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

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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 Extraluminal lipoma (IEL), not associated with spinal dysraphisms, 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 extraluminal 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.2,3 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.1,4,5 Magnetic resonance (MR) imaging represent the reference standard for the IEL diagnosis.6,7 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 followed 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.8 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 levels10-12 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

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

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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.

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 myelo-spinal 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 infiltrating16 and treatment is by total surgical excision.17 The term lipoma,
Table 1. Review of the literature.

| Study            | Case | Sex | Age (Year) | Risk Factors | Symptoms                  | Level involved | Complication | Surgical treatment | Neurological Follow Up Outcome (month) |
|------------------|------|-----|------------|--------------|---------------------------|----------------|---------------|-------------------|----------------------------------------|
| Maier HC et al. 1962 | 1    | F   | 1          | Intrathoracic lipoma | Paraparesis             | D7             | -             | PDME              | Complete recover 96                     |
| de Bruijn IF 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 Y et al. 1996 | 8    | M   | 20         | Paraparesis     | D6                      | PDME           | Complete recover 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         | Obesity, Lumbar trauma, L5 fracture | LW, UR           | L5             | Ankle deficit | PDME              | Improvement 30                                 |
| Schiaza C et al. 2003 | 11   | F   | 80         | -             | Paraparesis, AW          | C5-D2          | LW and AM   | PDME              | Improvement 18                                 |
| 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                               |
| Zygargidis D et al. 2008 | 14   | F   | 60         | Sciatica, LW    | L5                       | -             | PDME              | Complete recover 18                     |
| Grilliciou NC 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         | PDME              | Complete recover 12                         |

PDME: Posterior Decompression with Mass Excision; APDME: Anterior and Posterior Decompression and Mass Excision; LW: Lower Weakness; BP: Back Pain; UR: Urinary Retention; 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. 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 13 (58.7%) patients, followed by the thoracic vertebral fractures, and thus, 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.[6-13]

**Treatement 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-pedicule 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.

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