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Case Report

Unexpected intradural disc herniation instead of space-occupying tumor at L3-L4 level: a case report and literature review

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Abstract: Background: Lumbar disc herniation (LDH) is a common disease, with a conventional treatment method, as well as well-established surgical procedure, when necessary. However, some rare cases of LDH, such as intradural disc herniation (IDH), accounting for a very small proportion (approximately 0.3%) of all LDH cases, could lead to intra-operation or post-operation complications, which requires a more circumspect pre-operational radiology analysis and overall management. Herein, we reported a case with L3-L4 IDH identified by pathological examination. Recent studies on PubMed were reviewed to summarize the unique characteristics of IDH, as well as diagnosis and treatments. Case introduction: A 69-year-old male was admitted to our department due to complaints of chronic low back pain for the past one month, along with radiating pain along the left lower hip and posterolateral left lower extremity. Disk herniation and space-occupying mass inside the canal at the L3-L4 level were confirmed by both lumbar CT and MRI. In surgery, after resection of the disc at L3-L4, further exploration revealed unsatisfactory volume of disk tissue and local eminence posterior to ventral dura, which emphasized the need for preoperatively identifying the mass inside the spinal canal. The tumor-like mass was found inside the dura. Finally, transforaminal lumbar interbody fusion (TLIF) was performed followed by resection of the mass. However, the histology examination showed a disc-like fibrocartilage tissue. The symptoms were immensely improved after the operation. Conclusion: IDH has a low incidence and is sporadically reported. Misdiagnosis is very common preoperatively as well as intraoperatively. IDH usually develops more rapidly compared with intradural tumors. Adhesion between dura mater and posterior longitudinal ligament may play a critical role in the disease onset.

Keywords: Intradural disc herniation, lumbar disc herniation, discectomy, dura mater, lumbar spine

Introduction

Lumbar disk herniation (LDH) is a common disease characterized by pain in the back and legs caused by spinal canal or nerve root compression due to dislocation of the degenerated disk material [1]. However, Intradural Disk Herniation (IDH) is a rare type since intradural migration of disk material is extremely rare, which indicates bypassing several obstacles including posterior longitudinal ligament and the dura layer. The first case was published by Dandy in 1942, after which some sporadic cases have also been reported [2]. It is often misdiagnosed as space-occupying tumor due to the typical imaging manifestations in computed tomography (CT), and magnetic resonance imaging (MRI), leading to incorrect treatment strategy and passive decision in operation. Intraoperative and post-operative pathological findings can confirm the diagnosis [3].

Herein, we reported a patient with IDH at L3-L4 level, and described the clinical presentation, stepwise surgical procedure and post-operation findings. We also reviewed the recent literature with new advances about IDH.

Case report

A 69-year-old male was admitted to our department due to complaints of chronic low back

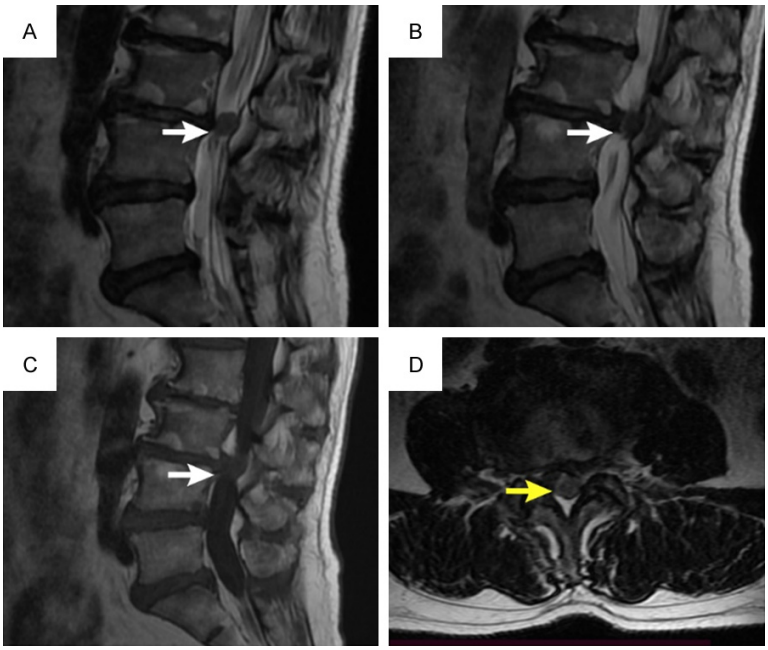

Figure 1. Preoperative MRI revealed partial intervertebral disk protrusion, and a mass in the spinal canal at the L3-L4 level with cauda equine compression. Arrows show the mass location in T2WI (A and B, white arrow) and T1WI (C, white arrow) sequences of sagittal position and corresponding transverse position (D, yellow arrow). Foraminal stenosis was not observed.

pain for around one month, along with progressive radiating pain along the left lower hip and posterolateral left lower extremity, with no response after conservative treatment such as nonsteroidal anti-inflammatory drugs (NSAIDs) or physical therapy. No cauda equina syndrome such as abnormal defecation or saddle region sensation, or other related clinical presentation was observed. Neurological physical examination revealed myotome reduction at L5 representative area, with strength level of 4/5. Lumbar CT and MRI examinations suggested space-occupying mass inside the spinal canal at the L3-L4 level. Both T1WI and T2WI sequences revealed partial intervertebral disk protrusion, and a mass in the spinal canal at the L3-L4 level with cauda equine compression [**Figure 1**]. The patient was diagnosed as prolapsed LDH, approaching inward to spinal canal, but benign intradural tumor could not be completely ruled out. The patient was advised to undergo transforaminal lumbar interbody fusion (TLIF) procedure, while rachitomy was also considered as an alternative, if necessary, after discussion with the department of neurosurgery.

During surgery, the disk was resected first as scheduled TLIF. However, very small amount of disk tissue was resected, as well as the volume of disk herniation into hinder spinal canal. No obvious herniation was observed. Further exploration showed severe adhesion between ventral dura and posterior longitudinal ligament (PLL), and a local eminence posterior to ventral dura. Based on these conditions, the surgical plan was revised to rachitomy before conducting the remaining TLIF. The posterior spinous process was removed and a median window was opened directly above the corresponding level of mass. The dorsal dura was carefully incised under surgical microscope. After dissecting cauda equina, the mass was found inside the dura in the previously identified area. The mass was circular, about 15*20 mm, with smooth surface, yellowish-white without capsule [**Figure 2A**], however, the texture was somewhat tough [**Figure 2B**], which was unlike a tumor lesion. The base of the mass was severely adhered to ventral dura wall and was carefully separated until adequate release [**Figure 2C**]. Both crevasses were sutured carefully after the mass was completely removed [**Figure 2D**]. Absorbable dura sealing glue was applied after a long strip of fascia tissue was placed on top of the sutured incision to fasten it and provide extra protection. A drainage pipe was retained before the incision was finally closed. The patient was required to remain in a supine position without pillow until the drainage pipe was removed after the drainage volume gradually reduced to <50 ml/day. Unexpectedly, one week post-operation, the histology examination showed a disc-like fibrocartilage tissue in the tumor-like mass by Hematoxylin and Eosin (HE) staining. Neuro neoplasia markers such as SOX10 were negative in further supplementary verification [**Figure 3**]. The symptoms were immensely improved after the operation.

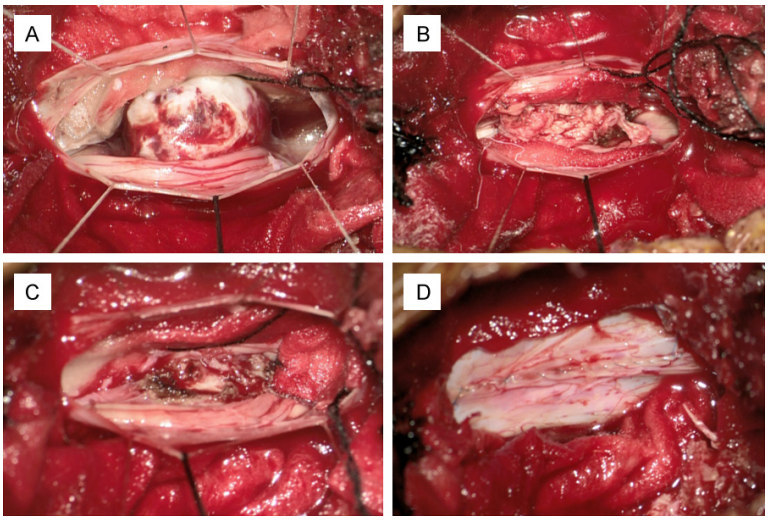

Figure 2. Intraoperative images. A. The exposed intradural mass behind the dorsal dura was opened. B. The texture of component was tough. C. Defect of ventral dura after the mass was resected. D. Both the ventral and dorsal dura were carefully sutured to prevent CSF leakage. Left side is oriented cranial end for all images.

Discussion

Lumbar disk herniation (LDH) has several subtypes depending on the location of herniated nucleus pulposus, such as bulging, protrusion, prolapse, and some rear types including intradural or transdural disk herniation. The incidence of LDH is approximately 0.26-0.30% of all degenerative disk herniations. The first case of LDH was reported by Dandy in 1942. Thereafter, LDH of L4-L5 is reported to be the most common region (55%), while L3-L4 (16%) are less reported, and L5-S1 (10%) are rarely seen at the level of L1-L2 and L2-L3 [3-5]. The patient in this report complained mainly of unilateral low back and leg pain, and presented with acute exacerbation within one month. Tushar Rathod et al. have previously reported the case of a 60-year-old male at the same level, with complaints of low backache and pain radiating to the right leg for two weeks [6]. Victor R. et al. presented the case of a 59-year-old man with lumbar and radicular pain, and recurrent lumbar herniation [7]. All these cases showed a rapid disease progression [8]. However, our patient showed no typical positive sign such as diminished ankle dorsiflexors and extensor hallucis longus (EHL), and straight leg raising test as seen in other cases.

This IDH was finally identified by pathological examination. Previously, few clinical characteristics, including complaints, physical examination (PE), and radiology features, were suggestive of the nucleus pulposus nature of intradural mass, which increased the limitation of differential diagnosis between space-occupying mass and IDH. Among the nine cases reported by Giancarlo, three were confirmed during surgery [9]. However, some valuable clues were identified when we reread the MRI images. In **Figure 1C**, similar signal between the disk and mass tissue was a clue, and in **Figure 1D**, the continuity of PLL seemed unsatisfactory as well as the rough edge. The potential pathophysiological mechanisms of IDH include congenital or acquired adhesions between the PLL and the ventral dura mater, which provide a gateway for prolapsed nucleus pulposus to approach the inner space of dura [10, 11]. We also found severe adhesion at the basal part of the mass, which seemed to be further attached to the anterior tissue in addition to ventral dura [**Figure 2B**]. Additionally, congenital narrowing of the spinal cord, and congenital or iatrogenic finesse of dura mater, are also considered as alternative reasons [12-15]. Lastly, some patients may have acquired adhesions between the annulus fibrosus, posterior longitudinal ligament, and dura mater, particularly when these are associated with postoperative adhesions caused by chronic flogosis [16-21] or due to previous surgery [18, 22-30].

In the present case, the main expected postoperative complication was cerebrospinal fluid (CSF) leakage from the sutured dura mater. Several preventive measures were taken to avoid this complication. First, both the dissected dorsal dura and isolated ventral dura due to adhesion were sutured tightly, hemostatic material was used to reduce effusion and related looseness. Fascia tissue was implanted to promote healing, along with absorbable dura sealing glue. Second, since the ventral dura is reported to be difficult to fully close by suture [31, 32], a drainage pipe was retained in order to release the pressure of spinal canal in case of any leakage. Lastly, the volume of

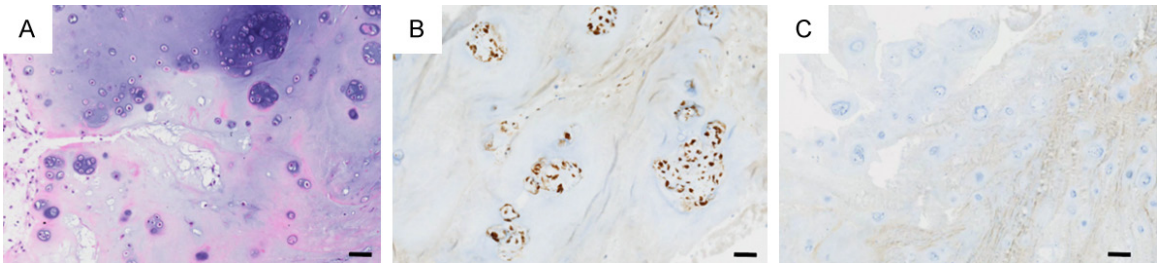

Figure 3. Histology examination showed typical cartilage manifestation by HE staining (A), which was further confirmed by positive S100 in immunohistochemistry, while SOX10 (C) expression was negative. Scale bars: 50 μm (A and B), 100 μm (C).

drainage was recorded daily, as well as the body temperature and all clinical manifestations such as headache and dizziness were comprehensively monitored. The patient was required to remain supine without pillow until the volume of drainage decreased to <50 ml/day.

Conclusion

IDH has a low incidence and is sporadically reported. Misdiagnosis is very common preoperatively and intraoperatively, which increases the risks during resection as well as post-operative complications such as cerebrospinal fluid leakage and related infections. IDH usually develops more rapidly over few weeks or months, with acute pain and numbness of lower back and limbs compared with intradural tumors. Adhesion between dura mater and posterior longitudinal ligament may play a critical role in the disease onset.

Acknowledgements

The authors certify that they have obtained all the appropriate patient consent forms.

Disclosure of conflict of interest

None.

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