CASE REPORT

Idiopathic spinal epidural lipomatosis without obesity: a case with relapsing and remitting course

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Study design: Case report of a rare form of idiopathic spinal epidural lipomatosis (SEL) manifesting with relapsing and remitting course.

Objective: To describe this very rare clinical and pathological condition and the results of surgical intervention.

Setting: A department of neurosurgery in Morocco.

Methods: A 24-year-old man presented with a 2-year history of mid-thoracic back pain and progressive neurogenic claudication with two episodes of remitting and relapsing course. Spinal magnetic resonance imaging revealed a fatty epidural mass extending from T4 to T9. Posterior decompression was performed, and he was followed for 3 years after the operation.

Results: Pathological examination of the surgical specimen revealed nodules of mature fat cells without neoplasm. The patient's symptoms completely resolved after surgical decompression.

Conclusion: Fluctuating clinical course has not been reported previously in SEL. Such pathology should be considered in the differential diagnosis of demyelinating diseases.

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Introduction

Spinal epidural lipomatosis (SEL) is a rare disease, defined as a pathological overgrowth of normal extradural fat.¹ It occurs in patients with exogenous steroid treatment, endogenous Cushing's syndrome, endocrinopathies or morbid obesity.^{1–3} Idiopathic SEL in non-obese patients is exceptional.⁴ We report an additional case, which is the only one presented fluctuating symptomatic course.

Case report

A 24-year-old man presented with a 2-year history of mid-thoracic back pain. He reported recently paresthesias in the lower limb extremities with progressive weakness lastly for 3 months duration. The pain restricted progressively his walking distance to 100 m. He denied sphincter dysfunction. He had no history of trauma and no other significant medical history apart from two episodes of leg numbness, which occurred 18 and 6 months ago; these symptoms resolved progressively and completely within few days. Physical examination revealed a non-obese patient (73 kg, 1.72 m) without any signs of Cushing's syndrome. Motor and sensory

function was normal but deep tendon reflexes of the lower extremities were hyperactive with bilateral Babinski's sign.

Spinal plain radiographs were unremarkable. Spinal magnetic resonance imaging (MRI) showed a posterior extradural mass, 8-mm thick, extending from T4 to T9 segments with mild compression of the thecal sac. This mass was homogeneously hyperintense in both T1 and T2-WI (Figure 1). Blood, biochemical and urine analyses were unremarkable. The patient was discharged with a prescription for analgesics and rehabilitation therapy. Four months later, he still had medullary claudication with a walking distance of less than 40 m. MRI features were unchanged.

A bilateral T4-T9 laminectomy was performed. Intraoperative findings revealed that the epidural space was filled by a thickness non-capsulated fatty lobular mass, which compressed the dura; some venous engorgement has been seen. After an easily complete resection, the thecal sac becomes pulsatile. Microscopic tumor studies showed nodules of mature fat cells. Post-operatively, the weakness of both legs improved remarkably. The patient remains neurologically intact and free of pain for more than 3 years follow-up.

Discussion

SEL may occur in some previously healthy patients in the absence of predisposing factors but obesity has been related

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Figure 1 Thoracic spinal magnetic resonance imaging. Axial T2-WI (a) sagittal T1 (left) and T2-WI (right) (b) showing a posterior epidural mass with mild compression of the dura extending from T4 to T9 segments. This mass was hyperintense at both T1 and T2-WI similar to that of subcutaneous adipose tissues.

to this idiopathic form.^{1,3,4} About 16 cases of idiopathic SEL in non-obese patients were reported previously.¹ Thoracic and lumbar canal were equally involved. Usually, the onset of symptoms was gradual with slow progression of neurological features for several months. Direct mechanical compression of the nervous structures and indirect vascular dysfunction (compression of the epidural blood vessels with venous engorgement) contribute to the evolution of the myelopathy.

Our patient's presentation was very unusual because he exhibited a relapsing clinical course with numbness of lower limbs that recovered completely after a few days mimicking a demyelination disease (such as multiple sclerosis). This similar fluctuating course was never seen in SEL, but reported previously in some cases of spinal epidural angiolipoma.⁵ The pathogenesis is probably multifactorial; venous stasis with thrombosis have been suggested as precipitin syndrome.

MRI demonstrated increased posterior spinal epidural fat causing variable anterior compression of the cord. MRI also measured the extradural fat thickness, which was more than 7 mm (normally, less than 6 mm).^{1,3} Administration of gadolinium may allow differentiation between epidural lipomatosis (which does not enhance) and spinal angiolipoma.^{2,5}

The treatment depends on the severity of the neurologic manifestations. In some reports,² there have been favorable response after weight reduction in some obese patients. However, long-term follow-up results are not available. Surgical decompression is the treatment of choice in symptomatic patients in whom nonsurgical therapies have failed or in those patients who show signs of myelopathy.^{1,2,4} The prognosis of patients with idiopathic SEL after surgical management is favorable and no case of recurrence has been reported.¹

Conclusions

Fluctuating clinical course has not been reported previously in SEL. Such pathology should be considered in the differential diagnosis of demyelinating diseases. The early diagnosis is essential, since in patients with minor symptoms conservative treatment may be sufficient, while in progressive disorders surgical decompression can produce more rapid relief of signs and symptoms without recurrences.

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