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CASE REPORT

Can ischemic hip disease cause rapidly destructive hip osteoarthritis? A case report

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Summary – Avascular osteonecrosis of the femoral head (AONFH) usually goes through the four stages described by Arlet and Ficat: normal radiographs, heterogeneity and sclerosis of the femoral head, subchondral fracture with an individualized sequestrum, and secondary osteoarthritis. Arlet and Ficat individualized a specific pattern of AONFH which they called ischemic hip disease, in which cartilage damage seen as concentric joint space loss precedes the bony alterations. Although radiological and pathological studies of ischemic hip disease have been published, no clinical data are available. We report the case of a 65-year-old man admitted for a 1-month history of severe hip symptoms with concentric joint space loss but no osteophytes. Laboratory tests and examination of fluid aspirated from the hip ruled out septic arthritis and inflammatory hip disease. Two magnetic resonance imaging (MRI) studies done 1 month apart showed diffuse edema involving not only the femoral head but also the neck and trochanter, as well as major synovial hypertrophy. This atypical MRI appearance prompted synovial membrane and pertrochanteric core biopsies, which showed reactive synovitis and stage IV osteonecrosis, respectively. The pain, disability, and joint space loss worsened. Total hip arthroplasty was performed 1 month after the biopsy. Histological examination of the femoral head showed diffuse necrosis; no evidence of another condition was found on histological sections of the entire synovial membrane. This case corroborates the hypotheses put forward by Lequesne that some cases of rapidly destructive hip osteoarthritis may be ascribable to ischemia. Joint Bone Spine 2002 ; 69 : 76-80. © 2002 Éditions scientifiques et médicales Elsevier SAS

hip / hip osteoarthritis / ischemia / osteoarthritis / rapidly destructive hip osteoarthritis

In avascular osteonecrosis of the femoral head (AONFH), cartilage damage classically occurs late, over the undamaged part of the head, in response to mechanical strain caused by fracture of the sequestrum, which is the hallmark of stage IV disease in the Arlet and Ficat classification [1, 2]. In the 1970s, Arlet and Ficat [3] coined the term 'ischemic hip disease' to designate concentric joint space loss secondary to diffuse damage to the femoral head cartilage occurring before the bony radiological abnormalities characteristic of AONFH. They observed this pattern in a few patients, who had histological findings consistent with diffuse subchon

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dral bone edema as the cause of the cartilage damage. Catto [4] reported similar cases, and Lequesne [5] described rapidly destructive hip osteoarthritis (OA) ascribable to AONFH. We report a case in which two magnetic resonance imaging (MRI) scans done 1 month apart provided some insight into the pathophysiology of ischemic hip disease with no well-individualized sequestrum.

CASE REPORT

A 67-year-old patient with coronary heart disease, non-insulin-dependent diabetes mellitus, Leriche stage I occlusive arterial disease of the lower limbs, and a history of smoking was admitted for a 1-month history of hip pain causing insomnia. The pain had started abruptly. Radiographs had shown moderate hip dysplasia and concentric joint space narrowing; there were no osteophytes, changes in femoral head geometry, or alterations in bone structure *(figure 1)*. At admission, range of motion of the hip was restricted and painful in all planes. Other physical findings were normal. There was no personal or family history of rheumatic disease, glucocorticoid therapy, or dyslipidemia. Daily alcohol consumption was about 20 g. Body temperature was normal. Laboratory tests showed moderate inflammation (erythrocyte sedimentation rate, 25 mm/h; C-reactive protein, 10 mg/L). Tests were negative for HLA B27, antinuclear antibody, and rheumatoid factor. Aspiration of the hip recovered a few drops of clear yellow fluid with a mechanical cell pattern (820 leukocytes/mm3 with 40% of neutrophils and 60% of lymphocytes), no crystals, and negative studies for bacteria including the tubercle bacillus. MRI showed a diffuse low signal from the head, neck, and trochanters on T1 *(figure 2)* and T2 *(figure 2)* images, clearly visible on the STIR sequence (which cancels out fat signals) *(figure 4)*; there was fluid in the joint, and the synovial membrane was abnormally thick. *(figure 3)*.

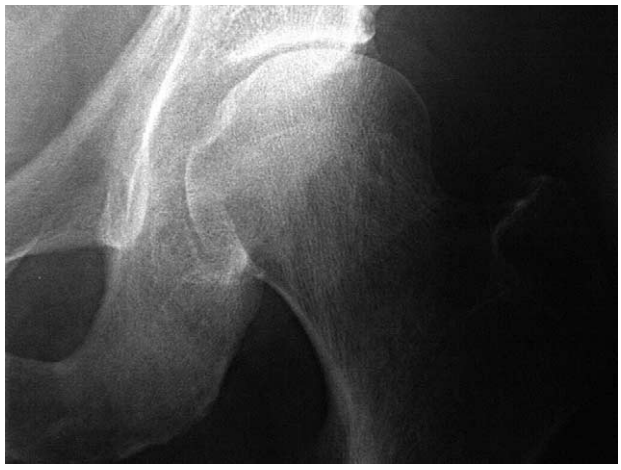

Figure 1. Anteroposterior radiograph of the hip (3 September 1999): concentric joint space loss without osteophytes or bone structure abnormalities.

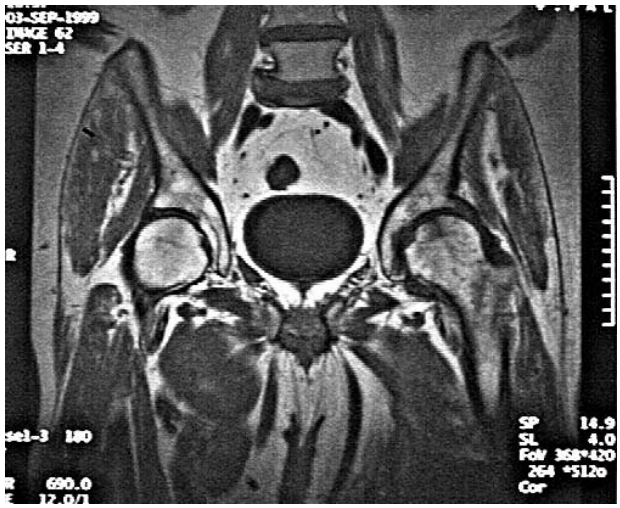

Figure 2. Magnetic resonance imaging (3 September 1999), T1-weighted sequences: diffuse heterogeneous low signal.

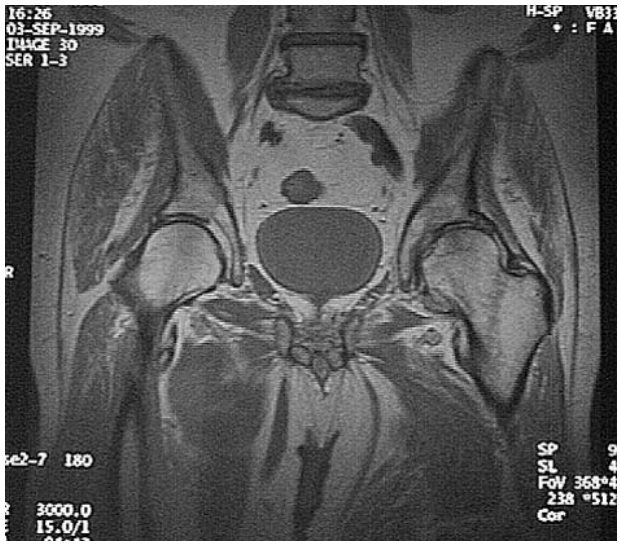

Figure 3. Magnetic resonance imaging (3 September 1999), T2-weighted sequences: low signal, hypertrophy of the synovial membrane.

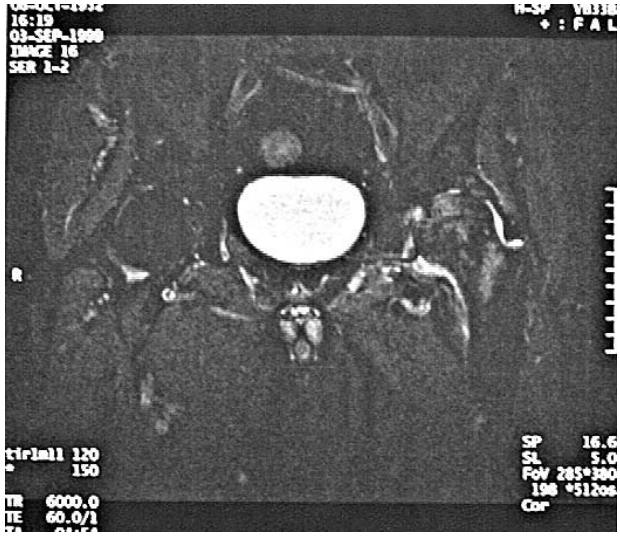

Figure 4. Magnetic resonance imaging (3 September 1999), STIR sequences: diffuse edema of the femoral head and neck, joint effusion.

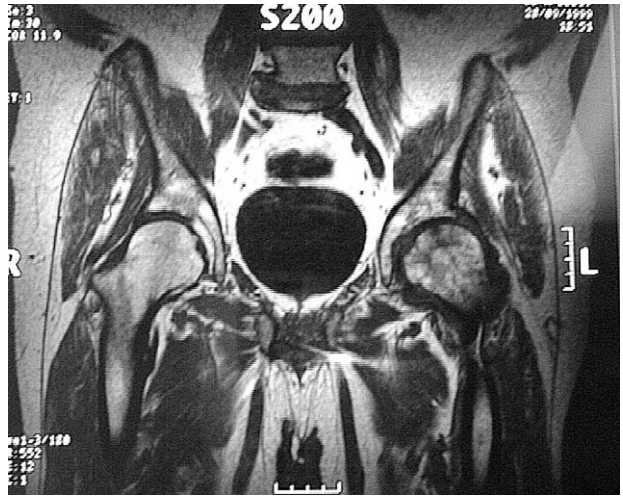

Figure 6. Magnetic resonance imaging (28 September 1999), T1-weighted sequence: moderate exacerbation of the signal abnormalities, persistent synovial thickening.

One month later, the pain had not abated, persisting at rest despite nonsteroidal anti-inflammatory therapy and a combination of acetaminophen and codeine. The physical findings were unchanged. A repeat MRI study showed a very small effusion, synovial membrane hypertrophy, and moderate exacerbation of the bone signal abnormalities, with no sequestrum *(figure 5)*. Further joint space loss was noted on the radiographs *(figure 6)*. Despite the negative findings from microbiological studies of the joint fluid aspirated 1 month earlier, a core biopsy and a biopsy of the synovial membrane were performed (Dr. Durroux) to rule out an infection before considering hip replacement surgery. The synovial membrane showed sclerosis and hyalinosis, with no inflammatory cells. Bone marrow changes consisted of fatty infiltration and edema of the intercellular spaces, the trabeculae contained no osteocytes, and there was no evidence of bone remodeling.

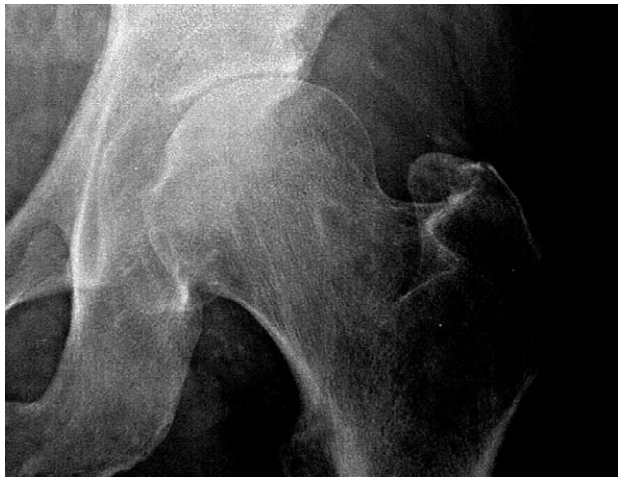

Figure 5. Anteroposterior radiograph of the hip (28 September 1999): further joint space loss.

One month later, the pain was unchanged and the Lequesne score was 15. Total hip arthroplasty was performed. The synovial membrane contained an abundance of macrophages phagocytizing cartilage debris. Other findings were congestion of the lamina propria and hyperplasia of the synoviocytes. The lymphoid cell population was too small to be taken as evidence of meaningful inflammation. The femoral head showed diffuse subchondral necrosis involving the marrow and trabeculae, meeting criteria for stage IV disease.

DISCUSSION

That the joint space long remains normal in patients with AONFH is often emphasized. Fracture of the sequestrum, which indicates passage to stage III in the Arlet and Ficat classification, widens the joint space, although this does not necessarily mean that the cartilage is intact. The classic tenet is that the damage to the femoral head cartilage is caused by mechanical strain related to alterations in femoral head geometry [2]. The cartilage damage, in turn, leads to osteoarthritis.

Arlet and Ficat used phlebography, intraosseous pressure measurements, and histological studies to show

that the circulatory disorders in AONFH extend beyond the sequestrum to the proximal femur [2]. Conceivably, diffuse ischemia of the subchondral bone may alter the vitality and structure of the femoral head cartilage, which derives its nutrients not only from the joint fluid but also from the subchondral blood vessels. Evidence that ischemia has deleterious effects on the cartilage has been obtained from autopsy studies in humans with peripheral occlusive arterial disease [6, 7]. Furthermore, in an experimental study in rabbits [8], occluding the blood vessels resulted in abnormal chondrocyte division with fragmentation of the cartilage.

Arlet and Ficat [3] conducted histological studies of core biopsies from patients with stage II or III AONFH, i.e., without joint space abnormalities. They found abnormalities in the joint cartilage: electron microscopy showed chondrocyte alterations including overfilled vacuoles, haphazard chromatin distribution and, in some cases, complete necrosis with karyolysis and rupture of the membrane. Weiss ([9] reported similar abnormalities.

In 1960, Serre and Simon [10] described three cases of hip disease with concentric joint space loss and diffuse osteonecrosis. Laboratory tests showed moderate inflammation, leading to the suggestion that osteonecrosis led to joint inflammation.

Arlet and Ficat [3] reported 40 cases of chronic, concentric, apparently primary hip disease with histologically documented osteonecrosis. Supero-medial or global superior joint space loss was present, simulating inflammatory hip disease in some patients. Bone structure was normal or showed sclerosis and geodes in most of the femoral head; histological studies indicated that these radiological abnormalities were produced by necrosis rather than by osteoarthritic geodes. The clinical and epidemiological characteristics in these patients were different from those in classic AONFH: the pain started abruptly, men and women were equally affected, mean age was 66 years as compared to 45 years in classic AONFH, and most patients had none of the classic risk factors for AONFH (alcohol abuse and glucocorticoid therapy). In another study [11], Arlet and Ficat noted that patients with peripheral occlusive arterial disease were at increased risk for this pattern of AONFH [11], which they called "ischemic hip disease."

Our case is similar to those reported by Serre in that the moderate increases in parameters for inflammation and the marked synovial membrane hyperplasia were reminiscent of inflammatory joint disease. In addition, similar to the patients studied by Arlet, our patient was

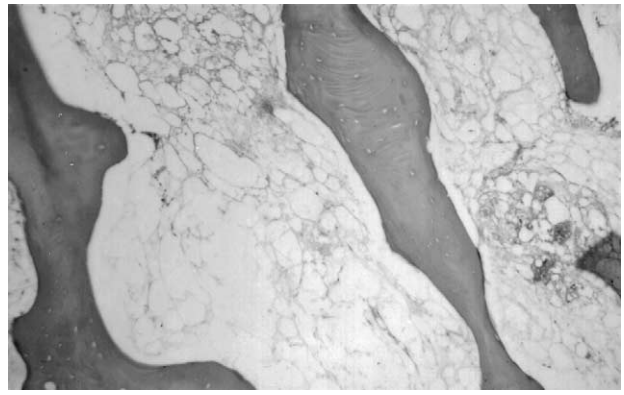

Figure 7. Core biopsy of the femoral head, hemalum-eosin stain, × 10: necrosis of the marrow and bone with empty osteocyte bays.

an elderly subject with hypertension and peripheral occlusive arterial disease but no history of alcohol abuse or glucocorticoid therapy. The abrupt onset, severe pain responsible for insomnia, and radiographic changes ruled out common osteoarthritis complicating hip dysplasia. MRI showed diffuse heterogeneous edema of the femoral head and neck, without the enhancement on T2 images seen in reflex sympathetic dystrophy syndrome; there was no individualized sequestrum, and the synovial membrane hypertrophy was more marked than is usually the case in AONFH. Histologic examination of the core biopsy and operative femoral head specimens showed advanced diffuse necrosis. This pattern is diagnostic for AONFH; although incipient necrosis confined to the marrow can be caused by delayed fixation of the specimen, extensive marrow necrosis with trabecular abnormalities *(figure 7)* occurs only in primary osteonecrosis, being readily distinguishable from the foci of marrow necrosis seen in patients with advanced osteoarthritis of the knee [12] or hip [13-15]. In our patient, histological studies of the synovial membrane ruled out an inflammatory or infectious disease. Neither a primary synovial disorder nor osteoarthritis complicating hip dysplasia could explain the MRI bone abnormalities or the histological changes found in our patient.

MRI evidence of edema can be found in osteoarthritic lesions, particularly at the knee. Zanetti [12] compared MRI and histologic abnormalities in 16 patients with advanced knee OA requiring replacement surgery. In most patients, areas with MRI evidence of edema were free from histological lesions, being composed either of fatty marrow (53%) or of normal trabe

culae (16%). Hofman [16] suggested that some patients with normal radiographs and MRI evidence of diffuse edema may have transient prenecrotic edema probably related to true reflex sympathetic dystrophy syndrome. Kim [17] made the same hypothesis but found no MRI signs of edema in 200 patients with AONFH. In our patient, examination of both histological specimens (core biopsy and operative specimen) showed that the MRI signs consistent with diffuse edema reflected extensive and advanced necrosis, rather than microfractures or reflex sympathetic dystrophy syndrome. This supports the suggestion by Arlet and Ficat that diffuse ischemia of the femoral head in a patient with peripheral arterial disease may be responsible for extensive and rapidly progressive damage to the femoral head cartilage with a secondary macrophagic synovial response. Thus the terms 'ischemic necrosis' or 'inflammatory hip disease complicating osteonecrosis' seem appropriate.

Rapidly progressive hip OA is characterized by severe joint destruction occurring within a few months [18]. The radiographic definition is a rate of joint space loss greater than 2 mm per year. The first radiographs obtained in our patient showed marked joint space loss. One month later, the increase in joint space loss exceeded 1 mm, suggesting rapidly destructive hip OA. Our case corroborates the suggestion by Lequesne that some cases of rapidly destructive hip OA may be ascribable to ischemia [19]. Core biopsy is probably not indicated in patients with rapidly destructive hip OA: given the diffuse cartilage damage, this procedure would probably fail to avoid progression of the disease to a stage requiring total hip arthroplasty.

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