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Polyarticular juvenile idiopathic arthritis following anterior cruciate ligament reconstruction with successful return to sport: a case report

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Case Report

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Abstract

Background

Juvenile idiopathic arthritis (JIA) is the most common chronic rheumatic disease in children, whereas new-onset JIA following orthopedic surgery is extremely rare. We report a case of polyarticular JIA that developed after anterior cruciate ligament (ACL) reconstruction, with successful return to sport.

Case presentation:

A 15-year-old female basketball player sustained a left ACL injury and underwent double-bundle ACL reconstruction using a semitendinosus tendon. Three weeks postoperatively, she developed polyarthralgia and joint swelling involving the fingers, wrist, and elbow. Ultrasonography and contrast-enhanced magnetic resonance imaging (MRI) demonstrated synovitis, and laboratory findings revealed elevated inflammatory markers, rheumatoid factor, and matrix metalloproteinase-3. JIA was suspected, and methotrexate therapy was initiated. Owing to persistent systemic inflammation, tocilizumab was added 2 months postoperatively, resulting in rapid improvement. The patient temporarily developed knee hemarthrosis 7 months postoperatively, which was managed conservatively. Serial MRI demonstrated gradual improvement in graft signal intensity. One year postoperatively, she was cleared to return to competitive sports, and at the final follow-up (15 months), she had returned to her pre-injury level without residual symptoms.

Conclusions

This case suggests that autoimmune diseases such as JIA should be considered when atypical joint symptoms occur after ACL reconstruction. Early diagnosis and appropriate treatment may enable favorable outcomes, including return to sport.

Background

Juvenile idiopathic arthritis (JIA), the most common chronic rheumatic disease in children, is defined as arthritis of unknown etiology that begins before the age of 16 years and persists for at least 6 weeks (1). JIA comprises a heterogeneous group of subtypes with different clinical presentations and prognoses. Polyarticular JIA, characterized by the involvement of multiple joints, is associated with a higher risk of persistent disease and joint damage (2).

Although the pathogenesis of JIA remains unclear, environmental factors such as infection, stress, and trauma, in addition to genetic susceptibility, may activate immune responses and contribute to disease onset (3). Participation in sports during childhood plays an important role in physical and psychosocial

development. Even in patients with JIA, appropriate disease control allows participation in physical activity and sports; however, their activity levels tend to be lower than those of healthy peers (4).

Anterior cruciate ligament (ACL) injury is a common injury among athletes, particularly in females (5, 6). Because ACL injury results in knee instability and functional impairment, ACL reconstruction is widely performed (7). Postoperative complications include graft failure, joint stiffness, infection, and deep vein thrombosis (8–10). However, no reports have described the onset of JIA following orthopedic surgery.

We report a case of polyarticular JIA after ACL reconstruction, presenting with polyarthralgia and joint swelling.

Case presentation

A 15-year-old female basketball player with no significant past medical or family history presented with knee instability and pain. The patient sustained a left knee injury during a game after contact with an opponent while dribbling. Magnetic resonance imaging (MRI) was performed (Fig. 1a, b), confirming an ACL injury. Preoperative evaluation revealed no abnormalities, and double-bundle ACL reconstruction using a semitendinosus tendon was performed 1 month after the injury (Fig. 2a–c).

The postoperative course was initially uneventful. The patient remained non-weight-bearing for 1 week, followed by gradual partial weight-bearing. Three weeks postoperatively, the range of motion of the knee improved to −10°/120°, and full weight-bearing was achieved.

At the same time, she developed swelling of the finger joints, left wrist, and elbow, and was referred to a pediatrician with suspected rheumatoid arthritis. At the pediatric evaluation, arthritis was observed in the bilateral metacarpophalangeal joints, wrists, and elbows. Ultrasonography demonstrated synovial hypertrophy with increased vascularity. Contrast-enhanced MRI of the elbow demonstrated synovitis and bone marrow edema in the olecranon (Fig. 3a–d). Laboratory findings revealed elevated C-reactive protein (1.66 mg/dL), high rheumatoid factor (RF) (324 IU/mL), elevated matrix metalloproteinase-3 (203.1 U/mL), and positive antinuclear antibodies (1:40). Based on these findings, JIA was suspected, and methotrexate treatment was initiated.

Two months postoperatively, tocilizumab therapy was started because of fever and elevated inflammatory markers. Inflammatory markers rapidly improved, and C-reactive protein levels normalized (0.04 mg/dL). Laboratory tests confirmed persistent RF positivity, leading to a diagnosis of polyarticular JIA.

Six months postoperatively, MRI demonstrated good continuity of the reconstructed graft, with a trend toward improved high signal intensity (SI) (Fig. 4a, b). On physical examination, the Lachman and pivot-shift test were negative, and no patellar ballottement was observed. The patient subsequently initiated advanced rehabilitation, including sprinting and ladder training.

Seven months postoperatively, she developed left knee swelling after training. Joint aspiration yielded approximately 27 mL of bloody fluid. MRI demonstrated preserved graft continuity but increased SI within the graft (Fig. 5a). Conservative management with rest was implemented. Follow-up MRI 8 (Fig. 5b) and 9 (Fig. 5c) months postoperatively showed a gradual decrease in graft SI. Rehabilitation was progressively advanced to include cycling and running. One year postoperatively, MRI demonstrated further improvement in graft SI (Fig. 6a, b), and the patient was cleared to return to competitive sports. Fifteen months postoperatively, the patient had returned to her pre-injury level of sports activity, and all knee-related symptoms had resolved. On physical examination, range of motion was 0°/140°. The Lachman and pivot-shift tests were negative. Instrumented laxity assessment using a KT-1000 arthrometer demonstrated a side-to-side difference of 1 mm.

Discussion and Conclusions

Case reports of autoimmune disease onset following ACL reconstruction surgery are extremely rare. Kembhavi et al. (11) reported a case of a 30-year-old male who developed rheumatoid arthritis (RA) following ACL injury. This patient had no history of rheumatoid arthritis before the injury and was diagnosed based on positive RF and anti-citrullinated protein antibodies 6 months after the initial sprain. The authors emphasized the necessity of thorough investigation when abnormal synovitis and cartilage damage are observed during arthroscopy. Kääb et al. reported chronic recurrent joint effusion in a patient diagnosed with Behçet's disease 7 years after ACL reconstruction, demonstrating the need to include chronic rheumatic diseases in the differential diagnosis of persistent joint dysfunction following ACL surgery (12). In the present case, joint swelling was observed postoperatively, and prompt referral to the pediatric department enabled early treatment, ultimately allowing successful recovery. To our knowledge, there are no previous reports of patients with JIA who achieved return to their original sport level.

The pathogenesis of JIA involves complex interactions among genetic, environmental, and immunological factors. Among genetic factors, associations with HLA alleles are most clearly established (13). Environmental factors include bacterial and viral infections, and it has been hypothesized that physical trauma and psychological stress may also affect disease onset in genetically predisposed individuals (14)

Multiple mechanisms have been proposed regarding the potential role of trauma and surgical stress as triggers for autoimmune arthritis. Karateev and Polishchuk (15) reported that physical injury can activate the immune system and manifest as autoimmune diseases such as psoriatic arthritis, rheumatoid arthritis, and axial spondyloarthritis in joints and tendon insertions through a “deep Koebner phenomenon.” From the perspective of post-traumatic osteoarthritis, Sunbol, et al. (16) reported that, following joint trauma, chondrocytes, synoviocytes, and immune cells release interleukin (IL)-1β, tumor necrosis factor-α, and IL-6, leading to cartilage matrix degradation and persistent inflammation induced by innate immune activation via toll-like receptors triggered by damage-associated molecular patterns.

This inflammatory cytokine cascade may induce autoimmune responses in genetically predisposed individuals.

When persistent abnormal joint inflammation or synovitis is observed following ACL reconstruction surgery, conducting thorough investigations with consideration of autoimmune joint diseases including JIA, rather than dismissing symptoms as mere postoperative complications, infectious arthritis, or reactive arthritis, is important (11,12).

A limitation of the present case is the difficulty in establishing a causal relationship between ACL reconstruction surgery and JIA onset, as the temporal association between these events may be coincidental. Future accumulation of similar cases and further basic research on immunological changes following ACL reconstruction surgery are needed.

In conclusion, polyarticular JIA developed after ACL reconstruction in a 15-year-old female patient, who was successfully treated with biologic therapy and returned to sport. Although whether ACL reconstruction contributed to the onset of JIA remains unclear, to our knowledge, such cases have not been previously reported. Clinicians should consider the possibility of autoimmune disease when patients present with atypical joint pain or swelling after surgery, as early evaluation and appropriate referral are important for timely diagnosis and management.

Abbreviations

ACL, anterior cruciate ligament; JIA, juvenile idiopathic arthritis; MRI, magnetic resonance imaging; RF, rheumatoid factor; SI, signal intensity

Declarations

Ethics approval and consent to participate

Ethics approval was not required for this case report in accordance with institutional guidelines. Written informed consent was obtained from the patient.

Consent for publication

Written informed consent was obtained from the patient and her legal guardian for publication of this case report and any accompanying images.

Availability of data and materials

Not applicable.

Competing interests

The authors declare that they have no competing interests.

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Authors' contributions

HKI drafted the manuscript and examined the patient. HO supervised the manuscript preparation and performed the clinical examination and surgery. ST managed the treatment from a pediatric perspective and provided clinical advice. ES collected the clinical data. HK assisted in manuscript preparation. YI provided overall supervision and critical advice. All authors read and approved the final manuscript.

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Figures

Fig. 1



Figure 1

Knee MRI at the time of injury

a: Coronal view demonstrating swelling of the ACL.

b: Sagittal view demonstrating ACL swelling and high signal intensity, suggesting femoral-sided rupture.

ACL, anterior cruciate ligament; MRI, magnetic resonance imaging.

Fig. 2

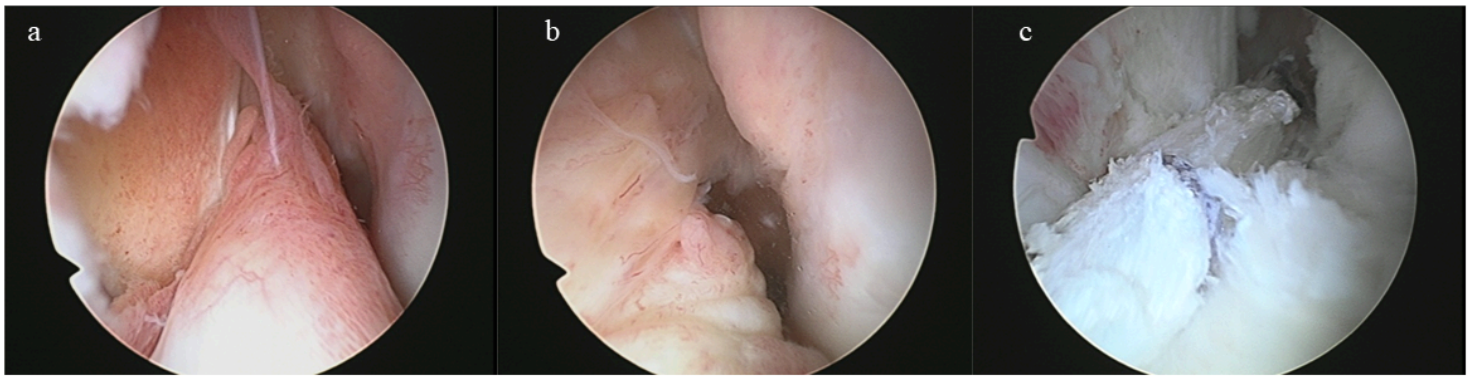


Figure 2

Arthroscopic findings at the time of ACL reconstruction

a, b: ACL remnant was preserved, and rupture was observed at the femoral side.

c: Double-bundle ACL reconstruction was performed using a semitendinosus tendon.

ACL, anterior cruciate ligament.

Fig. 3



Figure 3

Sagittal MRI of the elbow

a: T1-weighted image, b: T2-weighted image, c: Short tau inversion recovery (STIR), d: Contrast-enhanced T1-weighted fat-suppressed image.

Synovial proliferation was observed within the elbow joint. Bone marrow edema was also noted in the olecranon.

MRI, magnetic resonance imaging.

Fig. 4

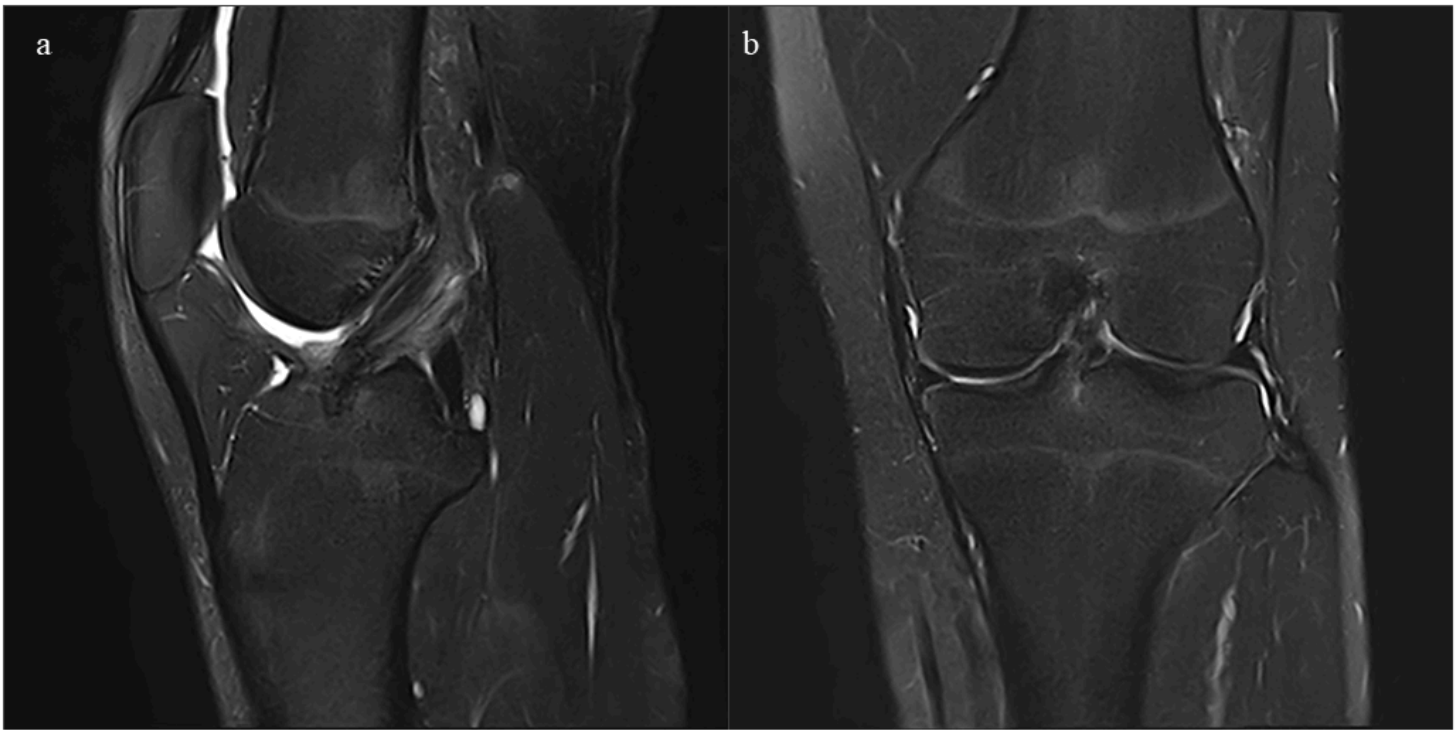


Figure 4

Knee MRI 6 months after ACL reconstruction

a: Sagittal view, b: Coronal view. High signal intensity persisted, and the continuity of the reconstructed ACL was preserved.

ACL, anterior cruciate ligament; MRI, magnetic resonance imaging.

Fig. 5

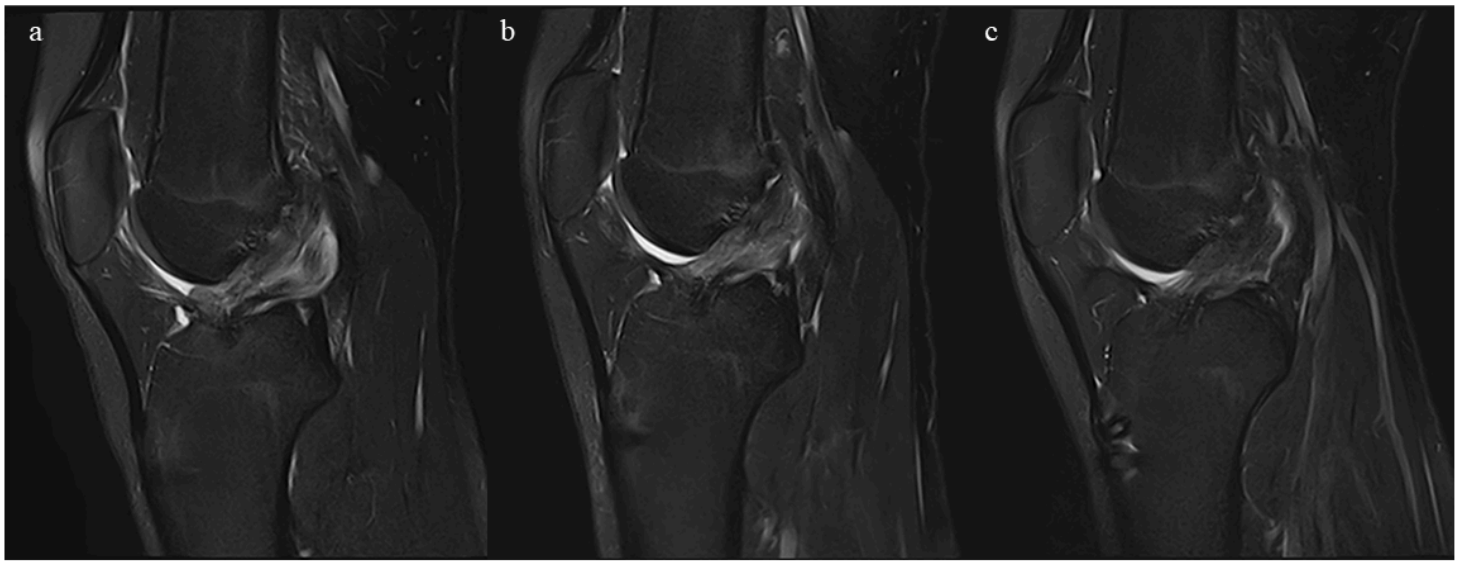


Figure 5

Sagittal knee MRI of the reconstructed ACL

a: Seven months postoperatively, increased signal intensity of the graft was observed. b: Eight months postoperatively, c: Nine months postoperatively. The signal intensity gradually improved over time.

ACL, anterior cruciate ligament; MRI, magnetic resonance imaging.

Fig. 6

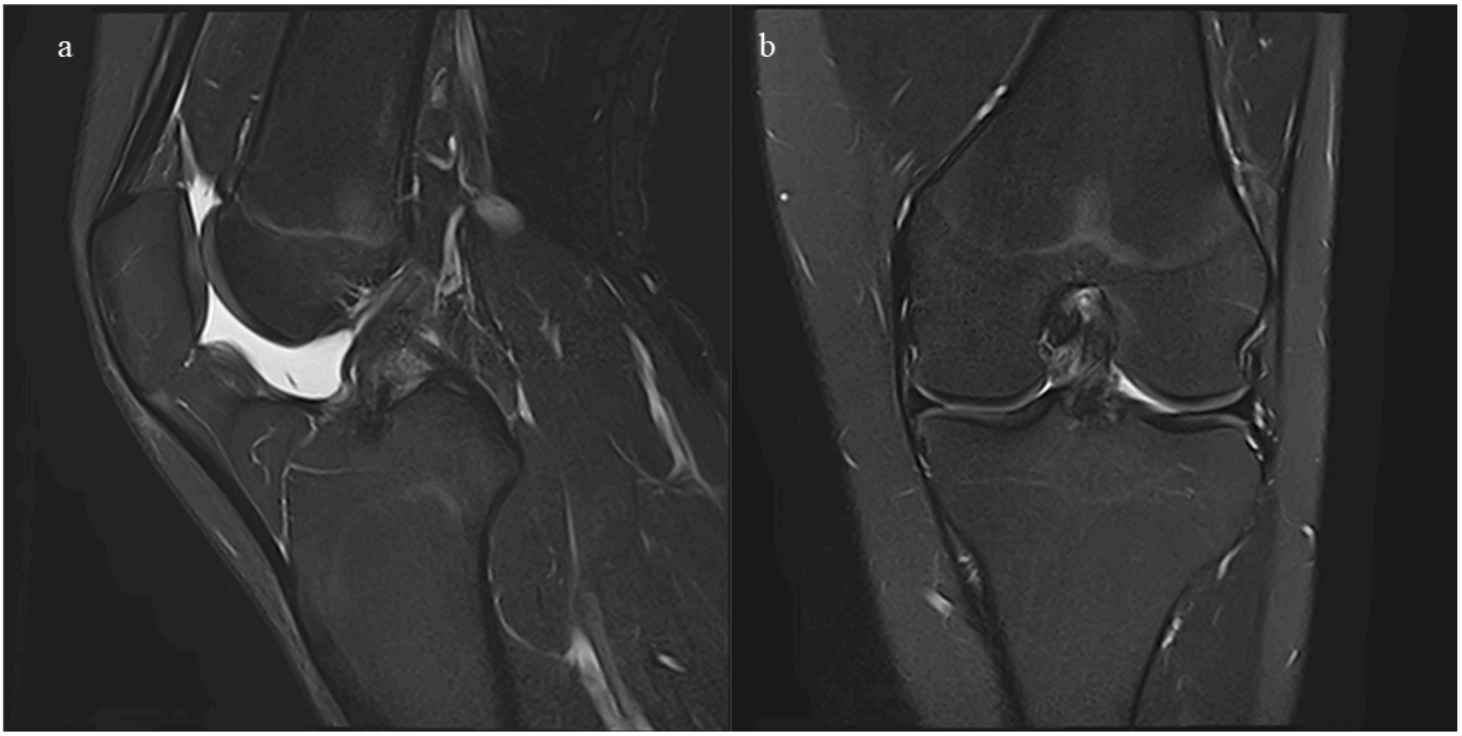


Figure 6

Knee MRI at 1 year after ACL reconstruction

a: Sagittal view, b: Coronal view. Mild joint effusion was observed, and the signal intensity of the reconstructed ACL had improved.

ACL, anterior cruciate ligament; MRI, magnetic resonance imaging.