Insidentally Diagnosed Osteopoikilosis After Knee Pain

Erdal Uzun¹, İlknur Uzun²

¹Orthopedic and Traumatology Clinic, Kayseri Research and Training Hospital, Kayseri, Turkey. ²Department of Internal Medicine, Faculty of Medicine, Erciyes University, Kayseri, Turkey.

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Abstract

Osteopoikilosis is a rare, benign, autosomal dominant disorder characterised by sclerotic bone lesions most commonly involving the hands, feet, pelvis, and ends of long bones. Lesions are typically found incidentally on imaging studies done for unrelated complaints. It may resemble bone tumor, metabolic disease, mastocytosis and tuberous sclerosis. Early recognition is essential to prevent unnecessary emotional distress and invasive testing. Herein we report a case presenting with knee pain whose radio¬logic investigation revealed typical osteopoikilotic lesions.

Key words: Osteopoikilosis, bone dysplasia, radiography, diagnosis.

Address for correspondence/reprints:

Erdal Uzun Telephone number: +90 (352) 336 8884 E-mail: nuzuladre@gmail.com

DOI:

Introduction

Osteopoikilosis is also named as "spotted bone disease" an asymptomatic osteosclerotic dysplasia initially described by Albers-Schönberg in 1915 (Resnick et al., 1988). It is a rare, benign, autosomal dominant disorder characterised by sclerotic bone lesions most commonly involving the hands, feet, pelvis, and ends of long bones (Woyciechowsky et al., 2012; Paraskevas et al., 2009). Skalp, vertebrae, costa and mandibular involvement were less commonly reported (Resnick and Niwayama, 1995). Prevalence has been estimated to be 1/50,000, male and female are equally affected, and it may occur at any age (Woyciechowsky et al., 2012). Lesions are typically found incidentally on imaging studies done for unrelated complaints (Carpintero et al., 2004). It may resemble bone tumor, metabolic disease, mastocytosis and tuberous sclerosis (Resnick et al., 1988). Therefore, patients with the diagnosis of mass history should be evaluated with physical examination, labolatuary and radiology together (Dabak et al., 2012). The symmetric distribution, lack of bone destruction, and location differentiates osteopoikilosis from other

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pathologies (Tuncel and Caner, 2012). Early recognition is essential to prevent unnecessary emotional distress and invasive testing (Carpintero et al., 2004). No routine follow-up or studies are necessary

Case Report

A 20-year-old female came to our hospital with right knee pain for about two months after a minor trauma. On physical examination her right knee medial joint space was painful with palpation accompanied by mild suprapatellar swelling and no other joints were affected. All joints had a full passive range of motion and other pyhsical examination was normal. He was in good health and had no history of illness or drug use. On X-ray the affected knee showed that there were circumscribed sclerotic areas near the ends of the tibia and femur (Figure 1). To explore possible intraarticular pathologies we considered to look Magnetic Resonance Imaging (MRI) of the affected knee. MRI showed no intraarticular signs but in T1 sequences there were multiple, welldefined, millimeter-sized sclerotic areas near the ends of the tibia and femur (osteopoikilosis) as shown on X-ray (Figure 2). Then bone scan done to explore the other parts of the body and showed multiple ovoid, radiodense bony lesions in the pelvis and proximal femurs, consistent with the diagnosis of osteopoikilosis. The patient used ibuprofen for 10 days and full resolution was achieved with 10 days rest.



Figure 1: On X-ray the affected knee showed that there were circumscribed sclerotic areas near the ends of the tibia and femur



Figure 2: MRI showed no intraarticular signs but in T1 sequences there were multiple, well-defined, millimeter-sized sclerotic areas near the ends of the tibia and femur (osteopoikilosis).

Discussion

Osteopoikilosis is a rare, benign, autosomal dominant disorder characterised by sclerotic bone lesions most commonly involving the hands, feet, pelvis and ends of long bones (Woyciechowsky et al., 2012; Paraskevas et al., 2009). Lesions are typically found incidentally on imaging studies done for unrelated complaints (Carpintero et al., 2004). In some patients with osteopoikilosis joint pain and effusions were reported but so far a clear relationship between osteopoikilosis and joint symptoms has not shown (Resnick et al., 1988). In this report the patient had medial knee pain with palpation accompanied by mild suprapatellar swelling but we thought this pain was not associated with osteopoikilosis. She had moderate pain for two months, bu she had no similar pain earlier.

Osteopoikilosis is an important disorder to be kept in mind during differential diagnosis with metastases. tuberous osteoblastic sclerosis. mastocytosis, osteopathy, melorheostosis. It can be distinguished by metaphyseal and epiphyseal involvement, symmetrical distribution and uniform size of the lesions (Appenzeller et al., 2007). In this report imaging of the affected knee showed that there were circumscribed sclerotic areas near the ends of the tibia and femur and also bone scan showed multiple ovoid, radiodense bony lesions in the pelvis and proximal femurs, consistent with the diagnosis of osteopoikilosis.

In conclusion, patients with osteopoikilosis remain asymptomatic and diagnosed incidentally on X-ray. The symmetric distribution, lack of bone destruction, and location differentiates

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osteopoikilosis from other pathologies. The physicians must advise the patients with osteopoikilosis that the disease has benign course, and similar pathologies can be found with their family members, In these cases to prevent unnecessary and invasive testing and emotional distress, early recognition is essential. No routine follow-up or studies are necessary but it is important to keep it in mind at differential diagnosis..

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