



Osteoid osteoma

Panagotis KITSOULIS, George MANTELLOS, Marianna VLYCHOU

From the Laboratory of Anatomy and Orthopaedic Department, University of Ioannina, Greece

Osteoid osteoma is a relatively frequent benign bone tumour, consisting of osteoid and woven bone, and surrounded by a halo of reactive sclerotic bone, with an average size of the nidus less than 1.5 cm. It is a condition of late childhood, adolescence and young adult age. It usually occurs in the appendicular skeleton and the spine, and is generally localised in or near the cortex. The lesion causes pain, especially at night, but can cause joint pain with synovitis and joint effusion if located in the vicinity of chondral structures, or painful scoliosis if located in the spine. Osteoid osteoma may have an unpredictable course, and may require treatment or resolve spontaneously. In some cases, the diagnostic approach is challenging ; there are different treatment methods, some of which have been recently introduced, with promising results. We review the literature about the natural history, clinical presentation, diagnostic approach and classical or modern treatment modalities of osteoid osteoma.

Keywords : osteoid osteoma ; review.

INTRODUCTION

Osteoid osteoma is a condition of youth (16). Its size is small and it is not uncommonly located in an area where a plain radiograph can miss the lesion. It is painful especially at night and it is not unusual, in a young active individual, to have the symptoms for quite a long time, while the case remains undiagnosed.

Complete surgical excision, without complementary therapies, is the gold standard and remains

the treatment of choice for osteoid osteoma, but we shall discuss some new therapeutic procedures recently introduced. The pathophysiology, clinical presentation and step by step diagnostic procedure will also be presented.

EPIDEMIOLOGY

Osteoid osteoma accounts for about 5% of all bone tumours, and 11% of benign bone tumours (32). It mainly affects adolescents and young adults (6, 16). Seventy percent of the affected young individuals are under the age of 20, but it is very rare under the age of 5 or in adults older than 40 years (8). Osteoid osteoma more commonly affects males, with an approximate male/female ratio of 2 to 1 (32, 40). It is uncommon in blacks.

LOCALISATION

Osteoid osteoma predominantly occurs in the appendicular skeleton. It is rare in the skeleton of the trunk, with the exception of the spine (6). The

■ Panagiotis Kitsoulis, MD, Orthopaedic Surgeon.

Department of Anatomy-Histology-Embryology, Medical School, University of Ioannina, Greece.

■ George Mantellos, MD, Orthopaedic Surgeon.

■ Marianna Vlychou, MD, Radiologist.

Department of Radiology, Medical School, University of Thessalia, Greece.

Correspondence : Panagiotis Kitsoulis, 21th October 28 str, 45332, Ioannina, Greece. E-mail : pkitsoulis@hotmail.com.

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lower extremity is much more frequently affected than the upper extremity. In the lower limb, the femur and tibia are involved in approximately 50% to 60%, followed by the foot, where a predilection site is the neck of the talus. Common sites of femoral involvement are juxta- or intra-articular regions of the femoral neck. In the upper limb, the hand is more frequently affected, usually at the phalanges. The spine is involved in approximately 7% to 10% of patients, with the lesion usually occurring in the posterior elements (22). Bones with intramembranous formation are almost never affected. Osteoid osteoma is very rare in the skull.

There is a predilection for the cortex of the long bone shafts (6). The localisation can be diaphyseal, metaphyseal or, rarely, epiphyseal (40). In areas of cancellous bone it is localised in or near the cortex rather than in the center of the bone.

The occurrence of two osteoid osteomas in the same patient is extremely rare (6).

CLASSIFICATION

According to the Musculoskeletal Tumour Society staging system for benign tumours, osteoid osteoma is a stage 2 lesion, which requires intervention or, if markedly symptomatic, treatment.

Osteoid osteoma is classified as cortical, cancellous, or subperiosteal. Cortical lesions are the most common. The nidus is within the cortical bone, surrounded by cortical sclerosis and thickening or periosteal reaction and bone formation. Cancellous lesions exhibit minimal sclerosis around the nidus.

PATHOPHYSIOLOGY

The tumour's histological similarity to osteoblastoma supports the belief that it is a benign tumour derived from the osteoblasts. The nidus contains high local concentrations of prostaglandins, which are considered to generate intense peritumoral reaction and to be related to the pain (6, 38).

Osteoid osteoma is an ovoid or spherical and hyperaemic reddish tumour. Its reddish colour of the nidus is contrasting to the whitish colour of the surrounding bone. A packed mesh of thin and con-

torted osteoid and woven bone trabeculae is observed on a background of highly vascularised connecting tissue containing numerous dilated capillaries, active osteoblasts and osteoclasts. The osteoid tends to calcify toward the center of the nidus, which corresponds to the radiodense nucleus (6).

The surrounding host bone appears as reactive bone, while the degree of sclerosis varies. Reactive bone may sometimes be minimal or absent.

Usually, the central nidus is easily enucleated from its skeletal bed. When the nidus is superficial it may protrude at the surface of bone.

CLINICAL PRESENTATION

The character of pain can help in establishing the diagnosis of a benign bone tumour. The history of a dull aching pain for weeks to months, which is worse at night and is relieved by aspirin or non-steroidal anti-inflammatory drugs (NSAID'S), is so common with osteoid osteoma as to be nearly diagnostic (11, 18). The dramatic pain relief that may occur with the administration of NSAID'S supports the theory that prostaglandins have an important pathophysiologic role for the patients with osteoid osteoma (7, 21). When the physician elicits such a history, the suspicion of an osteoid osteoma must be strongly considered even if initial radiographs of the site fail to reveal an abnormality. Pain can be from moderate to intense, with a tendency to steadily increase with time, and is increased by consumption of alcohol and vasodilation. Pain is not always well localised by the patient, can be referred to an adjacent joint or can radiate.

A patient with an osteoid osteoma involving the hip may complain of a dull aching pain in the knee, relieved with non-steroidal anti-inflammatory drugs. The combination of a high index of suspicion for a small tumour and knowledge of patterns of referred pain should lead the physician to obtain a bone scan and/or radiographs of the hip when radiographs of the knee fail to reveal a cause for the patient's symptoms.

When the spine is involved, muscular spasm may cause painful scoliosis. The lesion is typically

at the apex of the convexity. When the case stays undiagnosed for a long time, scoliosis becomes postural. Deformity resolves with early resection, within 18 months, especially in children under 11 years of age. In cases where the osteoid osteoma is superficial, it may compress a nerve root. Definite neurologic abnormalities are seen in 6.5% of patients with spinal osteoid osteoma.

If localised near a joint, synovitis with joint effusion and limited range of movement can be observed. Osteoid osteoma must be included in the differential diagnosis of persistent unexplained knee pain, especially when objective findings of the knee are vague (13). Plain radiographs have a low diagnostic value in the detection of the lesion whereas isotope bone scan and MRI are reliable imaging techniques.

IMAGING

In most cases, the plain radiographs, combined with the characteristic clinical history, are all that is required to establish the correct diagnosis. At least two orthogonal views centered over the lesion should be obtained. There are some limitations; the nidus in spinal involvement may be difficult to detect in plain radiographs. Also, intra-articular lesions may be difficult to find in plain films because of the limited sclerosis around the nidus. Usually the nidus has a radiodense central core, surrounded by a regular halo of bone sclerosis. This appearance can change, depending on the affected site and on the stage of evolution. Secondary radiographic signs can be observed, such as regional osteoporosis, broadening of the femoral neck, angular deformity of long bones or scoliosis which is concave to the side of the lesion.

During the last two decades, computed tomography (CT) scans have proved useful in the diagnosis of osteoid osteoma (22) (figs 1, 3). CT remains the best imaging modality for the diagnosis of osteoid osteoma (1). CT is recommended for its better spatial resolution, in view of surgery (24). The imaging picture is characteristic with a small rounded to oval radiolucent area ("nidus"), surrounded by a regular halo of bone sclerosis. Within the nidus there may be a central and irregular nucleus of

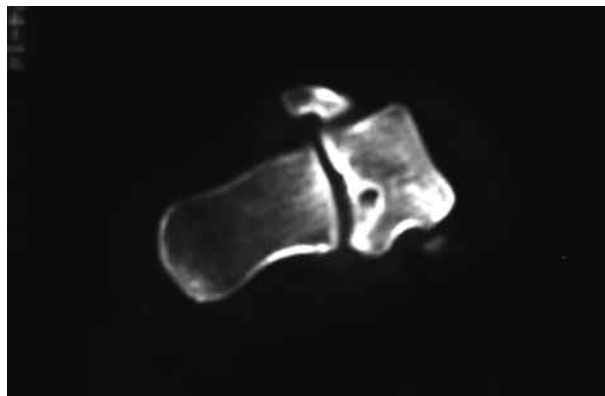


Fig. 1, 2. — CT and MRI scans of an osteoid osteoma located in the talus.

bony density, sometimes ring shaped (6). In cortical localisations there is fusiform thickening of the affected side of the shaft, with a smooth and regular surface, and homogeneous intense radiodensity. The nidus is located in the center of the thickening, always surrounded by the reactive bone sclerosis and almost always visible only with CT scan (6). If cancellous bone is affected, bone sclerosis surrounding the nidus is less remarkable. There are cases in cancellous bones where osteoid osteoma is protruding out of the bone due to minor periosteal



Fig. 3, 4. — Cortical localisation of osteoid osteoma in the diaphysis of the humerus.

reaction and bone formation around the nidus. CT may fail to diagnose osteoid osteoma when the nidus is in a cancellous location, due to the lack of perinidal density alteration (36).

MRI is inferior to CT scan in revealing the nidus, surrounding bone sclerosis and precise localisation of the tumour (figs 2, 4). MRI can be a highly valuable tool in bone tumour staging because it demonstrates cortical involvement and intramedullary and soft tissue spread (24). Also it is very good in detecting the nidus in cases of intra-articular lesions (2).

The role of technetium bone scanning in the evaluation of osteoid osteoma is to help define the precise location of the small pain-producing lesion in an area of complex anatomy (4). Also, in cases of

intra-articular osteoid osteoma, skeletal scintigraphy demonstrates both the lesion and the inflammatory involvement of the neighbouring soft tissue (2). The isotope bone scan is constantly positive, showing “double density sign”, consisting of a small rounded area of intense uptake centered in a more diffused halo (17). This examination may also be used to establish complete removal of the nidus.

TREATMENT

Complete surgical excision, without complementary therapies, is the treatment of choice for osteoid osteoma, with a low recurrence rate.

It is well known that osteoid osteoma may resolve spontaneously with time, so initial treatment consists of a trial of medical therapy using aspirin or nonsteroidal anti-inflammatory drugs. In a study of Kneisl *et al* (19), nine out of 24 patients were treated with non-steroidal anti-inflammatory medications, for an average of thirty three months (range, thirty to forty months). Six patients had resolution of the symptoms and remained asymptomatic for an average of 27 months after they discontinued taking the medication. Three patients were asymptomatic but still were taking medication after an average of 27 months. Fifteen out of 24 patients had operative treatment with complete relief of pain. The authors concluded that long-term administration of non-steroidal anti-inflammatory drugs can often be as effective as excision for the treatment of osteoid osteoma, without the morbidity that is associated with the operation, especially in patients in whom operative treatment would be complex or might lead to disability.

Despite these findings, a significant number of patients fail to continue with medical therapy because of intolerance to long-term administration of NSAID'S or because of the duration and severity of the pain and discomfort, pushing these young patients to a more aggressive therapeutic procedure. Possible complications and side effects after long-term therapies with non-steroidal anti-inflammatory drugs should be considered very carefully.

In most cases, when the patient keeps complaining of severe symptoms despite a trial of non-steroidal anti-inflammatory drugs, and the natural

history of the lesion has been explained to him, complete surgical excision is the classic treatment of choice.

The surgeon has to study CT scans preoperatively, in order to determine the surgical approach and the portion of bone to be resected (20). Intraoperative localisation of the nidus guided by computed tomography (CT) scan or intraoperative ^{99m}Tc bone imaging has been described (35, 39).

After exposure, the sclerotic bone covering the nidus is removed layer by layer with chisels. Sometimes, when removing reactive bone, one little vessel can be found. It is perforating the cortex from the periosteum to the nidus, and can be used as a guide for the surgeon to find the nidus (6). When exposed, the nidus appears as a punctiform reddish spot. It is well defined from the surrounding bone. Nidus removal can be wide, marginal or intralesional. When the technique is marginal or intralesional, the sclerotic bone is scraped out for a few millimetres by curettage (5, 6). The nidus should be preserved intact during removal. Campanacci *et al* (5) treated 89 patients by intralesional excision and eight by wide resection. They reported a rate of primary cure of 100%. Yildiz *et al* (42) treated 104 symptomatic patients operatively with either wide resection or curettage, and reported a success rate of 86.7% with an average follow-up of 2.5 years. In other series, incomplete nidus resection has been linked with local recurrence (14, 23, 25, 34). In a comparative study of Rosenthal *et al* (28), wide, aggressive resection had resulted in prolonged hospital stay, bone grafting, joint stiffness, and the possibility of postoperative fracture.

Burr-down technique requires less bone resection by progressively burring through the reactive bone to identify the nidus, which is then removed with curettes and burrs. The burr-down technique was associated with less postoperative immobilisation, a shorter duration of protected weight bearing, and an earlier return to activity (40).

Computed tomography-guided percutaneous resection of osteoid osteoma is another surgical technique. Sans *et al* reviewed 38 patients with a mean follow-up of 3.7 years and concluded that cure was obtained in 32 patients (84%) (31).

Donahue *et al* (10) reported only one minor complication and no recurrences occurred with a mean follow-up of 17 months, with computed tomography guided excision of osteoid osteoma in an outpatient setting, using an Einhorn biopsy set and a power drill. In another series of Roger *et al* (26), sixteen consecutive adolescent and adult patients underwent CT-guided percutaneous excision of a nidus with 14-gauge biopsy cutting needles, with local anaesthesia. The success rate was 87.5%.

New treatment modalities have been introduced during the last two decades, in order to reduce the potential morbidity seen with more traditional open techniques and to provide pain relief and early functional return, especially for tumours located in the vicinity of joint structures. The technique of percutaneous radiofrequency thermal ablation and percutaneous laser thermotherapy will be presented.

Percutaneous radiofrequency thermal ablation is based on the CT scan appearance of osteoid osteoma, its small size, and controlled thermal injury of bone (37). The probe is placed under CT guidance with the tip generating and sustaining a temperature of 90° C for 4 minutes. Rosenthal *et al* (27) described the technique in 1992. de Berg *et al* (9) and Rosenthal *et al* (29) reported a success rate of 94% and 88% respectively. Failures were related to suboptimal electrode positioning or short duration of heating. In a comparative study of open versus percutaneous radiofrequency ablation, there was a recurrence rate of 9% for open and 12% for percutaneous technique (28). No statistical difference in recurrence rates was shown. In the Barei *et al* (3) series, 10 out of 11 patients were treated with this technique. The authors suggested this technique for all patients with extraspinal osteoid osteomas that are not immediately adjacent to neurovascular structures (3). Recently published papers by Hadjipavlou *et al* (15) and Samaha *et al* (30), suggest that osteoid osteoma of the spine, adjacent to neural structures, can effectively and safely be treated by minimally invasive percutaneous CT guided radiofrequency coagulation.

Percutaneous interstitial laser thermotherapy with CT or MRI guidance is another minimally invasive procedure for the treatment of osteoid osteoma (12, 33, 41). With laser treatment the nidus is

destroyed through heating. Gangi *et al* (12) treated 15 patients with this technique ; a high-power semi-conductor diode laser (805 nm) with a 400-micron optical fiber was used. The fiber was introduced into the nidus through an 18-gauge needle. Around the fiber tip, well-defined coagulative necroses from 5 to 9 mm (energy delivery, 400-1.000 J) were obtained (12). The success rate was 95%. The smaller amount of bone destruction as compared with classic open surgery, allows for a short recovery time. Also the damage to cortical structures is minimal even if coagulated. Witt *et al* (41) using the same technique had about the same results.

Pain regresses immediately after surgery. If pain remains unchanged or there is only partial relief, this suggests that the nidus has not been excised or destroyed. If the pain returns after a period of a few months to a year, then the nidus has been subtotaly removed. The imaging techniques will reveal the presence of the nidus. Recurrence never occurs after removal or total destruction of the nidus.

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