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# Transdural spinal cord herniation: A rare cause of neurologic impairment: Report of two cases and review of the literature

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ABSTRACT

*Introduction:* Herniation of the thoracic spinal cord is a rare pathology related to the genesis of a breach in the anterior dura. Adding to the previously published cases, we report our department's experience with 2 cases of young adults who presented with transdural spinal cord herniation following spinal trauma.

*Presentation of the cases:* We report the cases of 2 men aged 27 and 57, with history of spinal trauma, who presented diverse clinical complaints (motor impairment, genito-sphincteric disorders, etc.). Spinal cord MRI showed an anterior displacement of the spinal cord. Both patients were operated. Peroperative findings and radiological features were concordant with transdural spinal cord herniation.

*Discussion:* Spinal cord herniation is a rare condition which is usually related to a misdiagnosed spinal trauma. Pathogenesis is still being debated. This condition may lead to a myelopathy potentially responsible for severe neurologic impairment, mainly presenting as a Brown-Sequard syndrome. Diagnosis is based on clinical and radiologic features on MRI. Surgical treatment is proposed when neurological signs are found. Long-term clinical and radiological monitoring is necessary for paucisymptomatic, non-operated patients.

*Conclusions:* Based on the described cases, we tried to review the main clinical, radiological and therapeutic features related to this condition.

## 1. Introduction

Spinal cord dysfunction, commonly designated as myelopathy, is mainly related to a spinal cord compression of degenerative, neoplastic or traumatic origin. Spinal cord herniation (SCH) is a rare and sometimes even unknown etiology of myelopathy, described for the first time in 1974 by Wortzman et al. [1]. We conducted a review of the literature, and found that since that date (1974), a total of 172 patients have been reported that presented with this pathology [2–4]. However, some other reports suggest that this condition might occur more frequently than previously assumed [5].

SCH is related to an anterior displacement of the spinal cord through a dural defect. It leads to neurological impairment, secondary to adhesion and vascular compromise [5,6]. Pathogenesis is still under debate, but a traumatic element seems to be involved as in our case.

In this paper, we report our institutional experience of two cases of thoracic spinal cord herniation leading to severe neurologic impairment. A review of the clinical, radiological and therapeutic features related to this condition was processed.

The work has been reported in line with the SCARE 2020 criteria [7].

## 2. Case reports

### 2.1. Case n°1

A 28-year old man, presented due to the progressive onset of pain of the upper thoracic spine for the past 2 years. About 18 months ago, he progressively developed walking disorders. Questioning revealed a history of mild spine trauma, several weeks before the onset of pain. Neurologic examination found spastic paraparesis (muscular testing at 3+ on both lower limbs), without any sensory impairment. The sagittal MRI of the spine showed anterior focal displacement of the spinal cord at the T8 level, producing a "C-shaped" deformation, erasing the anterior subarachnoid spaces and widening the posterior perimedullary spaces. Underneath this herniation could be seen an intramedullary hyperintensity ranging from T8 to T12 vertebral level. There was no evidence of underlying spinal cord injury (Figs. 1 and 2). We decided to operate the patient. Before surgery, the patient was placed in a lateral position.

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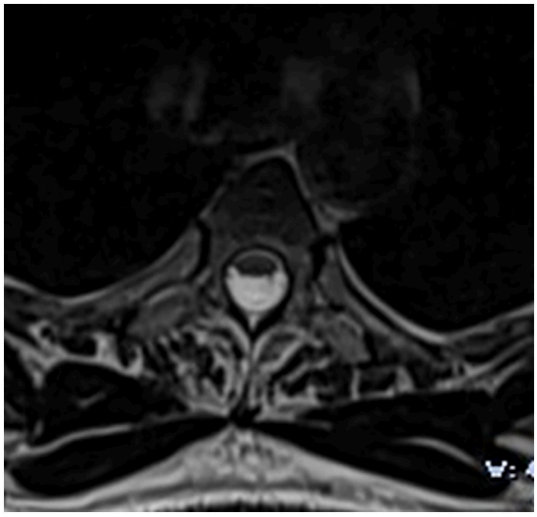

**Fig. 1.** Axial section of a thoracic spinal cord MRI on T2-WI at the level of T8 showing an enlarged posterior subarachnoid space associated to an anteriorly compressed spinal cord.

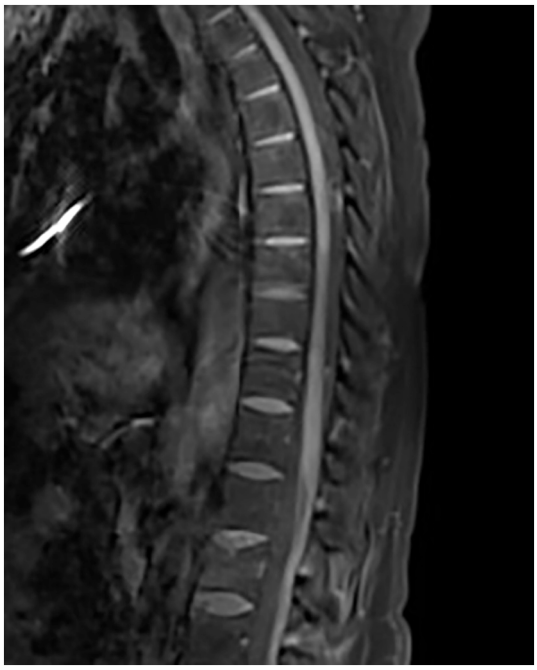

**Fig. 2.** Sagittal section of a thoracic spinal cord MRI showing the anterior displacement of the cord ranging between T8 and T12.

Peroperative, a laminectomy ranging from the T8 to T12 vertebrae was performed, followed by a wide durotomy. We noticed an adherence between the posterior part of the spinal cord and the dura mater due to the presence of fibrosis. The section of the dentate ligament, as well as a smooth dissection of this fibrosis by microscissors allowed a liberation of the spinal cord. A careful assessment of the spinal cord gave the possibility to examine the ventral dura, where the cord was too much adherent to the dura mater with limited possibilities to dissect the medulla from the ventral dura. Thus, an enlargement duraplasty was performed in order to give more room of liberty for the spinal cord, followed by a tight closure of all different plans. Postoperative course was uneventful. A 2-years follow-up showed an improvement of the patient's spasticity (muscular testing at 4- on both lower limbs).

### *2.2. Case n°2*

A 57-year-old man, with a medical history of diabetes and arterial hypertension, had a severe lumbar spine trauma for which he did not present for consultation. One month later, he presented with a progressive thoracic back pain. Six months later he presented with walking disorders, and two months after with sphincter dysfunction. At physical examination, the patient presented with a spastic paraparesis (muscular testing at 3- on both lower limbs), with no obvious sensory impairment. A sagittal MRI of the spine showed a posterior displacement of the spinal cord at the T3 vertebral level. The intramedullary T2-WI hyperintensity visualizes the spinal cord being displaced, pressing against the posterior wall of the vertebral canal. In consequence, the posterior subarachnoid spaces were enlarged, and the anterior subarachnoid spaces decreased in size (Fig. 3). Axial sections at the level of T3 (Fig. 4) revealed dural median and right paramedian dehiscence. In addition, we noticed the collapse of the L2 vertebra. Peroperative, a laminectomy of T2, T3 and T4 was performed. As for the first patient, we noticed an adherence between the posterior part of the spinal cord and the dura mater due to the presence of fibrosis. The section of the dentate ligament, as well as dissection of this fibrosis allowed a liberation of the spinal cord for better mobility. But in contrast to the previous case, after smoothly tracking the spinal cord, a defect within the ventral dura could be noticed, measuring 1 cm width and 3 cm length. This defect was plugged using aponeurosis reinforced by biological glue. Postoperative course was uneventful. A 8-month follow-up showed an improvement of the patient's walking disorders (muscular testing at 4- on both lower limbs).

## 3. Discussion

Transdural spinal cord hernia is a rare cause of progressive myelopathy. However, the incidence rate for this condition has raised

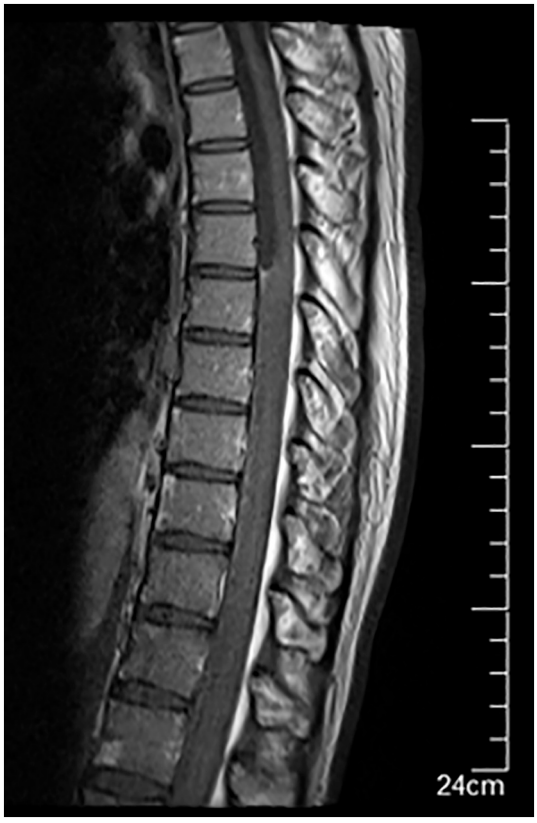


**Fig. 3.** Sagittal section of a thoracic spinal cord MRI on T1-WI showing an atrophy of the spinal cord at the level of T3.

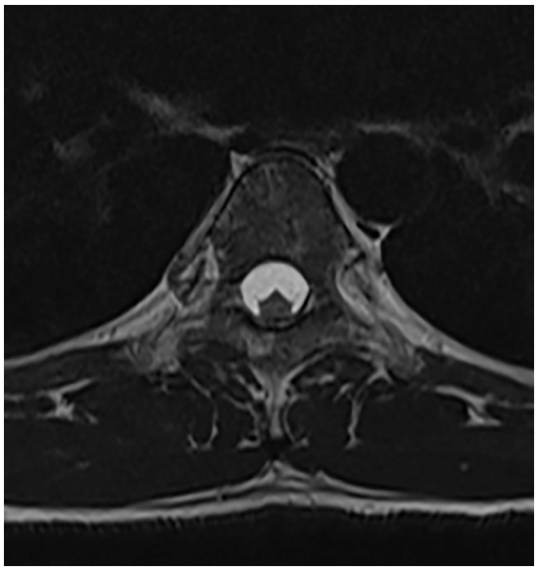

**Fig. 4.** Axial section of a thoracic spinal cord MRI on T2-WI at the level of T3 showing an enlarged anterior subarachnoid space associated to a posteriorly compressed spinal cord.

due to wider access to MRI nowadays. To the best of our knowledge, there were 158 cases that have been reported since Wortzman's case in 1974 [1,2,8].

This pathology is defined by the herniation of the spinal cord at the thoracic vertebral level, mainly ranging from T4 to T7. This herniation is related to an anterior or anterolateral defect in the wall of the dura, which may be congenital or acquired [4,5]. Myelopathy results due to adhesion and vascular disorders of the spinal cord [3].

The pathogenesis of the dural breach remains unknown. Several explanations have been proposed: 1) Congenital: related to a duplication of the dura mater associated to a posterior arachnoid cyst. However, this hypothesis does not explain the onset of the symptomatology most often occuring during the adulthood [9,10]. 2) Another explanation is a traumatic vertebral injury, a disc protrusion or thoracic osteophytosis [4,10,11]. However, less than 10% of the reported cases mentioned any past thoracic spine trauma. 3) A third explanation would be a "syringomyelia-like" theory which states, that the breach is related to pulsed adhesions between the dura mater and the thoracic medulla, thus extending with the respiratory rhythm of the cerebrospinal fluid [2]. In our experience, since both of our patients had a spine trauma previous to the onset of the symptoms, we suggest that this trauma might have caused a dural breach. However, other factors might take part in the pathogenesis. Association between traumatic, inflammatory and haemodynamic features would be involved. Once the dural breach appears, the anatomical disposition of the anteriorly placed thoracic medulla facilitates the adherence between the medulla and the ventral dura. The herniation is then accentuated by the physiological pulsation of the cerebrospinal fluid. Symptoms develop when the herniation evolves to a strangulation [3,12,13].

In the literature, the average duration of a patient having symptoms before being diagnosed is 40 months [3]. In our two cases, the patients presented and were consulted after 24 and 9 months, which is shorter than the average delay. We suppose that this may be related to the anterior spinal trauma that these patients had.

More than half of the patients present with a typical, slowly progressing Brown-Sequard syndrome [14,15]. It begins with motor disorders, followed by sensitive ones. Other signs and symptoms are reported such as sphincter disorders, headaches, thoracic back pain and spastic paraparesis [15].

Magnetic resonance imaging (MRI) is the gold standard for diagnosis. Sagittal sections of T2-weighted imaging (WI) are usually sufficient to suspect the diagnosis, as they show an anterior deviation of the spinal cord, with a C- or S-shaped deformity at the level of the hernia and without interposition of cerebrospinal fluid. An enlargement of the posterior subarachnoid spaces and a diminution of the anterior ones is usually present [16,17]. An intramedullary hyperintensity that indicates spinal cord suffering (myelomalacia) can be used for functional prognosis postoperatively. T1-W MR images are of a lower utility, but they may be able to show an atrophy of the spinal cord [17].

Axial sections complete the study by showing the invagination of the spinal cord into the right anterolateral epidural space [16,17].

Positive diagnosis of thoracic spinal cord herniation is mandatory. A misdiagnosis should be avoided as a surgical extradural approach could cause irreversible damage to the spinal cord [4,13].

The main differential diagnoses may be clinical, with symptomatology suggesting a possible transverse myelitis or an intramedullary tumor. Radiologic features might lead to an incorrect diagnosis of a posterior arachnoid cyst, particularly as they can be seen in association to spinal cord herniation in 20–25% [5,13,17].

The therapeutic approach depends on clinical features. Surgical indication is obvious when the patient has a progressive neurological disorder, mainly motor function. Surgery is able to halt the progression of an evolving symptomatology, sometimes even leading to a marked improvement, in particular if the diagnosis is made early. Nevertheless, complete recovery is rarely obtained. Otherwise, a conservative approach can be adopted for asymptomatic patients using close clinical monitoring [11].

The purpose of surgery is to reintegrate the invaginated spinal cord, and to prevent the recurrence [11,14].

Several techniques for this intervention exist. The technique of choice is based on an anterior transthoracic approach making it possible to repair the anterior defect by a duraplasty [10]. In some cases, when it is impossible to repair the breach in the dura, an enlargement of the orifice is performed, thus preventing further strangulation of the spinal cord [10]. Likewise, a posterior approach is realized in order to achieve an enlargement plasty of the dura mater [14]. In our experience, we chose to perform the latter approach which gave favorable postoperative outcomes.

Long-term follow-up after surgery is recommended in order to detect a possible recurrence.

Prognosis of transdural spinal cord herniation is related to several features. Referring to a meta-analysis of 126 reported cases, Groen et al. [11] demonstrated that Brown-Sequard syndrome and surgical reduction of the spinal cord hernia are factors associated to a favorable postoperative outcome. Summers et al. [6] conducted a review of the literature that included 174 patients. Out of these patients, 159 patients were operated: 74% showed neurological improvement, 18% remained unchanged in their status and 8% presented a worsening after a mean follow-up of 33 months.

Cases of symptoms recurring postoperatively have been reported after follow-ups ranging between 18 months and 10 years, emphasizing the importance of long-term clinical follow-ups and further imagistic investigations [2,11].

## 4. Conclusions

Transdural spinal cord hernia is a rare and still under-recognized condition. It is caused by a hernia of the thoracic spinal cord through a breach of the ventral dura. Furthermore, it represents an etiologies of progressive myelopathy. Brown-Séquard syndrome is a very common clinical presentation. Magnetic resonance imaging is the key element in diagnosing this condition. Surgery is indicated in the case of motor deficits or progressive neurological signs. Long-term clinical and radiological monitoring is necessary for asymptomatic patients or after surgery. Our two rare cases aim to make clinicians and radiologists aware of the existence and the diagnostic difficulties of this condition.

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NA.

## Consent

A written consent has been obtained from the patients.

## Author contribution

- GG wrote the manuscript
- MB made the bibliographic research
- MR and IZ corrected the manuscript

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Not applicable.

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There are no conflicts of interest to declare for this article.

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