

**ATYPICAL FEMUR LOCALIZATIONS OF OSTEIOD OSTEOMA: CASE REPORT AND LITERATURE REVIEW**

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**ABSTRACT**

Osteoid osteoma (OO) is a benign bone-forming tumor, it affects all age groups and especially young adults. Bones of lower extremity are predominantly involved. The proximal femur is involved in 20% of cases. Pain tends to become increasingly severe at night and is relieved by salicylates. Lesions involving the articulating surfaces are commonly associated with synovitis. When the tumor is clinically suspected, X-rays and Computed Tomography (CT) practically confirm the diagnosis but also specify the location of the lesion. Sought treatment for osteoid osteoma of the proximal femur should be simple, accurate, efficient and inexpensive as possible. We report 04 cases of articular and juxta-articular OO of the proximal femur, all treated by open surgery with very good evolution.

**INTRODUCTION**

Osteoid osteoma is a frequent benign bone tumor frequent. It represents 2 to 3% of all bone tumors and 10 to 20% of all benign bone tumors. It is preferentially located on the shaft and metaphysis of long bones, with a preferred location in the proximal femur. This small tumor (<1 cm) is characterized by a specific structure: the nidus. It usually occurs in young adults and is characterized by male predominance. The clinical manifestations are most often typical, made of pain nocturnal, insomnia, calmed by salicylates (aspirin).

Computed tomography is the most specific examination allowing the positive diagnosis.

Complete surgical excision of the nidus usually results in total healing and avoids recurrence.

Treatment has long been based on surgical resection, the modality of which differs from team to team, but with a significant frequency of failures and complications.

The increasing precision of computed tomography and bone scintigraphy has made it possible to treat this lesion with less invasive and more effective interventions. Transcutaneous techniques<sup>[1]</sup> under tomographic control are among the therapeutic advances in recent years.

The aim of this work is to assess the effectiveness of the

different techniques used by analyzing the clinical course, the recovery time and the recovery time activities.

**History**

This lesion was described by BERSTRANG in 1930 then by MILCH in 1934.

In 1935, JAFFE studied<sup>[2]</sup> this osteoblastic tumor which had no origin infectious, he named it osteoid osteoma, admitting its neoplastic nature.

The articular manifestations of osteomas have been known since SHERMAN in 1947.

In 1955, EDEIKEN classified the osteoid osteoma into three types according to the different histological aspects, then named it after its site of origin in bone.

The common type is the cortical osteoid osteoma which is seen in the cortex long bones.

In 1970 JAFFERS, COURTOIS and MAZABRAND provide histological proof of a chronic inflammatory non-specific synovitis in the vicinity of an osteoma of the femoral neck.

**METHODS**

This is a retrospective study of four cases of osteoid osteoma (OO) of femoral atypical localizations, treated

in the orthopedic surgery department in Ibn Sina Hospital in Rabat, Morocco.

All our patients presented clinical and radiological pathognomonic signs: permanent daytime pain, with nocturnal paroxysms, calmed down by taking aspirin.

X-Rays revealed localized osteocondensation.

Histological confirmation of the diagnosis has been successfully attempted in all procedures performed.

The length of hospital stay, as well as the result of the treatment on pain and limping were noted. The occurrence of complications and their influence on the final result were evaluated. Any incomplete disappearance of pain or its reappearance, as well as the need for revision surgery were considered as a therapeutic failure.

Patients with an incomplete clinical or radiological profile and a follow-up less than one year were eliminated from the study.

The minimum acceptable follow-up has been set at one year, as recurrences and failures of treatment reported to date in the literature appeared during the first year after treatment.

Patients must be treated surgically.

A total of four patients met the inclusion criteria.

## RESULTS

In our cases, there was no gender predominance, and none of our patients were followed for a disease.

Age was between 16 and 37 years with an average of 26, 5.

The femoral neck represents the preferred site with a frequency of 75% and the femoral head (25%).

Cortical localization represents 50% of cases and spongy localization in 50%.

As found in our four observations, patients with OO had clinical pathognomonic signs of daytime pain, with nocturnal paroxysms, subsided by taking salicylates and limping.

The clinical examination shows pain on palpation and a slight decrease in joint mobility. The other joints were free.

Radiologically, all of our patients underwent a standard hip X-ray which objectified the OO of the femoral neck by a small lytic image corresponding to the nidus

surrounded by reactive sclerosis in 3 cases and one pure lytic lesion in 1 case.

A patient underwent bone scintigraphy.

Two patients received a CT scan that shows the image of the nidus.

A patient underwent an MRI which showed a hypersignal of the lesion with significant articular effusion. (Fig.1, 2)

Our four patients underwent surgical treatment with a monobloc open excision.

The duration of hospitalization varied between 5 and 7 days with an average of 6.2 days.

All histological results were in favor of osteoid osteoma.

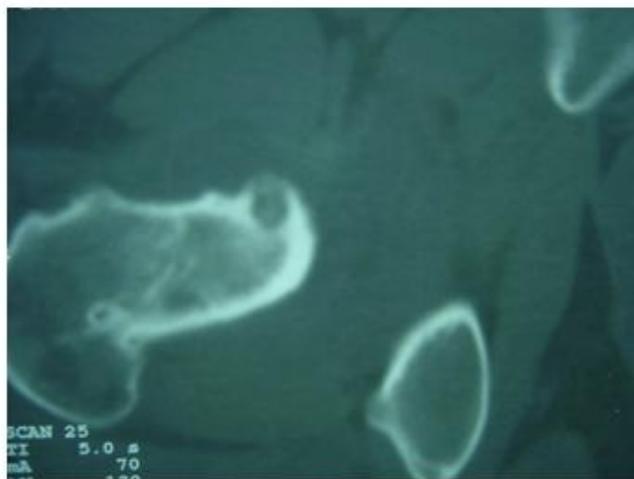
The procedure resulted in immediate and permanent relief (disappearance of the pain in a few days) with return to normal hip mobility area after rehabilitation.

Support was authorized at 45 days.

At three months of the operation, the patients remained asymptomatic with the disappearance of the nidus on the X-Rays.



**Figure 1: X-ray of the hip showing a nidus in the lower part of the femoral neck.**



**Figure 2: CT of the right hip showing a hypodense lesion of the antero-internal and superior part of the neck of the femur.**

## DISCUSSION

Osteoid osteoma is a common benign primary bone tumor. It represents 2 to 3% of all bone tumors and 10 to 20% of all benign bone tumors.<sup>[3][4][5][6]</sup>

It is mainly located in the long bones.<sup>[3][7]</sup> with a predilection for the lower limbs,<sup>[8]</sup> especially the tibia and femur.

Intra-articular localization is rare and its frequency is difficult to assess, approximately 10 to 13% of cases<sup>[3][9][10][11]</sup> It mainly affects the hip as found in our four observations<sup>[9][10][6][12][13][14]</sup> but also the knee.<sup>[11][15]</sup> elbow,<sup>[16][17]</sup> wrist and carpus.<sup>[7][8][18][19]</sup>

Rarely, the spine, flat bones, and skull are interested.

Clinically, pain, found in all of our patients, is constant and typically calmed by salicylates and nonsteroidal anti-inflammatory drugs (NSAID).<sup>[8][21]</sup>

Intra or juxta-articular lesions may present with a synovitis.<sup>[22]</sup> This is the case with one of our patients. When this persists, the patient may have a decrease in range of motion and limping.<sup>[21]</sup>

In children, case of one of our patients, pain and limping are the usual manifestations. The locations close to the epiphyseal plaques generate growth disorders such as misalignments and length changes with amyotrophy.<sup>[23]</sup>

## Radiology

on X-rays, OO appears as a small radiotransparent nidus (<1 cm) surrounded by a halo of condensation. The indirect manifestations of synovial inflammation and joint effusion, or symptoms mimicking those of osteoarthritis may be present.<sup>[25][26]</sup>

CT, performed in 02 cases, fundamental for the diagnosis of intra and juxta-articular OO.<sup>[40,41]</sup> with realization of too thin cuts to avoid false negatives.

Bone scintigraphy has a sensitivity of 100%.<sup>[27]</sup> It has been occurred in one case.

MRI, too influenced by the edematous reaction in the soft tissues, appears significantly less better than CT in the diagnosis of osteoid osteoma.<sup>[28][29]</sup>

## Pathology

Mass resection is preferable for the pathologist. It facilitates the diagnosis by the presence of the nidus which is specific to the lesion.

Curettage should be prohibited because in addition to the risk of recurrence and local dissemination, it does not provide valid material for pathological examination.

## Treatment – complications

Resection of the nidus alone is necessary and sufficient for healing, but its simple mechanical or physicochemical destruction is also possible.<sup>[30][20]</sup>

The problematic of the surgical treatment of OO comes from the small size of the nidus and difficulty in locating during surgery.

Non-operative treatment is an option since the natural history of osteoid osteoma is spontaneous involution after years.<sup>[31]</sup> Resolution of symptoms has been reported within 6 to 15 years of evolution.

The use of aspirin or NSAIDs reduces this duration to 2 to 3 years,<sup>[32][33]</sup> but there is some cases of osteoblastoma from OO medically treated.

In surgical treatment, the nidus can be radiographically spotted by an X-ray image intensifier but it lacks precision.

Scannographic spotting<sup>[42, 62]</sup> which is more chosen in practice, and isotopic<sup>[60, 61]</sup> are also possible.

Surgery can be open, with resection of the nidus and the need to fill the loss of bone or supportive osteosynthesis depends on the volume resected and the resulting weakening the ensuing weakening.

Approach depends on the location of the OO. In our cases, 2 patients underwent Watson Jones approach, Moore approach for the two others.

Anterior approach is usually performed for cervical and anterior OO, the lateral one for those localized on the great trochanter.

For the base of the femoral neck, anterior approach is preferable.

Mini-invasive technics are more and more performed.

CT-guided percutaneous drilling is a safe and reliable method of treating OO.<sup>[34]</sup>

Bone weakening and logistical logistical problem of assembling the orthopedic surgeon, radiologist, and anesthesiologist in the tomography room, are the principal limits of the procedure.

Percutaneous resection by radiofrequency or laser and CT-guided thermocoagulation are a major contribution in the treatment of OO but not available in orthopedic centers in Morocco.

The choice of technique depends on its availability, degree of invasiveness, the risk of failure associated with the technique after the first surgery<sup>[73,77]</sup> and the resumption of the patient's activity.<sup>[77,78]</sup>

## CONCLUSION

Osteoid osteoma is a relatively common benign osteoblastic tumor. It usually occurs in young adult males.

Easy to diagnose, OO poses, essentially, a problem of intraoperative spotting of the nidus.

The interest of our work is to bring 04 cases of relatively atypical femur localizations of osteoid osteomas, and to insist on the place of open surgery, the only and effective procedure in the absence of the possibility of other more modern techniques.

## Consent

The patients have given their informed consent for the case to be published.

## Competing Interests

The authors declare no competing interest.

## Authors 'Contributions

All authors have read and agreed to the final version of this manuscript and have equally contributed to its content and to the management of the manuscript.

## REFERENCES

1. Toriani M, Rosenthal Di: Percutaneous radiofrequency treatment of osteoid osteoma. *Pediatric Radiol*, 2002; 32: 615- 618.
2. HL. JAFFE Osteoid osteoma: benign osteoblastic tumor composed of osteoid and atypical bone. *Arch. Surg*, 1935; 31: 709-728.
3. Clavert JM. Ostéome ostéoïde et ostéoblastome. In : *Les cahiers d'enseignement de la SOFCOT. Conférences d'enseignement*.
4. Bonneville P, Ralhlac JJ. Ostéome ostéoïde, ostéoblastome. *Encyclopédie Médicochirurgicale, Appareil locomoteur*, Paris. Elsevier, 2001.
5. Laffosse JM, Tricoire JL, Cantagrel A, Wagner A, Puget J. Osteoid osteoma of the carpal bones - Two case reports. *Joint Bone Spine*, 2006; 73(5): 560-3.
6. Efstathopoulos N, Sapkas G, N Xypnitos F, Lazaretos I et al. Recurrent intraarticular osteoid osteoma of the hip after radiofrequency ablation: a case report and review of the literature. *Cases Journal*. 2009; 2: 6439.
7. Girard J, Bequet E, Limousin M, Chantelot C, Fontaine C. Osteoma osteoid of trapezoid bone: a case-report and review of the literature. *Chir Main*, 2005; 24(1): 35-8.
8. Saidi H, El Bouanani A, Ayach A, Fikry T. Ostéome ostéoïde du lunatum : à propos d'un cas. *Chir Main*. 2007;26(3):173-5.
9. Allen SD, Saifuddin A. Imaging of intra-articular osteoid osteoma. *Clin Radiol*, 2003; 58(11): 845-52.
10. Szendroi M, Kollo K, Antal I, Lakatos J, Szoke G. Intraarticular osteoid osteoma: clinical features,

- imaging results, and comparison with extraarticular localization. *J Rheumatol*, 2004; 31: 957-964.
11. Eggel Y, Theumann N, Lüthi F. Intra-articular osteoid osteoma of the knee: clinical and therapeutical particularities. *Joint Bone Spine*, 2007; 74(4): 379-81.
  12. Dae-Hee L, Woong-Kyo J, Soon-Hyuck L. Arthroscopic Excision of Osteoid Osteomas of the Hip in Children. *Journal of pediatric orthopaedics*, 2009; 29: 547- 551.
  13. Richardson M, Taylor J. Intra-articular Osteoid Osteoma in the Femoral Neck of an 18-year-old Man: A Case Report. *Journal of Manipulative and Physiological Therapeutics*, 2009; 32: 245-250.
  14. Papagelopoulos PJ, Mavrogenis AF, Kyriakopoulos CK, Benetos IS et al. Radiofrequency Ablation of Intra-articular Osteoid Osteoma of the Hip. *The Journal of International Medical Research*, 2006; 34: 537-544.
  15. Georgoulis AD, Papageorgiou CD, Moebius UG, Rossis J, Papadonikolakis A, Soucacos PN. The diagnostic dilemma created by osteoid osteoma that presents as knee pain. *Arthroscopy*, 2002; 18(1): 32-7.
  16. Daubinet G, Laredo JD, Djian P. Un cas rare d'ostéome ostéoïde de la coronoïde d'apparition et d'évolution Inhabituelle. *EMCRhumatologie Orthopédie*, 2004; 1: 261-263.
  17. Tounsi N, Trigui M, Ayadi K, Kallel S, Boudaouara Sallemi T, Keskes H. Ostéome ostéoïde de l'olécrane. *Rev Chir Orthop Reparatrice Appar Mot*, 2006; 92(5): 495-8.
  18. Niamane R, Lespessailles E, Deluzarches P, Vialat JF, Maitre F, Benhamou LC. Osteoid osteoma multifocally located and recurrent in the carpus. *Joint Bone Spine*, 2002; 69(3): 327-30.
  19. Kalb K, Schlör U, Meier M, Schmitt R, Lanz U. Osteoid osteoma of hand and wrist. *Handchir Mikrochir Plast Chir*, 2004; 36(6): 405-10.
  20. Ward WG, Eckardt JJ, Shayestehfar S, et al. Osteoid osteoma diagnosis and management with low morbidity. *Clin Orthop Relat Res*, 1993; 291: 229-35.
  21. Gitelis S, Schajowicz F. Osteoid osteoma and osteoblastoma. *Orthop Clin North Am*, 1989; 20(3): 313-325.
  22. Alani WO, Bartal E. Osteoid osteoma of the femoral neck simulating an inflammatory synovitis. *Clin Orthop Relat Res*, 1987; (223): 308-312.
  23. Chantal Job Deslandre-Pathologie de la hanche chez l'enfant –Revue du rhumatisme, 2009; 76: 361-3.
  24. Boscainos PJ, Cousins GR, Kulshreshtha R, et al. Osteoid osteoma. *Orthopedics*, 2013; 36: 792-800.
  25. Klein MH, Shankman S. Osteoid osteoma: radiologic and pathologic correlation. *Skeletal Radiol*, 1992; 21(1): 23-31.
  26. Schlesinger AE, Hernandez RJ. Intracapsular osteoid osteoma of the proximal femur: findings on plain film and CT. *AJR Am J Roentgenol*, 1990; 154(6): 1241-1244.
  27. Ziegler DN, Scheid DK. A method for location of an osteoid-osteoma of the femur at operation. A case report. *J Bone Joint Surg Am*, 1992; 74(10): 1549-1552.
  28. Assoun J, Richardi J, Railhac JJ. Osteoid osteoma: MR imaging versus CT. *Radiology*, 1994; 191: 217-223.
  29. Hayes CW, Conway WF, Sundaram M. Misleading aggressive MR imaging appearance of some benign lesions. *Radiographics*, 1992; 12: 1119-1134.
  30. Gangi A, Dietermann JL, Clavert JM, Dodelin A, Mortazavi R, Durcke L et al. Traitement des ostéomes ostéoïdes par photocoagulation au laser. À propos de 28 cas. *Rev Chir Orthop*, 1998; 84: 676-684.
  31. Golding JS. The natural history of osteoid osteoma: with a report of twenty cases. *J Bone Joint Surg*, 1954; 36: 218-29.
  32. Carpintero-Benitez P, Aguirre MA, Serrano JA, Lluch M. Effect of rofecoxib on pain caused by osteoid osteoma. *Orthopedics*, 2004; 27: 1188-91.
  33. A. Haddam et al. / Médecine Nucléaire, 2009; 33: 663. Success of intraoperative.
  34. Bonneville P, Assoun J, Chicoisne MP, Clement JL, Cahuzac JP, Railhac JJ et al. Intérêts de la technique de résection percutanée sous scanner des ostéomes ostéoïdes selon R Kohler. *Maîtrise Orthop*, 1993; 23: 1-12.