LATE DIAGNOSIS OF OSTEOID OSTEOMA IN ADOLESCENCE: A CASE REPORT

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[Diagnosi tardiva di osteoma osteoide in adolescenza: un case report]

ABSTRACT

Osteoid osteoma (OO) is a benign osteoblastic tumor commonly located in the long tubular bones of lower extremities.

We present an illustrative case of an unusual location of osteoid osteoma in the left anterior acetabolar wall in a 16-years-old male child initially treated for chronic radicular pain and subsequently for psychosomatic pain leading to a delay of one year in diagnosis.

Key words: Osteoid osteoma, bone tumor, night pain, adolescence, diagnostic delay.

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Introduction

Osteoid osteoma (OO) is a benign bone tumor that Bergstrand first described in 1930[(1)]. In 1935 Jaffe described it as a unique entity[(2)].

It is commonly located in the long tubular bones of lower limbs and it is an infrequent, but important, cause of musculoskeletal pain[(3)].

This tumor occurs in children and young adults, and since the clinical history may be difficult to elicit, frequent problems in early diagnosis can be found.

OO may have an unpredictable course, may require treatment or resolve spontaneously.

Symptoms of OO can last from weeks to years before diagnosis and eventual surgery. Pain is the principal symptom of both initial and recurrent disease.

The imaging studies (Radiography, Radionuclide scanning, CT scanning) are helpful in diagnosis. Initial treatment of OO remains non-operative, with medications consisting of aspirin or other non-steroidal anti-inflammatory drugs (NSAIDs). Surgical intervention, or CT-guided RFA, are generally indicated for patients whose pain is unresponsive to medical therapy[(4)].

Here we report an illustrative case of OO of the left anterior acetabolar wall in a 16-years-old male child initially treated for chronic radiculitis and subsequently for psychosomatic pain that masqueraded the osteoma leading to in one year diagnosis delay.

Case report

A 16-years-old young male patient presented with a persistent nagging pain with a duration of one year, located in the anterior left thigh and associated with ipsilateral lower limb hyposthenia. Lately, the symptoms worsened leading to several night awakenings due to the nocturnal pain, and responding to the analgesic medicament (paracetamol).

During the last year, the patient was admitted in a peripheral hospital where a diagnostic lumbosacral magnetic resonance imaging (MRI) scan was performed. It showed a mild disc bulge at L3/4 and L4/5; moreover an abdominal MRI scan excluded pelvic masses, and an electromyography of lower limbs showed signs related to a L4-L5 left chronic radiculitis. In addition, an ultrasound scan of the thigh and the X-ray of hip joint and femur

didn't reveal any abnormality. Subsequently he underwent repeated cycles of physical therapy without reasonable effects.

At the end of these investigations the neurologist prescribed an antidepressant treatment (amitriptyline) for suspected psychosomatic illness.

When he was admitted to our department he was complaining of pain. The physical examination was normal, although the referred hyposthenia, not neurological alterations were found. Routine blood tests were all normal too.

Considering characteristics and persistence of pain, we put the previous diagnoses up for discussion and decided for a femur computerized tomography (CT) scan which showed an oval mass of 8x7 mm in the left anterior acetabular wall: an hypodense area with a central hyperdense nidus that can be related with OO.

The patient was referred to a specialistic center to undergo CT-guided radiofrequency ablation (RFA).

No complications were observed during or immediately after the procedure. The patient was able to place weight on his left leg after 7 days and he started to walk after 15 days. Three months after the ablation, the patient still did not show signs of pain.

Discussion

Osteoid osteoma (OO) is a benign bone tumor most commonly located in the long tubular bones of lower extremities, and is an infrequent, but important, cause of musculoskeletal pain[3].

The femur, particularly the intertrochanteric intracapsular regions of the hip, is affected in two thirds of cases[5]. The diaphyseal part of the tibia and the humerus are other common sites.

It is rare in the trunk skeleton, with the exception of the spine[6]. Involvement here most commonly manifests as painfull scoliosis, but painless conditions can also occur.

Pettine et al noted that 50% of the lesions occur in the cervical spine and up to 78% of OO in the lumbar spine are associated with scoliosis[7]). Other location involved include the hand, talus, foot, and joints[8, 9].

OO accounts for about 11% of benign bone tumors; 70% of the affected young individuals are under the age of 20, and it is very rare under the age of 5 or in adults older than 40 years[6, 10]. OO incidence rate in males is higher than in females, with a male/female ratio of 2:1[11, 12]. Most patients present with pain, which worsens at night and is relieved by non-steroidal anti-inflammatory drugs (NSAIDs)[11]. This supports the theory that prostaglandins have an important pathophysiologic role for these patients[13]. The pain is described as continuouos, deep, aching, and intense with varying quality and severity. It is typically localized to the site of the lesion.

The 6.5% of patients present with neurological disorders with spinal OO: the osteoma of the hip may cause a reflective pain miming the hurt resulting from spinal disc hernia compressing nerve root.

When OO patients present with radicular pain and neurological signs as atrophy, weakness and diminished deep tendon reflexes in the affected limb, this can simulate lumbar spinal disease resulting in unnecessary neuroradiological investigations. In our case report, the evidence of a minimal disc protrusion provided by MRI, initially addressed to a chronic radicular pain, entailing the diagnosis of OO, undergoing the patient to a treatment failure.

The radiological evaluations are fundamental for the diagnosis in addition to the relevant clinical information. Bone scanning dramatically decreased the mean interval from the appearance of symptoms to diagnosis from months to 12 months[7].

OO elicits a profound osteoblastic response in surrounding medullary and cortical bone and shows the characteristic sclerosis around a lucent nidus framework. The radiographic findings may be characteristic including a cortical based central nidus of vascular osteoid tissue surrounded by reactive sclerotic bone. Occasionally, the radiographic pictures is negative, contributing to the delay in diagnosis[11]. In these cases, the CT scan is the best imaging modality for the identification of OO[6, 14].

The four diagnostic criteria to evaluate the lesion are:

- rounded /oval with neat boders
- less than 2 cm in diameter
- a dense and homogeneous central area
- a radiolucent circumferential of 1-2 mm

CT is more accurate than MRI. Sans et al reported that CT helped in confirming the diagnosis of OO in 74% of cases[15]. Radionuclide scans are reliable tools when radiographic findings are not diagnostic. Wells et al urged that the bone scans be performed when radiographic are normal or inconclusive, especially in pediatric patients[16].

The radiological differential diagnosis includes osteoblastoma, osteomyelitis, arthritis, fracture due to physical efforts, and enostosis. Cases of sponta-

neous regression, with an unknown mechanism, are described after a median time of 3-7 years[17]. The tumor infarction may be a possible hypothesis.

A rare complication of OO is a localized overgrowth and a deformity of bone as, for example, the scoliosis in presence of vertebral lesion[18].

Initial treatment of OO remains non-operative, with medications consisting of aspirin or other NSAIDs. The response to salicylates is not universal, however. Surgical intervention is generally indicated for patients whose pain is unresponsive to medical therapy[4]. Complete surgical excision without complementary therapies, is the treatment of choice for OO, with a low recurrence rate. Pettine et al added that the most important determinant for successful surgical removal is the exact localization of lesion[7]. New treatment modalities have been introduced during the last two decades, in order to reduce the potential morbidity seen with more traditional opened techniques and to provide pain relief and early functional return[6].

Radiofrequency ablation for the OO treatment was first described in 1992, and is currently the choice therapeutic treatment in both adult and pediatric cases; it is minimally invasive, safe and has a high technical and clinical success rate. The literature reports a therapeutic success rate range of 88-96%.

Possible side effects of RFA technique include adjacent tissues failure and/or treatment failure with consequent relapsing[17].

In conclusion, the articular pain in adolescence is often correlated to the physical activity or orthopaedic problems, but in few cases only, it hides a more important cause (i.e. chronic rheumatic pathology, lymphoproliferatives diseases, benign or malignant bone tumors).

However, more attention should be paid to the nocturnal character of limb pain, that is a strong clue of bone localization and nature of tumour.

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