Case Report

Osteoid Osteoma of the Distal Fibula: a Case Report of a Very Rare Location Treatment

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Abstract

Osteoid Osteoma (OO) is a very common small benign osteoblastic tumor. It represents 10% of all bone benign lesions, and is usually localized to the proximal metadiaphysis of the femur. We report the case of an OO localized in the distal inner metadiaphysis of the fibula. To our knowledge this is the first report in literature. We performed en-bloc resection because of the deep localization of the lesion, which was not reachable by minimally invasive procedures. At 6 months follow-up bone stock reconstitution was observed, and no relapse occurred at 1 year. Plain X-rays are generally sufficient to make a diagnosis. When radiological findings are not suggestive, computed tomography and bone scan represent second level examinations. Minimally invasive procedures represent a promising and valuable treatment, but open surgery shall be still taken in account when the location is not easy to reach and/or it is in proximity to neurovascular structures.

Key words:

Osteoid osteoma; Fibula; Benign tumor; Minimally invasive En-bloc resection

Introduction

Osteoid Osteoma (OO) is a very common small benign osteoblastic tumor, generally presenting with pain, often at night [1]. First described by Jaffe, it is characterized by a nidus composed of osteoid and woven bone, surrounded by cortical thickening due to reactive bone [2,3]. It is typical of young patients, with maximum incidence in the 2nd and 3rd decades of life, with a male predilection. It represents 10% of all bone benign lesions, and it is usually localized to the lower limb, especially in proximal metadiaphysis, proximal femur being its most common localization. Another less frequent localization is the hindfoot, followed by the upper limb and spine [4]. OO can be classified in 3 different types according to its localization: cortical, cancellous and subperiosteal. They can be further differentiated in intra- or extra-capsular [5]. The typical clinical presentation is nocturnal pain, sometimes accompanied with regional swelling. The pain is very sensitive to salicylates and NSAIDs [6] with pain easing in half an hour; this represents an important clue to diagnosis [7]. When OO is intra-capsular, it can cause specific arthritis or synovitis; in these cases the diagnosis may be very difficult and delayed [8]. All the clinical features of OO are due to its anatomo-pathological characteristics: recent immune-histochemical staining studies have demonstrated a high presence of COX-1 and COX-2 enzymes inside the nidus, producing high levels of prostaglandins [9]. These inflammatory agents could be responsible for pain, swelling, arthritis and reactive bone surrounding the nidus with a halo of sclerosis [3]. Plain X-rays represent first level imaging examination. The classic appearance is a radiolucent nidus (the tumor itself) surrounded by sclerotic reactive bone. This feature is typical of cortical OO. The cancellous and subperiosteal subtypes cause no or few reactive bone and are consequently unlikely to be seen in radiographs [10]. When symptoms are not typical or X-rays are not clear, bone scan can clearly identify the nidus with high sensitivity but poor specificity [11]. CT scan is the gold standard for diagnosis; it can clearly identify the nidus, its extension, and is very helpful in differential diagnosis [12]. The use of MRI is controversial since it often fails to identify the nidus, and because of intramedullary oedema it can lead to misdiagnosis (other aggressive or malignant tumors, osteonecrosis or stress fractures) [13,14]. Differential diagnoses include: osteoblastoma, stress fracture, abscess, intracortical osteosarcoma, enostosis [15]. Surgical excision is the most common treatment; en-bloc resection along with the surrounding reactive bone is associated with a lower rate of recurrence, even though it is associated with higher risk of fracture because of bone loss [16]. Recent studies report an equivalent rate of success to surgery using percutaneous CT-guided techniques such as laser or radio-frequency ablation [17,18]. The use of these treatments is limited when in proximity with neurovascular bundles, because of a high risk of lesions, and when the OO occurs in small bones, because o f a higher risk of osteonecrosis [19,20]. We present a case of a 28-year-old man, whose nocturnal local pain of the left ankle with improvement under salicylates led to the diagnosis of an osteoid osteoma.

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The patient's first symptoms started 3 years before him presenting to our outpatients' department. He referred diffuse left ankle pain, stronger in the lateral aspect, sometimes irradiated to the proximal leg; no swelling and no trauma was reported. The pain started almost always at night, only occasionally during the day. Paracetamol didn't ease the pain, so the patients started to take autonomously Nimesulide 100mg as the pain started, with an improvement in symptoms. The pain came back for several months, so he underwent his first orthopaedic visit. He performed leg X-Rays (Figure 1), with no pathological findings. No treatment was then started. The patient addressed to a second orthopaedic surgeon; a mild lower back pain started in the meanwhile. Since the patient has a heavy job and because

of back pain, the surgeon suspects a nerve roots compression and asked the patient to perform a lower limbs electromyography and a lumbar spine MRI. The former was negative, but the latter detected a L5-S1 disc herniation, so he started a treatment with Etoricoxib 90mg. The pain was under control as long as the patient took Etoricoxib, but it came back as soon as he stopped. Thus, he decided autonomously to perform a leg MRI and finally came to our clinic in November 2015. The MRI (Figure 2) showed aspecific signal alterations of the peroneal malleolus, best seen in the medial aspect of the fibula just over the tibiofibular syndesmosis. Oedema could be seen in the syndesmosis itself, probably on a reactive basis. Imaging gave us no clue, but the nocturnal pain sensitive to COX-2 inhibitors was consistent with osteoid osteoma, even though the distal fibula is not a typical site of presentation. So we prescribed a 99MTc-MDP Bone Scan that was completed by the radiologist with a Tomoscintigraphy with SPECT/MRI reconstructions (Figure 3) radionuclide uptake in the left distal fibula was seen both in the vascular and the static phases; the signal enhancement was stronger in the deep aspect of the distal fibula showing the typical "double density" sign. The suspect of an osteoid osteoma was thus confirmed. Because of the deep aspect position, and the close proximity of peroneal artery, sural nerve and small saphenous vein, we decided to perform an open procedure using a postero-lateral approach: the goal was en-bloc resection of the lesion along with the reactive bone, excising the medial aspect of the distal fibula, sparing the syndesmosis.

The patient was placed in a prone position with a sandbag under the affected leg. A pneumatic tourniquet was applied at the thigh. A longitudinal incision between the posterior border of the fibula and the lateral border of the Achilles tendon was performed. After incision of the peroneal retinaculum, the tendons and fibres of the peroneal muscles (brevis and longus) and flexor hallucis longus were retracted anteriorly and posteriorly respectively, passing through the internervous plane. The posterior aspect of the distal fibula was exposed (Figure 4) the nidus location was identified by a reactive cortical bone area of about 3x2cm. Under image intensifier, a cortical bone brick was resected and excised using a scalpel. We decided not to reinforce the fibula with a plate because it is not considered a weight-bearing bone and the syndesmosis had not been affected by the surgery. A control X-ray was performed at the end of procedure. No post-operative neurovascular injury was observed and the patient was allowed to walk with no weight-bearing on the affected limb. Since the first night after surgery the patient reported the disappearance of that specific pain he knew for 3 years. The patient was then evaluated at 7 days for a clinical check, and at 14 days to remove surgical sutures and perform a control X-ray. The hypothesis of OO was confirmed by Anatomopathology; microscopically the nidus was clearly identified (Figure 5A), surrounded by reactive sclerotic bone. At high-power magnification it was composed of woven bone trabeculae with lining osteoblasts and osteoid matrix (Figure 5B and 5C). In (Figure 6) post-operative X-Rays are reported. At a 1 month follow-up the patient was completely free from pain, no fracture occurred and he returned to his normal life. At 6 and 12 months the result was confirmed with no recurrence and radiological evidence of bone stock reconstitution.

Figure 1: Left Ankle X-rays .No gross pathologic signs are reported. A: With hindsight a small hypointensity can be seen in the Latero – lateral projection (red circle). B: A-P projection. No signs.
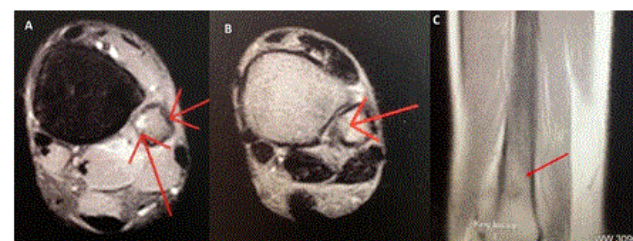

Figure 2: Left leg MRI images. A) COR STIR and B) Cor T2: Signal alternations of the peroneal malleolus, best seen in the deep aspect of the fibula just over the tibiofibular syndesmosis. Oedema can be seen in the syndesmosis itself on a reactive basis. Image C) SAG T1: a probably benign quiescent cyctic lesion is seen 6 cm from tibio-talar articulation.
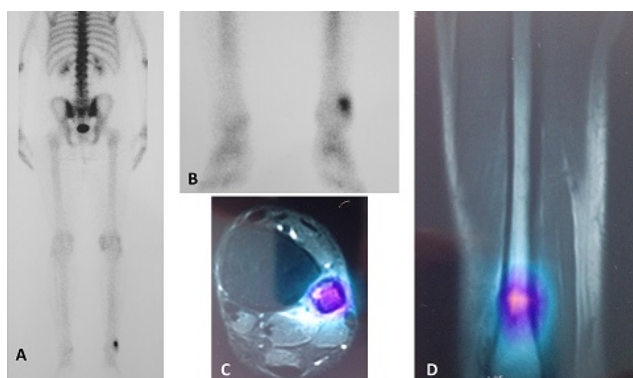

Figure 3: Images A and B: 99MTc-MDP Bone Scan : Radionuclide uptake in the left distal fabula, seen both in the vascular and the static phases . Image C and D: SPECT/RM reconstruction: signal enhancement is stronger in the deep aspect of the peroneal malleolus (double density sign).
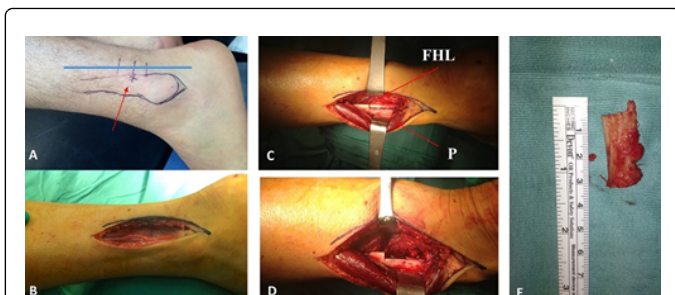
Figure 4: Image A) Incision line (blue) and point of maximum pain (arrow). Image B) Skin incision and muscles exposition. Image c) Fibres and tendons of Flexor Hallucis Longus (FHL) and Peroneal muscles (P) are retracted: cuts are made in the posterior half of the fibula. Image D and E) The lesion is excised.
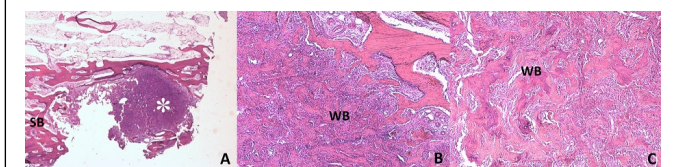

Figure 5: Haematoxylin and eosin stained microscope images. Image A) The nidus * is surrounded by reactive sclerotic bone (SB) (HE, original magnification x6). Image B and C) High- power magnification images (HE, x50 and x100 respectively) show the composition of the nidus; it's made of woven bone trabeculae (WB, red in HE) surrounded by osteoid matrix
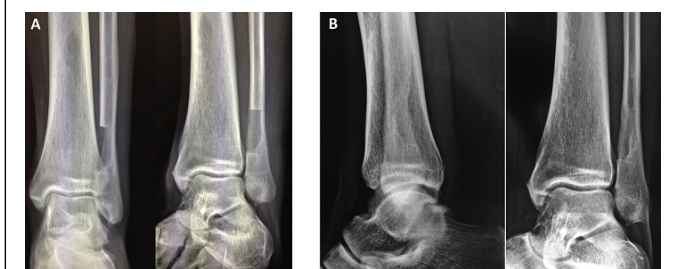

Figure 6: Image A) Post- operative A-P and oblique X-rays projections. Image B) 12 months X-rays follow- up.
Discussion

Typical locations of OO are the proximal meta-dyaphisis of the long bones in the lower limbs, the proximal femur being the most common site. There are few reported localizations to the fibula; in the series of 1666 cases of OO reported in the Rizzoli Hospital, only 47 were localized in the fibula [21]. We searched through Medline and EMBASE databases and found no case report of OO in the distal fibula. We think that the rarity of the localization and the therapeutic choice make this case of some interest, so we decided to report.Minimally invasive techniques are very commonly used in recent years. Many different approaches have been reported in literature and results are comparable to those of open surgery. The most commonly used procedures are CT-scan guided; the ablation

technique is generally radiofrequency thermoablation, preceded by K-wire drilling whenever the reactive bone was too sclerotic [22-24]. Tsoumakidou et al. report the use of Laser photocoagulation in the axial skeleton. Recent articles have described MRI guided techniques; Masciocchi et al. reported the use of Magnetic Resonance guided Focused Ultrasound Surgery (MRgFUS) in the treatment of OO in the appendicular skeleton, showing comparable result with the control group (treated with CT-guided radiofrequency thermoablation) [25]. Minimally invasive techniques have some important limits: the small dimension of the used needle could increase the risk of incomplete nidus removal; the necessity of multiple passages to completely remove the nidus increases the risk to damage important structures; previously thermoablated lesions are not eligible for MRgFUS; a complete histology is not achievable (most studies report no histologic examination). They have two main contraindications: the proximity of critical structures such as nerves, tendons, ligaments, tendon insertion points major than 1 cm shall be considered as an exclusion criterion; lesions located deeper than 1.2 cm from the bony surface are not eligible for MRgFUS because the distance would impede the penetration of the ultrasound beam [25]. Even though it is now demonstrated that less invasive procedures have comparable results with open surgery when performed in selected cases, we chose the latter in this case because of the deep location and the close proximity of peroneal artery, sural nerve and small saphenous vein. We chose en-bloc resection, instead of curettage, because it has proven to have a rate of relapse close to 0% [26]. Our patient experimented a complete remission of symptoms at a 12 months follow-up.In conclusion, even though OO has a preferential location in the proximal portion of lower limb long bones, it can be found in any segment. Minimally invasive procedures represent a promising and valuable treatment, but open surgery shall be taken in account when the location is not easy to reach and/or it is in proximity to critical structures.

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