Hemorrhagic Ligamentum Flavum Cyst Associated with Adjacent Bone Erosion: an Uncommon Subtype of Juxta-Facet Cyst—a Case Presentation

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Abstract
Ligamentum flavum cyst of the lumbar spine has seldom been described. The mechanism of formation of these cysts remains unknown, but it is thought to be part of the degenerative process. Although they are clearly visible on imaging studies, these cysts are frequently wrongly diagnosed as ganglion or synovial cysts. Bone erosion is rarely associated with this pathology. Most common symptom is back and radicular pain due to nerve root compression. We report a case of ligamentum flavum cyst associated with bone remodeling presented with back and radicular pain. It was correctly diagnosed pre-operatively and treated surgically with satisfactory clinical outcome.

Keywords Ligamentum flavum cyst · Spine · Juxta-facet cyst

Introduction
Ligamentum flavum cysts are considered a rare subtype of juxta-facet cyst along with cysts of the posterior longitudinal ligament and facet joints [11]. Most of the epidural cysts reported in the literature are synovial or ganglion cysts [11]. They were first reported by Moiel et al. in 1967 [10]. Although they are initially thought to arise from hypermobile facet joints and myxomatous degeneration of the ligamentum flavum, the exact pathophysiologic mechanism remains unclear [4, 13]. They arise usually dorsolateraly within the canal, and they may cause nerve root compression provoking back and radicular pain [13]. Although the ligamentum flavum is denser along the midline, it is an uncommon site for cyst development [13]. Moreover, bone remodeling is a rare radiological finding [1]. In this work, we report the case of an 83-year-old female harboring a large ligamentum flavum cyst associated with bone erosion. It was correctly diagnosed by the neuroradiologist, and it was confirmed intra-operatively and histopathologically.

Case Report
An 83-year-old female without any known medical problems other than sleep apnea presented with symptoms of chronic low back and radicular pain and progressive neurogenic claudication over a 12-month period. She denied weakness, bowel or bladder disturbances, or trauma/heavy weightlifting. Neurologic examination revealed right S1 radicular pain and sensory loss in the same dermatome as well as positive Lasègue sign and Achilles reflex loss.

An MRI and a CT were performed, revealing local degenerative changes and an expansive lesion behind L5 and S1 vertebral bodies, located in the anterior epidural space to the right and contiguous to the ligamentum flavum, extending to the L5-S1 right lateral recess and to the right L5-S1 and S1-S2 neural
foramina, compressing both L5 and S1 neural roots. This lesion had heterogeneous and predominant hyposignal T2 and hypersignal T1 and presented no enhancement after gadolinium administration, in keeping with a hemorrhagic cyst with subacute bleeding. All modifications were associated to local bone remodeling of the posterior surface of S1 vertebral body and its right neural foramen, depicting the slow growth of a benign lesion (Figs. 1, 2, 3, 4, and 5). Regarding differential diagnosis, the presence of hematic products, and the absence of contrast enhancement speak against a schwannoma, the bone remodeling speaks against a herniated disc, and the most probable diagnosis was considered a hemorrhagic arthro-synovial cyst or a hemorrhagic cyst of the ligamentum flavum.

The patient was taken to the operating room, and an L5-S1 fenestration was performed using standard microsurgical technique. An approximately 3 cm cystic mass was observed in the ventral surface of the dura. The lesion was adherent and it was arising from within the dorsal aspect of the ligamentum flavum. The affected ligamentum flavum was excised. Notably, there was no point of connection with facet joint capsules or dura mater. The lesion was resected in piecemeal fashion. The cyst contained some mucinous fluid and blood in various stages of coagulation. The L5 and S1 nerve roots were decompressed, and the patient recovered uneventfully after surgery. The histological analysis of the tissue from the surgical extirpation shows a cystic degeneration of the ligamentum flavum without synovial lining (Fig. 6). These results confirmed the initial hypothesis of ligamentum flavum cyst (Fig. 7).

On 12-month follow-up, she was doing well, without any radicular pain and no radiological findings of cyst recurrence or instability.
Discussion

Ligamentum flavum cyst is a result of the spectrum of degenerative process of the spine [6]. These lesions are found most commonly in the lumbar spine. The most common level is the L4-L5, followed by the L5-S1 and L3-L4 levels [12, 13]. Cervical spine is a rarer location of this entity [3]. The exact pathogenic mechanism is not yet fully understood [13]. Continuous stress to the ligamentum flavum due to minor chronic trauma in the context of hypermobility and spinal instability are thought to be associated with ligamentum flavum hypertrophy, myxoid degeneration, necrosis, calcification, and fibrosis [12–14]. Therefore, not surprisingly spinal degenerative disease, such as spondylosis, spondylolisthesis, and disk disease are commonly found in conjunction with these cysts [3, 12–14].

Ligamentum flavum cysts must be differentiated from the synovial or ganglion cysts which are common lesions occurring in the facet joints [12–14]. Synovial cysts have a synovial lining membrane and are continuous with the facet joints and remain outside the ligamentum flavum [8]. They have pseudostratified columnar epithelium, filled with clear and xanthochromic fluid [8, 12]. On the other hand, spinal ganglion cysts do not communicate with the facet joint cavity, have no synovial lining but fibrous tissue wall, and are filled with a viscous, gelatinous material [5, 12]. In contrast, ligamentum flavum cysts are embedded in the inner surface of ligamentum flavum with no epithelial lining and importantly no communication with facet joints [5, 7].

On CT these cysts are indistinct, appearing as hypodense round structures. Bone erosion and remodeling is rarely associated with these cysts. From our search, we have identified only one short case report of a ligamentum flavum cyst associated with bone remodeling [1]. As in our case (Fig. 5), calcium deposition may be detected on CT scans. The exact pathophysiological mechanism however remains unknown [1, 13–15]. Histological analysis of such cyst showed calcium triphosphate and calcium phosphate deposition associated with degenerative changes [1]. This hypothesis is based on reduction in elastic fibers and increase in collagen fibrils due to migration of hypertrophic chondrocytes. Lastly, metabolic diseases such as hyperparathyroidism, hypothyroidism hypophosphatemia, and hemochromatosis can induce calcium deposits [1, 12–14]. On MRI, which is the method of choice for the investigation, they are depicted as epidural cystic lesions adjacent to a ligament flavum that may be hypertrophied, characterized by a variable signal intensity on T1-weighted images according to its contents, most commonly hyperintense on T2WI and with peripheric enhancement.

Although it may be difficult to distinguish between ligamentum flavum cysts and synovial cysts on imaging studies, such a differentiation may be helpful to the surgeon for presurgical planning. The role of the neuroradiologist cannot be more emphasized for accurate pre-operative diagnostic and
However, in most cases, ligamentum flavum cysts are wrongly diagnosed as ganglion or synovial cysts, and the diagnosis is made intraoperatively followed by a histologic confirmation [1, 2, 4, 13]. In one case, the authors reported that this pathology was correctly diagnosed pre-operatively [11].

The clinical presentation of ligamentum flavum cysts depends on its location and its size. Most commonly, cysts cause radicular pain and sensory deficit at a rate of 97% and 55%, respectively [14]. Motor function weakness is a less usual presentation (39%). Lasègue sign can be presented in one-third and abnormal reflexes in one-fifth of cases [14, 15]. Our patient presented with gradually developing right-sided radicular pain involving the S1 distribution, positive Lasègue sign, and Achilles reflex loss.

The differential diagnosis of cystic lesions involving the ligamentum flavum includes inflammatory; granuloma, degenerative; intraligamentous amyloid deposition, ossification, myxomatous degeneration, tumoral; perineural cyst, intraspinal dermoid cyst, neurofibroma, infectious; and hydatid cysts or cysticercosis and hematoma [12–14].

Conservative therapy does not have favorable clinical outcomes [1, 12–14]. Most conservative therapies have temporal effect. Surgical removal is the treatment of choice [12–15]. The goal of surgery is decompression of the thecal sac and nerve roots and resection of the cyst [13]. Excision of the affected ligamentum flavum at the base of the insertion of the cyst is associated with minimal rate of recurrence. Cyst recurrence has been reported when the affected ligamentum

![Fig. 5](a) Sagittal and (b) coronal CT reformats depict the local bone remodeling, suggesting the slow growth and benign etiology of the expansive right L5-S1 epidural lesion. Moreover, panel b reveals remodeling of right S1 foramen (arrow)

![Fig. 6](On the right, we recognize the ligamentum flavum. On the left, there is a cystic degeneration with intraluminal deposition of fibrin (yellow-orange), without synovial lining (Van Gieson’s stain, ×200)

![Fig. 7](Post-operative coronal T2-STIR and axial T2-TSE MR images demonstrate the complete removal of the lesion and the decompression of the right L5 and S1 neural roots)
flavum was left behind [15]. Technical challenges reported in the literature lies in the presence of adhesions to the dural wall, which is the principal factor of incomplete resection [14]. In our case, complete removal of the cyst was performed including the base of insertion which it was dissected carefully from the adjacent dura. We did not remove the affected bone since we believe it is not associated with cyst recurrence. Moreover, on 12-month follow-up, the patient is completely asymptomatic.

**Conclusion**

Ligamentum flavum cyst is a rare cause of lumbar nerve root compression or spinal stenosis, rarely associated with surrounding bone reaction. They should be considered in differential diagnoses of cystic lesions of the lumbar spine. We describe a case of ligamentum flavum cyst with a significant bone erosion correctly diagnosed pre-operatively and successfully treated with radical surgical excision with complete resolution of symptoms.

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