



# METACHRONOUS OSTEOID OSTEOMA OF THE MID DIAPHYSIS AND PROXIMAL METAPHYSIS OF THE TIBIA: DOUBLE LOCALIZATION AT 5 YEARS INTERVAL

V. PAVONE<sup>1</sup>, G. TESTA<sup>1</sup>, F. ISFAN<sup>2</sup>, A. SESSA<sup>3</sup>, G. SESSA<sup>1</sup>, F. CANAVESE<sup>4</sup>

<sup>1</sup>Department of Orthopedics, University Hospital "Policlinico-Vittorio Emanuele", University of Catania, Catania, Italy

<sup>2</sup>Department of Pediatric Oncology, University Hospital Estaing, Clermont Ferrand, France

<sup>3</sup>Il Clinic, Biomechanics Laboratory, Rizzoli Orthopedic Institute, Bologna, Italy

<sup>4</sup>Pediatric Surgery Department, University Hospital Estaing, Clermont Ferrand, France

**Abstract** – *Osteoid osteoma (OO) is a benign osteoblastic tumor occurring frequently during growth. Treatment is based on miniminvasive techniques such as radiofrequency ablation, microwave ablation or cryoablation, nowadays preferred to surgical or arthroscopic excision. The disease may recur, mainly owing to incomplete tumor excision or ablation.*

*A girl aged 6 years presented with non-specific left anterior leg pain that had persisted for two weeks. OO of the left tibia at the level of mid-diaphysis was diagnosed, and surgical excision and curettage were performed. After five years the patient started complaining of pain at the proximal left tibia, with clinical characteristics similar to the earlier instance. Radiological findings were compatible with a new-onset OO. The patient underwent surgical excision of the tumor followed by bone grafting, resuming full daily activities after 18 months.*

*We report an unusual case of double localization of pediatric osteoid osteoma of the tibia at five years interval. Further studies may establish a systemic etiology and associated morbidities.*

**KEYWORDS:** *Osteoid osteoma, Surgical excision, Children, Double localization.*

## INTRODUCTION

Osteoid osteoma (OO) is a benign osteoblastic tumor frequently occurring during the growth years, and represents 10-12% of all benign bone tumors<sup>1,2</sup>. Its clinical presentation consists of localized nocturnal pain that disturbs rest and requires anti-inflammatory drugs, symptom intensity increasing in young adults<sup>3,4</sup>. The lesion can be localized at diaphysis, metaphysis, epiphysis or in periarticular surfaces<sup>2,3</sup> with subperiosteal region, cancellous or cortical bone frequently involved. Hip, elbow and ankle are the most often affected joints<sup>5</sup>.

Digital radiology and CT scan are important in its diagnosis, with typical findings being intracortical nidus with a variable amount of calcification, cortical thickening, sclerosis, and bone marrow edema. Diagnosis is based on clinical and radiological findings, and a biopsy should always be done when feasible.

In the past, treatment usually consisted in surgical or arthroscopic excision<sup>6,7</sup>; nowadays, miniminvasive techniques such as radiofrequency ablation, microwave ablation or cryoablation<sup>8</sup> are largely diffused.

Recurrences are described after both treatment options, owing to incomplete tumor removal<sup>9</sup> although recurrence many years later is very uncommon<sup>10</sup>. Multiple nidi may be present close to each other in a single bone or in adjacent bones, or may be present in separate bones in multicentric lesions. The latter is rarer and is usually called metachronous osteoid osteoma.

Here we present the case of a 6-year-old female with an atypical manifestation of OO. The tumor was characterized by a double localization with onset of the second lesion 5 years after the first one. The tumors were located in different regions of the tibia, namely mid-shaft and proximal metaphysis.



We report on the atypical and unique presentation of a double, metachronous OO, and describe our treatment strategy.

### CASE REPORT

A girl aged 6 years presented with non-specific anterior left leg pain that had persisted for two weeks and gradually increased in intensity. She underwent intensive physiotherapy for 4 weeks, with no improvement in pain or leg function. Resting pain at night worsened, and was resistant to non-steroidal anti-inflammatory drugs (NSAIDs).

Physical examination showed a limp and swelling of the anterior aspect of the left leg. Palpation of the anterior leg found marked tenderness. Neurovascular examination was negative, and blood results were normal. A radiographic evaluation of the affected leg was performed and compared with the contralateral side. An area of sclerotic bone surrounding a radiolucent area was identified at the level of the mid-diaphysis of the left tibia (Figure 1). A computed tomography (CT) scan evidenced a



**Fig. 1.** Area of sclerotic bone surrounding a radiolucent area at the level of the mid-diaphysis of the left tibia by digital radiology.



**Fig. 2.** CT scan of tibial mid-shaft osteoid osteoma.

nidus with a sclerotic rim localized in the mid-tibial region (Figure 2). Imaging was compatible with OO, and surgical excision and curettage were performed. Biopsy specimens revealed a focus of benign osteoblastic lesion characterized by a well-demarcated core (nidus) with a surrounding zone of reactive bone formation, confirming the diagnosis of OO. After surgery, partial weight-bearing was allowed at 24 h after the operation, and full weight-bearing was permitted at 2 weeks. Complete restoration of full articular functions was reported.

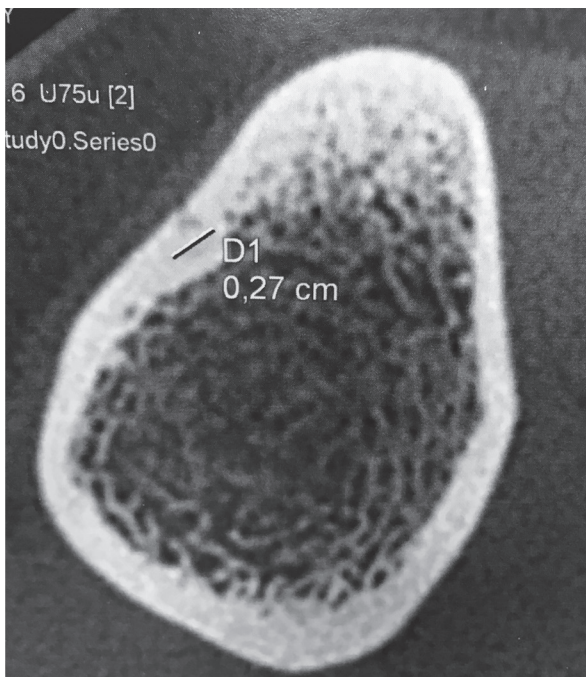
Six months later, the patient re-presented for a left undisplaced distal tibial fracture, corresponding to the surgical site (Figure 3). There was no fever or any other signs of infection. A short-leg cast immobilization was applied and maintained for 5 weeks, with early union and functional restoration. Three months after cast removal the patient was pain-free and had resumed full daily and sports activities.

Five years from the first appearance, the girl, now aged 11 years, reported new left leg pain with similar frequency and characteristics, but a different location (proximal left leg). No positive history of trauma could be identified. The symptoms, which worsened during the night, were relieved by treatment with NSAIDs. The CT scan showed the presence of a nidus cavity of 4-5 mm surrounded by a distinctive zone of reactive bone at the level of the proximal left tibia (Figure 4). These findings pointed to a new

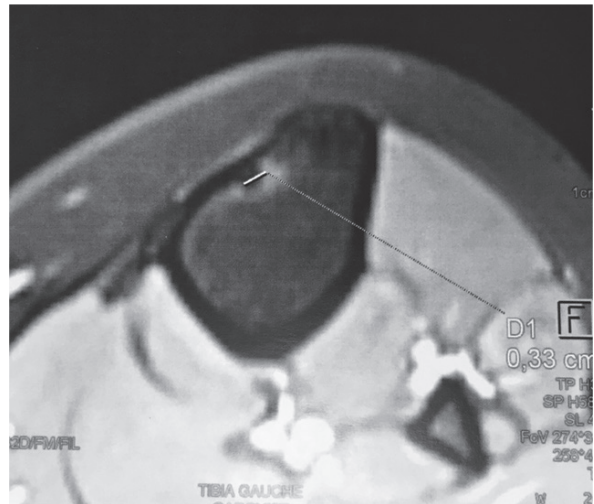


**Fig. 3.** Left undisplaced distal tibial fracture, corresponding to the surgical site by digital radiology.

OO. Magnetic resonance imaging (MRI) and bone scintigraphy were compatible with OO (Figures 5, 6). Importantly, radiographic images recorded at the time the first lesion was discovered were normal at the level of the proximal tibia (Figure 1).



**Fig. 4.** Distinctive zone of reactive bone at the level of the proximal left tibia by CT scan.

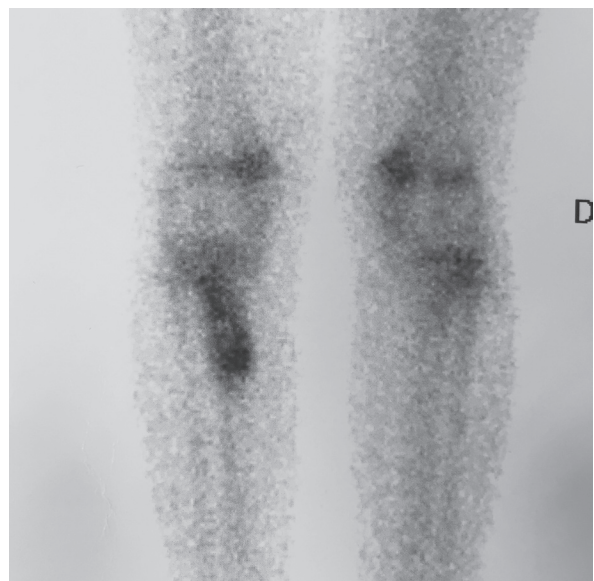


**Fig. 5.** MRI of osteoid osteoma of left tibia.

The patient underwent a new surgical treatment, consisting of surgical excision of the tumor and bone grafting. Biopsy confirmed the diagnosis of OO, and excluded malignancy or infection. Weight bearing was allowed 2 weeks after surgical procedure. Eighteen months after surgical treatment of the second lesion, the patient was pain-free and resumed full daily and sports activities.

## DISCUSSION

Osteoid osteoma is a benign tumor, common in young patients. Predilected localization is in the shaft of a long bone near the metaphyseal junction, especially in the lower limb<sup>11,12</sup>. In our patient the localizations of OO were extra-articular.



**Fig. 6.** Bone scintigraphy of left tibia.



Typical clinical features are nocturnal pain that prevents rest, and increasing recourse to analgesic drugs. Intra-articular presentation may be associated with muscle atrophy, tenderness and swelling, if diagnosis is delayed<sup>13</sup>. Conventional radiographs are the first diagnostic assessment, and show the nidus as a small lytic spot surrounded by a radiolucent ring<sup>14</sup>. If typical and radiological features are not present, a CT scan can be useful to make an accurate early diagnosis<sup>15</sup> as observed in our patient. MRI is not as accurate as a CT scan for defining the extent of the nidus<sup>14,16</sup>. It is important for differential diagnosis, because OO can have symptoms similar to chronic ankle instability, anterior impingement, stress fracture, osteomyelitis, osteonecrosis and monoarticular arthritis<sup>17</sup>.

In past years, OO was conservatively treated, because it was believed that symptoms could spontaneously resolve after more than ten years, based on radiological findings<sup>18</sup>. Although the tumor has been considered self-limiting, in young children surgical treatment is indicated to prevent growth disturbance, premature arthritis, and muscle atrophy<sup>12</sup>. In our patient, who came to our observation after physical therapy had failed, and with pain resistant to analgesic drugs, surgical treatment was chosen.

Different options were available. Surgical excision has been considered as the gold-standard treatment for OO, with good results in 88-100% of cases<sup>19,20</sup>. Arthroscopic excision is considered, being less invasive than arthrotomy, providing good visualization of lesions and shortening post-operative rehabilitation times. However, it carries the risk of damaging articular surface and of intra-articular dissemination of a potential malignancies or infections in case of doubtful diagnoses<sup>3-7</sup>. Nowadays, mini-invasive techniques such as radio-frequency ablation, microwave ablation or cryo-ablation, all performed under CT scan guidance, have largely taken the place of standard, en-bloc excision of the tumor having shown equivalent safety and efficacy but significantly shorter hospital stay. In particular, radio-frequency ablation offers an alternative to conventional surgical excision, with less risk than open surgery, and reported success rates of over 90%<sup>8,21,22</sup>.

In the patient described here, traditional surgery was chosen, to remove the tumor completely as alternative mini-invasive techniques were not available at the time of the surgical procedures. Following the first surgical procedure, partial weight bearing was allowed 24 hours after surgery. Partial weight bearing was allowed 2 weeks after the second surgery, as surgical excision of the tumor was more extensive, requiring bone grafting.

Recurrence of OO is a possible complication, usually owing to an insufficient excision or ablation of the tumor<sup>23</sup>. However, in our patient, the second OO appeared 5 years after the first one, in another region of the same tibia bone. The term recurrence,

defined as a new lesion formed at the site of the completely excised tumor, may therefore be inappropriate here<sup>12</sup>. It is known that patients undergoing piecemeal resection with intra-lesional margins are at higher risk of local recurrence than patients treated by local excision, which offers a better chance of complete excision of the nidus.

A similar case was reported by Allagui et al<sup>10</sup> in a 26-year-old woman, who presented a rare double localization of an OO of the ankle, the first focus in the tibial metaphysis and the second located in the neck of the homolateral talus, at the same time. Another case was reported by Niamane et al<sup>24</sup>. The authors report a case of OO located simultaneously at the right capitate and at the proximal part of the right third metacarpus. One year after the surgical excision of the tumor at the right capitate, a second intervention was needed for local recurrence. However, a third OO was discovered 16 months later at the proximal part of the right third metacarpal bone, requiring surgical treatment. The particularity of our case is that the two lesions occurred 5 years apart at two distinct sites of the same bone. The second interesting finding is that both localizations occurred when the patient was still skeletally immature<sup>24</sup>. To the best of our knowledge, such a case has never been reported in the English-language literature. On the other hand, several cases of OO with double nidus in children have been reported. Matera et al<sup>25</sup> reported on a 10-year-old female with spontaneous pain in her right hip and leg for a 6-month period. Clinical examination and imaging studies confirmed a diagnosis of OO with a double nidus at the level of the femur. The lesion was treated by CT-guided thermo-ablation with radio frequency under general anaesthesia. Similarly, Myiazaki et al<sup>26</sup> reported on a young adult boy with OO in the left tibia, containing double nidi. Plain radiography and CT showed two adjacent nidi surrounded by bony sclerosis within the distal left tibia. The tumor was successfully treated using two separate CT fluoroscopy-guided percutaneous radio-frequency ablations during a single session. Allieu et al<sup>27</sup> made the definitive diagnosis of double OO one year later, owing to the persistence of pain.

The case reported here is an unusual instance of double localization of pediatric osteoid osteoma of the ankle, reported five years apart. In a patient with a history of a previous OO, presenting with deep, continuous pain in a different site, increasing during the night and relieved by oral NSAIDs, an accurate evaluation of radiographic findings is thus recommended, because OO is a possible differential diagnosis. Further studies to establish systemic etiology and associated morbidities will be needed.

The patients and their families were informed that data from the case would be submitted for publication, and gave their consent.

**CONFLICT OF INTEREST:**

The authors declare that there is no conflict of interests regarding the publication of this paper.

**REFERENCES**

1. GANGI A, ALIZADEH H, WONG L, BUY X, DIETEMANN JL, ROY C. Osteoid osteoma: percutaneous laser ablation and follow-up in 114 patients. *Radiology* 2007; 242: 293-301.
2. ATESOK KI, ALMAN BA, SCHEMITSCH EH, PEYSER A, MANKIN H. Osteoid osteoma and osteoblastoma. *Am Acad Orthop Surg* 2011; 19: 678-689.
3. BOJANIC I, ROGOSIC S, MAHNIK A, SMOLJANOVI T. Removal of osteoid osteoma of the tibia using two-portal posterior ankle arthroscopy. *J Foot Ankle Surg* 2012; 51: 103-110.
4. RIZZELLO G, LONGO UG, MAFFULLI N, DENARO V. Arthroscopic removal of an intraarticular osteoid osteoma of the distal tibia. *J Foot Ankle Surg* 2010; 49: 398.e17-398.e21.
5. MORBIDI M, VENTURA A, DELLA ROCCA C. Arthroscopic assisted resection of juxtaarticular osteoid osteoma. *J Foot Ankle Surg* 2007; 46: 470-473.
6. FURUKAWA M, ANAZAWA U, HORIUCHI K, YABE H, MORIOKA H, MUKAI M, TOYODA T, CHIBA K, MORII T, SHIRAIISHI T, TOYAMA Y. Arthroscopic removal of intra-articular osteoid osteoma in the knee: case report and review of the literature. *J Orthop Sci* 2011; 16: 321-325.
7. GAO K, HUA Y, CHEN S, LI Y, XIN Q. Arthroscopic excision of juxtaarticular cancellous osteoid osteoma in the talar neck. *Knee Surg Sports Traumatol Arthrosc* 2013; 21: 1300-1303.
8. BRUNERS P, PENZKOFER T, GÜNTHER RW, MAHNKEN A. Percutaneous radiofrequency ablation of osteoid osteomas: technique and results. *Rofo* 2009; 181: 740-747.
9. FENICHEL I, GARNIACK A, MORAG B, PALT R, SALAI M. Percutaneous CT guided curettage of osteoid osteoma with histological confirmation: a retrospective study and review of the literature. *Int Orthop* 2006; 30: 139-142.
10. ALLAGUI M, BAUER T, ROUSSELIN B, JOUVE F, HARDY P. Double localization of an osteoid osteoma of the ankle: percutaneous treatment by CT-guided drilling. *Rev Chir Orthop Reparatrice Appar Mot* 2007; 93: 506-510.
11. YERCAN HS, OKCU G, OZALP T, OSIÇ U. Arthroscopic removal of the osteoid osteoma on the neck of the talus. *Knee Surg Sports Traumatol Arthrosc* 2004; 12: 246-249.
12. SAMPATH SC, SAMPATH SC, ROSENTHAL DI. Serially recurrent osteoid osteoma. *Skeletal Radiol* 2015; 44: 875-881.
13. FRANCESCHI F, LONGO UG, RUZZINI L, MARINOZZI A, RIZZELLO G, PAPALIA R, DENARO V. En-bloc retrograde resection of an osteoid osteoma of the patella using computed tomography under arthroscopic control. *J Knee Surg* 2008; 21: 136-140.
14. SHUKLA S, CLARKE AW, SAIFUDDIN A. Imaging features of foot osteoid osteoma. *Skelet Radiol* 2010; 39: 683-689.
15. FRANCESCHI F, MARINOZZI A, PAPALIA R, LONGO UG, GUALDI G, DENARO E. Intra- and juxta-articular osteoid osteoma: a diagnostic challenge: misdiagnosis and successful treatment: a report of four cases. *Arch Orthop Trauma Surg* 2006; 126: 660-667.
16. DONLEY BG, PHILBIN T, ROSENBERG GA, SCHILS JP, RECHT M. Percutaneous CT guided resection of osteoid osteoma of the tibial plafond. *Foot Ankle Int* 2000; 21: 596-598.
17. ROSENTHAL D, CALLSTROM MR. Critical review and state of the art in interventional oncology: benign and metastatic disease involving bone. *Radiology* 2012; 262: 765-780.
18. SIMM RJ. The natural history of osteoid osteoma. *Aust N Z J Surg* 1975; 45: 412-415.
19. CAMPANACCI M, RUGGIERE P, GASBARRINI A, FERRARO A, CAMPANACCI L. Osteoid osteoma. Direct visual identification and intralesional excision of nidus with minimal removal of bone. *J Bone Joint Surg Br* 1999; 81: 814-820.
20. CANTWELL CP, OBYRNE J, EUSTACE S. Current trends in treatment of osteoid osteoma with an emphasis on radiofrequency ablation. *Eur Radiol* 2004; 14: 607-617.
21. RIMONDI E, MAVROGENIS AF, ROSSI G, CIMINARI R, MALAGUTI C, TRANFAGLIA C, VANEL D, RUGGIERI P. Radiofrequency ablation for non-spinal osteoid osteomas in 557 patients. *Eur Radiol* 2012; 22: 181-188.
22. EARHART J, WELLMAN D, DONALDSON J, CHESTERTON J, KING E, JANICKI J. Radiofrequency ablation in the treatment of osteoid osteoma: results and complications. *Pediatr Radiol* 2013; 183: 29-33.
23. LINDNER NJ, OZAKI T, ROEDL R, GOSHEGER G, WINKELMANN W, WÖRTLER K. Percutaneous radiofrequency ablation in osteoid osteoma. *J Bone Joint Surg Br* 2001; 83: 391-396.
24. 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: 327-330.
25. MATERA D, CAMPANACCI DA, CALDORA P, MAZZA E, CAPANNA R. Osteoid osteoma of the femur with a double nidus: a case report. *Chir Organi Mov* 2005; 90: 75-79.
26. MIYAZAKI M, KURABAYASHI T, TSUSHIMA Y. CT fluoroscopy-guided radiofrequency ablation of osteoid osteoma with double nidi. *Jpn J Radiol* 2012; 30: 78-80.
27. ALLIEU Y, LUSSIEZ B, BENICHOU M, CENAC P. A double nidus osteoid osteoma in a finger. *J Hand Surg Am* 1989; 14: 538-541.