CASE REPORTS

The Value of the Bone Scan in Idiopathic Regional Migratory Osteoporosis

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Idiopathic regional migratory osteoporosis is a self-limited entity of unknown origin, which has received little attention in the nuclear medicine literature. It is characterized by severe joint pain, typically in a hip, with normal laboratory and normal early radiographic findings. Spontaneous recovery occurs, but recurrence is frequent in another joint, often the opposite hip. Recurrence in the same joint has never been reported. The bone scan is shown to be positive with first symptoms and is important in the diagnosis and management of this entity. Three cases are presented.

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Three cases of idiopathic regional migratory osteoporosis (IRMO) in middle-aged white males are presented. Two patients had involvement of first one hip and then the other. The third had one hip involved and was lost to follow-up.

This entity was first described by Curtiss and Kincaid in 1959 (1): Three patients presented with hip pain in the last trimester of pregnancy. Most cases reported since have been middle-aged males (2-8). Several authors have used different titles but we prefer the descriptive name idiopathic regional migratory osteoporosis.

The typical patient is a middle-aged male who presents with severe pain in a joint of the lower extremity, usually the hip. The onset is typically insidious, with no history of trauma; within days the pain reaches disabling proportions, but over a period of 2 to 6 mo there is spontaneous recovery, with no residual changes. The process may recur in another joint during the next 2 yr, often in the opposite hip. Recurrence in the same area has never been reported. Physical findings are minimal.

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Case 1. A 52-year-old white male was admitted in November 1976 with severe left buttock, left leg, and hip pain, exacerbated by motion and weight-bearing. He denied fever, chills, or weight loss. He walked with a marked antalgic gait, favoring the left leg. Moderate discomfort was noted on range of motion of the left hip. There was no fever, and laboratory studies were all normal. Radiographs showed osteoporosis of the left femoral head and neck (Fig. 1A). A Tc-99m pyrophosphate bone scan showed markedly

increased uptake in the left femoral head (Fig. 1B). Selective left external iliac arteriography showed slightly increased vascularity.

At 3 wk from onset of symptoms, biopsy of the synovium and left femoral head showed nonspecific chronic inflammation of the





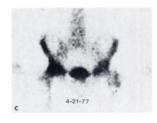




FIG. 1 Case 1. (A) Radiographs of pelvis in November 1976. Osteoporosis of left femoral head, neck, and trochanters is striking but ilium is also demineralized. (B) Anterior Tc-PPi bone scan on following day shows intense activity in left femoral head. Left pelvis and proximal femur are also abnormal. (C) Anterior bone scan with Tc-99m MDP in April 1977 shows return of left pelvis and hip uptake towards normal. Note persisting activity in trochanters, possibly due to hyperemia. Right hip and pelvis are now symptomatic and grossly abnormal. (D) Radiographs 6 wk later. Density of left femur is partially restored, and surgical defect in femoral neck and head persist. Right femoral head is demineralized.

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	TABLE 1.			
	Date	Clinical information	x-ray	Bone scan
	11/21/76	L. hip pain for several wk	Osteoporosis, left femoral head and neck	Markedly increased uptake left femoral head
	12/1/76	L. hip biopsy nonspecific		
Case 1	1/11/77	Better	Demineralized left femoral head	
	5/1/77	R. hip pain for 2–3 wk Symptom-free on left	None	4/21/77: increased activity right femoral head and neck; left returning to normal
	5/2/77	R. hip biopsy nonspecific		
	6/3/77	Continued R. hip pain	Demineralized right femoral head	
Case 2	Oct. 1976	Pain, severe, R. hip, biopsy R. hip nonspecific	Osteopenia with cortical thinning	Markedly increased activity righ hip, Marrow scan normal
	Jan. 1977	Improved	Improved	
	April 1975	L. Hip and thigh pain for several wk	Normal	No scan
Case 3	7/1/75	Continued L. hip pain	Demineralized left femoral head with poorly defined cortical margin	Markedly increased uptake in le femoral head
	10/9/75	Minimal discomfort L. hip	Normal	Normal
	Fall 1977	Similar symptoms R. hip	Demineralized right femoral neck; left normal	Increased uptake R. femoral head, neck, trochanters, acetabulum; left normal
	Spring 1978	Symptom-free		

synovial tissue. The femoral head showed "fat necrosis, scattered plasma-cell reaction, and endosteal new-bone formation. No changes of avascular necrosis were seen." Cultures were negative. Radiographs in January 1977 showed demineralization of the left femoral head. The patient became asymptomatic with conservative therapy.

On May 1, 1977, he was readmitted with a 2- to 3-wk history of pain in the opposite hip, described as a constant aching. A Tc PPi bone scan on April 21, 1977 (Fig. 1C) showed increased activity in the right femoral head and neck. The left side was returning toward normal and was asymptomatic. Radiographs on June 3, 1977 showed demineralization of the right femoral head (Fig. 1D). Physical examination showed a full range of motion of both hips but pain on the right side. Laboratory studies were normal. On May 2, 1977, biopsy of the right capsule showed fibrosis and mild lymphocytic infiltration. Biopsy of the bone showed fat necrosis and some new bone formation around the trabeculae. No changes of avascular necrosis were seen. Cultures were negative.

The patient recovered on conservative therapy and would not return for a follow-up bone scan. See Table 1 for chronologic summary.

Case 2. A 58-year-old white male complained of severe pain in the right hip, rapidly worsening. The pain was relieved by rest. There was no history of trauma, weight change, or underlying illness. He had slightly decreased range of motion in the right leg. All laboratory studies were normal.

Bone scan with 15 mCi of Tc-99m methylene diphosphonate

(Fig. 2A) showed marked increase in activity in the right hip region. Bone-marrow scan with 10 mCi of Tc-99m sulfur colloid showed normal activity (Fig. 2B). A concurrent radiograph (Fig. 2C) showed osteopenia with cortical thinning. Hip aspiration cultures were negative. Needle biopsy of the right femoral head showed hyaline cartilage and extensive new bone, with marrow fibrosis. He was discharged on pain medication, and improved. A radiograph 3 mo later (Fig. 2D) showed improvement, with no collapse or joint changes. He became asymptomatic and was lost to follow-up.

Case 3. A 50-year-old white male presented in April 1975 with increasing left hip and thigh pain of several weeks' duration. He had a noticeable limp favoring the left leg. There was no history of trauma. He had considerable discomfort with extremes of motion of the left hip. Laboratory studies and radiographs were normal. On July 1, 1975, radiographs showed demineralization of the left femoral head, with poor definition of the cortical margin. A Tc-99m polyphosphate bone scan showed markedly increased uptake in the left femoral head.

Symptoms resolved on conservative therapy, and on Oct. 9, 1975, a repeat scan was normal. Radiographs showed essentially normal mineralization.

In the fall of 1977 the patient developed similar symptoms in the opposite hip. Bone scan showed increased uptake in the right acetabulum, femoral head, femoral neck, and trochanters. A radiograph demonstrated demineralization of the right femoral head and neck, with a normal left hip. He has subsequently become symptom free with no specific therapy.

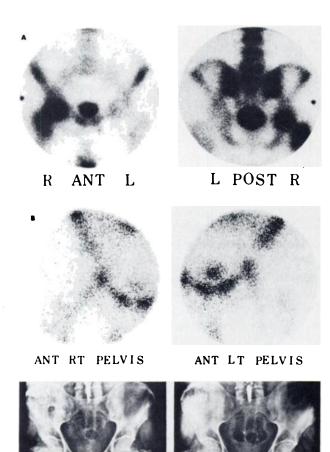


FIG. 2. Case 2. (A) Bone scan with Tc-99m MDP. Abnormal uptake in right femoral head, neck, and trochanters. Rest of the bone scan was normal. (B) Bone-marrow scan 5 days later with Tc-99m sulfur colloid. Marrow activity is normal in both proximal femurs. (Aseptic necrosis usually causes diminished or absent marrow uptake.) (C) Radiograph on day of bone scan shows osteopenia with cortical thinning in right proximal femur. (D) Radiograph 3 mo later shows improvement, with no collapse or joint changes.

DISCUSSION

Radiographs have been shown to be completely normal at the onset of IRMO (9), but serial radiographs have shown progressive, marked osteoporosis around the involved joints. Often the cortex in some areas is too thin to be identified. The earliest radiographic changes appear 3 to 6 wk after the first symptoms. Joints appear normal. The radiographs return to normal with no discernible residual changes, usually within 2 to 6 mo.

There is very little in the literature to suggest the importance of the bone scan in the early diagnosis and management of this entity. Several authors have briefly mentioned some increased activity at the involved sites. O'Mara and Pinals reported a case (9); they correlated the radiographic and bone-scan findings and demonstrated the superior sensitivity of the bone scan for early detection of IRMO.

Increased bone uptake generally indicates one or more of the following: increased osteoid formation, increased mineralization of the osteoid, or increased blood flow to the region. Because IRMO patients can show increased activity on the bone scan before any radiographic changes are discernible, it appears that there is

simultaneous excessive osteoclastic and osteoblastic activity. The radiographs show osteoporosis 3 to 6 wk after first symptoms; later, they show improvement, and the bone scan shows more intense activity. This suggests that the repair process is exceeding the destructive process. Eventually a normal balance is reached and both the radiograph and scan revert to normal.

All of the standard laboratory studies used to evaluate patients with joint pain are normal. Several biopsy reports in the literature agree with our two biopsied cases (three hips). Synovial membranes have been reported as normal or with minimal chronic nonspecific inflammation. Bone biopsy findings have been reported as consistent with osteoporosis.

The cause of IRMO is unknown. A sufficient number of cases of this symptom complex have been reported, with similar laboratory, radiographic, and bone-scan findings and with spontaneous recovery, to consider it an entity. Local hyperemia, either neurogenic or due to stasis, appears capable of producing a localized osteoporosis. Curtiss and Kincaid were unable to produce radiographic changes experimentally by compressing or severing obturator nerves in dogs (1). Lequesne (10) has suggested that IRMO is a nontraumatic variety of Sudeck's atrophy (reflex sympathetic dystrophy).

Several disease processes need to be considered in the differential diagnosis. A painful joint with localized demineralization suggests inflammatory or neoplastic disease as the prime consideration. IRMO patients show no periosteal involvement, no leukocytosis, no significant heat, and no ESR elevation. Tuberculosis of the hip can present with pain and demineralization, with increased uptake on the bone scan. Clinical presentation and joint involvement help to differentiate tuberculosis from IRMO. The age of the patient generally speaks against primary neoplasm.

The scan appearance, with the radiograph, help to rule out metastatic invasion. Disuse atrophy is slower in onset and the changes usually involve an entire extremity.

Synovial chondromatosis and villonodular synovitis may show little more than osteoporosis on the early radiographs. These disorders show no spontaneous regression. The patients are usually young, with severe stiffness and pain and typical radiographic findings later. Also the scan appearance is not one of generalized increased uptake throughout the femoral head and neck, as is typical of IRMO.

Both stress fracture and aseptic necrosis can imitate IRMO. The scan changes in stress fracture are typically localized to the cortex at the impending fracture site, rather than involving an area diffusely. Adults with aseptic necrosis often have a history such as sickle cell disease or steroid therapy. Also, in our experience, healing infarcts show more irregular uptake than does IRMO. A bone-marrow scan may help differentiate IRMO from marrow necrosis, as in Case 2. Radiographs are usually of help in the differential diagnosis of these conditions.

Since IRMO is a self-limited entity, there is no specific therapy. Immobilization is probably important in the early stages to prevent compression deformities, but the intense pain discourages movement. Steroids and antiinflammatory drugs seem to offer very little help, if any.

This report is prompted by the paucity of information on IRMO in the nuclear medical literature. The bone scan appears very useful in the diagnosis and management of this disorder. We believe that the diagnosis can be strongly suggested by correlation of history, laboratory findings, physical examination, radiographs, and bone-scan findings, and that many patients can be spared multiple invasive procedures such as biopsy and angiography if the diagnosis is considered.

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Diverse Bone Scan Abnormalities in "Shin Splints"

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Four young patients who presented with pain over the anterior compartment of the legs, gave a recent athletic history suggesting stress fractures. Although radiographs were initially normal in all four cases, the bone scintigrams were positive. The individual findings, however, were quite different. In one there was a single focal area of increased radioactivity in each of the tibias; the second patient had uneven uptake of radiotracer and several foci of accumulation in the fibulas; the third showed diffuse linear tibial uptake suggesting periosteal lesions; and the fourth case revealed uptake in the lateral malleolus and in bones of the foot.

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Although the term "shin splints" has been applied to stress fractures in horses (1), it has also become commonly used by the laymen to refer to anterior leg soreness occurring in athletes (2). We have performed bone scans on four young people with negative radiographs of the area, who turned out to have stress fractures of the leg.

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All four patients were seen because of pain over the anterior compartments of one or both legs. Each had a history of active participation in sports, and radiographs of the legs were negative. Only one of these people had any other problem (spondylolisthesis of L-5 in Case 2). After the intravenous administration of Te-99m methylene diphosphonate, images were obtained with a gamma camera. The pertinent data are summarized in Table 1, and scintigrams from each patient are shown in Figs. 1-4.

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RESULTS AND DISCUSSION

Stress fractures have been demonstrated early in their course by radionuclide imaging, before demonstrable radiographic changes (3.4). In each of the four cases presented here the initial



FIG. 1. Anterior scintiphotos in Case 1, demonstrating intense focal tibial uptake bilaterally.