Case Report
An uncommon cause of anterior knee pain in a child: Osteoid Osteoma of the patella and a review of the literature

Bir çocukta ön diz ağrısının nadir bir nedeni: Patellar Osteoid Osteoma ve literatürün gözden geçirilmesi

Nazan Kaymaz\textsuperscript{a}, Burak Kaymaz\textsuperscript{b}

\textsuperscript{a} Department of Pediatrics, Faculty of Medicine, Canakkale Onsekiz Mart University, Canakkale, Turkey
\textsuperscript{b} Department of Orthopedics, Faculty of Medicine, Canakkale Onsekiz Mart University, Canakkale, Turkey

ABSTRACT

\textbf{Introduction}: Osteoid osteoma (OO) is a small, benign bone neoplasm that has a well demarcated nidus surrounded by a reactive zone of sclerosis. It is most common in the long bones and only rarely in the patella.

\textbf{Case Presentation}: An eight-year-old girl was admitted to the outpatient clinic with the complaint of anterior knee pain. She had been suffering from pain for approximately one year. The patient had a patellar osteoid osteoma. Surgical curettage of the lesion was performed, and the patient was free of pain the day after the surgery.

\textbf{Conclusions}: Although Patellar OO is rare, it should be included in the differential diagnosis of persistent knee pain in children and young adults.

\textbf{Keywords}: Osteoid Osteoma, patella, knee pain, curettage, delayed diagnosis, bone neoplasms

ÖZ

\textbf{Giriş}: Osteoid Osteoma (OO) belirgin bir nidus ve onu çevreleyen sklerotik reaktif zon ile karakterize küçük selim bir kemik tümörüdür. Genellikle uzun kemiklerde yerleşen ve birlikte nadiren patellada da görülebilmektedir.


\textbf{Sonuç}: Her ne kadar Patellar OO nadir gözükse de çocuk ve genç erişkinlerde uzun süreli diz ağrısı ayrıntılı tamsızında düşünülmesi gerekir.

\textbf{Anahtar Kelimeler}: Osteoid Osteoma, patella, diz ağrısı, küretaj, geçici tanı, kemik neoplasması

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<td>Burak Kaymaz, M.D.</td>
<td><a href="mailto:kaymaz23@yahoo.com">kaymaz23@yahoo.com</a></td>
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\textbf{Correspondence}: Dr. Burak Kaymaz, Hamidiye Mah. Rauf Denktaş Cad. Konak kale Sitesi B2-23 Kepez/Canakkale-Turkey

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Key Points
1. Anterior knee pain in children is not always a benign condition.
2. Persistent knee pain in children requires further investigation like CT or MR imaging.
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Introduction
Osteoid osteoma (OO) is a small, benign bone neoplasm that has a well demarcated nidus surrounded by a reactive zone of sclerosis. It is most distributed in the long bones, such as femur and tibia [1]. Osteoid osteoma accounts for 13% of all benign bone tumors and is more frequently found in patients between 10–30 years of age with male predominancy [2]. Most patients suffer from pain with gradually increasing severity, and the pain often worsens at night [3]. Patella is a rare location of osteoid osteoma and the diagnosis of osteoid osteoma of the patella is critical for patient management. However, the diagnosis of osteoid osteoma of the patella is a challenging problem because patients often present with anterior knee pain, which is one of the most common musculoskeletal system symptoms [4,5]. Some patients may be examined at the sports medicine clinics and may not be diagnosed, even after arthroscopy, or some may be misdiagnosed as osteochondritis dissecans or meniscal pathologies [6,7]. The primary treatment of osteoid osteoma is surgical removal of the nidus and some of the surrounding bone after precise localization of the nidus. Open curettage and excision of the nidus or percutaneous nidus excision by CT guided radiofrequency ablation could be applied if technical opportunities are sufficient. Surgical excision provides relief from the pain immediately. When excision is made completely and the lesion is removed by peripheral bone tissue, the risk of recurrence is also decreased [8]. In this article, we reported a patellar osteoid osteoma case with delayed diagnosis. We used the direct curettage technique, and the patient was free of pain the day after the surgery.

Case Presentation
An 8-year-old girl was referred to the Orthopedics outpatient clinic from the Pediatric outpatient clinic in our hospital. The patient suffered from left knee pain for 11-12 months. The pain was severe and was getting worse at night. Previously, the patient was advised to take non-steroidal anti-inflammatory drugs and was told to rest, but the exact pathology could not be diagnosed, and the patient was not free of pain after the NSAID medications. When the patient admitted to our clinic, physical examination revealed slight atrophy of the left thigh. The range of motion of the left knee joint was within normal range. There was slight tenderness over the left patella. Laboratory tests were normal. The A-P and lateral X-ray showed no abnormality, but MR images showed a lesion in the upper 1/2 of the left patella that was suspicious for osteoid osteoma (Figure 1). A computed tomography (CT) scan showed the exact lesion with nidus that was located in the medullary cavity of the patella near the articular surface. (Figure 2). In consideration of the patient's symptoms and radiographs, the lesion was thought to be an osteoid osteoma. The lesion was decided to be curetted through the anterior open technique. Approximately 5 cm anterior incision was made to reveal the anterior surface of the patella. After grinding off the cortical bone, the center of the lesion was found and, under fluoroscopic control not to damage the articular surface, the curettage of the nidus together with reactive bone was performed (Figure 3). The patient was free of pain and mobilized the day after surgery. In the first month of control, there was no complaint from the patient. Direct radiography showed full ossification of the curedtted region of the patella (Figure 4).

Figure 1. Sagittal X-ray (a), sagittal MR image (b) and axial MR image (c) of the patient
Figure 2. Sagittal (a) and axial (b) CT images of the patient show the pathognomonic nidus located in a subchondral location.

Figure 3. Intraoperative photograph (a) showing the anterior open surgery and fluoroscopic image (b) showing the curettage of the lesion.
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Discussion

The diagnosis of patellar osteoid osteoma is exceedingly rare, and the diagnosis may be challenging, resulting in the delay of treatment. Because many other diseases of the patella are more commonly seen than osteoid osteoma of the patella, it is important to be aware of this entity. The clinical symptoms can mimic other more common diseases, such as chondromalacia patellae, osteochondritis dissecans, meniscal tears, osteomyelitis, focal abscess, osteoarthritis, and cortical stress fracture [9]. Therefore, a thorough patient history and careful examination of the radiographs are important to make a correct diagnosis. [10]. The imaging features of the intra-articular OO differ from those of the extra-articular OO. Direct radiography is not usually diagnostic in 80% of the cases since no nidus or periosteal reaction can be seen at the time that symptoms begin [11-13]. Excessive periosteal reaction occurs in typical intracortical localized osteoid osteoma, but in intraarticular or medullar osteoid osteoma, minimal or no periosteal reaction is seen [14]. In some cases, juxta articular osteopenia or osteoarthritic changes may be seen [15,16]. So that the conventional radiological diagnosis of osteoid osteoma may be difficult. If the X-ray findings are not diagnostic in young patients with typical knee pain, a CT or MRI scan may be necessary to confirm the diagnosis. However, it should be kept in mind that the scans still require a careful examination because the nidus may be too small to be noticed initially. [11,15]. It is reported in the literature that approximately 21% of intra-articular niduses cannot be identified and another 29% are poorly identified on initial MRI. CT scans are still considered the gold standard for diagnosing OO in both adults and children [16,17]. In our case, X-rays were also non-diagnostic so that first MRI and then CT images were required, and CT images were more accurate than MRI in the detection of the OO, showing the characteristic nidus. Newer techniques have shown promising results in the imaging of intra-articular OO. Single photon emission computed tomography and MRI gadolinium enhanced imaging have also been proven to be efficient in the detection of the nidus in intra-articular lesions [18]. Especially around the knee joint, the diagnosis of osteoid osteoma can be delayed for many months [11]. The average delay for diagnosis of intra-articular OO has been reported by Szendroi et al. at 26.6 months and by Rolvien et al. at 20.7 months [13,18]. In our case, the time interval between the onset of symptoms and the diagnosis was approximately one year.

High prostaglandin levels produced within the nidus are the cause of the pain in OO [20]. Transmission of these prostaglandins from the nidus to the synovium causes lymphofollicular synovitis, resembling rheumatoid arthritis histologically and monoarthritis of infectious, degenerative, or rheumatologic diseases clinically [19]. In some cases, OO relief with salicylates or NSAIDs may be seen, and the patient may be free of nocturnal pain. The diffuse pain due to synovitis and the lesion itself, accompanied by non-specific symptoms such as limited range of motion, gait and postural disturbances, joint effusion, and muscle atrophy around the joint, may be misleading for the clinician [15].

Surgical treatment is needed after the diagnosis of osteoid osteoma. Different surgical procedures may be appropriate, including open surgery and excisional biopsy, percutaneous resection with a cannulated skin-punch biopsy needle [21], or resection under CT and arthroscopic guidance [22], and rarely patellectomy. An excisional biopsy may result in patella baja, as described by Vallianatos et al [7]. Other minimally invasive procedures require precise localization of the nidus; otherwise, a small lesion may be missed. Patellectomy may affect the strength of the patellar tendon and result in a limited range of motion when the knee is extended.

Figure 4. Postoperative one-month radiography showing the fully ossified, curetted region.
Limitations
As a limitation, we should mention that the radiological image of the patient is not of high quality, so that the diagnosis of the patient is getting harder. Also, we do not have any idea about the long-term results of the treatment or about the risk of recurrence.

Conclusion
Although a rare entity, patellar OO should also be included in the differential diagnosis of persistent knee pain in children and young adults. Especially in adolescents presenting with anterior knee pain or long-standing non-specific complaints around the knee joint with non-specific MRI findings, a CT scan may be a reasonable step in the diagnostic procedure. High clinical suspicion is necessary to avoid delay in diagnosis and treatment of this rare lesion.

Patient’s consent: Yes
Conflict of interest: The authors declare that they have no conflict of interest.

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References