Idiopathic Spinal Cord Herniation Associated With a Thoracic Disc Herniation

Case Report, Surgical Video, and Literature Review

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Purpose: The aim of this publication is to present a case of idiopathic spinal cord herniation (ISCH) associated with a transdural disk herniation, demonstrate an operative technique used to treat this condition and provide an updated review the literature.

Background Context: ISCH is an infrequent condition that can cause progressive myelopathy leading to severe neurological dysfunction. This condition is characterized by ventral displacement of the spinal cord across a defect in the dura, either congenital or acquired, resulting in vascular compromise and adhesion that subsequently causes injury to the spinal cord. We present the management of such a patient, in addition to a review of the literature regarding management of ISCH.

Methods: This patient underwent surgery using the dural graft sling technique for repair of the dural defect and restoration of normal spinal cord position within the thecal sac. A review of the literature revealed a total of 171 patients supplemented by our 1 patient, which were then analyzed.

Results: The majority of patients, treated with a variety of surgical techniques, experienced improvements in symptomatology. Our patient experienced significant improvement in symptomatology.

Conclusions: Although ISCH is a rare clinical condition that causes myelopathy, patients managed with surgery generally, though not universally, have a favorable neurological outcome. The associated surgical technique video demonstrates the dural sling technique for the treatment of this rare disorder.

Key Words: idiopathic spinal cord herniation, transdural herniated disk fragment, dural defect, dural graft sling, surgical repair

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Idiopathic spinal cord herniation (ISCH) is an infrequent condition that can cause progressive myelopathy leading to severe neurological dysfunction.1–6 This condition is characterized by ventral displacement of the spinal cord across a defect in the dura, either congenital or acquired, resulting in vascular compromise and adhesion that subsequently results in injury to the spinal cord.5,6 Since its first description in the English literature by Wortzman and colleagues in 1974, ISCH has slowly become a more readily diagnosed entity with the availability of magnetic resonance imaging (MRI) along with increased awareness in the associated signs and symptoms.7–18

Here we describe a case of ISCH, which provides 2 valuable additions to the currently available literature on this disorder. First, this case demonstrates the presence of a transdural herniated disk fragment, a previously hypothesized etiology of the dural defect present with ISCH.5,6 Second, a video demonstrating the dural graft sling technique for repair of the dural defect is presented to assist surgeons not familiar with the surgical management of this rare entity.

CLINICAL CASE AND OPERATIVE TECHNIQUE

Clinical Presentation
A 50-year-old male with a history of multiple lumbar spine surgeries presented with new-onset and progressive myelopathy. MRIs of his spinal axis (Fig. 1A) demonstrated ventral displacement of the spinal cord, in the midthoracic region, in the pattern characteristic of ISCH. A computed tomography (CT) myelogram (Fig. 1B) was also consistent with this diagnosis. Given the neurological dysfunction present, the decision was made to perform a laminectomy and intradural exploration to repair the suspected dural defect.

Operative Technique
Following induction of general anesthesia and prone positioning on a radiolucent Jackson Frame, localizing fluoroscopy was performed to determine the level of interest. A complete thoracic laminectomy centered primarily at T (thoracic) 7 was carried out in the standard fashion, with partial laminectomies also performed at the inferior portion of T6, and superior T8. Ultrasound was used before performing a durotomy to evaluate the spinal cord position.

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This confirmed localization with a ventral displacement of the spinal cord at the center of our dural exposure. After opening and retracting the dura with tacking sutures, microdissection around the spinal cord was performed demonstrating a focal protrusion of the spinal cord through a ventral dural defect that appeared partial thickness in nature. The dentate ligaments were transected bilaterally to allow manipulation and gentle rotation of the spinal cord. Once the spinal cord was mobilized a small desiccated herniated disk fragment was noted at the base of the dural defect, which was resected. A strip of latex surgical glove was cut and passed under the spinal cord. This was used to elevate the cord out of the defect. A small Duragen Plus (Integra LifeSciences Co., Plainsboro, NJ) pledget was placed in the defect after which a thin Gore-tex (WL Gore & Associates Inc., Flagstaff, AZ) pericardial patch was then cut to the appropriate size and positioned under the spinal cord to prevent it from reherniating into the defect. The graft was then sutured to the sides of the thecal sac before closing the dura. This technique is demonstrated in the Supplemental Video (Supplemental Digital Content 1, http://links.lww.com/CLINSPINE/A117). The wound was closed in layers with absorbable sutures.

The patient’s postoperative course was uneventful and he was discharged home on postoperative day 3. His myelopathy improved rapidly over the course of the next several weeks and a postoperative MRI showed the spinal cord positioned centrally within the thecal sac (Fig. 1C). At last follow-up, his neurological symptoms have resolved though he continues to have mild nondisabling back pain.

**DISCUSSION**

ISCH is a rare cause of myelopathy, that occurs secondary to an anterior dural defect which allows the spinal cord to descend into the resulting cavity. The first report of ISCH was by Wortzman and colleagues in 1974. Since that time, the number of published cases have markedly increased, especially with the advent of MRI. In general, ISCH most frequently occurs in the thoracic spine. The unique features of the thoracic spine, which may predispose to this condition, compared with other spinal segments, include the anterior positioning of the thoracic spinal cord, the kyphosis of the thoracic spine, and the anterior physiologic movements of the spinal cord due to cardiac, pulmonary, and flexion and extension movements.

**Etiopathogenesis**

ISCH most commonly presents with pathology at the T4–T5 level, in women (67/33, female to male ratio), during the sixth decade of life (with a range of 22–78 y).

Although the etiology remains debatable, there are 3 types of defects described by Aizawa et al which include: a pseudomeningocele or epidural cyst, a full-thickness dural defect, and a defect in the layer of duplicated ventral dura. Any clinical or historic injury may precipitate a tear in the dura that grows over time. Alternatively, it has also been proposed that a herniated and calcified disk abutting the dura may initiate thinning, erosion, and eventual compromise or rupture of the dura. In this patient, it was noted that the dura appeared, in fact, to be duplicated and a herniated disk fragment was noted within the ventral dural defect perhaps giving credence to this potential mechanism of dural defect formation.

**Clinical Sequelae**

Thoracic myelopathy in a Brown-Séquard syndrome pattern is the most frequently cited presentation if ISCH. Additional manifestations include the full range of neurological signs and symptoms that one might expect from thoracic myelopathy. These signs and symptoms can include gait dysfunction, sphincter, and sexual disturbances, progressive paraparesis, and sensory loss.
| References            | Age (y) | Sex  | Clinical Symptoms | Duration (y) | Spinal Level | Treatment or Repair Procedure | Clinical Outcomes |
|-----------------------|---------|------|-------------------|--------------|--------------|--------------------------------|--------------------|
| Wortzman et al[20]    | 63      | Male | BS                | 1.5          | T7           | Direct suture closure         | W-IM               |
| Masuzawa et al[39]    | 36      | Male | BS                | 1            | T4/T5        | Graft                          | IM, IS             |
| Oe et al[30]          | 61      | Male | BS                | NA           | T4/T5        | Defect widening               | Same               |
| Isu et al[31]         | 43      | Female | BS                | 1            | T5/T6        | Arachnoid cyst resection      | IS                 |
| Tromnier et al[28]    | 45      | Female | BS                | 1.5          | T2/T3        | Arachnoid cyst resection      | IS                 |
| Nakazawa et al[42]    | 43      | Female | BS                | 5            | T2           | Defect widening               | IM+S               |
| White and Firth[29]   | 61      | Female | BS                | 1.5          | T4           | Graft                          | Same               |
| Kumar et al[30]       | 38      | Male | BS                | 3            | T7/T8        | Direct suture closure         | IM+S               |
| Borges et al[6]       | 68      | Female | BS                | 12           | T7           | Direct suture closure         | IM+S               |
| Isu et al[41]         | 43      | Female | BS                | 8            | T2/T3        | Direct suture closure         | IM                 |
| Tronnier et al[28]    | 45      | Female | BS                | 10           | T7           | Direct suture closure         | IM                 |
| Nakazawa et al[42]    | 61      | Male | BS                | 3            | T4/T5        | Defect widening               | IM                 |
| Hausmann and Moseley[44] | 56   | Female | BS                | 8            | T6           | Direct suture closure         | Same               |
| Nakaj et al[49]       | 57      | Male | BS                | 10           | T6/T7        | Hernia not confirmed surgically | Same              |
| Miyake et al[50]      | 49      | Female | SP                | 3            | T4/T5        | Hernia not confirmed surgically | Same              |
| Maita et al[51]       | 45      | Female | BS                | 3            | T3/T4        | Defect widening               | I                  |
| Uchino et al[26]      | 71      | Female | BS                | 2            | T4/T5        | Direct suture closure         | Same               |
| Baur et al[48]        | 66      | Female | BS                | 7            | T10          | Direct suture closure         | IM+S               |
| Chong et al[6]        | 56      | Female | BS                | 4            | T4/T5        | Patch                          | Same               |
| Dix et al[4]          | 44      | Male | BS                | 5            | T6           | Direct closure+biopsy         | W-I                |
| Brugier et al[23]     | 70      | Male | BS                | 0.5          | T5/T6        | Direct suture closure         | IS                 |
| Marshman et al[52]    | 55      | Female | BS—SP             | 6            | T7/T8        | Hernia not confirmed surgically | Same              |
| Abe et al[53]         | 58      | Male | BS                | 4            | T7/T8        | Defect widening               | IM                 |
| Tekkšk et al[54]      | 49      | Female | BS                | 3            | T3/T4        | Patch                          | IM                 |
| Wada et al[4]         | 59      | Male | BS                | 4            | T4/T5        | Defect widening               | IM                 |
| Pereira et al[55]     | 52      | Male | BS                | 6            | T5/T6        | Direct suture closure         | IM                 |
| Miyagushi et al[1]    | 54      | Female | BS                | 2            | T3/T4        | Hernia not confirmed surgically | IM+S              |
| Morkoff et al[57]     | 56      | Female | BS                | NA           | T            | Hernia not confirmed surgically | Same              |
| Egushi et al[58]      | 54      | Female | BS                | 5            | T8/T9        | Hernia not confirmed surgically | Same              |
| Aizawa et al[35]      | 44      | Male | BS                | 5            | T4           | Hernia not confirmed surgically | Same              |
| Watanabe et al[35]    | 43      | Female | BS                | 3            | T3           | Hernia not confirmed surgically | Same              |

(Continued)
TABLE 1. Summary of Reported Cases of Spinal Cord Herniation in the Literature 1974–2017 (continued)

| References      | Age (y) | Sex | Clinical Symptoms | Duration (y) | Spinal Level | Treatment or Repair Procedure | Clinical Outcomes |
|-----------------|---------|-----|-------------------|--------------|--------------|--------------------------------|-------------------|
| Cellerini et al59 | 47      | Male| SP                | 3            | T3           | Defect widening                | I                 |
| Massicotte et al2 | 63      | Male| BS                | 14           | T3/T6        | Observation                     | Same              |
| Massicotte et al2 | 59      | Female| BS                | 6            | T4/T5        | Observation                     | Same              |
| Massicotte et al2 | 33      | Female| BS                | 2            | T7/T8        | Observation                     | Same              |
| Massicotte et al2 | 57      | Female| BS                | 8            | T6           | Patch                           | Same              |
| Massicotte et al2 | 27      | Male | BS                | 1            | T9           | Patch                           | IM                |
| Massicotte et al2 | 46      | Female| BS                | 2            | T4           | Observation                     | Same              |
| Giuseppe et al60 | 28      | Female| BS                | 5            | T6           | Patch                           | W-I               |
| Massicotte et al2 | 64      | Male | SP                | 4            | T8           | Patch                           | W                |
| Najjar et al49   | 50      | Male | Numbness          | 6            | T4           | Observation                     | Same              |
| Massicotte et al2 | 39      | Female| BS                | 2            | T7/T8        | Observation                     | Same              |
| Massicotte et al2 | 50      | Male | BS                | 1            | T9           | Patch                           | IM                |
| Massicotte et al2 | 44      | Female| BS                | 8            | T8/T9        | Patch                           | W-IM              |
| Massicotte et al2 | 32      | Male | SP                | 1            | T7           | Primary closure                 | IM                |
| Massicotte et al2 | 36      | Female| BS                | 1.5          | T2-T3        | Patch                           | IM                |
| Massicotte et al2 | 50      | Female| SP                | 1            | T7-T8        | Patch                           | IM                |
| Massicotte et al2 | 50      | Male | BS                | 1            | T6-T7        |                                 |                  |
| Elleg er et al65 | 59      | Female| BS                | 2.5          | T2           | NS                             | IM                |
| Massicotte et al2 | 59      | Female| BS                | 1.5          | T6           | NS                             | IM                |
| Massicotte et al2 | 52      | Male | BS                | 3            | T4-T5        | Patch                           | Same              |
| Massicotte et al2 | 54      | Female| BS                | 7            | T2-T3        | Patch                           | IM                |
| Massicotte et al2 | 60      | Female| BS                | 2            | T2-T3        | Patch                           | IM                |
| Massicotte et al2 | 59      | Female| BS                | 1            | T5-T6        | Patch                           | IM                |
| Massicotte et al2 | 34      | Male | BS                | 5            | T7-T8        | Patch                           | Same              |
| Massicotte et al2 | 72      | Male | BS                | 5            | T4-T5        | Sling                           | Same              |
| Morley et al58   | 28      | Female| BS                | 2            | T5-T6        | Patch/graft                     | IM                |
| Saito et al69    | 68      | Female| BS                | 32           | T6-T7        | Defect widening                 | IM                |
| Saito et al24    | 57      | Male | BS                | 14           | T2-T3        | Patch                           | IM                |
| Arts et al60     | 58      | Female| SP                | 5            | T7-T8        | Sling                           | IM                |
| Akaza et al71    | 56      | Male | BS                | 5            | T2-T3        | NS                             | IM                |
| Kim et al72      | 38      | Female| BS                | 3            | T4-T5        | Patch                           | IM                |
| Senturk et al73  | 38      | Female| SL                | 0.5          | T4           | No treatment                    | Stable            |
| Ulil et al38     | 50      | Male | BS                | 2            | T2-T3        | Graft/patch                     | IM                |
| Hassler et al75  | 51      | Female| BS                | 2            | T5-T6        | Patch                           | IS                |
| Hassler et al75  | 49      | Female| BS                | 3            | T5-T6        | Patch                           | W                 |
| Hassler et al75  | 46      | Male | SP                | 12           | T2           | Patch                           | Same              |
| Hassler et al75  | 50      | Male | BS                | 4            | T4-T5        | Patch                           | Same              |
| Hassler et al75  | 52      | Female| SP                | 5            | T6-T7        | Patch                           | IS                |
| Hassler et al75  | 37      | Female| BS                | 4            | T4-T5        | Patch                           | Same              |
| Hassler et al75  | 54      | Female| SP                | 6            | T4-T5        | Patch                           | IM                |
| Hassler et al75  | 43      | Female| BS                | 1            | T6-T7        | Patch                           | IM                |
| Hassler et al75  | 54      | Female| BS                | 0.5          | T7-T8        | Patch                           | IM                |
| Hassler et al75  | 41      | Male | SP                | 4            | T3           | Patch                           | IM                |
| Hassler et al75  | 59      | Female| BS                | 5            | T8-T9        | Patch                           | IM                |
| Hassler et al75  | 51      | Male | BS                | 2            | T2-T3        | Patch                           | IM                |
| Hassler et al75  | 47      | Female| BS                | 3            | T6-T7        | Sling                           | IM                |
| Hassler et al75  | 42      | Female| BS                | 5            | T5-T6        | Sling/sleeve                    | IM                |
| Hassler et al75  | 68      | Male | BS                | 5            | T7-T8        | Sling/sleeve                    | IM                |
| Hassler et al75  | 72      | Male | SL                | 2            | T6           | Defect widening                 | W                 |
| Hassler et al75  | 49      | Male | SL                | 1            | T4-T5        | Defect widening                 | Same              |
| Hassler et al75  | 62      | Female| SL                | 10           | T6           | Defect widening                 | IM                |
| Hassler et al75  | 69      | Female| SL                | 1            | T4-T5        | Defect widening                 | Same              |
| Hassler et al75  | 48      | Female| BS                | 4            | T3           | Defect widening                 | IM                |
| Hassler et al75  | 58      | Male | BS                | 12           | T7-T8        | Defect widening                 | IM                |
| Hassler et al75  | 56      | Female| BS                | 2            | T4-T5        | Defect widening                 | IM                |
| Hassler et al75  | 65      | Female| SL                | 7            | T2-T3        | Defect widening                 | IM                |
| Hassler et al75  | 39      | Female| SL                | 15           | T3-T4        | Defect widening                 | IM                |
| Hassler et al75  | 75      | Male | BS                | 5            | T4-T5        | Defect widening                 | IM                |
| Hassler et al75  | 55      | Male | SL                | 2            | T4-T5        | Defect widening                 | IM                |

(Continued)
Imaging Workup

Currently, MRI is the most common imaging modality utilized in making the diagnosis of ISCH. Specifically, one can note on sagittal MRI the ventral angulation of the thoracic spinal cord along with enlargement subarachnoid space behind, giving it a “delta” configuration. One should also be cognizant of posterior compressive arachnoid cysts, which can appear similar to ISCH, and be better defined by phase-contrast MRI which allows for visualization of the dorsal pulsatile cerebrospinal fluid flow. Alternatively a CT myelogram can be performed to support the diagnosis of ISCH, by demonstrating ventral displacement of the spinal cord without a contrast block or defect that could indicate the presence of an arachnoid cyst which does not communicate with the subarachnoid space. A CT myelogram may also be a useful alternative in those with contraindications to MRI.2,23,24

Treatment

Surgical management is recommended in symptomatic patients with ISCH to prevent further neurological deterioration. Three surgical techniques have been described: use of primary sutures to close the dural defect,6,25–27 use of a dural graft sling to repair the defect3,23,28–34 and enlargement of the dural defect.1,22,35,36

| References | Age (y) | Sex | Clinical Symptoms | Duration (y) | Spinal Level | Treatment or Repair Procedure | Clinical Outcomes |
|------------|---------|-----|-------------------|-------------|-------------|-------------------------------|------------------|
| Sasani et al21 | 49 | Female | BS | 8 | T1–T2 | Defect widening | IM |
| Kwong et al80 | 56 | Female | SL | 3 | T3 | No treatment | Stable |
| Zairi et al33 | 41 | Female | M | 2 | T8 | Sling | IM |
| Prada et al32 | 50 | Female | BS, M | 3 | T3–T4 | Patch | IM |
| Kwong et al80 | 37 | Female | BS, M | 3 | T4–T5 | Patch | Same |
| 31 | Male | SL | 5 | T5 | Patch | IM |
| 38 | Female | SL | 2 | T2–T3 | Patch | IM |
| 53 | Female | SL | 3 | T6–T7 | Patch | Same |
| 58 | Male | SL | 2 | T8 | Patch | Same |
| 46 | Male | M | 7 | T8 | Patch | Same |
| 71 | Female | M | 3 | T8 | Patch | Same |
| 26 | Female | M | 2 | T6 | Patch | IM |
| 69 | Male | M | 2 | T8 | Patch | Same |
| 35 | Female | M | 1 | T8 | Patch | IM |
| 51 | Male | BS, M | 1 | T7 | Patch | IM |
| De Souza et al14 | 66 | Female | BS | 7 | T4 | Patch | IM |
| Yamamoto et al83 | 60 | Female | BS | 15 | T5–T6 | Defect widening | IM |
| Berg-Johnsen et al84 | 44 | Female | M | 3 | T4–T5 | Patch | IM |
| 63 | Female | M | 5 | T5–T6 | Patch | Same |
| 75 | Male | BS | 4 | T4–T5 | Patch | IM |
| 58 | Female | M | 4 | T4–T5 | Patch | IM |
| 57 | Female | BS | 6 | T4 | Patch | IM |
| 42 | Female | BS | 2 | T6–T7 | Sling | Same |
| 60 | Female | BS | 10 | T7–T8 | Sling | IM |
| Hawasi et al85 | 32 | Female | BS | 1 | T6–T7 | Sling | IM |
| 44 | Female | BS | 1 | T5–T6 | Sling | IM |
| 58 | Male | BS | 3 | T4–T6 | Sling | IM |
| 36 | Female | BS | 0.25 | T6–T7 | Sling | IM |
| 44 | Female | BS | 0.33 | T1–T2 | Sling | IM |
| 58 | Female | BS | 7 | T5–T6 | Defect widening | IM |
| Carroll et al86 | 58 | Male | BS | 3 | T4–T6 | Sling | IM |
| Ju et al87 | 33 | Female | BS | 6 | T3–T4 | Patch | IM |
| Samuel et al88 | 58 | Male | SL | 1 | T6–T7 | Observation | Resolution |
| Kumar et al89 | 58 | Male | BS | 0.25 | T7–T8 | Dural graft | IM |
| Delgado-López et al90 | 33 | Female | BS | 1.5 | T7–T8 | Titanium microstaples | IM |
| Alkhamesa et al91 | 50 | Female | BS | 3 | T3 | Patch | IM |
| Payer et al92 | 60 | Male | SL | 2 | T5–T6 | Patch | Stable |
| Gkekas et al93 | 55 | Male | BS | 5 | T5–T6 | Defect widening | IM |
| Martinez-del-Campo et al13 | 61 | Male | M | 0.5 | T3–T4 | Dural graft | IM |
| Current study | 50 | Male | M | 1 | T7 | Dural graft sling | IM |

171 patients Male: 61

BS indicates Brown-Séquard syndrome; I, improved; IM, improved motor function; IM+S, improved motor and sensory; IS, improved sensation; M, myelopathy; NA, not available; NS, not specified; SL, sensory loss; SP, spastic paraparesis; W, worse; W-I (M or S), worse than improved (motor or sensory).
Surgical Outcomes

A meta-analysis by Groen et al.\(^37\) looked at surgical results of 121 ISCH patients. They demonstrated that 73% had neurological improvement, 20% being unchanged, and 7% with a neurological decline. A more recent review of the literature by Summers et al.\(^38\) showed that 74% of a patient diagnosed (119/159) with ISCH that underwent surgery demonstrated clinical improvement postoperatively. Overall, 18% showed no clinical changes, and 8% demonstrated worsening postoperative exam findings.\(^38\) Subsequent case reports demonstrate a similar theme of improvement with surgical management. These reports spanning from 1974 to 2015 are summarized in Table 1. Unfortunately, detailed reporting of the techniques employed in individual cases has not uniformly been carried out. Therefore, while all 3 of the described surgical approaches to ISCH appear relatively safe and effective, drawing conclusions regarding the optimal mode of surgical repair is not possible at this time.

CONCLUSIONS

Although ISCH is a rare clinical condition that causes thoracic myelopathy, patients managed with surgery generally, though not universally, have a favorable neurological outcome. The case presented demonstrates the transdural extension of a herniated thoracic disk as a potential cause for dural defect formation. The associated surgical technique video demonstrates the dural sling technique for the treatment of this rare disorder.

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