We present four cases of IDH, including one case involving the thoracic spine.

Preoperative characteristic imaging findings can be useful for diagnosis, but intraoperative findings are required for definitive diagnosis.

It is useful to confirm the anterolateral aspect of the dural sac from the annulus fibrosus of the intervertebral disc during surgery.
**Introduction:**

Disc herniation is a common disease, but cases of intradural disc herniation (IDH) are very rare, with an incidence of 0.2%–2.2% among all cases of herniated discs.\(^1\)\(^-\)\(^2\) IDH was first reported in 1942 by Dandy.\(^4\) The typical symptoms include acute exacerbation of chronic back pain, and there is a higher incidence of cauda equine syndrome in IDH than in extradural herniations.\(^5\)-\(^7\) Preoperative diagnosis is difficult because of variable clinical and radiological presentations.

**Case reports:**

A summary of the cases is presented (Table 1).

**Case 1:**

A 33-year-old man presented with lumbago and bilateral L5 radiculopathy for 1 month. He had undergone right L5–S1 disc resection 5 years ago, left L5–S1 disc resection 2 years ago, and right L4–L5 disc resection 1 year ago. A lesion was observed in the canal at the L5 level by MRI (Fig. 1 a), and L4–L5 complete block was observed in myelography (Fig. 1 b,c). An intradural and extradural lesion was not observed during surgery but was confirmed by examination with an echo device. After dural incision,
intradural disc sequestrum was observed and removed. Postoperatively, his symptoms improved, and his course has been good at 1 year after surgery.

**Case 2:**

A 67-year-old woman presented with continuous bilateral thigh pain for 4 months. Gaolium-enhanced MRI revealed an extradural mass with peripherical enhancement at the L2–L3 level. The mass was thought to represent an extruded disc. We performed an L2–L3 discetomy. During surgery, after L2–L3 fenestration, we observed a hole in the ventral dura mater due to herniation; therefore, after incising the dorsal dura mater, we resected the intradural disc herniation. The defect in the ventral aspect of the dural sac was subsequently repaired. Postoperatively, her pain improved, and recurrence has not been observed for 3 years.

**Case 3:**

A 74-year-old man previously underwent L3–L4 fenestration and discectomy in another hospital by L3-L4 disc herniation. However, after initial improvement, his pain increased 1 month after surgery, and he consulted our institution. He was suffering from lumbago and bilateral L4 radiculopathy. Magnetic resonance imaging (MRI) revealed an intradural mass lesion at the
L3–L4 level (Fig. 2a,b) and gadolinium showed ring enhancement (Fig. 3 a,b).

Computed tomography (CT) revealed air in an intervertebral disc (Fig. 3 c).

Therefore, we performed a re-operation. During surgery, after a dorsal dural incision, a hole was observed in the ventral dura that was connected to the intervertebral disc. We removed the herniated nucleus pulposus, sutured the ventral and dorsal dura, with L3-4 instrumented transforaminal lumbar interbody fusion (TLIF). The high degree of adhesion, protrusion of the hernia into the foramen, and difficulty with adequate preservation of the facet led to the decision to resect the facet and perform TLIF. (Fig. 4)

Pathological analysis showed degenerative fibrocartilage in the resected herniation, including capillary proliferation in part, interstitial edema, and inflammation partly in the ventral dura mater (Fig. 5). Two years after the re-operation, symptoms have recovered, and recurrence of the hernia did not occur.

Case 4:

An 85-year-old woman suffered from gait disorder due to bilateral proximal leg muscle weakness for 1 month, which deteriorated rapidly. She also had sensory deficit and insensate incontinence of urine. Manual muscle testing of
the quadriceps and hamstrings gave a score of almost 3/5. MRI revealed a mass lesion in the Th12–L1 level, and the “tumor” was located in the ventral spinal cord (Fig. 6). We initially scheduled a surgery for treatment of meningioma. During surgery, after Th12–L1 laminectomy, a dorsal dura incision was made, which confirmed that herniation had occurred through a hole in the ventral dura. We resected as much as possible and then repaired the dura mater. Pathological analysis showed fibrocartilage consistent with nucleus pulposus. Two years after surgery, recurrence has not been observed; however, bilateral leg strength has not improved.

Discussion:

1 Etiology:

IDH was first reported in 1942 by Dandy, and later it was found to be a rare complication of disc protrusion. The incidence of IDH is 0.2%–2.2%, and males are four times more affected than females. Oztürk et al. reported that 92% of all IDH cases occur in the lumbar region, with only 5% occurring in the thoracic region and 3% in the cervical region.

The pathogenesis of IDH is not clearly understood, but it reportedly involves dural adhesions between the posterior longitudinal ligament and
intervertebral disc annulus, dural adhesions caused by postoperative scarring, and vulnerability of the dura to iatrogenic or congenital factors such as dural thickness and epidural adhesions along with the longitudinal ligament.\textsuperscript{10,11} In previous reports, IDH occurred most often (60\% of all cases) at L4–L5 level, because the dura mater and ventral posterior longitudinal ligament are anatomically closest at this level,\textsuperscript{12} and lumbar disc herniation frequently occurs at this level.\textsuperscript{6} On the other hand, in our report, there were two cases without surgical history, and in two of the three lumbar spine cases, IDH occurred at the L2–L3 level. In comparison with previous reports, our cases are not typical in etiology. In addition, in two cases with no previous surgery, the cause of the IDH is still unclear.

2 Symptomatology:

For lumbar level occurrence, there are no differences in clinical signs between extradural disc herniation and IDH,\textsuperscript{13} most cases are acute, and features include severe leg pain and chronic low back pain.\textsuperscript{14} However, there is a higher incidence of cauda equina syndrome in IDH than in extradural herniation.\textsuperscript{6,11} Concerning this phenomenon, Ducati et al. reported that resistance against massive nucleus pulposus was decreased and that the disc
fragment entered the dural sac and permitted herniation of a larger amount of the nucleus pulposus. Thus, this may be consistent with most observed neurological symptoms. Cauda equina syndrome reportedly occurs in 30%–60% of IDH cases. However, in our case, nerve root symptoms were also observed.

For thoracic level occurrence, >90% patients with thoracic disc herniation have occurrences at T6–T11. Thoracic disc herniations most commonly present with back pain, radicular pain, sensory impairment, and/or motor change. In contrast, our thoracic level case involved the Th12–L1 level, which is rare, and myelopathy such as bilateral leg muscle weakness, sensory deficit, and bladder rectum disorders were present. One previous report stated that only unilateral leg muscle weakness without sensory deficit had occurred at the same site. There is no specific symptom for the thoracic level IDH.

3 Diagnosis:

MRI is the gold standard for neuroimaging studies. Typical IDH imaging features include ring enhancement by gadolinium-enhanced MRI, which is essential for diagnosis and differentiation between herniated discs and
tumors, such as schwannoma and meningioma.\textsuperscript{17,18} Ring enhancement is caused by chronic granulation tissue and peripheral neovascularization.\textsuperscript{17} In IDH, on the other hand, air images with the sequestrum into the intradural space or spinal canal are sometimes seen on CT at six times the frequency observed in normal disc herniation.\textsuperscript{19} It can migrate to the epidural space through ruptures in the annulus, in most cases together with hernia fragment.\textsuperscript{20} And it may be contained in pseudocysts in which no large amounts of disc material appear to be present.\textsuperscript{21} In previous reports, complete block often occurs in myelography. Kataoka et al. reported that a common myelographic finding in IDH was complete block, which occurred in 71% patients, and incomplete block in 15%.\textsuperscript{14} However, similar images also can be seen in huge midline herniation and intradural tumor diseases.\textsuperscript{5,22} And sometimes, in discography, dural perforation can be seen.\textsuperscript{23} In our cases, ring enhancement was observed in case 3, CT air image in case 2, and complete block in case 1. As mentioned above, these characteristic images could be useful in diagnosis.

From the above, although it is not easy to accurately detect if disc herniation is located intradurally, there are several features of imaging, and IDH
imaging can help distinguish intradural from extradural lumbar cases.⁶,¹⁷,¹⁸

4 Surgery:

The final diagnosis of IDH is typically made intraoperatively period.⁹

Surgical treatment of IDH involves firm removal of the intradural disc sequestrum. It is also important to initially find the anterolateral portions of the dural sheath. All patients in this series showed adhesions between the PLL and disc. Dense adhesions between the disc space and the dural sac detected in the intraoperative phase have also been reported.¹² In addition, the nerve roots and intervertebral foramen require careful attention to avoid injury in cases of lumbar herniation.

With regard to the treatment of ventral dural defects, case report details vary widely and include closure with an autogenous fascial or fat graft, closure with an artificial membrane, and direct suturing.¹⁶ In our series, suture and TLIF were performed in one case each. There have been no previous reports of patients treated with TLIF in IDH. It was possible to perform discectomy with interbody fusion, which should prevent recurrence.

5 Outcome:

In our cases, at the lumbar level, because there was no muscle weakness
preoperatively, recovery of symptoms was observed in the early postoperative period. Previously, patients who presented preoperatively only with radicular pain had complete recovery without neurological deficits after the surgical procedure. On the other hand, in the case involving the thoracic level, leg muscle weakness remained after surgery. In this case, surgery was performed after 1 month and the patient was paralyzed; therefore, he was vulnerable to long-term compression of the spinal cord at the level of conus, which may have led to his poor postoperative recovery.

Conclusion:

We present four cases of IDH, including one case involving the thoracic spine. In preoperative imaging, differentiating intradural tumors from large disc hernias is difficult. Preoperative characteristic imaging findings can be useful for diagnosis, but intraoperative findings are required for definitive diagnosis. Thus, it is important to be aware of pathogenesis, and it is useful to confirm the anterolateral aspect of the dural sac from the annulus fibrosus of the intervertebral disc during surgery.

References:


4 Dandy WE. Serious complications of ruptured intervertebral disks. JAMA 1942;119:474-477.


8 Lesoin F, Duquennoy B, Rousseaux M, Servato R, Jomin M. Intraderal


15 Stillerman CB, Chen TC, Couldwell WT, Zhang W, Weiss MH. Experience


Table 1

Summary of the cases

Figures:

**Fig. 1** (Case1) Preoperative sagittal (a) T2-weighted MRI showing a mass in the canal at the L5 level. L4–L5 complete block was observed by myelography (b, c).

**Fig. 2** (Case3) Sagittal (a) and axial (b) T2-weighted magnetic resonance imaging show expansion and an intradural lesion at L3–L4.

**Fig. 3** (Case3) Sagittal (a) and axial (b) T1-Gd-weighted magnetic resonance imaging show ring-enhanced sign. Sagittal (c) computed tomography showing air in the disc space and air in the intradural sequestrum.

**Fig. 4** (Case3) L3–L4 posterior lumbar interbody fusion was performed. Postoperative front X-ray (a), lateral X-ray (b), and sagittal (c) T2-weighted magnetic resonance images are shown.

**Fig. 5** (Case3) The resected intradural disc hernia shows degenerative
fibrocartilage, including capillary proliferation in part (a). Interstitial edema and inflammation can be observed in a part of the ventral dura mater (b).

(Hematoxylin and eosin staining, ×40)

**Fig. 6** (Case 4) Preoperative sagittal (a) and axial (b) T2-weighted magnetic resonance imaging (MRI) showing a disc herniation at Th12–L1. Postoperative sagittal (c) and axial (d) T2-weighted MRI show a hernia resection and expansion of the spinal dura matter.
# TABLE: Summary of the cases

<table>
<thead>
<tr>
<th>Case</th>
<th>Age(y), Sex</th>
<th>Number of past surgeries</th>
<th>Herniation level</th>
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<td>4</td>
<td>85, F</td>
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<td>Th12-L1</td>
<td>Weakness of bilateral leg muscles, sensory deficit and bladder rectum disorders (1 month)</td>
<td>N/A</td>
</tr>
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</table>

N/A: not applicable
Figure 1

(a) X-ray image showing vertebrae.

(b) MRI scan of the spine.

(c) Radiograph of the lumbar spine.
Figure 2

(a)

(b)
Figure 3

a) 

b) 

c)
Figure 5

a)

b)
Figure 6

a) Th12
   L1

b) d)