

Intradural Disc Herniation Concurrent with Ossification of the Posterior Longitudinal Ligament, Ossification of the Ligamentum Flavum, and Cauda Equina Schwannoma at the L1-L2 Level: A Case Report

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Intradural disc herniation (IDH) accounts for <0.3% of all cases of disc herniation¹⁾. In this study, we report an extremely rare case of IDH concurrent with multiple compressive lesions at the L1-L2 level that presented as severe cauda equina syndrome.

A 66-year-old man without significant medical history presented to our hospital with low back pain and numbness and weakness in both lower limbs for the past 4 days, with rapid progression of paralysis of the lower extremities. Neurological examination has revealed marked hypesthesia and paresthesia of both lower limbs and weakness of the distal muscles in the lower extremities (manual muscle testing [MMT] grade: 3/5 on the right side and 2/5 on the left side). Preoperative computed tomography revealed ossification of the posterior longitudinal ligament (OPLL) at the L1-L3 levels and ossification of the ligamentum flavum (OLF) at the L1-L2 level (Fig. 1). Preoperative magnetic resonance imaging (MRI) indicated a space-occupying lesion in the spinal canal at the L1-L2 level (Fig. 2). The patient could not undergo contrast-enhanced MRI as planned due to severe pain.

Urinary retention and worsening of paralysis in his lower limbs (MMT grade of the distal muscles: 2/5 on the right side and 0/5 on the left side) were noted 19 hours after hospitalization; emergency surgery was performed 30 hours after hospitalization based on a preoperative diagnosis of progressive cauda equina syndrome due to compression of the

cauda equina at L1-L2. The dural tube appeared to be tense and bulged, even after posterior decompression involving removal of OLF at L1-L2, with tight adhesion between the ventral side of the dura and OPLL. The dural sac was opened posteriorly, where a cauda equina tumor was observed in the dorsal part of the intradural space; a yellowish white mass that appeared like IDH was also seen in the ventral area of the intradural space (Fig. 3A). After removing the intradural yellowish white mass (Fig. 3B), the cauda equina tumor was excised using the intracapsular enucleation procedure (Fig. 3C), and the dura mater was closed posteriorly; we could not perform primary suture of the anterior dural tear because it was determined to be technically difficult, as has been reported previously^{2,3)}.

Histopathological examination revealed that the cauda equina tumor was a schwannoma (Fig. 4) and that the yellowish white mass was the nucleus pulposus of the herniated disc. Four months postoperatively, the patient was able to walk without aid, with almost full recovery of motor function of the lower limbs and the urination disorder resolved.

IDH concurrent with other compressive lesions at the same spinal level has been identified to be rare^{4,5)}, and to the best of our knowledge, this is the first report of a case of IDH concurrent with an intradural tumoral lesion at the same spinal level. Currently, the pathogenesis of IDH remains unclear^{6,7)}; several reports have described that adhesions between the posterior longitudinal ligament and ven-

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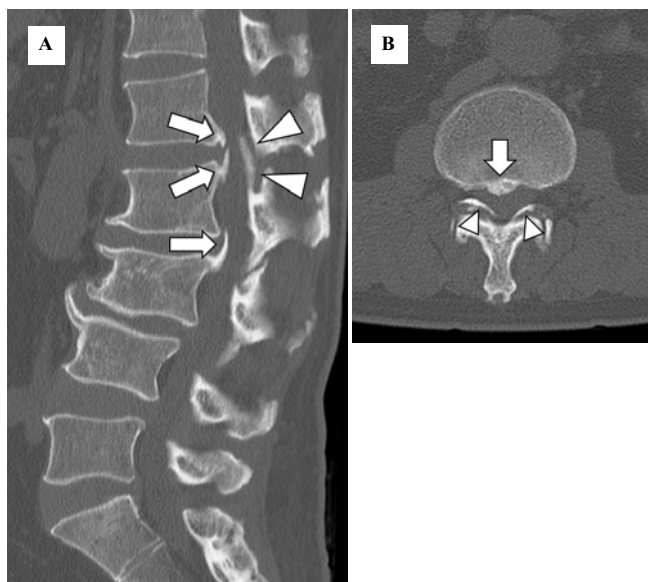


Figure 1. A) Preoperative mid-sagittal CT scan showing OPLL at the L1-L3 levels (arrows) and OLF at the L1-L2 level (arrowheads). B) Preoperative axial CT scan at the L1-L2 level showing OPLL (arrow) and OLF (arrowheads).
CT, computed tomography; OLF, ossification of the ligamentum flavum; OPLL, ossification of the posterior longitudinal ligament

tral dura mater may play an important role^{2,6-8}). Thus, OPLL may be associated with the development of IDH in this case.

Though MRI has been widely used, preoperative accurate diagnosis of IDH remains difficult; thus, IDH is definitively diagnosed based on surgical findings only after dural incision^{4,8,9}). Several reports have shown the difficulty in using MRI findings alone to distinguish between IDH and spinal tumors^{3,6,9,10}); similarly, we could not diagnose the coexistence of IDH and schwannoma with the use of preoperative plain MRI findings.

Our patient presented with severe cauda equina syndrome probably because IDH developed concurrently with subclinical spinal canal stenosis due to OPLL, OLF, and cauda equina schwannoma. Good postoperative neurological recovery was achieved possibly because of early surgery performed after symptom onset.

Conflicts of Interest: The authors declare that there are no relevant conflicts of interest.

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Author Contributions: Masayuki Tanabe wrote and prepared the manuscript. Satoshi Ogihara contributed to the conceptualization and design of the study, collected the patient's data, and edited the manuscript. Shunpei Iida, Satoshi Ikemune, Jun Kikuchi, and Kazuo Saita collected the patient's data. All authors have read, reviewed, and approved the article.



Figure 2. A) Preoperative mid-sagittal T2-weighted MRI scan showing an intra-spinal canal space-occupying lesion compressing the cauda equina (arrowheads) at the level of L1-L2, associated with spinal canal stenosis due to OPLL and OLF. B) Preoperative axial STIR MRI scan at the level of the L1-L2 disc showing a heterogeneous high-intensity lesion (arrowhead) nearly compressing the entire spinal canal. C) Preoperative axial STIR MRI scan at the level of the upper L2 endplate showing that the nerve rootlets of the cauda equina (arrows) are laterally pushed to the right and the left sides.

MRI, magnetic resonance imaging; OLF, ossification of the ligamentum flavum; OPLL, ossification of the posterior longitudinal ligament; STIR, short-TI inversion recovery

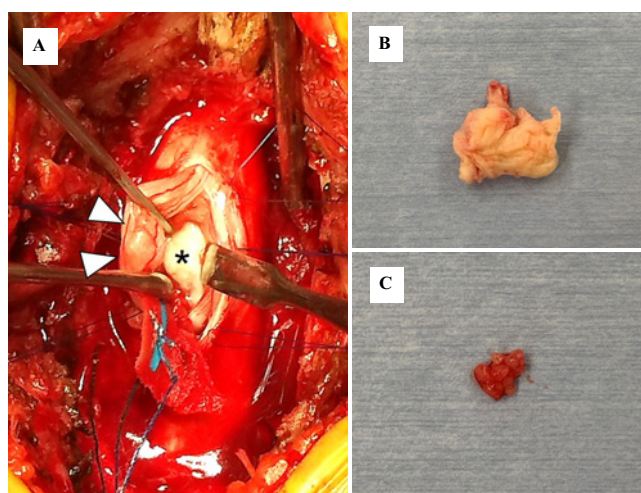


Figure 3. A) Intraoperative photograph showing that the lesion is similar to the intradural disc hernia (asterisk) and the tumoral lesion continuous with a cauda equina rootlet (arrowheads). B) Excised specimen of the lesion is similar to the intradural disc hernia. C) Specimen of the parenchyma of the cauda equina tumor excised using the intracapsular enucleation procedure.

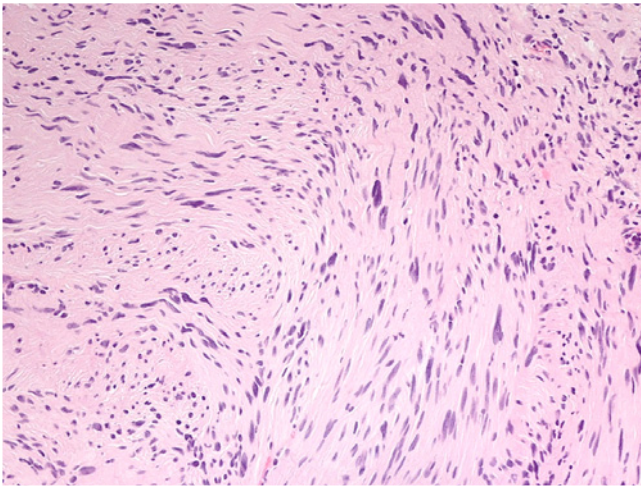


Figure 4. Hematoxylin and eosin staining of the resected specimen of tumor in the cauda equina. High-power field (200×) reveals the proliferation of spindle cells with mild polymorphism.

Ethical Approval: Not required.

Informed Consent: Informed consent was obtained from the patient in this study.

References

1. Floeth F, Herdmann J. Chronic dura erosion and intradural lumbar disc herniation: CT and MR imaging and intraoperative photographs of a transdural sequestrectomy. *Eur Spine J.* 2012;21(Suppl 4):S453-7.
2. Rodrigo V, Claramonte M, Martín M, et al. Intradural disc herniation: how I do it. *Acta Neurochir (Wien).* 2018;160(5):945-7.
3. Aydin MV, Ozel S, Sen O, et al. Intradural disc mimicking: a spinal tumor lesion. *Spinal Cord.* 2004;42(1):52-4.
4. Wang D, Wang H, Shen WJ. Spontaneous cervical intradural disc herniation associated with ossification of posterior longitudinal ligament. *Case Rep Orthop.* 2014;2014:256207.
5. Hsieh JH, Wu CT, Lee ST. Cervical intradural disc herniation after spinal manipulation therapy in a patient with ossification of posterior longitudinal ligament: a case report and review of the literature. *Spine (Phila Pa 1976).* 2010;35(5):E149-51.
6. Luo D, Ji C, Xu H, et al. Intradural disc herniation at L4/5 level causing cauda equina syndrome: case report. *Medicine (Baltimore).* 2020;99(7):e19025.
7. Inoue T. Pure conus medullaris syndrome without lower extremity involvement caused by intradural disc herniation at L1/2: a case report. *Spine Surg Relat Res.* 2018;3(4):392-5.
8. Choi JY, Lee WS, Sung KH. Intradural lumbar disc herniation-is it predictable preoperatively? A report of two cases. *Spine J.* 2007;7(1):111-7.
9. Liu CC, Huang CT, Lin CM, et al. Intradural disc herniation at L5 level mimicking an intradural spinal tumor. *Eur Spine J.* 2011;20(Suppl 2):S326-9.
10. Lee JS, Suh KT. Intradural disc herniation at L5-S1 mimicking an intradural extramedullary spinal tumor: a case report. *J Korean Med Sci.* 2006;21(4):778-80.

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