CASE REPORT



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Heterotopic bone formation following anterior cruciate ligament reconstruction with BPTB autograft

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In this study, we present a 36-year-old male patient who developed heterotopic ossification after anterior cruciate ligament (ACL) reconstruction performed using bone-patellar tendon-bone autograft harvested from the 1/3 middle part of the patellar tendon. This ossified part, which restricted range of motion of the affected knee, was excised surgically 1 year after diagnosis. Physical examination, conducted 36 months later, revealed the achievement of full range of motion without any complaints or recurrences. Heterotopic ossification following ACL reconstruction is a very rare complication, which should be removed with open surgery.

Key words: ACL reconstruction; bone-patellar tendon-bone autograft; heterotopic ossification.

Reconstruction of the anterior cruciate ligament (ACL) using patellar tendon autografts was considered the gold standard till the late 1990s. However, as with every surgical modality, ACL reconstruction with bone-patellar tendon-bone (BPTB) graft has many complications. Local sensitivity at the donor site, infrapatellar nerve damage, rupture or inflammation of the patellar tendon, loss of terminal extension, patellar fracture, anterior knee pain and difficulty in sitting on the knees have been reported as the most significant complications.^[1-9] Few studies, however, have revealed heterotopic ossification following ACL reconstruction with BPTB.^[4,10,11]

In this study, we present the rare complication of heterotopic ossification after ACL reconstruction performed using patellar tendon autograft.

Case report

A 36-year-old male patient injured his left knee while playing football. After initial orthopedic physical and radiological examinations, he was diagnosed with ACL rupture. Three months after the injury, in December 2005, the patient underwent arthroscopic ACL reconstruction using patellar tendon autograft in another hospital. He was rehabilitated in the hospital for 7 days postoperatively and a home-exercise program was instituted for 1 month. No problems were detected during the early postoperative period. Follow-up evaluations, 6 weeks postoperatively, revealed the presence of a painless knee without any instability, and a knee range of motion (ROM) between 30 and 90 degrees. An area of ossification localized in the patellar tendon was observed on radiographs.

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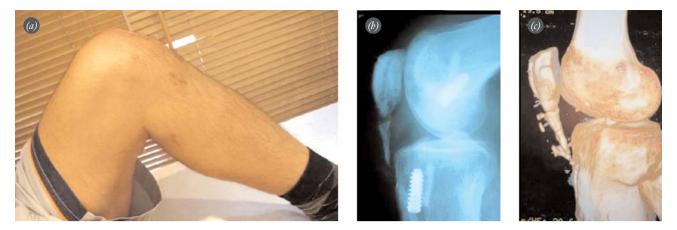


Fig. 1. (a) Clinical picture, (b) radiograph, (c) CT of the patient 4 months after the index operation. [Color figure can be viewed in the online issue, which is available at www.aott.org.tr]

The patient was referred to us with related complaints and diagnosed with heterotopic ossification. The patient did not receive indomethacin or radiotherapy because of late diagnosis. Diagnostic arthroscopy under general anesthesia was planned in April 2006 due to the persistence of the patient's complaints and restricted ROM (Fig. 1). Physical examination of the knee revealed no laxity and intra-articular abnormality was not seen. As knee ROM was between 30 and 90 degrees, full ROM could only be obtained through manipulation of the knee. Close observation was decided upon to see the completion of the maturation of bone formation. A continuous passive motion (CPM) device was applied under epidural anesthesia to initiate knee motions during the immediate postoperative period. The patient was discharged when a knee ROM of 0 to 130 degrees was achieved following 3 weeks of rehabilitation.



Fig. 2. Lateral radiograph of the patient, taken at the 1st year follow-up visit after the index operation.

The patient reapplied to our institution in July 2006 with the same complaints. Clinical examination revealed the absence of instability with a knee ROM of 20 to 90 degrees. Radiological evaluation suggested the presence of inadequate bone maturation. To regain normal knee ROM, manipulation of the knee performed under general anesthesia resulted in a knee ROM of 130 degrees. A circular cast was applied with the knee at full extension. The cast was removed on the third postoperative day, and full ROM exercises were started with the aid of a CPM device placed under epidural anesthesia. Following 3 weeks of rehabilitation, range of knee motion was 0 to 130 degrees with 10° loss of terminal active extension (0-10-130 degrees). In December 2006, twelve months after the index operation, the patient applied again to our clinic with restricted knee ROM (10 to 90 degrees) without pain or instability. However, a considerably hard ossification area was noted during palpation of the affected patellar tendon. A marked ossified mass without any problem related to fixation materials was radiographically observed (Fig. 2). Blood tests were within normal limits. Lysholm score (79 points), Tegner activity level (4 points) and IKDC (International Knee Documentation Committee) score (D) were assessed. These findings suggested complete maturation of bone formation and open surgery was planned for the next day. The patellar tendon was exposed and two mass lesions with dimensions of 10x45 mm and 10x20 mm were excised (Fig. 3). Biopsy material was obtained from the excised bone for pathological and microbiological analyses. After meticulous hemostasis, the tendon sheath was sutured. A subcutaneous drain was placed and the layers were closed properly. The knee was immobilized in full extension for 3 days. Then a controlled rehabili-

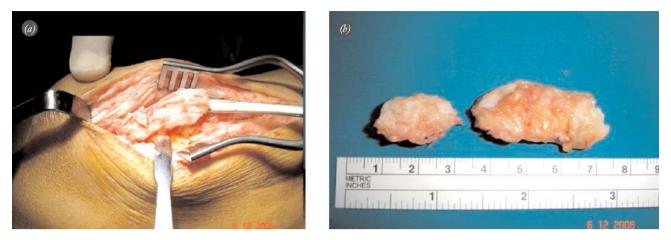


Fig. 3. (a, b) Pictures from the operative procedure. [Color figure can be viewed in the online issue, which is available at www.aott.org.tr]

tation program was applied to improve knee ROM. Oral indomethacin prophylaxis (35 mg, TID) was used for 6 weeks postoperatively. Microbiological examination of the specimens did not reveal any microbial growth, while the histopathological examination indicated "metaplastic bone formation in hyalinized connective tissue" (Fig. 4). At the end of the 3rd postoperative month, full range of knee motion was achieved and the knee regained its load-bearing capacity without any new bone formation. On the final follow-up in November 2009, a knee ROM of 0 to 135° (0-5-135 degrees) with normal muscular forces was detected. However, a 5° loss of terminal extension, lack of instability with associated Lysholm score (95 points), Tegner activity level (9 points), and IKDC score (A) were noted (Fig. 5). Examination of knee radiographs revealed complete fusion between the graft and the affected bone, stable implants without any enlargement of the bone tunnels, and the development of newly formed bone tissue (Fig. 6). The patient was able to kneel and sit on his knees without problems.

Discussion

Currently, BPTB and hamstring tendons are the preferred materials in the very frequently practiced surgical management of ACL. Success rates above 90% have been reported for ACL reconstruction using middle 1/3 patellar tendon graft.^[1-9] However, ACL reconstruction using patellar tendon graft is associated with many problematic outcomes. In the literature, patellar fractures, patellar tendinitis and/or rupture, patella infera syndrome, muscular weakness of quadriceps, arthrofibrosis, anterior knee pain, patellar sensitivity, difficulty in sitting on knees, shortening of patellar

tendon, loss of terminal extension, and patellofemoral pain have been reported as the most common complications.^[1-3,5,9] Heterotopic ossification in the harvested site of the patellar tendon is a very rarely seen complication and literature related to the formation of ossified issue is scarce.^[4,10,11] Specchiulli et al.^[4] and Lerat et al.^[10] reported that formation of osteophytes on the edge of the patella following ACL reconstruction is frequently seen and does not necessitate therapeutic intervention. A case report presented by Erdoğan et al. had similar characteristics with ours.^[11] Their 43-yearold male patient developed patellar tendon calcification measuring 2x1 cm 18 months after ACL reconstruction and no complaint was reported for the first 136 months after excision of this lesion. In our 36year-old patient, heterotopic ossification was detected

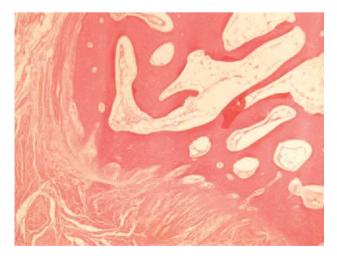


Fig. 4. Histologic appearance of the excised bone. (H-E stain; original magnification, x40). [Color figure can be viewed in the online issue, which is available at www.aott.org.tr]



Fig. 5. (a, b) Clinical pictures of the patient, taken at the 3rd year follow-up visit after excision of the heterotopic bone. [Color figure can be viewed in the online issue, which is available at www.aott.org.tr]

4 months after ACL reconstruction and surgical intervention was delayed until completion of its maturation process. At the final follow-up, 36 months after the last surgical intervention, we noted that the patient had no symptoms and/or recurrences and could perform daily activities without complaint.

Erdoğan et al. termed this new formation "patellar tendon calcification" following ACL reconstruction.^[11] In our case, we used the term "heterotopic ossification", as this bone formation developed after surgical trauma. In a study by Järvelä et al.,^[12] ACL reconstruction with patellar autograft was performed. At the end of the approximately 10-year follow-up, the patellar tendon site where graft material was harvested was examined using ultrasound and power Doppler US. In all but 3 patients, calcification, patellar tendon thicken-



Fig. 6. Lateral radiograph of the patient taken at the 3rd year follow-up visit after excision of the heterotopic bone.

ing and peritendinous changes in the patellar tendon where the graft material was obtained was detected. In our case, since the newly formed ossification lump was palpable and easily detectable on X-ray, we did not use US or Doppler US. Histopathologic examination of the biopsy material excised from the mass was reported as "metaplastic bone formation". We preferred to use the term "heterotopic ossification" for the new bone formation because of its development after surgical trauma, its progressive nature, and characteristic histopathologic features.

The most important cause of heterotopic ossification following surgical trauma is inadvertently scattered bone debris in the operative working field. In addition, aggressive rehabilitation programs may cause micro tears in the tendon, leading to bleeding.^[2,10-13,19] We think that these bone fragments and hematoma result with time in progressive bone formation in the bone bed and tendon. Even if this complication is very rare, we suggest meticulous harvesting and preparation of the graft material to avoid unnecessary future surgeries.

Relevant studies in the literature reported extremely higher rates of heterotopic ossification after brain injury and surgical interventions. This newly formed periarticular ossification was reportedly encountered most frequently in the hip and shoulder and is localized at a much lesser rate (3%) around the knee joint.^[13-19]

Surgical management is indicated especially when the restricted range of knee motion is due to a heterotopic ossification. To remove the heterotopic ossification, completion of its maturation should be awaited. Literature findings recommend a close observation period of 6 to 12 months, excision of the bone mass after visualization of complete bone maturation on 2 or 3 radiographs, and the disappearance of related hot spots on bone scans.^[13-17,19] We monitored our patient for 8 months and performed open surgical excision at completion of bone maturation as evidenced by radiological and clinical examinations one year after reconstructive surgery. At the end of the third postoperative year, there was no recurrence of complaints.

Recurrence is a frequently encountered problem after excision of the heterotopic ossification. To avoid this post-excisional ossification, prophylactic radiotherapy and indomethacin therapy have been recommended.^[13-15,17] For our patient, we prescribed indomethacin for 6 weeks. After 3 years of follow-up, recurrence was not observed. However, we believe that a longer followup period is required to arrive at a definite conclusion.

In conclusion, although heterotopic ossification following ACL reconstruction performed using patellar tendon autograft is a very rarely seen complication, it is important because of its ensuing morbidity. To avoid this complication, the graft must be meticulously prepared and any bone debris must not be left inside the operative field. After completion of its maturation, the heterotopic bone should be removed with surgical excision.

Conflicts of Interest: No conflicts declared.

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