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Intra- and juxta-articular osteoid osteoma: a diagnostic challenge

Misdiagnosis and successful treatment: a report of four cases

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Abstract Osteoid osteoma is a benign tumour in young adults. The clinical manifestations are generally typical nocturnal pain that prevents sleep and that is alleviated with aspirin. When the typical clinical and radiological features are present, diagnosis is not difficult. Problems in the differential diagnosis may arise in connection with an unusual location. We report on the clinical features, radiographic and histopathological findings, treatment, and results of four patients who were managed for an intra- or juxta-articular osteoid osteoma at our medical centre between 2000 and 2002 and in whom the initial diagnosis was erroneous and delayed from 1 to 10 years. In order to remove these lesions, we performed a CT-guided en block retrograde resection under arthroscopic control for juxta-articular osteoid osteomas (knee) and an arthroscopy-assisted en block antegrade resection in two cases of intra-articular osteoid osteoma (elbow and shoulder). None of the osteoid osteomas recurred in the follow-up period, and each patient got relief from pain.

Keywords Intra-articular · Juxta-articular · Osteoid osteoma · Arthroscopy · En block resection

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Introduction

Osteoid osteoma, first described by Jaffe in 1935 [12], is a benign tumour‘ in young adults. It occurs in the second and third decades of life and the lesion is about 2.3 times more common in men [10, 22]. It is located preferentially in the shaft of the long bones near the metaphyseal junctions, with a predilection for the lower limbs [24].

About a quarter of osteoid osteomas are not detected on plain radiographs alone. In such cases, CT, bone scintigraphy, magnetic resonance imaging, and angiography are useful to make an early and correct diagnosis. A double-ring sign on scintigraphy is a characteristic of osteoid osteoma. MRI frequently does not provide a correct diagnosis and is not as accurate as CT that remains the technique of choice for identifying the extent of the nidus [2, 6, 9, 11, 17, 27, 28].

The clinical manifestations are generally typical nocturnal pain that prevents sleep and that is alleviated with aspirin. Conventional radiographs and CT scans generally suffice to establish the diagnosis, which is already strongly suggested by the clinical picture [2]. When the typical clinical and radiological features are

present, diagnosis is not difficult [18, 21]. Problems in the differential diagnosis may arise in connection with an unusual location [24], since it causes atypical pain [14, 23]. Clinically, juxta- and intra-articular osteoid osteomas may resemble more common entities such as traumatic or degenerative pathologies of the joint, with delay in the diagnosis [3, 5, 7, 26].

Delay in the diagnosis is therefore responsible for muscle atrophies, tenderness, localized swelling or contractures. Therefore, joint pain, which is non-responsive to conventional treatment, needs a thorough persistent diagnostic approach, taking into consideration a number of syndromes and pathologies [8].

The histopathological criteria for the diagnosis of an osteoid osteoma have been described previously in detail. These lesions typically consist of a small yellowish to red pea nidus of osteoid and woven bone with inter-connected trabeculae, and a background and rim of highly vascularized fibrous connective tissue. Variable-degree sclerotic bone reaction may surround the lesion [9, 13]. The lesions in our patients had a histopathological characteristic of osteoid osteoma.

The intra- or juxta-articular osteoid osteoma can lead the clinician to misdiagnosis. In fact depended on the localization, many initial presumptive diagnoses must be considered. We report on the clinical features, radiographic and histopathological findings, treatment, and results of four patients who were managed for an intra- or juxta-articular osteoid osteoma at our medical centre between 2000 and 2002 and in whom the initial diagnosis was erroneous and delayed from 1 to 10 years.

Materials and methods

Between 2000 and 2002, four patients were diagnosed with and managed for an articular or juxta-articular osteoid osteoma at our medical centre. Data on age, gender, symptoms, location of the lesion, treatment regimen, and clinical course were compiled for each patient from the medical records. We also reviewed the radiographic and histopathological features of the lesions.

Results

Radiographic appearance

Case 1: The knee radiographs performed at our hospital revealed the presence of a radiolucent nidus with a sclerotic rim in the posterior area of the lateral tibial plateau. MRI and CT scan (Fig. 1) localized a subchondral lesion under the posterior horn of the lateral meniscus showing a maximum horizontal diameter of 8 mm and a height of 6 mm.

Case 2: CT scan (Fig. 2a) and MRI (Fig. 2b) of the knee revealed a sub-chondral lesion (localized 1 mm under the articular cartilage and showing a maximum horizontal diameter of 8 mm and an height of 9–10 mm) with perinidal sclerosis in the central region of the patella. Bone scan showed increased uptake corresponding at the lesion of the patella.

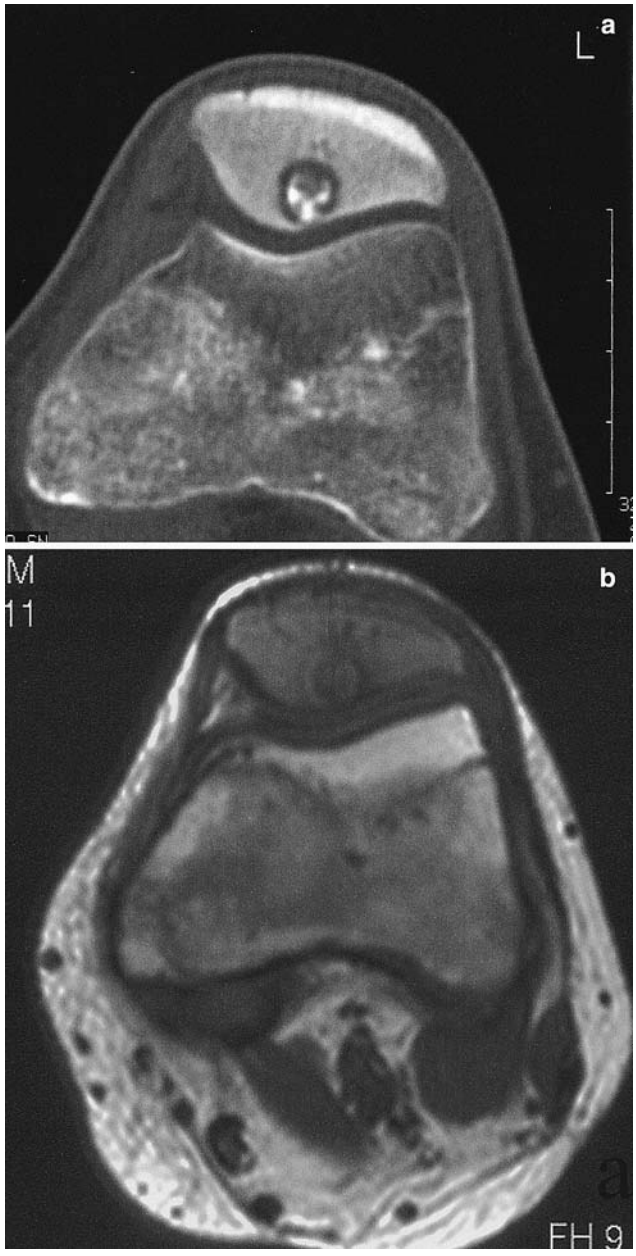

Fig. 1 Axial spiral CT scan (**a**) and axial T1-weighted MRI (**b**) from Patient 2 depicted a sub-chondral lesion with perinidal sclerosis in the central region of the patella. CT (**a**) showed a low attenuation ring and a calcified nidus

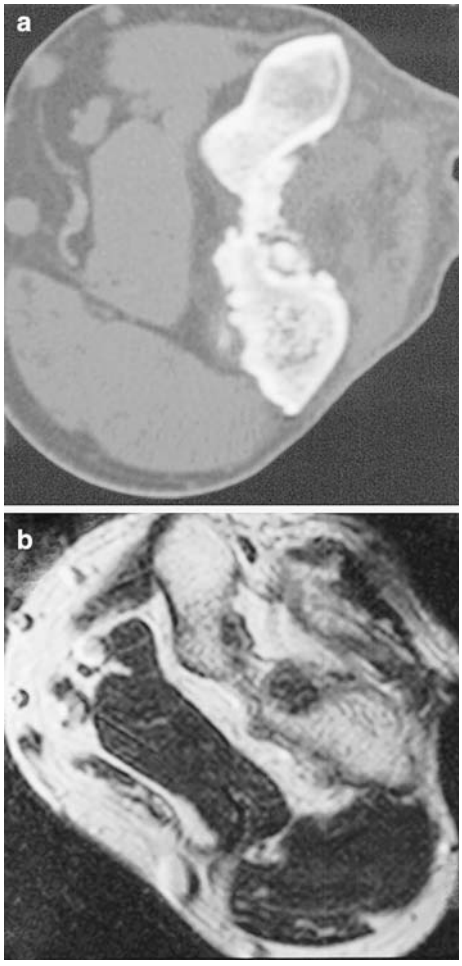

Fig. 2 Axial spiral CT (**a**), and axial MRI GET2-weighted (**b**) showed a lesion with a central nidus and perinidal sclerosis in the olecranic fossa of the distal epiphysis of the humerus (case 3)

Case 3: CT (Fig. 3a) and MRI (Fig. 3b) showed a lesion with a central nidus and perinidal sclerosis in the olecranic fossa of the distal epiphysis of the humerus.

Case 4: At scintigraphy, there appeared a little area of captation on the proximal humerus. CT (Fig. 4) showed a lesion measuring 8 mm at the head of the humerus, with a resorption of bone.

Follow-up

The duration of follow-up, defined as the time from the operation to that of the most recent clinical evaluation, ranged from 35 to 48 months (mean, 41.5 months) (Table 1).

Age and gender

The patients' ages ranged from 16 to 44 years, with a mean of 32.5 years (Table 1). There were two female and two male patients.

Fig. 3 Coronal MPR CT image showed a lesion (*arrow*) with perinidal sclerosis, consistent with an osteoid osteoma, in the head of the humerus (case 4)

Fig. 4 A Kirschner guide wire was percutaneously inserted

Symptoms and laboratory examinations

At the time of the initial presentation, all the patients had light pain that became worse at night and the administration of aspirin typically helped to alleviate the pain. Furthermore, all the patients had a limitation of the R.O.M. Results of laboratory examinations (biochemical and serologic tests) were normal for each patient, with no signs of inflammation or increased bone metabolism.

Location of the lesion

Case 1. Posterior area of the lateral tibial plateau.

Case 2. Central region of the patella.

Table 1 Case summary of the osteoid osteomas presented in this paper

Case	Gender and age (years)	History and symptoms	Duration of symptoms (years)	Site of lesion	Radiographic and other imaging findings	Pre-operative misdiagnosis	Duration of follow-up (months)	Hospital stay (days)	Clinical course
1	M 28	Persistent anterior knee pain, worse at night. The year before the patient had a negative arthroscopic diagnosis for -articular joint lesions	2	Posterior area of the lateral tibial plateau	Axial spiral CT scan showed the nidus with marked cortical sclerosis thickening localized in the posterolateral sub-chondral area of the tibial plateau (Fig. 1)	Meniscal tears	48	3	CT-guided en block retrograde resection under arthroscopic control of osteoid osteoma and filling of the bony defect with an autologus bone graft harvested from the proximal tibial metaphysis; patient returned to full activity and full-time employment
2	F 16	Persistent anterior left knee pain The pain was associated with effusion and swelling	1	Central region of the patella	No malalignment at axial XR view. CT scan (Fig. 2a) and MRI (Fig. 2b) from case 2 depicted a sub-chondral lesion with perinidal sclerosis in the central region of the patella	Femoro-patellar dysplasia	35	2	CT-guided en block retrograde resection under arthroscopic control for osteoid osteoma and filling of the bony defect with an autologus bone graft harvested from the proximal tibial metaphysis; patient subsequently relieved from pain
3	M 42	Elbow pain worse at night	10	Olecranic fossa of the distal epiphysis of the humerus	CT (Fig. 3a) and MRI (Fig. 3b) showed a lesion with a central nidus and perinidal sclerosis in the olecranic fossa of the distal epiphysis of the humerus	Post-traumatic periosteitis	46	1	En block antegrade resection arthroscopy; patient returned to full activity and full-time employment
4	F 44	Shoulder pain worse at night	3	Head of the humerus	MRI showed a lesion measuring 8 mm at the head of the humerus, with a halo of resorbed bone surrounded by marrow oedema	Rotator cuff tear	37	2	Arthroscopy-assisted en block antegrade resection and filling of the bony defect with heterologous bone. Patient subsequently relieved from pain

Case 3. Olecranic fossa of the distal epiphysis of the humerus.

Case 4. Head of the humerus.

Misdiagnosis in other hospitals

Case 1: Two years before the patient had a negative arthroscopic diagnosis for suspicion of meniscal lesion; however, at his follow-up visit, 6 months later, he still complained of knee pain.

Case 2: The year before the patient was treated by isometric rehabilitation of the quadriceps for a suspected femoro-patellar dysplasia.

Case 3: The patients was treated for 10 years for a post-traumatic periosteitis.

Case 4: A continuous night pain was attributed for 3 years to a rotator cuff pathology.

Pre-operative diagnosis, treatment, and outcome

The diagnosis of osteoid osteoma had not been established pre-operatively in one patient (case 3). However, for all the patients, a diagnosis of osteoid osteoma was considered in the differential diagnosis. Two of these patients (cases 1 and 2) had a CT-guided wide en block retrograde resection under arthroscopy. In the other two (cases 3 and 4) we performed an arthroscope-assisted surgical technique of en block antegrade resection. The bony defect was filled with an autologus bone graft harvested from the proximal tibial metaphysis in the cases 1 and 2, while in the cases 3 and 4 we used heterologous bone graft. After the surgery all the patients had a complete relief of pain.

Illustrative case report (case 2)

In July 2002 a 16-year-old female professional skater, came to us complaining of a persistent anterior left knee pain for about 1 year. The onset of the symptoms was light pain that became worse at night associated with knee effusion and swelling that limited the patient's daily activities and exercises. The administration of aspirin typically helped to alleviate the pain. A year before the patient visited another hospital for a suspected femoro-patellar dysplasia. At the hospital X-rays in standard and axial projections were taken as well as an MRI of the knee. The MRI examination revealed a widespread alteration of the signal in T1 localized in the sub-chondral bone. The medical report attributed the image to sub-chondral oedema secondary to contusive trauma. On the basis of the MRI the orthopaedist ordered the patient to stop agonistic sport exercises and to start medical therapy with light drugs for inflammation and isometric rehabilitation of the quadriceps. The patient, however, at her follow-up visit, 6 months later, still complained of knee pain associated with functional restriction.

When the young woman skater was presented in our hospital she limped and had a limited R.O.M. as well as swelling and pain when the Hoffa fat pad was palpated. Laxity and meniscus tests were negative, but femoropatellar signs were positive. Laboratory examinations were within the normal range. Radiographs, CT (Fig. 2a), and MRI (Fig. 2b) of the knee revealed the presence of a nidus with a sclerotic rim in the central region of the patella, localized 1 mm under the articular cartilage and showing a maximum horizontal diameter of 8 mm and an height of 9–10 mm. Bone scan showed increased uptake corresponding to the lesion of the patella.

Surgical procedure

The patient was administered a loco-regional anaesthesia and a single intra-venous dose of a fourth generation cephalosporin and was transported to the CT radiology room. Sections with a thickness of 1–3 mm were obtained in order to determine the precise localization of the nidus. With a 20-gauge needle in place a single CT cut confirmed the correct approach. A skin incision was made at the puncture site and access to the nidus was established using an 11-gauge Jamshidi hollow biopsy needle and a 2 mm coaxial drill system, depending on the hardness of the adjacent bone. Additional CT images located the position of the inserted instrument with the patient lying supine. A kirschner guide wire was percutaneously inserted (Fig. 5) under CT control entering perpendicular to frontal plan corresponding the central zone of the patella. The following axial (Fig. 6) and coronal views of CT showed the central placement of the Kirschner wire until the distal edge of the lesion after three attempts. We measured the intra-osseous length (20 mm) of the Kirschner wire (starting from the external cortex till the distal edge of the lesion) comparing a new Kirschner wire with the first one. Following this procedure the patient was transferred to surgery in order to surgically remove the lesion. An arthroscopic examination of the knee was carried out showing normal conditions of the articular cartilage of the patello-femoral joint and the remaining structures. A vertical 1.5-cm surgical incision in correspondence of the entry of the Kirschner wire was made and using an elevator the cortical bone was exposed. A 1-cm diameter hollow drill bit was then used on the guidance of the Kirschner wire (Fig. 6) to remove a bone block of calculated depth and dimensions (about

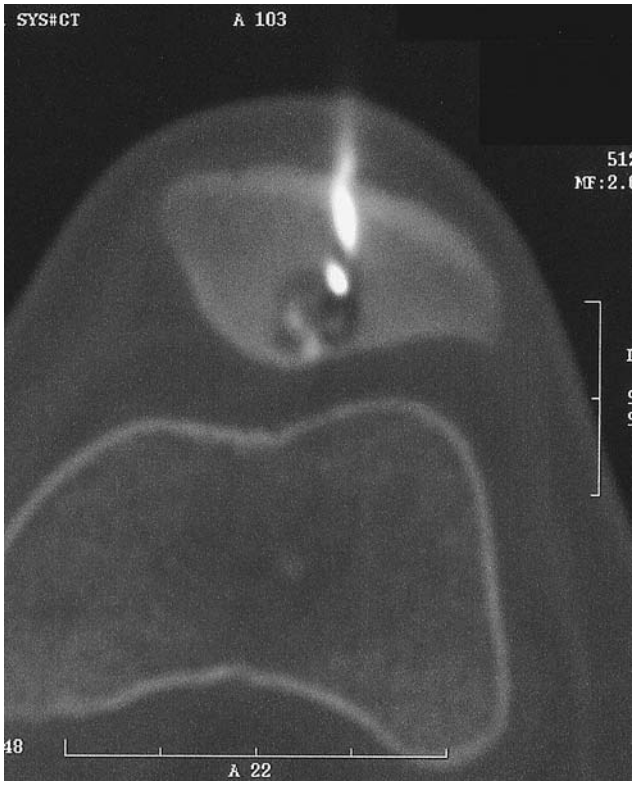

Fig. 5 Axial view CT control showed the central placement of the Kirschner wire into the lesion

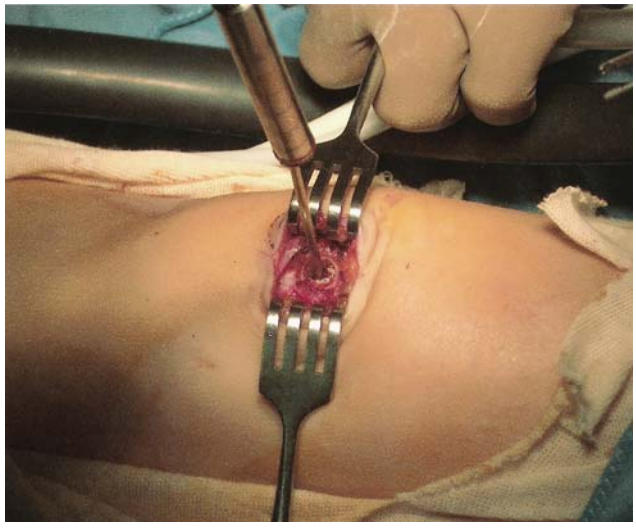

Fig. 6 A 1-cm diameter hollow drill bit was used on the guidance of the Kirschner wire to remove a bone block

18 mm^3). Curettage of the remaining defect was performed with the arthroscope and a small retrograde curette into the tunnel. The gross bone specimen removed revealed the incorporated lesion and the patella articular cartilage integrity was then demonstrated arthroscopically. In order to fill the bony defect an autologus bone graft was harvested from the proximal tibial metaphysis using an osteochondral tube harvester, 1 mm wider (11 mm) than the size of the defect. The bone plug was press-fitted directly into the defect. Post-operative X-rays in a-p and lateral views showed the correct positioning of the graft and no fractures of the patella. The removed gross bone specimen confirmed the pre-supposed osteoid osteoma diagnosis.

Discussion

In all patients of our series the initial diagnosis was erroneous and delayed for on average 4 years (range 1–10 years). Initial plain radiographs were not diagnostic in all patients. In accordance with Georgoulis et al. [8] we found that the radiographic findings may not diagnose an osteoid osteoma when the lesion is so small that it is not detectable on plain radiographs. As pointed out by Kransdorf et al. [19], when the radiographic changes occur late, there may be a long delay in the definite diagnosis.

Furthermore, when the symptoms precede the radiological findings it is always necessary to perform further investigation with bone scintigraphy, CT, and MRI. Although MRI is not much helpful in diagnosing osteoid osteoma, in our cases it was useful, particularly in showing the inflammatory reaction produced by osteoid osteoma and in excluding other associated pathologies.

In cases 1 and 2 knee pain was misdiagnosed, respectively, as a meniscal tear and a femoro-patellar dysplasia, but no history of trauma and no patellar malalignment were present in these cases.

As other authors reported [8], in case 1 the patient underwent a diagnostic arthroscopy without any resolution of symptoms before correctly diagnosed as osteoid osteoma.

In case 3 the first diagnosis was post-traumatic periosteitis. Osteoid osteomas are rare in the elbow and cause joint dysfunction that can simulate other conditions [20]. Our patient who had the lesion in the distal aspect of the humerus had a loss of flexion and extension, as described by many authors [20]. Other findings such as swelling and weakness were non-specific, demonstrating that physical examination alone frequently fails to accurately localize the lesion. The differential diagnosis in this case included chronic inflammatory arthritis, osteochondritis dissecans, tendonitis, rheumatoid synovitis, epicondilytis, golfer elbow, loose bodies, olecranon osteophytosis associated to olecranon fossa impingement, osteoarthritis, osteomyelitis. A history of an injury can make the diagnosis of osteoid osteoma even more difficult, particularly when the symptoms

occur soon after an injury. The correlation between injury and the onset of osteoid osteoma has been discussed, but remains unclear and difficult to elucidate [1, 16]. In our case an injury precedes the onset of the lesion. The trauma leads to the low index of suspicion and value of the osteoid osteoma diagnosis. Clinically, the patient complained of elbow pain during the day and at night. Diurnal pain can be due to a synovitis of the humeroulnar compartment. The pathogenesis of the synovitis seen in intra-articular osteoid osteoma is not clear. Synovitis slowly leads to cartilage destruction, which causes a definitive osteoarthritis [4, 25]. Owing to the neurovascular structures retrograde removal of the lesion was impossible and the antegrade approach was mandatory.

In case 4 the woman complained of night shoulder pain. For 3 years the night pain was attributed to rotator cuff pathology. An MRI enabled us to diagnose an osteoid osteoma surrounded by bone marrow oedema. Night pain may be often attributed to rotator cuff pathology but this pathology is less likely on young patients. In fact osteoid osteoma is more frequent in adolescence, whereas degenerative cuff pathology occurs more often in elderly patients [15]. Retrograde removal was impossible to perform for the angle of the approach that could cause a great tuberosity fracture.

In order to remove these lesions we performed a CT-guided en block antegrade and retrograde resection under arthroscopic control. These techniques allow minimal and generally complete bone resection owing to precise and reliable localization. The identification of the nidus during the surgical procedure may be difficult; therefore, less invasive, percutaneous proceedings under CT guidance have been developed. Moreover, CT-guided percutaneous drilling of the guide wire may present some problems, like infection of the surgical wound and the necessity of performing the procedure in two steps in a not-sterile condition of the radiology room.

However, complete resection of the nidus remains decisive and in our cases the arthroscopic technique was necessary to detect and remove all the remaining bone tumour particles. Under arthroscopic control it is even possible to work in a small room (a 10-mm tunnel) like we did in cases 1 and 2 and it is possible to avoid destroying open approaches in order to reach hidden lesions that are difficult to reach by open surgery. Using this procedure the hospital stay was reduced to 1–3 days and the patients returned quicly to the normal daily activities.

In conclusion, juxta- or intra-articular osteoid osteoma can simulate several other articular pathologies with different clinical presentations. Patients complaining of a persistent joint pain that increases during the night must be considered for more accurate examination and diagnosis in order to avoid unnecessary surgical procedures.

References

1. Ambrosia JM, Kernek CB (1985) Osteoid osteoma of the foot: presentation following trauma. Orthopedics 8:684–685
2. Assoun J, Richardi G, Railhac JJ, Baunin C, Fajadet P, Giron J, Maquin P, Haddad J, Bonnevialle P (1994) Osteoid osteoma: MR imaging versus CT. Radiology 191:217–223
3. Corbett JM, Wilde AH, McCormack LJ, Evarts CM (1974) Intra-articular osteoid osteoma; a diagnostic problem. Clin Orthop Relat Res 98:225–230
4. Cronemeyer RL, Kirchmer NA, De Smet AA, Neff JR (1981) Intra-articular osteoid-osteoma of the humerus simulating synovitis of the elbow. A case report. J Bone Joint Surg Am 63:1172–1174
5. Franceschi F, Marinozzi A, Rizzello G, Papalia R, Rojas M, Denaro V (2005) Computed tomography-guided and arthroscopically controlled en bloc retrograde resection of a juxta-articular osteoid osteoma of the tibial plateau. Arthroscopy 21:351–359
6. Frassica FJ, Waltrip RL, Sponseller PD, Ma LD, McCarthy EF Jr (1996) Clinicopathologic features and treatment of osteoid osteoma and osteoblastoma in children and adolescents. Orthop Clin North Am 27:559–574
7. Freyschmidt I Ostertag H (1988) Knochenbildende Tumoren. In: Freyschmidt I, Ostertag H (eds) Knochentumoren. Springer, Berlin Heidelberg New York, pp 97–106
8. Georgoulis AD, Papageorgiou CD, Moebius UG, Rossis J, Papadonikolakis A, Soucacos PN (2002) The diagnostic dilemma created by osteoid osteoma that presents as knee pain. Arthroscopy 18:32–37
9. Goldman AB, Schneider R, Pavlov H (1993) Osteoid osteomas of the femoral neck: report of four cases evaluated with isotopic bone scanning, CT, and MR imaging. Radiology 186:227–232
10. Healey JH, Ghelman B (1986) Osteoid osteoma: current concepts and recent advances. Clin Orthop 204:76–85
11. Helms CA, Hattner RS, Vogler JB 3rd (1984) Osteoid osteoma: radionuclide diagnosis. Radiology 151:779–784
12. Jaffe HL (1935) Osteoid osteoma of bone. Radiology 45:319
13. Jaffe Hl (1953) Osteoid-osteoma. Proc R Soc Med 46:1007–1012
14. Joyce MJ, Mankin HJ (1983) Caveat arthroscopos: extra-articular lesions of bone simulating intra-articular pathology of the knee. J Bone Joint Surg Am 65:289–292
15. Kaempffe FA (1994) Osteoid osteoma of the coracoid process. Excision by posterior approach. A case report. Clin Orthop Relat Res 301:260–262
16. Kendrick JI, Evarts CM (1967) Osteoid-osteoma: a critical analysis of 40 tumors. Clin Orthop Relat Res 54:51–59
17. Khurana JS, Mayo-Smith W, Kattapuram SV (1990) Subtalar arthralgia caused by juxtaarticular osteoid osteoma. Clin Orthop Relat Res 252:205–208
18. Kiers L, Shield LK, Cole WG (1990) Neurological manifestations of osteoid osteoma. Arch Dis Child 65:851–855
19. Kransdorf MJ, Stull MA, Gilkey FW, Moser RP Jr (1991) Osteoid osteoma. Radiographics 11:671–696
20. Marcove RC, Freiberger RH (1966) Osteoid osteoma of the elbow–a diagnostic problem. Report of four cases. J Bone Joint Surg Am 48:1185–1190

21. Moberg E (1951) The natural course of osteoid osteoma. J Bone Joint Surg Am 33:166–170
22. Pikoulas C, Mantzikopoulos G, Thanos L, Passomenos D, Dalamarinis C, Glampedaki-Dagianta K (1995) Unusually located osteoid osteomas. Eur J Radiol 20:120–125
23. Roberts P, Davies AM, Starkie CM, Grimer RJ (1990) The nidus of an osteoid osteoma mimicking an os supratrochleare dorsale. Br J Radiol 63:899–902
24. Sim FH, Dahlin DC, Beabout JW (1975) Osteoid-osteoma: diagnostic problems. J Bone Joint Surg Am 57:154–159
25. Snarr JW, Abell MR, Martel W (1973) Lymphofollicular synovitis with osteoid osteoma. Radiology 106:557–560
26. Torg JS, Loughran T, Pavlov H, Schwamm H, Gregg J, Sherman M, Balduini FC (1985) Osteoid osteoma. Distant, periarticular, and subarticular lesions as a cause of knee pain. Sports Med 2:296–304
27. Yamamura S, Sato K, Sugiura H, Asano M, Takahashi M, Iwata H (1994) Magnetic resonance imaging of inflammatory reaction in osteoid osteoma. Arch Orthop Trauma Surg 114:8–13
28. Winter PF, Johnson PM, Hilal SK, Feldman F (1977) Scintigraphic detection of osteoid osteoma. Radiology 122:177–178