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Case Report Open Access

Transient Osteoporosis of the Hip in Non-Pregnant Woman

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Abstract

Introduction: Transient osteoporosis of the hip is an uncommon condition characterized by pain in the hip and pronounced osteopenia of the femoral head and neck. TOH is a self- limiting condition typically associated with the third trimester of the pregnancy. The authors present a case of transient osteoporosis of the hip that was complicated by a pathologic neck fracture.

Case Report: A 26-year-old woman, non-pregnant, was present with a history of right hip pain after minor trauma. Radiographs showed fracture of right neck femur and a severe osteopenia. She underwent closed reduction and internal fixation of the right hip without complications. In follow-up at 3 months, the patients referred hip pain and the radiography showed non-union fracture. Before excluded diagnosis of oncologic disease, she was submitted a new surgery. In follow up at 4 years, the hip pain and limited mobility in right hip was persisting.

Discussion: Although pathological fractures are rare, they are the most serious consequence of TOH. In most cases this condition is self-limited with a completed recovery. This case is very rare because the hip fracture as consequence of TOH is uncommon, it is a no pregnant woman and the patient didn´t have complete recovery.

Abbreviations : TOH: Transient Osteoporosis of the Hip; BMI: Body Mass Index; CT: Computed Tomography Scan

Manuscript

Introduction

Transient osteoporosis of the hip (TOH) was described for the first time in 1959 by Curtis and Kinkade, they presented three cases of transient osteoporosis of the hip in women on the third trimester of pregnancy [1,2]. TOH is a rare disease of unknown etiology. It mainly affects pregnant women on the third trimester and middle-aged men. It rarely affects children and non-pregnant women. Its evolution is usually benign, being the hip fracture the most serious complication. The authors present a clinical case of TOH in a non-pregnant woman, complicated with hip fracture.

Clinical Case

Female patient, 26 years old, caucasian, with a body mass index (BMI) of 37.7. She had a vaginal delivery two years ago and breastfed for the first 3 months. Nine months after delivery she was submitted to a gastric band placement. No history of chronic medication. The patient was admitted to the Emergency Room with complaints of severe pain and functional impotence of the right hip after a minor trauma. On examination she presented pain and shortening of the right leg. The X-ray of the pelvis showed fracture of the right femoral neck with pathological characteristics (Figure 1). The Axial Computed Tomography Scan (CT) revealed T2 hypersignal in the femoral, and acetabular region. The blood tests and Bone Scintigraphy did not show relief changes. At Magnetic Resonance Imaging (MRI) was observed in the femoral region bone marrow infiltration. Bone biopsy was performed and revealed osteomalacia without neoplastic cells (Figure 2).

Hormone assays, bone densitometry, parathyroid scintigraphy, head, neck and chest CT scan, none revealed any changes. Two weeks after the fracture diagnosis a closed reduction and internal

fixation with an intramedullary rod (UFN Spiral Blade) was performed (Figure 3). The patient was discharged hospital on 4th postoperative day medicated with bisphosphonates and oral calcium supplement. At the follow up 3Th months after surgery, she resented a walking disability with radiographic signs of osteosynthesis failure. CT scan demonstrated extensive bone rarefaction to the entire right femur region, with evidence of extrusion of the femoral neck prosthesis, without signs of fracture onsolidation. In MRI having been observed cellular infiltrate and adenopathy of the lymphatic chains. The hypothesis of cancer was again placing. In this context, the bone biopsy was repeated but the result of which was concordant with the previous one: osteoporosis with exclusion of mphoproliferative disease, Gohram-Stout Dengue, infections, primary tumors or metastases. Given the extension of the lesion, the patient was submitted to a Total Hip Arthroplasty with prosthesis (Reef®), together with appropriate medical treatment (Figure 4). The histology of the femoral head was osteoporosis, excluding Gohram-Stout. Four years after surgery, the patient still had pain and limited mobility of the hip with signs of prosthesis integration.

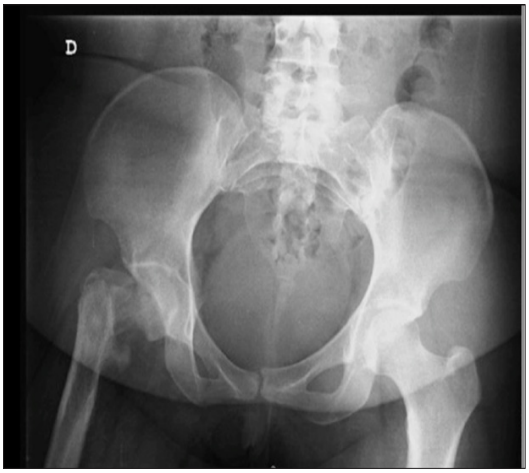

Figure 1: X- Ray Shows Osteosporosis of Right Hip and Right Femur Fracture.

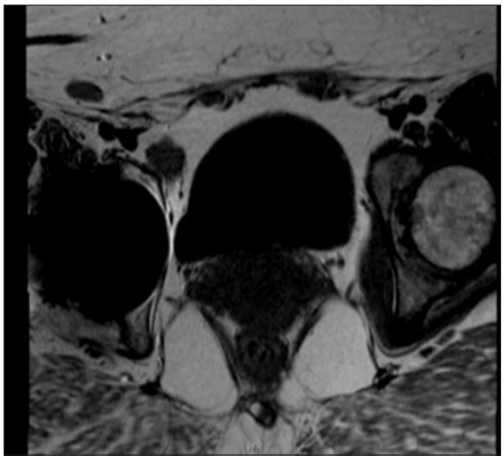

Figure 2: MRI of the Hip is Observed Hyperintensity of Right Femur Head with Cell Infiltration.

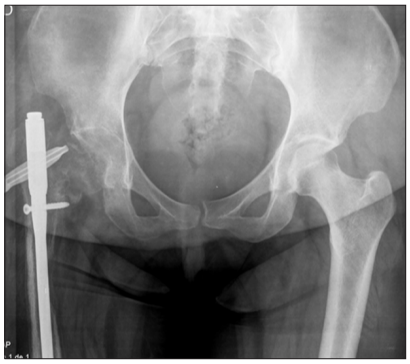

Figure 3: X ray - Intramedullary Rod (UFN Spiral Blade), Osteopenia in Right Femur.

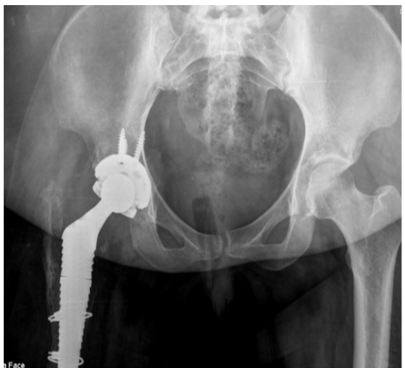

Figure 4: Total Hip Arthroplasty with Prosthesis (Reef®).

Discussion

Although TOH was first described more than half a century ago, its etiology remains unknown [3]. However, different theories have been presented, but none is proven. One of the most frequently mentioned is the Ischemic Osteonecrosis due to venous congestion associated with a stage of hypercoagulability [4,5]. Other researchers argue that THO and the Sympathetic-Reflex Dystrophy are the result of same neurogenic process [6]. Pregnancy is the only recognized precipitating factor [7,8]. The pathogenesis of THO in pregnant women is difficult to understand. Both the pregnancy and the breastfeeding period are characterized by a loss of bone mass [9]. Postmenopausal osteoporosis is associated with hypoestrogenism, however, THO occurs more frequently during the third trimester of pregnancy, at a time when estrogen production is high.

Thus, weight gain, venous stasis and hypercoagulability occurring during pregnancy may be the cause for THO in pregnancy [10]. Other precipitating factors such as genetic predisposition, Sudeck's atrophy, compression of the obturator nerve, small vessel ischemia, spinal hypertension, consumption of alcohol, obesity, hemoglobinopathies, negative balance of calcium and changes in adrenal function have been pointed out, though in a less consensual way than pregnancy [11,12]. In THO there is a stimulus (as yet unidentified) that activates a high number of osteoclasts in the femoral head region, with significant loss of mass bone.

The interval between loss of bone mass and its formation is characterized by a fragile bone, vulnerable to microfractures and stress fractures of the femoral neck [13].

The left hip is more often hit than the right, however, other joints such as the ankle, foot, shoulder and spine can also be affected. In 25 to 30% of cases could be bilateral [1]. The diagnosis is made by the clinical history, objective examination and complementary diagnosis exams, being most often an exclusion diagnosis. The clinical presentation is not very specific, presenting as main symptoms the hip pain with increasing in intensity, claudication and muscular atrophy in the absence of trauma [3,14]. In the objective examination there is a decrease in mobility in the abduction and rotation of the hip [5]. The blood tests in most cases are within the parameters with the exception of a possible increase in alkaline phosphatase and erythrocyte sedimentation rate. In pregnancy usually both tests are high but its eaning continues unknown.18 Imaging diagnosis through radiography, CT scan or MRI allows to do the diagnosis.19 In the X-ray, is observed a peri-articular osteoporosis of the interarticular line. MRI is the gold standard exam presenting in T1 low sign in the head of the femur and high T2 sign.

These changes are suggestive of bone marrow edema [15,16]. Lequesne and Mauger describe three phases in TOH evolution: on the first stage there is an increase of pain with progressive deteriorates and some functional disability without radiologic findings. On the second stage, pain and functional disability reach the maximum and radiologically an osteopenia is observed, on the third stage a regression of the symptoms and radiologic findings occurs [17]. The normal evolution of THO is benign, presenting a medium interval between the onset of symptoms and clinical resolution of 4-24 months. The recovery usually is completed and without complications [18]. Differential diagnosis includes avascular necrosis, Gohram-Stout disease, septic arthritis, osteomyelitis, stress fractures and primary or secondary neoplasia [4,5]. Many treatments have been suggested, but all with little effectiveness. Currently, the recommended therapy is the symptomatic treatment with analgesics, anti-inflammatories non-steroids, limb discharge and physical therapy. In pregnant women, it is also not recommended to breastfeed after the delivery [19].

Other problems of pregnancy are that most analgesics and anti-inflammatories are contraindicated as therapeutic which makes it difficult to control pain in this group. Other tested therapies with cortisone, phenylbutazone, and prednisolone improve symptomatology but do not change the evolution of the disease. Scheinberg et al. showed that the use of calcitonin may not improve only the symptoms but also the evolution of the disease, presenting a faster recovery [20]. In our clinical case, the only risk factor presented by the patient was obesity. However, the fact of having placed a gastric band is not described in the literature as a factor precipitant for THO.THO is a rare disease and is mainly underdiagnosed during pregnancy due to its less specific clinical presentation [21]. In the case described there was difficulty in establishing the diagnosis due to 2 factors, the patient did not belong to the risk groups and the evolution of the disease was not the usually. As previously described, THO begins by manifesting pain associated with progressive functional disability in the hip with a spontaneous resolution of the symptoms within 24 months [22].

In this case, the presentation was a proximal fracture of the femur with pathological characteristics, being the first hypothetic diagnosis a neoplastic process. After its exclusion and as the clinical evolution was not favorable, there was a need to exclude Gohram-Stout Syndrome. The etiology of Gorham-Stout Disease is unknown and is characterized by an uncontrolled and expansive proliferation of blood vessels and lymphatic cells within the bone, causing bone reabsorption and replacement by angioms or fibrosis [23]. Gastroham-Stout Disease as in THO occurs a focal osteoporosis. However, the most affected parts are the shoulder, the skull, the pelvis, the jaw, the rib cage and the spine. Clinical presentation may be of pain in the affected region or fracture. The diagnosis is made by histology. In the clinical case we have three histological exams that exclude Goraham-Stout Disease, which leads to the diagnosis of THO [24]. The authors with this case want to alert to a pathology that is misdiagnosed and for the need for diagnosis mainly in pregnant women. The second aim was to present the less benign evolution of this pathology that despite being self-limiting may leave irreversible sequels.

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