Neuropathic Arthropathy of the Spine in Diabetes

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Neuropathic arthropathy, as originally described by Charcot (1), is typically associated with syringomyelia, syphilis, and leprosy. The first time it was described as existing in a diabetic patient was in 1936 by Jordan (2).

In the diabetic population, neuropathic arthropathy is an uncommon but well-recognized manifestation. Neuropathic joints are classically associated with loss of pain sensation, loss of tendon reflexes, neuropathic foot ulceration, arthropathy, and autonomic neuropathy. The distribution of affected joints is commonly distal; tarsal, metatarsal phalangeal, and interphalangeal joints are those most frequently involved. Charcot joint of the spine associated with diabetic neuropathy has rarely been documented. In a collection of 109 cases reported by Robillard et al. (3), there was only 1 case of spinal involvement. Here we report the clinical and radiological features of two cases of Charcot arthropathy of the spine in type I diabetic patients.

CASE 1 — A 57-year-old Caucasian woman with type I diabetes of 45 years' duration presented with sudden-onset severe central lumbar back pain and restricted back movement. She had developed a painful neuropathy of the lower limbs 8 years earlier and a right Charcot ankle 1 year previously. On examination, there was evidence of glove and stocking neuropathy bilaterally but no neurological evidence of cord compression or sphincter disturbance. Background retinopathy was present on fundoscopy with preserved visual acuities. Blood pressure was 135/86 mmHg. Previously, she was shown to have established nephropathy with proteinuria of 0.55 g/24 h and an isotopic glomerular filtration rate of 24 ml/min.

Plain X ray of the lumbar spine demonstrated a marked loss of disc space at L4/5 with some destruction of the adjacent anterior margins of the vertebral bodies. This was felt to be highly indicative of infection. Blood urea was 18.9 mmol/l and creatinine 155 μmol/l. Hematology, bone biochemistry, and plasma electrophoresis were normal. Chest X ray was normal and Mantoux negative at 1 in 1,000.

Computerized tomography (CT) of the lumbar spine demonstrated a vacuum phenomenon at L4/5 with a soft tissue mass around the disc. Large-bore biopsy was carried out at the level of L4/5 disc space. Histology demonstrated necrotic bone but no evidence of infection or neoplasia. Acid-fast bacilli were not identified, and the specimen was sterile on subsequent culture. The patient gained mobility slowly using conventional analgesia and a transcutaneous nerve stimulator. The back pain became progressively less troublesome, and in 3 months, she was requiring no analgesia. Repeat radiology with magnetic resonance imaging 3 years after her original presentation (Fig. 1) shows no change in the appearance of bony disorganization and loss of disc space at L4/5. There is no local edema or soft tissue swelling to indicate an inflammatory process.

CASE 2 — A 40-year-old diabetic Caucasian woman, insulin-dependent for 13 years, presented with acute thoracolumbar back pain. The patient had established diabetic nephropathy with proteinuria of 1.2 g/24 h and an isotopic glomeral filtration rate of 39 ml/min, and she had been treated for proliferative retinopathy. At presentation, she had clinical diabetic autonomic neuropathy and signs of a glove and stocking peripheral sensory neuropathy but no motor loss or sphincter disturbance.

Plain X ray of the thoracic spine demonstrated a destructive process at the level of T12/L1. A lumbar myelogram was performed, which showed a complete block at that level and a collapsed vertebral body. CT of this area confirmed a paraspinal mass with marked destruction of T12/L1 vertebral bodies. Clinical reexamination confirmed no neurological deficit attributable to the lesion at this site. Investigations showed the following: a normochromic normocytic anemia, erythrocyte sedimentation rate 45 mm/h, creatinine 106 μmol/l, calcium 1.93 mmol/l with albumin of 26 g/l, phosphate 1.7 mmol/l, and normal hematinics. Blood and sputum cultures were consistently negative as was Mantoux 1 in 1,000.

A diagnosis of spinal osteomyelitis was made on the basis of the radiological

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CT, computerized tomography.

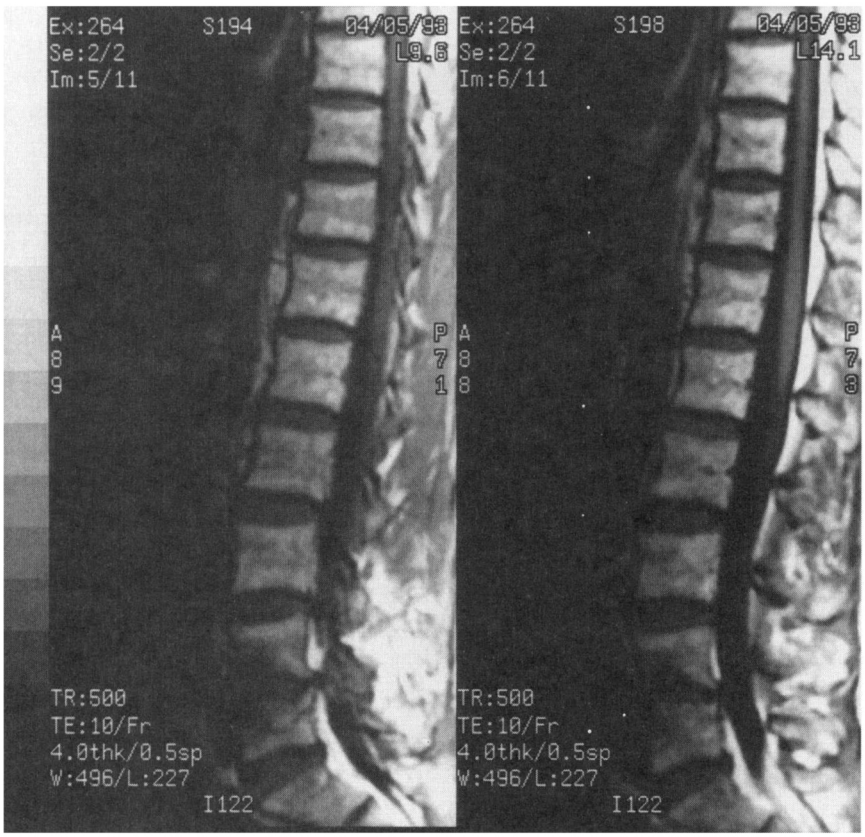

Figure 1—*Case 1: magnetic resonance imaging scans performed 3 years after the patient's original presentation. The T1 sagittal scans show destruction of the L4/5 disc and vertebral bodies, which had not changed since the original X ray.*

appearances, and a left hemilaminectomy was performed with exploration of interspinal joints. At operation, there was little evidence of compression of the theca by the paraspinal mass, and biopsies taken were subsequently sterile for acid-fast bacilli and other cultures. Biopsy tissue was described macroscopically as fragments of hard gray material. Microscopically, these were bony particles with marrow and dense collagenous material without evidence of granulomata or metastatic tumor; no organisms were seen. There was no evidence of chronic or acute inflammatory infiltrate.

Postoperatively, she continued to complain of back pain but was able to move slowly, with the pain alleviated by simple analgesia. Despite the effects of the peripheral and autonomic neuropathy, her mobility continued to improve with no back pain. Repeat CT showed unchanged destruction of the T12/L1 disc.

CONCLUSIONS — When a diabetic patient presents with acute back pain, the less common causes require consideration. Latent spinal infection is an important cause not infrequently seen in this potentially predisposed group of patients (4,5). As illustrated by these two cases, however, diagnostic uncertainty may arise when an acute spinal neuropathic degeneration mimics infection. It is possible that the two pathologies may coexist.

The first documented case of Charcot spine in diabetes was reported in 1952 by Zucker et al. (6) in a patient with established neuropathy. While local soft-tissue swelling is a common feature of peripheral Charcot joint involvement, this may not be clinically apparent in spinal disease. Pain is generally not a primary feature of peripheral Charcot arthropathy because of the presence of distal hypoesthesia, and when there is pain it is disproportionately less than that caused by an equivalent degree of joint disruption due to infection. However, pain does seem to be a major feature in the few reported cases of diabetic spinal arthropathy, as in our two. The extent of the spinal disease in diabetes is variable, as cases have been documented with both generalized and localized disease (7,8).

The radiological evaluation of a disorganized spinal joint can be difficult in this clinical setting, in which infection cannot be confidently excluded in a patient who may have features predisposing to Charcot change. The typical features associated with neuroarthropathy are osteopenia (although this may be difficult to assess), destruction of articular surfaces, gross disorganization, fragmentation, fractures, periosteal new bone formation, bone resorption, and vascular calcification. Changes relating specifically to the spine include anterior and lateral subluxation, dislocation, and kyphoscoliosis. A vacuum phenomenon, as seen in case 1, suggests a noninfectious cause (8). None of these changes, however, are specific; the differential diagnosis must include infection, herniated disc, tumor metastases, Paget's disease, or "pseudo-Charcot's" due to nonsteroidal anti-inflammatory drugs (9). Hemodialysis can also be associated with similar spinal changes (10).

Scintigraphy is commonly used in an attempt to differentiate infection from neuropathic problems. Being nonspecific (11), technetium 99^{m}-MDP scans are generally unhelpful. By using white cell–labeling techniques (12,13), the specificity in differentiating infection from diabetic osteoarthropathy is improved, but these tests are unreliable in rapidly pro-

gressive disease processes, be they infective or neuropathic problems.

These two cases demonstrate the difficulty in reaching a diagnosis in this condition. Investigations should be directed at the exclusion of other causes, particularly infections. Bone biopsy, either with open or with CT- or fluoroscopy-guided large-bore needle technique, is essential to reduce sampling error and to obtain enough material for both histological study and formal culture. The natural history of the condition is relatively benign. Of those cases reported, none developed neurological deficit in addition to their peripheral neuropathy. In our cases, using supportive measures alone, the prognosis from the lesion likewise appears good. Neither case has developed any permanent disability as a direct consequence of the acute event.

In conclusion, we have described two women with type I diabetes and an acute neuroarthropathy of the spine, in which an infectious process was the initially suspected diagnosis. Subsequent investigation excluded infection, and both women spontaneously recovered from the acute episode but demonstrated persistent radiological changes consistent with Charcot spine.

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