



Patellar ligament rupture during total knee arthroplasty in an ochronotic patient

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Ochronotic arthropathy mainly involves the spine and large joints. Along with blackening of the joint, degeneration rapidly progresses mostly in the knee, resulting in symptoms by the 4th or 5th decade. As the role of medical treatment and joint conservation surgeries are limited in the early stages, joint replacement is the only effective option in one third of patients. We present a case of the unique complication of patellar ligament rupture during total knee replacement (TKR) of an ochronotic joint. A 51-year-old male presented with bilateral severe tricompartmental osteoarthritis with varus deformities and restriction of motion. Bilateral TKR was performed. At the 28-month follow-up, the patient was walking pain free with acceptable position of implants in radiographs. To our knowledge this is the first report of rupture of the patellar ligament during TKR of an ochronotic joint. We propose appropriate preoperative preparation and greater care in the handling of the tendon during TKR of an ochronotic joint in order to avoid complication.

Key words: Ligament rupture; ochronosis; total knee replacement.

Blackening of the joint may be due to endogenous ochronosis (deficiency of homogentisic acid oxidase) or, rarely, exogenous ochronosis (due to accumulation of hydroquinone, resorcinol, phenol, mercury or picric acid).

^[1] Endogenous ochronosis is a rare autosomal recessive disorder with the triad of alkaptonuria, ochronosis and ochronotic arthropathy.^[2] Clinical signs of ochronosis are due to the arthropathy of the spine and large joints.^[3,4]

Medical treatment of this rapidly progressive arthritis is limited to vitamin C in high doses, nitisinone, N-acetylcysteine, chondroitin or glycosaminoglycan and symptomatic treatment with analgesics and physiotherapy.^[5,6] Arthroscopic debridement and drilling may be helpful in the early stage, but most of these patients will eventually require joint replacement.^[3]

Considering that the knee is the most commonly replaced symptomatic joint, any complication occurring during total knee replacement (TKR) is of great importance. In this report, we present the unique complication of patellar ligament rupture during TKR of an ochronotic joint. To our knowledge this is the first report of ligament rupture during TKR of an ochronotic knee.

Case report

A 51-year-old male presented with pain and restriction of movement of both knees for 6 months. On examination, both knees had mild swelling, joint line tenderness, crepitus, 10° of varus deformities and restriction of terminal 20° of flexion and 10° of extension. Radiographs showed bilateral severe tricompartmental involvement,

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joint space reduction and osteophyte formations (Fig. 1). A diagnosis of primary osteoarthritis was made and bilateral TKR was planned. Pre-anesthetic investigations were normal.

The left knee was operated first. On exposure, parts of the patella, femoral and tibial condyles and portions of the joint capsule showed black discoloration (Fig. 2). The possibility of ochronosis was considered and surgery was continued. A cruciate-sacrificing, cemented TKR (PFC Sigma; DePuy, Warsaw, IN, USA) was performed, along with patellar resurfacing and deformity correction. The right knee showed more extensive involvement than the left. In the right knee, the distal half of the patellar ligament was heavily pigmented and very fragile, resulting



Fig. 1. Preoperative radiographs of the (a) right and (b) left knee.



Fig. 2. Intraoperative photograph of the knee showing blackened areas of the joint including undersurface of the patella and femoral condyle. [Color figure can be viewed in the online issue, which is available at www.aott.org.tr]

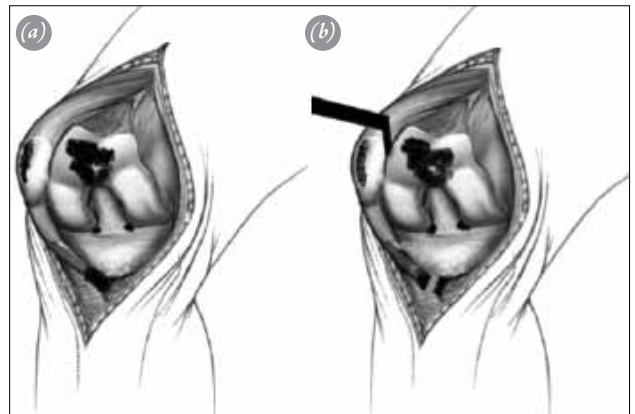


Fig. 3. (a) Involvement areas of the right knee and patellar ligament. (b) Rupture of the ligament at the distal attachment.



Fig. 4. Postoperative radiograph of the knee after 28 months.

in rupture during retraction with mild retraction force (Fig. 3). Replacement of the femoral, tibial and patellar surfaces was performed. The ruptured tendon was repaired with non-absorbable suture (Ethibond) using the Bunnell stitch through the tendon and a drill hole in the tibial tubercle. This was augmented with relaxing suture as described by West et al.^[7] The blackened articular cartilage was sent for histopathological study. The right knee was protected in extension with a brace. Ambulation was begun at the postoperative 4th week and range of movement exercise at the 6th week. At the final follow-up at 28 months, the patient was walking pain free. There was no extension lag, knee flexion range was up to 90° and implants were in acceptable position on radiographs (Fig. 4).

A retrospective provisional diagnosis of ochronotic arthritis was made. On further evaluation, the patient had a history of urine turning black after a few minutes, blackish pigmentation of the sclera and mild restriction of lumbosacral spine movements. Radiographs of the

lumbosacral spine showed calcification in the disc spaces (Fig. 5). Urine was positive for ferric chloride, sunlight standing and Benedict's tests. HLA-B27 test was negative. Histopathology showed pigment-laden tissues and giant cells confirming the diagnosis of ochronotic arthritis.

The patient's younger brother, aged 45, had experienced non-radiating low back pain for 1 year with a history of urine turning black on contact with air and blackish pigmentation of the sclera. The lumbar spine had mild scoliosis and restriction of movement with normal neurology. Radiographs showed diminished disc spaces with calcifications of disc materials. HLA-B27 test was negative. Urine was positive for ferric chloride, sunlight standing and Benedict's test. The patient's 2-year-old grandson also had blackish discoloration evident in his diapers and positive ferric chloride and Benedict's tests for urine, indicating alkaptonuria.

Discussion

Ochronosis is a hereditary disorder with male predominance and was evident in three cases reported from one family.^[8] As the extraskelatal features are subtle in many patients, the initial diagnosis of ochronosis may be missed.^[5,9] In the present case, the diagnosis could only be made intraoperatively. Arthropathy is the main cause of symptoms in alkaptonuric patients.^[3,8,10] Ochronosis



Fig. 5. Lateral view of the lumbosacral spine.

is the manifestation of a long-term process of homogentisic acid deposition in the cartilage, increasing fragility and thus allowing fissuring and early degeneration. The increased apoptosis and NO liberation by the affected chondrocytes and toxic granules liberated from the affected endothelium may also contribute to the damage.^[2,11] Together, these reactions result in fissuring and flaking of the cartilage. The flakes then attach to the synovium, producing thickening, fibrosis and chondromatosis.^[1] A rapidly progressive arthritis then ensues by the 4th or 5th decade of life.^[8]

Treatment of ochronotic arthropathy is symptomatic. Rapidly progressing tricompartmental involvement does not allow other joint-sparing procedures such as arthroscopic debridement or high tibial osteotomy.^[3] Kontinen et al. performed the first TKR in an ochronotic knee with cementless prosthesis.^[4] Since then, there have been many reports of successful TKRs in ochronosis cases. Though most of these authors reported no difficulty during the TKR, Spencer et al. reported one case in which the patellar tendon was stiff and attenuated, making patellar dislocation difficult.^[10] In the present case, the distal part of the right patellar tendon was heavily pigmented and attenuated and ruptured without any excessive retraction force or intraoperative accidental injuries. In the opposite knee, there was no involvement of the patellar tendon and the surgery was uneventful. Although there have been no previous reports of rupture of patellar tendon during TKR, Manoj Kumar et al.^[12] reported a case of spontaneous rupture of the patellar tendon at the proximal part and 3 spontaneous ruptures of the Achilles tendon in 2 patients. The authors noted pigmentation at the rupture sites and fibrillation and degeneration of the tendons and proposed that in these tendons, which primarily contain collagen Type 1, the homogentisic acid deposition inhibits collagen cross-linking. This leads to reduction of the structural integrity of the collagen, thus increasing the likelihood of rupture. Ando et al. examined a case of bilateral Achilles tendon rupture and found that the ruptured sites were devoid of normal collagen.^[13] Jebaraj et al. noted loss of fibrillary pattern in the tendon, increased thickness and small foci of calcification during ultrasonography in a case of Achilles tendon rupture.^[14]

Patellar tendon rupture during TKR is a major complication that may occur when the knee is tight or has scarring due to previous surgery and when flexed with the dislocated patella. Various treatment options for these ruptures have been described, including fixation with suture or staples, autologous tissue augmentation with hamstring tendon, turndown of the quadriceps

tendon or medial gastrocnemius flap or reconstruction with Achilles tendon allograft.^[15] In the present case, as the rupture occurred without any iatrogenic cause, we believe that the preexisting tendon degeneration could be responsible. We used the direct repair method and protected it by immobilizing the knee in extension for 4 weeks. The patient showed satisfactory results at final follow-up of 28 months.

Unfortunately, in the present case, preoperative diagnosis could not be made due to an inadequate history and lack of radiograph examination of the spine. Therefore, we suggest that ochronosis should be included in the differential diagnosis of patients with degenerative arthritis involving multiple joints and the necessary investigations should be performed to exclude it.

In conclusion, considering the high frequency of TKRs in ochronotic arthritis, the risk of intraoperative patellar tendon ruptures due to attenuation and degeneration of tendons can be significant. Therefore, preoperative diagnosis is essential so that greater care can be taken in handling the tendon during TKR and repair can be undertaken in case of any rupture. A preoperative ultrasonography of the patellar and Achilles tendons may also be helpful in anticipating such complications in advance.

Conflicts of Interest: No conflicts declared.

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