

Osteoid osteoma of the proximal phalanx of the hallux: a case report of a challenging diagnosis

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Summary. Osteoid osteoma is a benign bone lesion that accounts for approximately 10% to 12% of all benign bone tumors. More than 80% of lesions occur in patients between 5 to 25 years old; males are more commonly affected with a ratio of 3:1. The foot is rarely involved: its involvement is less than 4% in the foot and of 1.7% in the metatarsals. In this paper we discuss the case of a 27-years-old woman with a 12 months follow-up, presented with an osteoid osteoma of the proximal phalanx of the great toe that underwent an en-bloc excision of the lesion and subsequent filling with cancellous autograft from the ipsilateral calcaneus. (www.actabiomedica.it)

Key words: osteoid osteoma, foot, hallux, phalanx, excision

Background

Osteoid osteoma (OO) is a common benign bone lesion that accounts for approximately 10% to 12% of all benign bone tumors analyzed by biopsy (1). These tumors were first recognized as a distinct group in 1932 thanks to Jaffe's work published only in 1935 (2). Its peak of incidence is during the second decade of life although more than 80% of lesions occur in patients between 5 to 25 years old, while rare lesions have been observed in patients as young as 2.5 years of age. Males are more commonly affected with a ratio of 3:1. It is an osteoblastic tumor; at least 50% of these tumors occur in the diaphyseal and metaphyseal regions of long bones of the lower extremities. The foot is rarely involved, 3 indeed in a review Jackson found an incidence of less than 4% in the foot and of 1.7% in the metatarsals (4). It is histologically characterized by a central region of fibro-osseous tumor (usually smaller than 1, 5 cm) surrounded by a distinctive zone

of reactive bone sclerosis (5). The characteristic clinical feature is a local pain pattern, which is rapidly alleviated by the administration of NSAIDs or aspirin. This pain is typically referred at rest and at night. The ability of NSAIDs to alleviate pain from osteoid osteomas is due to the well-described production of prostaglandin E2 and prostacyclin by the tumor. When the lesion is superficial, it could be also associated with local swelling (6, 7). Not usual locations and unclear symptoms could delay diagnosis. The typical x-ray appearance for lesions localized in cortical bone shows a "nidus", consisting of a central lucency surrounded by a sclerotic rim of bone. When the OO is located in the cancellous bone or in the sub-periosteal region of small bones of the foot, x-ray could not exhibit the typical sclerotic image around the nidus (8, 9). Moreover radiographically the OO must be distinguished from a Brodie abscess and from a stress fracture. This article describes the case of a 27- years-old woman with an uncommon OO of the proximal phalanx of the great toe.

Case Report

A 27-years-old woman presented in September 2016 with dull aching pain at the proximal phalanx of the great toe of the left foot for over 12 months, with no previous history of trauma, no other diseases, no allergies, no previous surgery. The patient didn't refer about other joint pain and family history. She didn't show any local sign, except for a slight painful motion at metatarsophalangeal joint. The pain was worse at night, but remitted partially with NSAIDs or aspirin assumption. Local palpation exacerbated the pain. There were no neurovascular disorders in the limb, laboratory parameters and inflammatory markers were within normal limits. Plain x-rays (Fig. 1) didn't show any lesion at the hallux. Otherwise CT scan and MRI revealed a focal lesion (maximum diameter 2, 5 mm) with a central hypodense area and sclerotic peripheral component. (Fig. 2). Bone scintigraphy with technetium-99 (Fig. 3) revealed a small area of hypermetabolic activity at the base of the proximal phalanx of the great toe to the left foot. These appearances were suggestive of an OO. After discussion with the patient, due to poor pain control, the decision was made to undertake an excisional biopsy of the lesion (No-

vember 2016). The definitive diagnosis could only be confirmed by histologic examination. A medial incision was made over the proximal phalanx of the hallux. The area of the lesion was identified and an en-bloc excision was performed using a chisel (Fig. 4). The cavity was then filled with a cancellous autograft from the ipsilateral calcaneus (Fig. 5). The bone wedge was sent for histopathological examination. Evaluation with hematoxylin-eosin staining revealed a hyper-cellular central area ("nidus") without atypical cells, surrounded by areas with thickened trabeculae and osteoblastic rims (Fig. 6-7), confirming the diagnosis of OO. The patient began weight-bearing after surgery with postoperative flat-soled shoe for 4 weeks. The pain remitted immediately after surgery. Early rehabilitation showed marked improvement of joint mobility. At 12 months post-operative follow-up (November 2017), the patient remained asymptomatic with no signs of joint degeneration or any evidence of OO recurrence. She resumed her usual activities and had no complications.



Figure 1. Preoperative AP x-rays of the left foot

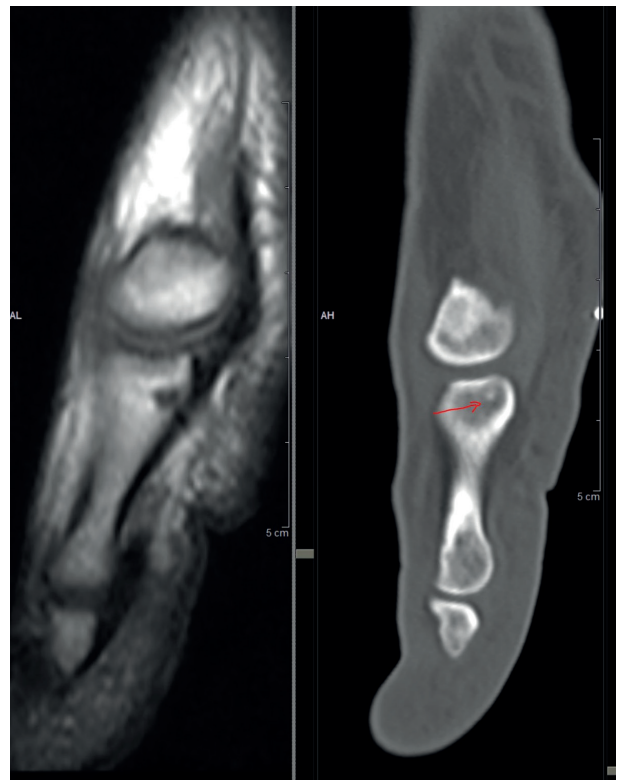


Figure 2. Preoperative TC and MRI images of the left foot showing the lesion at the proximal phalanx

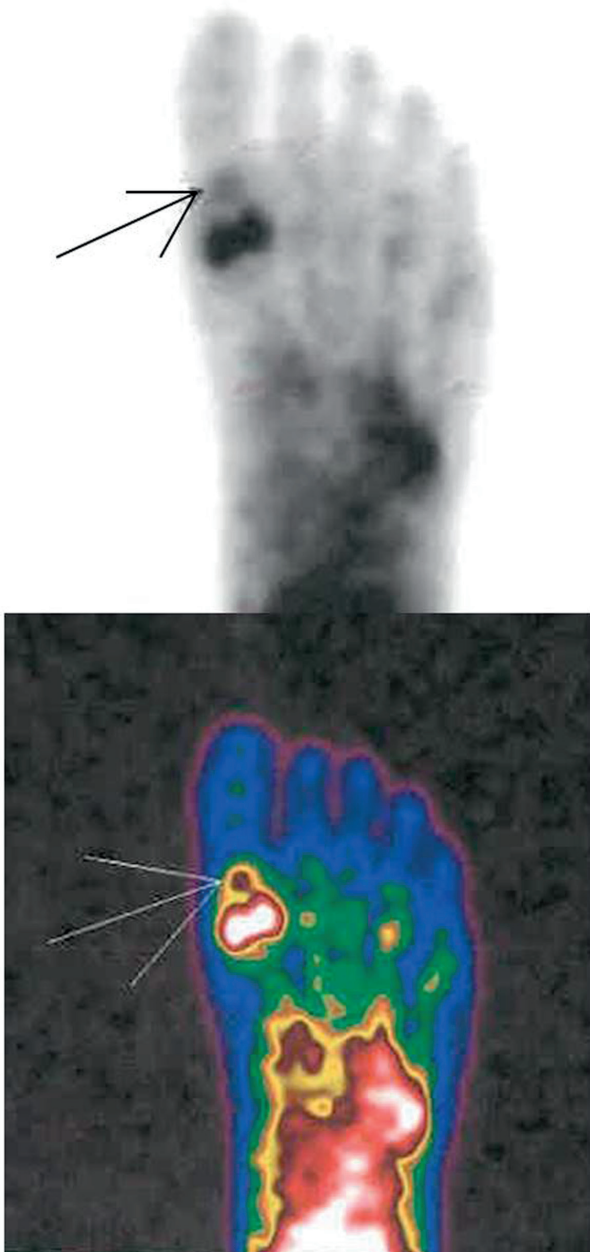


Figure 3. Bone scintigraphy with technetium-99 of the left foot showing the lesion at the proximal phalanx

Discussion

Osteoid osteoma (OO) of the foot is a rare event. The talus is the most commonly involved, but other bones as the calcaneus, cuboid, navicular, and phalanges may be affected in a few cases (4, 7, 10-12).



Figure 4. Intraoperative image showing the complete excision of the lesion of the proximal phalanx

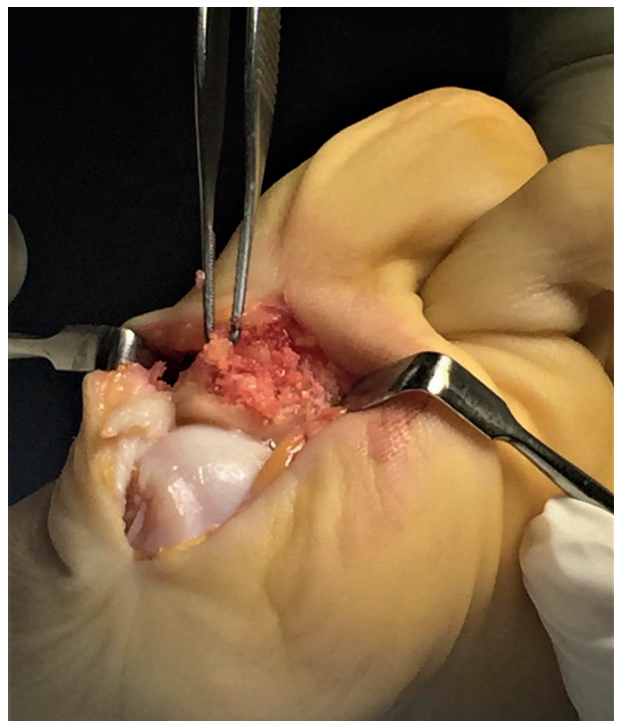


Figure 5. Intraoperative image showing the cavity filled with a cancellous autograft from the ipsilateral calcaneus

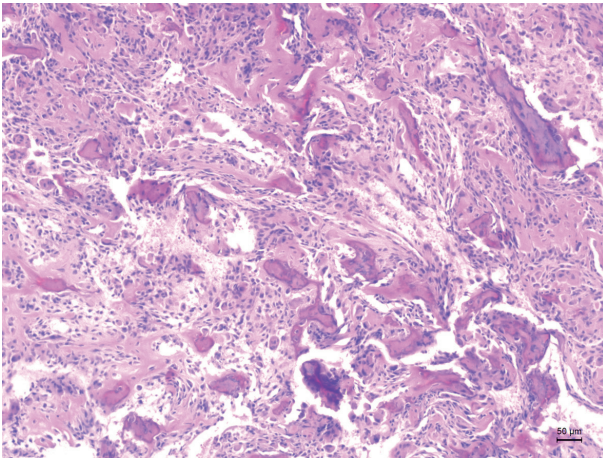


Figure 6. Histopathological examination with hematoxylin-eosin revealing a hyper-cellular central area (“nidus”), surrounded by thickened trabeculae and osteoblastic rims

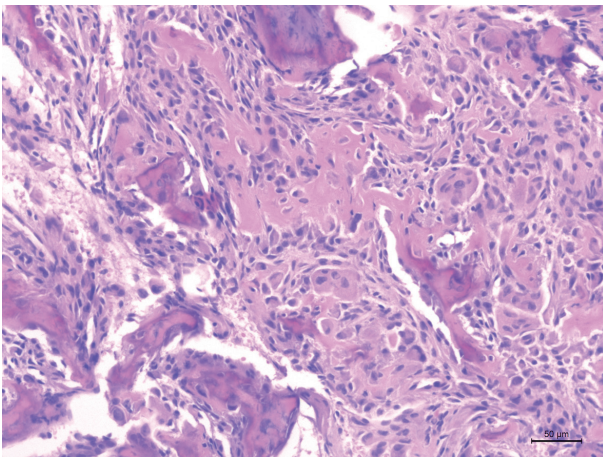


Figure 7. Enlargement of figure 6 showing the nidus surrounded by thickened trabeculae and osteoblastic rims

Sproule reported a case of OO of the second toe (12). Lacroix et al. in 2001 reported a sub periosteal OO at the distal phalanx of the fourth toe (13). With regard to the great toe, cases involving the distal phalanges seem to be rare. Case reports showed OO in the proximal phalanx with involvement of the interphalangeal joint (7) and in the distal phalanx (14-17). In this paper we report our experience with an osteoid osteoma of the proximal phalanx of the hallux, without articular involvement. The cortical type of OO is the most common, but the cancellous and sub-periosteal varieties appear to be more frequent in the small bones

of the hand and foot (18, 19). Local swelling and temperature increase can be confused with an infectious process, but in OO there is no increase in C-reactive protein. Kahn et al. (20) reported a case of a young female with a dull ache in the great toe, associated with local erythematous changes. X-rays revealed an area of radiolucency on the distal phalanx of the hallux. The lesion was excised and microscopy revealed signs consistent with an osteoid osteoma. In our case, the patient didn't show local sign that could be misdiagnosed with an infectious process. Although an uncommon finding in the foot, OO it should always be considered a differential diagnosis in patients presenting with a history of pain and a local increase in temperature. The x-ray appearance of OO in small bones of the foot is different from long bones. In these cases the nidus is usually located in cancellous bone, and osteosclerosis is mild or absent and may be far from the lesion (14). The case reported in this paper in fact didn't show any sign on plain x-rays. CT scan might be helpful for the diagnosis, showing in many cases the contrast between the nidus and the normal trabecular tissue around it (21). MRI may aid, but it is usually less useful than CT in defining bone lesions. Moreover the images may be confused with a malignant process or osteomyelitis due to edema of the cancellous bone. However further investigation with bone scintigraphy should be always performed to reach a correct diagnosis (6). During last decades, noninvasive techniques have been used in the treatment of these lesions, such as CT guided radiofrequency ablation or thermal RFA. The reported results are similar to those obtained in open surgery, though they can't provide a definitive histologic diagnosis. Moreover the CT-guided thermal RFA in a small area like proximal phalanx could damage the surrounding structures including the overlying skin and adjacent nerves structures. Other complications that must be taken into account are the incomplete ablation or recurrence of the lesion, or the fracture of the treated bone. For this reason, surgical resection of the lesion could be considered the treatment of choice and often lead to a resolution of the symptoms. After surgery, recurrence occurs only if the resection of the nidus is not complete (6, 7). Our patient reported a complete resolution of symptoms after surgical resection of the lesion.

The phalanx of the hallux is an uncommon localization for OO, but this entity should be always considered as a possible diagnosis in cases of local dull nocturnal pain without associated etiology. A timely approach followed by clinical and radiological evaluation could avoid delays in diagnosis and in the definitive treatment of the lesion, which is only confirmed by surgical excision and histopathological examination. Even if plain x-rays don't show any significant sign, in cases of persistent pain CT scan, MRI and a SPECT exam should be performed to identify the lesion and carry out a correct diagnostic and therapeutic approach.

Conflict of interest: Each author declares that he or she has no commercial associations (e.g. consultancies, stock ownership, equity interest, patent/licensing arrangement etc.) that might pose a conflict of interest in connection with the submitted article

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