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Idiopathic and Chronic Epidural Hematoma in the Lumbar Spine: A Case Report and Review of Literatures

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Abstract

Spontaneous and chronic epidural hematoma (SSEH) in the lumbar spine is rare, and idiopathic and chronic SSEH in the lumbar spine is extremely rare disease. Most of lumbar SSEH were acute and secondary with trauma, hematologic disorders, drug, and surgical procedure. Only 20 cases of chronic SSEH in the lumbar spine have been reported and 14 cases among them were considered to be idiopathic. Definitive guidelines for management of this condition are not clear and surgical total evacuation was performed in most of the cases. Some authors reported the epidural bleeding originates in the rupture of Batson's plexus due to a rise in intra-abdominal pressure, but the mechanism is not clearly clarified. We report a surgical case of idiopathic and chronic SSEH. A 61-year-old woman suffered a sudden onset of severe lumbar pain during sleep. She had no history of trauma, spinal surgery, or hypertension. Magnetic resonance imaging revealed a lumbar chronic epidural hematoma which compressed the dural sac behind and extended from L2 to L5. This patient underwent the partial evacuation of the hematoma with partial hemilaminectomy on left at L2/3, resulting in immediate pain relief and resolution of symptoms and almost absorption of the hematoma within 1 week of the procedure. We presented this rare case and reviewed idiopathic and chronic epidural hematoma in the lumbar spine.

Key words: idiopathic, epidural hematoma, chronic, lumbar spine

Introduction

Spinal epidural hematoma (SEH) is a rare condition described in associated with trauma, hematologic disorders, anticoagulation and antiplatelet therapy, vascular malformations, neoplasm, trauma, or medical intervention such as epidural catheterization or spinal surgery. Most cases of spontaneous SEH were acute and secondary caused by the aforesaid risk factors. In 2009, Sarubbo et al.[1)] reviewed spontaneous and idiopathic chronic spinal epidural hematoma in the lumbar spine and one case was reported later in English.[2)] Surgical evacuation was performed in most of the cases and led to the good results. Here, we report a case of chronic and idiopathic SEH, which was successfully treated with surgical partial evacuation and reviewed about such cases.

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Case Report

A 61-year-old healthy woman presented to our hospital with a history of sudden onset of severe lumbar pain and left buttock and leg pain during sleep. She denied previous lumbar trauma, spine surgery, or other spinal disorders. She had a history of mild lumbar pain 2 months before and fully recovered without any treatment within 2 weeks. On physical examination, she had hypalgesia at L2–5 bilaterally, severely at L4, in the dermatome and no motor weakness, bladder, or rectal dysfunction. She never took drugs, such as antiplatelet and anticoagulant agents. All blood and coagulation tests (platelet counts, prothrombin time, partial prothrombin time, and fibrinogen) showed normal values.

Computed tomography (CT) scans revealed an extensive high-density mass behind the dural sac from L2 to L5 body (Fig. 1a). On magnetic resonance imaging (MRI, Philips, Intera Achieva 3.0T Quasar Dural), the mass showed low intensity on T1 (TR 500 ms; TE 9.0 ms; NSA 2; FOV 320; Matrax 448), very low on T2 (TR 3000 ms; TE 90 ms; NSA 2; FOV 320;

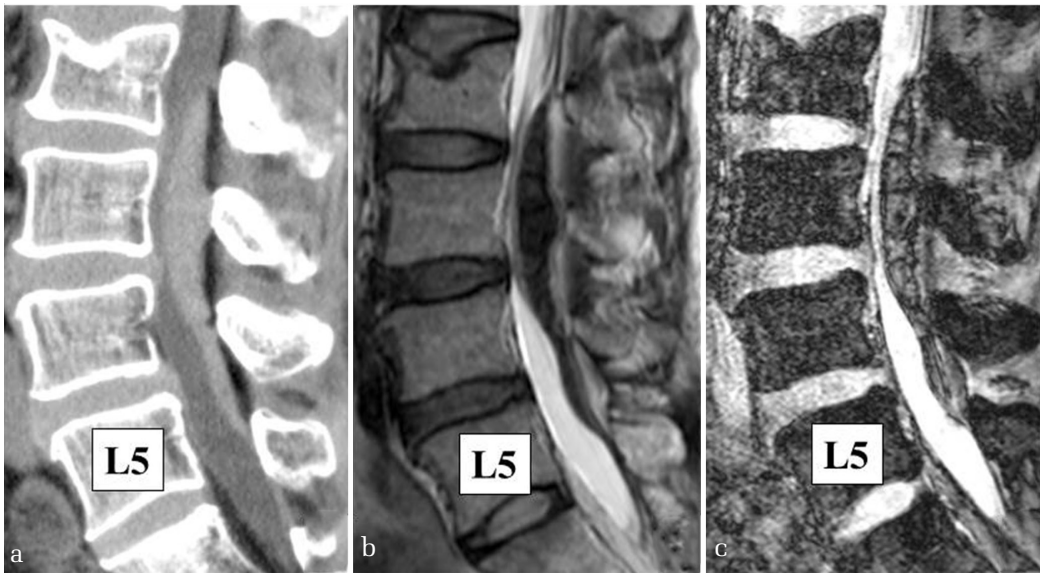

Fig. 1 Preoperative CT (a) and MR images (b: T2, c: T2*) on sagittal view.

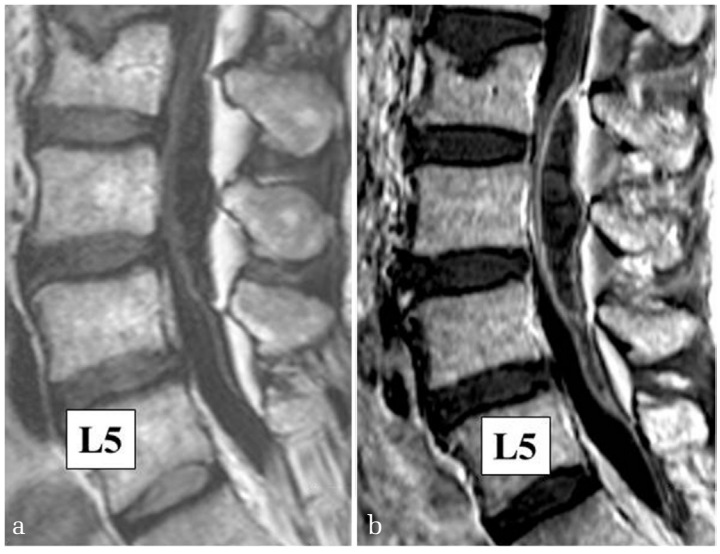

Fig. 2 Preoperative T1 (a) and Gadolinium-enhanced T1 (b) images on sagittal view.

Matrax 400), and T2* (TR 600 ms; TE 16 ms; NSA 2; FOV 180; Matrax 320) and surrounded by a thin membrane (Figs. 1b and 1c, Fig 2a). Gadolinium-enhanced T1 images showed slight enhancement in the membrane (Fig. 2b). Computed tomography angiography (CTA) and magnetic resonance angiography (MRA) did not show vascular malformation and positron emission tomography (PET), CT showed no hot areas in the lesion. Based on the radiological findings, the mass was considered as chronic epidural hematoma and conservative therapy was chosen at first. But, the severe lumbar pain did not improve with enough medicine and bed rest, and surgical procedure was performed 1 week after admission.

A 3.5 cm midline skin incision was made at L2/3 disc space. The fascia was incised left to the midline, sequential dilators inserted and a quadrant retractor (Medtronic, Minneapolis, MN, USA) was placed at L2/3. When the yellow ligament was partially removed after partial laminectomy on left at L2 and L3 (Fig. 3), a jelly-like dark brown hematoma with a capsule was seen (Fig. 4a). After cutting the capsule, the hematoma was carefully removed piecemeal. Histopathologically, the membrane of the capsule was granulation tissue with neogenesis of blood capillary and had hyperplasia of fibroblast and invasion of inflammatory cells and no tumorigenesis (Fig. 4b). The hematoma showed only blood cells and fibrin, and the membrane was adherent to epidural fat tissue. Therefore, the hematoma was diagnosed as chronic SEH.

Lumbar pain was decreased immediately after the operation. Postoperative MRI on day 2 showed the hematoma was removed almost completely at L2/3, but remained mainly at L3/4 (Fig. 5a). The MRI on postoperative day 7 showed that the hematoma

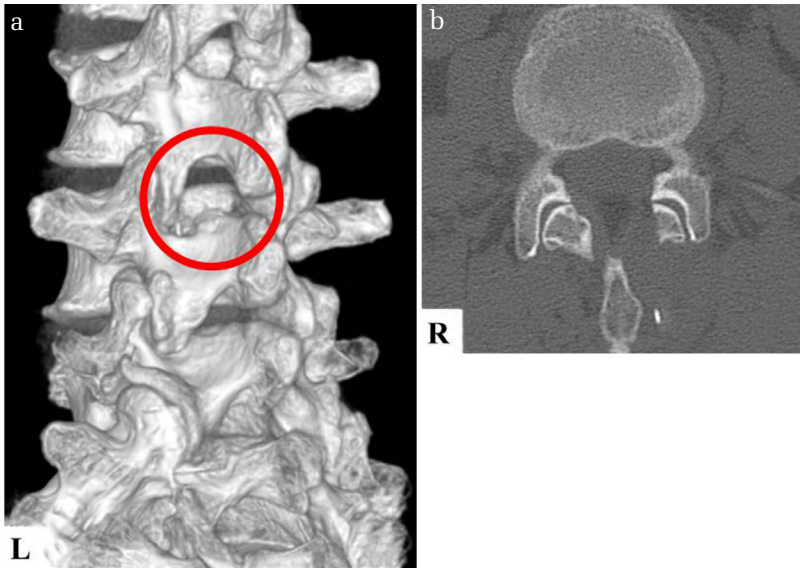

Fig. 3 Postoperative 3D CT (a) and CT on axial (b).

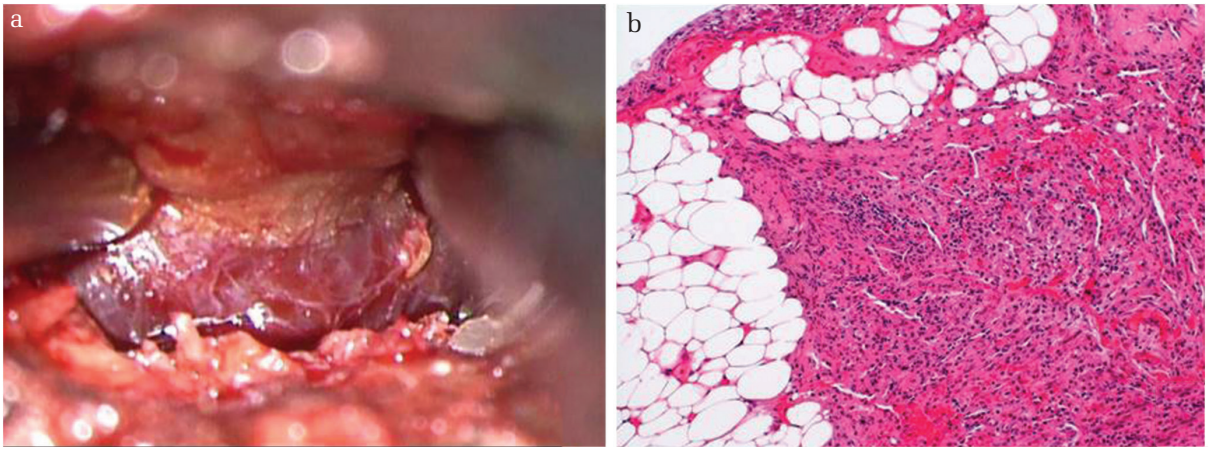

Fig. 4 Intraoperative image shows an encapsulated fibrous dark brown hematoma (a) and photomicrograph of the membrane shows granulation tissue and epidural fat tissue (b).

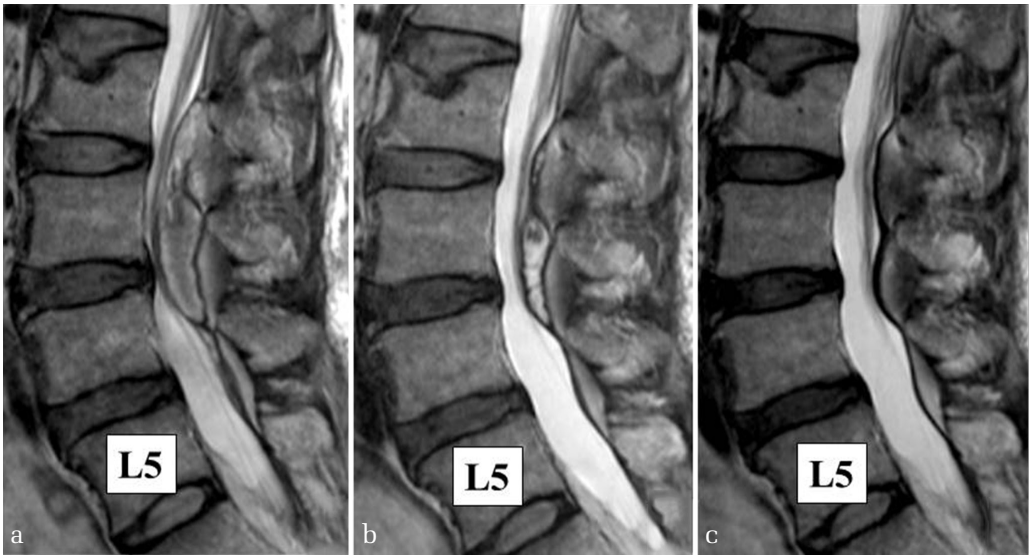

Fig. 5 T2 images on sagittal view after operation [(a) 2 days after operation, (b) 7 days, (c) 1 month].

disappeared almost completely and her lumbar pain vanished and the hematoma completely absorbed in 1 month after operation (Figs. 5b and 5c).

Discussion

Spinal epidural hematoma is uncommon condition and can be classified as spontaneous, secondary, and idiopathic but the definition was not clear. The most important causes of secondary SEHs are with hematologic disorders,[3–6] anticoagulation[7–8] and antiplatelet therapy[9–11], vascular malformations[12], neoplasm, trauma[13], or medical intervention, such as epidural catheterization or spinal surgery.[14,15] Spontaneous SEH is defined as SEH with causative risk factors, such as minor trauma, chiropractic manipulation,[16] Paget's disease,[17,18] ankylosing spondylitis,[19,20] and rheumatoid arthritis.[21] Our case never had above risk factors and was recognized as idiopathic SEH.

Chronic SEH is defined as spinal compression for months or years with mild symptom and all of the reported cases in the past occurred in the lumbar spine. In the past reports, 17 cases of chronic and spontaneous SEH were reported in English literature.[1,2,22–31] As shown in Table 1, 14 cases of idiopathic and chronic SEHs were reported. Three cases were diagnosed on myelogram, four were on CT, and seven cases were on MRI. Though the hematoma was shown as various images on T1 and T2 images, chronic hematoma was in general low intensity on both T1 and T2 images. In our case, SEH which was spread from L2 to L5 body level was like crescent and showed low intensity on both of T1 and T2 images. Therefore, it was in chronic phase based on MRI even though the symptom occurred suddenly. The MR images in addition to the shape of the SEH indicated that the hematoma was enlarged little by little since the first onset of lumbar pain was 2 months before.

The locations and shapes of SEHs diagnosed with myelogram and CT were not correctly expressed. The SEHs were located at the lateral recess in five, at lateral behind the dural sac in five, at midline behind the dural sac in three, and not described in one. The SEHs were located at only one level in 11 and at two levels in three. In our case, SEH was spread from L2 to L5 body level. All of SEH cases presented with pain in the lumbar, buttock, and lower leg, and only two cases showed sudden severe pain and the other showed gradual progressive pain. Three cases complicated with slight motor weakness. The evacuation of SEHs was performed because of the severe pain in all the cases and none of the procedure was emergent. In one case, the operation was performed 3 days after admission because the pain was severe. In our case, it was 7 days after because of the same reason. The evacuation can be considered when enough medicine and rest therapy are not effective.

In past reports as shown in Table 1, 14 cases of idiopathic and chronic SEHs were operated, but two cases were not described in detail. About 11 cases were localized in the single level and three cases were spread at 2 levels. The SEH in our cases were spread from L2 to L5 body level. In past literatures, surgical evacuation resulted in good clinical courses and laminectomy in nine, hemilaminectomy in two and partial hemilaminectomy in one were performed. The SEHs located laterally were removed with laminectomy or hemilaminectomy and the SEHs located on midline were with laminectomy. The complete evacuation was done in five cases and partial evacuation was in one and the other eight cases were not described. Although the follow-up time was not described, the SEH removed partially vanished completely. Our case showed sudden severe lumbar pain and hypalgesia, and we choose surgery 1 week after the onset because the symptom did not improve. Because the SEH compressed dural sac the most severely at L2/3, partial hemilaminectomy at L2/3 and partial removal of SEH at the level was performed. The remaining SEH was completely disappeared 1 month after surgery. The residual chronic subdural hematoma in the cranial cavity after drainage is gradually absorbed and the hematoma does not need complete removal. For the same reason, we believe that partial removal and release of the capsule and epidural space can lead the drain to the epidural space and gradual absorption of the hematoma. Our procedure was considered as less invasive and effective to get enough results.

Recently, less invasive surgery is recommended and endoscopic surgery may one option for chronic SEH.[32–34] In this case, the SEH was preoperatively diagnosed as idiopathic and chronic because MRA, CTA, and PET CT denied that vascular malformation and tumor caused the SEH. But the conclusive evidence was not preoperatively obtained and microscopic removal was chosen. As a result, partial removal of the SEH led to the release of lumbar pain and hypalgesia, and early absorption of the hematoma 1 week after operation and complete absorption 1 month after operation.

The cause of idiopathic SEH was not clearly clarified. Some authors reported the epidural bleeding originates in the rupture of Batson's plexus due to a rise in intra-abdominal pressure.[17,27,30,31,35–40] On the other hand, other authors proposed that arterial bleeding or the rupture of epidural vascular malformation lead epidural hematoma.[41] In this case, the cause of the epidural hematoma was not clearly

Table 1 The summarize of all cases published in the literature of idiopathic and chronic epidural spinal hematoma in the lumbar spine

No.	Author	Year	Age	Sex	Level	Location	Shape	Duration	Tool	Surgery	Removal rate	Pathology
1	Harris	1969	66	M	L5/S1	?	?	Several M	Myelogram	Laminectomy L5–S1	?	Yes
2	Boyd	1972	66	M	L4–5	Posterior lateral	?	7M	Myelogram	Partial hemilaminectomy L4–5	?	No
3	Boyd	1972	75	F	L3/4	Posterior lateral	?	6M	Myelogram	Laminectomy	?	Yes
4	Levitan	1983	58	F	L4	Lateral recess	?	Sudden	CT: high	Yes but unknown	?	No
5	Levitan	1983	90	F	L3/4	Posterior lateral	?	1.5M	CT: high	Yes but unknown	?	No
6	Nehls	1984	74	M	L3/4	Lateral recess	?	2M	CT: iso	Laminectomy L3–4	?	Yes
7	De Almeida	1989	88	M	L3/4	Posterolateral	?	4M	CT: high	Laminectomy L3–4	?	No
8	Nakgami	1992	58	F	L4	Posterior	Crescent	2M	T1, T2: high	Total laminectomy L4, partial laminectomy L3, L5	Total	Yes
9	Lunardi	1995	45	M	L2–3	Posterior midline	Crescent	2M	T1, T2: high	Laminectomy L2–3	Total	Yes
10	Riffaud	1999	70	F	L4/5	Lateral recess	Nodular	Sudden	T1: iso, T2: low	Laminectomy L4	?	Yes
11	Vazquez-Barquero	2000	75	F	L2–3	Posterior	Nodular	7M	T1: iso, T2: low	Laminectomy L2–3	Total	Yes
12	Sarubbo	2009	65	F	L3/4	Lateral recess	Nodular	3M	T1: high	Laminectomy L3–4	Partial	No
13	Sarubbo	2009	85	M	L3	Lateral recess	Nodular	2M	T2: low	Hemilaminectomy L3	Total	No
14	Matsui	2014	78	M	L4	Posterior lateral	Nodular	9M	T1: high, T2: low	Hemilaminectomy L4, PLIF	Total	Yes
15	Our case		61	F	L2–5	Posterior	Crescent	Sudden	T1, T2: low	Partial hemilaminectomy L2–3	Partial	Yes

proved in pathological findings. But it seemed to be the rupture of venous plexus because arterial bleeding was not seen intraoperatively and the hematoma was jelly-like dark brown colored substance. The membrane was adherent to the epidural fat tissue and it may be considered that the venous bleeding from the epidural fat tissue caused the hematoma. The pathogenesis was not proved, but it seemed to be similar to chronic subdural hematoma of the intracranial cavity.

Conclusion

Epidural hematoma of the lumbar spine is rare condition and idiopathic and chronic epidural hematoma is even rarer. We must investigate the cause of the hematoma, such as vascular malformation and tumor. When the hematoma is diagnosed as idiopathic and chronic, partial removal of the hematoma can be one option as less invasive surgery depending on the condition of the hematoma.

Conflicts of Interest Disclosure

No potential conflicts of interest were disclosed.

References

1) Sarubbo S, Garofano F, Maida G, Fainardi E, Granieri E, Cavallo MA: Spontaneous and idiopathic chronic spinal epidural hematoma: two case reports and review of the literature. *Eur Spine J* 18: 1055–1061, 2009
2) Matsui H, Imagama S, Ito Z, et al.: Chronic spontaneous lumbar epidural hematoma simulating extradural spinal tumor: a case report. *Nagoya J Med Sci* 76: 195–201, 2014
3) Nojiri H, Kim S, Tsuji T, Uta S: Spontaneous spinal epidural hematoma as the initial presentation of leukemia. *Eur Spine J* 18 Suppl 2: 220–223, 2009
4) Stanley P, McComb JG: Chronic spinal epidural hematoma in hemophilia A in a child. *Pediatr Radiol* 13: 241–243, 1983
5) Bamford CR: Spinal epidural hematoma due to heparin. *Arch Neurol* 35: 693–694, 1978
6) Harik SI, Raichle ME, Reis DJ: Spontaneously remitting spinal epidural hematoma in a patient on anticoagulants. *N Engl J Med* 284: 1355–1357, 1971
7) Lederle FA, Cundy KV, Farinha P, McCormick DP: Spinal epidural hematoma associated with warfarin therapy. *Am J Med* 100: 237–238, 1996
8) Van Schaeybroeck P, Van Calenbergh F, Van De Werf F, Demaerel P, Goffin J, Plets C: Spontaneous spinal epidural hematoma associated with thrombolysis and anticoagulation therapy: report of three cases. *Clin Neurol Neurosurg* 100: 283–287, 1998
9) Bhat KJ, Kapoor S, Watali YZ, Sharma JR: Spontaneous epidural hematoma of spine associated with clopidogrel: a case study and review of the literature. *Asian J Neurosurg* 10: 54, 2015
10) Morales Ciancio RA, Drain O, Rillardon L, Guigui P: Acute spontaneous spinal epidural hematoma: an important differential diagnosis in patients under clopidogrel therapy. *Spine J* 8: 544–547, 2008
11) Sung JH, Hong JT, Son BC, Lee SW: Clopidogrel-induced spontaneous spinal epidural hematoma. *J Korean Med Sci* 22: 577–579, 2007
12) D'Angelo V, Bizzozero L, Talamonti G, Ferrara M, Colombo N: Value of magnetic resonance imaging in spontaneous extradural spinal hematoma due to vascular malformation: case report. *Surg Neurol* 34: 343–344, 1990
13) Devadiga KV, Gass HH: Chronic lumbar extradural haematoma simulating disc syndrome. *J Neurol Neurosurg Psychiatry* 36: 255–259, 1973
14) Spanier DE, Stambough JL: Delayed postoperative epidural hematoma formation after heparinization in lumbar spinal surgery. *J Spinal Disord* 13: 46–49, 2000
15) Sweasey TA, Coester HC, Rawal H, Blaivas M, McGillicuddy JE: Ligamentum flavum hematoma. Report of two cases. *J Neurosurg* 76: 534–537, 1992
16) Solheim O, Jorgensen JV, Nygaard OP: Lumbar epidural hematoma after chiropractic manipulation for lower-back pain: case report. *Neurosurgery* 61: E170–E171, 2007
17) Lee KS, McWhorter JM, Angelo JN: Spinal epidural hematoma associated with Paget's disease. *Surg Neurol* 30: 131–134, 1988
18) Hayem G, Deutsch E, Roux S, Palazzo E, Grossin M, Meyer O: Spontaneous spinal epidural hematoma with spinal cord compression complicating plasma cell myeloma. A case report. *Spine* (*Phila Pa 1976*) 23: 2432–2435, 1998
19) Gustafsson H, Rutberg H, Bengtsson M: Spinal haematoma following epidural analgesia. Report of a patient with ankylosing spondylitis and a bleeding diathesis. *Anaesthesia* 43: 220–222, 1988
20) Wu CT, Lee ST: Spinal epidural hematoma and ankylosing spondylitis: case report and review of the literature. *J Trauma* 44: 558–561, 1998
21) Farhat SM, Schneider RC, Gray JM: Traumatic spinal extradural hematoma associated with cervical fractures in rheumatoid spondylitis. *J Trauma* 13: 591–599, 1973
22) Boyd HR, Pear BL: Chronic spontaneous spinal epidural hematoma. Report of two cases. *J Neurosurg* 36, 239–342, 1972
23) de Almeida GM, Bianco E, Shibata MK: Encapsulated spinal epidural hematoma in an octogenarian. *Surg Neurol* 32: 394–395, 1989
24) Harris ME: Spontaneous epidural spinal hemorrhage. *Am J Roentgenol Radium Ther Nucl Med* 105: 383–385, 1969
25) Kotilainen EM, Pajulo O: Spontaneous epidural hematoma as a cause of sciatic pain in a schoolboy. *Pediatr Neurol* 17: 350–352, 1997

26) Levitan LH, Wiens CW: Chronic lumbar extradural hematoma: CT findings. *Radiology* 148: 707–708, 1983
27) Lunardi P, Mastronardi L, Lo Bianco F, Schettini G, Puzzilli F: Chronic spontaneous spinal epidural hematoma simulating a lumbar stenosis. *Eur Spine J* 4: 64–66, 1995
28) Nakagami W, Yokota S, Ohishi Y, et al.: Chronic spontaneous lumbar spinal epidural hematoma. *Spine* (*Phila Pa 1976*) 17: 1509–1511, 1992
29) Nehls DG, Shetter AG, Hodak JA, Waggener JD: Chronic spinal epidural hematoma presenting as lumbar stenosis: clinical, myelographic, and computed tomographic features. A case report. *Neurosurgery* 14: 230–233, 1984
30) Riffaud L, Morandi X, Chabert E, Brassier G: Spontaneous chronic spinal epidural hematoma of the lumbar spine. *J Neuroradiol* 26: 64–67, 1999
31) Vázquez-Barquero A, Abascal F, Garcia-Valtuille R, Pinto JI, Figols FJ, Cerezal L: Chronic nontraumatic spinal epidural hematoma of the lumbar spine: MRI diagnosis. *Eur Radiol* 10: 1602–1605, 2000
32) Komp M, Hahn P, Oezdemir S, et al.: Bilateral spinal decompression of lumbar central stenosis with the full-endoscopic interlaminar versus microsurgical laminotomy technique: a prospective, randomized, controlled study. *Pain Physician* 18: 61–70, 2015
33) Markovic M, Zivkovic N, Spaic M, et al.: Full-endoscopic interlaminar operations in lumbar compressive lesions surgery: prospective study of 350 patients. "Endos" study. *J Neurosurg Sci* 2016 Jun 30. [Epub ahead of print]
34) Ying GY, Yao Y, Shen F, Ren YC, Chen CM, Zhu YJ: Percutaneous endoscopic removal of a lumbar epidural angiolipoma via interlaminar approach: a technical report. *World Neurosurg* 99: 59–62, 2017
35) Batson OV: The vertebral vein system. Caldwell lecture, 1956. *Am J Roentgenol Radium Ther Nucl Med* 78: 195–212, 1957
36) Cohen JE, Ginsberg HJ, Emery D, Schwartz ML: Fatal spontaneous spinal epidural hematoma following thrombolysis for myocardial infarction. *Surg Neurol* 49: 520–522; discussion 523, 1998
37) Gundry CR, Heithoff KB: Epidural hematoma of the lumbar spine: 18 surgically confirmed cases. *Radiology* 187: 427–431, 1993
38) Lawton MT, Porter RW, Heiserman JE, Jacobowitz R, Sonntag VK, Dickman CA: Surgical management of spinal epidural hematoma: relationship between surgical timing and neurological outcome. *J Neurosurg* 83: 1–7, 1995
39) Pear BL: Spinal epidural hematoma. *Am J Roentgenol Radium Ther Nucl Med* 115: 155–164, 1972
40) Johnston RA: The management of acute spinal cord compression. *J Neurol Neurosurg Psychiatry* 56: 1046–1054, 1993
41) Beatty RM, Winston KR: Spontaneous cervical epidural hematoma. A consideration of etiology. *J Neurosurg* 61: 143–148, 1984

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