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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.

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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 of 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.

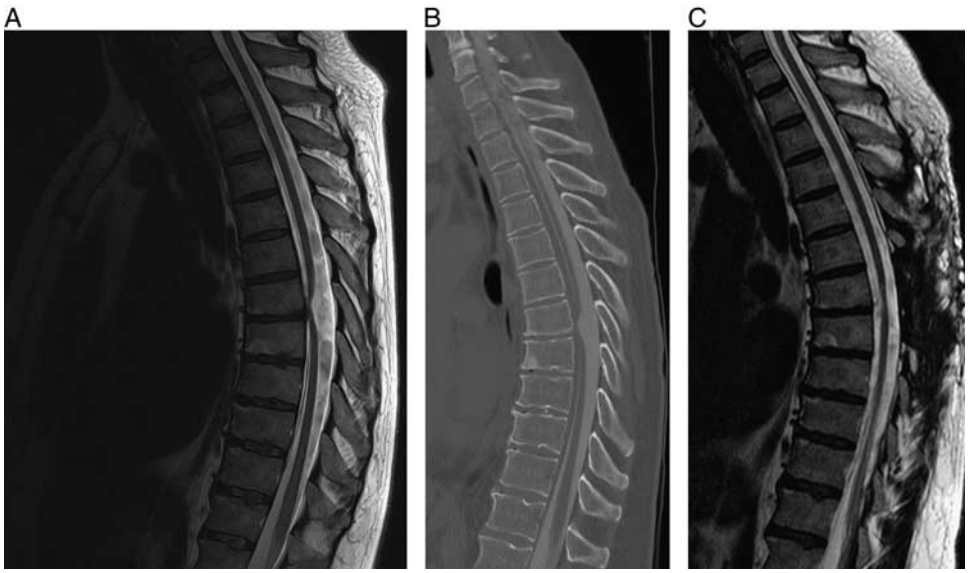

FIGURE 1. A, Preoperative T2 sagittal magnetic resonance imaging demonstrating ventral thoracic spinal cord herniation. B, Preoperative computed tomography sagittal myelogram demonstrating the ventral thoracic spinal cord defect and herniation. C, Postoperative T2 sagittal magnetic resonance imaging demonstrating normal positioning of the thoracic spinal cord.

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 Gor-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 in 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.[19] 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.[20] 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.[19]

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).[21] Although the etiology remains debatable, there are 3 types of defects described by Aizawa et al[22] which include: a pseduomeningocele 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.[22] 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.[21]

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

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[40]	61	Male	BS	NA	T4/T5	Defect widening	Same
Isu et al[41]	43	Female	BS	1	T5/T6	Arachnoid cyst resection	IS
	45	Female	SP	1.5	T2/T3	Arachnoid cyst resection	IS
Tronnier et al[28]	45	Female	BS	3	T3/T4	Patch	W-IS
Nakazawa et al[42]	43	Female	BS	5	T2	Defect widening	IM+S
	39	Female	BS	3	T4/T5	Defect widening	IM
White and Firth[29]	61	Female	BS	1.5	T4	Graft	Same
	39	Male	SP	1.5	T8	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
	69	Male	BS	8	T2/T3	Direct suture closure	IM
	48	Female	BS	10	T7	Direct suture closure	IM
Batzdorf et al[43]	23	Female	BS	2	T6/T8	Patch	IM
Hausmann and Moseley[44]	56	Female	BS	8	T6	Direct suture closure	Same
	36	Male	SP	10	T6/T7	Resection of hernia	W
	51	Female	BS	1	T6/T7	Hernia not confirmed surgically	Same
	49	Male	SP	3	T4/T5	Hernia not confirmed surgically	Same
Matsumara et al[45]	63	Female	SP	NA	T3/T4	Defect widening	I
Miura et al[25]	49	Male	BS	1.25	T5/T6	Defect widening	IM
Urbach et al[46]	44	Male	S	2	T5/T6	Laminectomy	I
Sioutos et al[31]	34	Female	BS	3	T7	Graft	W-IM
Slavotinek et al[47]	22	Female	BS	4	T5	Laminectomy	I
Uchino et al[26]	71	Female	BS	2	T4/T5	Direct suture closure	Same
	61	Female	BS	2	T6	Direct suture closure	NS
Baur et al[48]	66	Female	BS	7	T10	Direct suture closure	IM+S
Najjar et al[49]	57	Male	BS	NA	T2/T3	Graft	NA
	56	Female	SP	NA	T3/T4	Graft	NA
	68	Male	SP	NA	T7/T8	Graft	NA
Miyake et al[50]	45	Female	BS	3	T3/T4	Patch	IM+S
	53	Male	BS	6	T2/T3	Patch	IM
Dix et al[5]	44	Female	BS	NA	T7/T8	Graft	IM
Watters et al[27]	55	Female	BS	10	T3/T4	Direct suture closure	IM
Vallée et al[51]	28	Female	BS	2	T3/T4	Defect widening	IM
	58	Female	BS	6	T4/T5	Patch	IM
	40	Female	BS	2	T5/T6	Patch	Same
	49	Female	BS	4	T4/T5	Patch	Same
Brugières et al[23]	54	Female	BS	5	T6	Direct closure+biopsy	W-I
	70	Male	BS	0.5	T5/T6	Direct suture closure	IS
Marshman et al[52]	55	Female	BS—SP	14	T7/T8	Patch	IM
Abe et al[53]	58	Male	BS	4	T7/T8	Defect widening	IM
	28	Female	SP	1.5	T3/T4	NA	I
	58	Female	BS	NA	T4/T5	NA	I
Tekkök et al[54]	49	Female	BS	3	T3/T4	Patch	IM
	51	Female	BS	2	T6	Patch	IS
Wada et al[4]	59	Male	BS	4	T4/T5	Defect widening	IM
	63	Female	BS	10	T3/T4	Defect widening	IM
	48	Male	BS	2	T5/T6	Defect widening	IM
Pereira et al[55]	55	Male	SP	4	T2/T3	Fill defect with teflon	IM+S
Miyagushi et al[1]	54	Female	BS	2	T3/T4	Graft	I
Berbel et al[56]	56	Male	BS	NA	T	Could not be reduced	Same
Morkoff et al[57]	33	Female	BS	NA	Na	NA	I
Egushi et al[58]	54	Female	SP	NA	T4/T5	Laminectomy	Same
Aizawa et al[22]	44	Male	BS	5	T8/T9	Defect widening	IM
	60	Female	BS	3	T4/T5	Defect widening	IM
	59	Female	BS	20	T4/T5	Defect widening	IM
Watanabe et al[35]	43	Female	BS	5	T4	Defect widening	I
	39	Female	BS	3	T3	Defect widening	I
	54	Female	BS	4	T4	Defect widening	I
	71	Female	SP	5	T4	Defect widening	W
	49	Male	BS	5	T4	Defect widening	I
	47	Female	BS	5	T5	Defect widening	I
	78	Female	SP	4	T4	Defect widening	I
	56	Male	BS	2	T6	Defect widening	I

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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
	47	Male	SP	3	T3	Defect widening	I
Cellerini et al[59]	53	Male	BS	1	T8/T9	Patch	IM
	37	Female	BS	0.5	T4/T5	Patch	IM+S
Massicotte et al[2]	63	Male	BS	14	T5/T6	Observation	Same
	39	Female	BS	NA	T6/T7	Patch	IM+S
	50	Male	Numbness	6	T4	Observation	Same
	44	Female	SP	11	T5/T6	Patch	Same
	33	Female	BS	2	T7/T8	Observation	Same
	57	Female	SP	8	T6	Patch	Same
	27	Male	BS	1	T9	Patch	IM
	46	Female	BS	2	T4	Observation	Same
Giuseppe et al[60]	28	Female	SP	5	T6	Patch	W-I
	64	Male	SP	4	T8	Patch	W
Najjar et al[49]	32	Male	SP	8	T8/T9	Patch	W-IM
Spissu et al[61]	56	Female	BS	1	T7	Primary closure	IM
Sugimoto et al[62]	48	Male	BS	1	T4–T5	Defect widening	IM
Karadeniz-Bilgili et al[63]	36	Female	BS	1.5	T2–T3	Patch	IM
Ammar et al[64]	50	Female	SP	1	T7–T8	Patch	IM
	50	Male			T6–T7		
	51	Female		1.5	T2		
Ellger et al[65]	59	Female	BS	2.5	T2	NS	IM
Francis et al[66]	28	Female	BS	1.5	T6	NS	IM
Barrenechea et al[67]	65	Female	BS	3	T4–T5	Patch	Same
	32	Male	BS	1	T7–T8	Patch	IM
	54	Female	BS	7	T2–T3	Patch	IM
	60	Female	BS	2	T2–T3	Patch	IM
	59	Female	BS	1	T5–T6	Patch	IM
	34	Male	BS	5	T7–T8	Patch	Same
	72	Male	BS	5	T4–T5	Sling	Same
Morley et al[68]	28	Female	BS	2	T5–T6	Patch/graft	IM
Saito et al[69]	68	Female	BS	32	T6–T7	Defect widening	IM
Saito et al[24]	57	Male	BS	14	T2–T3	Patch	IM
Arts et al[70]	58	Female	SP	NA	T7–T8	Sling	IM
	43	Male	BS	1	T4–T5	Sling	IM
Akaza et al[71]	56	Male	BS	5	T2–T3	NS	IM
Kim et al[72]	38	Female	BS	3	T4–T5	Patch	IM
Senturk et al[73]	38	Female	SL	0.5	T4	No treatment	Stable
Uhl et al[74]	50	Male		2	T2–T3	Graft/patch	IM
Hassler et al[75]	51	Female	BS	2	T5–T6	Patch	IS
	49	Female	BS	3	T5–T6	Patch	W
	46	Male	SP	12	T2	Patch	Same
	50	Male	BS	4	T4–T5	Patch	Same
	52	Female	SP	5	T6–T7	Patch	IS
	37	Female	BS	4	T4–T5	Patch	Same
	54	Female	SP	6	T4–T5	Patch	IM
	43	Female	BS	1	T6–T7	Patch	IM
	54	Female	BS	0.5	T7–T8	Patch	IM
	41	Male	SP	4	T3	Patch	IM
Chaichana et al[76]	59	Female	BS	5	T8–T9	Patch	IM
Selviaridis et al[77]	51	Male	BS	2	T2–T3	Patch	IM
Ghostine et al[78]	47	Female	BS	3	T6–T7	Sling	IM
Groen et al[37]	42	Female	BS	5	T5–T6	Sling/sleeve	IM
	68	Male	BS	5	T7–T8	Sling/sleeve	IM
Imagama et al[79]	72	Male	SL	2	T6	Defect widening	W
	49	Male	SL	1	T4–T5	Defect widening	Same
	62	Female	SL	10	T6	Defect widening	IM
	69	Female	SL	1	T4–T5	Defect widening	Same
	48	Female	BS	4	T3	Defect widening	IM
	58	Male	BS	12	T7–T8	Defect widening	IM
	56	Female	BS	2	T4–T5	Defect widening	IM
	65	Female	SL	7	T2–T3	Defect widening	IM
	39	Female	SL	15	T3–T4	Defect widening	IM
	75	Male	BS	5	T4–T5	Defect widening	IM
	55	Male	SL	2	T4–T5	Defect widening	IM

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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
	49	Female	BS	8	T1–T2	Defect widening	IM
Sasani et al[21]	45	Female	BS	NA	T8	Primary closure	IM
Kwong et al[80]	56	Female	SL	3	T3	No treatment	Stable
Zairi et al[81]	41	Female	M	2	T8	Sling	IM
	48	Female	BS	4	T4–T5	Sling	IM
	56	Female	BS	2	T8	Sling	IM
Prada et al[82]	50	Female	BS, M	3	T3–T4	Patch	IM
	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 al[14]	66	Female	BS	7	T4	Patch	IM
Yamamoto et al[83]	60	Female	BS	15	T5–T6	Defect widening	IM
Berg-Johnsen et al[84]	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
Hawasli et al[85]	32	Female	BS	1	T6–T7	Sling	IM
	44	Female	BS	1	T5–T6	Sling	IM
	58	Male	BS	3	T4–T6	Sling	IM
	44	Female	BS	0.25	T6–T7	Sling	IM
	36	Female	BS	0.33	T1–T2	Sling	IM
Carroll et al[86]	58	Female	BS	7	T5–T6	Defect widening	IM
Ju et al[87]	33	Female	BS	6	T3–T4	Patch	IM
Samuel et al[88]	58	Male	SL	1	T6–T7	Observation	Resolution
Kumar et al[89]	58	Male	BS	0.25	T7–T8	Dural graft	IM
Delgado-López et al[90]	33	Female	BS	1.5	T7–T8	Titanium microstaples	IM
Alkhamees et al[91]	50	Female	BS	3	T3	Patch	IM
Payer et al[92]	60	Male	SL	2	T5–T6	Patch	Stable
Gkekas et al[93]	55	Male	BS	5	T5–T6	Defect widening	IM
Martinez-del-Campo et al[13]	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 Female: 110					

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).

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 defect[3,23,28–34] and enlargement of the dural defect.[1,22,35,36]

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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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