BRIEF REPORT

SPINAL CORD BLOCK WITH A DESTRUCTIVE LESION OF THE DORSAL SPINE IN ANKYLOSING SPONDYLITIS

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Extensive destructive lesions in ankylosing spondylitis (AS) that involve an intervertebral disc space and adjoining vertebral bodies have been reported since 1937 (1,2). The cumulative experience was reviewed in 1972 (3). The roentgenographic appearance may closely simulate that of infective spondylitis, but the lesions are almost invariably bacteriologically sterile. Trauma may be the dominant or possibly the sole etiology of these lesions (3), which evolve from purported ununited fractures occurring most commonly at sites in the lower dorsal spine, the juncture of rigid segments of the lumbar and dorsal spine. By inference, a prerequisite for this mechanism is obvious AS at the stage of spinal ankylosis.

A search of the literature disclosed evidence of spinal cord involvement accompanying the disc space lesion of ankylosing spondylitis in only a single case report (4).

Our report concerns a patient admitted for progressive paraparesis who had an epidural mass at the site of an extensive destructive lesion at D8–9, mistakenly interpreted as infective. Since the spine was neither deformed nor rigid, the diagnosis of AS came to light incidently, late in the patient's convalescence.

Case History. A 59-year-old laborer noted progressive weakness of the legs 6 months before coming to another hospital in November 1979. He had extreme weakness of the legs, a sensory deficit below D8, and bilateral Babinski signs. The sedimentation rate was elevated, and a radiograph showed a widened irregular disc space at D8–9 with destructive changes in the adjoining vertebral end plates (Figures 1A and B). Minor degenerative changes were diagnosed in films of the lumbosacral spine. A myelogram showed complete, apparently posterior extradural block at D9 (Figure 2).

After the patient was transferred to the Ann Arbor Veterans Administration Medical Center, laminectomies were performed at the D7, D8, and D9 levels, where there were no abnormalities of the laminae or ligamentum flavae. However, the posterior epidural space in this region was filled with what appeared to be compact adipose tissue, 1-cm thick at the D8 level, that indented the dural sac but which reexpanded readily after removal of the fatty tissue by blunt dissection. Portions of sclerotic material from the D8–9 disc space were also extracted.

Results of Gram stain and stain for acid-fast bacilli of the various tissues, as well as cultures for pyogens, fungi, and tubercle bacilli, were negative. Histopathologic sections showed adipose tissue in which there were foci of chronic inflammatory cells, primarily lymphocytes. Small bits of bone and adjacent fibrous tissue were included with reactive new bone formation and nonspecific chronic inflammatory cell infiltrates (Figure 3).

The bulk of the tissue removed was mature

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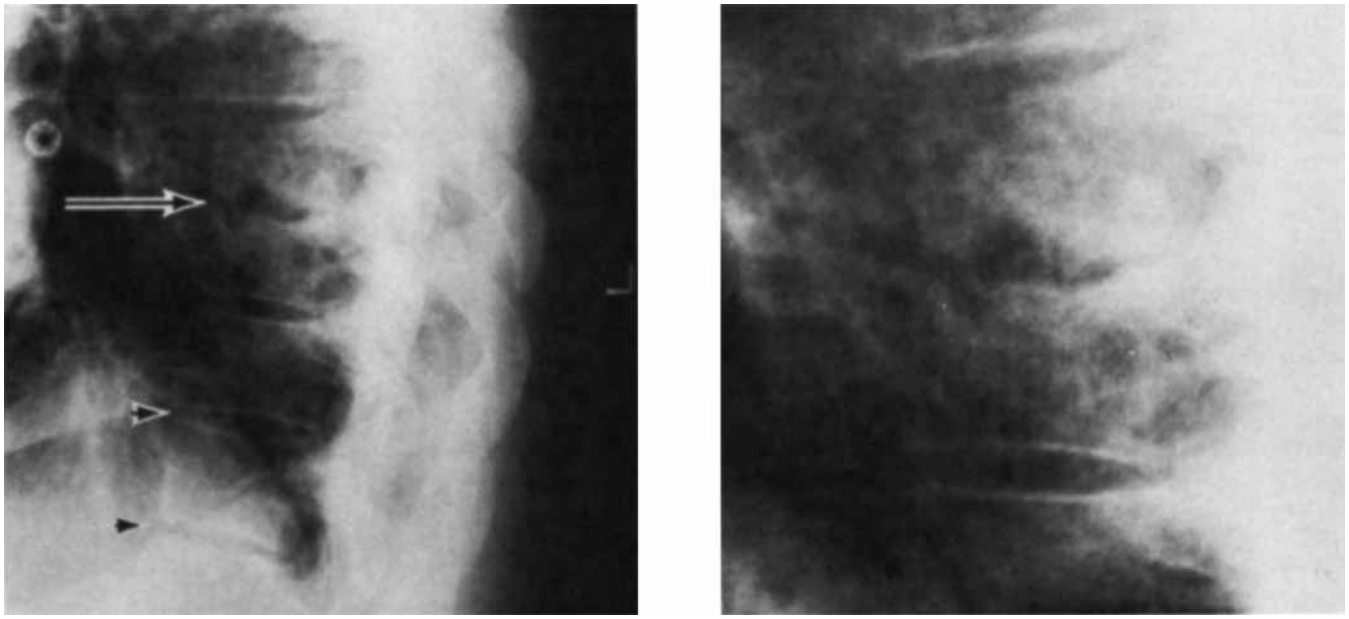

A B

Figure 1. **A**, Lateral radiograph of thoracic spine shows irregular widening of the D8–9 disc space, accompanied by sclerosis and destructive change of adjacent vertebral endplates, consistent with spinal pseudarthrosis (**arrow**). No definite posterior element fracture can be seen. Notice squaring of vertebral bodies with delicate new bone apposition (**arrowheads**) in the distal thoracic spine, consistent with ankylosing spondylitis. **B**, Enlarged view of D8–9 disc space.

adipose tissue with scattered aggregates of chronic inflammatory cells and fibrous tissue as noted above (Figure 4). The inflammatory nature of the tissue removed was definite but mild and nonspecific. The new bone could have reflected healing trabecular fractures or periosteal bone formation due to continued low-grade inflammation.

After the cultures obtained at surgery all resulted in negative findings, the patient was followed as an enigmatic case of noninfective inflammation and destruction at a disc space with cord compression due to thickened epidural inflammatory tissue. Postoperatively, he recovered strength rapidly, and by December 1980 there were no abnormal neurologic signs.

The diagnosis of underlying AS was given 8 months later, when the patient returned for reevaluation of military "service-connected" sacroiliitis. History from the patient at this time and from available army medical records disclosed that pain in the left heel and low back started in 1937. Dull lumbar pain persisted with severe exacerbations biannually until 1943 when radiographs showed bilateral sacroiliitis, and he was given a medical discharge from the army. In recent decades, lumbar pain persisted but was usually mild and overlooked.

Physical examination 8 months after his laminectomy showed an obese, short man without kyphosis, scoliosis, or a gibbus. Cervical range of motion was normal, and chest expansion was 2 cm. The lumbar spine was severely limited in mobility in all directions but was not rigid. The hips and peripheral joints were normal. HLA typing revealed A2,11; B27,—. The available spinal radiographs showed squaring of the bodies of the 10th, 11th, and 12th dorsal vertebrae (Figure 1) and bilateral obliteration of the sacroiliac joints (Figure 5).

Discussion. Cauda equina lesions have been increasingly recognized in ankylosing spondylitis, albeit poorly comprehended. Myelographic interpretations in these cases are variously reported as "normal" (5), "wide dural sacs" (6), and "dilation" or "diverticula" along nerve root sheaths (5,6); histopathologic observations include intradural fibrous tissue with lymphocytes (6), arachnoid-lined diverticula eroding the posterior surface of spinal canal without inflammation (7), adhesions of nerve roots to a dense fibrous arachnoid (8), and epidural "fatty material" without inflammation (9).

There is evidence that cervical and lumbar areas of the spinal cord have been similarly affected in

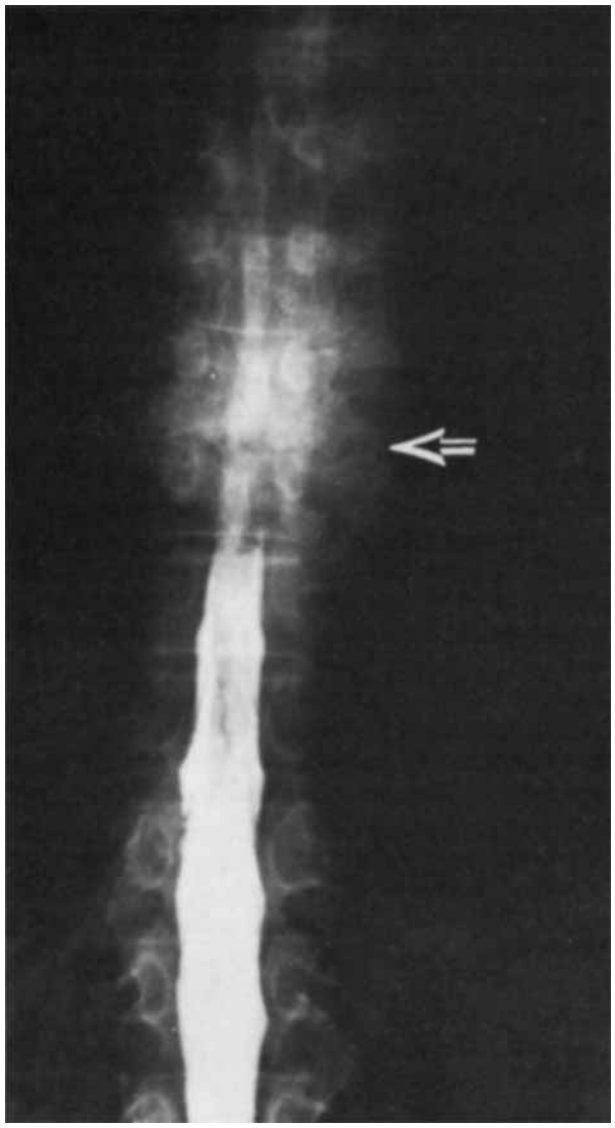

Figure 2. Myelogram showing complete block inferior to the affected interspace (**arrow**). Notice endplate irregularities on anteroposterior projection as well. The paravertebral widening at D8–9 may represent chronic inflammatory or fibrous response to the pseudarthrosis.

AS. Periradicular arachnoiditis affecting cervical segments was described by Bechterew in 1893 (10). Bailey and Casamajor in 1911 (11) and Parker and Adson in 1925 (12) included cases of AS under the generic term "osteoarthritis," among which were several patients with cord compression of the dorsal spine. They described the mechanism of the cord compression as "overgrowth of soft tissue" and "chronic osteochondritis."

Surgeons performing osteotomy of the spine for correction of kyphosis in AS have reported that the dura is apt to be thickened and adherent both to the overlying ossified ligamentum and to extensive adhesive arachnoidits (13,14). According to a published report (15), doctors performing surgery on a patient discovered thick adhesions between dura and cord at D7–8; later, after a myelogram showed a complete obstruction, similar changes at D12 as well as a large arachnoid cyst were revealed during repeat surgery. In contrast to our patient, there was no evidence of a destructive disc space lesion. Histologic sections showed merely fibrous proliferation. An account of a destructive intervertebral disc space lesion of AS accompanied by cord compression was published in 1969 (4). The authors described a man with "an immobile kyphotic thoracic and lumbar spine" who developed paraparesis and a sensory level deficit at the umbilicus associated with a disc lesion at the T9–10 interspace. The surgeons found "granulation tis-

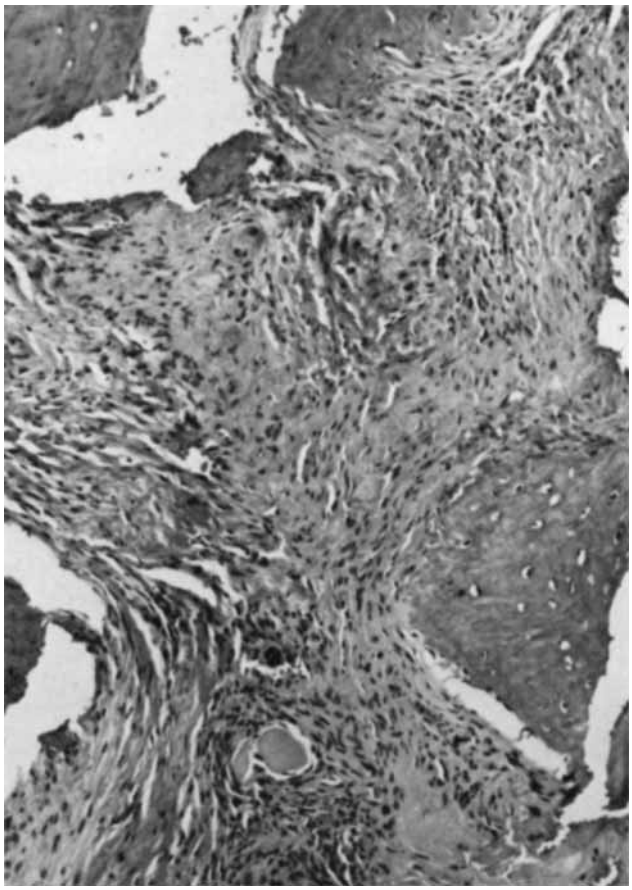

Figure 3. Reactive bone formation, fibrosis, and mild chronic inflammation were noted in adipose tissue. There is questionable palisading of cells and fibrinoid change adjacent to the new bone.

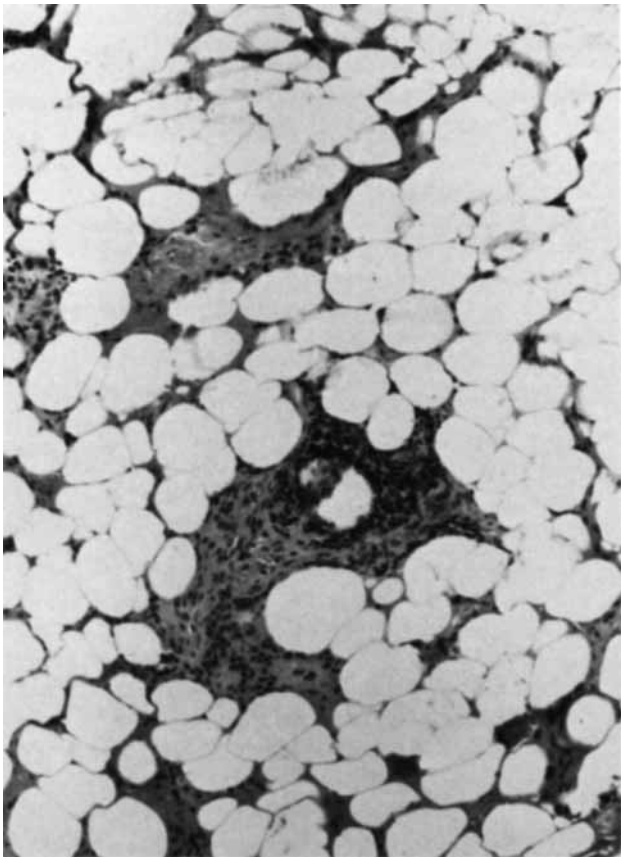

Figure 4. Adipose tissue with only a few foci of chronic inflammatory cells, primarily lymphocytes.

sue" in the involved interspace and adjacent epidural space; this material comprised largely fibrous tissue with areas of fibrocartilage, new bone, palisading of fibroblasts, questionable fibrinoid degeneration, and chronic inflammatory cells. The severe spondylitic deformity and rigidity in that patient contrasts with the absence of deformity in ours. His physicians hypothesized that a callus at the fracture site progressively enlarged due to repeated trauma and ultimately compressed the cord. The patient recovered only partially neurologically before dying suddenly of a pulmonary embolus 5 months later.

Inflammatory cells are not commonly found in meningeal lesions of AS; perhaps most tissues are taken during the later stages of the disease after purported inflammation has subsided. Elevated spinal fluid protein in early, but not late, cases of AS suggests that inflammation of the leptomeninges is common but limited in duration (16).

Except for limitation of lumbar spinal mobility, which was difficult to evaluate when our patient was encountered postoperatively, his objective spondylitic involvement of the spine was limited to squaring of the distal dorsal vertebra. Even though this finding is radiologically unobtrusive, it is a marker for the Romanus lesions, i.e., osteitis at the margin of the disc space (17). From our experience, it seems that in some cases the remarkable, extensive, destructive discovertebral lesions in AS may be initially inflammatory rather than merely reactive to trauma. Because ankylosis was absent in our patient, we may question the prevailing explanation that the disc lesion is basically a pathologic fracture at the juncture of rigid segments of dorsal and lumbar spine (3).

In summary, there is evidence of spinal meningeal disease with ankylosing spondylitis that may sometimes evolve to compress the cord or nerve roots, particularly in the lumbar segment. Also, there is a potential for an extradural mass of fibroadipose tissue associated with a noninfective destructive disc space lesion, a process found predominantly in the lower thoracic spine. The latter mechanism is established in our patient by myelographic and surgical findings. Inasmuch as the dura was not opened, we have no proof that purported meningeal changes did not play a minor role, as well, in facilitating cord compression. In the study of this patient, we can again recognize the diagnostic confusion surrounding disc space lesions

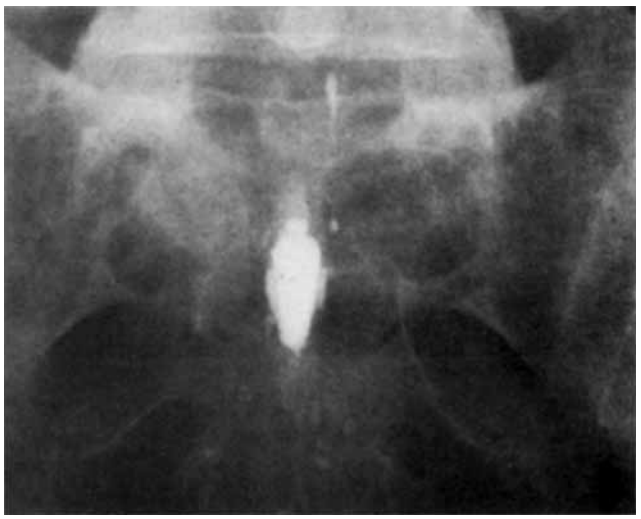

Figure 5. Anterior view of pelvis showing ankylosing and partial obliteration of the sacroiliac joints.

and emphasize that the underlying spondylitis may be mild, apparently inactive, and easily overlooked.

Anatomists have warned that a small lesion in the ventral wall of the thoracic vertebral canal may damage the cord, which is proximal to bone in this area (18). Thus, patients with destructive disc space lesions should be given a careful neurologic examination. Prompt immobilization until healing occurs is probably advisable in order to minimize bone overgrowth at the site.

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