STRESS FRACTURE OF THE FUSED LUMBO-DORSAL SPINE IN ANKYLOSING SPONDYLITIS

A Report of Three Cases

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According to Romanus and Ydén (1955) the first description of a locally destructive lesion involving both surfaces of a vertebral body in ankylosing spondylitis was in an article by Andersson (1937) reporting two cases. Since then there have been occasional reports of similar lesions, notably by Edström (1940) and Forestier, Jacqueline and Rotes-Querol (1956). Both of these authors commented on the radiological resemblance to tuberculosis, but the pathology of the "destructive lesions" was not established. In 1952 Baggenstoss, Bickel and Ward reported four patients in an article entitled "Rheumatoid granulomatous nodules as destructive lesions of vertebrae", and presented a detailed histological account based, however, on the single case of rheumatoid arthritis; the other three were cases of ankylosing spondylitis in which no histology was available. In their own words "the lesions were presumed to be caused by similar rheumatoid granulomatous bodies". That this presumption is erroneous was well stated by Lorber, Pearson and Rene (1961), who reported another histologically documented case of rheumatoid granulomatous nodules found in vertebrae. In their equally well documented case of ankylosing spondylitis "a different type of destructive lesion was found", and they pointed out that these lesions "are not nearly so rare as they are in rheumatoid arthritis". In 1969 Kanefield and his colleagues described three cases in ankylosing spondylitis and gave good evidence that the lesion might represent a response to delayed union or non-union of a fracture.

In the last three years we have seen three cases in ankylosing spondylitis that lend support to Kanefield's proposition. We report them in view of their relative rarity, and explain why we believe these lesions are not truly destructive but rather a failure to reconstruct as in non-union.

CASE REPORTS

Case 1—A Chinese man aged twenty-eight presented in May 1969 at Queen Mary Hospital with a four years' history of low back pain which had become persistent. He complained of no radiation of the pain, which was aggravated by walking. There was no history of pyrexia or of night sweats, no urinary symptoms and no loss of weight. There were no complaints regarding the peripheral joints.

Physical examination revealed a rigid lumbar spine with a tilt to the right and tenderness over the lumbo-sacral area. Straight-leg-raising was 70 degrees on the right and 80 degrees on the left. There were no neurological signs. Haematological investigations were negative apart from an erythrocyte sedimentation rate of 36 millimetres in the first hour. Urinary examination was negative, as were radiographs of the chest.

Radiographs of the spine and sacro-iliac joints showed the classical appearances of advanced ankylosing spondylitis, with extensive calcification of the ligamentum flavum and of the interspinous and paravertebral ligaments. The lumbar vertebrae were osteoporotic. The adjacent surfaces of L2 and 3 showed extensive destruction with subjacent sclerosis (Fig. 1). The disc space was not narrowed. Tomography revealed bone eburnation and disc calcification but no sequestration.

A diagnosis of both ankylosing spondylitis and infective spondylitis, probably tuberculosis, was made.

At exploration via the anterior approach in June 1969 no sign of inflammation at the site of the lesion was found. The space between L2 and 3 was filled with a mass of scar tissue; when this was excised some degenerate nucleus pulposus was found. Spreaders were inserted and a little movement
between the two vertebral bodies was demonstrated. An autogenous iliac graft was inserted across the L.2-3 space.

Comment (made at the time of operation)—‘There does not seem to be any evidence of infection at the site of the lesion and most likely this was a stress fracture of the ankylosed spine.’

Fig. 1
Case 1—Antero-posterior and lateral views taken in May 1969 showing the lesion at the L.2-3 interspace.

Fig. 2
Case 1—A photomicrograph of the biopsy specimen showing a typical area of proliferation of connective tissue cells and collagenisation. (Haematoxylin and eosin, ×180.)

Full bacteriological investigation of the material obtained at operation was entirely negative. The histological report read as follows: ‘Fibrocartilage and neighbouring bone show areas of degeneration and devitalisation. Proliferation of connective tissue cells and collagenisation are seen. Many vascular spaces are present in the proliferating connective tissue. Chronic inflammatory cells and macrophages are present in the granulation tissue in the narrow cavity.’ (Fig. 2.)
Case 1. Figure 3—A film taken in October 1969 showing sound fusion between L.2 and L.3 and now a defect between the spinous processes of L.3 and L.4. Figure 4—A lateral view taken in July 1970 showing sclerosis around the posterior defect and the beginning of a lesion of the L.4-5 interspace.

Case 1—Films taken in November 1971 showing the lesion now well established both anteriorly and posteriorly.
The patient was treated by immobilisation in a plaster corset for six weeks. At follow-up eighteen months after the operation he had long since returned to work but had had a return of low back pain for a few months. Radiographs now revealed a well-established new lesion between the third and fourth lumbar vertebrae. A careful review of the previous films then showed the following sequence of events. By October 1969 a defect in the posterior element over L.4 could be clearly seen, though the L.4–5 disc space was unchanged (Fig. 3). By July 1970 sclerosis around the space was recognisable (Fig. 4), and by November 1971 the classical appearance of a “destructive lesion” had developed (Fig. 5). Meanwhile the L.2–3 disc space had remained soundly fused.

This man was seen in June 1972 when clinically and radiologically there was no change and he had not stopped work. A further radiograph in September 1973 showed sound fusion at the L.2–3 level.

Case 2—A Chinese mechanic aged forty-five was first seen at Queen Mary Hospital in November 1969 with a ten years’ history of progressive stiffness and kyphosis of the dorsal spine, as well as an eighteen months’ history of numbness along the anterior aspect of both thighs aggravated by movement and relieved by rest. A corset and analgesics had proved ineffective. He had been an opium addict for twenty-five years.

Clinical examination revealed a slight kyphosis at the level of T.12 and L.1 with no local tenderness. Movement of the spine was however diminished due to associated pain. There were no abnormal neurological findings. Haematological investigation was negative apart from an erythrocyte sedimentation rate of 34 millimetres in the first hour.

Radiography of the dorsal spine showed osteoporosis with obliteration of the costo-vertebral articulations and paraspinal ossification. The sacro-iliac joints were fused. There was partial fusion of the symphysis pubis and irregular hyperostosis of the inferior margins of the ischial tuberosities and of the trochanteric regions. The lumbar spine showed opposing marginal destruction of T.12 and L.1 with marked sclerosis and osteophyte formation (Fig. 6). The rest of the lumbar spine showed paraspinal and ligamentous calcification which was also found in the several interspaces between L.1 and L.4. Again a diagnosis of both ankylosing spondylitis and infective spondylitis, probably tuberculous, was made.

Exploration via the anterior approach in November 1969 revealed large osteophytes between T.12 and L.1 and scar tissue filling up this space. There was movement between the two vertebrae. The scar tissue was excised and a cavity filled with serous fluid was found on the far side of the interspace. Again there was no evidence of inflammation. The avascular bone was excised from the adjacent surfaces of T.12 and L.1 and an autogenous iliac graft was inserted.

After the operation the paraesthesia of the thighs disappeared but he was then lost to follow-up, due no doubt to his addiction to opium. However, a radiograph taken in September 1973 showed sound fusion at the T.12–L.1 level.
Case 3—A Chinese mechanic aged forty-one was first seen at Queen Mary Hospital in July 1971, with a nine years' history of a progressive kyphosis with spinal pain and dyspnoea on exertion.

Examination revealed a rigid spine with a generalised kyphosis from the sacrum upwards. The chest expansion was one centimetre. There were no other positive findings. Haematological examination was negative apart from an erythrocyte sedimentation rate of 44 millimetres in the first hour.

Radiology confirmed the clinical diagnosis of ankylosing spondylitis. No destructive lesion could be seen (Fig. 7).

![Case 3-Antero-posterior and oblique views of L.2 and L.3 showing well established ankylosing spondylitis but no sign of a fracture or destructive lesion.](image)

Operation was undertaken to correct the kyphosis in July 1971. The L.2–3 space was approached extraperitoneally; the annulus was not fused, disc remnants were seen and the spine was mobile at this level. The incision was then extended posteriorly towards the midline to reveal the posterior elements and a mobile fracture was found in the posterior elements of L.2 and 3 (Fig. 8). There was no evidence of inflammation. After wedge excision of the posterior elements of L.2 and 3 just below the fracture site, removal of disc remnants and correction of the kyphosis, an iliac graft was placed in the disc space.

He wore a plaster corset for the immediate post-operative period and at follow-up in December 1971 he was well with no complaint of pain except after walking for a long distance. A radiograph taken in September 1973 showed sound fusion at the L.2–3 level.
DISCUSSION

Fracture of the ankylosed spine (Stiasny 1933; Bergmann 1949; Martel and Page 1960; Weiss and Freehafer 1964; Grisolia, Bell and Peltier 1967) was first reported by Abdi in 1903, who referred to a fracture of the lumbar spine, while that of “a destructive lesion” has been attributed to Andersson by Romanus and Ydén (1955). Since then both topics have been reported infrequently. Most authors point out that “the ankylosed spine breaks like a long bone, transversely, as a result of a bending force” (Bergmann 1949, Woodruff and Dewing 1963). All the fractures occurred through what had been the interspace rather than through the substance of the body itself. Unlike the cervical spine, fracture of the thoraco-lumbar spine in ankylosing spondylitis is rarely reported, whereas “destructive lesions” are relatively frequent (Wholey, Pugh and Bickel 1960). The reverse is equally true of the cervical spine. There is no logical reason why this should be so unless we are dealing with the same condition. The only difference is that whereas in the cervical spine one sees it at the acute stage, in the thoraco-lumbar spine it is seen at a late stage with established non-union. A good reason for this is that in cervical injuries spinal cord injury and death are common, being eleven and ten respectively out of twenty in Woodruff’s series (1963).

These destructive lesions can heal. Romanus and Ydén point out that the destructive phase is followed relatively soon by a reparative phase that may result in complete healing. Because radiographs are taken at infrequent intervals the intermediate phases may be undetected and are therefore rarely seen. There is good reason to believe that these lesions occur more often than the literature would lead one to believe. Seaman and Wells (1961) reported eleven patients with destructive lesions in a survey of 110 patients with ankylosing spondylitis. We have seen three in three years.

One outstanding difficulty in reviewing the past literature is the fact that some authors have included in their studies both rheumatoid arthritis and ankylosing spondylitis (Baggenstoss et al. 1952, Martel and Page 1960, Lorber et al. 1961). Because the pathogenesis of both diseases is not completely understood and because the clinical, haematological, histological and radiological pictures show distinct differences (Edeiken, DePalma and Hodes 1964), it is wise to consider them separately. We believe the result of this confusion is that the lesion seen in ankylosing spondylitis has been described together with that seen in rheumatoid arthritis as “destructive”.

The only two histologically proven cases of rheumatoid nodules found in the vertebrae that were indeed destructive were reported by Baggenstoss et al. and by Lorber et al. Both patients died, and rheumatoid lesions were found in more than one vertebrae. Radiologically the appearances were different from those of ankylosing spondylitis. On the other hand the histology of the “destructive lesion” of ankylosing spondylitis as reported by Lorber et al. and Kanefield et al., and in our Case 1, shows a very different picture, which, as Kanefield and his colleagues point out, can be interpreted as “fibrous union of a fracture”. In addition we have had the opportunity to observe the macroscopic appearance of these lesions in our three cases because we adopted the anterior approach for spinal fusion. In all three cases we found no evidence of inflammation and at the site of the lesion only fibrous scar tissue with sclerotic bone. In Case 1 an infective etiology was ruled out by culture and guinea-pig inoculation. Movement was demonstrated in all three cases and in Case 3 an obvious fracture was seen. All three patients had lumbar spines which were otherwise truly ankylosed.

Case 1 also shows firstly that these lesions heal or unite following inter-body fusion by bone graft, and secondly, as opposed to the opinions of some authors (Romanus and Ydén 1955, Seaman and Wells 1961), that the process starts in the posterior elements and only later involves the anterior portion of the adjacent surfaces of two vertebrae and their disc space. Indeed a posterior fracture was found in Case 3 before it had become apparent radiologically. In other words the pseudarthrosis as such is of slow onset, like a fatigue fracture, and precedes the destructive phase. If a lesion is successfully fused, fracture and later
another destructive lesion are often seen above or below the fused vertebral space. Case 1 in this report and the Kanefield paper bear this out. This is hardly surprising. Our case reports therefore suggest that the etiology of “destructive lesions” in ankylosed spines is initially a stress fracture of the posterior elements, which if it fails to heal or unite results in a pseudarthrosis between two vertebral bodies.

The ideal surgical treatment is anterior fusion at the site of compression force rather than posterior fusion at the site of distraction. The latter can fail (Kanefield et al. 1969) while the former succeeds, as in this series. Fusion by the anterior route also permits observations on the pathology, both macroscopic and microscopic.

**SUMMARY**

1. The aetiology of a destructive lesion through a former interspace in a spine fused by ankylosing spondylitis is reviewed.

2. From the findings in three patients treated by anterior spinal fusion, evidence was obtained to show that a stress fracture, originating posteriorly between two fused spinous processes, leads to a pseudarthrosis between two vertebral bodies that may simulate a tuberculous lesion.

3. If spinal fusion is indicated, the anterior approach is recommended, both for direct observation of the lesion and to achieve sound union.

**REFERENCES**


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