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Epidural Spinal Lipomatosis With Acute Onset of Paraplegia in an HIV-Positive Patient Treated With Corticosteroids and Protease Inhibitor

Case Report

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Study Design. Case report.

Objective. To report a case of HIV-related lipodystrophy with a rapid onset of symptoms from epidural lipomatosis in the wake of protease inhibitor and steroid treatment.

Summary of Background Data. Symptomatic spinal epidural lipomatosis is considered to be a rare condition usually presenting with slowly progressive cord or nerve root compression. Only 2 cases of spinal lipomatosis in HIV-related lipodystrophy have been reported.

Methods. We describe the case of a 41-year-old male with HIV who received protease inhibitor medication and had neurologic deficits rapidly develop.

Results. The patient had complete paraplegia develop within 12 hours from admission following a 1-day history of unsteady gait and a 3-day history of leg numbness. After diagnosis of epidural lipomatosis on magnetic resonance imaging, the patient underwent decompressive thoraco-laminectomy. He recovered well and was able to walk by postoperative day 4.

Conclusion. It is important to maintain an awareness for the possible association between HIV lipodystrophy and symptomatic epidural lipomatosis.

Key words: epidural spinal lipomatosis, HIV lipodystrophy. **Spine 2005;30:E524–E527**

Symptomatic spinal epidural lipomatosis is considered a rare condition that is characterized by a pathologic accumulation of epidural fatty tissue within the spinal canal. This excess fat can lead to slowly progressive cord or nerve root compression[1–11] The first report of steroid-induced spinal lipomatosis in a patient receiving immunosuppressive treatment following renal transplant dates back to Lee *et al*[12] Since then, most cases in the literature have been linked to the exogenous Cushing syndrome from long-term steroid treatment.[13,14] The condition has been reported in cases of morbid obesity,[9,15] rheumatoid arthritis,[16] chronic obstructive pulmonary disease,[14] and dermatomyositis.[17] Epidural spinal lipomatosis is uncommon in endogenous hypercortisolism,[18,19] and only a few cases occurred in the absence of steroid treatment.[7] Ebright[20] and Cersosimo[21] *et* al report on patients who had symptoms develop from spinal lipomatosis following steroid and protease inhibitor treatment for HIV. We describe a case of a patient with HIV who received protease inhibitor medication and had neurologic deficits rapidly develop.

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Case Report

History

A 41-year-old HIV-positive male with a CD4 T cell count of 414×10^6 cells/L and a virus load HIV-1 RNA level less than 200 copies/mL presented to the Neurologic Department with an acute onset of leg weakness and unsteady gait. On admission, he reported a 3-day history of progressive numbness of the legs. He did not have pain.

HIV infection related to intravenous drug abuse was diagnosed 8 years previously, and treatment with the protease inhibitor Sequinavir (1200 mg/day) (Louston Intl., Inc., Linwood, PA and nucleoside-analogue reverse-transcriptase inhibitors (NRTI) lamivudine (300 mg/day) and stavudine (80 mg/day) was begun 6 years later, 2 years before neurologic signs developed. Sequinavir was discontinued when the virus load was less than 400 copies/mL, 8 weeks before neurologic symptoms of myelopathy began. NRTI medication was continued.

One year previously, the patient had received long-term corticosteroid treatment following the diagnosis and operative treatment of a cerebellar metastasis of an adenosquamous carcinoma of the lung. Steroid medication (methylprednisolone 24 mg/day) was continued for 2 months after microsurgical removal of the metastasis and during consecutive radiotherapy. Afterward, steroids were decreased to 18 mg/day for an additional 6 weeks before being stopped altogether, 7 months before neurologic symptoms of lipomatosis began. The primary tumor in the left-upper lobe of the lung was surgically removed and treated by adjuvant chemotherapy.

Examination

Sagittal T1-weighted magnetic resonance imaging (MRI) showed accumulation of epidural fatty tissue with spinal cord compression (Figure 1). Both MRI and plain radio-

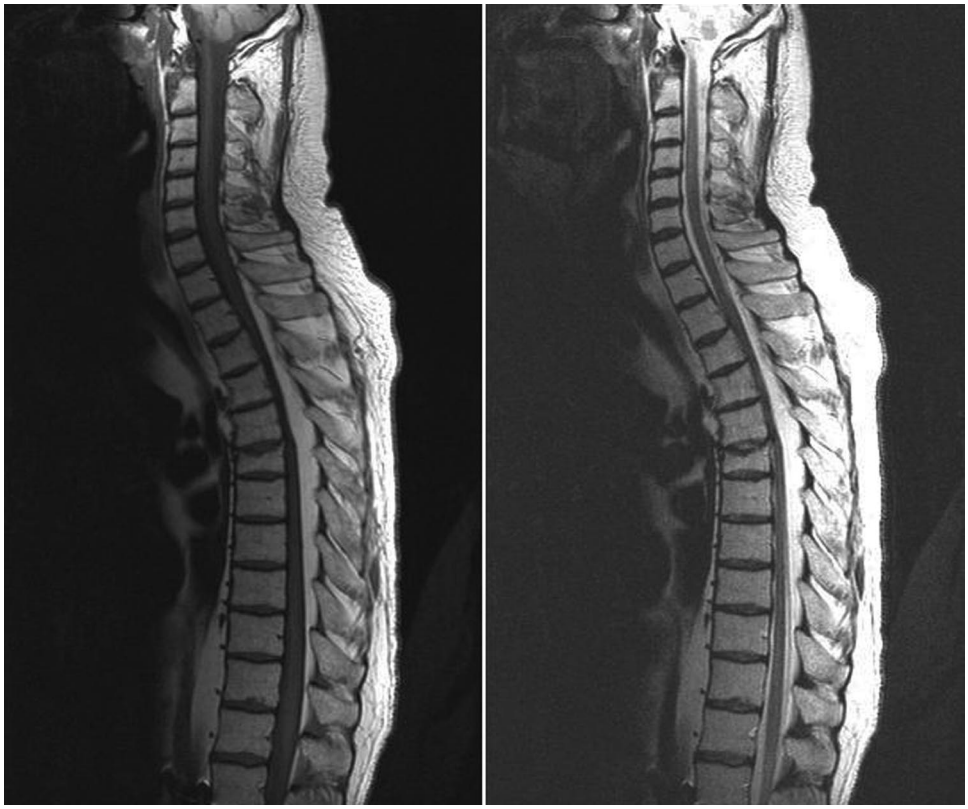

Figure 1. MRI examination. **Left**, Sagittal T1-weighted MRI shows extensive thoracic epidural lipomatosis and the old osteoporotic fracture of the sixth thoracic vertebral body. **Right**, On the T2-weighted image acquired in the midline, cerebrospinal fluid surrounding the cord is not visible. However, it is not possible to define a circumscribed point of maximum cord compression.

graphs of the thoracic spine suggest older osteoporotic compression fractures of T3, T4, and T6. Myelography was performed to determine the point of maximum compression, which was located to the level of T5–T7 (Figure 2). MRI showed a tumor lesion at the costovertebral joint of T2/T3, however, with no signs of cord compression (Figure 3). Laboratory examination of cerebrospinal fluid excluded carcinoma cells but showed an increased protein concentration of 757 mg/dL. Throughout the protease inhibitor treatment period, triglyceride and cholesterol levels were not increased. A routine serologic examination performed 7 days before admission confirmed normal triglyceride (157 mg/dL) and cholesterol (222 mg/dL) levels.

Clinical Course and Outcome

Under the initial worst case assumption of spinal cord compression by a metastasis from the previous bronchial adenocarcinoma, a single prednisolone dose of 1000 mg was administered on admission before diagnostic workup began. From admission, the neurologic condition of the patient deteriorated within 12 hours, to the point of paraplegia and urinary incontinence. In view of this rapid neurologic deterioration and the radiologic signs of spinal cord compression, decompressive laminectomy T5–T7 and the removal of the epidural fat were performed. The patient recovered well and was transferred to the rehabilitation unit 4 days later, at which point he was already able to walk again. At 8 months following surgery, he was still neurologically stable and able to walk. NRTI medication was continued.

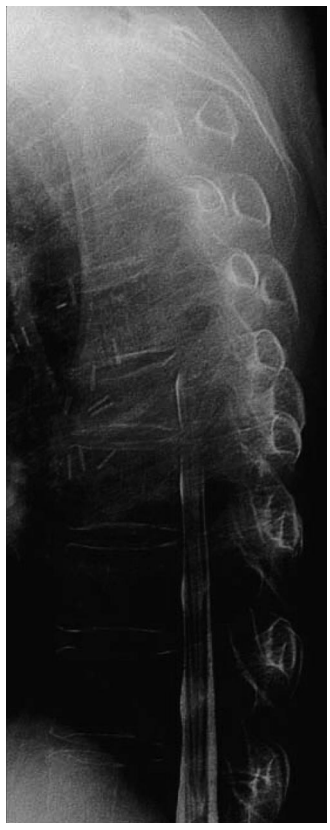

Figure 2. On myelography, contrast medium passage was impaired at the T6 level.

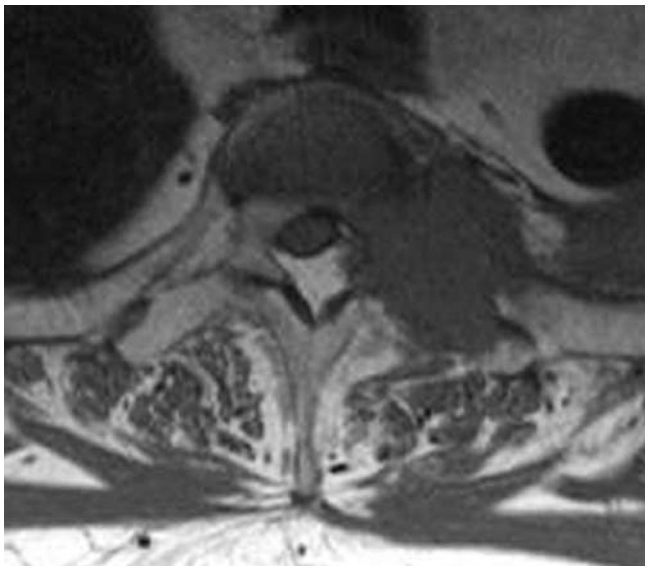

Figure 3. The metastatic lesion at the costovertebral joint T2/T3 does not compress the spinal cord.

■ Discussion

Since 1996, the generally accepted treatment of HIV-1 infection is the use of 3 antiviral drugs. In most cases, 2 NRTI are combined with one or more protease inhibitors.[17] This treatment regimen has led to an unsurpassed reduction in HIV-related morbidity and mortality.[17,22] However, lipodystrophy is a known and clinically troublesome adverse effect of protease inhibitor treatment characterized by a redistribution of peripheral fatty tissue, mostly from the face and extremities to central areas of the body.[17,20] Typical findings include the accumulation of fat in the abdominal viscera, mediastinum, supraclavicular fossae, and dorsocervical soft tissue, similar as in the exogenous or endogenous Cushing syndrome.[20]

Recent studies support the hypothesis that the NRTI component of antiviral medication may by the initiating factor of lipodystrophy, which is then aggravated by protease inhibitor medication.[17] However, it does remain the subject of research and discussion whether HIV lipodystrophy is the actual cause of spinal epidural lipomatosis. It is interesting that the presented patient had neurologic symptoms develop 8 weeks after protease inhibitor and more than 7 months after steroid medication had been terminated. Acute neurologic deterioration is not typical of spinal epidural lipomatosis,[5] and it may be speculated, whether the acute onset of symptoms was related to the additive effect of corticosteroids, anti-HIV medication, and the osteoporotic compression fracture of T6, albeit MRI is more suggestive of an older fracture (Figure 1).

To date, only 2 case of HIV-related spinal lipomatosis are reported in the literature, both of which presented slowly developing neurologic deficits over a period of several months.[20,21] Treatment recommendations for symptomatic spinal epidural lipomatosis in cases with different pathologies range from conservative management, with treatment of the underlying condition and weight reduction,[9,11,19,23,24] to decompressive surgery.[1,2,6,7,14] Ebright *et al*[20] report marked, but not complete recovery, following decompressive thoracolaminectomy. Kurt and Bakker-Niezen[22] show 1 case and a review of 12 additional patients, as well as a comprising argument favoring early operative intervention.

In the case of the HIV-related symptomatic lipomatosis, Cersosimo *et al*[21] report spontaneous gradual recovery following the discontinuation of protease inhibitor medication. However, as of yet, there are no clear guidelines whether antiviral treatment ought to be modified or stopped. The risk of neurologic deterioration from continuing NRTI and protease inhibitor medication will have to be carefully weighed in the individual case. In view of the increasing percentage of successfully treated patients with HIVs,[12,22] it is important to recognize the potential association between HIV-related lipodystrophy and symptomatic epidural lipomatosis.

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