

Case Report

Idiopathic Chondrolysis of the Hip Treated by Immunosuppressive Therapy and Arthroscopic Intervention

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Idiopathic chondrolysis of the hip (ICH), a very rare disorder of unknown etiology, occurs mainly in female adolescents. Characterized by pain, limp, stiffness and radiological narrowing joint space from the rapid destruction of the articular cartilage, ICH sometimes results in ankyloses. We present the case of a 10-year-old girl diagnosed with ICH based on arthroscopic inspection and synovium biopsy. The femoral deformity appeared gradually, like a cam-type femoroacetabular impingement. She was treated with intensive rehabilitation and immunosuppressive drug. We later performed an arthroscopic bumpectomy for residual symptoms. She achieved a favorable outcome as a 15-year-old at the latest follow-up.

Key words: idiopathic chondrolysis, hip joint, medication, bump, arthroscopy

Chondrolysis of the hip was first described in 1930 as cartilage necrosis complicated with acute slipped capital femoral epiphysis (SCFE) [1]. In 1971, Jones reported 9 similar cases, 7 of which were of unknown cause [2]. Duncan *et al.* reported nine cases with the same phenomenon, and they called this disorder 'idiopathic chondrolysis of the hip (ICH)' [3]. Chondrolysis of the hip in adolescents occurs secondary to other disorders including trauma, SCFE, Perthes disease, infection, transient arthritis, immobilization, and juvenile rheumatoid arthritis. Thus, the diagnosis of ICH is not easy and is made in part by excluding other diseases with similar symptoms and based on the radiographic features.

Segaren *et al.* reviewed the literature and reported that new cases of ICH are rare because the worldwide

occurrence is decreasing [4]. To the best of our knowledge, ICH is very rare in Japan, according to research by the Japanese Paediatric Hip Research Society meeting in 2016. The mainstay of treatment for ICH in the acute stage is conservative treatment. However, patients for whom conservative treatment has failed would need surgical intervention, and the optimal surgical treatment for ICH is a matter of controversy.

Here we report a case of ICH in a 10-year-old girl who was treated successfully by intensive conservative treatment, drug therapy, and additional arthroscopic surgical intervention.

Case Report

A 10-year-old Japanese girl presented with right coxalgia and gait disturbance. She had grown normally

without mental retardation and had no history of developmental dysplasia of the hip (DDH), Perthes disease, *etc.* None of her family had immune disorders or bone disease. When the patient was 8 years old, she complained of right coxalgia and visited another orthopedic clinic, where she was diagnosed with transient synovitis of the hip based on the results of an X-ray examination (Fig. 1). Her symptoms then showed spontaneous remission, and she had no complaint for nearly 2 years after that. However, the hip pain suddenly returned after physical activity at school; she visited the previous clinic and was treated immediately with conservative treatment that continued for 2 months. However, the coxalgia and gait disturbance continued to worsen.

At the patient's presentation to our hospital, magnetic resonance imaging (MRI) showed remarkable joint effusion of the affected hip (Fig. 2). The radiograph (Fig. 3) and computed tomography (CT) (Fig. 4) showed abnormal bony changes of the femoral head. At her first visit to our hospital, the patient could not walk without the aid of 2 crutches, and the range of motion (ROM) of her right hip joint was severely limited by pain: (right/left) flexion 70°/120°, extension -10°/0°, abduction 15°/40°, adduction -10°/10°, internal rotation 0°/50°, external rotation 60°/70°. The results of all of the laboratory tests were normal, including the blood counts, ESR, CRP, and matrix metalloproteinase-3.

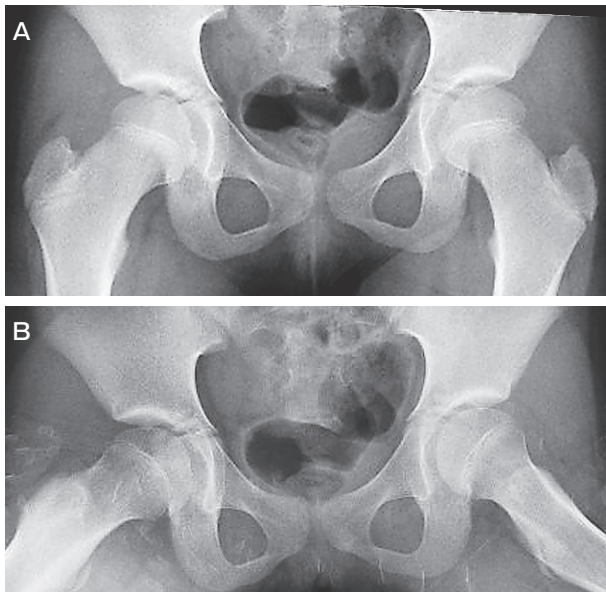


Fig. 1 A, B: The two-direction X-rays of both hips, taken when the patient was 8 years old.

The rheumatoid factor and antinuclear antibody results were also negative.

We recommended to the patient and her family that she should undergo hospitalization for treatment with continuous traction and physical therapy. After 2 months of hospitalization, the joint effusion was improved on MRI (Fig. 5). However, the patient's hip motor pain had only decreased a little and the range of motion was not improved.

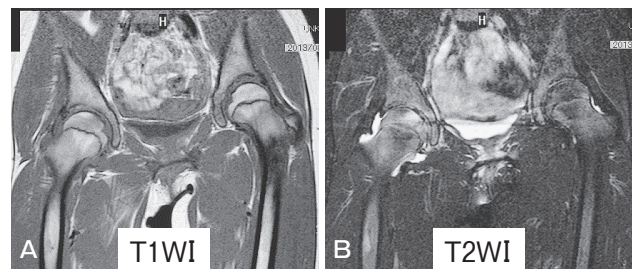


Fig. 2 MRI of the patient's hips, at the age of 10 years old. A, Coronal T1-weighted view; B, Coronal T2-weighted view of the right hip, showing remarkable joint effusion and bone marrow high intensity.

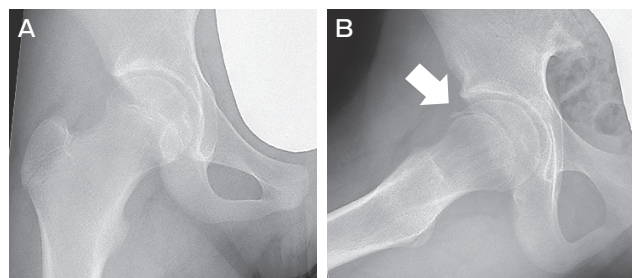


Fig. 3 A, B: The two-direction X-rays of the right hip, taken when the patient was 10 years old. Abnormal ossification can be seen in anterior-lateral side of the distal epiphysis of the femoral head on the right hip (*white arrow*).

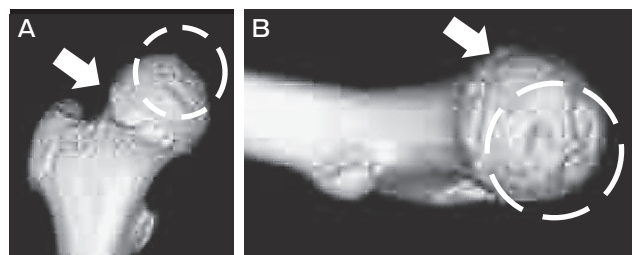


Fig. 4 A, B: CT of the right hip showing abnormal protrusion in the axial view (*white arrow*). Three-dimensional CT of the femoral head showing an abnormal irregular lesion at the medial side (*white pointed circle*).

The diagnostic hypotheses were as follows: chronic infection, juvenile rheumatoid arthritis, synovial chondromatosis, pigmented villi nodular synovitis, and idiopathic chondrolysis. At 4 months from the patient's first visit, we performed hip arthroscopy to carry out a biopsy of the synovium. However, the patient's hip joint space was not wide enough, even under traction with general anesthesia. We thus abandoned our attempt at accessing the central compartment.

We were able to perform an arthroscopic biopsy and a synovectomy of the proliferating synovium by gaining access to the peripheral compartment (Fig. 6). The joint fluid was clear and slightly red; the culture was negative, and the laboratory data were also within the normal ranges. The histopathological examination of the synovium showed non-specific inflammation. Tumor disease of the intra- and extra-hip joint was excluded. We therefore diagnosed ICH based on the total findings.

After the hip arthroscopy, we consulted a pediatrician (Dr. Yashiro); he administered oral medication with methotrexate (MTX) 12 mg/week to the patient,

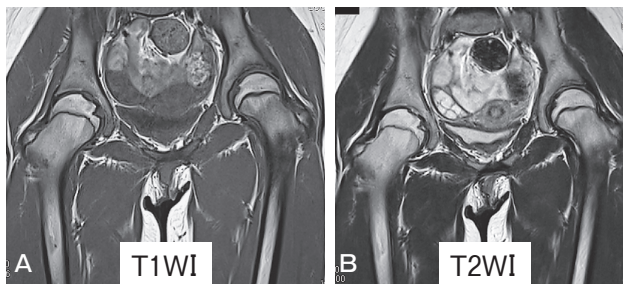


Fig. 5 Two months after hospitalization, MRI of the hips. **A**, Coronal T1-weighted view; **B**, Coronal T2-weighted view showing the improvement of the joint effusion and the bone density of the right hip.

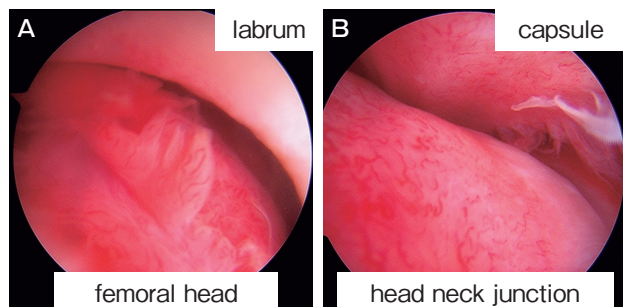


Fig. 6 **A**, **B**: Arthroscopic inspection. The abnormal synovium had proliferated at the surface of the femoral head and head neck junction.

and this continued until the latest follow-up. The non-weightbearing and motor physical therapies were continued in concert with the medication and hospitalization. At 1 year after the initial hip arthroscopy, the patient left the hospital because her clinical symptoms were improved, and we allowed her to go to primary school with 2 crutches.

However, the ROM limitation of the affected hip remained, especially in flexion (70°) and internal rotation (5°). Additionally, the anterior impingement sign was positive, and an abnormal bony bump was clearly visible on lateral X-ray (Fig. 7). The patient could not sit on the floor holding her knees. We thus performed an arthroscopic bumpectomy via the peripheral compartment (Fig. 8). After the second hip arthroscopy, the ROM was improved in flexion (100°) and internal rotation (30°), and the anterior impingement sign disappeared. At 3 months after this operation, we allowed the patient to go to school without any support. At the final follow-up, 5 years after her first visit, the ROM of the patient's right hip joint was still slightly limited: (right/left) flexion 110°/120°, extension 0°/0°, abduction 30°/40°, adduction 0°/10°, internal rotation

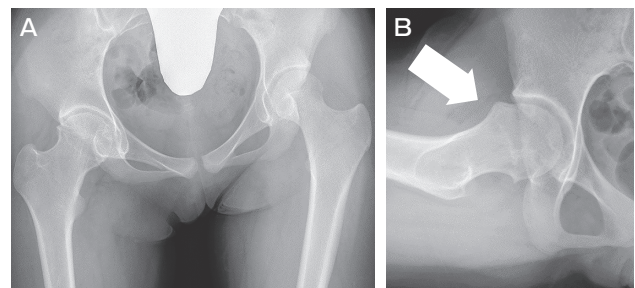


Fig. 7 **A**, **B**: The X-rays of the right hip at 18 months after the first hip arthroscopy; the joint space was maintained. The abnormal bony bump is observable in the lateral view (*white arrow*).

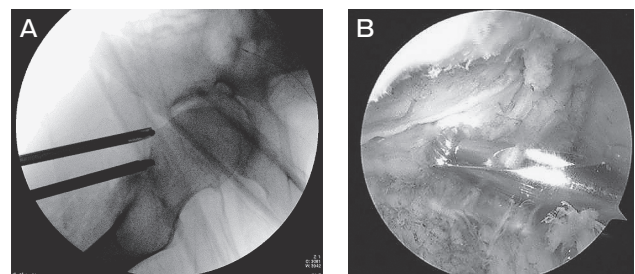


Fig. 8 **A**, The intraoperative view of the image intensifier at the arthroscopic bumpectomy; **B**, The arthroscopic view when the bump was resected with the abradar.

30°/50°, external rotation 60°/70°.

At that point, the patient could sit on the floor holding her knees. In the final radiographs, the osteopenia had disappeared and no remarkable bump was observed (Fig. 9). Although we still recommend that the patient avoids strenuous exercise, she reported that she does not experience any limitations now in her daily life.

Discussion

ICH was first described by Jones in 1971 [2]; nine cases that had no clear causation. Hoven *et al.* reported three cases of patients who had deposits of immuno-complexes in the synovial membrane [5]. It has been speculated that abnormalities of the immune system and cartilage metabolism, induced by incidents such as trauma or infection, may lead to the occurrence of ICH. However, the cause of ICH is still unknown, and the diagnosis remains difficult. There are few reports of ICH cases in Asia [6, 7].

The radiographic and clinical diagnostic criteria of ICH were first described by Duncan in 1975. In 1989, Daluga reported the radiographic features of ICH from early to late stage [8]. Regarding the specific features of ICH that are visible on MRI, Johnson suspected regional muscle atrophy, bone remodeling, cartilage loss, and joint effusion [9]. Laor *et al.* also reported bone edema in the central part of the epiphysis of the femoral head on early T2-weighted MR images in ICH [10]. In our patient's case, we diagnosed ICH based on the radiographic joint space narrowing, the chondral defect and bone edema of the epiphysis of the femoral head in the early MRI, the blood data, and the results of the histopathological examination by arthroscopic

synovectomy.

In their review, Segaren *et al.* described the natural history of ICH and divided it into 3 groups [4]. The extreme cases achieve a complete recovery; the second group develop a stiff hip that has limited function but no pain, and in the third group the disorder progresses to a painful, malpositioned ankylosed hip. The prognosis is unpredictable, and the long-term result in the chronic phase is quite different in each case.

The treatment for ICH in the acute phase is basically conservative management, *i.e.*, active rehabilitation to maintain the ROM, rest with traction for offloading, and medication with one or more nonsteroidal anti-inflammatory drugs for pain relief. Khoshhal *et al.* reported 2 cases of ICH treated with an injection of botulinum neurotoxin-A for spastic muscles, combined with intensive rehabilitation [11]. Ruiz Picazo *et al.* reported an unusual case of ICH in which common bacteria of the mouth were revealed by a joint biopsy [12]; they treated the patient with antibacterial drugs and MTX, and the result was satisfactory. Appleyard *et al.* reported a case of ICH that was successfully treated with the biopharmaceutical etanercept [13]. Thus, medication with methotrexate would also be effective for the immunologic abnormality in our patient's case.

Surgical treatment is indicated for ICH patients who have undergone failed conservative treatment and still have pain and limited hip motion. Treatments for ICH involving tendon release and capsulotomy [14] have been reported. Garcia DC *et al.* reported that the long-term results were poor despite surgical intervention [15]. Less-invasive interventions for each of the various conditions in ICH should therefore be considered.

We were unable to find any reports of arthroscopic treatment for ICH. In our patient's case, we performed the first arthroscopic intervention to achieve a definitive diagnosis and synovectomy 3 months after the patient's hospitalization for intensive traction and rehabilitation. After the first surgical intervention, treatment with MTX was started by a pediatrician. Both the radiographic joint space and the ROM were improved by prolonged rehabilitation for 1 year with hospitalization. However, the abnormal bony bump on the femoral head was clearly enlarged. The bump had formed gradually, in a process similar to callus formation.

The relationship between bump formation and the synovitis caused by ICH was unclear. Subsequently, the patient complained of pain and limitation in maximum

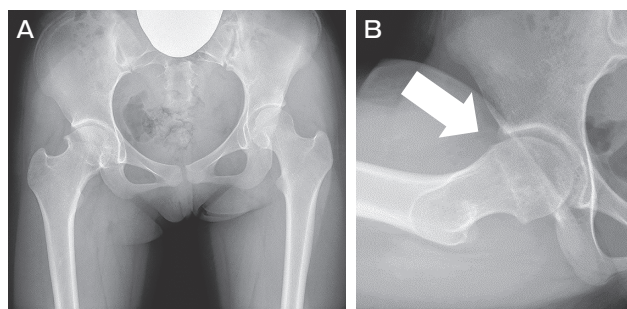


Fig. 9 A, B: The X-rays at 5 years after the second hip arthroscopy. The joint space was maintained and the pelvic obliquity was also improved. The abnormal bony bump is not present in the lateral view (white arrow).

flexion and internal rotation. We then performed a second surgical intervention, *i.e.*, arthroscopic bümpectomy. The patient was satisfied with the arthroscopic treatment. Further long-term follow-up will be necessary to watch for the potential development of osteoarthritis.

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