BRUCELLOSIS OF THE CARPUS

Report of a Case

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Brucellosis is typically a systemic febrile illness and has been well documented since first described in 1861 (Marston 1863). Bone and joint brucella infection is rare; the first case was recorded in 1932 by Kulowski and Vinke. Since then sporadic reports have appeared, these implicating *Brucella melitensis* more commonly than *Brucella abortus*. Infection may present as osteomyelitis or arthritis. Vertebral bodies, particularly in the lumbar region, are the usual bones involved, but almost any bone may be affected (Steindler 1940; Lowe and Lipscomb 1947; Adam, Macdonald and Mackenzie 1967). Brucellosis of the carpal bones is very unusual (Botreau-Roussel and Huard 1931; Hardy, Jordan and Borts 1936; Janbon and Bertrand 1953; Arct 1970). Arthritis typically affects large joints, the hip being most commonly affected (Coventry, Ivins, Nichols and Weed 1949). Monarticular arthritis with associated osteomyelitis involving a peripheral joint is much rarer. Prepatellar bursitis (Johnson and Weed 1954) and subacromial bursitis (Kennedy 1904) also occur.

Lowbeer (1949) first described the histology of brucella osteomyelitis as characterised by 1) granuloma with mononuclear reaction; 2) large foreign body type giant cells of bizarre appearance, though some resembling Långhans' giant cells; 3) widespread caseous necrosis (involving the spine in three of his four cases); and 4) scar formation and repair with new bone formation.

CASE REPORT

A farmer aged forty-eight gave a six weeks' history of a painful swelling of the right wrist. There was tenderness over the radial styloid. Radiographs were normal (Fig. 1), as were blood investigations: haemoglobin 15.2 grams per cent, leucocytes 7,000 per cubic millimetre (37 per cent lymphocytes), Westergren erythrocyte sedimentation rate 10 millimetres in the first hour, urea 40 milligrams per cent, uric acid 6.9 milligrams per cent, calcium 9.2 milligrams per cent, protein 6.5 grams per cent, albumin 4 grams per cent, alkaline phosphatase 18 King Armstrong units. A diagnosis of a tenosynovitis was made. The wrist was immobilised for six weeks and the patient was then discharged symptom-free.

Three and a half months later he returned with a history of six weeks' trouble in the right wrist, which was warm, stiff, painful to move and swollen. A radiograph now showed erosions of the carpus (Fig. 3). A provisional diagnosis of monarticular rheumatoid arthritis was made. Investigations showed: haemoglobin 14.8 grams per cent, leucocytes 6,000 per cubic millimetre (lymphocytes 58 per cent), erythrocyte sedimentation rate 38 millimetres in the first hour. The sheep cell agglutination test for rheumatoid arthritis was negative, and a protein electrophoretic strip was normal. Because of the patient's occupation, serological tests for brucellosis were done. The agglutination tests for *B. abortus* were positive at 1/400 in phenol saline and in 2-mercapto-ethanol and the complement fixation test was positive at 1/320. Blood culture was negative. A subsequent radiograph (Fig. 4) showed rapid progression of a destructive lesion in the carpus; so the wrist was explored.

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Operation—The carpus was found to be surrounded by thickened greyish synovium. The hamate was a mere shell containing yellowish granulation tissue. Granulation tissue and hypertrophied synovium were removed and material was sent for laboratory examination. *Brucella abortus* (Biotype I) was isolated after nine days' incubation of tissue in glucose liquid broth.

FIG. 1

FIG. 2

FIG. 3

FIG. 4

FIG. 5

Radiographs of the right wrist. Figure 1 shows the synovial stage of the infection. There is slight synovial swelling shown as displacement of fat pads on the medial and lateral margins of the wrist. Figure 2 shows the active erosive stage one month later, with subarticular erosions of the hamate and pisiform bones and rarefaction of the lunate and the tip of the hamate. Figure 3 shows the healing stage six months after Figure 1 was taken; the changes are now confined to adjacent subarticular margins of the hamate and capitate bones. Figure 4 shows articular reactivation complicated by osteomyelitis one month later. There is destruction of the hamate and lunate bones and there is extensive intercarpal erosion. The distribution of subarticular involvement is interesting, and in all areas involves opposing joint surfaces (*arrowed*). Figure 5 shows the healing stage again. The radiograph had a direct magnification $\times 4$ and was taken nine months later, after surgery and antibiotic treatment. Consolidation of the remaining portion of the hamate bone has occurred. Slight subarticular defects are still present. There is no extension of the disease process.

Histological features—The cortical bone showed lysis by osteoclasts, with new bone in other areas. The marrow cavity contained granulation tissue, showing a subacute inflammatory reaction of lymphocytes, plasma cells, histiocytes and smaller numbers of polymorphonuclear leucocytes. The synovium showed a more florid reaction, with the cell types already described, and also areas of large epithelioid cells resembling those of tuberculosis but without the

typical follicular arrangement. Multinucleate giant cells were frequent, but only one Länghans' type cell was seen. Lymphoid follicles, like those of rheumatoid arthritis, were also present. The appearances were those of a subacute granuloma, typical neither of chronic suppurative osteomyelitis, tuberculosis, nor rheumatoid disease.

Progress—The wrist was immobilised for ten weeks in plaster and thereafter by a block leather support which was discarded when the wrist was pain-free. Tetracycline was given for five months, together with streptomycin for six weeks. Nearly full wrist movement was regained.

After five months, the agglutinations to *B. abortus* had fallen to 1/200 in phenol saline and 1/50 in 2-mercapto-ethanol.

Radiological features—Synovial swelling around the wrist joint was visible in the first radiograph (Fig. 1). Subarticular erosions subsequently developed in several sites in the carpus (Fig. 2). Five months later the radiographic appearances showed improvement, with erosions confined to the contiguous subarticular zones of the hamate and capitate (Fig. 3). This was followed by a rapid extension of bone destruction with multiple joint erosions and osteomyelitis of the hamate (Fig. 4). After operation, consolidation took place in the hamate, but six months later minor subarticular erosive deformities remained. There was no ankylosis (Fig. 5).

DISCUSSION

Two to three hundred new patients with brucellosis are reported in England and Wales each year; infection is particularly likely in young men whose work brings them into contact with brucella-infected cattle, especially farmers and veterinary surgeons, or in persons who drink unpasteurised brucella-infected milk (*British Medical Journal* 1971). Infection acquired in this country is almost always caused by *B. abortus*.

In 1964 about one in three British herds were infected with brucellosis (Ministry of Agriculture, Fisheries and Food 1964), with the highest incidence in the West Midlands. The Ministry of Agriculture introduced a voluntary eradication scheme in 1967 and a compulsory scheme in 1971, and there is every hope that brucellosis will be eradicated from the British Isles within the next two decades.

In spite of the high incidence of infected herds in the West Midlands, acute brucellosis in humans is uncommon. The diagnosis is certain when the organism is isolated, but this is only achieved in one in four patients with the acute disease. Cultures from the blood or infected tissue are best collected by a pathologist who must use a medium known to support the growth of brucella (for example, liquid broth containing 0.1 per cent glucose). Cultures must be incubated immediately, preferably in an atmosphere of 10–14 per cent carbon dioxide in air, and they may have to be continued for eight weeks. Several daily blood cultures, bone marrow and liver cultures increase the chance of isolating the organism.

When cultures are negative it can be extremely difficult and sometimes impossible to determine the significance of serological results, particularly if, as in this case, the patient is known to have been exposed to brucella in the course of his occupation. A titre of 1/400 is common in farm workers and not necessarily indicative of active disease. High titres of agglutinating and complement fixing antibodies may be present in fit persons (Henderson and Hill 1972); conversely, even in cases proved by positive culture, the serum may contain no detectable antibodies. Serology is best regarded as a screening test only.

The histological features described by Lowbeer (1949) were present in our patient, but caseation was not conspicuous.

Radiological textbooks contain few illustrations of articular involvement in this disease, so that little is recorded of the changes to be expected in the course of this infection. In our patient, in the early stages, the appearances of synovial swelling and subarticular erosions were indistinguishable from rheumatoid disease or tuberculosis. Sequential radiographs showed regression and then dramatic progression of radiological changes. Subarticular erosions involving opposing joint surfaces were a constant feature (Fig. 4).

The present case illustrates the problem of diagnosis of a peripheral inflammatory monarticular arthritis in the adult. Tuberculous arthritis is now less common than the various non-suppurative inflammatory arthritides, though it must always be considered. Brucella arthritis is even more rare and, as with other diagnoses, the clinical presentation, course, and effect of immobilisation do not help to differentiate it. Knowing the patient's work may cause the clinician to suspect brucellosis, as in this case, but it is essential to confirm the diagnosis by culturing a biopsy specimen.

Chemotherapy against bone and joint brucellosis is similar to that used in systemic brucellosis. The drug of choice is tetracycline. Co-trimoxazole (Lal, Modawal, Fowle, Peach and Popham 1970) has been used with some success. The length of treatment depends upon the clinical progress and may have to continue several months.

SUMMARY

1. A farmer who owned cattle infected with brucellosis presented with a painful, swollen wrist. Osteolytic lesions were seen radiologically. The wrist was explored. Histological features were those of a subacute granuloma. Brucella abortus biotype I was grown.
2. The clinical diagnosis, bone and joint radiology, pathology and microbiology are discussed.

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