Periosteal osteoid osteoma of the distal femur

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Abstract

Periosteal osteoid osteoma is extremely rare. The diagnosis is not always clear. We report a case of periosteal osteoid osteoma arising from the posterior surface of the right distal femur in a 21-year-old woman. After careful evaluation and excisional biopsy, histological examination confirmed the diagnosis of osteoid osteoma, showing the nidus, surrounding osteosclerosis, and catarrhal synovitis. The lesion was treated successfully with excision of the nidus.

Introduction

Osteoid osteoma (OO) is a benign osteoblastic tumor found predominantly in males between the ages of 10 and 25 years. Usually, OO is located in the medullary cavity of the flat and long bones, and a periosteal location is very rare. Several cases of periosteal location have been reported in the literature.[1-31] We present a case of periosteal OO located in the posterior aspect of the distal femur.

Case Report

A 21-year-old woman presented with a nine-month history of gradually increasing right intermittent knee pain, exacerbating at night and disappearing with nonsteroidal anti-inflammatory drug intake. The patient reported no history of antecedent trauma to the knee. A physical examination showed some mild swelling and tenderness over the posterior aspect of the right distal thigh, but no mass was palpated. The range of motion of the right knee was normal with 130° of flexion and 0° of extension. Biological examinations showed: CRP <4 mg/L, and normal white blood cell count. Infectious and immunological tests were negative. An anteroposterior and a 3/4 radiograph of the knee showed a radiolucent nidus of 4 mm surrounded by a fine zone of sclerosis. A lateral radiograph showed a lesion with increased soft tissue density adjacent to the posterior surface of the distal femur (Figure 1). Computed tomography (CT) scans revealed a subperiosteal mass, with some ossification, in the posterior aspect of the medial femoral supracondylar area. The lesion was attached to the underlying cortical bone, with erosive changes of the bone. There was no medullary involvement of the lesion (Figure 2). An excisional biopsy of the lesion was decided on. The lesion was approached by a posterior incision. The excision was accomplished under radiographic guidance (Figure 3). The histological examination confirmed the diagnosis of OO, showing the nidus, surrounding osteosclerosis, and catarrhal synovitis (Figure 4). The patient's symptoms disappeared immediately after surgery. Two years later, the patient is pain-free and has a good range of motion, and there is no local recurrence of the tumor.

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Key words: periosteal, osteoid osteoma, femur, nidus.

Contributions: all authors have contributed to and read the article, and have given permission for their names to be included as co-authors.

Conflict of interest: the authors report no conflicts of interest.

Received for publication: 4 March 2010.
Accepted for publication: 21 June 2010.

Orthopedic Reviews 2010; 2:e15
doi:10.4081/or.2010.e15

Discussion

Osteoid osteoma is a relatively uncommon osteoblastic lesion of bone that accounts for approximately 10% of all benign bone lesion conditions, is most commonly encountered in children and young adults, and the male-to-female ratio of occurrence is 3:1.[23,25,27,29] OO was distinguished from chronic inflammatory lesions and recognized as a benign bone tumor by Jaffe in 1935, and in 1958 he wrote: "An osteoid osteoma is a small but painful bone lesion, and it is its small size and consistent painfulness that strikingly characterise it clinically."[23] Classically, an initially intermittent pain becomes more constant and more intense as the lesion persists; and this was consistent with the history described by our patient. This evolution of pain associated with enlargement of the lesion has been attributed to the presence of prostaglandins, nerve fibers, and a rich capillary plexus within the nidal and perinidal tissues.[32,33] The high levels of prostaglandins are felt to be the reason that OOs tend to respond well to the use of salicylates and nonsteroidal anti-inflammatory drugs. More than half of OOs occur in the femur and tibia, and the proximal femur is by far the most common location.[2,3,9,22] These same lesions may be extra-articular or intra-articular. The roentgenographic picture constitutes the most valuable diagnostic guide to the lesion; the nidus appears as a relatively radiolucent focus in the affected part of the bone. It is usually, although

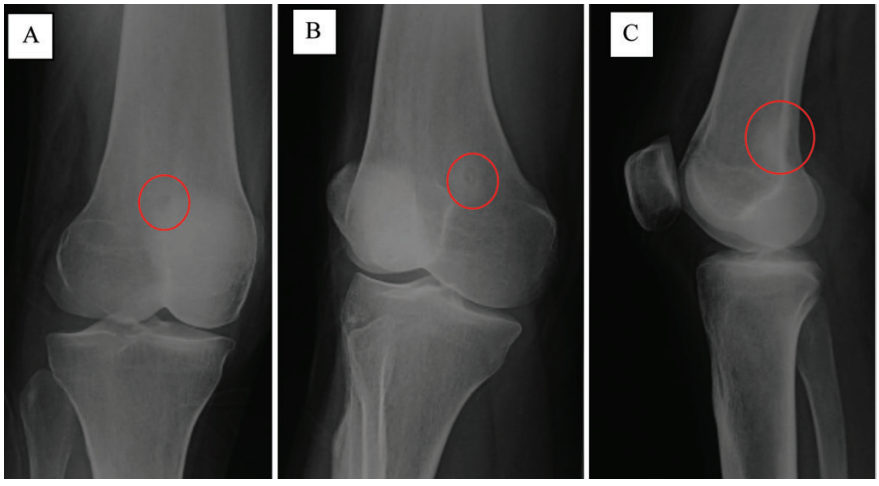

Figure 1. Anteroposterior (A), 3/4 (B), and lateral (C) radiographs of the periosteal osteoid osteoma of the distal femur.

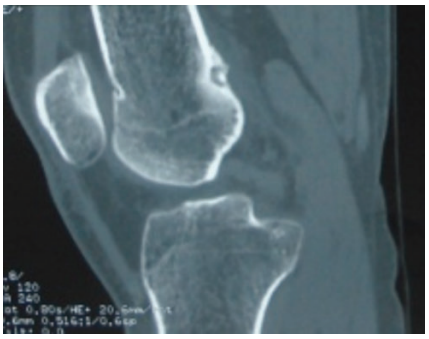

Figure 2. Computed tomography scan of the periosteal osteoid osteoma of the distal femur.

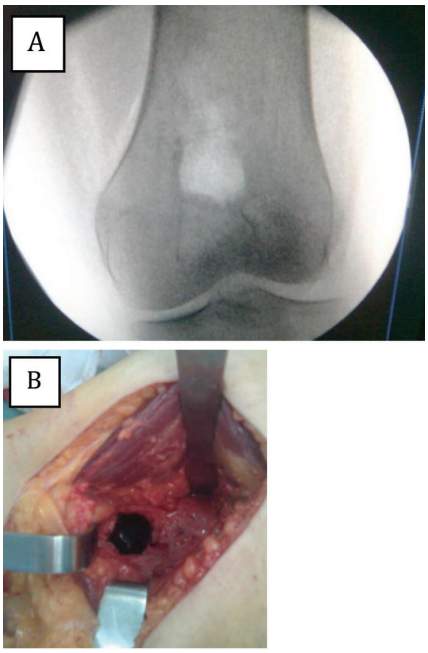

Figure 3. Radiographic preoperative aspect (A) and final aspect after excision (B) of the periosteal osteoid osteoma of the distal femur.

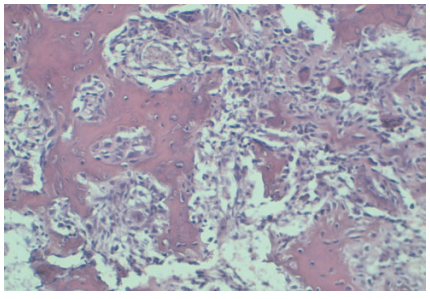

Figure 4. Histological features of the periosteal osteoid osteoma of the femur. The tumor consists of thick, irregularly shaped trabeculae of woven bone, and numerous osteoblast-like cells that fill the intertrabecular spaces. The osteoblast-like cells are devoid of cellular pleomorphism. The stroma contains many dilated small capillaries, loose fibrous connective tissues, and a few multinucleated giant cells and chronic inflammatory cells. (Hematoxylin and eosin stain; magnification: 200X.)

not always surrounded by a shadow reflecting reactive thickening or alteration of neighboring bone. The peculiar behavior of subperiosteal juxta-articular osteomas was first described by Sherman.[1] Edeiken *et al.*,[34] in 1966, distinguish three modes of presentation of OO according to the localization of the nidus in cortical, cancellous, or subperiosteal bone. Their description of the subperiosteal lesions includes: "Subperiosteal osteoid osteomas present as round soft-tissue masses immediately adjacent to bone. As a rule underlying

Table 1. Literature on osteoid osteoma.

Authors	Date	Site	Original diagnoses
Sherman[1]	1947	Elbow Distal radius	Infection
Dahlin and Johnson[2]	1954	Talus	Arthritis
Flaherty *et al.*[3]	1956	Talus Femoral neck	
Davison[4]	1956	Patella	
Cuevillas[5]	1957	Talus	Arthritis
Freiberger *et al.*[6]	1959	Femoral neck Distal fibula	
Guy *et al.*[7]	1959	Femoral neck	
Morton and Bartlett[8]	1966	Distal ulna	Osteomyelitis
Marcove and Freiberger[9]	1966	Elbow (×3)	Rheumatoid arthritis
Shifrin and Reynolds[10]	1971	Elbow	Entrapment of ulnar nerve
Snarr *et al.*[11]	1973	Femoral neck Olecranon fossa Distal humerus Elbow Hip	 Synovitis Ewing's tumor
Corbett *et al.*[12]	1974	Olecranon Femoral condyle	Rheumatoid arthritis Chondromalacia
Simon and Belier[13]	1975	Ankle	
Séruzier *et al.*[14]	1976	Olecranon fossa (×2)	Arthritis
Micheli and Jupiter[15]	1978	Distal femoral epiphysis	Chondromalacia
Mitnick *et al.*[16]	1979	Femoral neck	
Dinant and Desser[17]	1981	Talus Femur greater trochanter	Rheumatoid arthritis
Gould[18]	1981	Talus	
Bleifeld[19]	1981	Distal humerus	
Cronemeyer *et al.*[20]	1981	Distal humerus	Osteomyelitis
Apple and Loughlin[21]	1981	Distal tibia	Chronic sprain
Shereffet *et al.*[22]	1983	Foot (×4)	
Brabants *et al.*[23]	1986	Neck of talus (×3) Elbow	Chronic sprain Arthritis
Tudisco and Ippolito[24]	1986	Femur	Meniscus tear
Shankman *et al.*[25]	1997	Tibia (× 5) Talus (×2) Femur (×2) Hand Fibula	
Kayser *et al.*[26]	1998	Tibia (×8) Femur (×6) Fibula (×3) Ulna	
Chandak *et al.*[27]	2006	Humerus	
Weits *et al.*[28]	2006	Acetabulum	Arthritis
Ozturk *et al.*[29]	2008	Great toe	
Khan *et al.*[30]	2009	Talus	
Zampa *et al.*[31]	2009	Tibia (×3) Femur (×2)	Arthritis

bone reveals pressure atrophy or irregular bone resorption. Instead of invoking reactive bone, they affect neighbouring joints, causing synovitis, effusion and even joint deossification suggesting arthritis rather than osteoid osteoma." A cortical erosion may also be observed. The literature on subperiosteal OO is sparse (Table 1); only 81 cases have been reported, and in more than half of these the first diagnosis was wrong. The talus and distal humerus seem to be the most common sites.[23,27,28] Diagnosis usually is based on the typical pattern of pain, with relief by salicylates, and diagnoses especially on local tenderness to palpation. Local swelling may be seen, with concomitant joint effusion, muscle atrophy, and limitation of movement. Radiographic changes in bone are absent or very limited. The absence of bone sclerosis, which is typical of cortical OO, explains this. Local deossification, slight periosteal new bone formation, and joint narrowing may suggest inflammatory arthritis or even osteomyelitis.[1,2,8,16] Although the plain film manifestations may be confusing, three-phase bone scans and CT scans show findings characteristic of OO in all locations and serve to confirm the diagnosis. Magnetic resonance imaging may also show the nidus, often surrounded by extensive bone marrow and soft tissue edema, but OO remains a bone tumor best seen with CT scans owing to its small size and characteristic central mineralization.[26] Definitive treatment of OO focuses on excision of the nidus of the lesion and, as such, is considered rather straightforward. The main problem is that some locations are difficult to access. Thorough excision can provide immediate and permanent relief of symptoms, and this was the case in our patient. If the nidus is not completely removed, recurrence is likely to happen.[29] As with tumors in general, autogenous bone graft should be procured and the donor site wound closed before dissection and exposure of the tumor in question. The patient was symptom free and displayed no clinical or radiographic evidence of recurrence of the lesion after 24 months of postoperative follow-up.

Conclusions

Periosteal OO is a very rare clinical entity. A detailed history with a high index of suspicion can lead to a prompt diagnosis, which must be confirmed with histopathological examination. Adequate surgical excision of the nidus can be curative, and may give rise to complete remission of symptoms without recurrence.

References

1. Sherman MS. Osteoid osteoma: review of the literature and report of thirty cases. J Bone Joint Surg 1947b;29:18-30.
2. Dahlin DC, Johnson EW. Giant osteoid osteoma. J Bone Joint Surg Am 1954;36: 559-72.
3. Flaherty RA, Pugh DC, Dockerty MB. Osteoid osteoma. AJR 1956;76:1041-51.
4. Davison WR. Osteoid osteoma: report of atypical cases. Clin Orthop 1956;7:113-23.
5. Cuevillas AR. Osteoma osteoide juxta articular. Prensa Méd Argent 1957;44: 3385-7.
6. Freiberger RH, Loitman BS, Helpern M, et al. Osteoid osteoma: a report on 80 cases. AJR 1959;82:194-205.
7. Guy R, Fortier M, Boivin JM, et al. Observation d'un cas d'ostéome ostéoide avec pathologie articulaire. Union Med Can 1959;88:816-24.
8. Morton KS, Bartlett LH. Benign osteoblastic change resembling osteoid osteoma: three cases with unusual radiological features. J Bone Joint Surg Br. 1966;48:478-84.
9. Marcove RC, Freiberger RH. Osteoid osteoma of the elbow – a diagnostic problem: report of four cases. J Bone Joint Surg Am 1966;48:185-90.
10. Shifrin LZ, Reynolds WA. Intra-articular osteoid osteoma of the elbow: a case report. Clin Orthop 1971;81:126-9.
11. Snarr JW, Abell MR, Martel W. Lymphofollicular synovitis with osteoid osteoma. Radiology 1973;106:557-60.
12. Corbett JM, Wilde AH, McCormack U, et al. Intra-articular osteoid osteoma: a diagnostic problem. Clin Orthop 1974;98:225-30.
13. Simon WH, Belier ML. Intracapsular epiphyseal osteoid osteoma of the ankle joint: a case report. Clin Orthop 1975;108:200-3.
14. Séruzier E, Simonin JL, Ducastelle C, et al. Ostéome osteoide avec synovite: à propos de deux observations. Rev Rhum Mal Ostéoartic 1976;43:521-6.
15. Micheli LJ, Jupiter J. Osteoid osteoma as a cause of knee pain in the young athlete: a case study. Am J Sports Med 1978;6:199-203.
16. Mitnick JS, Braunstein P, Genieser NB. Osteoid osteoma of the hip: unusual isotopic appearance. AJR 1979;133:322-3.
17. Dinant HJ, Desser EJ. Osteoid osteoom: een ongewone oorzaak van chronische monarthritis bij twee jeugdige patienten. Ned Tijdschr Geneeskd 1981;125:1080-3.
18. Gould N. Articular osteoid osteoma of the talus: a case report. Foot Ankle 1981;1:284-5.
19. Bleifeld C. Osteoid osteoma: articular communication and minimal pain. NY State J Med 1981;81:1371-2.
20. Cronemeyer RL, Kirchmer NA, De Smet AA, et al. Intra-articular osteoid osteoma of the humerus simulating synovitis of the elbow: a case report. J Bone Joint Surg Am 1981;63;1172-4.
21. Apple DF Jr, Loughlin EC Jr. Osteoid osteoma of the ankle in an athlete. Am J Sports Med 1981;9:254-5.
22. Shereff MJ, Cullivan WT, Johnson KA. Osteoid osteoma of the foot. J Bone Joint Surg Am 1983;65:638-41.
23. Brabants K, Geens S, Van Damme B. Subperiosteal juxta-articular osteoid osteoma. J Bone Joint Surg Br 1986;68: 320-4.
24. Tudisco C, Ippolito E. Subperiosteal Osteoid Osteoma: A Case Report. Arch Orthop Trauma Surg 1986;104:398-9.
25. Shankman S, Desai P, Beltran J. Subperiosteal osteoid osteoma: radiographic and pathologic manifestations. Skeletal Radiol 1997;26:457-62.
26. Kayser F, Resnick D, Haghighi P, et al. Evidence of the Subperiosteal Origin of Osteoid Osteomas in Tubular Bones: Analysis by CT and MR Imaging. AJR 1998; 170:609-14.
27. Chandak RM, Phatak SV, Jain L, et al. Multifocal Osteoid Osteoma. A Case Report. Ind J Radiol Imag 2006;16:53-4.
28. Weits T, Van De Werf GJ. Subperiosteal osteoid osteoma of the right acetabulum. JBR–BTR 2006;89:162-3.
29. Öztürk A, Yalçınkaya U, Özkan Y, et al. Subperiosteal Osteoid Osteoma in the Hallux of a Nine-year-old Female. J Foot Ankle Surg 2008;47:579-82.
30. Khan JA, Vaidya S, Devkota P, et al. Subperiosteal Osteoid Osteoma of the Neck of Talus. J Nepal Med Assoc 2009;48: 58-61.
31. Zampa V, Bargellini I, Ortori S, et al. Osteoid osteoma in atypical locations: The added value of dynamic gadolinium-enhanced MR imaging. Eur J Radiol 2009; 71:527-35.
32. Sherman MS, Mcfarland G Jr. Mechanism of pain in osteoid osteomas. South Med J 1965;58:163-6.
33. Schulman L, Dorfman HD. Nerve fibers in osteoid osteoma. J Bone Joint Surg Am 1970;52:1351-6.
34. Edeiken J, DePalma AF, Hodes PJ. Osteoid osteoma: roentgenographic emphasis. Clin Orthop 1966;49:201-6.