Introduction

Idiopathic spinal cord herniation (ISCH) is a rare spinal pathology that often presents slowly progressive Brown Séquard syndrome or spastic paraparesis over years (1-5). It is most commonly observed in the thoracic spine and is characterized by ventral herniation of the spinal cord into a pseudomeningocele, ruling out the possibility of iatrogenic or traumatic reason (6-8). More than 200 cases have been reported since the first case was reported in 1970s (9-12). Despite advances in neuroimaging and graft materials, the natural history and optimal management strategy of ISCH is still under debate (13-15).

Surgical reduction is usually recommended for symptomatic patients with aggravation or signs of myelopathy. The goal of the surgical procedure lies is to...
detether the spinal cord, with measures taken to prevent reherniation (16). Laminectomy is a commonly used technique, and partial facetectomy can also be performed to increase the lateral exposure to gain access to the ventral dural surface without extensive spinal cord retraction (17-19). Adequate exposure is essential for the manipulation of herniated spinal cord detethering and dural defect repair, reducing undesired neurological deficits (20). The technique for dural defect management is still debatable, including enlarging or repairing by direct suture or graft materials, such as fascial patches, a muscle or fat graft, bovine pericardium, or artificial dural substitutes (19,21-24). Due to limited cases of this disease and on the variety of closure techniques, the optimal surgical strategy is still unclear.

Here, we present a case series of ISCH using the posterior midline approach with laminectomy. Fat graft with or without artificial dural patch fixed with stitches was used between the reduced spinal cord and the ventral dural defect. Notably, fat patches were used alone without artificial dural substitutes in most cases, which was unique from previous studies. Clinical and radiographic follow up was done. To the best of our knowledge, this is the largest case series using this technique to date. We present the following article in accordance with the STROBE and AME Case Series reporting checklists (available at https://atm.amegroups.com/article/view/10.21037/atm-22-3343/rc).

**Methods**

The present study was conducted in accordance with the Declaration of Helsinki (as revised in 2013). The study was approved by the ethics committee of Xuanwu Hospital, Capital Medical University [No. (2022)020]. Written informed consent for participation was not required for this retrospective study, in accordance with national legislation and institutional requirements.

A retrospective review of all cases of ISCH at our institution between January 2017 and June 2021 was performed. Patient demographic information, symptoms, imaging, operative details, perioperative and postoperative courses, clinical outcomes, and complications were reviewed. Japanese Orthopedic Association (JOA) score was calculated preoperatively and postoperatively for the comparison of outcomes. The preoperative score was calculated as follows: $RR = \frac{(postoperative \ JOA \ score - preoperative \ JOA \ score)}{(11 - preoperative \ JOA \ score)} \times 100\%$.

**Statistical analysis**

All data were descriptive statistics with mean ± standard deviation, calculated by SPSS version 22.0.

**Results**

**Patient demographics and radiographic presentation**

Seven patients (3 men and 4 women) aged 22–65 years, were included in the present study (Table 1). Patients presented with either Brown Séquard syndrome or spastic paraparesis that progressed over time prior to surgery. Six patients presented with weakness in the lower extremities. Six patients presented with temperature loss in the lower extremities. Two patients suffered from radicular pain. Urinary incontinence symptoms were not present in these patients. Three patients presented with typical Brown Séquard symptoms. The mean preoperative JOA score was 8.2 (range, 7–10). Dural defects were all located at the thoracic level, from T2 to T7 (Table 2).

Magnetic resonance imaging (MRI) was used for each patient and showed typical demonstration of widened posterior subarachnoid space and C-shaped ventral deformation of the spinal cord at the affected level (Figure 1A,1B). Computed tomography myelography was used for further differentiation if necessary to reveal dural defects (Figure 1C).

**Surgical technique**

With the patient in the prone position and under general anesthesia, baseline neurophysiological level on somatosensory-evoked potentials (SSEPs) and motor-evoked potentials (MEPs) were monitored for extremities and perianal regions. Through a midline incision, patients underwent 2–3 levels of laminectomy at appropriate spinal levels (Figure 2). Using an operative microscope, the dura was opened in the midline and secured with sutures. After the arachnoid and ipsilateral dentate ligaments were sharply opened throughout the field, the spinal cord was mobilized with a micro-dissector, so that the dural defect with spinal cord herniation was visible (Figure 3A). The limit of spinal cord retraction was dependent on SSEPs and MEPs. The procedure was paused when there was a latency increase.
of more than 10% or a reduction of amplitude >50% compared with baseline SSEP or MEP signals. After the careful dissection of surrounding adhesions, the edges of the dural defect were defined, and the herniated portion of the spinal cord was repositioned gently (Figure 3B). The defect could be enlarged slightly to avoid manipulating the herniation. A subcutaneous fat graft was placed through the dural defect, and the size of the graft was large enough to fill the cavity without protruding too much into the subdural space causing spinal cord compression. The graft was fixed to the edge of the defect by interrupted stitches (Figure 3C). In cases 1 and 2, the size of the dural defect was over 1.5 cm after the fat graft was positioned. We deployed an artificial dural patch intradurally overlapping the dural defect. The artificial dural patch covered the total dural defect and adhered circumferentially around the inner surface of the dura. No extra pressure on the spinal cord at that level was confirmed, and the dural patch was trimmed to fit the edge of the dural incision (Figure 3D). After thorough intradural rinsing, the dural incision was closed tightly together with an artificial dural patch by a running suture, so that the dural patch would be immobilized. Laminoplasty was then performed, and the wound was closed. Average operating time and estimated blood loss were 3 hours and 24 minutes and 88.6 mL, respectively (Table 2).

**Clinical outcomes**

Postoperative hospital stay length was 8.7 days (Table 3). One patient had a prolonged hospital stay due to COVID-19. One patient experienced CSF leakage, and a lumbar drain was placed for 1 week. One patient suffered

### Table 1 Patient demographic characteristics and presentation

<table>
<thead>
<tr>
<th>Case No.</th>
<th>Age (years)</th>
<th>Sex</th>
<th>Symptoms and duration (years)</th>
<th>JOA</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>61</td>
<td>Female</td>
<td>Right leg (6 years), left leg (1.5 years)</td>
<td>7</td>
</tr>
<tr>
<td>2</td>
<td>46</td>
<td>Male</td>
<td>Left thoracic radicular</td>
<td>9</td>
</tr>
<tr>
<td>3</td>
<td>50</td>
<td>Female</td>
<td>None</td>
<td>8.5</td>
</tr>
<tr>
<td>4</td>
<td>22</td>
<td>Female</td>
<td>Left leg (5 years)</td>
<td>None</td>
</tr>
<tr>
<td>5</td>
<td>63</td>
<td>Male</td>
<td>None</td>
<td>8.5</td>
</tr>
<tr>
<td>6</td>
<td>65</td>
<td>Female</td>
<td>None</td>
<td>7.5</td>
</tr>
<tr>
<td>7</td>
<td>41</td>
<td>Female</td>
<td>None</td>
<td>10</td>
</tr>
</tbody>
</table>

JOA, Japanese Orthopedic Association.

<table>
<thead>
<tr>
<th>Case No.</th>
<th>Level</th>
<th>Side of dural defect</th>
<th>Dural access</th>
<th>Graft</th>
<th>Duration (minutes)</th>
<th>EBL (mL)</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>T4–6</td>
<td>Ventral</td>
<td>3-level laminoplasty</td>
<td>Fat patch + artificial dural patch</td>
<td>228</td>
<td>50</td>
</tr>
<tr>
<td>2</td>
<td>T2–3</td>
<td>Ventrolateral left</td>
<td>2-level laminoplasty</td>
<td>Fat patch + artificial dural patch</td>
<td>180</td>
<td>100</td>
</tr>
<tr>
<td>3</td>
<td>T2–3</td>
<td>Ventrolateral right</td>
<td>2-level laminoplasty</td>
<td>Fat patch</td>
<td>135</td>
<td>100</td>
</tr>
<tr>
<td>4</td>
<td>T4–5</td>
<td>Ventrolateral left</td>
<td>2-level laminoplasty</td>
<td>Fat patch</td>
<td>240</td>
<td>100</td>
</tr>
<tr>
<td>5</td>
<td>T3–4</td>
<td>Ventral</td>
<td>2-level laminoplasty</td>
<td>Fat patch</td>
<td>150</td>
<td>20</td>
</tr>
<tr>
<td>6</td>
<td>T6–7</td>
<td>Ventrolateral right</td>
<td>2-level laminoplasty</td>
<td>Fat patch</td>
<td>220</td>
<td>200</td>
</tr>
<tr>
<td>7</td>
<td>T2–3</td>
<td>Ventral</td>
<td>2-level laminoplasty</td>
<td>Fat patch</td>
<td>180</td>
<td>50</td>
</tr>
</tbody>
</table>

EBL, estimated blood loss.

### Table 2 Operative details

Postoperative hospital stay length was 8.7 days (Table 3). One patient had a prolonged hospital stay due to COVID-19. One patient experienced CSF leakage, and a lumbar drain was placed for 1 week. One patient suffered...
from spinal canal stenosis on the surgical site 4 months later. After she underwent immediate decompression surgery, symptoms were relieved without sequelae.

The mean follow-up time was 23.4 months (range, 9–42 months). Clinical improvements were observed in 5 patients. Two patients remained unchanged. None of the patients achieved complete resolution of all symptoms, even after a long period of rehabilitation. Routine MRI was performed postoperatively, and all 7 patients demonstrated radiographic resolution of ISCH (Figure 4). The mean JOA score at the final follow up was 8.7 (range, 7–10). The mean RR of the JOA score was 17.9%.

Discussion

ISCH is a rare cause of spinal cord dysfunction for which the etiology and natural history has remained disputed. Many theories have been reported, including congenital defect, duplication defect, and acquired dural defects caused by herniated discs (20,23,25,26). Diagnosis is often difficult due to poor knowledge of the disease (27). In our case series, the greatest length of time between symptom onset to treatment was over 30 years. When the spinal cord is tethered by the ventral dural defect, CSF pulsation and pressure gradient will slowly exacerbate the injury (28). Reducing the rate of missed diagnosis requires better understanding of the imaging features of the disease. The typical characteristics of ISCH on T2 sequences are C-shaped spinal cord attaching to ventral part of dura, and enlarged dorsal CSF space, mainly in the thoracic region.

The presence of progressive neurological symptoms is recommended for surgical intervention (19,29,30). The goal of surgery is to detether the herniated spinal cord and keep the cord at a normal anatomic position, preventing reherniation. Various surgical techniques have been adopted to reduce manipulation of the spinal cord. Laminectomy is mostly used on its own or combined with facetectomy, costotransversecetomy, or pediculectomy to provide adequate exposure (31). In our experience, 2-level or 3-level laminectomy with meticulous dissection of the arachnoid and dentate ligament is sufficient for manipulation, and intraoperative neurophysiological monitoring could further improve surgical safety. Laminoplasty might not be necessary, as it can increase the risk of spinal canal stenosis. Based on the 1 case that required reoperation, obvious scar formation could be observed during operation due to laminoplasty. Optimal ventral dural repair strategies for ISCH are still unclear. The defect can be enlarged or closed primarily with sutures (25,32). However, enlargement will cause CSF leakage, while direct closure will increase the risk of retraction of the spinal cord due to the limited surgical window. Different materials have been used to cover the dural opening, including muscle, fascia, fat, bovine pericardium, teflon, and artificial dural grafts, with or without the use of glue or sealants, since duraplasty strategy of applying a fascial flap to the dura to cover the defect (9,33,34). Because of the limited number of cases and lack of comparative studies, the optimal graft material is not known.

Fat patch has been used widely for preventing CSF
leakage in varieties of procedures, such as transsphenoidal surgery, skull base surgery, and spine surgery with good results (35-37). We believe that fat patch is suitable for the treatment of ISCH. Autologous grafts are shown to have advantages over artificial grafts in duraplasty, with lower postoperative meningitis or CSF leakage rate (38). Moreover, the fat graft positioned in the herniation cavity provides supports and prevents reherniation. The unique part of our technique is we try our best to use fat patches alone, which will minimize the foreign-body reactions. However, sometimes the size of the defect is large, and the edge of the dural defect is too thin, so there would be a high risk of CSF leakage. In these circumstances, an artificial dural patch is required. Because of the limited number of cases, the 1.5-cm threshold was empirically determined, and should be supported by more clinical data.

**Figure 2** Surgical procedure of fat patch with an artificial dural graft. (A) Ventral dural deficit and spinal cord herniation; (B) laminectomy is performed; (C) midline dorsal dural opening is made, and the herniated portion of the spinal cord is repositioned; (D) fat graft is placed into the cavity outside the dural defect; (E) artificial dural graft is placed covering the dural deficit; (F) dorsal dural opening is sutured along with the dural graft.
The recovery of clinical after surgery in our study is limited, with a mean JOA RR 17.9%. Three patients showed no recovery of muscle strength postoperatively. The herniated spinal cord observed during the operation appeared to be deforming, with the nerve tract seriously damaged, resulting in difficult nerve function recovery. However, a halted progression of symptoms was observed in all cases. We believe that patients who are treated earlier will have better outcomes. To the best of our knowledge, this study is the largest series to date using fat graft with or without artificial dural patch material for the treatment of ISCH. The radiographic and neurological follow up showed reliability and a low recurrence rate.

The present study has several limitations. First, this was not a comparative study, so the advantages of this technique over others are unclear. Second, the series was small, which could result in bias when interpreting the results. However, considering the rarity of this disease, our study provides more clinical data on the fat patch technique for the treatment of ISCH.

**Conclusions**

ISCH is a rare, and progressive, but treatable, thoracic myelopathy. Surgical treatment is necessary for progressive cases. Using fat patch, with or without artificial dural patch, is an effective strategy for ventral dural defect repair of ISCH.
Acknowledgments

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Footnote

Reporting Checklist: The authors have completed the STROBE and AME Case Series reporting checklists. Available at https://atm.amegroups.com/article/view/10.21037/atm-22-3343/rc

Data Sharing Statement: Available at https://atm.amegroups.com/article/view/10.21037/atm-22-3343/dss

Conflicts of Interest: All authors have completed the ICMJE uniform disclosure form (available at https://atm.amegroups.com/article/view/10.21037/atm-22-3343/coif). The authors have no conflicts of interest to declare.

Ethical Statement: The authors are accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. The study was conducted in accordance with the Declaration of Helsinki (as revised in 2013). The present study was approved by the ethics committee of Xuanwu Hospital, Capital Medical University [No. (2022)020]. Written informed consent for participation was not required for this retrospective study, in accordance with national legislation and institutional requirements.

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References


Figure 4 Preoperative and postoperative imaging of a patient. (A) T2-weighted sagittal magnetic resonance imaging shows thoracic spinal cord herniation preoperatively; (B) postoperative imaging reveals repositioning of the spinal cord.

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