

Osseous sarcoidosis - A magnetic resonance imaging diagnosis

C.K. Rayner¹, S.P. Burnet,
J.D. McNeil

Department of Medicine, Modbury Public Hospital, Smart Road, Modbury, South Australia; ¹Department of Medicine, Royal Adelaide Hospital, Adelaide, South Australia, Australia.

Chris K. Rayner, MBBS, FRACP, Gastroenterology Clinical Fellow; Simon P. Burnet, MBBS, FRACP, Rheumatologist; Julian D. McNeil, MBBS, PhD, FRACP, Associate Professor of Medicine.

Please address correspondence to: Associate Professor J.D. McNeil, Department of Medicine, Modbury Public Hospital, Smart Road, Modbury, South Australia 5092, Australia.

Key words: Osseous sarcoid, diagnosis, MRI.

ABSTRACT

Bone involvement in sarcoidosis is not uncommon but may be overlooked as a cause of symptoms. Magnetic resonance imaging (MRI) is emerging as a sensitive diagnostic tool for osseous sarcoid. We document a case in which MRI suggested the diagnosis in the absence of abnormality with more conventional imaging techniques.

Case report

A 46 year old male presented to the Rheumatology clinic for assessment of bilateral groin discomfort. His past medical history included sarcoidosis, diet-controlled Type II diabetes mellitus, hypertension, hypercholesterolaemia and mild asthma. He was a non-smoker.

Sarcoidosis had been diagnosed three years earlier. A needle biopsy of an enlarged right preauricular lymph node had demonstrated non-specific granulomatous inflammation, suggestive of sarcoidosis. The following year he had complained of enlarging bilateral inguinal lymphadenopathy. Excision biopsy of a left inguinal node revealed nonnecrotizing granulomas with epithelioid and multinucleated giant cells, again consistent with sarcoidosis. The serum concentration of angiotensin converting enzyme at the time was 152 (range 41-139) U/L. He had complained of an intermittent productive cough with mild dyspnoea, but a chest radiograph was normal. Pulmonary function tests, however, had revealed a restrictive defect, with FEV 2.25 L (73% predicted), FVC 2.4 L (62% predicted), and no reversibility after bronchodilator. Persistent groin discomfort had been present for 12 months prior to review, worse on walking, and sufficient to interfere with work. A plain radiograph of the hips and pelvis was unremarkable, and computerised tomography (CT) of the lumbosacral spine showed only minor degenerative changes with minimal posterior disc bulging at L4-5. A CT scan of the chest showed extensive mediastinal and bilateral hilar lymphadenopathy, and nodular opacities in both lungs, and he had continued to experience minor respiratory symptoms and pulmonary function

tests were unchanged.

Prednisolone was commenced at a dose of 50 mg daily, with close attention to monitoring blood glucose levels. Over the 12 months prior to presentation, the average dose of prednisolone required was 25 mg daily, with improvement in pain on higher doses and return of symptoms on lower doses. Azathioprine had been added after 7 months but no reduction in the effective steroid dose had been achieved.

Assessment in the Rheumatology Clinic revealed bilateral inguinal lymphadenopathy, with an especially prominent right groin node. Examination of all joints, including the hips revealed a full range of movement and no evidence of synovitis, and there was no objective neurological abnormality. Serum calcium was 2.36 (range 2.10-2.55) mmol/L, alkaline phosphatase 63 (range 30-120) U/L, phosphate 1.26 (range 0.80-1.40) mmol/L and prostate specific antigen < 1.0 g/L. Serum protein electrophoresis was normal, and urinary Bence Jones protein was not detectable. A plain radiograph of both hips (Fig. 1) was again normal and a bone scan of pelvis and hips (anterior and posterior) using technetium 99m pyrophosphate was unremarkable (Figs. 2 and 3). Magnetic resonance imaging (MRI) of the pelvis and hips revealed a strikingly different picture (Figs. 4 and 5). Numerous areas of altered signal were clearly identified in the bone marrow, with lesions of low intensity on T1 and high intensity on T2 weighted images in both femurs. A diagnosis of osseous sarcoid was made on the basis of the clinical picture and MRI appearance, despite the normal plain radiographs and scintigraphy. A repeat MRI one year later showed a similarly dramatic, though virtually unchanged appearance. The patient continues to have variable bilateral groin discomfort for which he is maintained on low-dose prednisolone and azathioprine.

Discussion

Reports as to the prevalence of bone lesions in sarcoidosis vary between series, with a range of 1-14% (1-4) and an average of 5% (5), although this may be an underestimate given that bone



Fig. 1. Plain radiograph of the pelvis.

involvement is often asymptomatic (3). The tubular bones of the hands are most frequently involved (6), followed closely by those of the feet (2, 3, 5). Less common sites include the long bones (7), vertebrae (especially low thoracic and high lumbar) (5, 6), nasal bones, skull and sternum (8). Sites of bony involvement may be localised or disseminated (3). 80-90% of patients with bone involvement have associated pulmonary disease (5). Most have

chronic skin lesions, especially lupus pernio (8). Ocular and nasal mucosal disease is said to be more common than in other patients with sarcoid (7). Many patients are asymptomatic from sarcoid bone lesions, but bone pain may be experienced by up to 50% (2). Disease involving hands and feet is less likely to be painful, though some stiffness may occur (5-8), whereas vertebral involvement is usually painful (6). Soft tissue swelling may overlie the site of

bony disease, may contain granulomas on histologic examination and pathological fractures may occur (2, 8).

Plain radiographs of involved regions show variability and non-specific appearances (1). Lytic lesions are most common, including cyst-like areas and cortical defects, but areas of thickened cortex may also occur, with a reticular appearance (due to infiltration of granuloma into the Haversian systems) (2, 4). Sclerotic changes are seen less commonly, usually when the pulmonary disease is quiescent (5,9). Bone scans using ^{99m}Tc pyrophosphate are more sensitive for detecting osseous sarcoidosis than plain radiographs (2, 4), with affected areas appearing as "hot spots". Use of gallium-67 as the radioisotope is less sensitive than technetium, though gallium has a place in detecting pulmonary sites of disease in sarcoidosis (3, 10).

More recently, MRI has emerged as a sensitive diagnostic investigation for osseous sarcoid sometimes detecting disease in the absence of changes on the plain radiograph (7, 11). In both of these cases, however, technetium 99m scintigraphy revealed uptake in the clinically affected osseous site. Our case had both normal plain radiographs and scintigraphy, which is yet to be

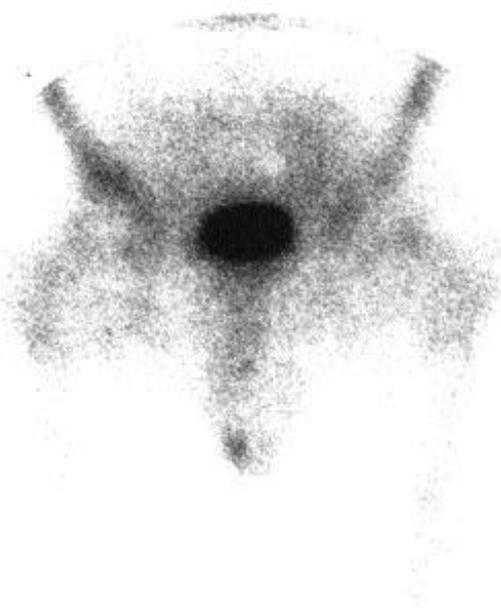


Fig. 2. Bone scan of the pelvis - anterior.

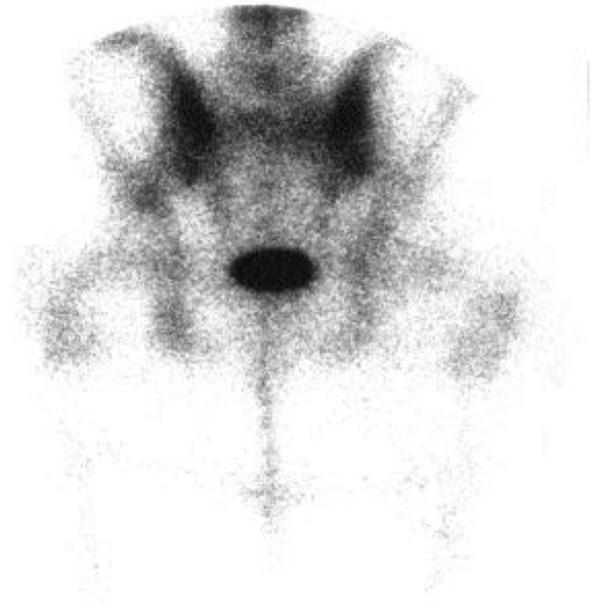


Fig. 3. Bone scan of the pelvis - posterior

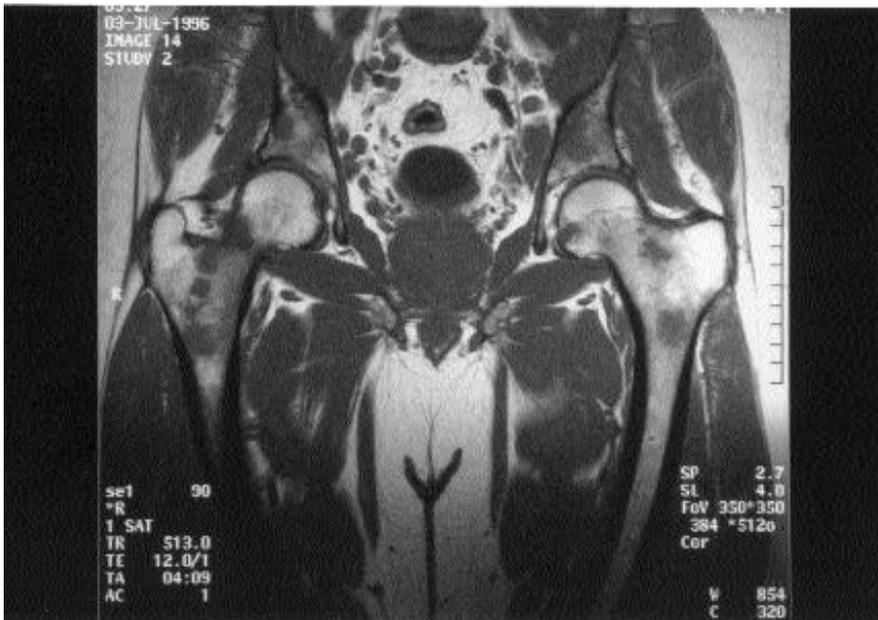


Fig. 4. T1 weighted MRI of the pelvis.



Fig. 5. T2 weighted MRI of the pelvis.

reported in the literature. T1 weighted images typically show hypointense lesions, which enhance to the intensity of adjacent marrow with gadolinium, while lesions are hyperintense with T2 weighting, sometimes with hypointense centre (5).

The radiological appearance of the bone lesions is non-specific, and similar changes may be seen with metastatic carcinoma, multiple myeloma, lym-

phoma, histiocytosis, osteomyelitis, tuberculosis, Paget's disease, traumatic bone lesions, bone infarction or fluorosis (1, 2, 4). Biopsy may be required, therefore, for definitive diagnosis.

Corticosteroids are indicated for the treatment of symptomatic osseous sarcoid; pain, disability, or deformity would be such indications. Healing of the lesions has been documented (9), but some series have suggested mortal-

ity four times greater than the general population of patients with sarcoidosis (8). Low dose methotrexate may be useful agent in osseous sarcoidosis, and resolution of lesions evident radiologically has been documented with this treatment (12).

Comment

While the plain radiographs and scintigraphy were unhelpful, the MRI appearance in the current case, in the context of the patient's illness and the behaviour of the lesions over time, would make the diagnosis of osseous sarcoid reasonable, without resorting to biopsy of an osseous lesion.

References

1. WILCOX A, BHARADWAJ P, SHARMA OP: Bone sarcoidosis. *Curr Opin Rheumatol* 2000; 12: 321-30.
2. SWEENEY DC, ROBERSON JC: Osseous and central nervous system sarcoidosis: Scintigraphic and radiographic findings. *J Nucl Med* 1995; 36: 464-6.
3. MANA J, SEGARRA MI, CASA R, MAIRAL L, FERNANDEZ-NOGUES F: Multiple atypical bone involvement in sarcoidosis. *J Rheumatol* 1993; 20: 394-6.
4. BEASLEY EW, PETERMAN SB, HERTZLER GL: An unusual form of tibial sarcoidosis. *Am J Roentgenol* 1987; 149: 754-6.
5. GOLZARIAN J, MATOS C, GOLSTEIN M, STALLENBERG B, DEPIERREUX M, STRUYVENJ: Case report: osteosclerotic sarcoidosis of spine and pelvis: Plain film and magnetic resonance imaging findings. *Br J Radiol* 1994; 67: 401-4.
6. BUNDENS DA, RECHTINE GR: Sarcoidosis of the spine. *Spine* 1986; 11: 209-12.
7. BLACKSIN MF, ACELLO AN, KOWALEE J, LYONS MM: Osseous sarcoidosis of the foot: Detection by MR imaging. *Am J Roentgenol* 1994; 163: 1444-5.
8. OVEN TJ, SONES M, MORRISEY WL: Lytic lesion of the sternum. Rare manifestation of sarcoidosis. *Am J Med* 1986; 80: 285-7.
9. ABDELWAHAB IF, NORMAN A: Osteosclerotic sarcoidosis. *Am J Roentgenol* 1980; 150: 161-2.
10. ROHATGI PK: Radioisotope scanning in osseous sarcoidosis. *Am J Roentgenol* 1980; 134: 189-91.
11. BLANK NM, STEININGER H, KALDEN JR, BURKHARDT H: Symptomatic bone lesion of the tibia head as onset of manifestation of sarcoidosis. *J Rheumatol* 1999; 26: 936-7.
12. KAYE O, PALAZZO E, GROSSIN M, BOURGEOIS P, KAHN MF, MALAISE MG: Low-dose methotrexate: An effective corticosteroid-sparing agent in the musculoskeletal manifestations of sarcoidosis. *Br J Rheumatol* 1995; 34: 642-4.