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Review

Transient regional osteoporosis

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ABSTRACT

Transient regional osteoporosis (TRO) is a disease that predisposes to fragility fracture in weight bearing joints of mid-life women and men. Pregnant women may also suffer the process, usually at the hip. The prevalence of TRO is lower than the systemic form, associated with postmenopause and advanced age, but may be falsely diminished by under-diagnosis. The disease may be uni- or bilateral, and may migrate to distinct joints. One main feature of TRO is spontaneous recovery. Pain and progressive limitation in the functionality of the affected joint(s) are key symptoms. In the case of the form associated with pregnancy, difficulties in diagnosis derive from the relatively young age at presentation and from the clinical overlapping with the frequent aches during gestation. Densitometric osteoporosis in the affected region is not always present, but bone marrow edema, with or without joint effusion, is detected by magnetic resonance. There are not treatment guidelines, but the association of antiresorptives to symptomatic treatment seems to be beneficial. Surgery or other orthopedic interventions can be required for specific indications, like hip fracture, intra-medullary decompression, or other.

Contents

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1. Introduction

Osteoporosis is a disease consisting of systemic deterioration of bone mass and microarchitecture, which predispose to fragility fractures. The disease associates with aging and exhibits a strong gender profile. The drastic fall of estrogens after menopause and the weaker bone constitution influences the higher prevalence in women. The pathophysiology of osteoporosis includes an array of additional risk factors, such as glucocorticoid therapy, chemotherapy, genetics, endocrine disorders, etc. [for review see 1].

Interestingly, some osteoporotic fractures may occur as a result of selective affliction of bones in weight bearing joints [2]. The original description defined the process as transient osteoporosis of the hip (TOH) during pregnancy [3]. The succeeding case reports revealed that TOH may be bilateral [4–12] and that may affect not only the hip, but also other joints in the lower extremities, like knee or ankle [13–15]. Moreover, it soon became clear that cases were not restricted to pregnancy [16–18], or even to women, since the disease was also observed in men, usually in their fifties or sixties [19–21].

As additional particularities, regional osteoporosis may migrate through different joints, especially when affecting men or non-pregnant women [22,23]. Consequently, the term TOH has been considered inadequate and the more inclusive transient regional osteoporosis (TRO) has been proposed by some investigators [24].

The prevalence is unknown, but it is believed that the low number of cases reported in the literature is possibly influenced by under-detection. The atypical age at presentation, together with the erroneous attribution to the frequent aches during pregnancy in the cases affecting pregnant women, are potential confounding factors [2]. A prospective study in France reported three TOH cases in 4900 pregnancies [25]. The coexistence of both systemic and regional osteoporosis has been documented [23,24,26–29], but this finding is not consistent in the literature.

The topic is of interest because, besides to a prevalence possibly higher than reported, the knowledge of the disease has improved in the latter years thanks to advances in diagnostic technology. In this review we will present the main clinical features of TRO, together with more recent data related with diagnosis and treatment.

2. Search strategy

2.1. The terms

We performed a comprehensive literature search using the PubMed database for articles published from 1980 up to December 18th 2013. Search terms were ("transient" or "transitory" or "migratory" or "regional") AND "osteoporosis". Language filters for English, French and Spanish were activated. Papers reporting clinical cases or series of cases on TRO were considered for inclusion. The process of article selection consisted of the following 2 steps: (1) three authors (AJCM, MAGP and JJT) independently screened the titles or abstracts to yield a list of candidate papers; (2) all potentially relevant papers meeting the predefined inclusion criteria were reviewed to further refine the search. Disagreements between investigators at each of the two steps were resolved by AC. The list was completed with a hand-search of reference lists of pertinent original or review articles.

2.2. Types of patients

Subjects who were diagnosed of TRO in either case reports or case series were considered eligible irrespective of whether a fragility fracture had occurred.

2.3. Data extraction

The extraction of data was performed by AJCM, MAGP and JJT and discrepancies were resolved by AC. Data collected was entered into a spreadsheet (Excel 2000, Microsoft, US). The profile of the retrieved information, where the studies were observational, conditioned a narrative review, which limited the use of the GRADE scoring criteria [30].

2.4. Search results

A total of 1427 papers were reviewed and 181 articles were selected for detailed assessment. Following a full-text review, 11 additional articles were included from manual bibliographic search. Fifty four papers were finally chosen for citation (Fig. 1).

3. Clinical presentation

The clinical picture was first described at the hip and in association with pregnancy in 1959 [3]. It was then subsequently reproduced in reports of single clinical cases or case series irrespective of whether they were related or unrelated with pregnancy [reviewed in 31]. Non-traumatic pain, which may be sudden or increase gradually, affects the hip. In the case of pregnant women, the phenomenon tends to occur during the third trimester or early puerperium. The pain, commonly localized in the groin, the anterior side of the thigh or the trochanter, begins with weight-bearing activities and increases with joint movements. Examination may reveal soft tissue swelling, effusion, and even muscle atrophy. The clinical picture progressively worsens, and the pain, which initially improves with rest, will persist at all times and become so invalidating to condition the use of a wheel-chair. The patient may suffer bilateral reduction in bone mineral density (BMD) and/or hip fracture [4–7,9,11].

Selective arthralgia of more or less acute onset is also the main symptom when the process initiates in other joints. The evolution, including the possible migration to other joints and the self-limiting condition, is similar to that affecting the hip.

A plain radiograph clearly confirms the diagnosis in case of fracture, or suggests the presence of the disease if there is a severe reduction in bone density without fracture.

4. Diagnosis

The information provided by new radiological techniques has helped to improve diagnostic accuracy and to gain knowledge on the mechanisms involved in the process [reviewed in 32]. Histological studies have been also performed to provide further evidence. However, some confusion may arise because of the considerable clinical and radiological overlapping with pathologies that may be distinct from TRO.

4.1. Radiological findings

Densitometric studies confirm that patients may suffer reduced BMD in the affected hip, and sometimes the contra lateral hip, which may be either symptomatic or non-symptomatic [33]. Bone density improves during follow-up, and the disease does not tend to recur. Conventional radiography and computed tomography (CT) show only subtle changes if there is no fracture [28,34].

Bone scintigraphy shows increased uptake of radiotracer [32,35]. The abnormality begins early and persists for weeks after symptoms abate. While this finding is clear, it is also rather non-specific, because it may result from inflammatory changes associated with infection, vascular necrosis, tumors or other causes.