Journal of Spine
ISSN: 2165-7939

Tofuku, J Spine 2014, 3:6
DOI: 10.4172/2165-7939.1000189

Case Report Open Access

Idiopathic Lumbar Epidural Lipomatosis

Katsuhiro Tofuku*

Department of Orthopaedic Surgery, Naruo Orthopedic Hospital, Japan

***Corresponding author:** Katsuhiro Tofuku, Department of Orthopaedic Surgery, Naruo Orthopedic Hospital, 12-24 Okadamachi, Chuoku, Kumamoto 862-0958, Japan, Tel: +81-96-371-1188; Fax: +81-96-372-2444; E-mail: tofuku@naruoseikei.com

Rec date: Nov 07, 2014, **Acc date:** Nov 12, 2014, **Pub date:** Nov 14, 2014

Abstract

We present two cases of idiopathic spinal epidural lipomatosis. Case 1: A 77-year-old man experienced tightness in bilateral lower extremities and neurogenic claudication for 1 year. He was overweight with a body mass index of 30.8 kg/m^2. Magnetic resonance imaging revealed extensive epidural lipomatosis of lumobsacral spine with significant constrictive compression of the thecal sac. Case 2: A 78-year-old man presented with low back pain along with progressive pain and numbness in the left lower extremity and neurogenic claudication for 6 months. He was diabetic with body mass index of 25.6 kg/m^2. Magnetic resonance imaging demonstrated circumferential compression of the thecal sac due to increased epidural fat tissue at the L4 level. We suggest that spinal epidural lipomatosis should be considered in the differential diagnoses when patients with neurological symptoms have conditions relating to hyperinsulinism such as obesity and exogenous steroid administration.

Keywords: Lumbar; Epidural lipomatosis; Obesity

Introduction

Spinal epidural lipomatosis (SEL) is a rare condition characterized by abnormal accumulation of unencapsulated epidural fat. Most patients with SEL are associated with the administration of exogenous steroids or several endocrinopathies including Cushing syndrome and hypothyroidism [1-9]. Idiopathic SEL, defined as cases without evidence of definite predisposing factors, seems to occur in obese patients but pathophysiology remains unclear [10-12]. We report two cases of idiopathic SEL with a review of literature.

Case Report

Case 1

A 77-year-old man experienced tightness in bilateral lower extremities and neurogenic claudication for 1 year. Over past few months, his gait disorder gradually worsened. He had no history of steroid use or endocrinopathy. On examination, he was overweight with a body mass index (BMI) of 30.8 kg/m2. He had no evidence of motor or sensory deficits. Magnetic resonance imaging (MRI) revealed extensive epidural lipomatosis of lumobsacral spine with significant constrictive compression of the thecal sac (Figure 1). An L4-L5 decompressive laminectomy with removal of the epidural adipose tissue was performed. The histopathological examination confirmed normal adipose tissue. The patient recovered completely.

Case 2

A 78-year-old man presented with low back pain along with progressive pain and numbness in the left lower extremity and neurogenic claudication for 6 months. He had diabetes but had no history of steroid administration or endocrinopathy. His BMI was 25.6 kg/m2. Neurological examination showed no muscle weakness and no sensory deficit. MRI demonstrated circumferential compression of the thecal sac due to increased epidural fat tissue at the L4 level (Figure 2). Histologic analysis revealed unencapsulated adipose tissue. An L4 laminectomy and removal of excessive adipose tissue was performed. After surgery, the patient's symptoms resolved.

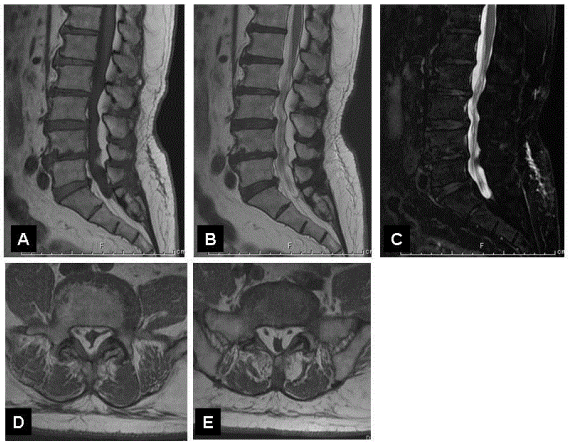

Figure 1: MRI showing increased epidural fat tissue, iso signal intensity to the subcutaneous fat. **A:** Sagittal T1-weighted images. **B:** Sagittal T2-weighted images. **C:** Sagittal fat suppressed T2-weighted images showing suppression of the fat signal. **D:** Axial T1-weighted images at the level of the upper edge of the L5 vertebral body. **E:** Axial T1-weighted images at the level of the upper edge of the S1

Discussion

Exogenous steroid administration and endocrinopathies such as Cushing syndrome and hypothyroidism are known predisposing factors for SEL [1-12]. Widespread use of exogenous steroid, increase in the prevalence of obesity with improving diagnostic modalities should have been the reasons for growing reports on SEL. Although the pathological mechanism of SEL still remains unknown, insulin seems to play an important role. The association of obesity and

diabetes in our patients suggests a metabolic syndrome associated with insulin resistance, leading to hyperinsulinism. Steroid also accelerates secretion of insulin, leading to inhibition of lipolysis. It is proposed that hyperinsulinism caused by metabolic syndrome or steroid intake inhibits mobilization of fatty acids (lipolysis) of the central adipose tissue at the site of abdominal or spinal epidural space.

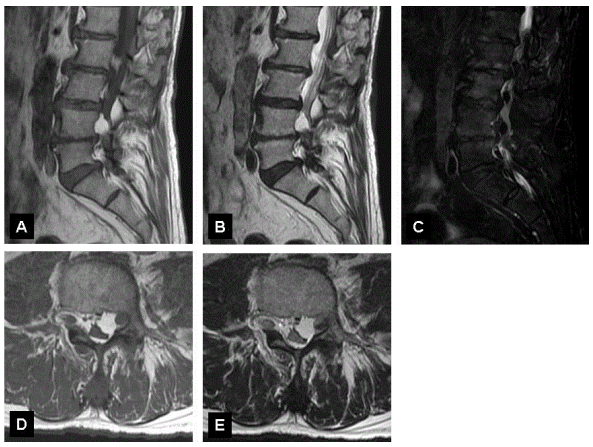

Figure 2: MRI showing deformation of the dural sac compressed by an excessive deposition of epidural fat tissue. **A:** Sagittal T1-weighted images. **B:** Sagittal T2-weighted images. **C:** Sagittal fat suppressed T2-weighted images. **D:** Axial T1-weighted images at the L4 level. **E:** Axial T2-weighted images at the L4 level

The symptoms depend on the level of canal compromise, with thoracic levels causing myelopathy and at lumbar levels resulting in radiculopathy or cauda equina syndrome. In general, the onset of symptoms is gradual and the condition progresses slowly. Some reported cases with acute onset or rapid progression of symptoms might be associated with vascular dysfunction such as epidural venous engorgement and bleeding [13]. Compression of the thecal sac could be clearly demonstrated on MRI images. A typical feature could be found on axial MRI images, where the overgrowth adipose tissue results in circumferential compression of the thecal sac, referred to as the "Y-sign" or "Stellate- sign" [14]. Epidural adipose tissue that has a thickness greater than 7 mm has been reported to be the diagnostic criterion for SEL [4,11].

The treatment of SEL depends on the severity of neurologic manifestations. Surgical treatment consisting of decompressive laminectomy with excision of epidural adipose tissue is required in patients who are refractory to the conservative treatment. Prognosis with surgical management looks favorable and no case of recurrence has been reported [15]. Conservative treatment including weaning of steroids and weight loss is recommended for patients without significant neurological deficits. Weight loss has been reported to be beneficial in obese patients [10-12]. Non-obese patients not receiving steroids should undergo endocrinological evaluation.

We suggest that SEL should be considered as a differential diagnosis when patients presenting with neurological symptoms have conditions pertaining to hyperinsulinism, may it be obesity or exogenous steroid administration. Early detection of SEL and conservative management including diet and specific insulin resistance treatment might avoid the need for surgical management.

References

1. Guegan Y, Fardoun R, Launois B, Pecker J (1982) Spinal cord compression by extradural fat after prolonged corticosteroid therapy. J Neurosurg 56: 267-269.
2. Fessler RG, Johnson DL, Brown FD, Erickson RK, Reid SA, et al. (1992) Epidural lipomatosis in steroid-treated patients. Spine (Phila Pa 1976) 17: 183-188.
3. George WE Jr, Wilmot M, Greenhouse A, Hammeke M (1983) Medical management of steroid-induced epidural lipomatosis. N Engl J Med 308: 316-319.
4. Roy-Camille R, Mazel C, Husson JL, Saillant G (1991) Symptomatic spinal epidural lipomatosis induced by a long-term steroid treatment. Review of the literature and report of two additional cases. Spine 16: 1365-1371.
5. Haddad SF, Hitchon PW, Godersky JC (1991) Idiopathic and glucocorticoid-induced spinal epidural lipomatosis. J Neurosurg 74: 38-42.
6. Bodelier AG, Groeneveld W, van der Linden AN, Haak HR (2004) Symptomatic epidural lipomatosis in ectopic Cushing's syndrome. Eur J Endocrinol 151: 765-769.
7. Noël P, Pepersack T, Vanbinst A, Allé JL (1992) Spinal epidural lipomatosis in Cushing's syndrome secondary to an adrenal tumor. Neurology 42: 1250-1251.
8. Sivakumar K, Sheinart K, Lidov M, Cohen B (1995) Symptomatic spinal epidural lipomatosis in a patient with Cushing's disease. Neurology 45: 2281-2283.
9. Chapman PH, Martuza RL, Poletti CE, Karchmer AW (1981) Symptomatic spinal epidural lipomatosis associated with Cushing's syndrome. Neurosurgery 8: 724-727.
10. Borstlap AC, van Rooij WJ, Sluzewski M, Leyten AC, Beute G (1995) Reversibility of lumbar epidural lipomatosis in obese patients after weight-reduction diet. Neuroradiology 37: 670-673.
11. Robertson SC, Traynelis VC, Follett KA, Menezes AH (1997) Idiopathic spinal epidural lipomatosis. Neurosurgery 41: 68-74.
12. Maillot F, Mulleman D, Mammou S, Goupille P, Valat JP (2006) Is epidural lipomatosis associated with abnormality of body fat distribution? A case report. Eur Spine J 15: 105-108.
13. Kawai M, Udaka F, Nishioka K, Houshimaru M, Koyama T, et al. (2002) A case of idiopathic spinal epidural lipomatosis presented with radicular pain caused by compression with enlarged veins surrounding nerve roots. Acta Neurol Scand 105: 322-325.
14. Kuhn MJ, Youssef HT, Swan TL, Swenson LC (1994) Lumbar epidural lipomatosis: the "Y" sign of thecal sac compression. Comput Med Imaging Graph 18: 367-372.
15. Fogel GR, Cunningham PY 3rd, Esses SI (2005) Spinal epidural lipomatosis: case reports, literature review and meta-analysis. Spine J 5: 202-211.