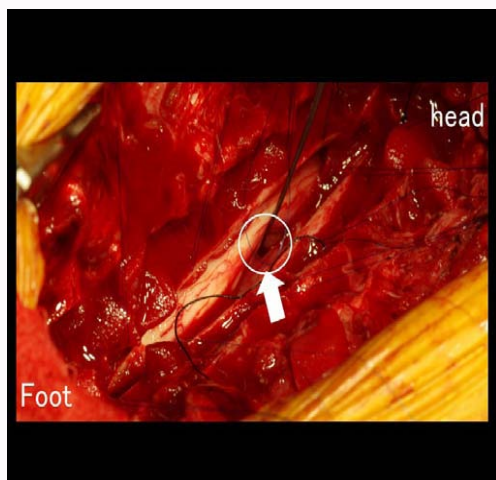


Figure 2: The dorsal dura was opened at the T5 level, and the ventral dural defect and incarceration of the spinal cord into the bone defect at the same level were identified.

myelography showed a hollow in the dorsal side of the T5 vertebra, and we suspected the presence of a bone defect and incarceration of the spinal cord into the bone defect (Figure 1). From the clinical history, physical findings, and images, we diagnosed him with incomplete paresis due to spinal cord herniation and performed surgery because his symptoms were progressive.

Operative findings

We performed laminectomy of T4-6. The dorsal dura was opened at the T5 level, and the ventral dural defect and incarceration of the spinal cord into the bone defect at the same level were identified (Figure 2). No duplicate dura or adhesion between spinal cord and dura was seen. After reduction of the cord herniation, the dural defect was closed using artificial dura (Figure 3).

Postoperative course

The patient had temporary progression of muscle weakness, which forced him to move around with a wheel chair, but the weakness improved gradually. By 5 weeks after surgery, the patient regained the ability to walk with a crutch and was discharged. Postoperative MRI showed the herniation at T4/5 had disappeared, and the spinal cord was reduced to its normal location (Figure 4). His JOA score, excluding the upper limb, improved to 8/11 at 1 year after surgery.

Discussion

Spinal cord herniation is defined as a segment of spinal cord

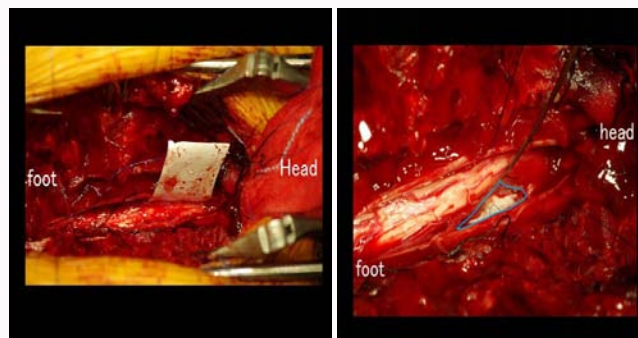


Figure 3: After reduction of the cord herniation, the dural defect was closed using artificial dura.

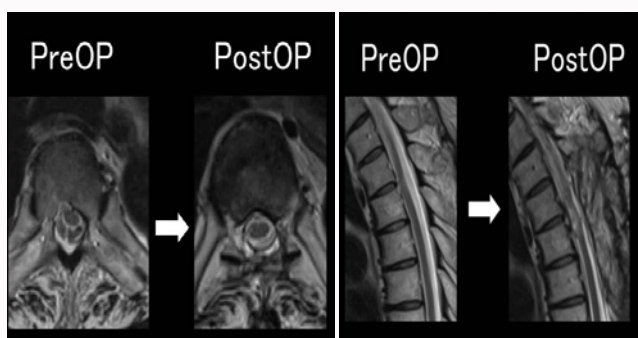


Figure 4: Postoperative MRI showed the herniation at T4/5 had disappeared, and the spinal cord was reduced to its normal location.

herniated through a ventral defect in the dura and often presents with Brown-Séquard syndrome, but patients present with paraplegia rather than hemiplegia. The pathogenesis is still unclear, but Summers et al. [2] classified the herniation under “congenital dural deficiency” “history of trauma” “pressure erosion of dura” and “duplication of ventral dura” [1]. Najjar et al. [3] and Barrenechea et al. [4] reported cases of spinal cord herniation with no duplicated dura, similar to the present case [3-7]. Our patient had no history of trauma or duplicated dura, and we presumed the pathogenesis was related to either a “congenital dural deficiency” or “pressure erosion of the dura” Either way; patients need to undergo surgical intervention because symptoms such as muscle weakness progress gradually.

In approximately 140 articles that we reviewed, seven cases involved incarceration into bone defects, and only reported multiple cases in one review Imagama et al. [5]. In our case, the positions of the dural defect and bone defect were within the same vertebra at the same level, so it is unlikely that the position was just a coincidence. However, even the reviews that reported bone defects did not discuss or propose a pathogenic mechanism, and the etiology remains unknown.

We proposed two hypotheses for the formation of the bone defect. The first hypothesis was that a continuous diapedesis of CSF through the congenital dural defect created the bone defect and forced the spinal cord to incarcerate and adhere. As a result, the spinal cord herniation was formed (Figure 5).

maintain that 60% of thoracic spinal cord herniations are seen at T4-6 Muraoka et al. [6]. Imagama et al. [5] did not mention the vertebral level with the bone defect; it remains unclear if bone defects are likely to be seen in T4-6, but judging from the anatomical

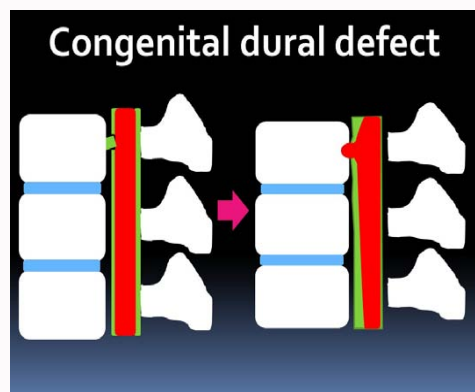


Figure 5: The formed spinal cord herniation.

alignment of the thoracic vertebra (kyphotic), the flow of CSF is slower on the dorsal side, resulting in the storage and turbulence of CSF. These mechanisms are most likely to occur at T4-6, suggesting that formation of a bone defect process may be more likely here.

The second hypothesis is that the dura mater was incarcerated into the pre-existing bone hole, and the adhesion that occurred between the dura and vertebra resulted in the secondary dural defect and spinal cord herniation (Figure 6). Pre-existing bone defects might result from the scalloping of a herniated disc or a hematoma or may occur developmentally during the prenatal period. Barrenechea et al. [4] guessed that arachnoid herniation may occur through an anterior dural defect into the extradural space and CSF would flow freely in and out. In time, as the spinal cord develops adhesions to the edges of the dural defect, CSF flow is impeded. Cardiac pulsations and respiratory movements then push the cord into the defect and cause the herniation [4].

Najjar et al. [3] took into consideration another hypothesis. An inflammatory process involving the spinal cord and/or the meninges is the initial event that leads to ventral adhesion of the cord to the dura. The spinal cord is gradually tethered and later herniated through a dural defect probably caused by the inflammation, adhesion, and pulsatility of the now fixed spinal cord [3]. The theory held by Najjar et al. [3] does not explain the coincidence of the close proximity of the bone defects to the dural defects and does not support the conclusion that the existence of pre-existing bone defects led to the formation of spinal cord herniation. Judging from time series, the first hypothesis advocating dural defects preceding bone defects seemed to affirm Barrenechea et al. [4] but further research is needed. In this case, we suspect the spinal cord herniation took place as a result of the entrapment of the spinal cord by incarceration into the acquired bone hole, which was probably made by the continuous leakage of CSF through the congenital dural defect, and resulted in the myelopathy.

Conclusion

We report a case of spinal cord herniation with slow progression of myelopathy and the underlying cause was determined to be

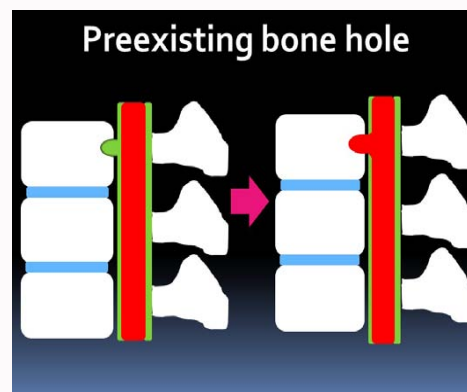


Figure 6: The second hypothesis is that the dura mater was incarcerated into the pre-existing bone hole, and the adhesion that occurred between the dura and vertebra resulted in the secondary dural defect and spinal cord herniation.

incarceration into the bone hole. We assumed that the spinal cord herniation more likely developed as a result of the incarceration into the bone defect made by the flow of CSF through the pre-existing anterior dural defect than for the spinal cord to have herniated after incarceration of the dura into the pre-existing bone defect and that adhesion between the dura and vertebra bereave the dura secondarily can be the pathogenesis of ISCH.

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