Case report

Percutaneous Ligamentum flavum cyst rupture by interlaminar Epidural approach - A case report and literature review

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Abstract.
A ligamentum flavum cyst (LFC) is defined as a cystic formation arising from the ligamentum flavum. The majority of symptomatic cysts usually present with lower leg radiating pain. An 84-year-old man presented with left lower leg radiating pain caused by a ligamentum flavum cyst. Magnetic resonance imaging demonstrated a hyperintense extradural mass with a peripheral hypointense rim on T2-weighted images at the L4/5 level. Percutaneous cystic rupture by interlaminar epidural injection was performed. The patient came to the clinic four weeks, 12 weeks, and one year after the procedure and was symptom-free at the last visit. In conclusion, LFC can be treated by the interlaminar epidural approach and this procedure could be suggested as a good option for treating an LFC.

Keywords: Ligamentum flavum cysts; Epidural injections; Low back pain; Minimal invasive procedures; Ages 80 and over.

INTRODUCTION
A ligamentum flavum cyst (LFC) was first reported by Moiel et al. in 1967 [1]. It represents a rare cause of lumbar nerve root compression or spinal stenosis. LFC is a juxtaposition facet cyst (JFC), which was reported in 1974 to describe synovial and ganglion cysts that are adjacent to the facet joints [2]. A previous study evaluated cystic origins intraoperatively in 58 cases and suggested that JFCs may originate from three anatomic structures and present as facet cysts, flavum cysts, and posterior longitudinal ligament (PLL) cysts [3]. The cyst causes back pain and lower leg pain. The treatment of choice is surgical intervention, and conservative treatment is also one of the treatments. Facet joint cysts (FJC)s can be ruptured with a conventional facet joint block, but it does not seem effective to apply the conventional approach to rupture LFCs due to differences in their anatomical origin. In this report, we describe a patient with radiculopathy pain caused by an LFC located at the L4–5 level. We attempted to treat the LFC with an interlaminar approach rather than the conventional method and successfully ruptured the LFC. The written informed consent statement was obtained for this report.

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CASE REPORT
An 84-year-old male patient presented with left lower leg pain. The pain was radiating along the lateral side from the left buttock to the dorsum of the foot, involving mostly the L5 root dermatome without low back pain. The patient had pain with an intensity of 8/10 on the visual analog scale (VAS) and had been having insomnia due to pain. The straight leg raising test (SLRT) was positive. The motor grade was G5/G5, and sensory remained intact. There was no tenderness around the lumbar area, such as the facet joints or paraspinal muscles, and he had no problems with walking for more than one hour by himself. The patient had been diagnosed with L4/5 spinal stenosis a year earlier. He had been taking 300 mg of gabapentin three times a day, and the symptoms slightly improved after an epidural steroid injection at another hospital. However, the pain aggravated again three months ago, and percutaneous epidural neuroplasty using a catheter was performed. He complained that previous procedures had little effect and the uncontrolled pain worsened, so he visited the pain clinic by way of the emergency room of the hospital.

There was no significant difference between the current X-ray images and previous images, which demonstrated grade I dynamic instability with degenerative spondylolisthesis findings in the flexion and extension views. Magnetic resonance imaging (MRI) was performed for further evaluation. Cysts were observed in two places at the L4–5 level on the MRI images. A cyst was observed near the ligamentum flavum and another was seen near the facet joint with sizes of about 8 × 9 mm and 4 × 5 mm, respectively (Fig. 1). There was no cord compression by the vertebral disc and there were no other abnormal MRI findings that may have caused the patient’s symptoms. The pain could be thought of as radiculopathy by LFC compressing the root. Because the cyst membrane was thick, and the cyst was located within the ligamentum flavum, it seemed difficult to access the cyst using a conventional approach. Therefore, we decided to induce cyst rupture mechanically through the epidural space and calculated the needle trajectory in the MRI image for advancing the needle to the LFC (Fig. 1C, D).

The patient was prepared for the procedure in a prone position, and sterile drapes were placed. First, a conventional facet joint block was performed on the L4/5 facet. A 1% lidocaine solution (3 mL) was used as the local anesthetic. Under C-arm guidance, a 22-gauge epidural...
Figure 1: Two cysts (white arrow) are located at the L4–5 level. Axial (A) and sagittal (B) views in T2-weighted magnetic resonance imaging revealed a left-sided 8 × 9 mm-sized hyperintense cyst. (C, D) The angle and distance for needle trajectory (yellow arrow) were calculated.

Figure 2: Interlaminar approach for ligamentum flavum cyst rupture in the (A) anteroposterior view and (B) the lateral view. (A) Contrast media was scattered around the skin in an interspinous area (dark, round shadow) due to high pressure. (B) The tip of the needle is located in the epidural space. (C) Lateral view of the conventional approach for juxtafacet cyst rupture after ligamentum flavum cyst rupture. No communication was observed between the cyst and the facet joint.

Block needle was advanced toward the facet joint until the needle penetrated the facet joint capsule. After confirming that the needle was in the proper position within the joint with contrast media, the mixture was gently injected with weak pressure to try to rupture the JFC. Pressure was heavily applied when the mixture was injected. As we expected before the procedure, the cyst was not ruptured mechanically by this conventional approach. Then, we tried to get the needle to advance toward the LFC using an interlaminar approach according to the needle trajectory calculated from the MRI image. Since the cyst was almost adjacent to the ligamentum flavum, we thought the needle might reach the cyst as soon as the needle penetrated the ligament and expected that it could be different from what we generally feel as loss of resistance (LOR). When the needle approached the ligamentum flavum, the C-arm was turned laterally to slowly progress the needle. We checked whether there was LOR, as well as the feeling of piercing the ligament when the needle tip was near the spinolaminar line. After positioning a needle in an appropriate position beyond the ligamentum flavum using the LOR technique, we injected contrast media. However, the pressure was rather high, and the contrast media did not spread out through the epidural space. Therefore, we thought that the needle was located in the cyst. When pressure was applied to inject the mixture, the patient was stimulated by the pattern of radiating pain.
that he usually felt. After several attempts, the pressure was reduced and contrast media was spread out smoothly through the epidural space (Fig. 2A, B). We could expect that the LFC was ruptured at this moment. Normal saline (5 mL) flushing was performed and a mixture of 5 mg of dexamethasone and 1,500 U of hyaluronidase with 5 mL of 0.3% mepivacaine was injected. After the LFC was ruptured, we injected contrast media into the facet joint once again using the conventional approach. But as in the first trial, no reduction in pressure was observed and the contrast media did not spread into the epidural space (Fig. 2C). There was no communication between the LFC and the facet joint, indicating that it was difficult to rupture the LFC by the conventional approach. We could not confirm that the LFC was ruptured after two attempts, but we terminated the procedure because we no longer thought that it was associated with the patient’s symptoms. The patient complained of pain with VAS scores of 4/10 in the injection area after the procedure and was noted to have normal motor/sensory function. Also, there were no other complications. We took care of the patient in the recovery room for one hour to observe changes in symptoms, and the patient was discharged without neurologic complications. At an outpatient visit after four weeks, the patient reported mild pain in the left buttock with VAS scores of 2/10. The patient stated that the pain had been much improved without any radiation. In an outpatient visit after three months, the symptoms appeared to be completely resolved. The patient was still symptom-free at his one-year visit after the procedure and was open for the next follow-up.

DISCUSSION
In this case, we successfully ruptured an LFC by percutaneous interlaminar epidural injection, which was a relatively simple procedure. The LFC was located inside the ligamentum flavum away from the facet joint and was not accessible by the conventional approach. Moreover, since there was no communication between the LFC and the facet joint, it was impossible to rupture the LFC through the facet joint. The position of the LFC was checked using MRI images and used to calculate the needle approach trajectory in the interlaminar approach to rupture the cyst. We did not perform a follow-up MRI. But, as contrast media spread well to the epidural space and the patient’s symptoms improved, it is estimated that the cyst inside the ligamentum flavum was successfully ruptured. No symptoms recurred even at the outpatient clinic visit one year after the procedure.

The term “juxtafacet cyst” refers to all cysts occurring in the posterior elements of the spinal canal. Ligamentum flavum cysts are a subtype of juxtafacet cysts [4] and are recognized to be the rare cause of cord and nerve compressions [5]. PLL cysts and FJCIs are also types of JFCIs. FJCIs have two histological subtypes. Some authors distinguish synovial cysts and ganglion cysts by the presence or absence of a synovial lining membrane. Synovial cysts communicate with the facet joint and are surrounded by the inner wall of the synovial lining membrane, while a ganglion cyst is a pseudocyst surrounded by a fibrous wall [2]. Almost all cysts of the ligamentum flavum and PLL cysts are pseudocysts, known as ganglion cysts [6].

The incidence of FJCIs is thought to be less than 0.5% in the general symptomatic population [5]. FJCIs arise from a defect in the synovial membrane and are thought to be caused by a combination of the degeneration of the zygapophyseal joint and destabilizing changes in the spine [4]. However, LFCs are rare and occur frequently in the lower lumbar region. The cause of LFCs has not yet been fully identified, but it has been suggested that persistent stress on the ligamentum flavum may lead to mild chronic trauma such as ligamentum flavum and degenerative disc disease in patients [6]. Therefore, the distribution of LFCs correlates with mobility and the incidence of degenerative changes. LFCs in the lumbar spine commonly occur at the L4–5 level, followed by the L5–S1, L3–4, and L2–3 levels. Only a few cases have been reported in the cervical spine [7].

There are no specific clinical symptoms for LFCs. The majority of symptomatic cysts usually present with lower leg pain. Wildi et al. [8] reported that 97% of the patients complained of radicular pain, and 76% of the patients had low back pain. Thus, LFCs can be confused with symptoms caused by herniated nucleus pulposus. Neuroimaging, especially MRI, plays an important role in the diagnosis of JFCIs. Generally, the cysts have a variable signal on T1-weighted and hyperintense T2-weighted images. A hyperintense extradural mass with a peripheral hypointense rim on T2-weighted images was consistent with an LFC observed in the histopathologic examination [9]. In our patient, MRI T2-weighted images showed thickened ligamentum flavum containing the cyst with a high-intensity signal and a low-intensity signal on T1-weighted images. LFCs are adjacent to or located within the ligamentum flavum and do not communicate with the facet joint. The differential diagnosis of imaging studies between LFC and FJC is helpful for surgeons because FJCIs are more difficult to resect, requiring exploration of the facet joint [10]. The histological features of the LFC, with an absence of a synovial lining membrane and no communication with the facet joint, helped us decide how we should perform the procedures.

Some cases of the spontaneous resolution of cysts have been reported, but most cases require treatment. The treatments reported for LFC range from conservative management to surgical decompression with stabilization. Surgical decompression is the treatment of choice. Surgical decompression may be required for the patient who has significant symptoms or is refractory to conservative care. The goal of surgery in a patient with an LFC is spinal decompression through laminectomy and the resection of the affected ligamentum flavum and the cyst. If complete excision of the cyst is performed, a minimal recurrence rate is assured. Even in the surgical department, endoscopic resection has been attempted due to the risk and complications of open surgery [11]. Conservative treatment can be an option for patients who cannot receive surgical intervention due to old age or systemic diseases, or patients who refuse surgical intervention. Conservative treatment includes medication, physical therapy, needle aspiration, and mechanical cyst rupture. Among the treatment modalities, minimally invasive procedures can be considered for patients with a high risk for surgery. Many percutaneous methods have been reported recently such as computed tomography-guided treatment, and epiduroscopic removal. Bureau et al. [12] reported that percutaneous steroid injections were often effective and could result in complete regression of the cyst. Shuang et al. [13] conducted a meta-analysis including 29 studies and reported that percutaneous cyst resolution procedures had the potential to be an alternative to surgery. The procedure we performed is a much
simpler and more accessible method compared to surgical intervention. Not only does it take less time, but it has the advantages of being less expensive, and less invasive. This case is thought to be meaningful because few cases of conservative treatment for an LFC have been reported in the literature.

There were some limitations to this case report. First, we could not confirm whether the FJC was ruptured after two attempts. The LFC seemed to press the nerve root and the FJC was relatively small in the MRI image. Therefore, we thought that the patient's symptoms were mainly caused by the LFC, not the FJC. But given the possibility that FJC can cause symptoms, it needs to be treated using other methods such as epiduroscopic removal [14]. Second, no histological examination of these cysts was performed. Since the cyst was estimated to be LFC from the histological, anatomical, and radiologic characteristics, this case may further contribute to the treatment modality of LFCs if the cyst is histologically identified. Third, a follow-up MRI was not performed due to the economic issues of the patient. It would have helped to identify how the cysts changed after the procedure. Further studies are needed to evaluate the long-term outcomes and incidence of complications.

In conclusion, LFC can be treated by the interlaminar epidural approach and this procedure could be suggested as a good option for treating an LFC.

Funding: Not applicable

Conflicts of interest: The authors declare no competing interests.

Reference