



## Review article

## Intradural lumbar disc herniation: A case report and literature review

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## ABSTRACT

**Study design and objection:** Intradural disc herniation is a unusual disease associated with spinal surgery. The definitive diagnosis of intradural herniation depends on intraoperative findings.**Summary of background data:** We present the case of a 63-year-old woman with backache and left sciatica radiation for more than two months. The L2/3 laminectomy and discectomy were performed after magnetic resonance imaging (MRI) study; however, no disc rupture was noted during surgery. Follow-up lumbar spine MRI revealed one large, ruptured disc. The patient underwent revision surgery with durotomy. The large intradural disc was found and removed piece by piece.**Methods, Results, and Conclusions:** Intradural disc herniation, especially large herniation, is hard to diagnose specifically despite the progression of neuroradiologic imaging techniques. A durotomy procedure should be considered if there is a missing ruptured disc or a palpable intradural mass during surgery.

## 1. Introduction

Intradural disc herniation (IDH) is a disease presented with the penetration of the ruptured intervertebral disc into the subarachnoid space, and it is a relatively rare degenerative spine problem [1, 2]. The name of lumbar IDH was first reported by Dandy in 1942, and more than 160 patients have been described in the English language literature [3, 4, 5]. Only 0.26%–0.30% cases of ruptured disc are IDH, and most (92%) occurs in the lower lumbar spine level, commonly at the L4/5 area. The typical presentation is acute progression of low lumbar backache or soreness with the sharp sciatica pain [5, 6, 7]. At present, the pathogenesis of IDH is uncertain [8].

The most common procedures for the diagnosis of ruptured lumbar discs are the images of computed tomography (CT) and magnetic resonance imaging (MRI). Despite progress in neuroimaging techniques, making a definitely diagnosis and preoperatively evidence whether the herniated disc is located at inside or outside the dura remains challenging [9]. This difficult diagnosis may result in an incorrect surgical procedure

or incomplete surgical decompression. For most clinical situations, the diagnosis is finally made according to the operative findings and post-operative pathological report [10].

Now, we presented a rare case of IDH at the L2/3 disc level and disclosure the clinical appearances, imaging presentations, treatment options, and results.

## 2. Patient description

A 63-year-old woman complained of acute exacerbation of low back pain (LBP) and radiating numbness to the left lower limb for more than six months. The progressive and constant pain occurred in the lower back, left thigh and left leg for another month. At first, she ever went to the local orthopedic clinic, and lumbar disc herniation was diagnosed. She underwent Chinese acupuncture therapy and rehabilitation for 1 month; however, the response was poor, and she had persistent LBP. The physical and neurological examination showed the numbness was below the L2 dermatome (left side was more than right side). The muscle power of low

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limb was all five. There was no fever, hematuria, urine/stool incontinence, sexual dysfunction. Therefore, she was transferred to our outpatient department for MRI study. The mass lesion was hypodense under the T1/T2-weighted images. The MRI showed a huge ruptured disc occupying nearly the all space of L2/3 spinal canal, resulting in the compression of the cauda equina and L4/5 spondylolisthesis (Figure 1A–D).

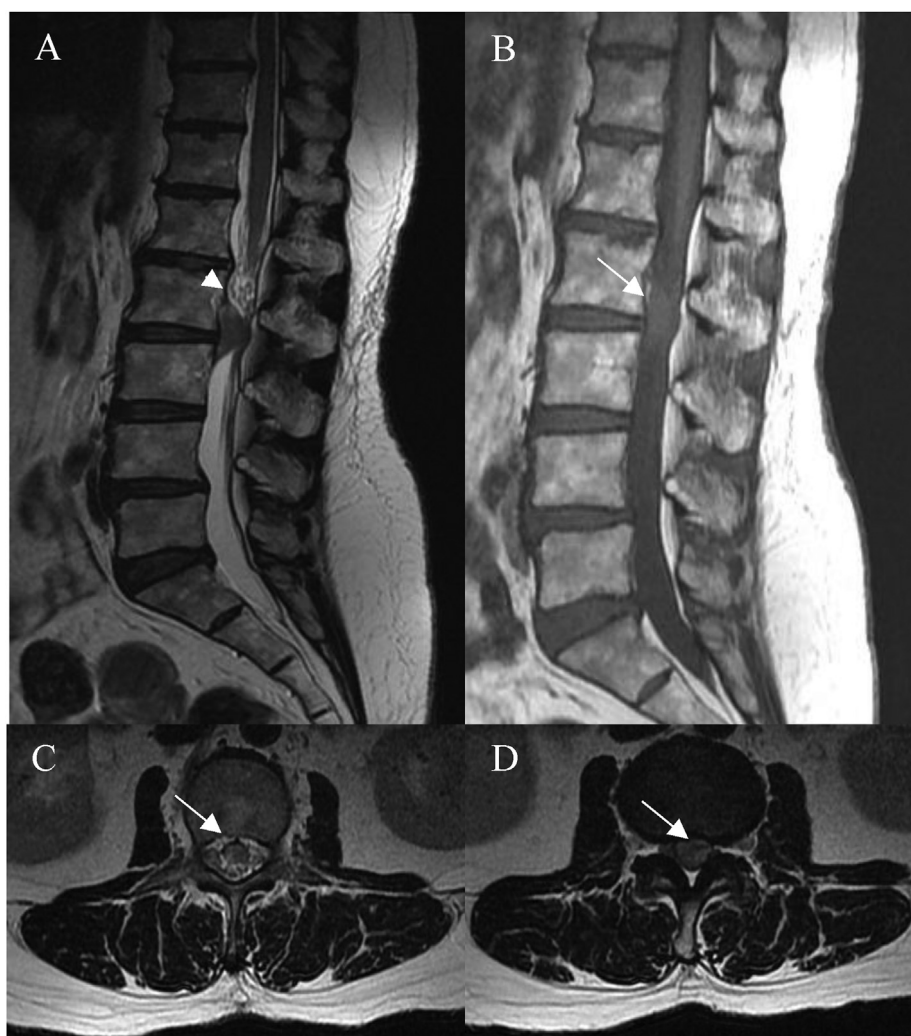
Based on the diagnosis of an L2/3 ruptured disc and L4/5 spondylolisthesis, L2/3 interlaminar microdiscectomy and L4/5 transpedicular screw fixation with cage fusion were performed. However, there was no obvious large, ruptured disc under the bilateral root at the L2/3 disc area during surgery. The correct operative spine level was double-checked using portal radiography. A repeated lumbar spine MRI was arranged because of the absence of a ruptured disc after the surgery. Follow-up MRI revealed that there was still one large L2/3 ruptured disc at the level of the laminotomy (Figure 2A, B).

According to the findings of the MRI and surgery, intradural disc herniation was suspected. This patient underwent a revisional surgery 1 week later. We enlarged the previous operative field by extending upward and downward hemilaminectomies. After opening the dura at the L2/3 level, we checked the nerve rootlets and discover the intradural disc pieces which proved a penetration from the ventral dura. The major part of disc fragments was inside the dura and adhered to few nerve rootlets due to inflammation. The bulging disc severely compressed the nerve fibers. The IDH was cautiously separated from the adjacent nerve tissue

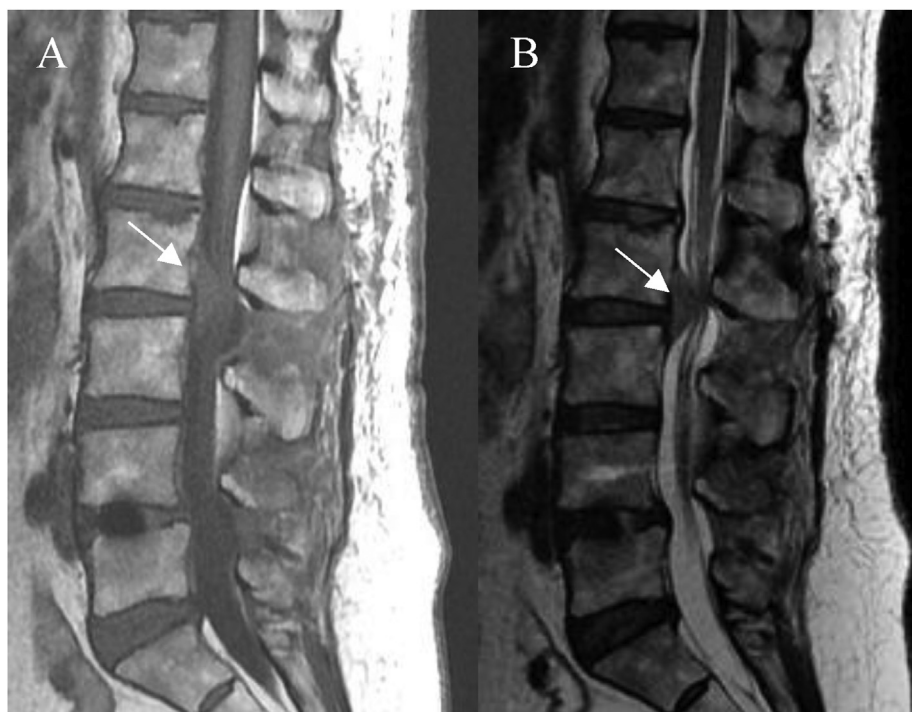
and the adhesion of dura matter. Finally, it was removed completely piece by piece (Figure 3A, B). There was no cerebrospinal fluid leakage from the ventral side perforating hole after removing rupture disc due to severe adhesion of the posterior longitudinal ligament (PLL) and fibrotic tissue. The pathologic study confirmed the finding of a degenerated ruptured intervertebral disc. The symptoms of severe LBP with left sciatica improved significantly after the second surgery.

### 3. Discussion

Lumbar IDH is a rare classification of disc herniation [11, 12]. In addition, IDH is difficult to diagnose preoperatively based on clinical symptoms or radiologic studies. Most IDH diagnoses are made intraoperatively. Only 25% of IDH cases in our literature review were diagnosed preoperatively (Table 1). When the adhesion of the dural sac was severe or the volume of the disc pieces gathered was less than that predicted by the images, the procedure of durotomy should be considered to check the intradural area for more free fragments and lesions [13]. Subsequently, a definite diagnosis could be made by the surgeon during the surgery. The most reported lumbar spinal level are L4/5 (55%), L3/4 (16%), and L5/S1 (10%). The L1/2 and L2/3 levels are rarely affected [14]. In our review of PubMed from 1999 to 2019, only 48 case reports of lumbar IDH were identified (Table 1). The male-to-female ratio was 31:1, and the average age was 52.3 years (range, 25–85 years). In this series,



**Figure 1.** MRI of the intradural disc. (A) Sagittal T2-MRI showed the continuity of the posterior longitudinal ligament and the anterior thecal sac at the level of the ruptured disc (arrowhead). (B) Sagittal T1-MRI showed a huge hypodense ruptured disc (arrow) with the disc totally occupying the spinal canal. (C, D) Axial T1&T2 MRI revealed one large central herniated disc. MRI, magnetic resonance imaging.



**Figure 2.** Follow-up MRI after the first surgery. MRI revealed that the ruptured disc still existed at the correct operative spinal level in (A) T1- and (B) T2-weighted images. MRI, magnetic resonance imaging.

T12/L1 was involved in four (8%) cases, L1/2 in four (8%) cases, L2/3 in six (12.5%) cases, L3/4 in five (10.4%) cases, L4/5 in 23 (47.9%) cases, and L5/S1 in six (12.5%) cases. L4/5 was the most common level at which IDH occurred. The surgical fusion rate was only 8/48 (16.7%). Hence, the most common procedures are simple laminectomy, durotomy, and discectomy.

The symptoms of IDH typically include chronic LBP combined with acute progression of low limb pain and/or motor weakness/incontinence [5, 15]. In our review series, the major symptom of IDH was LBP with leg pain (56%). Other symptoms included simple leg problems (27%), simple LBP (6%), cauda equina syndrome (4%), intracranial hypotension (4%), and acute paraplegia (2%). Two hypotheses about the pathophysiological mechanisms of IDH have been proposed [10, 16, 17, 18].

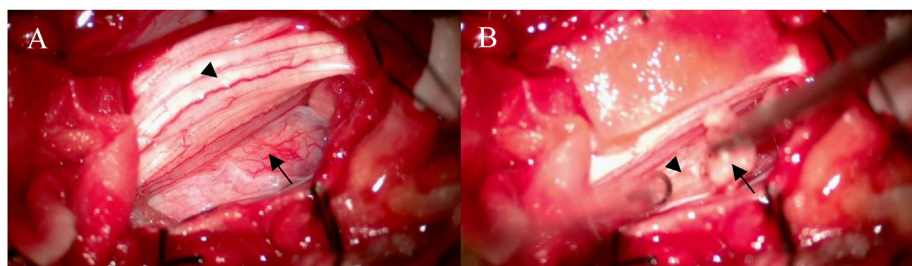
- (1) Adhesion occurs between the PLL, dural sac and fibrous annulus. The dura is eroded. A hole is created through the dura matter that causes the intradural disc penetration.
- (2) A narrow epidural space causes the congenital or iatrogenic inflammation of the dura matter, with highly susceptibility to IDH with progressive stenosis of the spinal canal.

Several MRI findings should be considered for a possible diagnosis of IDH: (1) the hawk-beak sign, a sharp-ended pathological mass

compressing the dural sac in the T2 sequence [19], (2) the “Y” sign, a “Y” shape on the ventral wall of the dural sac when the IDH separates the dura mater from the arachnoid mater [9, 20, 21], and (3) the crumble disc sign which may suggest an early stage of IDH [16]. However, no specific sign can confirm the diagnosis of IDH and, these signs may not be observed in most cases. It is important to note that IDH needs to be differentiated from other pathological tumors inside the spinal canal, such as meningioma, neuroma, metastasis, or cyst [21, 22, 23]. Gadolinium-enhanced MRI should be considered if IDH is suspected. The contrast medium promotes the differentiation of the lesion, especially for solid masses with homogeneous enhancement and fluid-filled cystic lesions [24]. However, it is not a routine examination for a simple intervertebral disc rupture [14, 25].

In most cases of IDH, a preoperative diagnosis is not possible. Most surgical experience was disc fragments, which were not found between the dura and disc level. The first impression that IDH might be present is the wrong surgical spine level. Thus, after checking the correct spine level, the operators should suspect the presentation of an IDH.

It is also critical to confirm the rupture hole of the dura mater before durotomy [26]. The ventral part of the dura mater needs to be examined because of the existence of a defective hole for disc penetration into the intra-dura. Intraoperative echography is the best and simplest procedure for the diagnosis of IDH as it can identify intradural lesions. Durotomy



**Figure 3.** Intraoperative images after durotomy under microscopic magnification. (A) The rootlets (arrowhead) were displaced by the bulging disc (arrow) from the ventral side. (B) The disc was removed piece-by-piece using forceps (arrow); the posterior longitudinal ligament (arrow-head) could be seen after the disc was removed.

**Table 1.** Review of lumbar intradural disc herniations (1998–2019).

Author	No. of cases	Age/sex	Preoperative symptoms	Level	Preop. Dx	Fusion
Ge CY [27]	1	68M	LBP with radiating pain	L4/5	N	Y
Inoue T [28]	1	52M	LBP with dysuria	L1/2	Y	N
Aprıgio RM [7]	1	57M	Chronic LBP and left thigh pain	L1/2	Y	Y
Baranowski P [19]	2	69F	LBP with low limb radiation	L3/4	N	Y
		37M	LBP, lower limb weakness	L4/5	N	Y
Fiechter M [29]	1	46F	Spontaneous intracranial hypotension	T12/L1	Y	N
Park YS [3]	1	43M	LBP with right sciatica	L3/4	N	N
Crivelli L [30]	1	73M	Right lumbosciatic pain	L2/3	N	N
Rodrigo V [10]	1	59M	Lumbar and radicular pain	L4/5	N	Y
Sharma A [16]	6	51M	LBP with bilateral radiculopathy	L4/5	N	N
		49M	LBP, cauda equina syndrome	L4/5	N	N
		33M	LBP, cauda equina syndrome	L4/5	N	N
		45M	LBP, sciatica	L4/5	N	N
		30F	Radicular pain	L4/5	N	N
		59M	LBP, radiculopathy	L4/5	N	N
Kasliwal MK [31]	1	25F	LBP	L4/5	N	N
Mailleux P [32]	1	66M	Bilateral L5 sciatica and paresthesia	L4/5	Y	N
Tamaki Y [33]	1	64M	Persistent LBP, right leg pain	L4/5	Y	Y
Kobayashi K [26]	4	33M	LBP, bilateral L5 radiculopathy	L4/5	N	N
		67F	Bilateral thigh pain	L2/3	N	N
		74M	LBP, bilaterally L4 radiculopathy	L3/4	N	Y
		85F	Weakness of bilateral leg muscles	T12/L1	N	N
Lee HW [34]	1	52M	Numbness in left calf and ankle	L1/2	N	N
Rapoport BI [35]	1	45M	Intracranial hypotension	T12/L1	Y	N
Jain SK [36]	1	40F	LBP, bilateral sciatica, urinary retention	L4/5	N	N
Krajewski KL [37]	1	70M	LBP, difficulty walking	L3/4	N	N
Kim HS [9]	1	54M	LBP, right sciatica	L2/3	N	N
Ducati LG [13]	5	44F	Urinary retention, right sciatica	L4/5	N	N
		71M	Urinary retention, L5/S1 sciatica	L4/5	N	N
		49F	Urinary incontinence, left sciatica	L5/S1	N	N
		68F	Left LBP, sciatica	L4/5	N	N
		46F	Left LBP, sciatica	L4/5	N	N
Singh PK [38]	1	30M	LBP, weakness in his both lower limbs	L4/5	Y	N
Floeth F [24]	1	47M	Recurrent LBP, acute left radiculopathy	L4/5	Y	N
Liu CC [21]	1	50F	LBP	L5	Y	N
Nagarıa J [39]	1	53M	Paraesthesia and weakness in his left lower limb	L5/S1	Y	N
Jang JW [23]	1	33M	Moderate right leg pain	L4/5	N	N
Chaudhary KS [40]	1	70M	Acute paraplegia	T12/L1	N	N
Oztürk A [6]	1	50F	Pain and weakness in both legs	L1/2	N	N
Choi JY [14]	2	65F	LBP with bilateral sciatica	L3/4	N	N
		30M	LBP with bilateral sciatica	L4/5	N	Y
Lee JS [41]	1	61M	LBP, bilateral sciatica	L5/S1	Y	N
Karabekir HS [42]	2	46M 37F	Severe leg pain, motor weakness	L2/3	N	N
			Severe leg pain, motor weakness	L5/S1	N	N
Hidalgo-Ovejero AM [43]	1	64F	Bilateral lumbosciatic pain	L2/3	Y	N
Aydin MV [22]	1	58M	LBP with right sciatica	L5/S1	N	N
Mut M [44]	1	32M	Lower back and right leg pain	L5/S1	N	N
This study	1	63F	LBP with left sciatica	L2/3	N	N

M, male; F, female; N, no; Y, yes; preop. Dx: preoperative diagnosis.

can be performed with evidence of intradural masses. A pathological study should be considered to confirm the diagnosis of IDH. However, IDH is hard to diagnose according to this information [3]. A definite diagnosis of IDH is often made based on the observation of the surgeon intraoperatively.

#### 4. Conclusion

We shared our surgical experience of a rare case of IDH. Opening and evaluation of the dural sac should be considered when an insufficient herniated disc is found after the correct spinal level is confirmed. Intraoperative echography may be helpful before performing durotomy.

#### Declarations

##### Author contribution statement

All authors listed have significantly contributed to the development and the writing of this article.

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### Data availability statement

Data included in article/supp. material/referenced in article.

### Declaration of interest's statement

The authors declare no conflict of interest.

### Additional information

No additional information is available for this paper.

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