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Is epidural lipomatosis associated with abnormality of body fat distribution? A case report

Received: 23 November 2004
Revised: 15 February 2005
Accepted: 19 April 2005
Published online: 9 July 2005
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Abstract Case report: To report a case of epidural lipomatosis in a patient with abnormal adipose tissue distribution, glucose intolerance and mixed hyperlipidemia. A 63-year-old male patient presented with low back pain radiating to the left calf on standing and walking (walking distance < 100 m). He weighed 97.5 kg, was 1.73 m tall (BMI 32.6 kg/m²) and had a waist circumference of 113 cm. He had a glucose intolerance after a 75-g glucose oral load test. CT-Myelography revealed voluminous epidural lipomatosis around L4–L5 and L5–S1. Low calorie diet and reduction in alcohol

intake achieved a weight loss of 17.5 kg in 7 months (80 kg, BMI 25.8 kg/m², waist circumference 94 cm) and dramatic improvement in low back pain, walking distance (> 500 m) and reduction of lipomatosis on CT-scan. Our case suggests a relationship between central obesity phenotype and epidural lipomatosis. Specific insulin resistance treatment might be proposed for these patients if this hypothesis is confirmed in further studies.

Keywords Lipomatosis · Epidural · Obesity · Hyperinsulinism

Introduction

Epidural lipomatosis is a rare condition defined by accumulation of non-encapsulated adipose tissue in the epidural space surrounding the spinal cord at the thoracic or lumbar levels. In most cases such lipomatosis is related to local [8] or systemic [4, 6, 11] administration of glucocorticoids, and a few cases have been reported associated with Cushing's syndrome [9, 12]. Idiopathic forms of the disease seem to occur in obese patients but their physiopathology remains unclear. Herein, we report a case of idiopathic epidural lipomatosis in a patient with central/android obesity, lipid metabolism abnormalities and glucose intolerance. Low calorie diet achieved a dramatic clinical improvement and reduction of lipomatosis on CT-scan survey, suggesting a relationship between central obesity phenotype and epidural lipomatosis.

Case report

The 63-year-old man was hospitalized in 1999 with a 3-year history of low back pain and pain in the lower limbs. The patient was receiving simvastatin for mixed hyperlipidemia. He had given up smoking in 1990 and confessed an alcohol consumption of ten drinks per week. Total hip replacement had been performed for bilateral hip osteoarthritis. There was no history of low back pain before 1996 when he presented with diurnal low back pain on standing and on walking. Walking was limited to 200 m at the onset of radicular pain in the left calf. Vascular investigations revealed thrombosis of the left superficial femoral artery and an atheromatous lesion. Surgical dilatation of the lesion was unsuccessful and the patient reported worsening of symptoms, with increased low back pain and reduction in the distance he could walk (100 m) in early 1997. Glucocorticoid had

not been administered at any point of time orally or locally.

Clinical examination revealed that the patient weighed 97.5 kg and was 1.73 m tall (BMI 32.6 kg/m²). Waist circumference was 113 cm, with a waist/hip ratio of 1.04. There was no clinical evidence of Cushing's syndrome. Spine examination demonstrated reduced lumbar spine flexion, with a floor/hand distance of 40 cm. There was no sensory-motor loss and no cauda equina syndrome. Tendon reflexes were present and Babinski reflex response negative. Passive manipulation of the hips revealed no pain or reduction on motion. Laboratory tests were normal for erythrocytes sedimentation rate and C-reactive protein. Fasting glycemia rate was 5.70 mmol/l and hyperglycemia test by oral administration of 75 g glucose indicated glucose intolerance, with glycemia at 7.72 mmol/l at 120 min. Cholesterolemia under simvastatin treatment was 5.62 mmol/l (normal ranges 4.37–6.73) and triglyceridemia was 1.04 mmol/l (normal ranges 0.51–1.83). Fasting cortisolemia at 08.00 hours was 496 nmol/l (150–610). Analysis of cerebrospinal fluid was normal. Radiological examination was performed in January 1999, demonstrating moderate lumbar scoliosis, associated with thinning of the L4–L5 and L5–S1 intervertebral disc space. Myelography demonstrated the dural sac compression particularly at the L5–S1 level (Fig. 1a, b). CT-Myelography displayed a voluminous epidural lipomatosis around L4–L5 and L5–S1 giving a three-leaved characteristic

aspect (Fig. 2a, b). There was a slight discopathy of L4–L5 but no evidence of disc herniation or lumbar osteoarthritis.

In addition to medical treatment combining rest and lumbar traction, the patient was offered a low calorie diet providing 1,600 Kcal/day, along with a reduction in alcohol intake. The diet was strictly adhered and resulted in a weight loss of 17.5 kg in 7 months. In November 1999 the patient weighed 80 kg, with a BMI of 25.8 kg/m². His waist circumference was reduced to 94 cm. The patient reported reduction in lumbar and radicular pain and increased walking range (> 500 m). CT scan of the lumbar spine showed a marked reduction of the lipomatosis (Fig. 3a,b).

Discussion

Apart from obesity, there were no signs indicating Cushing syndrome in the patient/case report analysed here and his fasting cortisolemia levels were normal. Moreover, the patient had not received any glucocorticoids, thus indicating that the lipomatosis was idiopathic and associated with obesity [10]. The symptoms of epidural lipomatosis are not specific and usually include low back pain, sometimes associated with neurological signs, i.e. radicular pain and cauda equina syndrome [7]. Radicular claudication in our patient was confused with signs of arteriopathy, which might have delayed diagnosis.

Fig. 1 a, b Myelography showing a dural sac compression particularly at the L5–S1 level

Fig. 2 a, b CT-Myelography demonstrates a voluminous epidural lipomatosis surrounding and compressing nerves roots and the thecal sac at L4 (**a**) and L5 (**b**) levels

Fig. 3 a, b The CT-scan from the patient seven months after treatment shows a reduction of the epidural lipomatosis at L4 (**a**) and L5 (**b**) levels

Clinical improvement is reported after weight loss, but the reduction in epidural lipomatosis on nuclear magnetic resonance is reported in only a few cases [1, 2]. Follow-up CT scan in our patient showed reduction in the accumulation of epidural adipose tissue around L4–L5 and L5–S1 after a weight loss of 17.5 kg, associated with disappearance of low back pain and improved walking distance. The low calorie diet thus made it possible to avoid laminectomy, which is sometimes proposed in this situation [7, 10].

More importantly, this case suggests a relationship between body fat distribution and the occurrence of epidural lipomatosis. The initial waist circumference of 113 cm indicated accumulation of abdominal adipose tissue [3]. This was reduced to 94 cm with the low calorie diet, parallel with the reduction in epidural lipomatosis. The bodily distribution of fat is not reported in the majority of cases described in the literature, and it is therefore difficult to associate idiopathic epidural lipomatosis with a particular obesity phenotype (central adiposity). It is nevertheless possible that, given that it is

found in Cushing's syndrome and in long-term treatment with glucocorticoids [8], central fat distribution is a common characteristic in all cases of epidural lipomatosis. Although not all the factors determining the localization of adipose tissue (abdominal vs. subcutaneous) are known, insulin appears to play a key role. The association of android obesity, glucose intolerance and mixed hyperlipidemia in our patient suggests a metabolic syndrome associated with insulin resistance [5] and probably hyperinsulinism (to be confirmed in further studies). It is possible that lower levels of insulinemia are achieved with a low calorie diet, thus permitting mobilization of fatty acids (lipolysis) of the central adipose tissue, whether abdominal or epidural. Specific insulin resistance treatment might be proposed for these patients if treatment by diet fails before contemplating surgery.

Acknowledgements The authors thank Daniel Bourry and the Département Communication et Multimédia, Faculté de Médecine, Université de Tours, for his technical assistance.

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