Report of a Rare Case: Ligamentum Flavum Cyst

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ABSTRACT
A rare case of ligamentum flavum cyst of the lumbar spine in an elderly male is reported. The patient presented with low backache and features of bilateral radiculopathy of a sudden onset. The cyst was lying in the extradural space. After surgery, the patient reported complete relief of symptoms.

Keywords: Etiology of lumbar canal stenosis, Extradural lesion, Ligamentum flavum cyst.


Source of support: Nil
Conflict of interest: None

BACKGROUND
Different etiologies for cystic lesions in the lumbar spinal canal have been reported in the literature, among them are hemorrhagic cysts, perineural cysts, dermoid cysts, and parasitic cysts. The most common lesion seems to originate from the facet joints: the synovial cyst, which represents a protrusion of the synovial membrane into the surrounding tissue. The literature remains imprecise about the histopathologic nature of cystic lesions in the lumbar region of the spine. Some authors differentiate between the terms 'synovial cyst' (with a synovial lining) and 'ganglion pseudocyst' (without any synovial lining). Others proposed the term 'juxtafacet cyst,' simply representing both. Also, evolution from a synovial cyst into a ganglion pseudocyst has been questioned. Ligamentum flavum pseudocyst, as a cystic lesion in the lumbar spine, has only rarely been mentioned.  

CASE PRESENTATION
A 68-year-old male presented to us with sudden onset of lower back pain with bilateral radiating pain to lower limb associated with difficulty in walking which was progressive in nature also complaining of weakness in both lower limbs. He was operated for cervical myelopathy in 1995. At present, there was no history of trauma or other constitutional symptoms.

Clinical examination revealed weakness (grade 3/5) of the both lower limbs ankle dorsiflexion and flexion and extension of the left great toe. Otherwise, motor examination of the other muscle groups was normal. Sensation of both lower limbs was intact. The knee reflexes were present and ankle reflexes were mute.

Plain radiographs of the lumbosacral spine showed degenerative changes. Blood parameters showed normal white cell count (WCC), erythrocyte sedimentation rate (ESR) and C-reactive protein (CRP) (Figs 1 to 3). Magnetic resonance imaging (MRI) scan showed a cystic lesion (where it is found to be mentioned), T1 hypointense and T2 hyperintense.

Posterior decompression by L3 to L5 laminectomy was performed. Intraoperatively, a cyst was noted in the epidural space spanning the whole of ligamentum flavum in a transverse and craniocaudal direction at L3/4 level. The dorsal side of the ligamentum flavum cyst extended up to L5 level space. The cauda equina was decompressed by excising the dorsal cyst wall and drainage of clear fluid inside dura repair done. The ventral wall could not be separated from the dura and was left in situ. Further decompression of bilateral lateral canals was performed by undercutting of the facet joints. Histological examinations suggestive of fibrocollagenous tissue with inflammatory cell infiltration (Fig. 4).

Postoperatively, the patient showed relief from the spinal claudication symptoms and improvements in ankle dorsiflexion, left great toe flexion and extension power to grade 3+/5. He could walk with stick with foot drop splint in bilateral lower limb. Bowl and bladder are intact.

DISCUSSION
Several studies have shown that the usual aging process of the ligamentum flavum causes thickening and loss of elasticity. Change in proteoglycans, loss of elastic fibers,
and increase in collagen tissue and chondroid metaplasia due to mechanical stress have been described.\textsuperscript{22} Additionally and closely related to age, amyloid can accumulate within the ligamentum flavum.\textsuperscript{25} This amyloid deposition has been reported in only a few cases to be associated with systemic amyloidosis.\textsuperscript{24} Similarly, age-related calcification of the ligamentum flavum can occur.\textsuperscript{26} A diffuse form of calcification contributing to the loss of elasticity and the thickening and a focal form of calcified material accumulation as well as granulomatous inflammation and tophaceous depositions of calcium pyrophosphate crystals can occur. These depositions have been ascribed to decreasing cellularity of the ligamentum flavum with age and resultant diminished calcification.
inhibiting factor production by fibroblastic-like cells.\textsuperscript{21} The tophaceous type of lesion seems to be closely related to previous degeneration of the affected ligament by minor trauma predisposing to calcium deposition. Activity of proteolytic enzymes within the ligament, produced by neutrophils localizing to calcified nodules, has been found.\textsuperscript{9} Wildi et al\textsuperscript{\textsuperscript{30}} found only four patients with calcium pyrophosphate depositions, suggesting that they play a minor role in the pathogenesis of ligamentum flavum pseudocysts. In addition to calcification, ossification of the ligamentum flavum might occur. This seems to follow a sequential process of chondroid metaplasia and eventual enchondral ossification mainly at the insertion site of the ligament. All these factors, resulting partially from mechanical stress, seem to contribute to loss of the natural structure of the ligamentum flavum, making them again susceptible to new mechanical stress, forming a vicious circle. The degenerated bony structure of the lumbar spine and the facet joints suggest a major pathogenic role of degenerative segmental instability in pseudocyst formation of the ligamentum flavum, as found by other authors too.\textsuperscript{18} Most ligamentum flavum cysts reported in the literature were also located laterally within the spinal canal. While possibly a consequence of chronic bony degenerative disease, this phenomenon may be further elucidated in certain cases by the observation that the yellow ligaments are not as thick laterally as they are medially. Furthermore, they form posterior recesses bilaterally to the vertebral bodies. These recesses are filled with epidural fat\textsuperscript{16} and offer an area of decreased resistance and may, as a result, tolerate cyst formation.

The pathogenesis of ligamentous degeneration remains to be elucidated, but it may be considered in the context of degenerative spinal changes. The spine is divided into alternating mobile and fixed segments, and the transitional zones between the mobile and fixed regions incur the most severe stress during motion. The anatomic disposition, histologic characteristics, and biomechanical properties of the ligamentum flavum indicate that it is markedly different from other spinal ligaments.\textsuperscript{10} The ligamentum flavum is a well-defined elastic structure composed of 80% elastic and 20% collagen fibers.\textsuperscript{31} This composition of dense connective tissue with elastic fiber predominance is rarely seen in other tissues, although it can be seen in the vestibular folds of the larynx and the media of large arteries.\textsuperscript{31} When a change occurs in the ligamentum flavum, regeneration of elastic fibers that includes the formation of collagen fibers and degenerative changes occurs, and this regenerative process leads to decrease in elasticity. Moreover, this process in the ligamentum flavum is markedly different from other spinal ligamentous reactions.\textsuperscript{10} Thus, chronic irritative or degenerative changes of the ligamentum flavum in the area of the cyst could predispose it to mechanical stress, even after a minor repeated injury.\textsuperscript{8}

Cysts of the ligamentum flavum have myxoid degeneration and arise from or are partially embedded in the inner surface of this ligament, and in contrast to juxta-articular cysts, are not related to the facet joint cavity. Pathogenesis of the cyst formation is secondary to ligamentous and fibrocollagenous tissue degeneration and hypermobility of the spinal segment, mainly at the transitional zones between the mobile and the fixed segments of the spine.\textsuperscript{14} These degenerative changes represent a histologically distinct entity different from ganglion or synovial cysts. Pathologic ligamentum flavum cysts can contain hemorrhage, and previous degeneration of the ligament may create conditions for the formation of hematoma. Rupture of vessels in degenerated lumbar ligamentum flavum may develop secondary to stretching forces on the back. The pathogenesis of the hematoma may originate from minor acute or chronic trauma, such as minor back injury, physical exertion or heavy lifting.\textsuperscript{20,26}

Intraspinal ligamentum flavum cysts are rare; they occur preferentially in the lower lumbar region,\textsuperscript{5,15,32} while cervical localization is uncommon.\textsuperscript{17} In most of the cases, ligamentum flavum cysts in the lumbar spine occur at L4–L5, the most mobile segment within the lumbar spine, and are frequently associated with lumbar degenerative spondylolisthesis. Cervical cysts are preferentially located in the cervicothoracic junction.\textsuperscript{29} Continuous stress to the ligamentum flavum due to minor chronic trauma, such as listhesis may predispose to the formation of the cyst.\textsuperscript{8} Only in a few cases is the localization of cysts C6–C7, C3–C4, and C5–C6 levels.\textsuperscript{29} No reports have described the appearance of these cysts in any region other than the mobile spine. The T2–T10 vertebrae mainly act with the ribs to form the thorax and are not generally considered to be part of the mobile spine (Table 1).

There are no specific clinical symptoms for ligamentum flavum cyst. Cysts in the spinal canal can impinge upon and displace neural structures and can lead to neurologic symptoms. The majority of symptomatic cysts usually presents with radiculopathy, such as sciatica in the case of lumbar cysts, and can mimic symptoms related to intervertebral disk herniation.\textsuperscript{15} In the study of Wildi et al,\textsuperscript{\textsuperscript{30}} 97% patients complained of radicular pain, 39% showed motor deficits, 55% had sensory changes, 18% had abnormal reflexes, and 33% showed a positive Lasèque sign. Our patient presented with gradually developing right-sided radicular pain involving mostly the L4 distribution with patellar reflex loss on the same side.
Neuroimaging is helpful in diagnosing cyst of the ligamentum flavum. On myelography, these lesions are recognized as intraspinal extradural masses and on postmyelogram computed tomography as a faint cyst adjacent to the ligamentum flavum.32 Magnetic resonance imaging provides the best images14,20,25,28: on T1-weighted images, the cysts have a variable signal, and on T2-weighted images, the cysts have a high-intensity signal.20,28 Differential diagnosis of imaging studies between ligamentum flavum cysts and synovial cysts is useful to the surgeon, as the latter are more difficult to resect, requiring exploration of the facet joint. Magnetic resonance imaging, in some cases of synovial cysts, reveals demonstrable communication with the facet joint with enhancement of the synovial cyst wall and of the adjacent facet joint. Synovial cysts often have a calcified rim, while ligamentum flavum cysts do not.

Differential diagnosis of intraspinal extradural mass lesions includes ligamentum flavum cyst, juxta-articular cysts (ganglion and synovial cysts), arachnoid cyst, perineural cyst, dermoid cyst, infectious cyst, schwannoma, meningioma, and metastasis or nontumorous-type mass lesions including neurofibromas, fibrous dysplasia, ependymal cyst, and rheumatoid arthritis pannus.15,20,23 The nomenclature of cysts in the spinal canal is somewhat unclear in the literature. Most intraspinal cysts reported are juxta-articular cysts. Ligamentum flavum and juxta-articular cysts can be definitely distinguished only by their pathological findings.

Conservative therapy appears to have no success.15,20 Most conservative therapies are temporary and have varying success in the short-term. Surgical removal is the first-choice therapy. The goal of surgery is spinal decompression as well as resection of the cyst and affected ligamentum flavum. Complete excision at the base of the ligamentous insertion of the cyst assures a minimal rate of recurrence. Wildi et al30 reported recurrence of the cyst in the remaining ligamentum flavum in two patients 1 year after surgery. While nearly 95% of all operated cysts can be removed in their entirety, a major reported intraoperative difficulty lies in the presence of adhesions to the dural wall, which is the main causative factor of incomplete resection.9

Complete removal of pseudocystic lesions generally has excellent results.2,9,14,27,28,30 Our patient showed complete postoperative resolution of symptoms. He is neurologically intact and symptom free to date.

CONCLUSION

To summarize, ligamentum flavum cysts represent a rare cause of lumbar nerve root compression or spinal stenosis. The lumbar ligamentum flavum undergoes lifelong mechanical stress. Similar to bony structures in this region, it degenerates with age. The degenerative changes in the lumbar ligamenta flava can be followed by cystic changes. Histologically, these degenerative changes represent a distinct entity different from ganglion or synovial cysts. Magnetic resonance imaging provides the best images. Radical removal of pseudocyst guarantees in nearly all cases complete relief of radiculopathy and seems to prevent recurrence of such a lesion at the same level.

REFERENCES


