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Epidural lipomatosis with cauda equina syndrome in chronic alcoholic patient: A case report

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ABSTRACT

INTRODUCTION: Epidural lipomatosis of the lumbar spine is a rare condition, which is described as the accumulation of fat in the extradural territory.**PRESENTATION OF CASE:** We report the case of a 60-year-old, non-obese, and chronic alcoholic man who was transferred to our spine department with cauda equina syndrome (CES) for 4 months. On magnetic resonance imaging (MRI), spinal epidural lipomatosis (SEL) was confirmed in the multilevel lumbar lesion. A decompression surgery was performed and the patient recovered significantly.**DISCUSSION:** The patient was not obese, had no abnormal liver laboratory test results, and no history of steroid injection or administration. The clinical signs at onset suggested bilateral lower cauda equina dysfunction, indicating a more diffuse involvement, consistent with lumbosacral epidural lipomatosis.**CONCLUSION:** This case report is the first description of SEL in a non-obese, chronic alcoholic patient who was neither receiving steroids nor had any kind of endocrinopathy.© 2017 The Author(s). Published by Elsevier Ltd on behalf of IJS Publishing Group Ltd. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

1. Introduction

Epidural lipomatosis of the lumbar spine is a rare condition, described as the accumulation of fat in the extradural territory [1]. This is usually secondary to an uncommon complication of long-term iatrogenic corticosteroid administration, endocrinological disorder, or obesity [2,3]. If neurological deficits develop, it is usually in the thoracic spine more than in the lumbar spine [1,4]. Spinal epidural lipomatosis (SEL) with cauda equina syndrome (CES) is not a common clinical condition [5,6]. Madelung disease (benign symmetrical lipomatosis) is diffuse deposits of fat arranged symmetrically in upper extremities, around the neck and shoulders. Middle-aged men who have a history of decades of heavy alcohol consumption occurred more frequently. Mechanism of etiology is assumed to be a malfunction in fat metabolism due to damage to mitochondrial DNA [7]. Here, we present a severe case of spinal epidural lipomatosis with cauda equina syndrome in a chronic alcoholic patient.

This article has been written according to SCARE criteria as described by Agha et al. for the SCARE group. 'The SCARE Statement: Consensus-based surgical case report guidelines. International Journal of Surgery 2016' [8].

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2. Case report

A 60-year-old non-obese, alcohol dependent man was transferred to our spine department with 4 months of progressive lower extremity weakness, neurogenic claudication with bladder bowel dysfunction, and hypoesthesia in the perineal region. He had a history of over four bottles of alcoholic drinks per day for 3 decades. He did not receive steroid medications or injection for other chronic illnesses nor had any definite liver disease, and was evaluated using plain radiography, computed tomography (CT), and magnetic resonance imaging (MRI). During admission to our hospital, he was not obese (weight 68 kg, height 178 cm, body mass index (BMI) 21.5 kg/m²). The MRI results demonstrated a pathological overgrowth of fat tissue in the spinal canal with a marked impingement of the dural sac, which showed severe central canal stenosis with space occupying epidural fat in the lumbar lesion (Fig. 1). Both leg and ankle weakness (power 2–3/5), and decreased sensation below L2 dermatome level were observed. Sphincter tone was decreased and saddle anesthesia was observed. L2/3 and L3/4 decompression and fat removal with partial laminectomy (Fig. 2), and L4/5 and L5/S1 total laminectomy with posterolateral fusion with pedicle screw fixation were performed without delay. After surgery, the patient showed delirium tremens and underwent additional treatment with a psychiatrist. Histopathological examination showed the presence of normal fat cells, in the absence of venous engorgement, hypervascularization, or bleeding (Fig. 3). With gradual improvement in the symptoms and signs, the patient returned to daily ambulatory activities with cane and almost recovered



Fig. 1. T1 weighted and short T1 inversion recovery (STIR) sequence MRI imaging: white arrow points to the epidural lipomatosis.

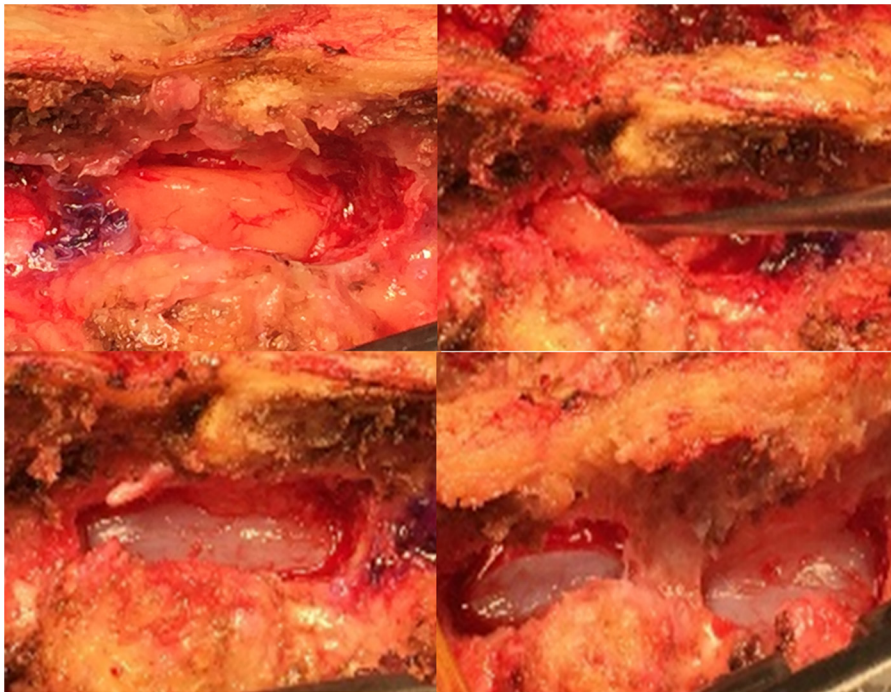


Fig. 2. Partial laminectomy and removal of hypertrophic fat tissue performed during surgery.

neurologically except for residual saddle anesthesia and voiding dysfunction at the 21-month follow-up.

3. Discussion

Epidural fat is soft and vulnerable; however, SEL is characterized by abnormal accumulation of unencapsulated fat in the epidural

space, which can cause a compressive CES [5,9]. In previous case reports, SEL was induced in most patients by steroids, obesity, or idiopathic causes [10,11].

SEL was first reported in 1974 with the use of steroids to prevent rejection reaction after a kidney transplant [12]. Thereafter, several reports have attributed the development of SEL to the administration of epidural steroid injections [13,14]. It has been reported that although the dose of steroid may be prescribed based on the time

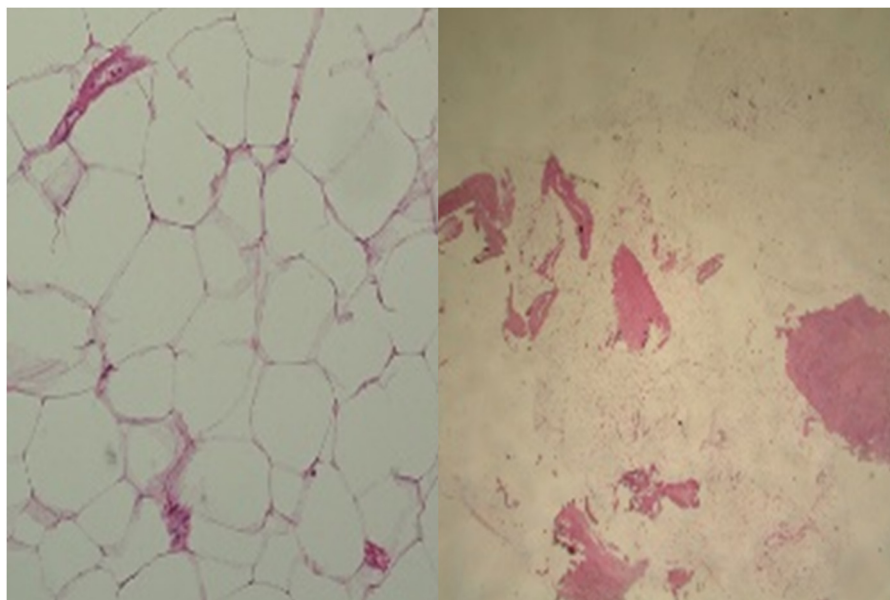


Fig. 3. Histopathology confirms benign adipose fat tissue under a microscope.

to onset of neurological symptoms, the mental acuity and neurological deterioration is not related to the rate or degree of recovery [10].

SEL is also associated with obesity and is the principal risk factor associated with lumbar epidural lipomatosis. It is thought that the obese state may be associated with elevated cortisol levels, with three times more likelihood of developing SEL in the lumbar region [2,15]. In this case, the patient had normal BMI, no abnormal liver laboratory test results, and no history of steroid injection or administration.

Although some cases of SEL are idiopathic, not many cases [4,16,17] demonstrate SEL in patients with no other suspected causative factors.

As expected, there is an intimate relationship between alcohol and fat deposition in several studies, which is convincing since SEL and metabolic syndrome share many components. A study on the physiology of cellular liporegulation in rodents sheds some light on the role of leptin in human metabolism of fat. If leptin is dysfunctional, then deposition of fat may take place in the liver, heart, muscles, and pancreas causing lipotoxicity or lipoapoptosis [2,18]. However, some studies have shown a beneficial effect of alcohol on the development of metabolic syndrome. In a study by Baik et al. [19], an increased risk of metabolic syndrome was associated with “obese, heavy liquor drinking persons”. Mild or moderate alcohol consumption may be protective, but only in persons who are not obese.

The clinical presentation of CES is usually gradual lower back pain, pain or sensory deficit in the perineum, sensorimotor signs of the lower extremities, and impaired autonomic function of the bowel and bladder. CES with lipomatosis often lasts for months to several years, followed by progressive or sudden neurological deficits [5,9,20]. In some patients, it may be difficult to distinguish CES from a conus medullaris syndrome, which is usually symmetric, spares leg function, and has a sudden onset [5]. In this case, the clinical signs at onset were indicative of bilateral lower cauda equina dysfunction, suggesting a more diffuse involvement, consistent with the final diagnosis of lumbosacral epidural lipomatosis.

MRI is the best imaging tool of choice. Compression with obliteration of the dural sac can be appreciated on the axial and sagittal MR images, and high-signal intensity on T1-weighted images and intermediate signal intensity on T2-weighted images are character-

istics of the adipose tissue. Moreover, short T1 inversion recovery (STIR) sequence may be useful for confirmation of the diagnosis as a lipid is hypointense in this sequence [19]. In our patient, STIR sequence was performed for confirming the diagnosis (Fig. 1).

The treatment of SEL may follow either a conservative therapy or surgery, depending on the severity of the clinical symptoms, underlying cause in the secondary forms, or presence of idiopathic SEL. Weight loss is recommended if the condition is associated with obesity, and a reduction in the steroid dose is recommended if SEL is associated with exogenous steroid use [21]. Most often, surgery, including laminectomy or resection of adipose tissue, is performed if the symptoms are rapidly progressive [16,22].

4. Conclusion

This case report is the first description of SEL in a non-obese, chronic alcoholic patient who was neither receiving steroids nor had any kind of endocrinopathy. This rare symptom may be a sign of severe cauda equina compression in a chronic alcoholic patient, and we recommend decompressive surgery with predictable good results.

Conflicts of interest

The authors have no conflict of interests to declare.

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This work was supported by a grant from Research 6 months of Inje University.

Ethical approval

Does not apply.

Consent

Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

Author contribution

Sung-Soo, Kim – study concept, design.

Dong-Ju, Lim – data collection, data analysis or interpretation, writing the paper.

Guarantor

Dong-Ju, Lim.

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