# Case Report / Olgu Sunumu

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# Bilateral Avascular Necrosis of the Knee Due to Corticosteroid Therapy in a Patient With Posterior Inferior Cerebellar Anterovenous Malformation

Posterior İnferior Serebellar Arterio-venöz Malformasyonlu Bir Olguda Yüksek Doz Kortikosteroid Kullanımına Bağlı Bilateral Diz Avasküler Nekrozu

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#### Summary

Osteonecrosis is a common complication of corticosteroid therapy. In this study, we report the case of a patient with Posterior Inferior Cerebellar Artery (PICA)-Arteriovenous (AV) malformation who has been diagnosed with knee osteonecrosis involving bilateral proximal tibial and distal femur due to high-dose corticosteroid use. A 27-year-old male patient presented with a history of subarachnoidal hemorrhage due to PICA-AV. He had undergone Gamma knife radiosurgery and received short-term high-dose corticosteroid therapy (dxamethasone 16 mg/kg for 21 days). Two years later, he developed bilateral anterior knee pain of insidious onset. Magnetic resonance imaging performed on admission showed osteonecrosis of the bilateral proximal tibial and distal femur, more pronounced in the tibia. Osteonecrosis is a fairly common complication in patients with the history of corticosteroid use for the treatment of variety of systemic and rheumatic disorders. The condition can manifest itself anywhere in the skeletal system, most commonly in the femoral head. Distal femur and proximal tibia with bilateral involvement is guite rare in the literature. Turk J Phys Med Rehab 2012;58:243-5. Key Words: Avascular necrosis, steroid, neurosurgery, knee

#### Özet

Osteonekroz kortikosteroid kullanımına bağlı sık görülen bir komplikasyondur. Literatürde sistemik ve romatizmal hastalıklarda oldukça sık rapor edilmiştir. Biz burada posterior inferior serebellar arterio-venöz malformasyonlu bir olguda yüksek doz kortikosteroid kullanımı sonucu bilateral femur distali ve tibia proksimalinde avasküler nekroz saptanan olguyu sunmayı amaçladık. Literatürde dizde yerleşim yeri olarak femur distali ve tibia proksimalinin birlikte tutulduğu avasküler nekrozlu olgu sayısı oldukça azdır. Olgumuz 27 yaşında erkek hasta, posterior inferior serebellar arterio-venöz malformasyonuna bağlı subaraknoidal hemoraji tablosu gelişmiş. Hastaya beyin cerrahi servisinde GAMA knife ışın tedavisi ve sonrasında 21 gün süre ile 16 mg/kg/gün dozunda deksametazon tedavisi uygulanmış. Tedaviden 2 yıl sonra hastanın her iki diz anterior bölgesinde ağrı yakınması başlamış. Manyetik rezonans görüntülemede bilateral femur distali ve tibia proksimalinde lokalize osteonekroz tespit edildi. Osteonekroz sistemik ve romatizmal hastalığı olan olgularda kortikosteroid kullanımına sekonder olarak gelişen ve sık görülen bir komplikasyondur. Steroid kullanımına bağlı osteonekroz en sık femur başında görülür. Bilateral femur distali ve tibia proksimalinin birlikte tutulumu ise oldukça nadir görülmektedir. Türk Fiz Tıp Rehab Derg 2012;58:243-5. Anahtar Kelimeler: Avasküler nekroz, steroid, nörocerrahi, diz

Address for Correspondence:/Yazsma Adresi: Ferdi Yavuz MD, Military Hospital of Etimesgut, Department of Physical Medicine and Rehabilitation, Ankara, Turkey Gsm: +90 544 343 54 58 E-mail: ferdiyavuz@yahoo.com **Received/Geliş Tarihi:** February/Şubat 2011 **Accepted/Kabul Tarihi:** April/Nisan 2011 ine and Rehabilitation, Published by Galenos Publishing. / © Türkiye Fiziksel Tip ve Rehabilitasyon Dergisi, G

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#### Introduction

Avascular osteonecrosis or non-traumatic necrosis is a fairly common bone disease, a devastating adverse effect of corticosteroid therapy, thought to be caused by an aseptic ischemic insult to the cellular elements of the bone marrow (1). It is considered to develop in patients who receive very high-dose shortterm, low-dose long-term corticosteroids or even intra-articular injections as widely reported in the literature (2). Osteonecrosis most commonly affects the hip, but may also affect other sites including the knee, shoulder, ankle, spine and other joints (3). The reported incidence of hip avascular necrosis is estimated to be between 0.3 and 0.6% (4). Several possible hypotheses for explanation of the pathogenesis of steroid-induced osteonecrosis, including increased size and number of fatty cells, increased intra osseous pressure, fatty degeneration of osteocytes, fat embolism and extra osseous arterial occlusion have been proposed in the literature (5). High-dose corticosteroid therapy causes an immediate and persistent decrease in bone formation, and a rapid and transient increase in bone resorption resulting in osteonecrosis (6). A decrease in bone mineral density (7) and apoptosis of osteoblasts and osteocytes (8) in response to high-dose corticosteroids are the other possible mechanisms for the development of osteonecrosis. There are specific areas of the skeleton for which this disease seems to have predilection. These include the femoral condyles, femoral and tibial heads, proximal tibia and some small bones of the ankle and the foot. The most commonly reported area is the femoral head followed by the tibial condyles. Physiotherapy, analgesics and anti-inflammatory drugs do give some symptomatic relief but do not alter the course of the disease.

## **Case Report**

Our patient a 27-year-old man was referred to the physical medicine and rehabilitation outpatient clinic with a 2-month history of bilateral severe knee pain which rendered him bed bound and forced to get admission into a hospital. There was no history of recent fever, chill or urinary symptoms. However, he gave a history of high-dose steroid intake in the past. Three years ago, when he reported to a neurosurgery emergency department

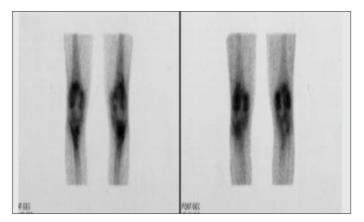


Figure 1. Isotope bone scan showing the areas of increased uptake in the distal femur and proximal tibia.

due to profound headache; he had been diagnosed as having posterior inferior cerebellar artery arteriovenous malformation based on Magnetic Resonance Imaging (MRI) findings. He underwent GAMA knife radiosurgery and was put on high-dose corticosteroids - dxamethasone 16 mg/kg - for 21 days during his post-procedure stay in an intensive care unit. Two years later, symptom of anterior knee pain started insidiously and he started experiencing progressive difficulty with his routine activities. However, his symptoms responded well to symptomatic treatment, no investigations were carried out at that time. One year later, he developed disabling knee pain rendering him unable to bear weight and walk. Both knees were tender to touch, especially on anterior aspect, joint range of motion (ROM) was too a bit reduced. Bilateral quadriceps wasting was appreciable. Rest of the systemic and musculoskeletal examination was unremarkable. Plain radiographs of the knee were unremarkable less mild osteoporosis in bilateral proximal tibia. Complete blood count also did not give any clue of an acute or chronic inflammatory process. 24-hour calcium level was 293 mg, dualenergy x-ray absorptiometry scan showed a T-score -1.4 (right knee), -2.0 (left knee) with normal serum alkaline phosphate level of 67 U/L. The patient was treated initially with conservative treatment comprising of 30 sessions of hyperbaric oxygen therapy, non-steroidal anti-inflamatory drugs (NSAIDs), cold packs, ROM exercises and bed rest, however, all this did not offer much in terms of relief. Keeping in view the severity of the pain, bone scan and MRI of both knees were carried out. Isotope bone scan showed an increased focal uptake in the proximal tibia and distal femur, bilaterally (Figure 1) MRI revealed bilateral proximal tibial and distal femoral osteonecrosis, more pronounced in the tibia, bilaterally (Figure 2). Core decompression of the bilateral proximal tibial regions was carried out by an orthopaedic surgeon, thus, his symptom of pain was reduced. Post-operatively, analgesics, isometric quadriceps and ROM exercises for the knee and hip were advised. At three-month follow up, patient was painfree with normal looking gait pattern; joint ROM was normal and his only limitation was feeling of mild discomfort on kneeling for his prayer activities.

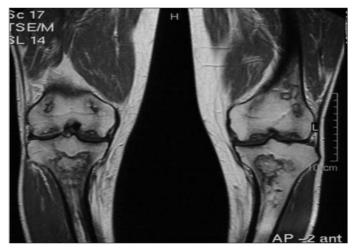


Figure 2. MRI showing the areas of osteonecrosis in the bilateral proximal tibia and distal focal areas of the femur.

# Discussion

Osteonecrosis has been reported during or after the course of steroid treatment in several conditions such as sickle cell disease, systemic lupus erythematosus, ulcerative colitis and Crohn's disease (9). Corticosteroids are believed to enhance the micro vascular ischemia by decreasing bone blood flow along with increased bone marrow pressure due to intramedullary lipocytes hypertrophy (10). The condition can manifest itself anywhere in the skeletal system, most commonly in the femoral head, but similar changes have been reported in the proximal knee, humerus, elbow and the foot (3). No clear cut rules exit regarding the dose and duration of corticosteroid treatment followed by manifestation of osteonecrosis. Reported cases have documented it as early as 6 months to as late as three years (11). On animal model, it is reported to be found one week after the initial steroid administration (12). Osteonecrosis begins insidiously and often the diagnosis is easily missed and delayed due to frequently normal plain radiographs in early part of the disease even in the presence of pathological changes. MRI has been reported to be more sensitive and specific to detect osteonecrosis in an early phase (13). In symptomatic patients with negative plain radiographs or MRI findings, the radionuclide bone scan is recommended. It is highly sensitive for demonstrating the areas of enhanced focal uptake before the changes are apparent on other imaging modalities (14). Conservative treatment options including analgesics, braces, reduced weight bearing, bed rest, deep heat modalities and ROM exercises are offered, but nothing has been proved to be of much significance besides offering a temporary symptomatic relief. None of the treatment options are believed to alter the course of the disease. If diagnosed at an early stage, collapse of the subchondral bone and progression of the disease may be averted in some patients by decreasing the joint stress and by training mobility. Different surgical approaches including core decompression, curettage, and bone transplant have been tried with contradictory outcomes, however, the ultimate treatment is often a joint replacement (15).

# Conclusion

Osteonecrosis is a common complication in patients with a history of short-term or long-term corticosteroid use but difficult to diagnose with simple conventional imaging techniques. A careful examination with high index of suspicion is essential while dealing with patients with anterior knee pain. MRI and radionuclide bone scan are helpful in detecting a plain radiograph negative lesion.

#### **Conflict of Interest:**

Authors reported no conflicts of interest.

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