Adult Idiopathic Chondrolysis of the Hip
— Report of Two Cases —

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Summary: Idiopathic chondrolysis of the hip is characterized by pain, limping and a progressive restriction in the range of motion of the hip. The paper describes the case of two female patients, age 28 and 37, who were referred to our hospital in October 1985 and May 1991 for pain and limitation of movement in the right hip. Neither patient had a previous history of systemic illness or trauma or medication such as steroids. On admission they underwent clinical and X-ray examinations. Both required an open biopsy to confirm the diagnosis. They have been followed for 4 years and 8 years, respectively.

Key words: idiopathic chondrolysis — young adult — osteoarthritis — open biopsy — progressive restriction of ROM

Introduction

Idiopathic chondrolysis of the hip is characterized by pain, limping and a progressive restriction in the range of motion (ROM) of the hip. Narrowing or the articular cartilage space is also usually noted. We report 2 rare adult cases of suspected idiopathic chondrolysis. There are very few reports of adult idiopathic chondrolysis, and open biopsy was needed to confirm diagnosis. No recognized treatment for idiopathic chondrolysis has yet been established.

Case 1

A 37-year-old female was referred to us in October 1985 with a 1-year history of right hip pain with restricted motion. She had previously been examined and medically treated in another hospital. After 1 month there, she was referred to our hospital for a detailed examination and further treatment. There was no history of fever, trauma, or other joint complaints, except for lumbar disc herniation in 1976.

Clinically, she had an antalgic gait, with slightly limited internal rotation to 10 degrees. The affected right leg was 1 cm shorter than the left. A laboratory exercise stress test showed results within normal limits. Radiographs revealed a narrowing or the joint space in the right hip and a normal shape of the femoral head (Fig. 1). Computed tomography revealed a cystic lesion in the right acetabulum with narrowing of the joint space in the right hip (Fig. 2a). This finding suggested primary osteoarthritis. A bone scintigram using 99mTc-MDP revealed an abnormal accumulation in
Fig. 1. Figures 1-4 show the findings in Case 1. Radiographs show a joint space narrowing in the right hip and a normal femoral head.

Fig. 2. a: Computed tomography shows a cystic lesion in the right acetabulum with joint space narrowing in the right hip. 
b: A bone scintigram using $^{99m}$Tc-MDP shows abnormal accumulation in the right femoral head and acetabulum.

She then underwent an open biopsy to confirm the diagnosis. Biopsy specimens from the capsule and the synovium grossly revealed the antero-lateral capsule was not strained and appeared to be normal. The synovial fluid was clear. Microscopically we found abnormal villous fibrillation and hyalinized connective and muscular tissue in the synovium, while the thickness of the synovium was normal (Fig. 3). These findings confirmed idiopathic chondrolysis as the diagnosis.

When she started to walk after 4 days, her hip pain had improved slightly and she could go up and down stairs. After 2 months of hospitalization, conservative therapy was continued on an outpatient basis. On follow-up examination at 4 years later, the ROM of the right hip was further decreased. Flexion was
decreased to 45 degrees, abduction to 5 degrees, internal rotation to 0 degrees and external rotation to 20 degrees. Her activities of daily living (ADL) were also affected. She could no longer kneel in the traditional Japanese style, nor could she reach her toes. Radiographs revealed a progressive advance in deformity of

Fig. 3. The figure shows the abnormal villous fibrillation and hyalinized connective and granulation tissue with fibrinoid change. There is no infiltration with inflammatory cells.

Fig. 4. Radiograph taken 4 years after that shown in Fig. 1. Progressive advance of osteolysis and increased joint space narrowing in the right hip.

Fig. 5. Figures 5-8 show the findings in Case 2. Radiographs of the right hip show a joint space narrowing, mild osteosclerosis in the acetabulum and ectopic calcification in the gluteal muscles.
Case 2

A 29-year-old female was referred to us in May 1991 with a 5 months’ history of progressive right hip pain and stiffness that had increased in severity in the prior 2 months. She had previously been examined and treated with conservative therapy for 3 months in another hospital. She was referred for detailed examination and treatment, and admitted as an in-patient after 2 months. There was no remarkable past history.

On physical examination she walked with a limp and the ROM of the right hip was restricted, with flexion limited to 110 degrees, external rotation to 20 degrees and internal rotation to 20 degrees. The left hip was normal. Laboratory data were normal. Radiographs of the right hip revealed a joint space narrowing, mild osteosclerosis in the acetabulum and ectopic calcification in the gluteal muscles (Fig. 5). Computed tomography revealed a cystic lesion in the right femoral head (Fig. 6a). A bone scintigram using 99mTc-MDP revealed an abnormal accumulation in the right femoral head and acetabulum (Fig. 6b).

Subsequently she underwent an open...
biopsy. Grossly we found normal articular cartilage, synovium, and synovial fluid. The biopsy specimen of the capsule and of the synovium microscopically revealed an abnormally thin layer of articular cartilage, in part of which we found invasion of fibrous connective tissue or loss of hyaline cartilage. The findings of the biopsy also included normal synovium and limited necrosis in the right femoral head (Fig. 7). From these findings, our diagnosis was idiopathic chondrolysis.

Twelve days after the open biopsy, she could walk with mild hip pain, and so could be followed on an out-patient basis. Follow-up evaluation for 3 years revealed lasting claudication and a progressive restriction in the ROM flexion to 70 degrees, abduction to 15 degrees and internal rotation to 0 degrees. Radiographically, the right hip joint space narrowing continued to progress (Fig. 8).

At present, she suffers a decreased capacity for ADL. For example she can no longer reach her toes and can no longer bow in a sitting style, so she now receives rehabilitation therapy.

Discussion

Chondrolysis of the hip is characterized by loss of a wide area of articular cartilage from the hip. Chondrolysis has been reported as a complication in slipped capital femoral epiphysis (SCFE). In 1971, Jones described cases of chondrolysis of the hip which were not associated with SCFE, immobilization, trauma, infection, or rheumatoid arthritis (Azuma et al. 1985; Ikai et al. 1988; Roy and Crawford, 1988; Daluga
and Millar, 1989), and he used the term "idiopathic chondrolysis of the hip" for the first time (Azuma et al. 1985; Daluga and Millar, 1989). In 1975, Duncan suggested the following diagnostic criteria (Azuma et al. 1985; Ikai et al. 1988) for idiopathic chondrolysis:

1) onset in adolescence
2) insidious onset of pain and limp
3) progressive restriction in ROM of the hip
4) radiographic joint space narrowing or a preserved contour of the femoral head
5) finding of non-specific synovitis (biopsy)
6) unknown origin

Since then, there have been case reports (Azuma et al. 1985; Roy and Crawford, 1988; Daluga and Millar, 1989) sporadically and most have involved children. In Japan, idiopathic chondrolysis has been considered a clinical entity after the suggestion of the diagnostic criteria (Azuma et al. 1985; Ikai et al. 1988) above, and has been differentially diagnosed from primary osteoarthritis. Though there have been several studies (Daluga and Millar, 1989) in regard to its pathogenesis, such as a theory of an immunopathological cause (Sivanantham and Kutty, 1977; Azuma et al. 1985) or of release of chondrolytic enzymes or of a metabolic disturbance in the chondrocytes, we still have not enough information to account for its etiology.

Our 2 cases had coxalgia, walking difficulties, and a slightly limited ROM. Their radiographs revealed joint space narrowing in the hip and a normal femoral head. From their first medical examination, clinical and laboratory findings, their disease was suspected to be primary osteoarthritis with young adult onset, pyogenic coxarthritis, and tuberculous coxarthritis. So, we performed an open biopsy for differential diagnosis.

Many investigators (Sivanantham and Kutty, 1977; Azuma et al. 1985; Ikai et al. 1988) have reported that the patho-histological findings of idiopathic chondrolysis of the hip were a thinning in the articular cartilage, fibrillation, and a swelling in the chondrocyte. They compared these findings with those of primary osteoarthritis, which are thinning or loss or fibrillation in the articular cartilage, and hypertrophic degeneration of the chondrocytes, thickening of the joint capsule with proliferation of synovium and villi, or bone necrosis in advanced cases.

The results of the biopsy in our 2 cases were an abnormal synovium with villous fibrillation or hyalinized connective and muscular tissue in Case 1, and an abnormal thin layer of articular cartilage, invasion of fibrous connective tissue, loss of hyaline cartilage, and bone necrosis in Case 2. There were no specific findings of synovitis. The hyalinized connective and muscular tissue and the invasion of fibrous connective tissue were not non-specific findings of inflammation. The histological findings in Case 1 resembled osteoarthritis. While in Case 2 the histological findings indicated both initial stage and terminal stage osteoarthritis at the same time. Although, the histological findings contradicted the findings of the medical examinations and the radiographs, we could not deny that they indicated primary osteoarthritis. Accordingly, although in these 2 cases primary osteoarthritis was a possibility,
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according to the criteria by Duncan (Azuma et al. 1985; Ikai et al. 1988), these 2 cases could not be diagnosed as idiopathic chondrolysis because of their late age of onset and because of the histopathological findings from the open biopsy.

Our 2 cases had coxalgia, slightly limited ROM in the onset, and had joint space narrowing in the hip, with a normal femoral head in the radiographs. And their symptoms were advanced.

Also we could not determine any cause of the disease, so overall these 2 cases resembled idiopathic chondrolysis of the hip. However discrimination from primary osteoarthritis and from rapidly destructive coxarthrosis was very difficult.

In 1977 Sivanantham and Kutty reported the case of a 20-year-old Amer-Indian man. He was not adolescent and had increasing hip pain and a progressively restricted ROM without any special history. Radiographical findings showed a uniform joint space narrowing with a normal femoral head. The destruction of the hip joint and limited ROM had advanced rapidly. Consequently, his hip joint had become ankylosed after 8 months. The result of a biopsy revealed avascular necrosis or medial hypertrophy in the arterioles of the capsule, and non-specific inflammation in the capsular and synovial tissue. From these findings the Amer-Indian man was diagnosed as having idiopathic chondrolysis although the onset age did not match the criteria by Duncan (Azuma et al. 1985; Ikai et al. 1988). They also performed an open biopsy and a histopathological examination. This case was informative for our two cases.

Bleck advocated (Azuma et al. 1985) that when the clinical and radiographical characteristics and the absence of signs of infection are indicative of idiopathic chondrolysis, a biopsy is not necessary. However, when an adult is suspected of having chondrolysis, an open biopsy is necessary for a differential diagnosis from primary osteoarthritis and from rapidly destructive coxarthrosis. According to the stage of the respective disease, a differential diagnosis is very difficult.

Our 2 cases had been treated with medication, and with rehabilitation including muscle strengthening exercises and ROM exercises. At the most recent follow-up, Case 1 displayed progressive restriction of ROM, but radiographically only slow progress in bone destruction. Her hip pain was improved. While Case 2 displayed a mild progression in the restriction of the ROM although a radiograph revealed no advance in the hip joint space narrowing. But in both these 2 cases the disturbance in ADL had become progressively worse.

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References


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