BRUCELLA SPONDYLITIS

Review of the Literature and Report on Two Cases

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The object of this paper is to draw attention to the bone and joint complications of brucellosis, to review the literature and to report on two cases.

In the Milroy lectures for 1950, Dalrymple-Champneys (1950) estimated that at least 400 to 500 cases of brucellosis occurred annually in Great Britain. According to other sources, the incidence of bone and joint complications of brucellosis is said to be about 30 to 40 per cent. A correspondingly high percentage of these complications could, therefore, be expected to occur in this country.

Gardner, Girdlestone and Gillespie (1932) reported brucella bone abscesses, and O'Donoghue (1933) reported septic arthritis, all bacteriologically confirmed by culture of the pus after drainage. Kulowski and Vinke (1932) reported a case of 'undulant fever spondylitis' with psoas abscess which was drained and yielded brucella melitensis. This was claimed to be 'the first proved case to be reported of brucella melitensis involvement of the human spine.' Kulowski (1936) gave the incidence of 'arthropathies' as being from 17 per cent. to 32 per cent. and stated that 'bone lesions also have been noted but stressed very little.' An unusual case was reported by Edward (1937) of an abscess around a plated femur from which a culture of brucella was obtained.

In an extensive review of the literature, Palagi (1934) collected 29 cases of spinal brucellosis to which he added 17 of his own. Bishop (1939) stated that of 56 cases of spinal brucellosis, 40 were situated in the lumbar vertebrae, 14 in the thoracic vertebrae and two in the cervical vertebrae. He also reported a further case involving both lumbar and cervical vertebrae, and drew attention to the characteristic lipping of the anterior vertebral border on the X-ray films. He also stated that only 12.5 per cent. of spinal cases developed an abscess. Four more cases of spondylitis due to

brucellosis, one of which developed an abscess were reported by Steindler (1940).

Reviewing the literature from 1906 to 1942, Dobelle (1943) found 64 cases of brucella spondylitis to which he added one of his own. This patient developed a psoas abscess and required spinal fusion, being only the fourth reported case to be treated in this way. Phalen (1942) stated that 'spondylitis is probably the most common complicating disorder of the bones referable to undulant fever.' In a comprehensive study, Pillmore (1946) gave the frequency of bone and joint involvement as being about 40 per cent., which agrees with the apparently independent calculations of Rimbaud and Serre (1947). Pillmore also pointed out the difficulties of the differential diagnosis as between brucella bone lesions on the one hand and pyogenic osteomyelitis and tuberculous bone and joint disease on the other. Brucellosis and its bone and joint complications were regarded as commonplace by Rimbaud and Serre. Of the 40 per cent. of osteoarticular manifestations they differentiated five types of which the spinal form may affect either the interarticular facets of the vertebral bodies, simulating Pott's disease. They stressed the importance of conservative treatment and considered that surgery was contra-indicated except in cases of abscess formation which they regarded as rare.

The radiological appearance of the osteoarticular manifestations of brucellosis of the spine were discussed by Bétoulières and Maléki (1947) who found that 30 of their 200 cases showed X-ray evidence of bone involvement and that 10 of these were located in the lumbar spine.

Janbon *et al.* (1950) reported 16 cases of cervical brucella spondylo-arthritis. Photographs of their original X-rays showed the typical diminution or obliteration of the intervertebral disc space, and later osteophyte formation confined to the anterior aspect of the vertebral body. Seven of these

cases showed nerve root involvement. Antelava (1951) observed 489 cases of brucellosis and their surgical complications. Spinal involvement occurred in 15 per cent. of them.

Case 1

On January 12, 1951, an Indian seaman, aged 36, was admitted complaining of vague abdominal pain, mainly in the right hypochondrium, and of occasional vomiting and general malaise for one month. For two weeks he had had pain in the left elbow.

Physical examination showed a moderately ill, thin man. T., 99° F. P., 88. The cardiovascular and respiratory systems were normal. The liver was tender and smoothly enlarged to two fingers breadth below the right costal margin. The spleen was not palpable. There was no lymphadenopathy. The left elbow was painful but showed no other abnormality and movements were full.

The urine showed no abnormality, the lung fields were radiologically clear and blood investigations gave the following results: E.S.R. (Wintrobe), 25 mm. in one hour; P.C.V., 42 per cent.; W.B.C., 8,800 with normal differential count; Wassermann and Kahn reactions negative; serum bilirubin, 0.5 mgm. per cent. The stool contained vegetative forms of *entamoeba histolytica*.

A provisional diagnosis of amoebiasis with hepatitis was made and treatment with emetine, 1 gr. intramuscularly daily was started. Although the liver tenderness disappeared the temperature, which had been fluctuating between 97.4° F. and 101° F., failed to settle. Brucella agglutinations were performed and found to be significantly raised, viz.: Br. abortus 1:3,840, and Br. melitensis 1:960. A blood culture grew Br. melitensis after 18 days and the organism was sensitive to penicillin, streptomycin, chloramphenicol and aureomycin *in vitro*.

A course of aureomycin, 3 g. initially, followed by 0.5 g. six-hourly, was given for 10 days from January 25, 1951, and his temperature settled. On January 28, 1951, he first complained of low back pain which gradually increased in severity. Examination showed marked tenderness and stiffness of the lumbar spine with spasm of the erector spinae muscles. Examination of the cerebrospinal fluid showed no abnormality; Wassermann reaction was again negative.

X-rays of the lumbar spine showed marginal erosion at the right antero-superior border of the body of the fourth lumbar vertebra (Fig. 1). The psoas shadow and disc spaces appeared normal. Further X-rays at monthly intervals showed some increase in the small area of erosion. The latest film on November 28, 1951, showed new bone forming an osteophytic spur adjacent to the area

FIG. 1.—Case 1. Oblique view of the spine showing erosion and some new bone formation on supero-anterior margin of L.V.4.

of erosion and some diminution of the intervertebral disc space L 3-4 (Fig. 2).

Since his back pain did not respond to bed rest alone he was put in a plaster bed. He then complained of pain in the neck and an X-ray showed compression of the bodies of the fifth and sixth cervical vertebrae with diminution of the intervening space (Fig. 3). The plaster bed was extended to include the head and neck and he remained immobilized for four weeks. After this he was symptom-free. The temperature had remained normal throughout the period of immobilization and he was allowed up.

On April 30, 1951, the E.S.R. was 36 mm. in one hour and agglutination titres were: Br. abortus 1:7,680, Br. melitensis 1:3,840. He had one further rise in temperature to 101° F. with pains in the back and extremities, and though the temperature settled spontaneously within four days the pain increased and further immobilization was necessary. A second course of aureomycin was given for 12 days. By June 30, 1951, he was

FIG. 2.—Case 1. Ten months after the onset of symptoms, showing osteophytic spur formation.

FIG. 3.—Case 1. Showing erosion and lipping of anterior borders of C.V.5 and 6 with considerable diminution of intervertebral disc space.

again symptom-free, was fitted with a Taylor brace and allowed up. He was discharged from hospital on July 26, 1951.

When seen four months later he had remained symptom-free. His agglutination titres were now: Br. abortus 1:960, Br. melitensis 1:120, and the E.S.R. normal at 3 mm. in one hour.

Case 2

On April 30, 1950, a man aged 30 was admitted to the Newmarket General Hospital under the care of Dr. R. Arden Jones. The main complaint was low back pain which had been associated with loss of weight, feverishness and sweating at night for several weeks.

Lumbar pain, which had radiated to the back of both knees, had first been noticed in November 1949, and improvement had followed physiotherapy. He had had a heavy cold with slight haemoptysis in February 1950, but chest X-ray had shown no abnormality.

Physical examination showed a pale, ill-looking,

well-built man. T., 99.2° F. P., 88. The cardiovascular and respiratory systems were normal. There was a well-defined, firm, tender swelling, 1½ in. in diameter, visible below the left inguinal ligament and an ill-defined swelling in the left iliac fossa just above the ligament. The spleen was palpable 1 in. below the left costal margin. Spinal movements were full and painless and straight leg raising was normal. There were no abnormal neurological signs.

The urine showed a trace of albumen, the lung fields were radiologically clear. X-rays of the lumbar spine showed narrowing of the disc spaces between the fifth lumbar and first sacral vertebrae, and bulging of the left psoas shadow. Blood investigations were as follows: E.S.R. (Westergren), 79 mm. in one hour; Hb., 80 per cent.; R.B.C., 4,270,000; W.B.C., 9,500 with normal differential; Br. abortus agglutinations were significantly raised to 1:1,000; blood culture (on two occasions) was sterile; Wassermann and Kahn reactions negative. Mantoux test negative to 1:100.

A course of chloramphenicol, 3 g. initially, followed by 0.5 g. six-hourly, was given for 16 days, combined for the first seven days with sulphadiazine, 5 g. daily. At first on complete bed rest, he was later put in a plaster bed. The temperature remained raised, the left inguinal swelling increased in size and became more painful. It was aspirated on May 26, 1950, and thick, glairy, slightly purulent fluid removed. A direct smear showed no organisms with either Gram or Ziehl-Nielsen stain and both aerobic and anaerobic cultures were sterile.

A course of penicillin, 60,000 units three-hourly, was then given with no apparent effect, and the abscess was drained. Culture of the pus was again sterile and guinea pig inoculation gave no evidence of a brucella or tuberculous lesion. The W.B.C. remained normal. Br. abortus agglutinations were now 1:5,000, and Br. melitensis 1:1,000.

Aureomycin was now given, 1 g. initially followed by 0.5 g. six-hourly for ten days, with no apparent improvement, low-grade fever persisting. A catheter was passed 12 in. up the sinus and opaque oil instilled. X-rays showed a cavity opposite the transverse process with some erosion of the lower border of the fifth lumbar vertebra. Curettage and biopsy of the sinus tract showed no evidence of tuberculosis.

On July 25, 1950, frank pus, growing staph. aureus on culture, was being discharged from the left inguinal sinus and an ill-defined swelling was now present on the right side posteriorly at the level of the third and fourth lumbar vertebrae. The sinus cleared and the swelling became smaller following a further course of penicillin, but on September 7, 1950, it recurred. As no improvement followed a further course of aureomycin, 0.5 g. six-hourly for 12 days, aspiration was performed and 10 oz. of thick, yellow, sterile pus removed from the right lumbar region. The E.S.R. was now 44 mm. in one hour and Br. abortus agglutinations had risen to 1:10,000.

Improvement now started, the fever settled, he became free from pain and the sinus began to heal. To be nearer home he was transferred to the Epsom District Hospital under the care of Mr. D. O. Davies.

He remained in a plaster bed for several more months. The sinus remained healed apart from a few days in January 1951, when pus was again discharged, containing staph. aureus on culture. Radiologically there was some progression of the lumbar lesion, there now being some erosion of the first sacral segment in addition to the previous findings (Fig. 4). In July 1951 a plaster jacket was applied and the patient has since been ambulant. The plaster was removed on November 3, 1951,

FIG. 4.—Case 2. Showing erosion of anterior border of S.V.I. Traces of opaque oil are seen adjacent to the lesion.

and X-rays showed some regeneration of bone in the previously rarefied area. His general condition was satisfactory and he was free from pain with an E.S.R. of 16 mm. in one hour. No further immobilization was considered necessary and he was allowed gradually increasing activity.

Discussion

A study of the literature shows that during the past 20 years many reports on the bone and joint complications of brucellosis have appeared in the American and continental literature. In the same period only two reports were found in the British literature on this subject, and only one case of brucella spondylitis was mentioned by Dalrymple-Champneys among the 980 cases he collected.

Minor non-specific X-ray changes in the vertebral bodies, similar to those reproduced here, are frequently seen in patients suffering from chronic backache of indefinite aetiology. These X-ray appearances, though sometimes indistinguishable from osteoarthritis, tuberculosis, etc., are said to

be consistent with a diagnosis of brucella spondylitis. It seems, therefore, that brucellosis should be considered as a possible aetiological factor in chronic spinal lesions and that a history of a febrile illness in conjunction with specific laboratory tests (blood cultures, agglutinations, etc.) and repeated X-ray examinations might reveal a higher incidence of this disease than published reports lead us to believe.

Some of these points are illustrated in the two cases reported. The time of onset of back pain is variable, it may apparently be the presenting symptom, as in Case 2, or it may accompany or follow the febrile stage of the illness. In Case 1 the diagnosis was suggested by the raised agglutination titres to brucella abortus and melitensis and confirmed by the isolation of *Br. melitensis* from the patient's blood. The course was benign and there was no abscess formation. In Case 2 the diagnosis rested on the history of a chronic febrile illness, the rising agglutination titres in the blood, the absence of a pyogenic blood picture and the unusual character of the pus initially aspirated. Tuberculosis could virtually be excluded by repeatedly negative cultures and guinea pig inoculations, by the absence of tuberculous changes in the sinus biopsy and by negative Mantoux tests. Secondary infection with staph. aureus might have occurred after exploration of the sinus tract with a catheter.

Persistent localized pain is the main symptom and is accompanied by local tenderness and muscle spasm. It is of interest to note that the original symptoms in Case 2 suggested a prolapsed intervertebral disc (lumbar pain with bilateral sciatic radiation) and Harris (1950) states that this has previously been noted. Both cases showed the relatively common involvement of the lumbar spine, and in addition Case 1 had a cervical lesion resembling that described by Bishop.

Most authors agree that treatment should be conservative except in the presence of abscess formation. Our experience suggests that complete rest in a plaster bed is beneficial and that abscess formation may be treated by aspiration initially

and, if necessary, later by surgery. Antibiotic therapy should be started as early as possible. A report by Killough *et al.* (1951) on the treatment of brucellosis suggested that aureomycin, chloramphenicol and terramycin were of equal value, though aureomycin had more side effects. It has been suggested by Knight *et al.* (1950) that the antibiotic treatment of brucellosis should be prolonged for at least four to six weeks. Where bone complications occur and tend to run a slow course, it would seem logical to continue treatment for even longer.

Brucella spondylitis appears to be a self-limiting process leading to recalcification and spur formation at the site of the original lesion (as Fig. 2).

Summary

Bone and joint complications of brucellosis have been reported with increasing frequency in America and on the continent of Europe.

The reports suggest that about 40 per cent. of all cases of brucellosis show some form of bone or joint involvement and that spondylitis is one of the commoner manifestations.

Two cases of brucella spondylitis occurring in this country, one with abscess formation, are reported in full and their treatment outlined.

It is suggested that brucellosis should be considered as a possible cause of chronic backache and that a higher incidence of brucella spondylitis might thus be revealed.

Acknowledgments

We should like to thank Sir Hugh Griffiths and Dr. Ronald Hartley, under whom the first case was admitted, for much helpful advice and criticism. We are greatly indebted to Dr. R. Arden Jones of White Lodge Hospital, Newmarket, and Mr. D. O. Davies of Epsom District Hospital, for permission to include their case in this article and for their help in the preparation of the case report. We are indebted also to Dr. G. L. Robinson for the laboratory investigations and to Dr. I. L. McKelvie for the X-ray reports.

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