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CASE REPORT

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Open reduction of the dislocated hip in juvenile idiopathic arthritis: a case report

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Abstract An 8-year-old girl with systemic-onset juvenile idiopathic arthritis (JIA) required surgical reduction for a dislocated left hip joint following failure of skin traction for 1 week. Unaided walking was achieved by 3 months postoperatively. Incongruence and irregularity of the hip joint remained but may resolve with maturation. Joint laxity caused by synovitis, flexion/adduction contracture with pain, and acetabular dysplasia by growth disturbance apparently caused hip dislocation.

Key words Hip dislocation · Juvenile idiopathic arthritis (JIA) · Surgical reduction

Introduction

Systemic-onset juvenile idiopathic arthritis (JIA) is difficult to treat and can eventually become indistinguishable from polyarticular-onset JIA.[1] Systemic- or polyarticular-onset JIA results in severe destruction of polyarticulation, including hip joints. As hip joint involvement can cause severe residual disability, treatment of this joint is extremely important for preserving the quality of life in young patients. The present article describes a rare case of dislocated hip joint in a patient with JIA for which surgical reduction was performed.

Case reports

A 3-year-old girl experienced high fever, spiking at 37°–39°C. Ultrasonographic examination showed no hepatosplenomegaly. High fever recurred several times annually until the patient was 8 years old, and JIA was diagnosed. There had been no use of corticosteroids or disease-modifying antirheumatic drugs (DMARDs). At 8 years of age she developed polyarthralgia in both hip joints and visited a hospital. Nonsteroidal antiinflammatory drugs were prescribed and a rehabilitation program was initiated to restore range of motion (ROM) in several joints. Four months later, leg-length discrepancy became apparent, and posterior dislocation of the left hip was revealed on radiography. The patient was then referred to our hospital.

Physical examination

Her body weight was 16.1 kg (−2.3 SD), and her height was 120 cm (−1.6 SD). The patient displayed eight painful joints (both wrists, hips, and knee and ankle joints), but only the bilateral knee joints were swollen. Neither skin rash nor uveitis was detected on her first visit to our hospital. Her left hip joint was contracted in flexion, adduction, and internal rotation, with severe pain. A leg-length discrepancy of 8 cm was apparent, and a small spherical bony prominence was palpable on her left buttock. Both wrists and both ankle joints were also contracted.

Laboratory findings

Blood counts were normal, but severely high levels of C-reactive protein were identified (9.9 mg/dl). The erythrocyte sedimentation rate was 90 mm/h. The negative rheumatoid factor, negative antinuclear antibody, and HLA-DR4 indicated systemic-onset JIA.

Imaging findings

Radiography revealed posterior dislocation of the left hip joint with severe osteopenia (Fig. 1A). However, destruction of the joint surface was not evident, and no destructive changes were detected on radiographs of other joints. Bilateral acetabula were dysplastic, and growth plates were

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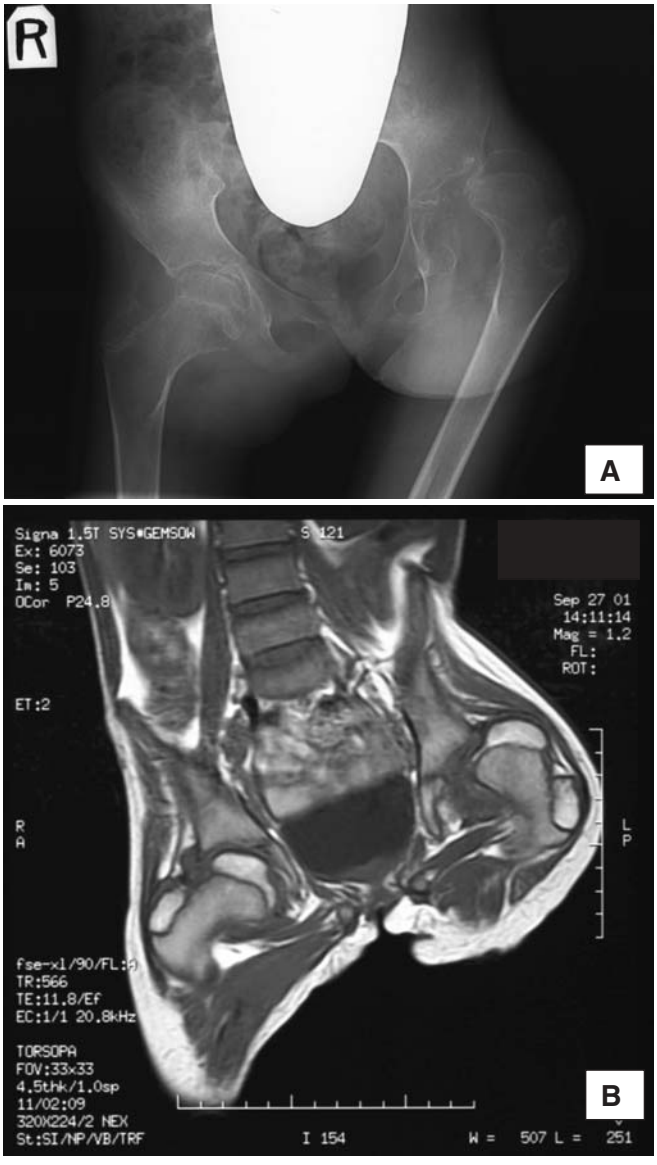

Fig. 1. Affected hip joint at first visit. **A** Radiography revealed severe osteoporosis with a dislocated left hip joint in addition to acetabular dysplasia. **B** Magnetic resonance imaging indicated normal intensity of the femoral head, suggesting the absence of idiopathic osteonecrosis

nearly closed. T2-weighted magnetic resonance imaging (MRI) indicated that avascular necrosis of the femoral head had not occurred (Fig. 1B).

Surgical treatment and postoperative course

Skin traction of the left limb for 1 week proved unsuccessful, so open reduction of the dislocated hip joint was performed. Using an anterolateral approach, and taking extreme care to avoid damaging the vascular structures to the femoral head, the affected hip joint filled with copious synovia was observed. An irregular articular surface of the acetabulum was seen, with numerous small fragments of cartilage in the joint cavity. Reduction of the femoral head was achieved after resecting the iliopsoas and adductor

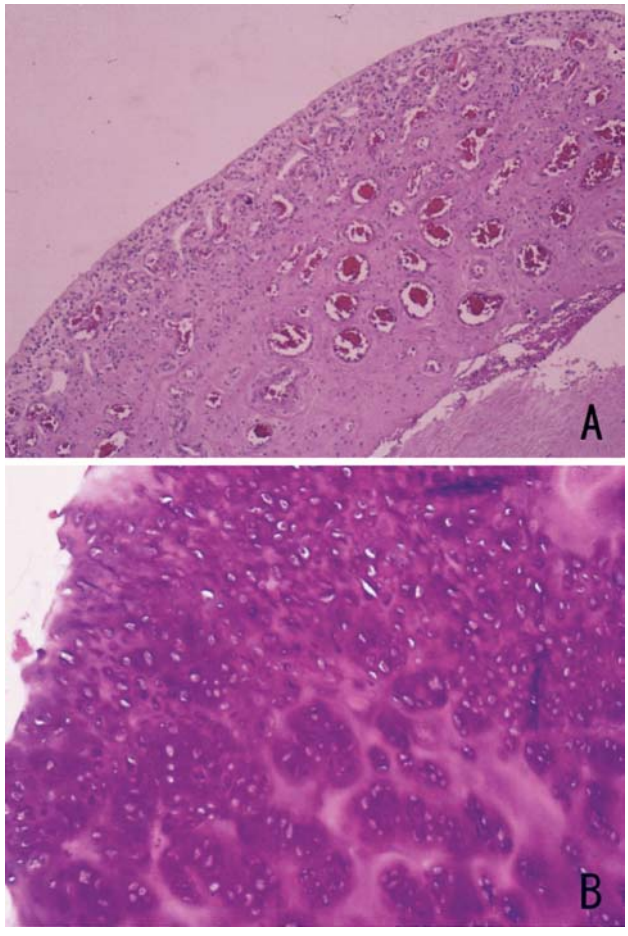

Fig. 2. Histological examination of tissue obtained from the affected hip joint at surgery. **A** Hypertrophic synovium was observed with proliferated and dilated vessels (×200). **B** Inside of joint displayed irregularity of the articular surfaces and numerous scattered cartilage fragments. Articular cartilage was severely degenerated (×400)

muscle tendons. Synovectomy of the affected joint was also performed. Histological examination revealed multiple cell layers with proliferated vessels in the synovium and severely degenerated articular cartilage (Fig. 2). A hip spica cast was applied for 3 weeks in the frog-leg position followed by 3 weeks in the functional position. Although the hip joint was considered at risk of spontaneous fusion, the reduced position was maintained with preservation of the ROM (Fig. 3). After removing the cast, standing and gait exercises were initiated using a long leg brace and crutches, as well as prescription of methotrexate, salazosulfapyridine, and glucocorticoid. The patient was able to walk unaided and independently by 3 months after the operation.

Discussion

In general, JIA leads to joint inflammation and growth disturbance. The former causes destruction of the articular cartilage and joint subluxation, and the latter causes loss

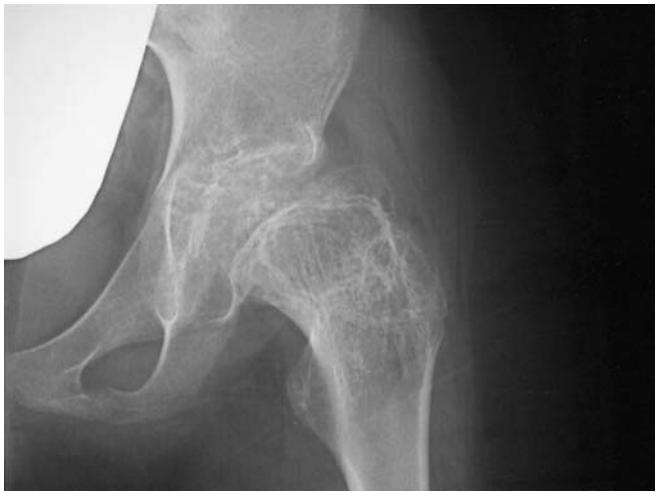

Fig. 3. Radiograph at the final follow-up. The reduced position was maintained, but joint congruence was not recovered

of congruence in the joint. Hip joints reportedly display several forms of destruction, including an Otto pelvis, subluxation, dysplastic hip, deformity of the femoral head or combinations of these problems. The ratio of hip involvement is reportedly 70% in JIA[2] and 50% in systemic JIA.[3] In most cases, hips display subluxation or central migration.[4]

Dislocation of the hip in JIA is rare, and to the best of our knowledge this is only the second report of such a case.[5] In this case, joint laxity caused by synovitis, flexion/adduction contracture with pain, and acetabular dysplasia due to growth disturbance contributed to dislocation of the hip joint.

The dislocated hip should be reduced by any means. In the first report in 1975,[5] the hip was successfully reduced using skin traction and subsequent cast immobilization. Once reduced, hip joint remodeling can be expected in the best-case scenario because of the young age of the patients, such as in the case reported in 2000.[6] If the best-case scenario does not occur, spontaneous ankylosing or severe destruction of the hip joint occurs, often leading to significant functional disability and subsequent joint replacement, even in young patients. At final follow-up, this girl was able to walk unaided, but radiography revealed inappropriate congruity with severe dysplasia of the acetabulum and deformation of the femoral head. Additional surgical treatment will probably be necessary in the future for her hip joint so she can have a normal social and school life, as do approximately 80% of children in Japan with systemic or polyarticular JIA.[7]

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