Transient osteoporosis of the hip with a contralateral delayed involvement: a case report

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Summary

We describe a case of non-simultaneous bilateral hip pain with bone marrow edema occurring in an adult male, with the contralateral hip being involved 12 years later after the onset of symptoms. On the basis of clinical and imaging findings, together with a complete resolution after conservative management, a post-hoc diagnosis of metachronous bilateral transient osteoporosis of the hip (TOH) was made. Non-simultaneous bilateral presentation of TOH is exceptional, and contralateral involvement with a 12-year delay has never been previously described.

KEY WORDS: transient osteoporosis of the hip; bone marrow edema; magnetic resonance imaging.

Introduction

Transient osteoporosis of the hip (TOH) is a rare, self-limiting clinical condition primarily affecting middle-aged men and also women during the last trimester of pregnancy. TOH consists of temporary osteopenia of the femoral head with progressive or acute hip pain, antalgic gait and severe functional disability (1). The etiology of TOH is still under debate, and, accordingly, no univocal treatment indications have been proposed to date (2). Although TOH is widely considered to be unilateral, a few previous descriptions of bilateral involvement have been reported (3). We report on a case of bilateral TOH occurring in an adult male, with the contralateral hip being involved 12 years after the onset of symptoms.

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Case presentation

In October of 2001, a 44-year-old employed, caucasian man was admitted to our institution complaining of continuous worsening pain and claudication of the left hip for 1 month, resulting in both a reduction in joint functionality and a disturbance of daily living activities. In the patient’s medical history, multiple contusions following an automobile accident in June of 2001 were noteworthy. Alcohol abuse and smoking as well as cortisone use were excluded. Both his developmental and family histories were unremarkable. Physical examination demonstrated the patient’s difficulty in rising from a sitting position, ambulation with an antalgic gait and a limp favoring the right hip. Both the passive and active ranges of motion were normal, but a muscular guarding with pain at the maximal passive range was detectable. A laboratory examination consisting of a complete blood cell count, serum chemistry, erythrocyte sedimentation rate testing, C-reactive protein testing, a coagulation profile and a complete assessment for rheumatic diseases showed values within the normal ranges. Plain radiographs of the left hip showed a diffuse osteopenia of the left femoral head and neck. Magnetic resonance imaging (MRI) scans (Figure 1) showed extensive bone marrow edema (BME) in the left femoral head and neck, as defined by the presence of an area of low-signal intensity on T1-weighted and high-signal intensity on T2-weighted and short T1 inversion recovery (STIR) images. Both the radiographic and MRI aspects of the right hip were unremarkable. A diagnosis of TOH was made, as the alternative diagnosis of avascular necrosis of the femoral head was excluded according to the distinguishing features proposed by Guerra and Steinberg (4). The patient was managed conservatively with non-weight bearing activity and magnetotherapy for 3 months and intramuscular injection of 100 IU calcitonin daily for a month. The symptoms resolved within 5 months, and post-treatment radiographic and MRI findings revealed a normal left hip.

The patient’s clinical history dealing with musculoskeletal diseases was unremarkable until January of 2013, when he was admitted to our institution complaining of spontaneous onset of pain in the right hip. Physical examination showed ambulation with an antalgic gait and a limp favoring the left hip. Both the passive and active ranges of motion were normal, but a muscular guarding with pain at the maximal passive range was detectable. The plain radiographs were unremarkable, whereas the MRI scans (Figure 2) showed BME in the right femoral head and neck. Again, the distinguishing criteria (4) suggested the diagnosis of TOH, with metachronous bilateral presentation. The patient was managed conservatively with non-weight bearing activity and magnetotherapy for 3 months and intramuscular injection of 200 mg sodium clodronate daily for a week then weekly for 3 months. The symptoms resolved within 6 months, and imaging findings (Figure 3) were unremarkable.
Figure 1 - MRI scan showing signs of left TOH: (a) coronal T1-weighted image, (b) axial T1-weighted image, (c) coronal STIR image and (d) axial STIR image.

Figure 2 - MRI scan showing signs of right TOH: (a) coronal T1-weighted image, (b) axial T1-weighted image, (c) coronal STIR image and (d) axial STIR image.
At the last available examination (June 2014), the patient exhibited painless hips, with a normal gait, and unremarkable physical examination for musculoskeletal diseases.

**Discussion**

TOH is an uncommon disease of an idiopathic nature, and the lack of early effective and specific diagnostic markers often complicates diagnosis (3). Although TOH usually presents with an acute onset of unilateral symptoms, bilateral and/or recurrent presentations may also occur (3); however, there is no consensus regarding their effective rate. Indeed, although the clinical review by Lakhanpal et al. (5) is usually referenced (3, 4), reporting a recurrence rate of 41%, it should be taken into account that this work involves cases of transient regional osteoporosis at different sites and does not report the exact rate in cases of only TOH. Hence, the proper comparison of rates is difficult. To the best of our knowledge, few cases of bilateral TOH have been described to date. Some previous case reports focus on simultaneous bilateral TOH without complications (3, 6) or leading to a fracture of the femoral neck during pregnancy (7).

Two distinctive characteristics of the present case are worth addressing. First, the occurrence of a bilateral TOH in a non-simultaneous manner is rare, with only 5 similar cases having been described to our knowledge. Garcia Garzón et al. (8) describe a case of right TOH in a 34-year-old man, with presentation at the left hip 4 months after the onset of symptoms. Similarly, the case report by Bolland (9) refers to a contralateral presentation of TOH in a 32-year-old man after 6 months. Owen et al. (10) focus on a case of a 34-year-old pregnant woman suffering from a left TOH, with presentation at the right hip 6 months later and evidence of a bilateral intracapsular femoral neck fracture. In addition, Dhaliwal et al. (11) report a case of bilateral TOH in a 20-year-old man, with the contralateral hip being involved 6 months after the onset of symptoms. Ikemura et al. (12) describe a patient who developed bilateral transient regional osteoporosis of the hip and subsequently experienced a relapse in the right hip 5 years later. We herein report a non-simultaneous TOH with contralateral presentation 12 years after the onset of symptoms, and no cases with a similar timing have been described to date.

The second major issue in this case addresses the lack of possible clinical and demographic factors that can pose a risk for TOH. Since its first description, TOH has been often observed in pregnant women, and Hadidy et al. (13) report on an interesting high proportion of medical doctors among patients suffering from TOH. Further risk factors, such as steroid intake, alcoholism and hypothyroidism (13), have also been recognized. However, it should be considered that in these reports, TOH was thought to be an early reversible stage of osteonecrosis of the femoral head (14). Moreover, minor trauma may cause TOH due to transient ischemia (15). Accordingly, in our patient’s medical history, a trauma preceding the left hip pain was remarkable, even though spontaneous right hip pain later manifested.

In conclusion, the etiology of TOH remains unclear. The atypical presentation we describe, together with the long time interval between the bilateral involvement of the hips, leads to the hypothesis of the presence of individual risk factors that are still unknown.
References

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