

Case Report

Lumbar Intraspinal Extradural Ganglion Cysts

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The lumbar intraspinal epidural ganglion cyst has been a rare cause of the low back pain or leg pain. Ganglion cysts and synovial cysts compose the juxtafacet cysts. Extensive studies have been performed about the synovial cysts, however, very little has been known about the ganglion cyst. Current report is about two ganglion cysts associated with implicative findings in young male patients. We discuss about the underlying pathology of the ganglion cyst based on intraoperative evidences, associated disc herniation at the same location or severe degeneration of the ligament flavum that the cyst originated from in young patients.

KEY WORDS : Ganglion cyst · Synovial cyst.

INTRODUCTION

Intraspinal cystic lesions have been noted to be as high as 1-10% in patients having low back pain or radiculopathy^{3,8,17,21}. The "juxtafacet cyst" has been the most common entity of intraspinal cysts and encompasses the synovial cyst and ganglion cyst¹². Extensive studies have been performed about the synovial cyst, however, very little has been known about the intraspinal extradural ganglion cyst^{1,2,19}. Two types of cysts share common symptomatic and radiologic characteristics but different histological background. Therefore, new name has been introduced to replace the "juxtafacet cyst" widely used since Kao first employed^{6,12}. Regarding the ganglion cyst, many authors used various names including facet cyst, posterior longitudinal ligament (PLL) cyst, disc cyst or ligament flavum (LF) cyst depending on the origin^{1,10,11,19}. On the other hand, some authors used the synovial cyst as a representative of the juxtafacet cysts including the ganglion cyst^{20,22}. Synovial cysts are based on the laxity of the synovial capsule of the facet joints and frequently associate with spinal instability⁸. The ganglion cyst is believed to arise from degenerated ligaments including the PLL and LF in the mobile spine^{1,2,15,19}. We use "ganglion

cyst", if the cyst originated from the ligamentous structures and lacked a true synovial lining without continuity to the facet joint, because they have common pathology regardless of the origin.

We report two cases of the ganglion cyst with intraoperative images matched to radiologic findings. To our knowledge, there has been no report with intraoperative images of underlying ligamentous degeneration or associated herniated disc at the same location.

We reviewed the literatures with focus on the underlying pathology of the ganglion cyst distinguished from the synovial cyst.

CASE REPORT

Case 1

A 37-year-old man presented with suddenly developed lower back pain and left leg pain after slip down during playing soccer 3 weeks ago. The pain radiated to the left L4 dermatome. Straight leg raise capacity was diminished to 45 degrees on the left. Sensation was diminished in the left L4 and L5 dermatomes. Motor function was normal. Magnetic resonance imaging (MRI) demonstrated a large cystic lesion and extruded disc at L3-L4 level. The cyst was noted along the L4 upper end plate and pressed down by the disc herniation. The cyst compressed the dural sac and left L4 root traversing the L3-L4 level. The lesion was isointense on a T1-weighted image and hyperintense on a T2-weighted image (Fig. 1A-C). Based on these findings, provisional

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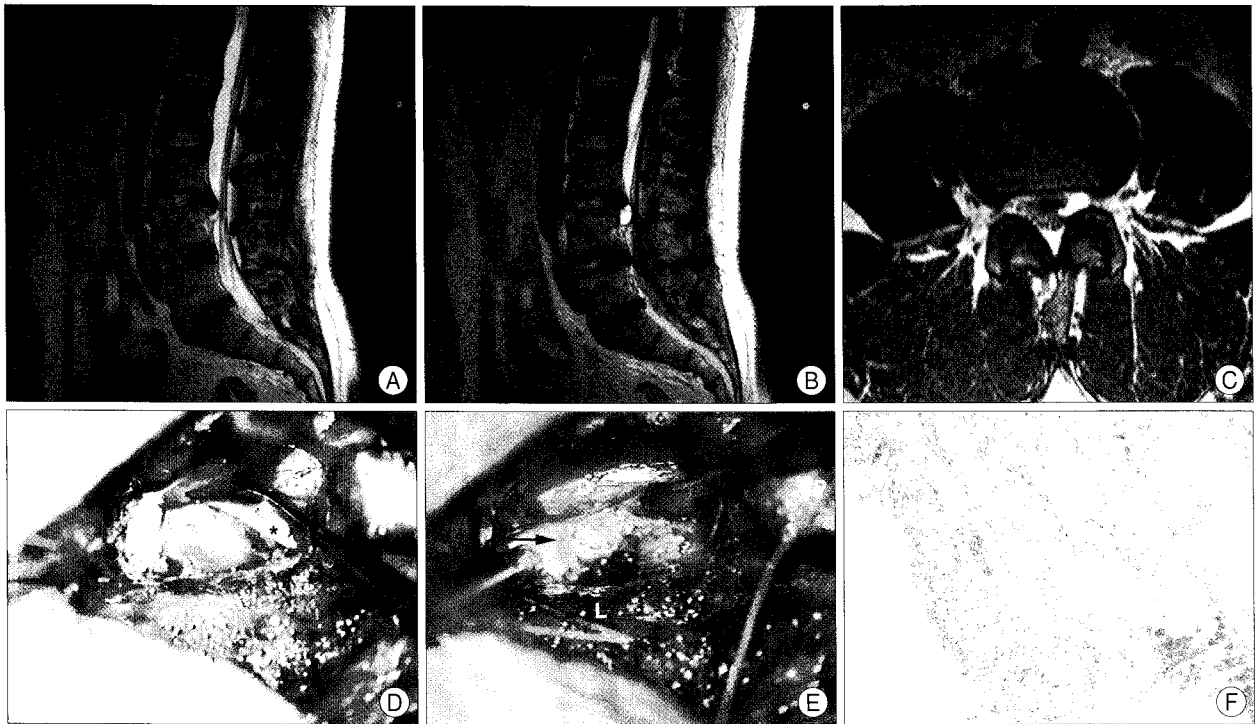


Fig. 1. Images of case 1. A and B : Preoperative sagittal T2-weighted magnetic resonance images showing a cystic lesion adjacent to the extruded disc material at L3-4. C : Preoperative axial T2-weighted magnetic resonance image showing disc herniation medial to the cystic lesion. D : Intraoperative photograph showing a large cystic mass filled with dark fluid. E : Intraoperative photograph showing the herniated disc material (arrow) just above to the origin of the shrank cyst (arrow head). F : Photomicrograph shows thick fibromembranous tissue with no lining epithelium. The thick walls show some fibroblasts and mild myxoid stromal change. H & E, original magnification $\times 100$. * : Nerve root, C : cyst, D : dura, L : lamina, SP : spinous process.

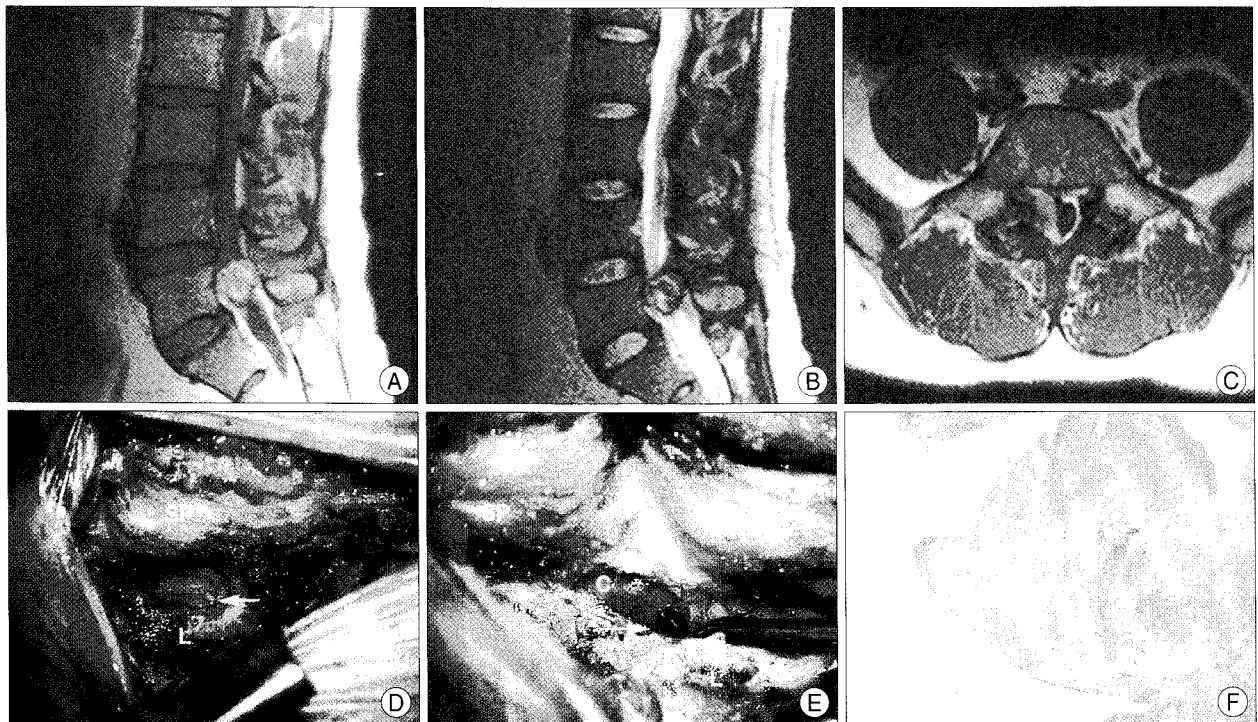


Fig. 2. Images of case 2. A and B : Preoperative sagittal magnetic resonance images showing a large round mass with high signal on T1-weighted image and heterogeneous high and low signal intensity on T2-weighted image. C : Preoperative axial T1-weighted magnetic resonance image showing the mass arising from the right ligamentum flavum of L5. D : Intraoperative photograph showing severely degenerated darkened yellow ligament (arrow). E : Intraoperative photograph showing fresh hematoma through an eye of a needle (arrow). F : Photomicrograph shows an irregular thick-walled cystic space with no lining epithelium. Old hemorrhagic areas are noted in the fibrotic stoma. H & E, original magnification $\times 40$.

diagnosis was herniated intervertebral disc combined with a hematoma or ganglion cyst. A standard microsurgical approach for the subtotal laminectomy was performed. The facet joint and the LF were grossly normal. 1.5 cm sized cystic mass was noted to compress the nerve root and has no connection to the facet joint capsule (Fig. 1D). The cyst was filled with thick yellowish fluid with dark brown colored components suggesting old hemorrhage. The cyst was aspirated and shrunken. The origin of the cyst was traced to the midline of the PLL under the surgical microscope. Herniated disc material and annular defect just above the origin of the cyst were identified (Fig. 1E). The origin of the cyst was completely excised from the beneath the thecal sac without complication. Histological examination of the resected material showed a wall of fibrous connective tissue with no lining epithelium. The thick walls had some fibroblasts and mild myxoid stromal changes (Fig. 1F). The radiating pain alleviated immediately after surgery without neurological deficit.

Case 2

A 39-year-old man visited a clinic with spontaneously developed acute leg pain 3 days prior to admission. He denied the history of low back pain. He was a cook working at a ski resort and physically active. Neurological examination demonstrated severe paresthesia on the right L5 dermatome and slight weakness of extensor hallucis longus was noted. The straight leg raising test was limited to lesser degree than 30 in the right leg. MRI demonstrated about $2 \times 1.8 \times 1.2$ cm sized round mass with broad base to the ligamentum flavum at the L5 body level (Fig. 2A-C). The cyst has high signal on T1-weighted images and heterogeneously mixed signal on T2-weighted images without contrast enhancement. The mass severely compressed the right L5 nerve root. Based on these findings, provisional diagnosis was juxtafacet cyst complicated with acute hemorrhage. A partial hemi-laminectomy was performed. The yellow ligament seemed severely degenerated. The ligament was loose and dark (Fig. 2D). A large cyst was noted to compress the L5 nerve root. The cyst was adherent to the dura but dissection was easily achieved. There was no obvious attachment to the adjacent facet joints. The cyst was filled with dark reddish hematoma that was not aspirated through the 18 gauge needle (Fig. 2E). The cyst was cleaved with a surgical blade and the hematoma was aspirated. The cyst was originated from the LF around the foramen. Photomicrograph showed an irregular thick-walled cystic space without a synovial lining. Old hemorrhagic areas were noted in the fibrotic stroma (Fig. 2F). The postoperative course was uneventful.

DISCUSSION

Juxtafacet cysts including the synovial cyst and ganglion cyst are uncommon cause of low back pain or leg pain^{3,8,11,17,13,20}. The synovial cyst is the most common cystic lesion in the lumbar spine and the ganglion cyst has been rarely reported. For the reason, main understanding about the clinical characteristics of juxtafacet cysts has been acquired through the investigation about the synovial cysts^{10,20}. Recently, more cases of ganglion cysts have been reported in detail under the histopathological evidence.

Common clinical manifestations of the intraspinal cysts are usually resulted from root compression or cauda equina syndrome. Radiologically, a typical cysts appears with low-intensity signal in T1-weighted images and a high-intensity signal in T2-weighted sequences^{10,20}. However, the signal can vary depending on the proteinaceous concentration of the fluid or even the presence of blood like in our second patient. Acute herniation of the disc can be associated with epidural hemorrhage, and the hemorrhage must be differentiated from the cystic lesion like in our first patient.

The differentiation between two types of the cysts is only of histological value, as they share clinical and radiological characteristics. Synovial cysts are thought to arise from peri-articular tissue such as synovial herniation through weakened or destroyed facet joint capsule with continued direct communication with the joint¹⁸. Synovial cysts have a pseudo-stratified columnar synovial cell lining and are supported by layers of villous nodular hyperplastic vascularized connective tissue¹⁸. On the other hands, typical pathological findings of the ganglion cyst are membranous and fibrocollagenous tissue with myxoid degeneration. There were also some degenerative clefts representing chronic granular formation. Necrotic tissues, histiocyte infiltration, hemosiderin deposits, and neovascularization were also present in the wall. There should not be a true synovial lining^{1,10,16,19}.

The involvement of the facet joints associated with intraspinal cysts may influence on the treatment of strategy. Ganglion cysts develop from mucinous degeneration of ligamentous tissue in the mobile spine and they have no direct communication with the facet joint. However, synovial cysts arise from laxity of synovium of the facet joints. Therefore, they have high correlation with segmental instability. Regarding to the ganglion cysts, to our knowledge, there is no report in patients with spinal instability.

Though the etiology of the synovial cyst remains controversial, segmental instability and trauma play a major role in the cyst formation^{3,8,14,17,20}. Synovial cysts are located most commonly at the L4-L5 level, the most mobile level, and common at the L3-L4 level and L5-S1 level in ord-

er^{3,13,17,20}. When they do appear in the cervical spine, it arise from the most mobile segments, C5-C6 or C7-T1, the level where a mobile segment is attached to a non-mobile^{3,5}. In addition, factors influencing the segmental instability such as disc degeneration, facet joint osteoarthritis, and sagittal orientation of facet joint are commonly identified in patients with synovial cyst at the level^{8,9,20}.

Lumbar intraspinal ganglion cysts are also most commonly identified at the L4-5 level^{4,7,12,19}. They are suggested to arise from the degeneration of the ligamentous structure in the mobile segments. We presented intraoperative photographs that imply ligamentous degeneration of the PLL and LF. Ganglion cysts that originate from the posterior longitudinal ligament are very rare¹⁶. According to Marshman's summary of ganglion cysts from posterior longitudinal ligament, there are only twelve reports with 25 cases of lumbar PLL/LF ganglion cysts until 2005¹⁹. Our first patient suggests that degenerated PLL is susceptible to tearing and herniation of the disc. To our knowledge, this is the first report of the ganglion cyst associated with ruptured disc material at the same location. The second patient demonstrated clear evidences of severely degenerated dark and sticky LF. Acute hemorrhagic ganglion cysts have been extremely rarely reported¹⁰. Our two patients were young and enjoyed playing soccer or skiing. Those activities may induce a trivial injury to the lower lumbar spine. There was no degenerative change noted in the zygapophyseal joints on the preoperative MRI. They did not have segmental instability. We hypothesize that minor traumatic lesion of the ligamentous structure in the mobile spine in young patients could be an important risk factor of the ganglion cyst.

In terms of surgical strategy, instrumentation has been frequently required for the synovial cysts because of wide excision of the facet joint and preexisting instability^{14,18}. However, the ganglion cysts have no correlation with segmental instability and connection to facet joints. Therefore, simple excision of the ganglion cyst could be successful through minimal access^{1,10,15,19}.

CONCLUSION

We presented two cases of lumbar intraspinal ganglion cyst showing clear evidences that the cysts originate from degenerated ligamentous structures in young patients. The patients could be successfully treated through the minimal access.

References

1. Asamoto S, Jimbo H, Fukui Y, Doi H, Sakagawa H, Ida M, et al. : Cyst of the ligamentum flavum-case report. *Neurol Med Chir (Tokyo)* 45 : 653-656, 2005
2. Baba H, Furusawa N, Maezawa Y, Uchida K, Kokubo Y, Imura S, et al. : Ganglion cyst of the posterior longitudinal ligament causing lumbar radiculopathy: case report. *Spinal Cord* 35 : 632-635, 1997
3. Boviatis EJ, Staurinou LC, Kouyialis AT, Gavra MM, Stavrinou PC, Themistokleous M, et al. : Spinal synovial cysts : pathogenesis, diagnosis and surgical treatment in a series of seven cases and literature review. *Eur Spine J* 17 : 831-837, 2008
4. Brish A, Payan HM : Lumbar intraspinal extradural ganglion cyst. *J Neurol Neurosurg Psychiatry* 35 : 771-775, 1972
5. Cheng YY, Chen CC, Yang MS, Hung HC, Lee SK : Intraspinal extradural ganglion cyst of the cervical spine. *J Formos Med Assoc* 103 : 230-233, 2004
6. Christophis P, Asamoto S, Kuchelmeister K, Schachenmayr W : "Juxtafacet cysts", a misleading name for cystic formations of mibile spine (CYFMOS). *Eur Spine J* 16 : 1499-1505, 2007
7. Dagain A, Dulou R, Dutertre G, Delmas JM, Pouit B, de Soultrait F, et al. : Surgical management of synovial cyst of the lumbar spine : retrospective study of 52 patients. *Rev Chir Orthop Reparatrice Appar Mot* 94 : 289-296, 2008
8. Doyle AJ, Merrilees M : Synovial cysts of the lumbar facet joints in a symptomatic population : prevalence on magnetic resonance imaging. *Spine* 29 : 874-878, 2004
9. Fujiwara A, Tamai K, An HS, Kurihashi T, Lim TH, Yoshida H, et al. : The relationship between disc degeneration, facet joint osteoarthritis, and stability of the degenerative lumbar spine. *J Spinal Disord* 13 : 444-450, 2000
10. Gazzeri R, Canova A, Fiore C, Galarza M, Neroni M, Giordano M : Acute hemorrhagic cyst of the ligamentum flavum. *J Spinal Disord Tech* 20 : 536-538, 2007
11. Hsu KY, Zucherman JF, Shea WJ, Jeffrey RA : Lumbar intraspinal synovial and ganglion cysts (facet cysts). Ten-year experience in evaluation and treatment. *Spine* 20 : 80-89, 1995
12. Kao CC, Uihlein A, Bickel WH, Soule EH : Lumbar intraspinal extradural ganglion cyst. *J Neurosurg* 29 : 168-172, 1968
13. Khan AM, Synnot K, Cammisa FP, Girardi FP : Lumbar synovial cysts of the spine : an evaluation of surgical outcome. *J Spinal Disorder Tech* 18 : 127-131, 2005
14. Kjerulf TD, Terry DW Jr, Boubelik RJ : Lumbar synovial or ganglion cysts. *Neurosurgery* 19 : 415-420, 1986
15. Kornberg M : Nerve root compression by a ganglion cyst of the lumbar anulus fibrosus. A case report. *Spine* 20 : 1633-1635, 1995
16. Lee Joe, Wisneski RJ, Lutz GE : A ganglion cyst causing lumbar radiculopathy in a baseball pitcher : a case report. *Arch Phys Med Rehabil* 81 : 837-839, 2000
17. Lyons MK, Atkinson JL, Wharen RE, Deen HG, Zimmerman RS, Lemens SM : Surgical evaluation and management of lumbar synovial cysts : the Mayo Clinic experience. *J Neurosurg* 93 (1 Suppl) : 53-57, 2000
18. Marion PJ, Kahanovitz N : Lumbar-sacral radiculopathy secondary to intraspinal synovial cyst. *Arch Phys Med Rehabil* 76 : 1011-1013, 1995
19. Marshman LA, Benjamin JC, David KM, King A, Chawda SJ : "Disc cysts" and "Posterior longitudinal ligament ganglion cysts" : synonymous entities? Report of three cases and literature review. *Neurosurgery* 57 : E818, 2005
20. Métellus P, Fuentes S, Adetchessi T, Levrier O, Flores-Parra I, Taliano D, et al. : Retrospective study of 77 patients harbouring lumbar synovial cysts : functional and neurological outcome. *Acta Neurochir (Wien)* 148 : 47-54; discussion 54, 2006
21. Sachdev VP, Savitz MH, Hindi AI, Goldstein HB : Synovial cysts of the lumbar facet joint. *Mt Sinai J Med* 58 : 125-128, 1991
22. Tabaddor K, Sachs D, Llana JF, Testaiuti MA : Ganglion cyst of the odontoid process. Case report and review of the literatures. *Spine* 21 : 2019-2022, 1996