

Case Report

Lumbar discal cyst causing bilateral radiculopathy

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Abstract

Background: Discal cyst is a rare lesion that can result in clinical symptoms typical of disc herniation manifesting as a unilateral single nerve root lesion. To the best of the authors' knowledge, this is the first reported case of discal cyst resulting in bilateral radiculopathy.

Case Description: A 48-year-old female presented with bilateral sciatica and neurogenic claudication for 3 months. Magnetic resonance imaging revealed an extradural cystic lesion compressing the ventral aspect of the thecal sac at the level of the L3-L4 intervertebral disc. The lesion showed low and high signal intensities on T1- and T2-weighted images, respectively. Total excision of the cyst was achieved after a left hemipartial laminectomy of L3, and an obvious communication with the disc space was found. Bilateral sciatica was immediately resolved after surgery, and was sustained at the two-year follow-up. The histological diagnosis was consistent with a discal cyst.

Conclusions: Although a discal cyst is extremely rare, the possibility of a discal cyst should be considered in differential diagnosis of patients with radiculopathy, particularly when encountering any extradural mass lesion ventral to the thecal sac. Surgical resection is the most employed therapeutic method for symptomatic lumbar discal cysts.

Key Words: Bilateral, discal cyst, lumbar spinal stenosis, radiculopathy

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INTRODUCTION

Low back and sciatic pain is commonly caused by degenerative conditions such as lumbar disc herniation or spinal stenosis. The discal cyst, which has distinct connection to the corresponding intervertebral disc in

the spinal canal, is a less common etiology of a lumbar radiculopathy.^[2,7,14] We recently encountered a case of discal cyst in which clinical and imaging features differed from those of previous reports. A brief review of previously reported discal cysts in the medical literature is also presented.

CASE REPORT

A 48-year-old female presented with low back pain radiating to both buttocks and the posterior thigh for 3 months. She also suffered from neurogenic intermittent claudication (NIC) within 10 minutes. The straight leg-raising test was positive at 60 degrees on both sides. However, sensory or motor of the lower extremity was normal.

Magnetic resonance imaging (MRI) demonstrated a cystic lesion measuring $6 \times 16 \times 16$ mm with low-signal intensity on T1-weighted imaging and high-signal intensity on T2-weighted imaging at the level of the L3-L4 disc. The cystic mass, located in the ventral aspect of the extradural space, displaced the thecal sac dorsally [Figure 1a and b]. Rim enhancement of the lesion was appreciated after administration of gadolinium [Figure 1c]. The L3-L4 intervertebral disc appeared to

have mild degeneration. After L3-L4 laminotomy, a tense dark blue-colored cystic lesion compressing the entire thecal sac was encountered. The cyst was found to be mildly adherent to both the thecal sac and the posterior longitudinal ligament (PLL). The bloody serous fluid was aspirated from the cyst. The cyst was traced back to its stalk, which communicated with the L3-L4 disc by means of a central annular tear. The cyst and stalk were excised completely at the base of the connection. Coagulation of the PLL surrounding a round defect was performed; however, the disc space was not entered. The patient achieved complete pain relief and was allowed to walk on the day after surgery. Histopathological examination of the cyst revealed thick fibrous connective tissue interspersed with areas of chronic inflammation. However, there was no evidence of specific lining cell layers or disc material [Figure 2]. MRI obtained 14 months after surgery showed no evidence of recurrence or progression of disc degeneration [Figure 3]. The patient



Figure 1: Sagittal (a) and axial (b) T2-weighted MRI demonstrating a cystic lesion at the level of L3-4 intervertebral disc. In sagittal (c) T1-weighted MRI, rim enhancement of the lesion was appreciated after gadolinium administration

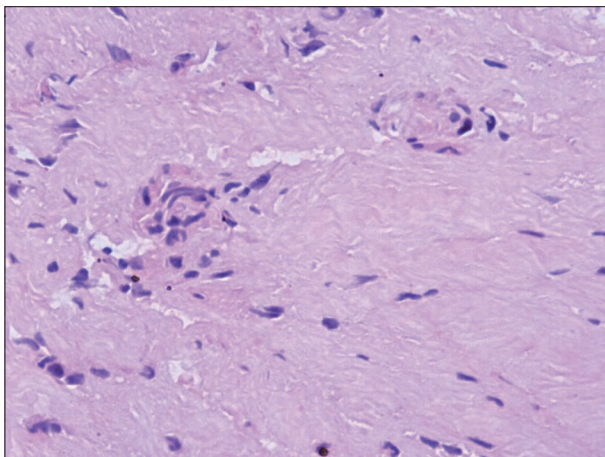


Figure 2: Histopathological examination of the cyst wall revealing fibrous connective tissue without lining cell (H and E, $\times 200$)



Figure 3: T2-weighted sagittal MRI images at 14-month follow-up after surgery. No recurrent cyst and no evidence of progression of degenerative change in the L3-4 disc were observed

has remained asymptomatic during a two-year follow-up period.

DISCUSSION

Discal cyst is extremely rare. To the best of our knowledge, only 57 cases of discal cyst including our case, have been reported in the literature.^[1-9,11-13,15-17] Fifty-one patients (89.5%) were males and 6 (10.5%) were females, and ranging age from 13 to 73 years (mean 33.8, median 31.1). The most common cyst locations were L4-L5 in 21 patients (36.8%), followed by L5-S1 in 16 patients (28.1%), L3-L4 in 12 patients (21.1%), L2-L3 in 5 patients (8.8%) and L1-L2 in 3 patients (5.2%). Clinical symptoms of discal cyst are indistinguishable from those of a typical disc herniation manifesting as a unilateral lumbar radiculopathy.^[2,12] Patients may suffer from NIC if the cyst becomes large enough to significantly compromise the diameter of the spinal canal. The present case is the only patient who presented with bilateral radiating pain and NIC.

Pathogenesis of discal cysts remains unknown. Two hypotheses for the development of discal cysts have been proposed. Chiba *et al*,^[2] hypothesized that an epidural hematoma is initially formed from hemorrhage of the epidural venous plexus that occurs in the space between the peridural membrane and the vertebral body, and discal cysts form most likely as a consequence of impairment of hematoma resorption. However, this theory does not explain the communicating stalk between the intervertebral disc and the cyst. Kono *et al*,^[12] proposed focal degeneration of an intervertebral disc with fluid production, similar to formation of meniscal cysts in the knee. Histologic findings from the cyst wall in a previous series and in our case demonstrated fibrous connective tissue without synovial lining cells, which supports this hypothesis.

MRI is the modality of choice for diagnosis of discal cyst. The cyst is round or oval in shape, with a low-intensity signal in T1-weighted images and a high-intensity signal in T2-weighted sequences that is consistent with a cyst containing liquid. This signal can vary depending on the proteinaceous concentration of fluid, or even the presence of blood. The peripheral rim of the cyst is enhanced on contrast-enhanced MRI.^[2,11,13] The cyst is a ventrolateral extradural lesion attached to a lumbar disc, and occasional extension into the lateral recess. In most reported cases, MRI has also revealed minimal degeneration of the involved disc. A connecting channel between the cyst and corresponding intervertebral discs can be demonstrated by discography and CT discography. On discography, contrast medium rapidly flowed into the cyst through a thin channel from the disc cavity, and severe radiating pain was simultaneously reproduced in the affected leg.^[11] Although discography can definitively

diagnose the discal cyst, we did not perform it due to MRI findings showing a high index of suspicion of discal cyst. Moreover, intraoperative findings of the apparent connection between the corresponding disc and the cyst also make it possible to differentiate discal cysts from other cysts.^[6,11,15]

Therapeutic guidelines have not been established. Spontaneous regression of the cyst has been reported,^[3,5] and intracystic steroid injections with successful resolution have also been attempted.^[11] However, surgical excision of the cyst has been employed in the majority of symptomatic discal cysts, and is highly effective for pain relief.^[2,6,13,15] An additional discectomy along with the associated cyst might depend on the rate of corresponding disc degeneration. Several authors emphasized the potential benefits to computed tomography or fluoroscopic-guided percutaneous aspiration of discal cyst.^[4,8,11] Percutaneous aspiration could be another initial option for discal cysts due to potential advantages including faster recovery, low complication rates, and avoidance of general anesthesia. In patients who failed initial percutaneous trial or recurred cases, surgical excision could be performed subsequently. Because of limited results of percutaneous aspiration by a small sample size and short-term follow-up period, careful analysis and follow up with additional cases are required for establishment of a proper therapeutic strategy.

Kono *et al*,^[12] have suggested that the discal cyst could not develop in the midline because the PLL prevents the cyst from developing dorsally to it. In almost all reported cases, cysts were located between the midline of the posterior vertebral bodies and the pedicles. However, in three cases, including ours, cysts that crossed the midline were reported.^[7,10] A median septum, connects ventrally with the thickened periosteum and dorsally with the PLL. However, it is not present in the intervertebral disc space in which the PLL is strongly attached to the annulus fibrosus. Considering the discal cyst crossing the midline, we suggest two possible pathways for growth of discal cysts: 1) when discal cysts arise from the area covered by peridural membrane only, they may grow between the midline of the posterior vertebral bodies and the pedicles, and spread to the lateral recess, causing unilateral root symptoms and 2) when discal cysts originate from the area covered by the PLL, perforation of annulus fibrosus, peridural membrane, deep PLL, and superficial PLL are required for bilateral growth of a midline crossing discal cyst. Under these circumstances, bilateral radiculopathy and NIC are likely to occur.

In summary, we present a rare case of a lumbar discal cyst with bilateral radiculopathy. Our case suggests that a discal cyst may cross the midline, resulting in bilateral radiculopathy and NIC. Surgical excision of the cyst offered immediate symptomatic improvement, which was sustained at the two-year follow-up.

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Commentary

Lumbar discal cyst causing bilateral radiculopathy

Discal cyst is certainly a rare pathology. In addition to the two origins of discal cysts noted by the authors,^[4] two additional possible origins have been proposed. Kobayashi *et al*, have proposed that one origin is the incomplete absorption process of a herniated disc,^[3] while Gadan *et al*, have proposed a traumatic process to the posterior longitudinal ligament, noting that their ten cases were found in young athletic subjects and that the microscopic findings were compatible with such a diagnosis.^[1] Differentiating between the origin of a discal cyst seems important in determining the best surgical approach and when to leave the disc space alone in the process, as was done by the authors in the current case presentation.

Also, from the standpoint of surgical technique, Kim and Lee,^[2] in their paper regarding 14 cases, have noted the value of CO₂ laser-assisted ablation of the discal cyst.

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Commentary

Lumbar discal cyst causing bilateral radiculopathy

A review of case report: Lumbar discal cyst causing bilateral radiculopathy

The authors present a case report concerning a 48-year-old female with bilateral sciatica and neurogenic claudication of 3 months duration secondary to an MR-

documented lumbar discal cyst at the L3–L4 level. Low signal T1 and high signal T2 MR studies documented the anatomy of the cyst which was subsequently adequately excised, without the disc space being entered, through a left hemipartial L3 laminectomy. Postoperatively, the

patient's symptoms totally resolved and she remained asymptomatic after two postoperative years.

When the authors reviewed the literature, they found references which included 57 discal cysts. Most patients were males, averaging 34 years of age, and had discal cysts predominantly at the L4–L5 and L5–S1 levels (64.9%). Symptoms typically included unilateral radiculopathy, unlike the present case which involved bilateral radiculopathy and neurogenic claudication. The authors quoted different hypotheses for the pathogenesis of discal cysts. Although the first hypothesis invoked epidural hematoma as the cause of the cyst, it did not explain why/how the “communicating stalk” developed between the disc and the cyst. The second, more likely hypothesis, offered focal degeneration of the disc with resultant “fluid production”; anatomical data demonstrating a fibrous cyst without a synovial lining supported this hypothesis.

However, the literature offers additional hypotheses regarding the pathoanatomical evolution of discal cysts, while also providing multiple differential diagnoses. In a study by Kobayashi *et al.*, discal cysts communicated with an adjacent disc herniation on both MR and CT studies; this finding was confirmed intraoperatively.^[7] The lesion located within the epidural compartment, compressed a single nerve root. Following excision of the cyst, histology and electron microscopy revealed residual disc tissue within the cyst wall. Their hypothesis was that macrophages contributed to the formation of a cyst following resorption of herniated disc tissue; subsequent hemorrhage within the cyst wall was the source of the serous fluid that replaced the resorbing disc. In another study by Cho *et al.*, lumbar intraspinal epidural ganglion cysts contributed to low back/leg pain.^[2] In another study by the same authors, two male patients with unilateral radiculopathy attributed to “rounded” intraspinal cysts on MR (hypointense on T1 and hyperintense on T2) not associated with significant disc herniations, also had ganglion cysts.^[3] Both patients exhibited significant degeneration of the ligamentum flavum. Furthermore, the authors also acknowledged that both ganglion and synovial cysts can present as juxtafacet cysts, and that differentiating between them may be difficult.

Gas-filled intradural cysts may also contribute to unilateral radiculopathy.^[8] In one case study, a 67-year-old female with marked right thigh pain had an enhanced MR scan which documented a cystic, gas-containing lesion within the spinal canal at the L2–L3 level. The CT-discogram documented contrast filling the cerebrospinal fluid pathways around but not penetrating the cyst. Removal of the cyst was accomplished by dissecting away the firmly adherent nerve root (from the capsule), defining the margin between the cyst and dura, and ultimately by removing the cyst and its discal contents.

In yet another study, an intradural disc herniation

containing gas, which appeared as a ring-enhancing “cystic” intradural lesion at the L3–L4 level on a contrast-enhanced MR study, was surgically excised.^[1] A gas-filled intradural cyst contributing to lumbar radiculopathy was also seen in a further study that discussed the similarities of gas-containing pseudocysts, herniated intradiscal gas, and free gas within the epidural compartment.^[6]

What diagnostic studies best document discal cysts? Although CT-discography readily documents the connection between the disc space and the cyst *via* an attenuated channel, it poses multiple inherent risks which include durotomy, root injury, infection, allergic reactions, etc. Notably, several non-invasive modalities, including MR and CT-based studies, with/without intravenous contrast, would likely adequately confirm the location and etiology of many of these lesions.

Although there are multiple therapeutic options offered to manage discal cysts, open surgical procedures, and not the minimally invasive ones, offer the most versatile therapeutic alternatives. Certainly, non-surgical treatment in minimally and/or asymptomatic patients may be a reasonable alternative, particularly since some discal cysts will spontaneously resorb.

However, other minimally invasive approaches such as intracystic steroid injections, or fluoroscopic/CT-guided cyst aspiration, are likely to fail. Reasons for failure include first, their inability to address the more complex pathology (e.g. disc herniations, synovial cysts, etc.). Second, the thecal sac and/or nerve roots, located dorsal to the discal cyst (with/without disc herniations), may be perforated with these percutaneous techniques, resulting in cerebrospinal fluid fistula formation, and/or root trauma. Third, these techniques provide minimal access to potentially more complex pathology, and therefore increase the risk to the patient of incomplete, inadequate cyst decompression, and/or cyst recurrence. Although Dasenbrock *et al.* observed that an L5-S1 discal cyst was successfully aspirated under CT-guidance, they acknowledged that few surgeons were opting for this approach as another study cited cyst recurrence following a similar approach.^[4] Although three of four patients with epidural/intradiscal gas were successfully treated with percutaneous needle aspiration of the gas, this could have resulted in inadvertent durotomy and/or other unanticipated local neural trauma.^[5] The optimal operation for discal cysts should be individually tailored to the patient's specific needs. Furthermore, as open procedures offer more flexibility, visibility, and effectiveness, they should not be replaced by minimally invasive alternatives simply because of the smaller size of the incision. It is critical here, that “marketing” of the minimally invasive techniques does not take the place of good judgment and safer surgery.

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Commentary

Discal cysts: Rare causes of nerve root compression

A 48-year-old Asian female presented with a 3-month history of bilateral lumbar radiculopathy. An LS spine magnetic resonance imaging (MRI) revealed an L3-4 discal cyst (hyperintense on T2-weighted sequences and hypointense on T1-weighted images with a slight rim enhancement) crossing the midline and causing bilateral nerve root compression. The patient underwent hemilaminectomy for cyst resection, and aspiration of bloody serous fluid. The cyst was traced back to its discal fistulous attachment where an annular tear was discovered and coagulated. The disk space itself was not violated. Clinical and radiographic follow-up, at 12 and 14 months, respectively, revealed excellent symptomatic and cyst resolution.

Degenerative conditions of the lumbar spine are common causes of radiculopathy. A number of rare conditions exist that may present identically; discal cysts, intraspinal extradural cysts that communicate with the intervertebral disc, are a rare cause of radiculopathy. First reported in English in 1999,^[7] only less than 100 patients with discal cysts have been published until now, primarily as case reports and small case series.^[1] This is the first reported case of a discal cyst causing bilateral symptoms.

Discal cysts are seen most commonly in young Asian men.^[1] The fact that discal cysts occur at more rostral levels than disc herniations (the L3-4 level in this case), can present in the absence of additional spinal pathology and most commonly affect young males, may suggest a pathophysiology distinct from that of degenerative conditions.

The etiology of discal cysts is debatable. Some suggestion underlying traumatic epidural hematoma with deficits in its resorption leading to cyst formation.^[2] Others believe that focal degeneration of the intervertebral disc leads to leakage of fluid into the spinal canal prompting a marked

inflammatory reaction, causing the formation of a pseudomembrane around the fluid.^[6] Finally, Marshman et al. have argued that discal cysts are synonymous with posterior longitudinal ligament/annulus fibrosus ganglion cysts, likely degenerative in origin.^[8]

MRI is the diagnostic study of choice, typically revealing an extradural intraspinal ventrally located cyst, hypointense on T1-weighted and hyperintense on T2-weighted images, with rim enhancement after gadolinium administration (as seen in this case). However, hemorrhagic cysts may be hyperintense on both T1- and T2-weighted images, and the rim of contrast enhancement is not always present. Discography, albeit invasive, is used diagnostically to confirm a connection between the intervertebral disc and the cyst.

The natural history of this condition has yet to be defined, providing little guidance on prognosis. Spontaneous regression of a discal cyst has been reported,^[4] as well as regression after intradiscal and epidural steroid injections.^[5] Although most reported cases have failed a trial conservative therapy, it is not clear if this represents a publication bias or if discal cysts are less responsive to conservative therapy. Nevertheless, an initial trial of conservative therapy should be pursued in neurologically intact patients.

Treatment options exist, though surgical resection is the most commonly employed modality. Successful CT-guided percutaneous aspiration of discal cysts has been reported.^[3] Its proponents point to its many potential advantages: decreased infection rates, avoidance of general anesthesia, and faster recovery. They argue that surgical excision should be reserved for patients who fail percutaneous drainage attempts. However, surgical excision remains the most commonly performed therapeutic modality used for symptomatic discal cysts, with complete irradiation

of the fistulous connection between the disk and the cyst in order to avoid recurrence.

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