

# Limping in twenty years old male A Case Report

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## ABSTRACT

**Background:** Osteoid osteoma(OO) is a relatively common benign skeletal neoplasm of unknown etiology that is composed of osteoid and woven bone, usually seen in adolescent and young males. Although, the clinical, radiological and scintigraphic features of OO have been well described, these features may be misleading or altered in the cases of lesser trochanter of the femur which is relatively uncommon location for OO with a few number of cases reported up to date.

**Case Presentation:** We report a case of a 20-year-old man who presented with painful limp. The pain had begun six months earlier and was made worse by walking and by exercise, with normal initial X-rays. The diagnosis was made after six months when typical Computed tomography, magnetic resonance imaging findings appeared, showing the nidus.

**Treatment:** A complete block resection accomplished successfully and the whole tumor with lesser trochanter of the femur in one mass was removed.

**Outcome and Follow-up:** the post-operative period was uneventful. The pathological examination is consistent with

osteoid osteoma. The patient was asymptomatic at one year follow up.

**Conclusions:** lesser trochanter OO can mimic lots of pathologic entities related to the affected area, and presents a diagnostic challenge and cause a delay in the diagnosis. The delays in the diagnosis and treatment can be avoided with a high index of suspicion. Treatment should be individualized according to the surgeon's preference and the characteristics of the case.

**Key words:** Osteoid osteoma OO, lesser trochanter, enblock excision

*Al-Kindy College Medical Journal 2014: Vol. 10 No. 2  
Page: 100-103*

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*Received 3rd May 2013, accepted in final 14 June 2013*

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Osteoid osteoma first described by Jaffe in 1935, is a benign osteoblastic tumor, mostly seen in adolescent and young males<sup>1</sup>. The lesions being more common in the lower extremity, children present with painful limping<sup>1,2,3</sup>. Conventional radiographs are effective in diagnosis, the radiological picture is of a central radiolucent area [nidus] surrounded by an area of cortical thickening. The imaging modality of choice is CT scan which is considered far superior to an MRI in diagnosis<sup>1,4</sup>. Surgical resection is the treatment of choice though spontaneous regression has been reported<sup>5</sup>. A high index of suspicion is helpful to make the proper diagnosis in painful limping in a child.

Theoretically, OO can involve any bone, however there is a site predilection for appendicular skeleton, and more than 50% of the cases occur in the tibia and femur<sup>5</sup>. OO is categorized into three types based on the radiographic localization of the nidus; cortical OO, cancellous OO (also referred to as medullary) and subperiosteal OO. Cancellous and subperiosteal OOs typically arise in an intra-articular or juxta-articular location<sup>6</sup>.

The essential point in the radiological diagnosis is the identification of central calcifications surrounded by ovoid translucency. The average diameter of the nidus is between 5mm to 1.5 cm. If the nidus is more than 2cm it belongs to the category of osteoblastoma. Intracortical osteoid osteoma produces dense sclerosis around the nidus. Subperiosteal type produces periosteal reaction. Subarticular osteoid osteoma simulates arthritis as it produces synovial reactions.

**Case report.** A 20-year-old man was referred to our clinic with a history of left hip pain for duration of six months. Three months ago, he had admitted to another medical center with the same complaints. Conventional pelvis radiograph was taken, however, it revealed no abnormal findings (Figure 1). The physician thought about the

avascular necrosis of the femoral head and ordered hip joint MRI as next step diagnostic work-up. MRI showed nonspecific finding. Consequently, the patient was treated with antibiotics. After 15 days of antibiotic therapy with ceftriaxone, no improvement observed in patient clinical picture.



**Figure 1.** Pelvis antero-posterior x-ray of the patient on initial admission. No abnormal finding was observed.



Figure 2: CT scan Images on coronal and axial planes.

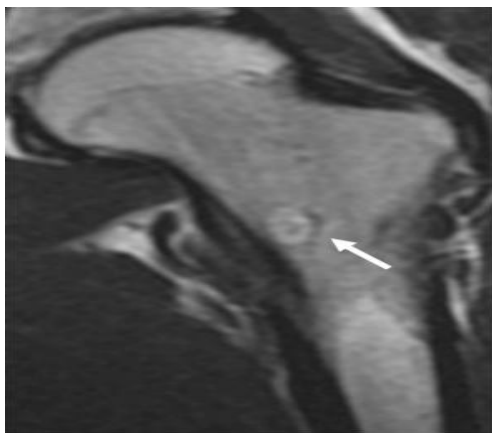


Figure 3: MRI femur showing the nidus in osteoid osteoma

The patient consult our clinic for further evaluation. His initial symptoms were worsened over several weeks, and he reported resting pain at the presentation. Physical examination revealed reduced left hip range of movements in all planes, with end-range pain in all directions especially flexion and external rotation . There was 3 cm atrophy of the left thigh compared to the right and grade 4/5 power of the left hip . Neuro-vascular examination was normal. Routine laboratory studies, including a complete blood count, erythrocyte sedimentation rate (ESR), alkaline phosphatase, blood urea, calcium, creatinine, glucose serum, and uric acid were all within normal limits. C-reactive protein (CRP) was slightly elevated upper limit. Based on these clinical and imaging findings, other diagnoses including inflammatory arthritis, specific bacterial infections such as tuberculosis and brucellosis were also investigated, Rose Bengal test, all yielded negative results. No pathologic correlation of a specific infection or a neoplastic lesion was established. Electromyography and lumbosacral MRI revealed mild lumbar nerve root compression and multi-level disc bulging. Therefore, patient received a physical therapy and rehabilitation . CT scan was performed and showed a focal area of increased uptake in the medial aspect of the left lesser trochanter of the femur (figure 2) also MRI reveals the lesion clearly (figure 3) . Open surgical excision of the lesion was performed by medial approach to the hip. Subperiosteal dissection of the iliopsoas tendon from the lesser trochanter ,the lesion was completely excised with an adjacent normal

bone from the lesser trochanter and tenodesis of the iliopsoas tendon with remaining bone, the defect was filled with hydroxyapatite granules.

The postoperative course was uneventful, drain removed on second post-operative day and the stitches of the wound removed on the 14<sup>th</sup> day, no infection occurred and the patient stayed non weight bearing on the left leg for 3 months.

The pathological examination confirmed the diagnosis of osteoid osteoma. Immediately after the recovery period 1 month after the operation the hip pain disappeared. At the final follow-up 12 months after the surgical excision, the patient was still free of pain with full-range of hip movements, and ambulating unaided.

**Discussion.** There are many causes of painful limping including infection, trauma and tumor<sup>2,7</sup> Osteoid osteoma, a benign osteoblastic tumour usually less than 15mm in size. It is most commonly seen in the long tubular bones of the lower extremities of patients in the second and third decade<sup>1&5</sup>, can present as limping. Pain is the predominant symptom and worse at night. Lesions around the proximal femur can present with painful limping<sup>1,2,3</sup> and wasting of the thigh muscles. Most of the cases of osteoid osteoma in the proximal femur have nonspecific symptoms. Dramatic relief with salicylates is documented.<sup>1,3,7,8</sup> Salicylates can accelerate healing in osteoid osteoma and is a good diagnostic and therapeutic consideration<sup>8</sup>. Typical radiographic appearance is a round lucent nidus with surrounding reactive sclerosis. The nidus on maturity may be radio dense due to mineralization. The nidus is better demonstrated in a CT scan than in an MRI<sup>1,4</sup> CT scanning with 2 mm cuts delineates the nidus properly<sup>8</sup>

The presence of area of sclerosis around the lucent area, due to reactive bone formation is typical of osteoid osteoma.<sup>1</sup> The nidus is the area of vascular osteoid tissue, and typically measures less than 10 mm. In children, there is extensive periosteal reaction around the nidus.<sup>4</sup> The pathophysiology of the characteristic pain of osteoid osteoma is becoming better understood. Immunohistochemical studies, prompted by the clinical response to nonsteroidal anti-inflammatory drugs, have shown that osteoblasts within osteoid osteomas

contain prostaglandins<sup>9,10</sup>. Similar results have been found in other benign and malignant bone tumors, such as fibrous dysplasia and osteosarcoma, however, so prostaglandins alone probably do not account for the distinctive pain profile of osteoid osteoma. Additional immunohistochemical studies in which antibodies are used against nerve fibers have shown that osteoid osteomas have an increased density of nerve fibers, particularly in the fibrovascular zone, as compared with other benign and malignant bone neoplasms, including osteoblastomas.<sup>11</sup> Therefore, the pain of osteoid osteomas may result from an interaction between prostaglandins and nerve fibers. The high levels of prostaglandins within the nidus and the unmyelinated nerve fibers identified in the fibrous zone around the central nidus or in the nidus itself are believed to be the cause of pain<sup>5</sup>. This is the reason why NSAID's can dramatically relieve the pain. On the other hand, the classical nocturnal pain found in around 80% of extra-articular OOs is absent and the pain is less responsive to salicylates when the lesion is intra-articular<sup>5,12</sup>. Radiographs in nondiaphyseal osteoid osteomas may be misleading and the tumor may be missed in most of the cases.<sup>13</sup> In a small series by Davidson et al there was a delay of six months in making the diagnosis because of the atypical radiographic appearance.<sup>13</sup> The delay in diagnosis can range from six months to two years.<sup>2</sup> There was delay in diagnosis of our case as there was no reactive bone in the area of the lesser trochanter. Non diaphyseal lesions do not produce the same amount of new bone as in diaphyseal lesions<sup>13</sup> which is associated with more marked reactive formation compared to medullary and subperiosteal osteoid osteomas. Nidus in cancellous bone maybe difficult to visualize as there is less periosteal reaction and new bone formation.<sup>1</sup>

The natural history of osteoid osteoma differs from that of most other skeletal tumors in that osteoid osteoma does not grow appreciably. This is an important distinction from osteoblastoma, another benign bone tumor that may have a similar appearance.<sup>14</sup>

Fine-cut CT imaging is the best method of identifying the nidus when the lesion is small or not apparent on plain X-ray particularly in the spine, pelvis and femoral neck or in intra-cortical lesions<sup>6, 15</sup>. However, formation of the nidus may take a longer time in cancellous bone, thus CT may not provide pathognomonic findings. On the other hand, the excellence in soft tissue imaging makes MRI an alternative radiographic method for evaluating joint disorders and demonstrates the tumor nidus of the intra-articular OO.<sup>16</sup> The sclerotic bone surrounding the nidus demonstrates low signal intensity on T1- and T2-weighted images.

Technetium bone scanning has high sensibility but low specificity<sup>15</sup>. It commonly shows a double-density sign with increased uptake centrally that distinguishes the lesion from osteomyelitis or abscess. This sign is characterized by a focal hot spot of the nidus and an area of low peripheral radionuclide accumulation around it, which is related to the sclerotic bone.<sup>17</sup>

Osteoid osteoma tends to regress over a period of time even without treatment.<sup>8</sup> The standard treatment of osteoid osteoma is en bloc resection, though other modalities of treatment including radio ablation have been practiced<sup>1</sup>. Because of the nonspecific symptoms and the lack of characteristic radiological features of OO of the lesser trochanter and in intra-articular lesion, there are up to 2.5-3.5 years delays in the diagnosis and treatment in the published cases<sup>5</sup>. In our case, the patient's diagnostic process took almost 6 months after the first onset of complaints. In this delaying and challenging diagnostic pathway, the pathologic entities that can cause similar symptoms and signs should

be taken under consideration in differential diagnosis. The differential diagnosis of a small lytic lesion in the cortex includes infection<sup>18</sup> several benign neoplasms including osteoid osteoma, chondroblastoma, eosinophilic granuloma, hemangioma, periosteal chondroma, and the very rare cancer, intracortical osteosarcoma. The lytic lesions caused by infection are usually larger and more irregular in shape than this one. Chondroblastoma may be indistinguishable from an osteoid osteoma on imaging. However, in most cases, chondroblastoma is found in the epiphysis or apophysis of the bone rather than in the shaft. It has a lobulated, rather than a rounded, boundary and is usually larger than this lesion. Similarly, eosinophilic granuloma is rarely this small at presentation, is sometimes multifocal, and often exhibits periosteal chondroma usually sits on the surface of the bone, producing a superficial indentation that has been described as similar to an egg in a cup. In very rare instances, a chondroma may be purely intracortical and may resemble an osteoid osteoma. Cortical hemangiomas may also closely simulate osteoid osteomas, but they are usually less painful. Osteoid osteoma is usually a small spherical or oval lytic lesion (1 mm to 15 mm), surrounded by a variable amount of reactive bone and soft-tissue edema. Reactive bone may take the form of medullary sclerosis or periosteal new bone. Osteoid osteomas are considerably more common than the other entities mentioned.<sup>19</sup>

These include acute, subacute and chronic inflammatory arthritides like septic or tuberculous arthritis, gout, rheumatoid arthritis,<sup>5</sup> Brodie's abscess and intracortical chondroblastoma<sup>5</sup>. Painful limp in a child is a serious concern for the parents and clinicians. Osteoid osteoma should be considered in the differential diagnosis even though the plain X rays are normal and a CT scan should be considered. Complete excision alleviates the symptoms.

In conclusion, there are significant differences in the symptoms and imaging features of lesser trochanter OO which can mimic lots of pathologic entities, that presents a diagnostic challenge and cause a delay in the diagnosis. As in our case, in the initial phases of the disease, the imaging features may not correlate with the symptoms and a specific finding may not be distinctive. The delays in the diagnosis and treatment may be avoided with a high index of suspicion and patience of the surgeon. The CT is a useful imaging modality to demonstrate an OO.

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