Eskisehir Medical Journal
Eskişehir Tıp Dergisi

Eskisehir Med J. 2023; 4(2): 151-153
doi: 10.48176/esmj.2023.124

Case Report / Olgu Sunumu

OSTEOPOIKILOSIS AS A RARE CAUSE OF BILATERAL KNEE PAIN: A CASE REPORT AND REVIEW OF LITERATURE

BİLATERAL DİZ AĞRISININ NADİR NEDENİ OLARAK OSTEOPOİKİLOZ: OLGU SUNUMU VE LİTERATÜRÜN İNCELENMESİ

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ABSTRACT

Background: Osteopoikilosis is a benign, asymptomatic disease, often diagnosed incidentally. It is characterized by symmetric, multiple, well-defined, oval sclerotic lesions distributed in the periarticular epiphysis and metaphysis of long bones. In symptomatic patients, treatment usually relies on pain relief. This case report presented an example of this rare disorder.

Case Report: A 24-year-old male patient with unremarkable personal, trauma, or family history was observed at the orthopedics and traumatology department due to bilateral knee pain. Physical examination revealed no specific findings. Antero-posterior (AP) and lateral radiographic views revealed several sclerotic oval blastic lesions in both knees. The patient was diagnosed with osteopoikilosis and oral naproxen sodium was prescribed for joint pain.

Conclusions: Osteopoikilosis is a frequently asymptomatic bone dysplasia. Its clinical importance originates from differentiating other sclerosing pathologies, especially osteoblastic metastatic conditions. Joint pain is the most common symptom and is usually treated well with nonsteroidal anti-inflammatory drugs.

Keywords: Bone abnormalities; Bone dysplasia; Osteopoikilosis; Sclerotic lesions; benign

ÖZET

Giriş: Osteopoikilosis benign, asemptomatik bir hastalıktır ve genellikle tesadüfen teşhis edilir. Uzun kemiklerin periartiküler epifiz ve metafizlerinde dağılım gösteren, simetrik, çok sayıda, iyi sınırlı, oval sklerotik lezyonlarla karakterizedir. Semptomatik hastalarda tedavi genellikle ağrının giderilmesine dayanır. Bu vaka raporu, bu nadir bozukluğun bir örneğini sunmaktadır.

Olgu Sunumu: Kişisel, travma veya aile öyküsünde özellik olmayan 24 yaşında erkek hasta, bilateral diz ağrısı nedeniyle ortopedi ve travmatoloji polikliniğine başvurdu. Fizik muayenede spesifik bir bulgu saptanmadı. Ön-arka (AP) ve yan radyografik görüntülerde her iki dizde birkaç sklerotik oval blastik lezyon görüldü. Hastaya osteopoikiloz tanısı kondu ve eklem ağrısı için oral naproksen sodyum reçete edildi.

Sonuç: Osteopoikilosis sıklıkla asemptomatik bir kemik displazisidir. Klinik önemi, özellikle osteoblastik metastatik durumlar olmak üzere diğer sklerozan patolojileri ayırt etmesinden kaynaklanmaktadır. Eklem ağrısı en sık görülen semptomdur ve genellikle nonsteroidal antiinflamatuar ilaçlarla iyi bir şekilde tedavi edilir.

Anahtar kelimeler: Kemik anormallikleri; Kemik displazisi; Osteopoikiloz; sklerotik lezyonlar; benign

INTRODUCTION

Osteopoikilosis, also known as osteopathy condensans disseminate or “spotted bone disease” is a rare osteosclerotic dysplasia. It is a benign and usually asymptomatic disease. Although there are cases without a family history, it is inherited in an autosomal dominant manner (1). It is usually an asymptomatic disorder and is often diagnosed incidentally. However mild articular pain with or without effusion was reported in 15 to 20% of cases (2). The vast majority of patients are asymptomatic and treatment is not necessary. It is characterized radiologically by the presence of symmetric, multiple, well-defined, oval, or rounded sclerotic lesions distributed periarticular epiphysis and metaphysis of the long bones. In symptomatic patients, treatment usually relies on pain relief through the use of non-steroidal anti-inflammatory drugs. The coexistence of other rheumatic diseases has been described in the literature (3, 4). In this care report, an example of this rare disease is presented.

CASE REPORT

A 24-year-old male patient with unremarkable personal, trauma, or family history was observed at the orthopedics and traumatology department due to bilateral knee pain. Oral and written informed consent was taken from the patient. Physical examination revealed a full range of motion, but no specific findings. Antero-posterior (AP) and lateral radiographic views were performed on both knees which revealed several sclerotic oval blastic lesions of approximately 2 to 4 mm in diameter in the periarticular areas (Figure 1). Blood tests for calcium-phosphor metabolism and rheumatic diseases were taken and resulted in normal values. In MRI view lesions were small and dark on both T1- and T2 sequences, which shows mature dense bone (Figure 2). Bone scintigraphy was not necessary as no malignancy was differentiated in diagnosis. The patient was diagnosed with osteopoikilosis and oral naproxen sodium was prescribed for joint pain. The patient was called for a radiographic investigation of possible lesion sites and

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Received date: 20.03.2023 **Accepted date:** 29.05.2023

Cite as: İmre E. Osteopoikilosis As A Rare Cause Of Bilateral Knee Pain: A Case Report And Review Of Literature. Eskisehir Med J. 2023; 4(2): 151-153 .doi: 10.48176/esmj.2023.124.

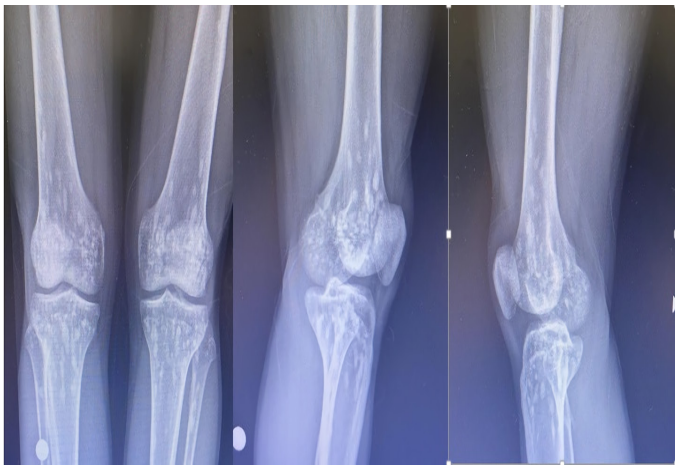

Figure 1. Multiple sclerotic oval lesions in AP and lateral x-ray views of both knees.

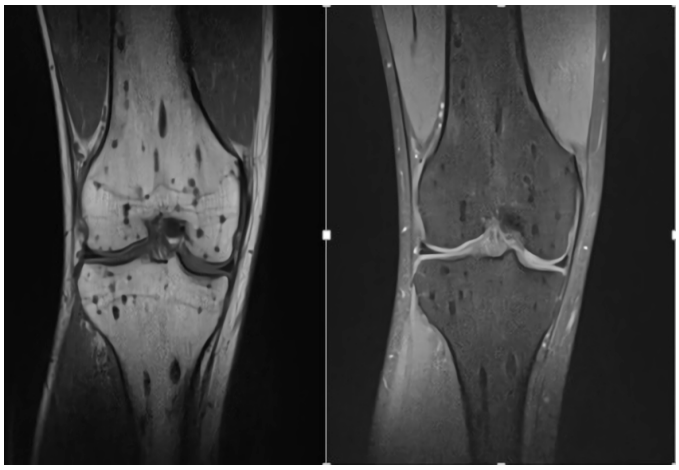

Figure 2. Small and dark lesions in both T1 and T2 sequences of MRI.

familial investigation but was out of follow-up.

DISCUSSION

Osteopoikilosis is a rare, benign condition, usually diagnosed by radiographic imaging of multiple ovoid sclerotic opacities on the metaphyseal area of bones incidentally (5). Osteopoikilosis is first described by Stieda in 1905 (6). The more anatomical-pathologic basis term osteopathia condensans disseminata was suggested by Lowrey and Booth (6). Osteopoikilosis, meaning “spotted bone” is descriptive of the x-ray view. Even though it is a sclerosing bone dysplasia, it is a benign disease with no specific treatment required. It is not associated with skeletal symptoms of fractures, scoliosis, hip osteoarthritis, and osteomyelitis, unlike osteopetrosis (7).

Osteopoikilosis is known to be a benign disease, however, if presented, mild joint pain with or without effusion is the most reported (%15 to 20) clinical manifestation just as the only symptom in this case (1, 2). Non-steroidal anti-inflammatory drugs are used for pain relief. In literature, other orthopedic conditions such as spinal stenosis, hip fracture, and malignant degeneration were also reported (8, 9). Besides the musculoskeletal conditions, skin and organ abnormalities were reported which wasn’t presented in this case (9-13). However, its association with these conditions has not yet been confirmed. In this case, the patient has mild joint pain which was treated with a non-steroid anti-inflammatory drug, Naproxen sodium. There was no other symptom; neither musculoskeletal nor dermatologic or any other.

A most important finding is multiple sclerotic ovoid lesions in 1-10 mm diameter. Mostly this evidence is found in metaphyses of long bones, distal femoral metaphysis in this case. Those findings have never been reported in the cranium, unlike osteopetrosis. Though, osteoblastic metastatic conditions, such as prostate carcinoma, should be excluded at a differential diagnosis. Bone scintigraphy is important in differentiating as results are normal in osteopoikilosis. In this case, the patient age was 24 and there was no other complaint but mild joint pain, so bone scintigraphy wasn’t considered necessary.

Genetically osteopoikilosis is associated with LEMD3 gene mutation. Although it is known to have an inheritance in an autosomal dominant manner, sporadic cases are reported (3, 14, 15). In this case, the patient was out of follow-up, so we didn’t have any other clue if this is a sporadic case or not.

This study is a level 5 case report study, with a low-level evidence value. Since osteopoikilosis is a rare disease, these kinds of cases were reported in the same manner or as case series. Follow-up time was a short period. For contributing more to the literature, case series with longer follow-up time and meta-analysis of those case reports are required. It is believed that this case report will contribute to literature as a sample of this rare,but important to differentiate, disease and as a case for further systemic review.

CONCLUSIONS

Osteopoikilosis is a frequently asymptomatic benign bone dysplasia. Its clinical importance originates from differentiating other sclerosing pathologies, especially osteoblastic metastatic conditions considering age and symptoms. Joint pain is the most common symptom and is usually treated well with nonsteroidal anti-inflammatory drugs.

Informed Consent: Informed consent was taken from the patient.

Authorship Contributions: Idea/Concept: Eİ, Design: Eİ, Supervision: Eİ, Data Collection or Processing: Eİ, Analysis or Interpretation: Eİ, Literature Search: Eİ, Writing: Eİ, Critical Review: Eİ, References And Fundings: -, Materials: Eİ.

Conflict of Interest: No conflict of interest was declared by the authors.

Financial Disclosure: The authors declare that they have no relevant financial.

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