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Spinal cord compression complicating aseptic spondylodiscitis in ankylosing spondylitis

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ABSTRACT

Aseptic spondylodiscitis is a well recognized complication of ankylosing spondylitis. Neurological complications of such discovertebral lesions are uncommon. We report a new case with a diagnosis of T12-L1 spondylodiscitis which developed ten years after a spinal cord compression. Such neurological complications of aseptic spondylodiscitis may be explained by proliferative epidural tissue without predominant inflammatory infiltrates and also the development of new bone reaction, suggesting the contribution of mechanical factors.

Introduction

Aseptic spondylodiscitis is a rare complicating feature of ankylosing spondylitis (AS) (1). The clinical presentation may vary from asymptomatic to localized back pain. Serious spinal cord damage has exceptionally been described in association with spondylodiscitis in AS (2). We report here a new case with a previously diagnosed spondylodiscitis which developed ten years after spinal cord compression and required surgical treatment.

Case report

The patient was a 58-year-old man with a 30-year history of HLA-B27 positive AS with axial involvement. His treatment consisted of intermittent administration of non-steroidal anti-inflammatory drugs (NSAIDs). He was seen in 1995 for increasing back pain without neurological symptoms. On x-rays, the intervertebral disc space between Th12 and L1 was reduced with erosions and a dense sclerosis of adjacent end plates (Fig. 1). He was not febrile and was thus diagnosed as having aseptic spondylodiscitis. With NSAIDs, he experienced progressive relief of back symptoms. He was seen again in 2005 for increasing back pain with gait disturbance, altered sensation when defecating without anal incontinence and pain located in the anterior aspect of his right thigh. He had no recent history of trauma or unintentional acute twisting movements with his back. At clinical examination, he was apyrexial with a marked ankylosed spine and reported percussion tenderness in the thoraco-lumbar transitional area. Neurological examination found moderate weakness of his right thigh, no sensory loss including the sacral area or loss of tendon reflexes. The anal sphincter was normal. Erythrocyte sedimentation rate was normal as well as CRP levels. Thoraco-lumbar spine radiographs showed the same changes at the Th12-L1 level than observed 10 years previously, without progression of the spondylodiscitis. Magnetic resonance imaging (MRI) was performed and showed decreased signal intensity in the disc space and the adjacent margins of Th12 and L1 on T1-weighted images (Fig. 2A). On T2-weighted images, there was increased signal intensity on vertebral end plates but also in the adjacent epidural space with spinal cord compression (Fig. 2B). The spinal canal was narrowed by a similar process around the facet joints. Due to the gradual deterioration of his gait and the persistence of defecation dysfunction, a surgery was decided. During the operation, it was discovered that there was a dense sclerosis of the vertebral end plates and the disc space was completely narrowed with rare persistent discal fragments; facet joints were hypertrophic and a granulation tissue encapsulated the dura. Laminectomy of

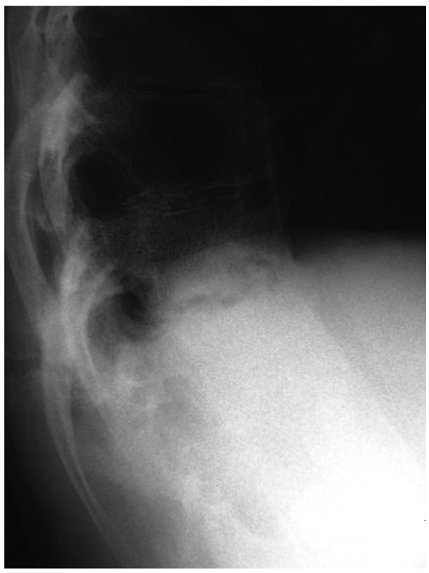
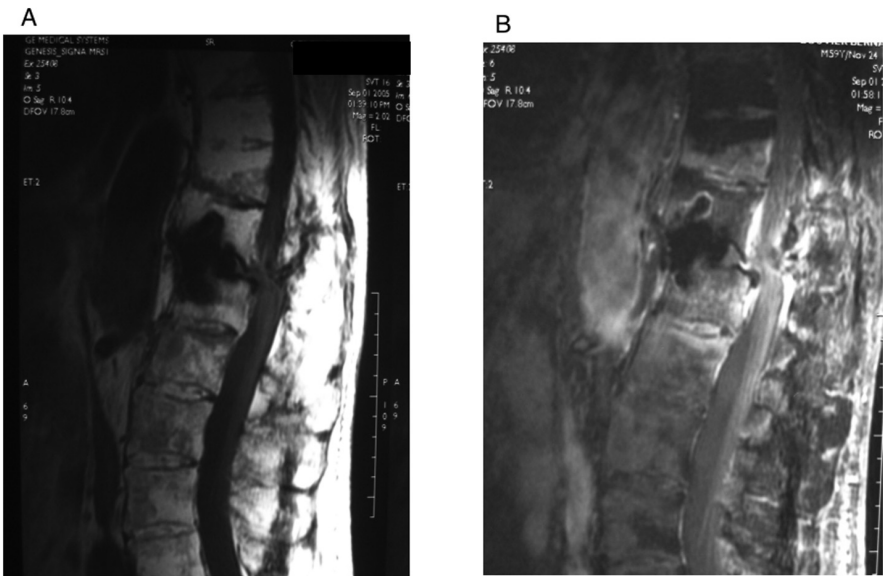


Fig. 1. Lateral radiograph of the thoraco-lumbar spine showing widening of the Th12-L1 space, sclerosis and destructive changes of adjacent vertebral endplates, consistent with the diagnosis of spondylodiscitis.



Figs. 2A and 2B. Sagittal T1 and T2 weighted magnetic resonance image of the thoraco-lumbar spine. Decreased signal intensity in the disc space and the adjacent margins of Th12 and L1 was observed on T1-weighted images with major destructive lesions of the vertebral bodies (Fig. 2A). On T2-weighted images, there was increased signal intensity on vertebral end plates but also in the adjacent epidural space with spinal cord compression (Fig. 2B). The spinal canal was narrowed by a similar process around the facet joints.

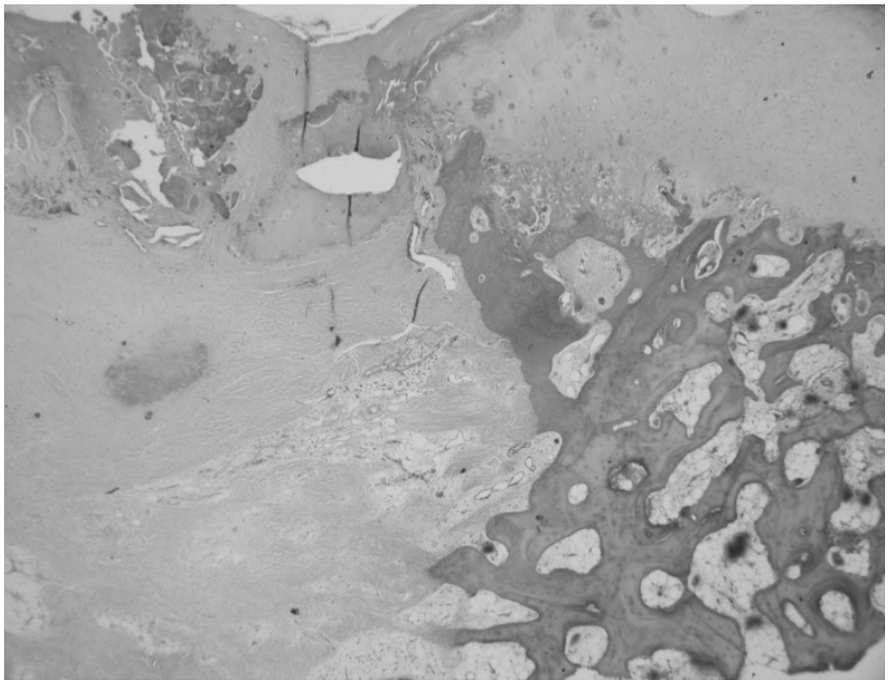


Fig. 3. Histopathological findings of the Th12-L1 biopsy showing fibrous tissue, necrotic bone and reactive new bone formation.

Th12 and L1 was performed with posterolateral fusion from Th11 to L2 with pedicle screw system. Histology showed no inflammatory cells but degenerative changes, with fibrous tissue, necrotic bone and reactive new bone formation (Fig. 3). Bacterial cultures of disco vertebral specimens remained negative. After surgery, the patient reported relief of his back pain and exhibited progressive recovery of motor strength in his thigh and normal control of defecation.

Discussion

Aseptic spondylodiscitis is a rare complication of AS defined by radiological abnormalities that included destructive foci throughout the discovertebral junction, bony sclerosis of adjacent vertebral end plates, widening or narrowing disc space, osteolysis of the vertebral bodies or in some cases, pseudarthrosis (3, 4). The prevalence of spondylodiscitis in AS has been estimated in retrospective study between 1 and 10% with a mean of 4.5% (3). In a retrospective study performed on hospitalized AS patients in a teaching hospital, the prevalence was found to be 18% (4). Cawley found a higher prevalence, reaching 28%, but this radiological analysis also included vertebral lesions such as Romanus osteitis (1). The clinical presentation of spondylodscitis is variable: it may be asymptomatic and found fortuitously on spine x-rays, or it may be responsible for acute and localized backache which may be interpreted as a common symptom of AS (3, 4, 5). However, backaches are usually intensified by exercise and relieved by rest. It is admitted that spondylodiscitis in AS occurred mainly in patient with long standing disease with a mean age at diagnosis around 40 and a mean disease duration ranging from 7 to 25 years (1, 3, 4). A history of trauma before the diagnosis of spondylodiscitis was found in certain studies but not all (1, 3). The discovertebral lesions are mainly described at the thoracic or lumbar level and exceptionally at the cervical spine (3, 4).

The pathogenesis of spondylodiscitis in AS is still controversial. Both mechanical factors, trauma and inflammation has been proposed for explaining such discovertebral lesions (3). Cawley emphasized the major role for manual labor as a predisposing factor but this was not confirmed by all authors (1, 3). Mechanical factors may play a role in patient with longstanding disease and rigid spine and may be responsible for abnormal motion between two fused segments, leading to progressive discovertebral destruction (3). The traumatic pathogenesis is supported by the absence of inflammatory lesions on histopathological analysis (6). Conversely, histological changes consistent with inflammation have been observed (4). Taken together, it is currently admitted

Table I. Spinal cord compression related to aseptic spondylodiscitis in ankylosing spondylitis.

Author (reference)	Sex	Age	Profession	Disease duration	Location of spinal cord compression	Delay between diagnosis of spondylodiscitis and neurological symptoms	Bamboo spine	Trauma	Clinical symptoms	MRI	Surgery observation	Biopsy	Treatment
Good (8)	M	59	labourer	40	T8-T9	Simultaneous diagnosis	no	no	Progressive paraparesis	ND	Presence of adipose tissue in the epidural space	Adipose tissue with chronic inflammatory cells Reactive new bone formation	Laminectomy
Wise (9)	M	59	ND	39	T8-T9	Simultaneous diagnosis	ND	no	Paresthesia Urinary incontinence Progressive lower limb weakness	ND	ND	Inflammatory destruction of bone	Surgical treatment
Jean-Baptiste (10)	M	38	ND	22	T11- T12	5 months	yes	no	Progressive paraparesis Intermittent urinary incontinence	ND	Spinal compression due to epidural adipose tissue and bone formation	ND	Surgical treatment
Jobanputra (11)	M	56	ND	30	T9-T10	9 years	no	no	Leg weakness Poor control of micturition (urinary incontinence)	ND	Granulation tissue encircling the dura	Fibrous and granulation tissue Mild chronic inflammatory cell infiltrate New bone formation	Laminectomy
Sakaura H (12)	M	55	ND	27	T11-T12	Simultaneous diagnosis	yes	no	Gait disturbance Urinary retention	Decreased signal intensity disc and vertebra T1 and T2 weighted images	Instability at T11-T12	ND	Laminectomy
Personal case	M	58	labourer	30	T12-L1	10 years	yes	no	Gait disturbance altered sensation when defecating, pain in the right thigh	Decreased, signal intensity in T1 and increased signal in T2 weighted images disc and vertebra	Granulation tissue around the dura	Fibrous tissue, necrotic bone and new bone formation	Laminectomy

that a multifactorial pathogenic process is associated with the development of spondylodiscitis in AS (3). This is supported by the variability of histological and radiological findings (7).

Spinal cord compression is uncommon in AS with spondylodiscitis. A Medline search performed throughout the 1980-2007 period identified 5 similar cases (Table I) (8-12): they corresponded to 5 male patients with a mean age of 53.4 and a mean disease duration of 31.6 years. Bamboo spine was noticed for 2 patients. They all had spondylodiscitis at the thoracic level. Clinical symptoms were paraparesis in 2 cases (8,10), urinary dysfunction in 4 cases (9-12), lower limbs weakness in 2 cases (9, 11). Similarly to our patient, spondylodiscitis was diagnosed 9 years before the development of neurological complications in one case (11). Another patient developed paraparesis due to exacerbation of preexisting but recently diagnosed spondylodiscitis and this paraparesis occurred while he was treated by anti-TNF-α agent (infliximab) for advanced AS (12). Spinal compression in AS patient with discitis may result from bone instability or granulation tissue growing from the epidural space. In the 5 cases from the literature search, only 3 had disco-vertebral histological analysis, showing growing fibrous or adipose tissue and new bone formation. Only one case was associated with predominant inflammatory lesions (11). In our case, no inflammatory changes were observed but fibrous and reactive bone tissues. It is though that the development of such tissues is directly responsible for spinal cord damage. Inflammatory infiltration of the epidural space without contiguous discovertebral changes has also been described in AS (2, 13). On the other hand, vertebral instability may be the main mechanism for neurological complication associated to aseptic spondylodiscitis (12). However, it is noteworthy that such neurological complications are very exceptional in the course of aseptic discitis in AS and that the histological features did not differ significantly between patients with or without spinal compression. Finally, neurological complication may occur several years after the diagnosis of spondylodiscitis, as it was observed in our patient and in another case (11). There is no evident explanation and particularly no trauma, but mechanical factors could probably better explain this late complication of preexisting spondylodiscitis, rather than uncontrolled disease activity.

Therefore, aseptic spondylodiscitis may induce neurological complications both at its initial phase or secondarily, after a variable delay. In addition, infective spondylodiscitis, a differential diagnosis of aseptic spondylodiscitis, may also develop into neurological involvement. Thus, clinicians must be aware of this late complication of aseptic spondylodiscitis in AS.

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