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CASE REPORT

Ankle and toe weakness caused by calcified ligamentum flavum cyst: A case report

Ho-Young Jung, Geon-U Kim, Yong-Won Joh, Jun-Seok Lee

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Abstract

BACKGROUND

Ligamentum flavum cysts, which are most common in mobile junctional levels of the spine, can be a rare cause of spinal stenosis. There have been several case reports of ligamentum flavum cysts. However, there is yet to be a documented case report of a calcified ligamentum flavum cyst. Herein, we report the first case of a calcified ligamentum flavum cyst causing ankle and toe weakness.

CASE SUMMARY

A 66-year-old male visited our hospital complaining of claudication as well as thigh and calf pain in his left leg, all beginning two weeks prior. Physical examination revealed motor weakness of the left ankle dorsiflexion and great toe dorsiflexion. Lumbar spinal computed tomography scans showed spinal stenosis combined with a calcified mass at the left side of the L4-5 level. Magnetic resonance imaging showed dural sac compression caused by the calcified mass at the left ligamentum flavum of the L4-5 level. We performed decompressive laminectomy and excision of the calcified mass combined with posterior lumbar interbody fusion at the L4-5 level. Intra-operatively, we found a firm and nodule like mass originating from the ventral surface of ligamentum flavum. Pathological examination suggested a calcified pseudocyst without a capsular lining. After the operation, the patient's motor weakness in the ankle and great toe improved gradually.

CONCLUSION

The patient's ankle and great toe weakness were improved successfully after surgical removal of the calcified cyst.

Key Words: Ligamentum flavum; Calcified cyst; Spinal stenosis; Claudication; Motor weakness; Spine; Case report

Core Tip: Ligamentum flavum cyst in lumbar spine can be a rare cause of spinal stenosis. There have been several case reports of ligamentum flavum cysts. However, to the best of our knowledge, no calcified ligamentum flavum cyst has been reported. We report the first case of a calcified ligamentum flavum cyst causing neurological claudication and motor weakness of lower extremities. We believe that our report would be useful for a diagnostic approach and treatment in patients who have spinal stenosis with neurological claudication and motor weakness.

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INTRODUCTION

Ligamentum flavum cysts are a rare cause of spinal stenosis, which causes neurological claudication and pain in the lower extremities[1]. Ligamentum flavum cysts appear on the ventral surface of the ligament without any connections to articular facets. The precise pathogenic mechanism underlying the formation of ligamentum flavum cysts remains unclear. However, the development of these cysts has been suggested to be associated with persistent micro-instability at the motion segment and concomitant micro-trauma, which can increase the risk of cyst formation[2]. There have been several reports of ligamentum flavum cyst[3-8]. However, to our knowledge, there has yet to be a report of a calcified cyst of ligamentum flavum. Herein, we report the first case of a calcified ligamentum flavum cyst presenting ankle and toe weakness.

CASE PRESENTATION

Chief complaints

A 66-year-old male presented with claudication, thigh and calf pain and motor weakness in his left leg.

History of present illness

He complained of weakness in his left great toe and ankle beginning two weeks prior to his visit, and the complained that his symptoms had become gradually aggravated with time.

History of past illness

Hypertension as underlying disease.

Personal and family history

There was no specific family health history.

Physical examination

Motor weakness of the left ankle dorsiflexion (grade 2) and great toe dorsiflexion (grade 0 to 1).

Laboratory examinations

When the patient admitted to our hospital, laboratory investigations revealed low levels of hemoglobin (10.5 g/dL, normal range: 14.0-18.0) with low levels of hematocrit (30.7 %, normal range: 42.0-52.0). White blood cell count was 6.70 × 10^9/L [normal range: (4.0-10.0) × 10^9] and platelet count was 240 × 10^9/L [normal range: (150-450) × 10^9]. Coagulation function test: Prothrombin time (PT): 10.0 s (normal range: 9.7-13.3); PT%: 119% (normal range: 77-120); international normalized ratio: 0.90 (normal range: 0.88-1.20); activated partial thromboplastin time: 28.8 s (normal range: 23.1-37.3). These results were all within normal ranges.

Imaging examinations

Plain radiograph of the lumbar spine showed degenerative changes and spondylolisthesis L4 on L5 (Figure 1). Magnetic resonance imaging (MRI) showed protrusion disc and dural sac compression attributable to a mass at the L4-5 level (Figure 2). Lumbar spinal computed tomography (CT) scans showed spinal stenosis combined with dural sac compression caused by a calcified mass at the left side of the L4-5 level (Figure 3).

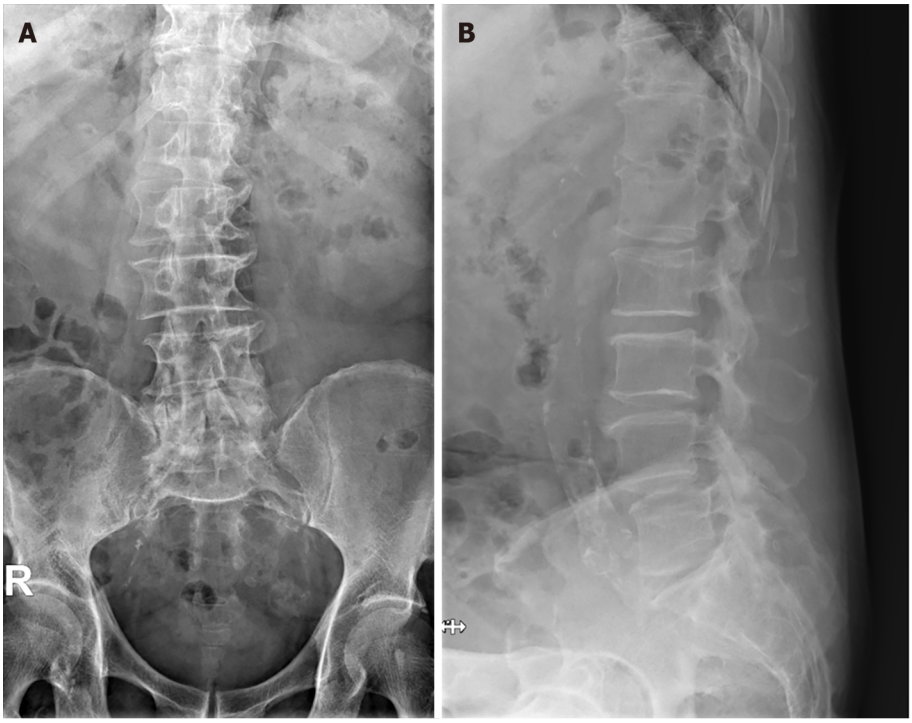

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Figure 1 Plain radiographs of the lumbar spine. A: Lumbar spine anteroposterior X-ray; B: Lumbar spine lateral flexion X-ray showing degenerative changes and spondylolisthesis L4 on L5.

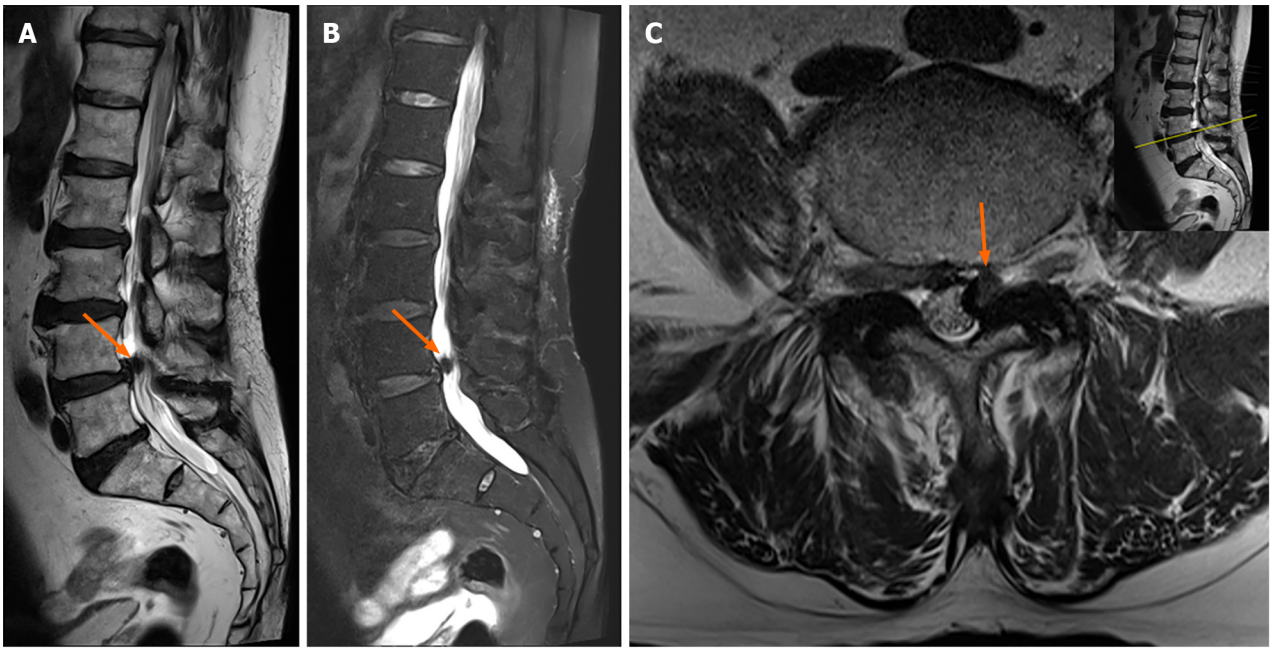

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Figure 2 Preoperative magnetic resonance imaging of the lumbar spine. A: Sagittal T2-weighted magnetic resonance imaging (MRI) showing a mass lesion with a hypointense signal at the L4-5 level (arrow); B: Sagittal fat-suppressed T2-weighted MRI showing a mass lesion with a hypointense signal at the L4-5 level (arrow); C: Axial T2-weighted MRI of the corresponding section showing left side dural sac compression by a mass (arrow).

FINAL DIAGNOSIS

Calcified pseudocyst of ligamentum flavum was confirmed by histological examination after surgery (Figures 4 and 5).

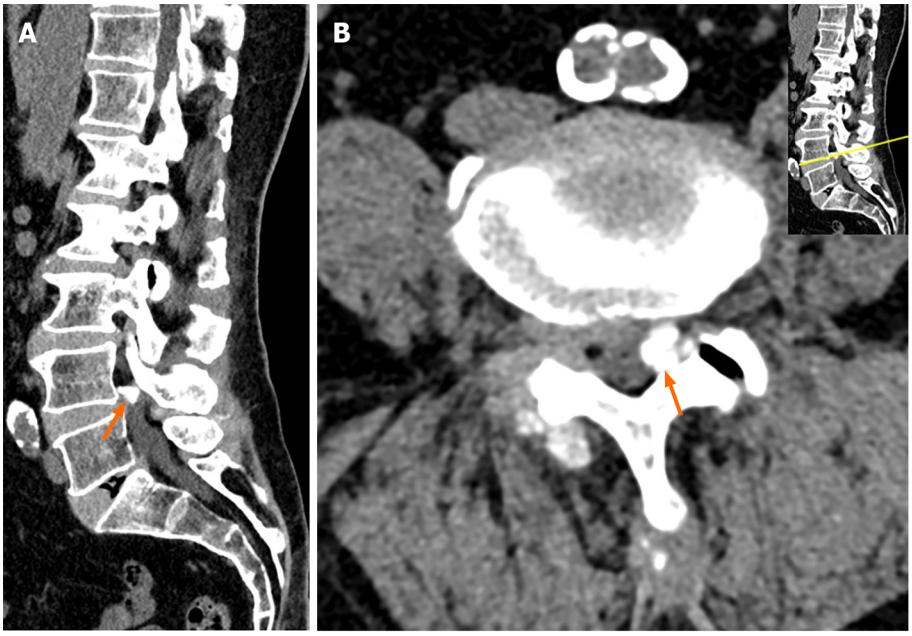

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Figure 3 Computed tomography scans of the lumbar spine. A: Sagittal computed tomography (CT) scan showing a calcified mass at the L4-5 level (arrow); B: Axial CT scan showing a calcified mass at the left side of the L4-5 level (arrow).

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Figure 4 Intraoperative clinical photographs of the excised calcified cyst. A firm, brown-colored, nodule-like mass originating from the ventral surface of the left side ligametum flavum was found. A: Ventral surface of the excised ligamentum flavum; B: Cross-section of the excised calcified cyst.

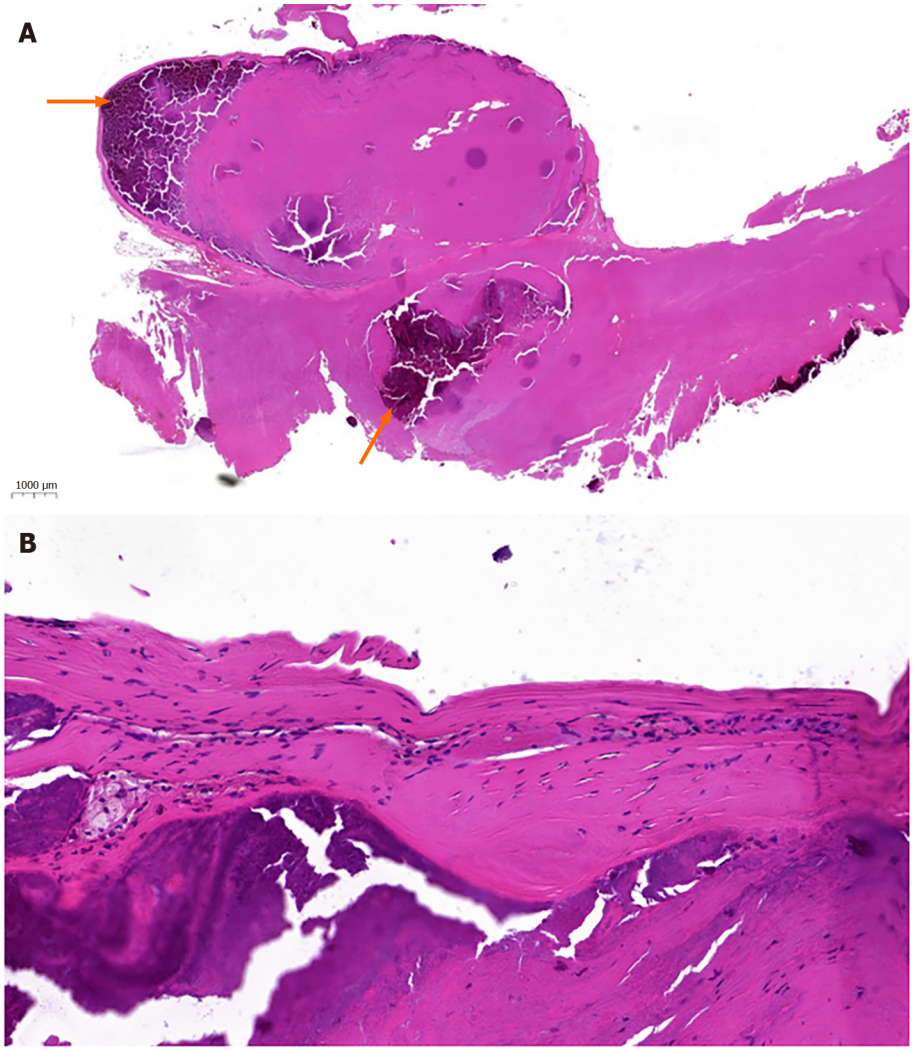

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Figure 5 Microscopic images of the excised calcified cyst. A: Low magnification (× 8) image showing a cyst in the ventral surface of ligamentum flavum with dark purple-colored calcified material in the cyst (arrow); B: Higher magnification (× 20) image showing no identifiable epithelial cell lining.

TREATMENT

Decompressive laminectomy and excision of the calcified mass for lumbar spinal stenosis, along with posterior lumbar interbody fusion at the L4-5 level for spondylolisthesis were performed.

OUTCOME AND FOLLOW-UP

Immediately after surgery, the pain in his left thigh and calf improved, but the motor weakness in his left ankle and great toe remained. On the fourth postoperative day, histological examination confirmed a calcified pseudocyst of ligamentum flavum (Figure 5). Two weeks after the operation, the motor weakness in the patient's ankle and great toe had improved gradually, and he was discharged from the hospital.

DISCUSSION

Ligamentum flavum cyst was first described by Moiel *et al*[8] in 1967, in a case where it compressed dural sac and presented as testicular pain and leg radiating pain. Ligamentum flavum cyst can be distinguished from facet joint cyst by its location on the ventral surface of the ligamentum flavum without any connections to articular facet[9,10]. The pathogenic mechanism of ligamentum flavum cyst has yet to be clearly identified. Segmental instability and micro-trauma have been described as major causes of the formation of ligamentum flavum cysts[9-11]. Segmental instability and micro-trauma at motion segment are believed to predispose one to cyst formation. Ligamentum flavum consists of elastic fiber with a reduced amount of collagen fibers. When lesions occur due to micro-trauma or segmental instability, the

regeneration of elastic fibers is extremely limited. This limited regeneration leads to a decrease in the elasticity of ligamentum flavum along with the deposition of collagen[12]. The accumulation of collagen in ligamentum flavum leads to progressive degeneration, and the myxoid changes can form cysts[13]. In our case, we found a segmental instability with grade I spondylolisthesis at the L4-5 level in our patient. It could also be considered a possible cause of cyst formation. However, the mechanism of calcification of ligamentum flavum cyst is unknown. It is presumed that calcification additionally occurred in the degenerative change of ligamentum flavum.

In general, ligamentum flavum cyst can be diagnosed using MRI. Ligamentum flavum cyst typically appears as hyperintense on T2-weighted images and as hypointense on T1-weighted images, where it appears as a cyst arising from the ventral surface of ligamentum flavum[6]. In our case, axial images of the L4-5 level showed left side dural sac compression by a mass that appears as hypointense on T2-weighted and T1-weighted images. Based on these MRI findings, we considered the mass to be a sequestrated disc material. However, lumbar CT scans revealed dural sac compression by the calcified mass at the left side of the L4-5 level. It was then surgically confirmed that the mass was a calcified cyst of ligamentum flavum cysts. In this way, performing CT scans along with MRI can aid one in making an accurate diagnosis.

Conservative treatment, such as that consisting of medication and percutaneous steroid injections, can be performed in ligamentum flavum cysts. However, these conservative therapies have not shown successful long-term results[4,14]. Surgical decompression with cyst excision is the treatment of choice when conservative treatment fails[4,14]. In this case, we decided to perform surgical removal of the calcified ligamentum flavum cyst due to the progression of motor weakness.

CONCLUSION

To our knowledge, the treatment reported herein is the first surgical treatment for calcified ligamentum flavum cyst causing ankle and toe weakness. We performed surgical decompression and excision of the calcified cyst. After the surgery, the patient's motor weakness gradually improved.

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FOOTNOTES

Author contributions: Lee JS conceived and designed the study, and performed the surgery; Jung HY, Kim GU, and Joh YW prepared the figures and collected the data; Jung HY and Lee JS wrote the manuscript; and all authors read and approved the final manuscript.

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