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Intradural lumbar disc herniation with cauda equina syndrome: Case report and recent advances

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ABSTRACT

Introduction: Intradural lumbar disc herniations (IDDH) are unusual and represent less than 0.3% of all lumbar disc herniations. They have a higher incidence of cauda equina syndrome. The possibility of intradural disc herniation is often missed on Magnetic Resonance Imaging (MRI) and during surgery if the size of the disc prolapse is inconsistent with the compression seen on the MRI. In such situations, the possibility of IDDH should be suspected.

Research question: How to diagnose intradural disc herniation pre-operatively? Surgical techniques if the intra-dural disc herniation is encountered intra-operatively.

Material and methods: In this article, we describe a case report of an intradural disc herniation (IDDH) causing cauda equina syndrome at the L4-5 level and who underwent surgical decompression. This case report highlights that by doing a dorsal durotomy and by using microsurgical techniques, excision of the intradural disc fragment can be achieved without any rootlet injury.

Results: At a 2-year follow-up, the patient has recovered completely from motor weakness and bowel and bladder incontinence.

Discussion and conclusion: Though uncommon intra-dural disc herniation can be diagnosed pre-operatively by its characteristic signs or by using newer techniques like 3-dimensional constructive interference in steady state (CISS) MRI. Intra-operative ultrasonography (IOUS) is a handy tool to localise and diagnose intra-dural disc herniation intra-operatively and its use is encouraged. Timely intervention can lead to acceptable outcomes even with cauda equina syndrome.

1. Introduction

Intradural disc herniation (IDDH) is a rare condition characterized by the protrusion of the nucleus pulposus of an intervertebral disc into the dural sac. IDDH accounts for only 0.3% of all cases of disc herniations, with a majority occurring in the lumbar spine (92%) (D'Andrea et al., 2004). Intradural lumbar disc herniation commonly presents with a combination of low back pain, radiculopathy, and paresis. While traditional radiculopathy is associated with lumbar disc herniations in general, cauda equina syndrome occurs more frequently in cases of IDDH. Cauda equina syndrome affects around 0.5%–1% of all lumbar disc herniation cases overall but has a higher incidence rate of 30%–60% specifically in patients with IDDH(2). This entity is typically identified at the perioperative or postoperative period, we report a case study of a 54-year-old male patient who presented with complete cauda equina syndrome and was subsequently diagnosed with intra-dural disc herniation.

2. Materials & methods

A written informed consent has been obtained from the patient regarding this case report publication.

The individual, a 54-year-old farmer, sought medical attention at the Urology OPD due to acute urinary retention and underwent immediate catheterization. Further examination revealed a two-day history of intense back pain and walking difficulties. The patient had been

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previously diagnosed with chronic back pain and right thigh discomfort for three months and had been managing it conservatively. In addition, he had a ten-year history of uncontrolled diabetes mellitus with irregular treatment.

Upon clinical examination, the patient exhibited tenderness in the lumbosacral region accompanied by muscle spasms in the paraspinal muscles. Bilateral positive nerve root tension signs were observed at 40° on the left side and 50° on the right side. Motor examination revealed bilateral weakness in ankle dorsiflexors (L4 myotome) with a power of grade 4/5 according to Medical Research Council Grading, as well as extensor hallucis longus (L5 myotome) and ankle plantar flexors (S1 myotome) with a power of grade 0/5 based on MRC grading system. Sensory examination indicated reduced sensations in both L5 and S1 dermatomes bilaterally. The patient required a urinary catheter due to urinary retention, and there was also decreased perianal sensation along with diminished anal tone.

Radiographic imaging of the LS spine showed degenerative changes with calcified posterior annulus at L1-2, L2-3, and L4-5 levels. The disc heights were reduced, and there were anterior bridging osteophytes at multiple levels indicating no signs of instability (Fig. 1). Additional tests such as urodynamic studies and an MRI of the lumbosacral spine were recommended. Magnetic resonance imaging demonstrated a significant herniation of the intervertebral disc at the L4-L5 level causing compression on the dural sac contents, foraminal stenosis, and disruption in cerebrospinal fluid flow seen on sagittal images. A separate posterolateral disc protrusion was observed at L1-2 and L2-3 along with moderate narrowing (Figs. 2, 3 and 4). Further examination using urodynamic studies confirmed that there was bladder hypo-contractility which aligned with a diagnosis of cauda equina syndrome.

3. Surgical technique

The patient underwent surgery under general anaesthesia in a prone position using the Relton-Hall frame. A posterior midline incision was made from L1 to L5, and the paraspinal muscles were erased bilaterally. Using a microscope, laminectomies were performed at the L1 and L2 levels to achieve decompression. At the L4 level, a wide laminectomy was required due to cauda equina syndrome while taking precautions to minimize further damage. The calcified posterior annulus was observed and separating the highly resistant adhesions between the posterior longitudinal ligament and ventral dura proved challenging even with blunt dissection techniques (see Fig. 5).

Despite extensive exploration, no disc fragment was discovered and there was a discrepancy between the MRI results and intraoperative findings. In consideration of a large disc herniation observed on the MRI scan, there was suspicion of an intradural disc herniation. The level of the disc was reconfirmed with intraoperative imaging to ensure correct surgical placement. At this point, based on advice from the senior author, intraoperative ultrasonography using Aloka Arietta 850 was employed. Utilizing ultrasound imaging, an intradural mass was identified; it appeared as a well-defined solid lesion separate from the dura mater layer. To access this mass and remove it successfully (Figs. 6 and 7), a posterior midline durotomy was done which revealed an irregular white hard mass characteristic of an IDDH. Careful dissection allowed for the separation of rootlets and extraction of the fragments.

Upon completion of the debulking procedure, which involved removing a portion of the intradural fragment of the lesion, an anterior dural tear was discovered. This revealed a fissured intervertebral disc at the L4-L5 level. The loose fragments within the disc were extracted through this tear in its outer rim. Unfortunately, it was not possible to suture the ventral dural defect. However, after ensuring that all fragments had been completely excised from their respective locations, primary repair was performed of the iatrogenic durotomy using continuous non-absorbable sutures. This repair was then validated by conducting a Valsalva manoeuvre to confirm its integrity and functionality. Adequate hemostasis was obtained and wound was closed in water-tight fashion.

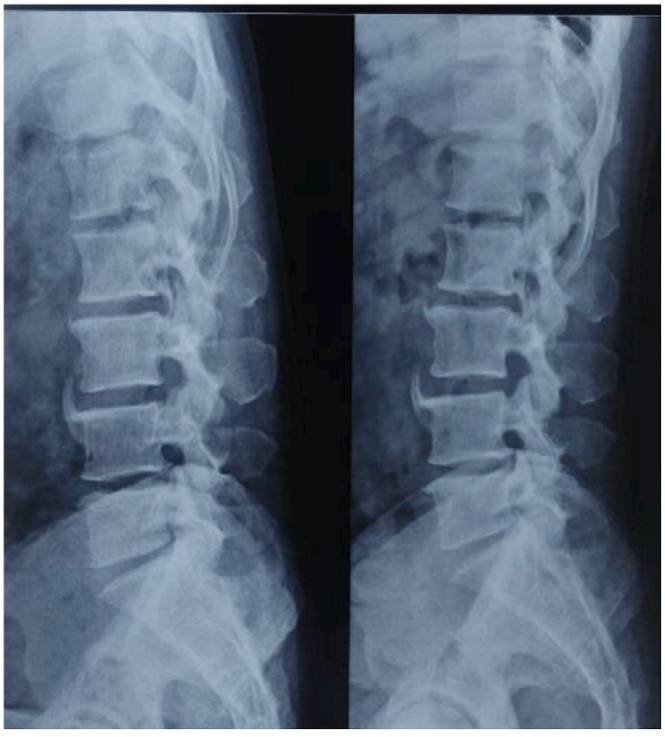


Fig. 1. Preoperative X-ray showed calcified annulus at L1-2, L2-3 and L4-5 with anterior bridging osteophytes.

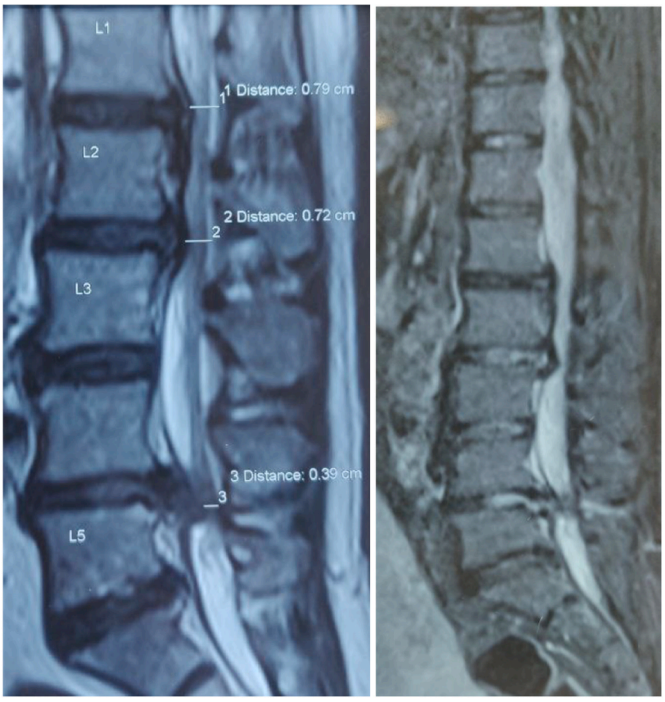
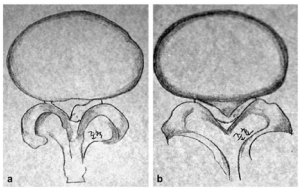
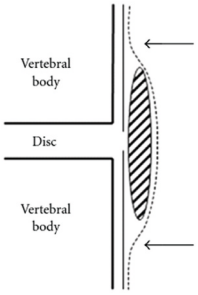
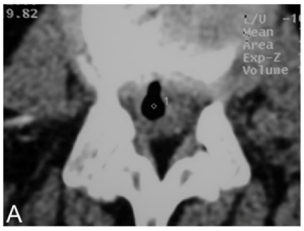
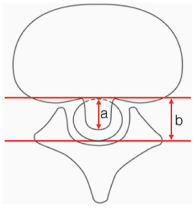
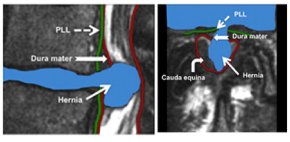


Fig. 2. Sagittal T2 weighted and STIR(Short tau inversion recovery) MRI showing abrupt disruption of PLL and intradural location of disc fragment at L4-5 level. Canal stenosis at L1-2 and L2-3 levels is also observed.

Table 1
Table showing radiological signs of Intra-dural disc herniation (IDDH)

Author's	Sign	Description	Additional comments/Figures
Choi et al. (Kataoka et al., 1989)	Hawk-beak sign	Abrupt loss of continuity of PLL and sharp beak-like appearance on T2-weighted axial imaging. Increased risk of dural tear.	
Mailleux et al. (Choi et al., 2007)	Crumble disc sign	Crumbled fragment of intra-dural herniated disc is suggestive of a sequestered fragment. Rare but specific sign of IDDH.	In chronic disc there is a peripheral ring enhancement on contrast MRI, which is not seen in acute disc herniations. The cause for ring enhancement is granulation tissue around the disc as a part of chronic inflammation. This finding can help differentiate with other intradural extramedullary tumours which enhance homogenously.
Sasaji et al. (Matsumoto et al., 2016)	Y-sign	One line of the dura and arachnoid was divided into two lines of the dura and the arachnoid. The branch of the ventral dural line appeared as "Y." This branch may have been a characteristic appearance of intradural extra-arachnoid lumbar disc herniation, and, thus, the authors called it the "Y - sign."	
Hidalgo-Ovejero et al. (Mailleux et al., 2006)	Intradural gas	Gas in disc or canal on CT is associated with intra-dural disc herniation. The gas is made up of nitrogen (90–92%) and carbon dioxide. In some cases, it can be associated with pseudocyst.	
Nam et al. (Hidalgo-Ovejero et al., 2004)	MHDD/CCD ratio	Ratio of Maximum herniated disc diameter to central canal diameter. If the ratio exceeded 0.6, IDDH is likely.	
Crivelli et al. (Arrigo et al., 2011)	Use of 3D CISS Sequences (3dimensional constructive interference in steady state)	Three-dimensional high-resolution CISS should be considered when IDH is suspected on conventional MRI to improve surgery planning.	

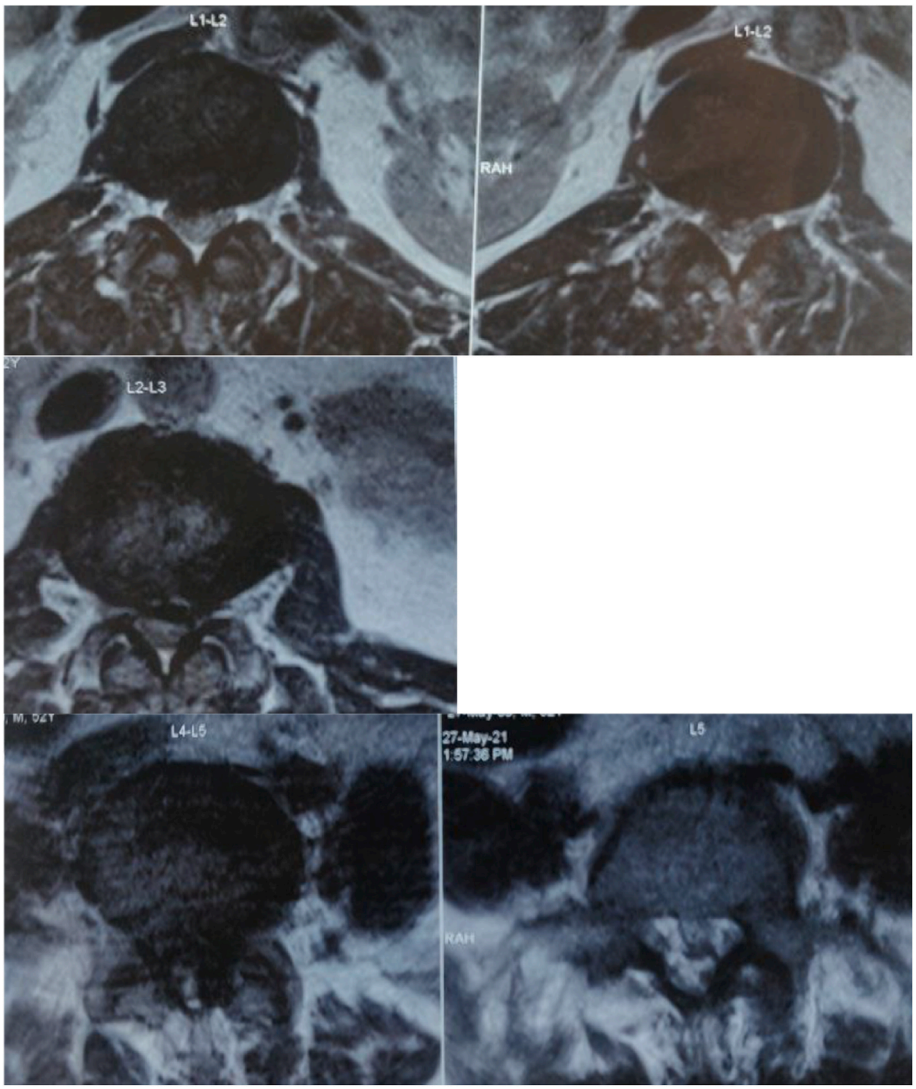


Fig. 3. Axial images of L1-2 and L2-L3 show canal stenosis with compression of the exiting nerve root. Axial MRI of L4-5 showing beak-like left posterolateral extrusion of disc fragment protruding into the dural sac occupying most of the canal diameter at L4-5 level (hawk-beak sign).

4. Results

The post-operative period was uneventful. The specimen was proved to be of discogenic origin by histopathological examination. Patient was followed-up regularly and at 02-year follow-up patient has regained complete bowel and bladder control with near normal motor and sensory power with mild perineal anaesthesia.

5. Discussion

Intradural disc herniation (IDDH), was first described by Walter E. Dandy in 1942 (Dandy, 1942), and only a few hundred cases have been documented in English literature to date. This term is actually misleading, as it represents a natural progression of disc herniation. It is more commonly observed in the lumbar region (92%), followed by the thoracic (5%) and cervical spine (3%). The L4-L5 level is the most frequently affected site (55%), with rare involvement of L3-L4 and L5-S1 levels, and even rarer occurrences at L2-L3 or L1-L2 levels (Oztürk et al., 2007). Intradural lumbar disc herniations are uncommon and account for approximately 0.26–0.3% of all disc herniations. Intradural disc herniation in thoracic spine may manifest as giant calcified thoracic discs due to dense calcification. The occurrence of IDDH in cervical spine can lead to significant neurological impairments such as Brown-Sequard syndrome and incomplete/transient quadriparesis (Guan et al., 2018). In the lumbar spine, two-thirds can present with cauda equina (67%) and one-third can present with radicular symptoms mimicking tumours thus the importance of frozen-section in diagnosing them (Aydin et al., 2004).

Despite the advances in neuroimaging techniques, it is very common to miss it on pre-operative imaging. D'Andrea reviewed 122 cases of lumbar IDH and found that only 8 cases could be diagnosed pre-operatively (D'Andrea et al., 2004). Matsumoto further quotes that only 13% of cervical and 7% of lumbar IDDH are diagnosed pre-operatively (Matsumoto et al., 2016). Association with multiple disc herniations is common as in our case. This not only highlights the fact that it has increased sensitivity for IDH. Another factor for not diagnosing pre-operatively is its very low incidence, it is often not suspected by the surgeon before surgery as it is uncommon, leading to difficulties during surgery. Other spinal pathologies that must be considered in the differential diagnosis of this Intradural extramedullary (IDEM) lesion are neurofibroma, lipoma, meningioma, epidermoid tumour, arachnoid cyst, arachnoiditis and metastasis (Sasaji et al., 2012).

IDDH can also be divided into intra-arachnoid and extra-arachnoid depending on its relationship with the subarachnoid membrane. In the case of intradural extra-arachnoid disc herniation, which is rare, the arachnoid is peeled off from the dura by the disc herniation and the disc

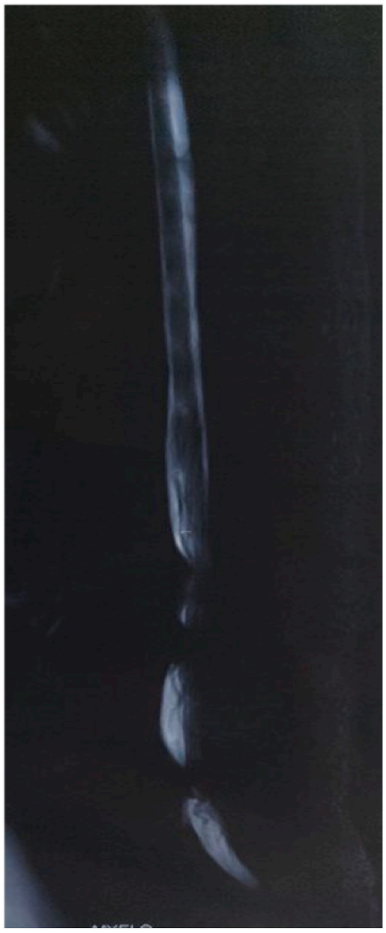


Fig. 4. Myelogram showing canal stenosis at respective levels.

fragment enters this virtual space but does not reach the rootlets and the CSF. In such cases, a CSF leak might not occur despite durotomy. In 2001, Mut et al. proposed a classification based on dural anatomy. He classified them into Type A which can be herniation of a disc into the dural sac and herniation of a disc into the dural sheath in the preganglionic region of the nerve root, named intraradicular disc herniation. (Type B) (Mut et al., 2001). The majority of Type B herniations occur at L5-S1 and Type A at L4-L5 level.

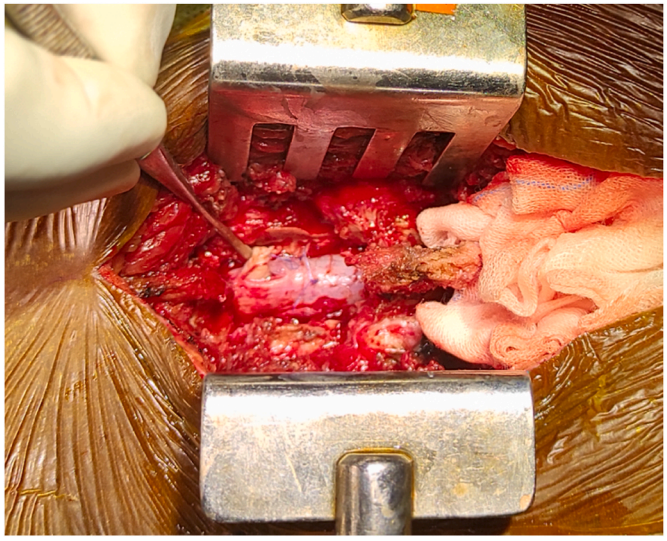


Fig. 6. Intraoperative image showing tense dura after laminectomy with disc fragment seen after dorsal durotomy. Careful dissection of rootlets from the disc fragment to facilitate its expulsion.

The etiopathogenesis of IDDH is not yet clearly understood. Several authors have found in autopsies, the presence of adhesions between the ventral dural sheath and the posterior longitudinal ligament (PLL). However, in some cases, these adhesions may be extremely resistant, and unable to be separated through blunt dissection, being an important predisposing factor. These adhesions can be present congenitally as demonstrated by Akhaddar et al. It can also be acquired due to chronic degenerative disc disease or inflammatory disease such as rheumatoid arthritis (Akhaddar et al., 2010). Ducati demonstrated that the dura mater and PLL were anatomically closest at the L4/5 level, and such adhesions occurred more often at this level, which may explain the higher incidence of IDDH in this region. Huge dimensions of the herniated fragment especially if calcified in nature, congenital or acquired spinal canal stenosis, congenital reduction of dural thickness and previous lumbar spine surgery all form a perfect setting for intradural extension of a herniated disc (Ducati et al., 2013) (see Fig. 3).

MRI with a high index of suspicion will be able to differentiate between intradural and extradural herniation to a considerable extent.

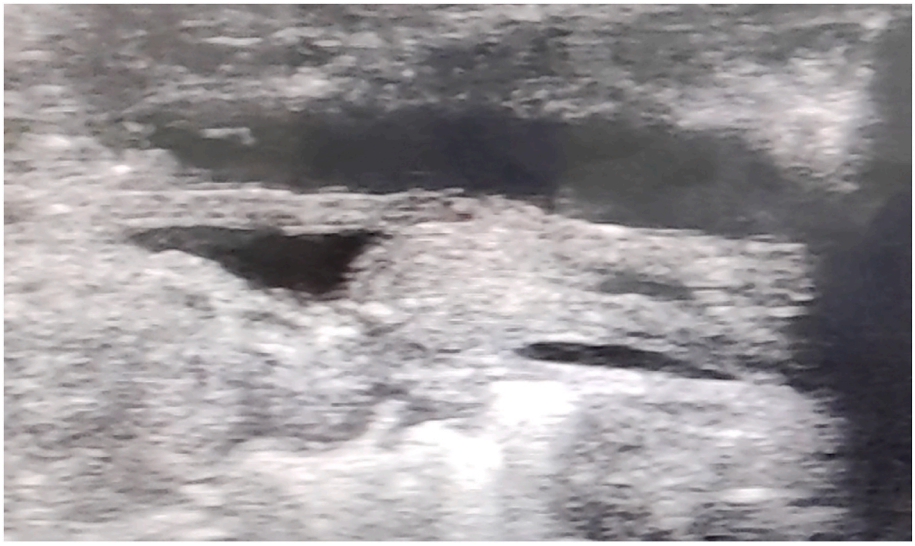


Fig. 5. Intra-operative Ultrasonography picture showing rootlets with an intra-dural mass.

Fig. 7. Disc fragment excised measuring about 2.7cm × 0.8mm from the dorsal durotomy.

Contrast MRI can help in differentiating in scars and disc fragments. Various pathognomonic features are described in the literature which are tabulated in Table 1. CT scan sometimes demonstrates air shadows located in the epidural or intradural space according to discal fragment migration. This occurs at 6 times the frequency observed in normal disc herniation and aids us to suspect an IDDH. On myelography, Kataoka et al. found a complete block in 71% of the cases of IDDH and an incomplete block in 15% (Kataoka et al., 1989). Our case demonstrated similar findings (Fig. 4). Discography and disco-CT are both useful as preoperative diagnostic tools for the diagnosis of IDH. They demonstrate leakage of the contrast medium into the intradural space. Crivelli et al. concluded that when there is a suspicion of intra-dural disc herniation, it is better to employ three dimensional constructive interference in steady state sequence(3D CISS, Table 1) to improve surgical planning, In conclusion, a poor margin, a beak-like herniated disc, an abrupt posterior longitudinal ligament discontinuity, Y-sign on ventral dura, disc material beyond PLL, disc calcification or ossification of a herniated disc and a high MHDD/CCD ratio were found to be associated with the presence of IDH (Choi et al., 2007) (Sasaji et al., 2012) (Nam et al., 2021) Diagnosis of this uncommon entity is difficult unless it is suspected preoperatively. MRI with gadolinium contrast is the investigation of choice if this is suspected preoperatively. In summary, radiologic diagnostic tools are helpful in defining the localization of herniated disc fragments in the spinal canal, but it is not always possible to predict with certainty if it is extradural or intradural.

In the present case, we did not suspect it preoperatively, which led to some difficulty and delay in intraoperative decision-making, prolonging the operative time. Only on a retrospective review of MRI data, did we note various findings suggestive of intradural herniation, such as a complete block in T2 sagittal and axial images; loss of continuity of the PLL; “beaklike” appearance of the extruded disc and a high MHDD/CCD ratio suggesting the probability of an intradural disc herniation. Such unrecognized variants of the presentation can lead to unsuccessful surgery necessitating revision procedures and possibly misidentification as failed back syndrome. Thus, this highlights the importance of diagnosing such rare presentations and knowing about their diagnostic clues and management strategies.

Surgical management includes laminectomy, dorsal durotomy, careful separation of disc material from rootlets (to reduce the risk of iatrogenic nerve root injury from surgery inside the dura), excision of the intradural disc fragments, a check for any extradural disc fragments, and repair of both dorsal dura and ventral dura (if possible). Precaution must be taken in case of cauda equina syndrome by doing wide laminectomy and using small size rongeur to prevent iatrogenic damage to already compromised neural tissue. Treatment of ventral dural defects varies in the literature. In many cases, direct suturing of a large defect is impossible, and repair is done with an autogenous fascial or fat graft or by closure with an artificial membrane. The anterior dural tear is difficult to close by primary suturing.

The use of the surgical microscope is essential for good visualization of the herniation after the dural incision, minimizing injuries to the nervous rootlets. Histopathological examination is sine qua non in such discs to reconfirm the diagnosis. Sometimes frozen section may be needed if there is doubt in the diagnosis as demonstrated by Lee et al. and Yu et al. (Lee and Suh, 2006)(Yu et al, 2021) There are other ways to reach the end-point as reported by Kobayashi et al. who reported that a discectomy with interbody fusion was performed to completely remove the disc for a recurrent herniated disc which was intra-dural in location (Kobayashi et al., 2014). Kim et al. performed a transforaminal endoscopic technique to manage intradural disc herniation at an L2-L3 level and obtained good clinical outcomes. They had sealed the antero-lateral dural defect through which the fragment was received with Gelfoam and sealants. Advantages of Endoscopic over traditional methods include less collateral damage and early mobilization of patient (Kim et al., 2018). Huliyappa et al. suggest that in minimal invasive cases, a muscle patch with fibrin glue alone may be sufficient rather than suturing the dural defect in transdural herniation (Huliyappa et al., 2017). Anterior lumbar interbody fusion was another alternative in the treatment of IDDH that can avoid unnecessary retraction of the nerve root and dura as has been reported by Choi JY et al. However, it is limited to the patients in whom the definite diagnosis was made preoperatively (Choi et al., 2007).

An intraoperative ultrasound (IOUS) may be used to confirm an intradural mass in such a situation. Harel et al. stated their experience with the use of IOUS in 16 cases of thoracic disc herniation. They state it is low-cost, easy to use and has a short learning curve. It was also used in our case, and we encourage its use in doubtful scenarios (Harel and Knoller, 2016). Wei et al. recently described the use of intra-operative ultrasonography (IOUS) when the dorsal dura appears tense or if the disc fragment is not found and there is suspicion of intra-dural fragment. Advantages include knowing the location of fragment, anatomy and displacement of nerve roots and planning the durotomy. In the above-stated case report after L3 & L4 Laminectomy for a migrated L3-4 disc herniation after intra-operative ultrasonography, a para-median durotomy was done instead of a routine dorsal durotomy (Wei et al., 2023).

The prognosis of this condition is related to the duration of symptoms, presentation (radiculopathy vs. CES) and the presence of previous surgery. In cases where there are symptoms of cauda equina compression (CES), the recovery takes longer and can be incomplete. In CES, the prognosis is mainly related to the time elapsed from the beginning of the symptoms till the surgery, which must be performed within the first 48 hours (Arrigo et al., 2011). Our patient had undergone surgery within this ideal period which significantly relieved the symptoms and led to successful neurologic recovery. Postoperatively, there was no neuro worsening, demonstrating that gentle manipulation of the rootlets under a microscope does not cause any neurological deterioration. He was on intermittent self-catheterization until the end of 6 months and returned to near normal albeit with perineal anaesthesia.

6. Conclusion

So, in conclusion, despite the advancements in radiological imaging, intradural disc herniation is often missed preoperatively. The possibility of an IDDH should be suspected when the size of the disc prolapse is inconsistent with the compression seen on the MRI. In such cases, dorsal durotomy is mandatory for the excision of the disc fragment and to achieve a favourable neurological outcome (Yuan et al., 2020). Having preliminary knowledge can make a surgeon adequately prepared. In such a scenario, intraoperative ultrasonography can be handy. The likelihood of pre-operative diagnoses can be improved with the advent of newer imaging techniques like 3D CISS sequences. (Crivelli and Dunet, 2017).

Declaration of competing interest

None for all authors.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.bas.2023.102724.

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