Thoracic spine fracture associated with an extradural lipoma: Case report and systematic review of the literature

Giovanni Noia,[1] Mattia Basilico,[2] Raffaele Vitiello,[2] Andrea Perna,[1] Antonio Leone,[3] Nicolò Rumi,[3] Francesco Ciro Tamburrelli[1]

[1]UOC Chirurgia Vertebrale, [2]Istituto di Clinica Ortopedica, [3]Istituto di Radiologia, Fondazione Policlinico Universitario A. Gemelli IRCCS, Rome; Università Cattolica del Sacro Cuore, Rome, Italy

Abstract

Isolated extradural lipoma (IEL), not associated to spinal dysraphisms, is a rare condition. Frequently IEL was confused with much more frequent diffuse lipomatosis. The lesion can be completely asymptomatic and occasionally diagnosed with magnetic resonance (MR) imaging. This paper describes a case of a patient with an axial compression fracture of the thoracic spine associated with an extradural lipoma. We also performed a systematic review of the pertinent literature in order to retrieve the key information regarding: the diagnosis, the clinical features and the treatment.

Introduction

Isolated Extradural lipoma (IEL), not associated with spinal dysraphism, is a rare lesion accounting for 0.4-0.8% of all intraspinal tumors; however, the true incidence is not established and, probably, underestimated since extradural lipomas rarely cause clinical symptoms,[1] on the other hand, many pathologies of the spine can cause non-specific symptoms such as low back pain.[2-5] It is important to differentiate IEL from spinal epidural lipomatosis (SEL) which is instead a hyperplasia of epidural fat which rarely causes compression of the spinal nerve structures. In most cases SEL is associated with obesity, chronic steroid therapy or some endocrinopathies.[1,6,7] Magnetic resonance (MR) imaging represent the reference standard for the IEL diagnosis.[7,8] The aim of the study is to describe a case of a patient with an axial compression fracture of the thoracic spine associated with an IEL. We also performed a systematic review of the literature in order to retrieve the key information regarding: the diagnosis, the clinical features and the treatment.

Methods

A systematic review of the literature indexed in PubMed, MEDLINE and Cochrane Library databases using as search-terms "*Extradural" OR "Epidural" AND "Lipoma*" AND "Spine" OR "Vertebral" OR "Spinal", and their mesh terms combinations was performed. The Preferred Reporting Items for Systematically Reviews and Meta-Analyses (PRISMA) was follow as reported in Figure 1. Only English publications were evaluated. The inclusion criteria of the review was the presence in the article of: demographic characteristics, onset symptoms, diagnostic information, implemented treatment, possible complications and outcomes in patients with IEL. Abstracts and full-texts were independently screened by two authors (A.P. and R.V.), any discordance was solved by consensus with a third author (F.C.T).

Case Report

A 46-year-old man was admitted to our emergency department following a high-velocity motor vehicle accident. He presented with severe axial back and chest pain worsening with the breath; Numerical Rating Scale score was 8/10. On clinical examination there was motor weakness (MRC 4/5) of both lower limbs, sensory reduction below T6 level and hyporeflexia in the lower limbs. Total body CT examination revealed sternum and ribs fractures, bilateral pleural effusion and mediastinal hematoma, as well as T5 and T6 vertebral body fractures due to an axial compression mechanism. Basing on CT images, we classified these fractures as A3 (incomplete burst fracture) according to the AO spine injury classification system. [9] MR imaging confirmed the T5 and T6 vertebral bodies fractures without associated ligamentous disruption and showed a posterior, rounded, encapsulated, epidural soft tissue mass with high signal on T1-weighted images, causing spinal cord compression at the same level of the fractured vertebral bodies (Figure 2). The spinal cord was anteriorly dislocated contacting the posterior aspect of the vertebral body. These MR features were suggestive of an epidural lipoma producing spinal cord compression. Basing on MR images, we reported anteroposterior diameter of the spinal canal (SpCa) and anteroposterior diameter of the epidural fat (EF). We calculated EF/SpCa*100% which was 51% (Figure 3), giving an idea of spinal canal stenosis due to the lipoma [2]. The presence of lipoma and neurologic symptoms led us to treat the patient with a surgical decompression and pedicle screw fixation. We performed a T5-6 laminectomy, an en-bloc resection of the lesion and fixation from T3 to T11. Pedicle screws were placed with a hybrid technique: percutaneous at the T7, T10, T11 levels [10-12] and free-hand open technique at T3 and T4 levels. [13] Pedicle screws were connected with two roads, bended according to the thoracic curve of the patient. The removed lesion was

Correspondence: Andrea Perna, Fondazione Policlinico Universitario A. Gemelli IRCCS, Roma, Università Cattolica del Sacro Cuore, Largo A. Gemelli 8, 00168, Rome, RM, Italy.
Tel.: +390630156638, +393278371443.
E-mail: perna.andrea90@gmail.com

Key words: spine; extradural lipomas; trauma; magnetic resonance imaging; stabilization; personalized medicine.

Contributions: GN: Data collection, text writing, figures processing; MB: Data collection, text writing, text formatting; RV: Study design, revision of the text, review of the literature; AP: Study design, text revision, review of the literature, bibliographic research; AL: Data collection, bibliographic research, Text revision, figures processing; NR: Data collection, figures processing; FCT: Study design, review of the literature, text revision.

Conflict of interest: the authors declare no potential conflict of interest.

Funding: none.

Availability of data and materials: All available data are present in the text.

Ethics approval and consent to participate: The study respects national ethical standards and the Helsinki Convention.

Informed consent: Written informed consent for scientific purposes and clinical data collection was obtained according to institutional protocol.

Received for publication: 11 April 2020.
Accepted for publication: 17 June 2020.

Orthopedic Reviews 2020; 12(s1):8684
doi:10.4081/or.2020.8684

yellow, soft, lobulated with a thin capsule. Histopathology showed normal fatty tissue with no evidence of atypia (Figure 4). The histological diagnosis was a true lipoma. The patient recovered without complications and was discharged 7 days after surgery with a custom-made orthopedic cast. Post-operative radiography and MR (Figure 5) were performed; the latter showed complete decompression of the spinal canal. The patient was followed-up at regular intervals of 15 days, 1, 3, 6 months, and 1 year after surgery. At 3-month follow-up, a CT of the thoracic spine showed a good placement of the pedicle screws without encroachment of the spinal canal and advanced healing of the fractures. At 6 months from surgery, the patient regained autonomy in all his Activities of Daily Living and remained stable at 1-year with complete recovery of daily activities.

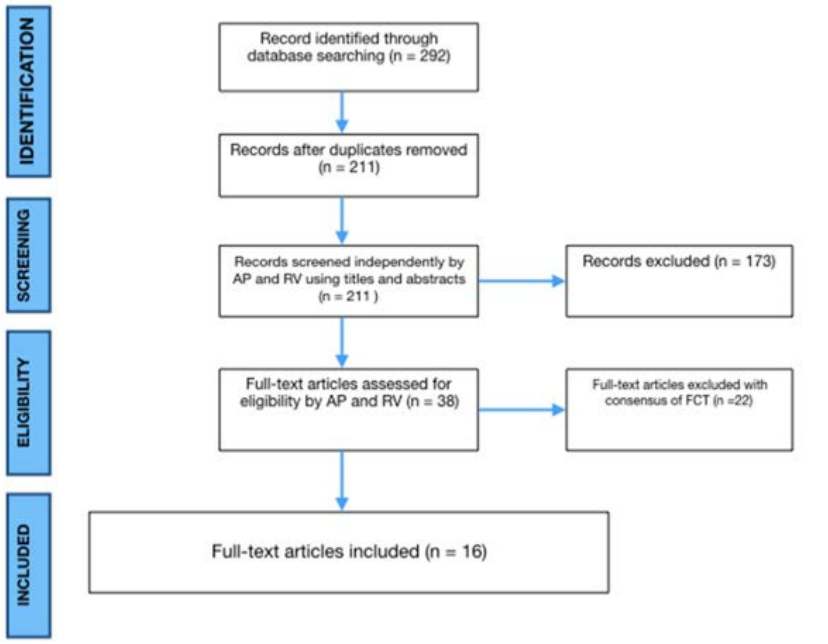

Figure 1. Preferred Reporting Items for Systematically Reviews and Meta-Analyses (PRISMA) flow-chart.

Discussion and systematic literature review

Background

Lipomas are widespread benign tumor with uncertain pathogenesis.[14] Their localization in epidural space of the spine are rarely observed in the absence of myelospinal dysraphism.[1] The presence of fat tissue into the epidural space should be distinguished in SEL and lipomatous masses (angiolipoma and IEL).[1,15] Spinal epidural angiolipomas are rare, benign tumors composed of both mature fatty tissue and abnormal vascular elements. These tumors can be categorized into two subtypes: non-infiltrating and infiltrating[16] and treatment is by total surgical excision.[17] The term lipoma,

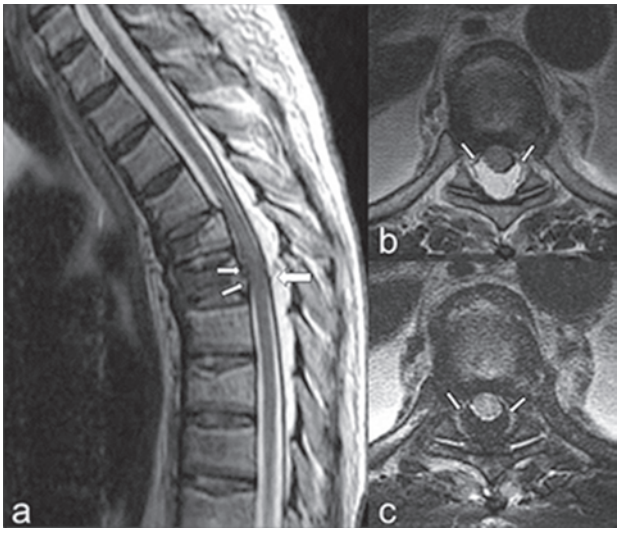

Figure 2. a) Sagittal T2-weighted MR image showing distraction of the PLC at T4-5 level (arrow) associated with mild retropulsion of the T5 and T6 posterior vertebral body wall (small arrows). b) Axial T2-weighted and c) corresponding axial T2-weighted fat-saturated MR images show the posterior, rounded, oblong encapsulated soft tissue mass with lipomatous signal intensity (small arrows).

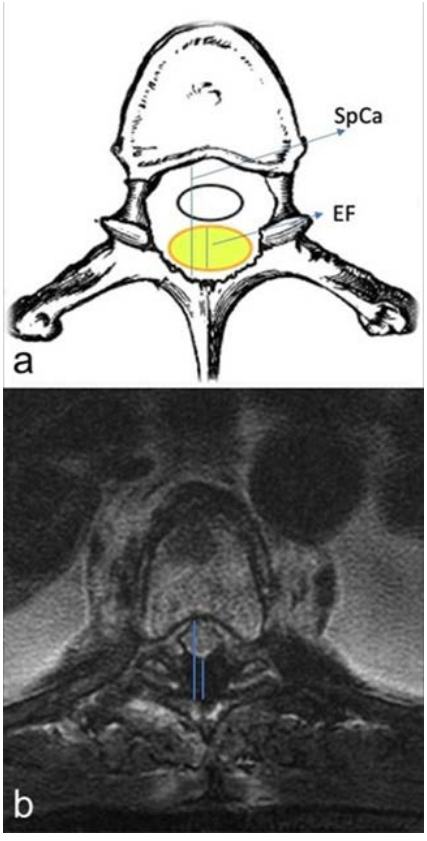

Figure 3. a) Schematic representation and b) axial T2-weighted MR images show the anteroposterior diameter of the spinal canal (SpCa) and anteroposterior diameter of the epidural fat (EF). Note: the calculated EF/SpCa*100% is 51%.

instead, should be reserved to well localized masses of fatty tissue with the same histological aspects of the lipoma of the other sites of the musculoskeletal system.[1]

Generally asymptomatic and occasionally discovered during an MR examination of the spine, IEL can become, much more rarely, the cause of neurologic impairment syndrome due to the compression of the neural elements inside the spinal canal particularly in the thoracic spine.[18] IEL as a cause of spinal compression must be considered in the differential diagnosis with a large variety of pathologies, specifically epidural haematomas and the most frequent metastatic vertebral tumors.[15,19]

Included patients and demographical data

Only a few cases of symptomatic epidural lipomas have been described. A total of 211 reports, excluding duplicates, were screened, thereby 16 were retrieved and finally included in our review (Figure 1). Numbers software (Apple Inc., Cupertino, CA) was used to tabulate the data obtained.

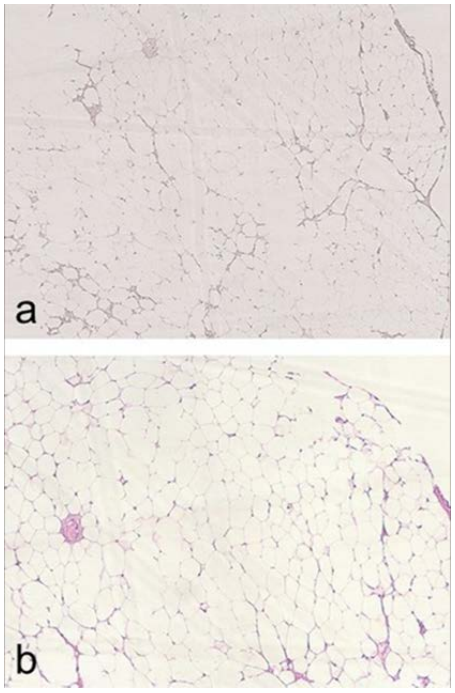

Figure 4. Microscopic examination (a, and b) reveals mature adipose tissue; the fat contains few small capillaries within thin fibrous stands and a thin fibrous strands.

Table 1. Review of the literature.

Study	Case	Sex	Age (Year)	Risk Factors	Symptoms	Level involved	Complication	Surgical treatment	Neurological Outcome	Follow Up (month)
Maier HC *et al.*. 1962	1	F	1	Intrathoracic lipoma	Paraparesis	D7	-	PDME	Complete recover	96
de Brüine JF *et al.* 1983	2	M	50	Obesity	BP, Sciatica, LW	L2-L3	-	PDME	Complete recover	24
Butti G *et al.* 1984	3	M	44	_	AW	C5-C6	_	PDME	Complete recover	60
	4	F	50	_	Paraparesis	L4-L5	_	PDME	Complete recover	24
Marks SM *et al.* 1985	5	M	43	_	BP, NC	L5	_	PDME	Complete recover	3
	6	M	60	_	BP, LW	L5-S1	_	PDME	Complete recover	10
	7	M	56	_	BP, Sciatica, LW	L3-L4	_	PDME	_	_
Meisheri YV *et al.*. 1996	8	M	20	_	Paraparesis	D6	_	PDME	No neurological improvement	3
Ashkan K *et al.* 2002	9	M	18	Klippel-Trenaunay-Weber syndrome	LW, UR	D5-D10	Sensory loss	PDME	Improvement	-
Subramaniam P *et al.* 2002	10	M	38	-	Paraparesis, AW	C5-D2	LW and AM	PDME	Improvement	18
Schizas C *et al.* 2003	11	F	80	Obesity, Lumbar trauma, L5 fracture	LW, UR	L5	Ankle deficit	PDME	Improvement	36
Park JS *et al.* 2005	12	F	60	Neck Mass	AW	C5-C6	-	APDME	Complete recover	12
Al-Zain F *et al.* 2008	13	M	27	_	Blurred vision, diplopia, BP, Papilloedema	L4-S3	_	PDME	Complete recover	4
Zevgaridis D *et al.* 2008	14	F	60	_	Sciatica, LW	L5	-	PDME	Complete recover	18
Iplikcioglu AC *et al.* 2008	15	M	45	_	-	D3-D7	-	PDME	Improvement	_
Kim HK *et al.* 2012	16	M	55	Obesity	Sciatica, LW	L5	_	PDME	Improvement	24
Missori P *et al.* 2012	17	F	49	_	LW	D6-D7	_	APDME	Improvement	32
Loriaux DB *et al.* 2015	18	M	38	_	Sciatica, LW	L5-S1	_	PDME	Improvement	10
	19	M	40	_	Sciatica, LW	L3-L4	-	PDME	Improvement	12
	20	M	44	_	Sciatica, LW	L4-L5	_	PDME	Improvement	12
Tateiwa D *et al.* 2018	21	M	76	_	Sciatica, LW	L4-L5	_	PDME	Improvement	12
Present case	22	M	46	Thoracic trauma, T5-T6 compression fractures	LW, BP	D5-D6	_	PDFME	Complete recover	12

APDME: Anterior and Posterior Decompression and Mass ExcisionAW: Arm Weakness BP: Back Pain; LW: Leg Weakness; NC: neurogenic claudication ; PDFME: Posterior Decompression with Fusion and Mass Excision; PDME: Posterior Decompression and Mass Excision; UR: urinary retention.

Only 22 cases, including our patient, of IEL were reported in literature.[1,14,15,18,20-30] The mean age of included patients was 45.5 (+/- 18.1) years, one patient was pediatric (<18yo); the Male/Female ratio was 2.6. The mean follow up time was 16.7 months (+/- 9.9). Demographic and clinical features are summarized in Table 1.

Localization, symptoms and diagnosis

IEL is prevalent in the lumbar segment (13 patients,58.7%), followed by the thoracic spine (6 patients, 27.5%) and the cervical spine (3 patients, 13.8%). Presentation symptoms were: lower extremity weakness in 13 (58.7%) patients, sciatica in 8 (36%), back pain in 5 (22.5%) and paraparesis in 4 (18%). Radiological diagnosis was obtained in 8 patient (36%) with myelography, in 14 patient (64%) with RM imaging. The reported predisposing conditions were: obesity in 3 patient (13.5%), vertebral fractures in 2 patients (9%), Klippel-Trenaunay-Weber syndrome in one patient (4.5%).

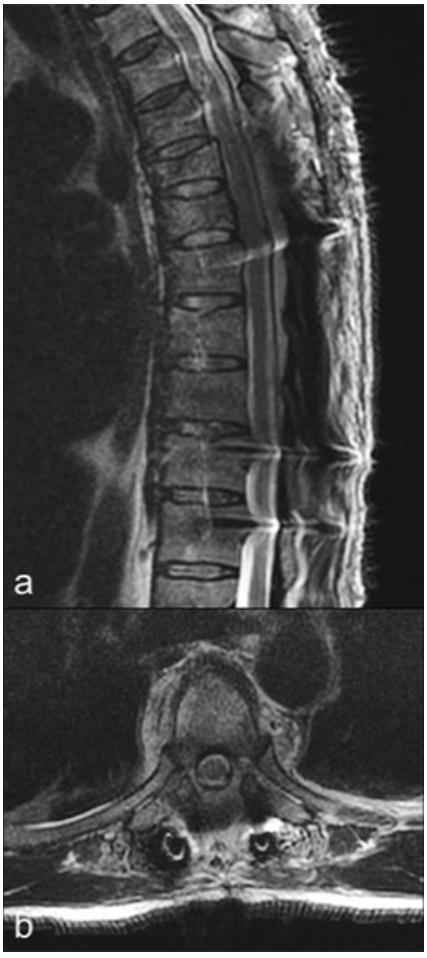

Figure 5. a) Sagittal and b) axial T2-weighted MR images show complete removal of the lipoma.

Treatment and outcomes

All patient were surgically treated: in 19 patient (85.5%) the chosen treatment was posterior decompression with en-bloc tumoral mass excision, in 2 patients (9%) anterior and posterior decompression with en-bloc tumoral mass excision, in one patient (4.5%) posterior decompression with en-bloc tumoral mass excision and trans-pedicle screw fixation due to associated post traumatic vertebral fractures. Ten patients (45%) completely recovered from the neurological deficit at the last follow-up visit; in 11 (49.5%%) patients only partial neurological recovery were reported, whereas one patient (4.5%) did not demonstrate any neurological recovery at the last follow-up visit.

Present case

In the reported case, MR imaging played an important role in the treatment decision making showing the epidural lipoma causing spinal cord compression at level of two thoracic vertebral fractures, and thus, necessitating decompressive laminectomy and en-bloc resection of the lesion in addition to a long posterior stabilization. We used a combined open-percutaneous approach in order to minimize the invasivity of a completely open approach and blood loss.[10-13]

Conclusions

In case of neurological symptoms associated with diagnosis of IEL, early surgical decompression with en-bloc excision of the tumor mass represent the treatment of choice. MR imaging represent the gold standard for radiological diagnosis of IEL. Finally, considering the concept of personalized medicine, the MR imaging allowed to obtain patient-tailored treatment with the highest safety in order to improve clinical outcomes and prevent further treatment.

References

1. Loriaux DB, Adogwa O, Gottfried ON. Radiculopathy in the setting of lumbar nerve root compression due to an extradural intraforaminal lipoma: a report of 3 cases. J Neurosurg Spine 2015;23:55–58.
2. Perna A, Ricciardi L, Barone G, et al. Medical management of acute non-specific low back pain: comparison of different medical treatments, one center's retrospective analysis. J Biol Regul Homeost Agents 2018;32:121-129.
3. Ricciardi L, Sturiale CL, Pucci R, et al. Patient-Oriented Aesthetic Outcome After Lumbar Spine Surgery: A 1-Year Follow-Up Prospective Observational Study Comparing Minimally Invasive and Standard Open Procedures. World Neurosurg 2019;122:e1041-6
4. Proietti L, Schirò GR, Sessa S, Scaramuzzo L. The impact of sagittal balance on low back pain in patients treated with zygoapophysial facet joint injection. Eur Spine J 2014:628-33.
5. Yang Z, Zhang Y, Xu D et al. Percutaneous vertebroplasty combined with interstitial implantation of 125I seeds in banna mini-pigs. World J Surg Oncol 2013;11:46
6. Vitiello R, Perisano C, Covino M, et al. Euthyroid sick syndrome in hip fractures: Valuation of vitamin D and parathyroid hormone axis. Injury 2020:30013-9.
7. Borré DG, Borré GE, Aude F, Palmieri GN. Lumbosacral epidural lipomatosis: MRI grading. Eur Radiol 2003;13:1709-21.
8. Brisson M, Kashima T, Delaney D et al. MRI characteristics of lipoma and atypical lipomatous tumor/well-differentiated liposarcoma: retrospective comparison with histology and MDM2 gene amplification. Skeletal Radiol 2013;42:635-47.
9. Vaccaro AR, Oner C, Kepler CK et al. AOSpine Thoracolumbar Spine Injury Classification System: Fracture Description, Neurological Status, and Key Modifiers. Spine 2013;38:2028–37.
10. Tamburrelli FC, Perna A, Proietti L, et al. The Feasibility of Long-Segment Fluoroscopy-guided Percutaneous Thoracic Spine Pedicle Screw Fixation, and the Outcome at Two-year Follow-up. Malays Orthop J 2019;13:39-44.
11. Proietti L, Scaramuzzo L, Schirò GR, et al. Posterior percutaneous reduction and fixation of thoraco-lumbar burst fractures. Orthop Traumatol Surg Res 2014;100:455-60.
12. Scaramuzzo L, Tamburrelli FC, Piervincenzi E, et al. Percutaneous pedicle screw fixation in polytrauma patients. Eur Spine J 2013;22:933-8.
13. Tamburrelli FC, Scaramuzzo L, Genitiempo M, Proietti L. Minimally invasive treatment of the thoracic spine disease: completely percutaneous and hybrid approaches. Minim Invasive Surg 2013;2013:508920.

14. Park JS, Shirachi I, Sato K, et al. Lipoma with dumb-bell extradural extension through the intervertebral foramen into the spinal canal. Case report. J Neurosurg Spine 2005;2:69-71.
15. Butti G, Gaetani P, Scelsi M, Pezzotta S. Extradural spinal lipomas. Report of two cases and review of the literature. Neurochirurgia (Stuttg) 1984;27:28-30.
16. Ghanta RK, Koti K, Dandamudi S. Spinal epidural angiolipoma: A rare cause of spinal cord compression J Neurosci Rural Pract 2012; 3:341-3.
17. Lin JJ, Lin F. Two entities in angiolipoma. A study of 459 cases of lipoma with review of literature on infiltrating angiolipoma. Cancer 1974; 34:720-7.
18. Schizas C, Ballesteros C, Roy P. Cauda Equina Compression After Trauma: An Unusual Presentation of Spinal Epidural Lipoma. Spine 2003;28:E148–51.
19. Tamburrelli FC, Meluzio MC, Masci G, et al. Etiopathogenesis of Traumatic Spinal Epidural Hematoma. Neurospine. 2018;15:101-7.
20. Maier HC. Extradural and intrathoracic lipoma causing spinal cord compression. Successful treatment by surgical excision. JAMA 1962;181:610-2.
21. Marks SM, Miles JB, Shaw MD. Idiopathic spinal extradural lipomas: Three cases and review of the literature. Surg Neurol 1985;23:153–6.
22. Meisheri YV, Mehta S, Chattopadhyay K. Acute paraplegia due to an extradural spinal lipoma: Case report. Spinal Cord 1996;34:633–4
23. Ashkan K, Moore AJ. Spinal cord compression caused by an extradural lipoma in Klippel-Trenaunay-Weber syndrome. Case illustration. J Neurosurg 2002;97:269.
24. Subramaniam P, Behari S, Singh S, et al. Multiple subpial lipomas with dumb-bell extradural extension through the intervertebral foramen without spinal dysraphism. Surg Neurol. 2002;58:338-43.
25. Al-Zain F, Gräwe A, Meier U. Papilloedema in association with spinal lipoma and bilateral chronic subdural bleeding. Spinal Cord 2008;46:392-4.
26. Zevgaridis D, Nanassis K, Zaramboukas T. Lumbar nerve root compression due to extradural, intraforaminal lipoma. An underdiagnosed entity? J Neurosurg Spine 2008;9:408–10.
27. Iplikcioglu AC, Hatiboglu MA, Ozek E, et al. Surgical removal of spinal mass lesions with open door laminoplasty. Cent Eur Neurosurg 2010;71:213-8.
28. Kim HK, Koh SH, Chung KJ. Solitary epidural lipoma with ipsilateral facet arthritis causing lumbar radiculopathy. Asian Spine J 2012;6:203–6.
29. Missori P, Pandolfi S, Antonelli M, Domenicucci M.Epidural neural fibrolipoma of the thoracic vertebral canal. J Neurosurg Spine 2012;17:449-52.
30. Tateiwa D, Yamasaki R, Ariga K, et al. An intraspinal extradural lipoma with spinal epidural lipomatosis: A case report and a review of literature. Surg Neurol Int 2018;9:212.