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The rough walking: an interesting symptom of myositis ossificans traumatica in a patient with Missouri syndrome; succint review

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ABSTRACT

Heterotopic ossification (HO) is the formation of lamellar bone inside soft-tissue structures where bone normally does not exist. It is not an inflammatory process, but a reactive and benign response to an injury. We can distinguish three different types: myositis ossificans circumscripta, myositis ossificans progressiva and localized myositis ossificans traumatica (MOT).

MOT, which is more frequent in men than in women, can occur at any site, but most frequently in the hip following total hip arthroplasty. However, this is also the most common site of involvement in patients with a traumatic brain or spinal cord injury, and has also been described after an avulsion fracture, and also has been reported as associated with contact sports or those at risk of repetitive trauma.

Key words: **Calcifications, osteoarthritis, traumatisms, Missouri syndrome.**

INTRODUCTION

Heterotopic ossification (HO) is the formation of lamellar bone inside soft-tissue structures where bone normally does not exist. It is not an inflammatory process, but it is a reactive and benign response to an injury. We can distinguish three different types: myositis ossificans circumscripta, myositis ossificans progressiva and localized myositis ossificans traumatica (MOT).

We report a slightly symptomatic case of MOT with more than 20 years of evolution, accompanied with a brief review of the scientific literature. In addition, our patient was also diagnosed with Missouri syndrome, which makes this case report even more interesting because of its peculiarity.

CASE REPORT

A 56-year old man presented to our outpatient clinic with a story of more than four years of pain in the small joints of his hands (especially at metacarpal location), accompanied sometimes with morning stiffness lasting for less than 30 minutes. He was a healthy individual otherwise and the family history was irrelevant. Now working in a metallic carpentry for more than 30 years, he used to be a semi-professional soccer goalkeeper player and had suffered a fracture of his left wrist and tibia plus fibula.

During the physical examination the hands showed an aspect of severe osteoarthritis on his second and third metacarpophalageal joints, but with some synovitis sensation in his wrists; nevertheless the range of movement was complete, and Fabere test was normal and not painful. He did not present pain elsewhere, though the walk was slightly rough; he admitted to have been walking like that since he was young. Blood test and radiological studies were requested. Erythrocyte sedimentation rate (ESR), C-reactive protein (CRP), rheumatoid factor (RF), anti-cyclic citrullinated peptide (anti-CCP) antibodies, antinuclear antibodies (ANA) and HLA-B27 were all normal or negative. By ultrasound it was observed a slight quantity of synovitis affecting metacarpal joints but without Doppler signal; we also found images consistent with osteophytes, which were confirmed in the X-ray (figure 1). However, and in spite of not finding chondrocalcinosis images anywhere, the hip X-ray showed gross calcifications affecting the pelvis, mainly surrounding the joints (figure 2).

Reinterrogating the patient, he admitted to have suffered repetitive traumatisms on his hips, due to his activity as a goalkeeper, but never had asked for medical attention as within some days of rest and anti-inflammatory drugs (NSAIDs) he used to recover promptly. He denied any remaining local pain, and we again confirmed a complete range of motion on his lower limbs (hips and knees). Study of calcium, iron and thyroid metabolism was normal, with which we could exclude hyperparathyroidism and hemochromatosis as a cause of secondary osteoarthritis. No other studies were taken, since the patient was asymptomatic.

Finally the patient was discharged with the diagnosis of Osteoarthritis in his second and third metacarpal joints (Missouri syndrome)[1] and myositis ossificans traumatica (MOT)[2] secondary to a repetitive soccer player injury on his hips. We just prescribed NSAIDs and pain relievers if needed for the symptoms in his hands.

FIGURE 1

HAND X-RAY: IMAGES CONSISTENT WITH OSTEOPHYTES

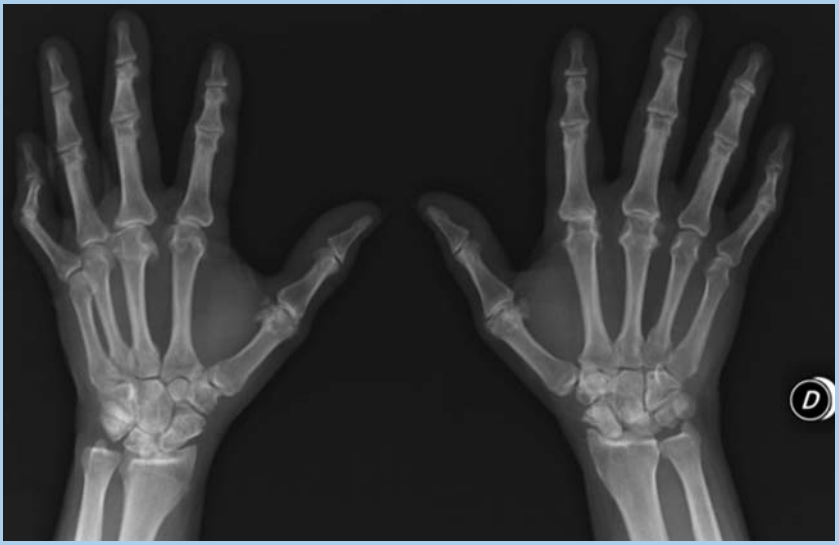
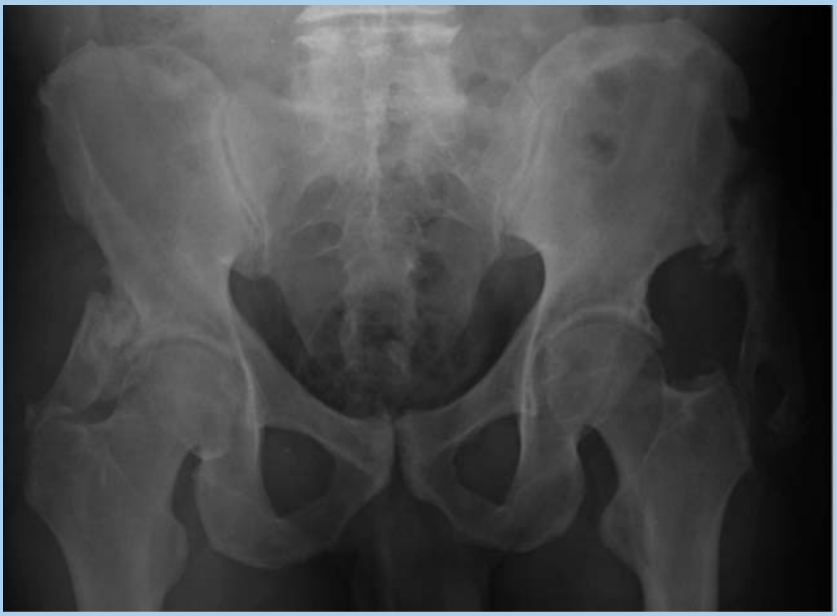


FIGURE 2

HIP X-RAY: GROSS CALCIFICATIONS AFFECTING THE PELVIS, MAINLY SURROUNDING THE JOINTS



DISCUSSION

MOT, which is more frequent in men than in women, can occur at any site, but most frequently in the hip following total hip arthroplasty. However, this is also the most common site of involvement in patients with a traumatic brain or spinal cord injury, and has also been described after an avulsion fracture (quite frequent between adolescents, young adults, athletes and ballet dancers), as a consequence of an isolated contusion or a repeated one[3]; hence, we should not miss this diagnosis in athletes. The most frequently affected muscles are rectus femoris, the medial head of grastrocnemius, ischiotibial muscles, adductors and, less frequently, arm and forearms muscles[4]. This phenomenon has also been described in people affected with Ankylosing Spondylitis, Forrestier disease, burns and some hereditary illnesses[5].

The etiology of HO remains still unknown[6]. Nevertheless, plenty of studies have been done, where a genetic predisposition has not yet been established. HO has been described after some usual vaccines (diphteria tetanus pertussis) in children with fibrodysplasia ossificans progressiva. Local hemorrhage and a history of significant local injury might be some important precipitant factors in the occurrence of HO.

The clinical signs and symptoms can develop form 3 to 12 weeks after a musculoskeletal injury, spinal cord injury or another related factor. As the initial symptoms we can find increased joint stiffness (which may be accompanied by local pain), limited range of motion, warmth, swelling and erythema, which often makes it difficult to distinguish from deep venous thrombosis (DVT); they can even both coexist after some processes. Even rare, some complications have been described, such as DVT, nerve or vascular compression, spasticity and lymphatic obstruction leading to lymphedema[7].

Histologically HO cannot be differentiated from callus formation of a healing fracture. The onset of the ossification process lies in fibroblastic metaplasia: there is a well-delineated zone of fibroblastic proliferation, followed by chondroblasts and eventually osteoblasts with blood vessels and Haversian canals. The new bone may be contiguous with the skeleton, but does not involve periostium. Mature HO shows cancellous bone and mature lamellar bone with blood vessels and bone marrow, with only a small amount of hematopoiesis. So, at the beginning, we should not mislead the diagnosis with a highly differentiated sarcoma or osteosarcoma (if there is a lot of osteoid). Using a biopsy we can find what Akerman[8] described in 1958, the "zonal phenomenon": beginning in the second week to second month, bone is formed surrounding the injuried area and then moves inward toward the site of injury giving layers of varying tissue activity; this way, the outside layer is bone, the middle layer is an area of extensive osteoblastic and fibroblastic activity,

and the central layer is an area of highly cellular tissue with a lot of cell differentiation and necrotic cells (not matured bone, which does not exist in the ostoesarcoma). Other reasonable differential diagnosis include extraoseous sarcoma, synovial osteosarcoma, osteochondroma, posttraumatic periostitis, osteomyelitis and tumoral calcinosis[9].

To confirm the diagnosis[6], at the beginning of the process, the most sensitive imaging technique is the three-phase bone scintigraphy, we can also use ultrasonography (US), computer tomography scan (CT) and magnetic resonance imaging (MRI). Prostaglandin E2 (PGE2) excretion in 24-hour urine[6], though unspecific, is felt to be a reliable bone marker, both for early detection and to confirm treatment efficacy. Plain X-ray studies are useless initially, since it is after several weeks when we can find some diagnostic images.

The most accurate treatment[6] is not clear jet, thus prevention has been highly promoted. Kinesitherapy, manipulation under anesthesia and localized shock waves might help. In addition, it has been highly recommended the protection of the athlete from recurrent injury by restriction of its activity and, as much as possible, using an extra padding over the affected area (from 3 to 6 months). No clinical trials have been taken to prove any specific treatment, just small case series have been published. Here we mention the most frequent tried therapies.

Indomethacin (50-75 mg/day for at least 3 weeks) and COX-2 inhibitors have sown a reduction in both the early and late diagnosis[10]; the former has a two-fold action: it directly inhibits the differentiation of mesenchymal cells into osteogenic cells, and indirectly by suppressing the prostaglandis-mediated remodelling in a post-traumatic bone.

Not in use for several years, etidronate[11] (300 mg/iv x 3 days + 20 mg/kg/day orally for about 6 months) used early and maintained at high dose for a prolonged time significantly reduces this situation, since it blocks the aggregation, growth and mineralization of calcium hydroxyapatite crystals in 95% of the cases[12]. However, it was just recommended for the inflammatory phase, because of its secondary effects and because the effect disappears when the treatment is stopped[6].

Warfarin at therapeutic dose (or even to keep the prothrombine time between 1 and 1.5) has been tried in patients with medullar lesions, in which it seems to exist a hipercoagulability state; this situation might be the link with HO in this group of patients. Nevertheless, without definitive results[13,14]. The reason of having tried warfarin as a preventive treatment for HO is explained with osteocalcin; this protein is essential to build the bone matrix, and during its make up process it suffers a post-translational vitamin K-dependent carboxylation reaction. As an anti-vitamin K, warfarin limits the osteocalcin proper production.

Radiation therapy has also been tried, since irradiation interferes with the differentiation of pluripotential mesenchymal cells into osteoblasts and suppresses the tissue inflammation combined with the ablation of pain receptors[15].

Finally, in case of severe functional impairment or intractable pain, surgical removal[6] can be considered, but always after at least 6-12 months of evolution, to avoid its reproduction.

In conclusión, myositis ossificans traumatica (MOT) is a relatively common injury associated with contact sports or those at risk of repetitive trauma, though short series of cases are the usual findings when searching in the literature. This process needs to be kept in mind after local injuries, especially when the patient complains of joint stiffness, limited range of motion, warmth, swelling and erythema. However, final calcification lesions would remain for god.

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