CASE REPORT/*OPIS PRZYPADKU*

Symptomatic ganglion cyst of ligamentum flavum as a late complication of lumbar fixation

Objawowa torbiel galaretowata więzadła żółtego jako późne powikłanie stabilizacji kręgosłupa w odcinku lędźwiowym

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Abstract

We report the case of a 72-year-old man who underwent surgery for a mobile spondylolisthesis L4-L5. Six months later, magnetic resonance imaging revealed an extradural cyst of the ligamentum flavum at L5-S1, which was then removed. Histological examination revealed a ganglion cyst of the ligamentum flavum. Cyst formation could be explained primarily according to the natural history of chronic degenerative disease of spine elements. Nevertheless, we could also consider the cyst formation as demonstrative of an adjacent segment syndrome: hypermobility of the L5-S1 segment just below three fixed vertebral segments would have triggered the mechanical stress necessary for L5-S1 ligamentum flavum degeneration.

Key words: ligamentum flavum, ganglion cyst, lumbar fixation, adjacent segment syndrome.

Streszczenie

W pracy opisano przypadek 72-letniego mężczyzny operowanego z powodu ruchomego kręgozmyku L4-L5. W badaniu za pomocą rezonansu magnetycznego wykonanym 6 miesięcy po operacji stwierdzono zewnątrztwardówkową torbiel więzadła żółtego na poziomie L5/S1, którą następnie usunięto chirurgicznie. Na podstawie badania histopatologicznego rozpoznano torbiel galaretowatą więzadła żółtego. Powstanie torbieli można by tłumaczyć głównie naturalnym przebiegiem przewlekłego procesu zwyrodnieniowego elementów kręgosłupa. Należałoby rozważyć również możliwość powstania torbieli w mechanizmie zespołu sąsiedniego segmentu – nadmierna ruchomość w odcinku L5-S1 bezpośrednio poniżej trzech segmentów poddanych stabilizacji mogłaby wywołać mechaniczne obciążenie skutkujące zwyrodnieniem więzadła żółtego w odcinku L5-S1.

Słowa kluczowe: więzadło żółte, torbiel galaretowata, stabilizacja kręgosłupa w odcinku lędźwiowym, zespół sąsiedniego segmentu.

Introduction

Cysts of the ligamentum flavum rarely cause neurological signs and symptoms [1-3]. They can be histologically classified as either synovial or ganglion cysts [4]. The first are true cysts, characterised by an inner synovial lining layer, which is common with other cystic lesions such as facet-joint cysts and posterior longi-

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tudinal ligament cysts [5,6]. Conversely, ganglion cysts are pseudo-cysts that result from the mucoid degeneration of the ligamentum flavum [5,7]. They are typically not in communication with the synovial layer of facet joints, because their walls lack a synovial layer. They are more frequent in aged people and in hypermobile vertebral segments, because the degeneration of the ligamentum flavum is facilitated by the continuous mechanical stress.

This is a rare case of symptomatic cystic degeneration of L5-S1 ligamentum flavum, secondary to surgical fixation of an adjacent segment.

Case report

We describe the case of a 72-year-old man who was admitted to our department with persistent low back pain. Magnetic resonance imaging (MRI) and dynamic X-ray revealed signs of diffuse degeneration of the lumbar spine with a mobile spondylolisthesis L4-L5 and lumbar stenosis of L3-L4 (Figs. 1A-B). The preoperative MRI also revealed a small cyst of the ligamentum flavum at L4-L5 (Fig. 1C), which was not considered significant. The patient underwent laminectomy L3-L4-L5 and corrective surgery in which pedicle screws and rods were inserted at levels L3-L5, with a posterior lumbar interbody fusion (PLIF) of L4-L5 (Fig. 2A). During surgery, the small cyst of the ligamentum flavum was recognised and removed with flavectomy of the segment. The L5-S1 segment was not fixed.

The clinical course was uneventful and the patient was discharged with complete resolution of his symptoms. Six months after surgery, the patient presented with a progressive right L5 radiculopathy with crippling sciatica. A second MRI assessment revealed the presence of an extradural posterior cystic lesion measuring 2 cm at the L5-S1 level, which compressed the dural sac and partially occupied the right neural L5-S1 foramen (Figs. 2B-C).

The patient underwent a second operation to remove the lesion and to decompress the dural sac and the right L5 root using a median posterior approach. The patient refused completion of surgical correction at the L5-S1 segment.

The cystic lesion was removed 'en bloc' together with the L5-S1 ligamentum flavum, in which it was completely included (Fig. 2D). Histological and immuno-histochemical examination revealed a ganglion

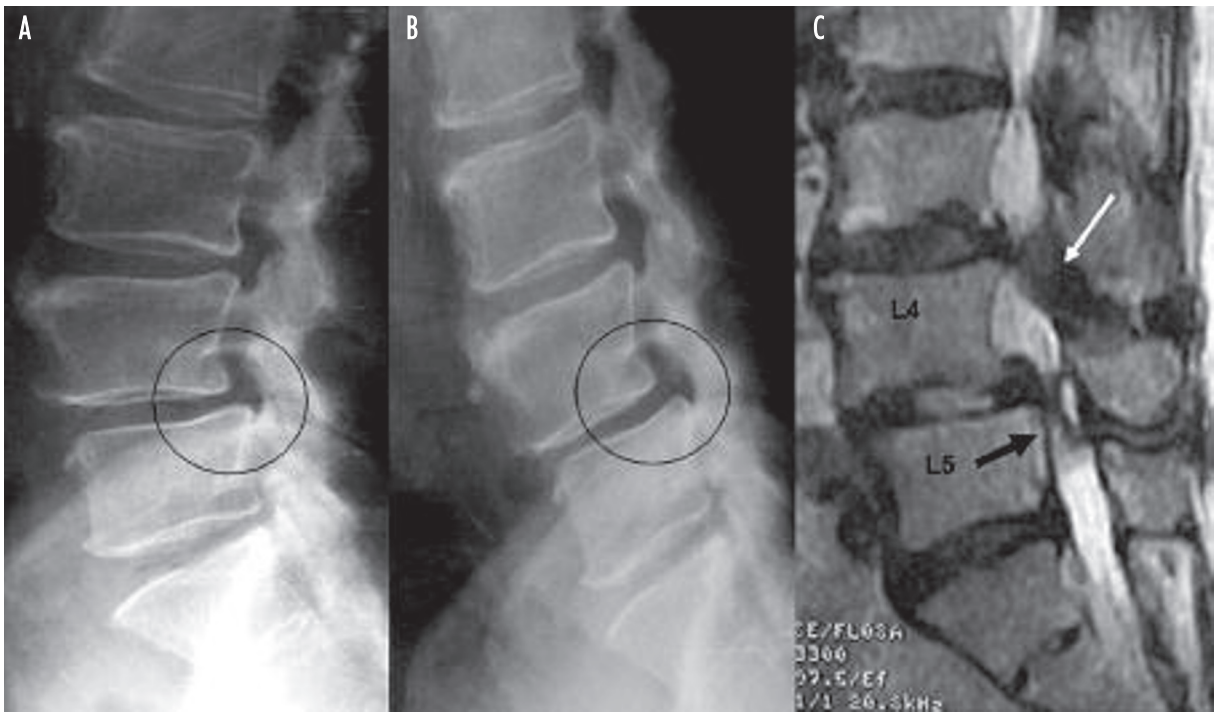

Fig. 1. Preoperative dynamic lateral X-ray of lumbar spine (A – extension and B – flexion): note the L4-L5 anterolisthesis in flexion (black circle); C) – preoperative T2-weighted MRI in sagittal plane showing L4-L5 listhesis and L3-L4 and L4-L5 canal stenosis. A hypertrophic ligamentum flavum is evident at L3-L4 (white arrow) and a small ganglion cyst of the ligamentum flavum is visible at L4-L5 (black arrow)

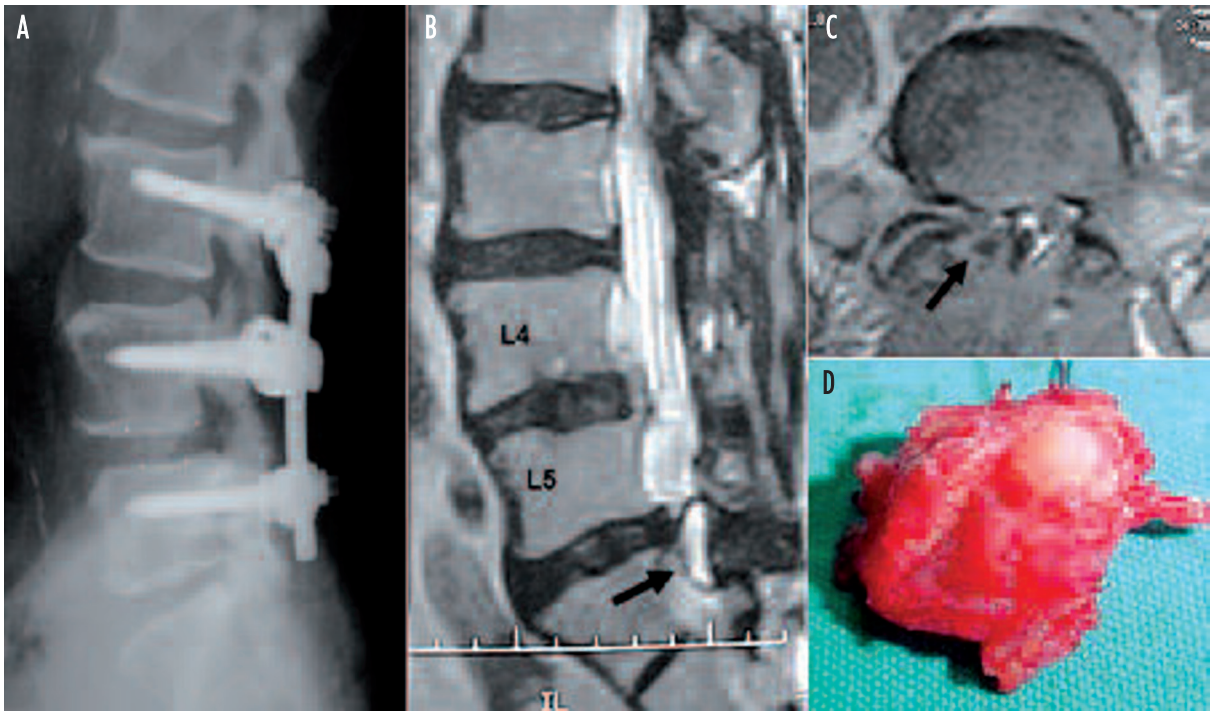

Fig. 2. A) Postoperative lateral X-ray of lumbar spine showing the pedicle screws at L3-L5 levels connected by postero-lateral rods and the L4-L5 posterior inter-body fusion; B) and C) T2-weighted MRI in sagittal plane and T1-weighted MRI in axial plane performed six months after the operation of spinal fixation. The canal is well decompressed and the listhesis reduced, but at the L5-S1 level a larger ganglion cyst with mass effect on L5 root is evident (black arrows); D) intraoperative photograph of the cyst, removed en bloc with all the L5-S1 ligamentum flavum; the cyst was filled with a viscous, gelatinous material

cyst of the ligamentum flavum. The cyst was filled with a viscous material. The ligamentum flavum was found to have diffuse mucoid degeneration, with multilocular pseudocystic spaces lacking a synovial layer (Fig. 3). The following clinical course was characterised by the complete resolution of L5 radiculopathy.

Discussion

The ligamenta flava are paired structures connecting adjacent lamina. Unlike all other ligaments, they are composed principally of elastin (80%), as are the vestibular folds of the larynx and the media of large arteries. When degenerative changes occur due to aging, the ligamentum flavum undergoes an increase of collagen fibres and an accompanying decrease in elasticity; further mechanical stress due to continuous minor trauma or degenerative micro-instability may lead to chronic mucoid degeneration of the ligament and to the formation of a ganglion cyst [4,7].

Ganglion cysts of the ligamentum flavum are rare, and a differential diagnosis of a benign intraspinal extradural mass lesion includes synovial or ganglion cysts, perineural cysts, intraspinal dermoid cysts, neurofibroma, schwannoma with cystic degeneration, hydatid cysts, or rheumatoid arthritis pannus; meningiomas with cystic necrosis are usually more diffusely enhanced by contrast medium and metastases are generally associated with osteolysis [6-9].

Cysts of the ligamentum flavum are typically described in mobile regions of the spine, confirming the importance of mechanical factors in their pathogenesis [3,7,10,11].

They may present with acute and/or chronic symptoms [12]. A cyst of the ligamentum flavum is most likely to produce radicular compression, but vertebral canal stenosis and vertebral bone erosion resulting from such cysts have also been described [13,14].

The timing of symptoms in our patient is consistent with chronic ligament degeneration and the slow formation of a cyst. Moreover, the absence of erythrocytes in the cyst indicates that acute traumatic progression of the cyst was unlikely. Progressive haemorrhage of the cyst wall due to continuous trauma or in the case of epidural anaesthesia can explain the sudden progression of cyst volume and related symptoms in some cases [15-17].

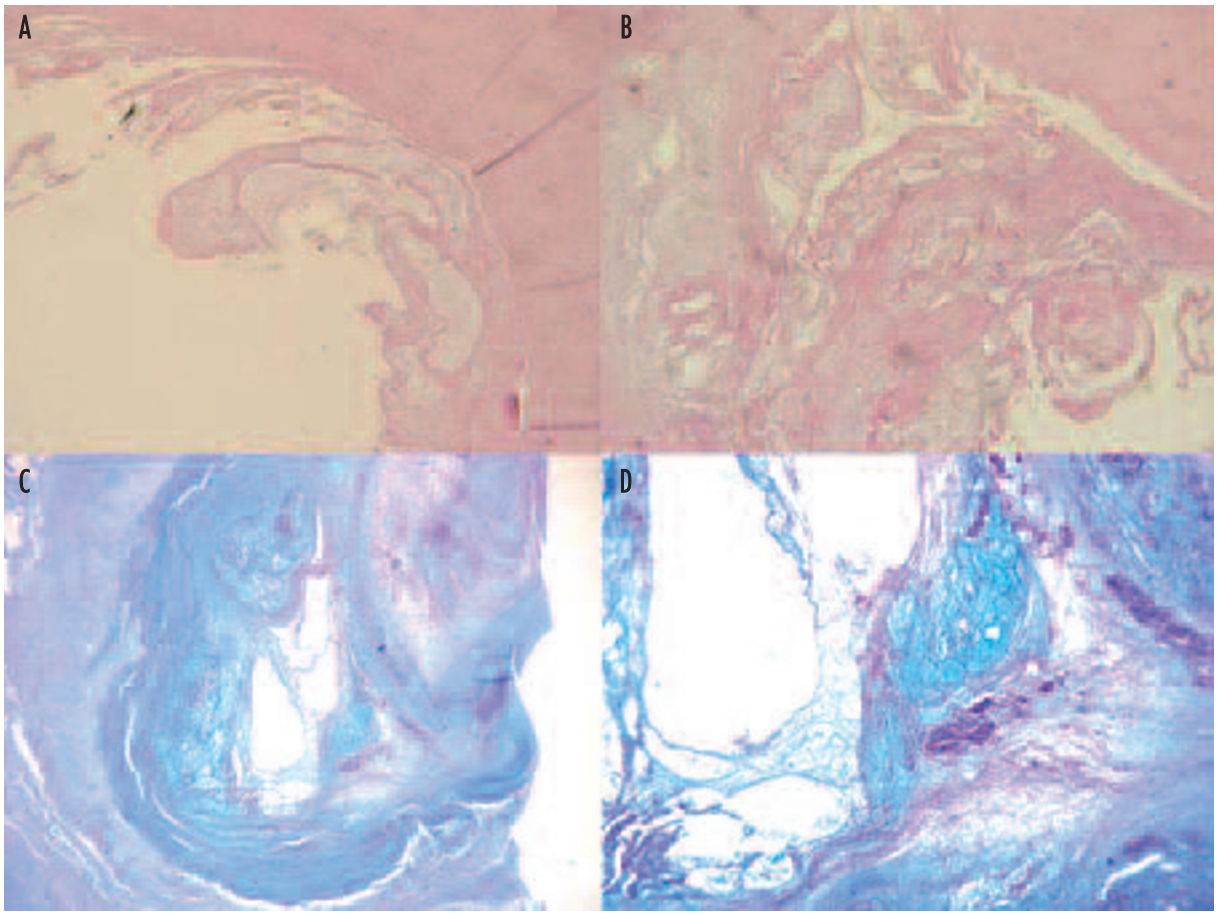

Fig. 3. Photomicrographs of cyst and ligamentum flavum sections; A) and B) – multilocular pseudocystic spaces in the context of ligament's fibrous connective tissue. Note the absence of a synovial layer of the cystic walls (haematoxylin and eosin, original magnification ×10 and ×20); C) and D) – mucoid degeneration of the ligament, characterized by an increase in the mucoid ground substance in the connective tissue containing glycoprotein and mucoprotein, which stains blue to bluish green (Alcian blue-PAS, original magnification ×5 and ×20)

Our patient presented with a diffuse chronic degenerative disease of the lower lumbar spine and an instability of the L4-L5 segment, and underwent surgery to repair levels L3-L5. The L5-S1 segment was not repaired. In the first six months after the operation, our patient developed a ganglion cyst of the ligamentum flavum at the L5-S1 segment, which produced crippling sciatica owing to a right L5 root compression.

Cyst formation could be explained primarily according to the natural history of chronic degenerative disease of spine elements. Furthermore, the presence of a smaller ganglion cyst of the ligamentum flavum at the unstable L4-L5 segment, revealed by MRI before the first surgery, would suggest that the ligamentum flavum was predisposed to cystic degeneration.

Nevertheless, the natural hypermobility of L5-S1 just below three fixed vertebral segments may have produced a continuous mechanical stress on a degenerated ligamentum flavum to trigger its cystic degeneration in the following six months.

In this case, the symptomatic cystic mucoid degeneration of the ligamentum flavum could have been stimulated by the surgical fixation of adjacent lumbar segments; given this possibility, we could consider the cyst formation as demonstrative of an adjacent segment syndrome.

When the cyst is symptomatic, treatment should be a total excision coupled with complete flavectomy. Since cyst formation may be triggered by abnormal segmental mobility, it is recommended to evaluate spinal motion by dynamic X-rays, to eventually plan a surgical fixation of involved segments [3,7,10,11].

In conclusion, we presented a case of a symptomatic ganglion cyst of the ligamentum flavum secondary to

lumbar fixation in an old patient with degenerative spondylolisthesis. Cyst formation could be finally considered expression of an adjacent segment syndrome.

Disclosure

Authors report no conflict of interest.

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