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CASE REPORT

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Surgical management of idiopathic thoracal ventral spinal cord herniation: a case report

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Abstract

Introduction Idiopathic spinal cord herniation is a rare entity that is characterized by the displacement of the spinal cord through a defect in the dural layer and presents with symptoms of myelopathy. Surgical management usually results in good outcomes.

Case presentation A 58-year-old female patient presented with weakness of both legs since four months. Her neurological examination showed paraplegia in the lower extremities, and ventral herniation was detected in the T9 vertebral level. The patient underwent surgery. Left T9 hemilaminectomy was followed by the reduction in the herniated spinal cord into its physiological location and the covering the anteriorly located dural defect via circumferentially covering the dural surface of the corresponding vertebral level. Postoperatively, the patient's neurological status improved gradually and radiological scans showed a total reduction in the herniation.

Conclusion Despite the rarity of the pathology, spinal cord herniation should be included in the differential diagnosis of the patients presenting with myelopathy or Brown–Séquard syndrome.

Keywords Spinal cord herniation, Ventral cord herniation, Myelopathy, Dural repair, Case report

Introduction

Idiopathic spinal cord herniation (ISCH) was first described by Wortzman in 1974 after coinciding that the spinal cord was displaced ventrally in a surgical intervention for thoracic disk herniation [21]. It is a condition that presents with signs of myelopathy such as motor or sensory symptoms, urinary or gait disturbances, Brown–Sequard syndrome, and radiculitis due to nerve root entrapment and is rarely encountered in the clinical setting [2, 6]. The improvements in imaging technologies and increased awareness about the disease led to an increase in the diagnosis in the last two decades [19]. Despite the fact that consensus has not been reached for the management of asymptomatic cases, surgery is indicated for symptomatic patients [6, 18]. Surgical treatment results in a good outcome in symptomatic patients.

In this case report, we present a female patient that was diagnosed with idiopathic ventral spinal cord herniation through a dural defect and underwent dural repair with reduction in the herniated spinal cord.

Case presentation

A 58-year-old female patient was admitted with a complaint of insidious weakness in both legs for four months. She neither had a chronic disease nor was using medication. In her past medical history, there was no relevant disease or surgery. She could not walk or stand still during admittance. In her neurological examination, the cranial nerves were intact. She had full muscle strength in both upper extremities. Sensory and deep tendon reflex examination of upper extremities was normal. However, in both lower extremities, the patient's muscle strength was 1/5 in all muscle groups. Patellar and Achilles tendon reflexes were hyperactive bilaterally. Babinski's reflex was

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positive in both extremities. She did not have pain, temperature, or light touch sensation in the lower limbs.

A brain and spinal cord MRI was obtained. In the sagittal T2-weighted sequence, it was observed that the spinal cord had a kink ventrally through the dura mater in the thoracic T9 vertebral level. She did not have any osseous or discal pathology at the corresponding level (Fig. 1a,b). CT myelogram showed that cerebrospinal fluid CSF flow was occluded; however, accumulation of CSF or a cyst was not observed (Fig. 1c).

The patient underwent surgery under neuromonitorization and general anesthesia. Baseline data of somatosensory evoked potentials (SSEP) and motor evoked potentials (MEP) were obtained from all of the extremities. She was positioned prone, and a midline incision was made through T8 and T10 vertebrae. Paraspinal fascia and muscular tissue were dissected on the left side, and left T9 laminectomy was performed. Intraoperative ultrasound was performed to localize the dural defect. A vertical dural incision was made at the dorsal surface of the dura mater. Dissection of the dentate ligaments was followed by the reduction in the herniated cord via a dissector into its normal anatomical position. A bovine pericardium dural graft was inserted into the ventral surface of the spinal column at the level of herniation, and the graft was positioned to cover the whole spinal cord in 360 degrees. The dural incision was sutured, and no CSF leak was observed following positive ventilation. Neuromonitoring parameters showed improvement at the end of the surgery. Immediate postoperative MRI showed that the normal physiological alignment of the spinal cord was achieved and the dura mater was covered circumferentially at the corresponding level (Fig. 2). However, a follow-up MRI scan 2 months later revealed a contusional area in the spinal cord at the segment where the dural graft was inserted and there was myelomalacia extending both cranially and caudally, despite dramatic neurological improvement in the patient. 6th-month follow-up MRI showed remission of the contusion and myelomalacia. The patient's neurological status improved with muscle strength of 3/5 in both lower extremities, and she could stand and walk with a walker device.

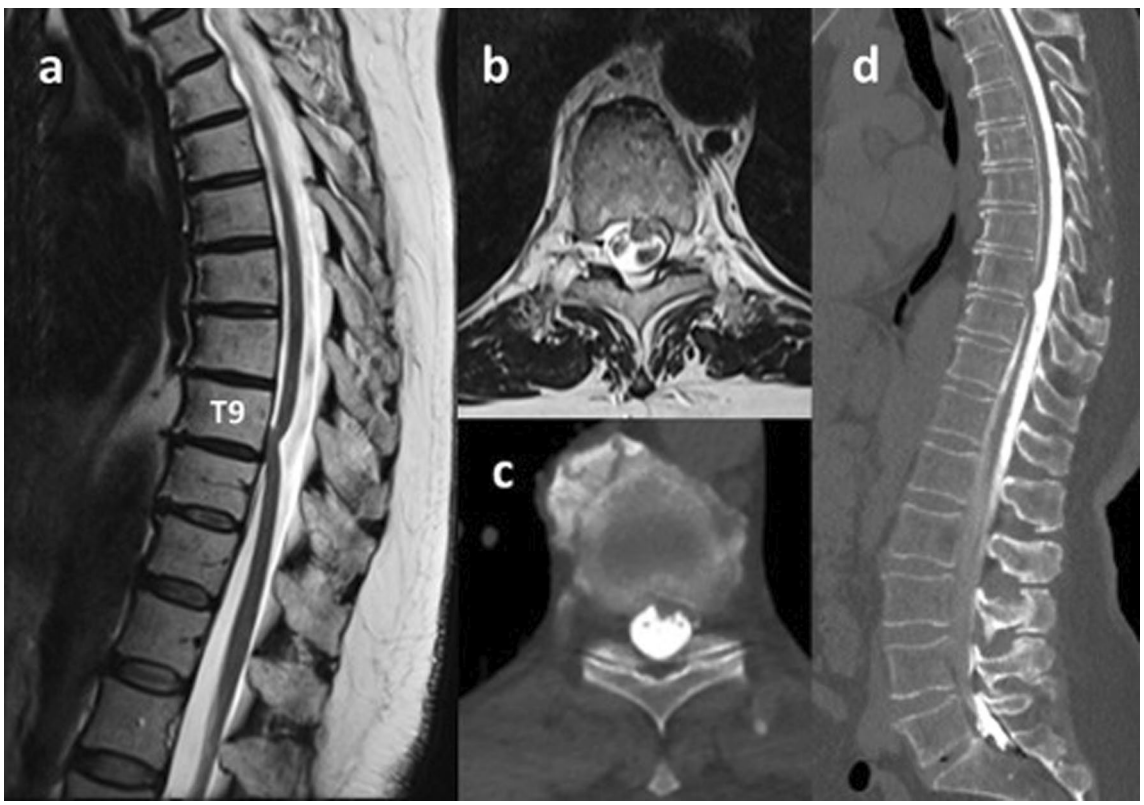

Fig. 1 Preoperative MRI and CT-myelogram. **A** T2-weighted sequence sagittal MRI showing the herniated spinal cord ventrally. **B** T2-weighted sequence axial MRI showing the central herniation of the spinal cord through the ventral wall of the dura mater. **C** CT-myelogram confirms that there is no accompanying arachnoid cyst. **D** Sagittal CT-myelogram imaging of the spinal column

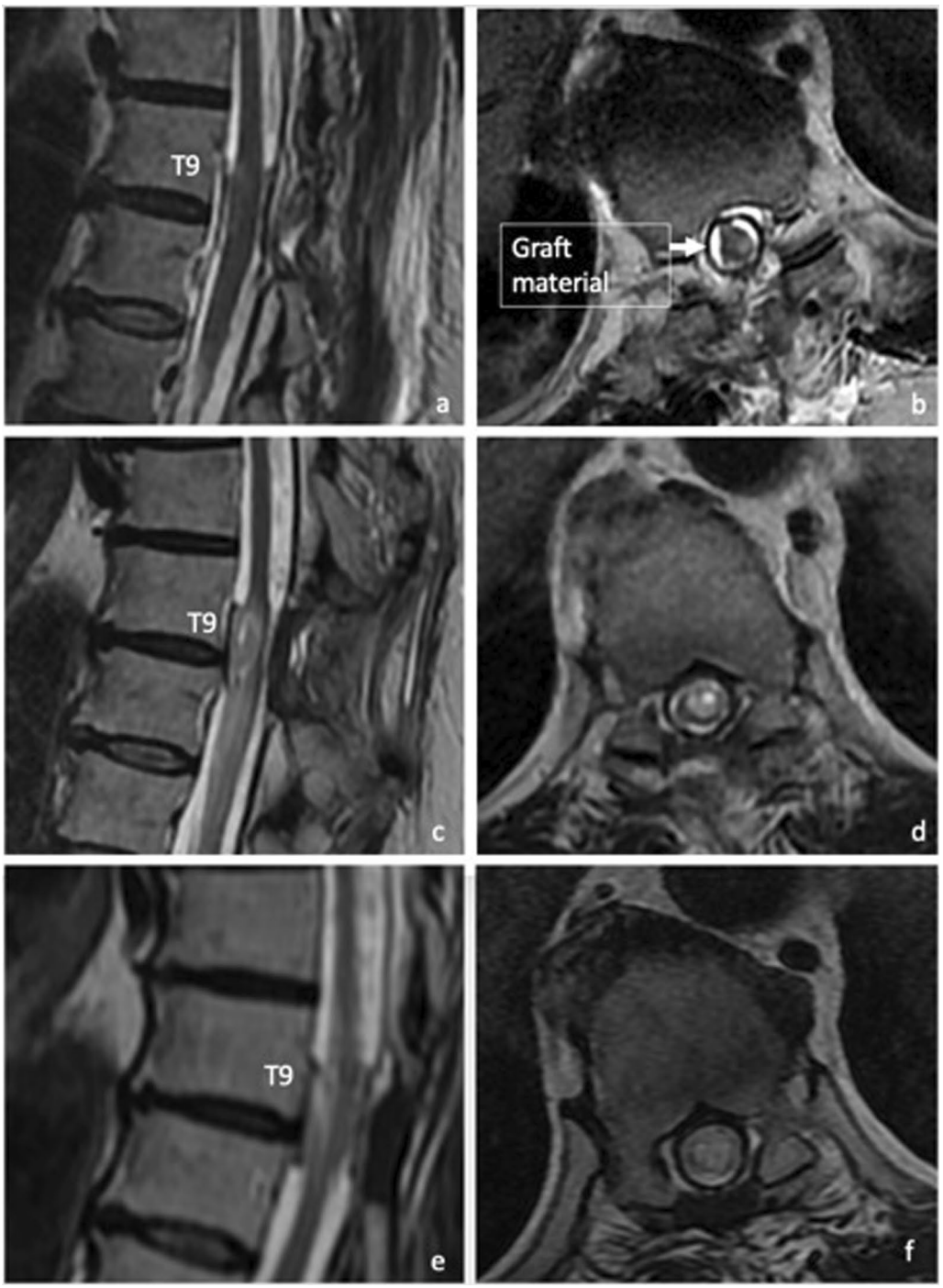

Fig. 2 Postoperative follow-up MRI scans of the thoracic spine. **A**, **B** Axial and sagittal T2-weighted MRI imaging of the surgical site. The spinal cord is reduced to its normal anatomical position, and the bovine pericardium graft was circumferentially covering the dural surface and the ventral dural defect. **C**, **D** Sagittal and axial T2-weighted MRI imaging of the follow-up MRI scan 2 months postoperatively showing contusion and myelomalacia in the spinal cord rostrally and caudally. **E**, **F**: Sagittal and axial T2-weighted MRI imaging of the follow-up MRI scan 8 months postoperatively. Images show remission of the contusion and myelomalacia. The patient's neurologic status improved dramatically

Discussion

Spinal cord herniation is well described in the literature and may be categorized as idiopathic, traumatic, or iatrogenic. Over a hundred cases are described for idiopathic spinal cord herniation through a dural defect, most commonly at the ventral or ventrolateral thoracic

level [4]. Predisposal for the thoracic region is mostly attributed to kyphosis and anterior positioning of the thoracic spine [1]. Iatrogenic herniation mostly occurs after spinal surgery, especially in the presence of pseudomeningocele [4]. Rarity and atypical presentation of the pathology may cause a delay in the diagnosis and treatment of these patients [6, 14].

Etiology and pathophysiological mechanisms

There are different hypotheses on the pathophysiology of the disease. Bartels et al. resected the herniated neural tissue under intraoperative neuromonitorization and showed that there was no altering in either motor or sensory signals; therefore, they suggested that the herniated tissue was lacking functionality [2]. They also revealed that there was no significant myelinization in the tissue after histopathological examination.

A ventral dural defect is the main problem in the evolution of spinal cord herniation. It is unclear whether this defect exists congenitally or is acquired later in life [19]. Pre-existing ventral pseudo-meningocele, a meningeal diverticulum, or an extradural arachnoid cyst supports the existence of the defect congenitally [16]. However, Finneran et al. reported on a case that presented with cervical ventral herniation after corpectomy surgery with neither signs of a dural defect or CSF leak. They suggested that along with the adhesion of the spinal cord and accompanying structures, CSF pulsation and hydrostatic factors aggravate the process [4].

Secondly, duplicated ventral dura with cord herniation between the two layers is suggested to be a possible mechanism for the disease. In a literature review by Derber et al., 18% of 89 cases had duplication of the dura mater. In these cases, the dural defect was in the inner layer of the dura mater and the herniation of the neural tissue extended into the space between the layers [3].

Thirdly, a dorsally located arachnoid cyst may accompany ISCH [3]. It was suggested that the dura mater is eroded due to the pressure conveyed by the arachnoid cyst, therefore predisposing to a dural tear [8]. In these cases, reduction in the herniated spinal cord and repair of the dural defect are mandatory along with excision of the arachnoid cyst to improve the outcome of the surgery [17].

Clinical features

ISCH causes myelopathy with varying levels of severity [19]. Motor symptoms are more prominent than paresthesias with Brown–Sequard syndrome exhibited in two-thirds of the reported patients [15]. Early deterioration of the lateral columns and sparing of the dorsal columns which are manifestations of Brown–Sequard syndrome might be explained by the herniated ventral part of the spinal cord through the dural defect [3]. Bowel and/or bladder symptoms are reported in 10 percent of the patients [15]. Radicular pain in the corresponding dermatome may also be observed [6]. Headaches due to decreased intracranial pressure have also been reported [12].

Radiological features

Until recently CT myelography and standard MRI have been utilized for the diagnosis of ISCH; however, new developments in MRI technology such as 3D fast imaging employing steady-state acquisition (3D-FIESTA) and true fast imaging with steady-state precession (true-FISP) provide optimal contrasting between the CSF and neural tissue [19]. The most feasible gold standard technique currently is standard MRI conjoined with CSF flow study because physiologically CSF flow is directed to the anterior of the cord in the cervical and posterior of the cord in the thoracic region. However, this mechanism may be altered in patients owing ISCH. Moreover, a CT myelogram may determine the CSF cutoff level if there is a block by a dorsal arachnoid cyst [19].

Imagama et al. classified ISCH based on its radiological appearance into 3 groups: type K resembles ventrally kinked spinal cord, type D (discontinuous type) resembles cessation of cord continuity at the herniation site, and type P (protrusion type) resembles the disappearance of subarachnoid space ventrally without any kinking. They also categorized the hiatus in the ventral dura as central or lateral, based on axial images. They suggested that a centrally located hiatus predisposes poor outcomes after the surgery [7]. Our case corresponded to type K with a centrally located dural defect. This might have been the explanation for the severe neurological deterioration of our patient prior to surgery.

Surgical techniques

The main choice of treatment for ISCH is surgical intervention. The surgery aims to detether the spinal cord and implement adjunctive stages to decrease the risk of re-herniation [13]. Excessive retraction of the neural tissue should be avoided whilst reaching the ventral wall of the spinal cord. The ventral wall in the thoracic region has been reached via costotransversectomy and bilateral pediculectomy. Bakhsheshian et al. suggested performing left sided transpedicular approach [1]. The dentate ligament should be sectioned following a laminectomy.

Different techniques for dural repair have been described. The most common technique is to repair the dural defect with a dural graft. Fascia lata, muscular tissue, and artificial dural grafts have been used for this purpose [9, 10, 20]. In a case series with 5 patients, Hawasli et al. described covering the dural defect with a bovine

pericardium placed into the subdural space [6]. In our case, we also utilized a bovine pericardium to cover the spinal cord circumferentially in order to prevent the recurrence of herniation. In this technique, careful dissection of the neural elements should be achieved prior to dural repair in order to prevent postoperative radicular pain [6]. Conversely, widening the dural defect has also been described in order to release the spinal cord. However, in this technique, CSF may accumulate ventral to the spinal cord and may cause clinical symptoms [5]. If there is a dura mater duplication, the dural layer's inner layer should be resected to release the constricted neural tissue [3].

Prognosis and surgical outcomes

ISCH often results in good outcomes following early surgical intervention. In a series of 54 patients, Gaurav et al. reported symptomatic improvement in neurological impairment in 94% of cases [19]. Motor function improvement was observed prominently in patients with Brown–Séquard-like spastic paraparesis [1]. Furthermore, tethering of the cord may trigger ischemia in the ventral spinal cord due to distortion or damage to the blood vessels and this might be the reason for partial recovery in patients that achieved a total reduction in the spinal cord [3].

Factors associated with the poor outcome are a long duration of symptoms and pre-existing spastic paraplegia. Centrally located herniation based on the classification by Imagama is also reported to result in a poor outcome [7]. Additionally, the persistence of the cord extension through the dura mater, which is defined as persistent anterior displacement, may cause the persistence of neurological impairment. Moreover, this displacement may result in the formation of ventral arachnoid adhesions; therefore, new neurological symptoms may arise in 12.6% of the patients [11].

Conclusion

Idiopathic spinal cord herniation is a rare entity that presents with signs of myelopathy and Brown–Séquard symptoms. Early surgical intervention increases the chances of an acceptable outcome. However, the long-term outcome of these patients is still not well documented in the literature.

Abbreviations

ISCH	Idiopathic spinal cord herniation
MRI	Magnetic resonance imaging
CT	Computerized tomography
CSF	Cerebrospinal fluid
SSEP	Somatosensory evoked potentials
MEP	Motor evoked potentials
3D-FIESTA	3D fast imaging employing steady-state acquisition
True-FISP	True fast imaging with steady-state precession

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Author contributions

AO and SD performed the clinical interventions and follow-up of the patient. BS was involved in data interpretation and writing the draft of the manuscript. All authors read and approve the final manuscript.

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Declarations

Informed consent

Written informed consent was obtained from the patient for all the medical interventions that were planned to be performed. Also, written informed consent was obtained for publishing any of the data relevant to the patient's clinical condition.

Competing interests

The authors declare that they have no competing interest.

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