



Contents lists available at ScienceDirect

International Journal of Surgery Case Reports

journal homepage: www.casereports.com

Case report of early aseptic loosening of total hip arthroplasty in monostotic paget disease, a diagnostic challenge

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ARTICLE INFO

Article history:

Received 1 March 2016

Received in revised form 26 May 2016

Accepted 27 May 2016

Available online 31 May 2016

Keywords:

Paget disease of bone

Implant loosening

Biphosphonates

ABSTRACT

Paget's disease of bone is a localised chronic osteopathy which produces bone deformities, bone hypervascularity, structural weakness and altered joint biomechanics. Although radiological diagnosis of Paget's disease of bone is usually straightforward, monostotic cases may potentially raise specific problems which require invasive and expensive procedures such as bone biopsies. The pelvis and upper femur are frequently affected, resulting in disabling hip disease that may require total hip arthroplasty. We report a case of Paget disease of bone in an 84-year-old woman, which was initially identified as avascular necrosis of the hip, reason for which she underwent total hip arthroplasty. During follow up, the patient complained about hip pain and in a few months she was not able to walk because of an early loosening with bone destruction. Radiological and laboratory exams were carried out with normal results except for alkaline phosphatase (AP). After treatment with biphosphonates hip pain relieved but hip reconstruction was not possible. In this paper we present an early aseptic loosening of hip arthroplasty due to monostotic Paget's disease of bone, a rare ethiology of loosening which poses particular diagnostic difficulties prompting an excessive use of excisional biopsies.

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1. Introduction

Paget's disease of bone (PD) was first described in 1877 by Sir James Paget at St Bartholomew's Hospital in London. Sir Paget described patients with multiple deformities of the bone and noticed that some patients developed bone sarcomas.

Nowadays PD is a localised chronic osteopathy characterised by an osteoclastic dysfunction with increased bone resorption and a subsequent compensatory formation of new bone with a defective microstructure. The disease may appear in its monostotic (only one third of patients) or polyostotic variants and the most common symptom is bone pain, which is typically constant, present at rest, and worse at night [1]. Patients may also present pathologic stress fractures, bony enlargement, gait disturbances or secondary osteoarthritis. Physiologic markers of bone turn over may be elevated. Suspicion should arise in case of bone pain with characteristic radiological findings.

We present a case of early failure of total hip arthroplasty, following hip osteonecrosis diagnosis and constant hip pain, more severe at rest despite the arthroplasty; subsequently, alkaline phosphatase was measured and elevated serum levels were found which

was reflective of a rapid new bone turn over. Therefore, a new diagnosis was reached: early implant loosening because of PD.

2. Case report

An 84-year-old woman was referred to our department because of pain in her left hip. The pain was of a constant nature and, although she was able to walk, it had risen gradually causing an antalgic gait. The pain got worse at rest and there was no history of trauma reported. Clinical examination showed a decreased active and passive range of motion, associated with relative left thigh atrophy, most likely because of a lack of use of the quadriceps due to the pain.

Anteroposterior and axial radiographs (Fig. 1) showed incipient hip osteoarthritis. Consequently, medical treatment was started with non-steroidal anti-inflammatory drugs (NSAID). The X-ray showed mixed radiolucent and radiopaque areas. Three months later the patient complained about hip stiffness; a new radiological exam and magnetic resonance imaging were carried out revealing cephalic bone necrosis. This X-ray shows lytic areas (Fig. 2). Before surgical treatment a bone biopsy was practiced to confirm the initial diagnosis: avascular necrosis of the hip. During surgery, a biopsy specimen was taken from the femoral head of the right hip due to excessive bleeding of the acetabular bone. The histological findings of the specimens and bones showed no evidence of malignant cells.

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Fig. 1. Radiographs of the left hip in anteroposterior view showing hip osteoarthritis and mixed radiolucent and radiopaque areas (white arrow).

Following the total hip arthroplasty (THA), she had pain-free and improved clinical functions. As demonstrated in Fig. 3, the immediate postoperative radiograph shows a correct implant positioning. Two months later, she developed a creeping right groin pain and a limp in spite of NSAID. The radiograph demonstrated an acetabular loosening with migration of acetabular component. She was advised non-weight bearing and crutches or wheel chair use. Regardless, three months later hip pain increased. Since we suspected another etiology other than hip osteoarthritis, we decided to broaden the imaging studies to discard any lesions in other bones. The radiograph and the computerized tomography (CT) showed acetabular and iliac bone destruction. The CT scan shows esclerotic and lytic areas on the right iliac bone (Fig. 4). In order to make certain it was not a malignant tumor or a bone infection a new biopsy was carried out and we obtained a large blood volume showing a profuse local vascularity. The histological section can be seen in Fig. 5.

The patient underwent laboratory investigations with blood and urinary tests looking for myeloma or lymphoma. Serum bone AP was high (three times greater than the normal value), which is reflective of the rapid new bone turnover. Other laboratory studies showed normal levels of calcium, phosphate, parathormone and vitamin D levels. A complete succession of bone radiographs (skull, humerus, spine, femur, pelvis, tibia) was performed, showing



Fig. 3. Immediate postoperative radiograph, cementless implants well positioned.

pathological findings exclusively in the pelvic bone: generalized mixed radiolucent and radiopaque areas with characteristic cotton-wool appearance (Fig. 4). No bone scan was done.

All these findings support monostotic PD as the diagnosis that caused acetabular loosening with bone destruction. After treatment with bisphosphonates hip pain decreased but because of the severe bone destruction the patient could not walk. We offered the patient a replacement of her hip arthroplasty after the treatment with bisphosphonates due to the obvious loosening of the implant. She did not accept surgical treatment to achieve hip reconstruction. At present she needs two crutches to stand up.

3. Discussion

Paget's disease of bone is a rare condition with an approximate incidence of 2–3% in patients older than 40 years [2–4]. The etiology is unknown, several observations suggest a viral cause (measles, respiratory syncytial virus and canine distemper virus) [5] but genetic and environmental factors have been implicated in the development of PD (mutations in the sequestosome 1 or p62 gene, SQSTM1) [6–9]. There is strong evidence that susceptibility is determined by variants within or close to genes that regulate osteoclast function, moreover the frequency of familial aggregation is high (in some studies 40% of affected patients have a family history of PD) [10–12].

It is a focal disorder of bone metabolism, characterized by an initial phase of bone resorption that begins in subchondral bone

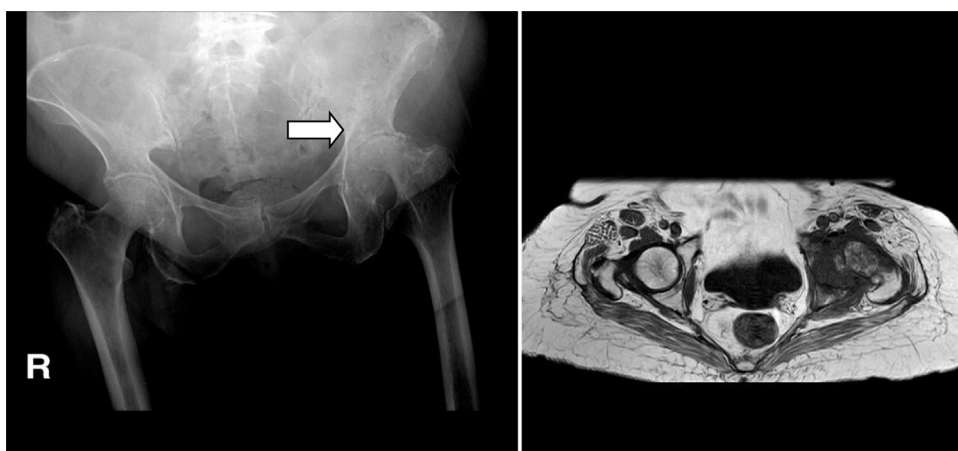


Fig. 2. Preoperative radiograph showing cephalic bone necrosis and magnetic resonance image demonstrating avascular necrosis and acetabular bone changes.

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