

LIGAMENTUM FLAVUM HEMATOMA: A CASE REPORT AND LITERATURE REVIEW

HEMATOMA DE LIGAMENTO AMARELO: RELATO DE CASO E REVISÃO DA LITERATURA

HEMATOMA DEL LIGAMENTO AMARILLO: CASO CLÍNICO Y REVISIÓN DE LA LITERATURA

ERICSON SFREDDO¹, MARCELO TEODORO EZEQUIEL GUERRA²

ABSTRACT

The aim is to present a rare case of ligamentum flavum hematoma in the lumbar region, discuss its physiopathology and treatment and review the literature. A woman aged 68 presented with neurogenic claudication due to degenerative lumbar spondylolisthesis that evolved into a sudden worsening with cauda equina syndrome. The magnetic resonance imaging (MRI) showed signs of degeneration of the lumbar spine, with a narrow spinal canal from L2 to S1, anterolisthesis L4 L5 and an expansive lesion hyperintense on T1-weighted and hypointense on T2-weighted images considered compatible with hematoma in the topography of the yellow ligament in L1-L2. The patient underwent laminectomy and lumbar fixation. Her evolution was good in the postoperative period and at 18 months of follow-up she walked alone, despite the pain that is controlled with simple medications. Even though rare, it seems that ligamentum flavum hematoma has a relationship with the degeneration and rupture of small vessels associated with micro trauma to the spine. Its physiopathology is not well defined and treatment is similar to other spine compression processes.

Keywords: Spine; Cauda Equina; Ligamentum flavum; Hematoma; Laminectomy; Spondylosis.

RESUMO

O objetivo é apresentar um caso raro de hematoma do ligamento amarelo na região lombar, discutir sua fisiopatologia e tratamento e revisar a literatura. Uma mulher de 68 anos apresentou-se com claudicação neurogênica devido à doença degenerativa lombar e espondilolistese que evoluiu para uma piora súbita com a síndrome da cauda equina. A imagem por ressonância magnética (IRM) mostrou sinais de degeneração da coluna lombar, com um canal vertebral estreito de L2 a S1, ântero-listese de L4 L5 e, posteriormente no nível da L1-L2, um processo expansivo arredondado e hiperintenso em T1 e com bordas hipointensas em T2 ponderada, compatível com hematoma na topografia do ligamento amarelo. A paciente foi submetida a laminectomia e fixação lombar. Sua evolução foi boa no período pós-operatório e, aos 18 meses de follow-up andou sozinha, apesar da dor que é controlada com medicamentos simples. Mesmo sendo raro, parece que o hematoma do ligamento amarelo tem uma relação com a degeneração e ruptura de pequenos vasos associadas a microtraumas na coluna vertebral. Sua fisiopatologia ainda não é bem definida e o tratamento é semelhante ao de outros processos de compressão da coluna vertebral.

Descritores: Coluna Vertebral; Cauda Equina; Ligamento Amarelo; Hematoma; Laminectomia; Espondilose.

RESUMEN

El objetivo es presentar un caso raro de un hematoma ligamento flavum en la región lumbar, discutir su fisiopatología y el tratamiento y revisión de la literatura. Una mujer de 68 años presentó claudicación neurogénica debido a la espondilolistesis lumbar degenerativa y que se convirtió en un repentino empeoramiento con el síndrome de cauda equina. Una imagen de resonancia magnética (RM) mostró signos de degeneración de la columna lumbar, con canal espinal estrecho de L2 a S1, anterolistesis L4 L5 y en la L1-L2, un proceso expansivo redondeado e hiperintenso en T1 y hipointenso en los bordes en T2, compatible con hematoma en la topografía del ligamento amarillo. La paciente fue sometida a laminectomía y fijación lumbar. Su evolución fue buena en el postoperatorio y, a los 18 meses de seguimiento caminaba sola, a pesar del dolor que se controla con medicamentos simples. Aunque raro, parece que el hematoma del ligamento amarillo tiene relación con la degeneración y la ruptura de pequeños vasos asociadas a micro-traumas en la columna vertebral. Su fisiopatología no está bien definida y el tratamiento es similar a otros procesos de compresión de la columna vertebral.

Descriptores: Columna Vertebral; Cauda Equina; Ligamento Amarillo; Hematoma; Laminectomía; Espondilosis.

INTRODUCTION

Ligamentum flavum hematoma (LFH) is a rare cause of radicular or spine compression. It usually occurs in mobile segments of the spine, and it seems to bear a relation with smaller traumas; however, this relation does not occur in some cases as indicated by some reports, as this one. From the pathologic standpoint, there is a degeneration of the ligament with the subsequent rupture of small winding vessels. The present report it is a case of a patient presenting with a rapid neurological deterioration because of a cauda equina syndrome which was due to an advanced degenerative disease; the

patient underwent a laminectomy and fixation of lumbar instability. The pertinent literature is discussed and reviewed.

CASE REPORT

A 68-year-old woman had a history of lumbar pain irradiated to the posterior surface of the thighs at rest; the pain was associated to progressive limping. More recently, the patient presented with an unbearable pain and a decrease in quality of life, despite pharmacological treatment. During hospitalization for treatment and investigation, she presented with high and sudden lumbar pain and

1. Preceptor in Neurosurgery, Hospital Cristo Redentor, Porto Alegre, RS, Brazil.
2. Professor of Orthopaedics and Traumatology, ULBRA, Porto Alegre, RS, Brazil.

loss of strength in lower limbs. The neurological exam showed crural paraparesis degree I and urinary incontinence, characterizing cauda equina syndrome. The patient underwent magnetic resonance imaging (MRI), which showed degenerative signs of the lumbar spine with a narrowed spine canal of L2 and S1, anterolisthesis of L4 upon L5 and, at the level of L1–L2, a posterior lesion hyperintense in T1-weighted and with hypointense in T2-weighted (Figure 1 and 2).

The patient underwent decompression of the cauda equina by laminectomy of L1 to L5 and posterolateral fixation with pedicular screws from L1 to S1 plus intertransversal graft from iliac. During surgery, it was observed that the interior of the lesion between L1–L2 had hemorrhagic content in different stages of absorption, which characterized several episodes of bleeding at different times (Figure 3).

Presently, the patient is at 18 months of follow up and, after several physiotherapy sessions, is gradually recovering strength. Today, she walks unassisted and makes eventual use of analgesic medication for lumbalgia.



Figure 1. MRI T1-weighted: Degeneration of multiple lumbar levels with anterolisthesis of the body of L4 and retrolisthesis of the bodies of L2 and L1. Hyperintense image between as spinal apophysis of L1 and L2 causing compression and narrowing of the spine canal.

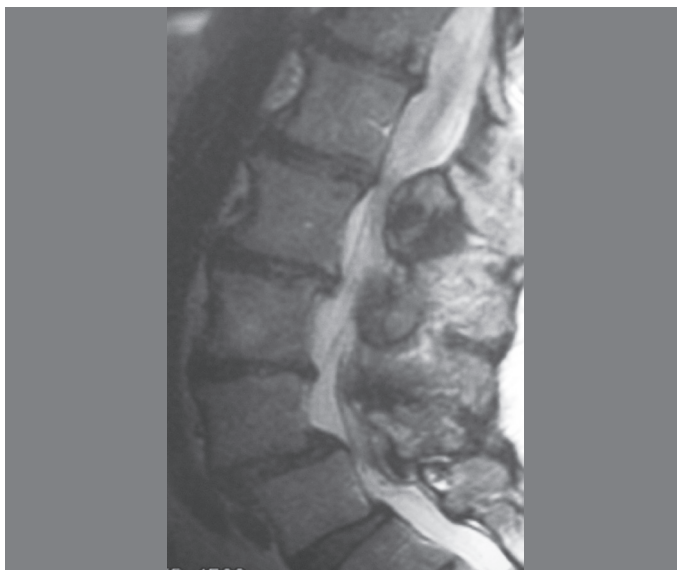


Figure 2. MRI T2-weighted: Aspect of the L1–L2 lesion with a peripheral hypointense image and hyperintense in its interior.

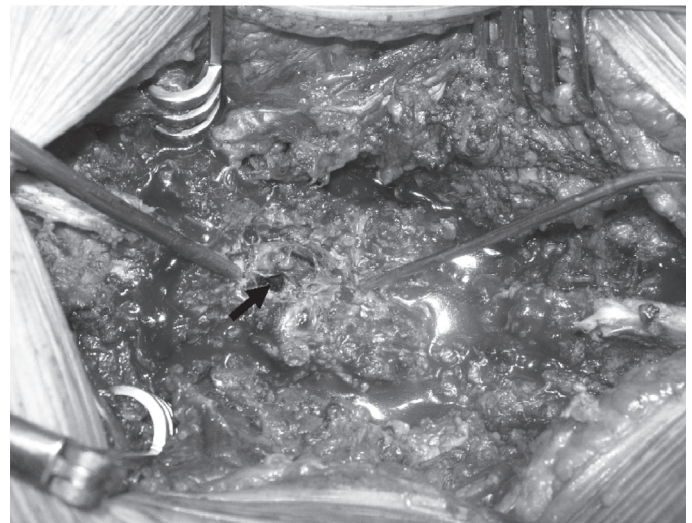


Figure 3. Transoperatory image: Broad laminectomy from L1 to L5. The arrow points to the open hematoma of the ligamentum flavum and its content being aspirated. Note the various phases of absorption of the hematoma.

DISCUSSION

Currently, a few cases of ligamentum flavum hematoma have been reported in the English literature indexed¹: one case in the cervical spine², 3 cases in the thoracic spine^{3–5}, and the remaining, as well as the present case, in the lumbar spine (Table 1). The most distinguishing characteristic is that most patients are male¹. Most cases presents a small trauma as the probable genesis of the hematoma; however, one of the 3 cases of Spuck et al.⁴ did not present a history of trauma or intensive physical exercises; it was related to the complication of a percutaneous procedure for relief of low lumbar pain. The case presented herein was related only to a severe lumbar disease, although the hematoma was located at a superior level when compared to the alterations.

The ligamentum flavum consists of elastic fibers and collagen. It is poorly vascularized, and few small vessels go through it⁶. The ligamentum flavum is an unlikely site of bleeding because of its poor vascularization and the constant tension forces to which the ligaments of the vertebral spine are submitted. An exception to this is to the presence of a severe degenerative process⁴. Minamide et al.⁷ suggests that bleeding is related to irregular vessels of thin walls in a ligament that is degenerated and hypertrophied. An increase in abdominal pressure after a small trauma would transmit the pressure to these thin-walled vessels, then the proliferated vessels may be susceptible to rupture, causing the hematoma. Miyakoshi et al.³ suggests that the poor alignment of the vertebral spine submits the ligamentum flavum to an excessive axial load at a given point and relates this fact to the genesis of the hematoma that occurred according to his report at the level of T9–T10. A small trauma and poor vascularization may be important predisposing factors that increase the risk of hematoma⁸; however, in the present case there was no injury of any kind, and the hematoma occurred far from where the greatest degenerative compromise was, which contradicts the present theory. Moreover, according to Yamaguchi et al.¹, if the genesis of the hematoma depends on the presence of lumbar degeneration and small injuries, why is it so rare?

At the lumbar levels, tolerance to compression seems to be higher than at other levels. As result, the gradual formation of the hematoma or granuloma may take weeks to become symptomatic¹. The differential diagnosis includes synovial cyst, ganglion cyst, spontaneous epidural hematoma, neoplasia, and other cystic lesions^{7,9}.

Magnetic resonance imaging is the method of choice for diagnosis. In T2-weighted the hematoma appears as a hyperintense cyst that is very similar to a juxtafacet cyst⁵. Images depend on the size, location and mainly age of the hematoma. In the acute stage, it appears hypointense in T2-weighted and isointense in T1-weighted.

Table 1. Reported Cases of Ligamentum Flavum Hematoma.

Reference	Ag / sex	Level	Symptom	Treatment	Outcome
Sweasey et al ¹⁶	43 ♂	L4-5	Back & leg pain	R	Excellent
	60 ♂	L2-3	Leg pain & Weakness	R	Excellent
Cruz-Conde et al ¹⁶	67 ♂	L4-5	Leg pain	R + Fu	Excellent
Minamide et al ⁷	76 ♂	L3-4	Leg pain	L	Excellent
Mahallati et al ⁹	30 ♂	L3-4	Leg pain	R	Excellent
Yuceer et al ¹⁷	67 ♂	L2-3	Leg pain	R	Excellent
Hirakawa et al ⁸	50 ♂	L4-5	Low back pain	F + Fu	Excellent
Maezawa et al ⁵	66 ♂	T11-12	Epiconus Syndrome	L	Numbness
Chi et al ¹⁸	64 ♀	L3-4	Radiculopathy	L	Excellent
Mizuno et al ¹³	45 ♀	L4-5	Leg pain & Weakness	L	Excellent
Miyakoshi et al ³	66 ♀	T9-10	Leg Weakness & numbness	L	Excellent
Chen et al ²	72 ♂	C3-4	Neck/Arm pain & Weakness	L	Excellent
Yamaguchi et al ¹	62 ♂	L3-4	LBP & Weakness	HL	Excellent
Spuck et al ⁴	64 ♀	L2-3	Claudication	IL	Excellent
	62 ♂	L4-5	Leg pain & Weakness	IL	Excellent
	60 ♂	T12-L1	Thigh pain	IL	Excellent
Albanese et al ¹⁴	70 ♀	L1-2	Leg pain & Weakness	L	Excellent
Miyakoshi et al ¹⁹	67 ♂	C3-4	Weakness & Numbness	R	Numbness
Kotil et al ²⁰	80 ♂	L4-5	LBP & Hypoesthesia L5-S1	L	Excellent
	74 ♂	L4-5	LBP & Hypoesthesia L5-S1	L	Excellent
Lee et al ²¹	72 ♂	T7-8	Paraparesis	L	Excellent
Takahashi et al ²²	63 ♀	L3-4	LBP & Sciatic pain	L	Excellent
	63 ♂	L5-S1	LBP & Sciatic pain	HL	Excellent
Tamura et al ¹⁵	69 ♂	C2-3	Fall & Unsteady gait	L	Frankel D
Kim et al ²³	67 ♂	L2-3	Cauda equina syndrome	L	Numbness
This paper	68 ♀	L1-2	Cauda equina syndrome	L + Fu*	Frankel D

(R) Resection; (F) facetectomy; (L) Laminectomy; (HL) Hemilaminectomy; (IL) Interlaminectomy; (Fu) Fusion; (LBP) Low back pain. * Fusion with pedicular screws for treatment of vertebral instability in the inferior levels.

Between the third and seventh day, the meta-hemoglobin makes the image hyperintense in T1-weighted. In the next weeks, the hemolysis of erythrocytes results in the accumulation of extracellular meta-hemoglobin that appears hyperintense in T1-weighted and T2-weighted¹⁰⁻¹².

The treatment of choice is laminectomy or partial laminectomy, when the objective is the removal of the expansive effect^{1-3,6,13,15,23}, interlaminectomy with the removal of the ligamentum flavum⁴ or associated to arthrodesis when there are the criteria for spine instability^{6,8}. In the case presented herein, we opted for laminectomy for the removal of the hematoma and fixation with pedicular screws for

treatment of spine instability in the inferior levels, especially at L4–L5 with the extension of the fixation up to L2–L3 where the laminectomy was performed.

Albanese et al.¹⁴ reviewed the literature and found excellent follow up on all cases of epidural hemorrhage due to ligamentum flavum hematoma. The authors points out that early decompression is essential for improvement of symptoms, however reviews all cases of epidural hemorrhage, regardless of cause. Tamura et al.¹⁵ suggest that if a patient presents with a lesion and symptoms such as those described here, the lesion should be surgically removed as soon as possible.

REFERENCES

1. Yamaguchi S, Hida K, Akino M, Seki T, Yano S, Iwasaki Y. Ligamentum flavum hematoma in the lumbar spine. *Neurol Med Chir (Tokyo)*. 2005;45(5):272-6.
2. Chen HC, Hsu PW, Lin CY, Tzann WC. Symptomatic hematoma of cervical ligamentum flavum: case report. *Spine (Phila Pa 1976)*. 2005;30(16):E489-91.
3. Miyakoshi N, Shimada Y, Okada K, Hongo M, Kasukawa Y, Itoi E. Ligamentum flavum hematoma in the rigid thoracic spinal segments: case report. *J Neurosurg Spine*. 2005;2(4):495-7.
4. Spuck S, Stellmacher F, Wiesmann M, Kranz R. Case reports: a rare cause of radicular complaints: ligamentum flavum hematoma. *Clin Orthop Relat Res*. 2006;(443):337-41.
5. Maezawa Y, Baba H, Uchida K, Kokubo Y, Kubota C, Noriki S. Ligamentum flavum hematoma in the thoracic spine. *Clin Imaging*. 2001;25(4):265-7.
6. Cruz-Conde R, Berjano P, Buitron Z. Ligamentum flavum hematoma presenting as progressive root compression in the lumbar spine. *Spine (Phila Pa 1976)*. 1995;20(13):1506-9.
7. Minamide A, Yoshida M, Tamaki T, Natsumi K. Ligamentum flavum hematoma in the lumbar spine. *J Orthop Sci*. 1999;4(5):376-9.
8. Hirakawa K, Hanakita J, Suwa H, Matsuoka N, Oda M, Muro H, et al. Post-traumatic ligamentum flavum progressive hematoma: a case report. *Spine (Phila Pa 1976)*. 2000;25(9):1182-4.
9. Mahallati H, Wallace CJ, Hunter KM, Bilbao JM, Clark AW. MR imaging of a hemorrhagic and granulomatous cyst of the ligamentum flavum with pathologic correlation. *AJNR Am J Neuroradiol*. 1999;20(6):1166-8.
10. Dorsay TA, Helms CA. MR imaging of epidural hematoma in the lumbar spine. *Skeletal Radiol*. 2002;31(12):677-85.
11. Harbury OL, Provenzale JM, Barboriak DP. Vertex epidural hematomas: imaging findings and diagnostic pitfalls. *Eur J Radiol*. 2000;36(3):150-7.
12. Sarrazin JL. [Imaging of postoperative lumbar spine]. *J Radiol*. 2003;84(2 Pt 2):241-50.
13. Mizuno J, Nakagawa H, Inoue T, Hashizume Y. Ligamentum flavum hematoma in the lumbar spine. *Neurol Med Chir (Tokyo)*. 2005;45(4):212-5.
14. Albanese A, Braconi A, Anile C, Mannino S, Sabatino G, Mangiola A. Spontaneous hematoma of ligamentum flavum. Case report and literature review. *J Neurosurg Sci*. 2006;50(2):59-61.
15. Tamura T, Sakai T, Sairyo K, Takao S, Kagawa S, Katoh S, et al. Hematoma in the cervical ligamentum flavum. Report of a case and review of the literature. *Skeletal Radiol*. 2010;39(3):289-93.
16. Sweasey TA, Coester HC, Rawal H, Blaivas M, McGillicuddy JE. Ligamentum flavum hematoma. Report of two cases. *J Neurosurg*. 1992;76(3):534-7.
17. Yuceer N, Baskaya MK, Smith P, Willis BK. Hematoma of the ligamentum flavum in the lumbar spine: case report. *Surg Neurol*. 2000;53(6):598-600.
18. Chi TW, Li KT, Chieng PU. Post-traumatic ligamentum flavum hematoma: a case report. *Kaohsiung J Med Sci*. 2004;20(1):41-4.
19. Miyakoshi N, Shimada Y, Kasukawa Y, Ando S. Ligamentum flavum hematoma in the cervical spine - case report. *Neurol Med Chir (Tokyo)*. 2006;46(11):556-8.
20. Kotil K, Bilge T. A ligamentum flavum hematoma presenting as an L5 radiculopathy. *J Clin Neurosci*. 2007;14(10):994-7.
21. Lee HW, Song JH, Chang IB, Choi HC. Spontaneous ligamentum flavum hematoma in the rigid thoracic spine: a case report and review of the literature. *J Korean Neurosurg Soc*. 2008;44(1):47-51.
22. Takahashi H, Wada A, Yokoyama Y, Fukushi S, Sakurai T, Shibuya K, et al. Ligamentum flavum haematoma: a report of two cases. *J Orthop Surg (Hong Kong)*. 2009;17(2):212-5.
23. Kim HS, Kim SW, Lee SM, Shin H. Ligamentum flavum hematoma in the adjacent segment after a long level fusion. *J Korean Neurosurg Soc*. 2011;49(1):58-60.