

Case report

Neurogenic claudication by epidural lipomatosis:
a case report and review of literature

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Abstract

Epidural lipomatosis is most frequently seen in patients on chronic steroid treatment. Only twelve cases of idiopathic spinal epidural lipomatosis have been described. In this report we present an additional case of this condition in a middle-aged male presenting with neurogenic claudication.

Keywords: Epidural lipomatosis; Steroid therapy; Neurogenic claudication; MRI

1. Introduction

Spinal epidural lipomatosis is a rare condition defined by pathologic accumulation of fat in the extradural space of the vertebral canal; generally it is associated with the administration of exogenous steroids and has been reported in patients receiving immunosuppressive therapy after organ transplantation [1–3] and in patients requiring steroids for systemic disease (e.g. SLE [4,5], dermatomyositis [6], rheumatoid arthritis [7], chronic obstructive pulmonary disease [3,8]). Epidural lipomatosis is uncommon in patients with an endogenous elevation of steroids, as in Cushing's syndrome [9].

There are few reports of this syndrome occurring in the absence of steroid treatment. It has been described in hypothyroidism [10], in morbid obesity [11], and in 2 patients undergoing radiotherapy for bronchial carcinoma [8] and testis seminoma [12].

We present a new case of this condition in a patient without endocrine disease and review the literature on idiopathic spinal epidural lipomatosis.

2. Case report

A 52-year-old male, presented with an almost 2-year history of back pain and unilateral neurogenic claudication: He described pain, irradiating to the lateral side of the upper right leg. During the period of complaints the pain was progressive and associated with numbness and tingling. After walking 50 meters the pain became so severe that he was forced to sit and rest. The leg symptoms were alleviated by sitting or resting. There were no complaints of bladder or bowel dysfunction.

His medical history was negative for endocrinopathy, and he had not received any medication prior to the onset of symptoms. In 1953 an appendectomy had been performed, and in 1990 he was operated for an abscess in the right inguinal region.

Examination

Physical examination revealed a nervous man, in no acute distress. Body length was 1.75 m and weight 89 kg. Blood pressure, pulse and respiration were normal. Spine examination revealed a severe torsion scoliosis and fixation of the lumbar spine. Straight leg raising test was positive at 45 degrees on the right, negative on the left. There was no muscular weakness or loss of sensibility. The knee reflex and achilles tendon reflex were slightly

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diminished on the right side. The peripheral pulses were normal symmetrical and there was a good capillary refill.

Except for hypercholesterolemia (8.6 mmol/l) and hypertriglyceridemia (2.88 mmol/l), laboratory investigations were normal, including normal levels for thyroxine, TSH, cortisol, testosterone and ACTH.

Neuroimaging

The myelogram demonstrated epidural compression behind the corpus of L4, with complete myelographic block (Fig. 1). The compressing mass was just behind the posterior wall of the 4th lumbar vertebral body, and anterior to the dura. CT-scan revealed an epidural mass, with the characteristic density of fat. Transversal (Fig. 2) and sagittal (Fig. 3a) MRI demonstrated an epidural mass, beginning at the level of the upper side of the 3th lumbar vertebral body, and expanding downwards to the level of the second sacral vertebra. It surrounded the thecal sac, occupying more than 50% of the spinal canal. The lesion showed increased intensity on both T1- and T2-weighted images, compatible with fat.

Surgical procedure

A decompressive laminectomy from L2 to S1 was performed. After exposure an abnormal accumulation of



Fig. 1. Myelographic images show nonspecific narrowing and total compression of the thecal sac behind the corpus of L4.



Fig. 2. Transversal T1-weighted MRI through the body of L4, showing epidural fat with high signal intensity surrounding the thecal sac.

fatty tissue presented over the entire exposed trajectory, also encasing the nerve roots L5, S1 and S2. Proximal to the disc L2-L3 normal epidural fat was discerned. Debulking of the fatty tissue was performed with the CUSA, and after decompression the dural sac was seen to expand and the initially absent pulsations returned. Histologic sectioning confirmed normal adipose tissue.

Postoperative course

During the initial postoperative period there was no improvement. Two months postoperatively the complaints of back and right leg pain had disappeared, but he developed symptoms of the left leg, suspect for meralgia paraesthetica. Shortly thereafter however the symptoms on the right recurred, although less intense than before the procedure, while the pain in the left leg disappeared completely. Control MRI (Fig. 3b) performed seven months postoperatively, showed some remnant epidural lipomatosis anterior to the dura. There were no signs of intervertebral disc disease or lumbar stenosis.

3. Discussion

Although spinal epidural lipomatosis is a rare disorder, it has been well described in the literature. The etiology of the disease is almost invariably secondary to exogenous corticosteroid administration. Duration and dosage of corticosteroid administration varies in different reports, suggesting a threshold for the hypertrophic development of epidural fat. Fessler et al. [3] remark that epidural lipomatosis should be considered in patients with low back pain, leg pain, and/or radicular symptoms, when there is a history of corticosteroid use, even when this history is of short duration.

The diagnosis of epidural lipomatosis can be established by myelography, CT scanning and MRI. Myelography can reveal the epidural nature of the process and

the level of obstruction, but can not identify the specific cause. The characteristic low density of fat on CT enables an accurate diagnosis, but does not show the relationship between the epidural lipomatosis and the anatomical structures surrounded by this fat. MRI is considered the imaging procedure of choice [13,14]: a high contrast between fat and the thecal sac permits an accurate evaluation of the extent of the lipomatosis.

Only 14 cases of epidural lipomatosis have been reported in the absence of corticosteroid use. Badami and Hinck [11] suggest that morbid obesity may be a predisposing factor, but this still remains to be proven. Toshniwal and Glick [10] reported a case of epidural lipomatosis secondary to hypothyroidism, in which they pointed out that lower levels of thyroid hormone reduce the rate

of lipolysis. It would be of great interest to study the presence of clinical relevant abnormal fat in the spinal canal in patients with thyroid deficiency state. The remaining 12 cases are without endocrinopathy; Table 1 summarizes the characteristics of these patients, including the 13th case described by us.

Twelve of the patients described were male, with a mean age of 49.8 years (range 18–75). The lipomatosis involved at least 2 levels, affecting in 8 cases the lumbosacral spine, and in 5 cases the thoracic spine. Mean body mass index ($\text{weight}/[\text{height}]^2$) was 30.8 kg/m^2 with a range of $27.7\text{--}36.5 \text{ kg/m}^2$; this is above the limit of 27.5 kg/m^2 , which is the National Institutes of Health's definition of obesity.

In cases of steroid administration Butcher [16] and

Table 1
Clinical summary of reported cases of idiopathic spinal epidural lipomatosis. LE = lower extremity.

Author (ref.)	Sex, age (yrs)	Level of radiographic compression	Symptoms	BMI (kg/m^2)	Treatment	Outcome
1. Quint et al. [8]	M, 34	Th4-8	Th-6 radicular pain, back pain		Laminectomy, fat debulking	Improved pain Returned to work
2. Stambough et al. [17]	F, 54 black	L4-5	LE pain, back pain, neurog. claudicatio	34.1	Laminectomy, fat debulking	Near complete-relief of symptoms/pain
3. Bednar et al. [15]	M, 57	L4-S1	LE pain, back pain, motor weakness of lower limb		Exploration after transection of lig. flavum, blunt resection of fat. No bone resection.	Complete relief of complaints
4. Gero and Chynn [18]	M, 64 black	L3-5	LE pain, radicular distribution		Laminectomy, fat debulking	Not specified
5. Haddad et al. [4]	M, 40	Th1-10	Th-5 radicular pain	31.8	Weight reduction, diet with 15 kg weight loss over 1 year	Improved pain
	M, 18	Th6-8	Back pain, LE numbness, gait difficulty	29.2	Laminectomy, fat debulking	Improved pain
	M, 54	L5-S1	LE pain, decreased sensation	27.7	Laminectomy, fat debulking, intradural exploration	Improved pain pseudomeningocele necessitating 2 reoperations
8. Beges et al. [13]	M, 59	Below L3	Leg weakness neurog. claudicatio	36.5	Weight reduction, diet with 19 kg weight loss over 8 weeks	Relief of complaints
9. v. Rooij et al. [14]	M, 49	L4-L5	Backpain, LE pain, Numbness	29.7	Laminectomy, fat removal	Complete relief of complaints
	M, 41	L4-S1	Back pain, LE pain	28.6	Weight reduction, diet with 23 kg weight loss	Complete relief of complaints
11. Stern et al. [19]	M, 51	Th1-10	Bilateral LE numbness, weakness		C7-Th11 laminectomy 24 mo follow-up	Marked improvement
	M, 75	Th6-8	LE weakness, clonus, spastic gait, urinary incontinence		Th6-8 laminectomy	Resolution of incontinence, improved LE strength, 24 mo follow-up
13. Kurt and Bakker (this report)	M, 52	L3-S2	LE pain, back pain, neurog. claudicatio	29.1	Multilevel laminectomy, fat debulking with CUSA	Initially improved pain, later recurrence



Fig. 3a (left) and 3b (right). Sagittal T1-weighted MRI pre-operative (left) and post-operative 7 months (right). Before operation there is an abnormal amount of epidural mass, beginning at the level of the 3th lumbar vertebral body, and expanding downwards. The post-operative MRI demonstrates adequate decompression and reduction of the epidural fat.

later George [1] described that reduction in the dose of steroids can result in improvement of the clinical symptoms. In cases of idiopathic epidural lipomatosis, 2 modalities of treatment exist: conservative management with weight reduction and operative therapy consisting of decompressive laminectomy and fat debulking. In the patients described with idiopathic spinal epidural lipomatosis ten underwent operative treatment and three were treated conservatively. Haddad [4] was the first who described a patient with complete resolution of his Th-5 radicular pain after losing 13.5 kg in a year, and later Beges [13] and Van Rooij [14] confirmed that weight reduction with a low calory diet may be successful. In our patient with an almost a 2-year history of complaints there was a clear delay in time before the correct diagnosis was established and the patient presented to the neurosurgeon. This may be one of the factors causing residual complaints in the patient described, but the operative therapy can insofar be considered successful that the progression of symptoms was halted and the development of a cauda syndrome prevented. The etiology of the disease in our patient remains unknown.

4. Conclusion

In patients with spinal epidural lipomatosis the accumulation of epidural fat is an ongoing process, that may cause a broad scale of symptoms over time. Early recognition of the disorder creates the possibility to intervene at an early stage. Although conservative management with weight reduction can be attempted, it is our opinion

that surgical decompression should be considered and in any case not delayed too long, because of the significant risk of irreversible progression of neurological deficit.

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