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Subchondral insufficiency fracture with rapid collapse of the femoral head in a patient with Turner’s syndrome

Sir,

Primary subchondral insufficiency fracture (SIF) has been described infrequently in the limited literature since 1999 [1–4]. In the past it had always been misdiagnosed as osteonecrosis (ON). SIF of the femoral head characteristically presents as an acute painful hip disability without a history of obvious antecedent trauma, steroid therapy or alcoholism. It has been identified in elderly obese women and renal transplant patients [1]. We describe here a patient with Turner’s syndrome (TS) and consequent generalized osteopenia whose femoral heads successively underwent rapid collapse within 3 months.

A 31-yr-old woman visited our clinic for routine surveillance following right hemiarthroplasty performed at another regional hospital 1 month previously due to a rapidly collapsed femoral head. There was no evidence of antecedent trauma or known systemic conditions usually associated with non-traumatic ON. The patient’s height was only 140 cm; she weighed 35 kg, and the body mass index (18) indicated she was under weight. On physical examination, the range of motion (ROM) of bilateral hips was painless and full. A radiograph of the pelvis demonstrated a subchondral lytic lesion (Fig. 1A). The patient was treated conservatively and referred to an endocrinology clinic for evaluating her abnormally short stature, short neck, osteopenia and poor sexual maturation. After chromosome analysis, the patient was diagnosed with TS and then treated with hormone replacement therapy.

Key messages

• IVIG almost completely restores the altered phenotype of scleroderma fibroblasts.

Rheumatology

Fig. 1. (A) An anteroposterior radiograph of the left hip showing no definite hip lesions except for a suspected subchondral lytic area (arrow) over the superolateral portion of left femoral head. (B) An anteroposterior radiograph of left hip made 5 weeks later showing advanced collapse of the femoral head. The collapsed zone (arrows) seems to originate from the suspected lytic lesion and extend distally at a 45° inclination with good preservation of the dome. (C) Photomicrograph of the specimen obtained from the subchondral area of the cupola (asterisk in Fig. 1B) revealing viable cartilage and trabecular bone, implying that there is no antecedent ON. (Haematoxylin and eosin stain, original magnification ×40.) This figure is available in colour as supplementary data at Rheumatology Online.
Unfortunately, she had a sudden onset of painful disability of the left hip while bending to pick up something from the floor 1 month later. Over the succeeding weeks the pain gradually increased. Because of intractable symptoms, she visited our clinic again. At that time she needed crutches for walking as little as 100 m. Physical examination revealed a positive Fabere test and limited ROM with 40° flexion, 20° abduction, 0° adduction, 20° external rotation and 20° internal rotation. Laboratory test results were normal. Repeated radiographs demonstrated a collapsed femoral head (Fig. 1B). She subsequently underwent left hemiarthroplasty with bipolar endoprosthesis.

On gross examination, the excised femoral head revealed a deformed configuration. Articular cartilage at the superolateral portion was detached but not lost from subchondral bone. The cupula above the collapsed zone was well preserved. Histologically, thin and discontinued bone trabeculae indicating osteopenia were evident throughout the head. The specimen obtained from a subchondral area of the cupula revealed viable cartilage and trabecular bone, implying that there was no antecedent ON (Fig. 1C). In the collapsed zone, fragments of fractured bone had undergone necrosis. There was no evidence of chondrolysis, chondrocalcinosis, pannus formation or apatite crystal deposition.

An insufficiency fracture means a stress fracture that occurs in weakened bones due to osteoporosis, osteomalacia etc. SIF of the femoral head has been reported recently, mainly in the elderly, in overweight patients or in renal transplant recipients [1]. Almost all the cases of SIF had been previously considered as ON due to very similar clinical presentation and radiographic interpretation. Sometimes the histopathological differentiation between these two lesions is still confusing because all fractures will ultimately lead to some bone and marrow necrosis [5].

TS is a relatively common disorder, with an incidence of 1 in 2500 live female births. It is a result of the absence of an X chromosome or the presence of a structurally abnormal X chromosome. Its most consistent clinical features are short stature and ovarian failure [6]. Affected women are susceptible to a number of medical tribulations. Documented orthopaedic problems include osteoporosis, scoliosis and asymptomatic valgus of elbows and knees. For women with TS, there is a reduction in peak bone mass of 25% [7]. Without definite treatment they will continue to show evidence of a reduced bone mass throughout adulthood, and this has also been shown to be associated with an increased risk of fractures [6, 7].

The concluding diagnosis depends up on the histopathological findings [1, 3, 5, 8, 9]. Grossly, the necrotic lesion did not show a well-demarcated wedge-shaped, opaque-yellowish infarcted region such as that found in classic primary ON. On microscopic examination, the specimen obtained from the subchondral area of the cupula revealed viable cartilage and trabecular bone, implying that there was no antecedent ON.

Since the concept of SIF is a new one, the natural history, prognostic factors and treatment options have not yet been established. In general, elderly patients, who tend to have osteopenic bone and/or to be overweight, suffering acute hip pain without antecedent trauma are thought to be the patients at risk [1–5, 8–10]. We highlight that patients with generalized osteopenia, such as TS patients, even if young and under weight, might also be at risk. It is important to distinguish primary SIF from primary ON due to the potentially different treatment philosophy [1]. For such a case, knowledge of SIF, early recognition with MRI studies [9], avoidance of analgesics [1] and sufficient immobilization with non-weight-bearing and aggressive treatment for osteoporosis [6, 7] are probably the best ways to prevent the catastrophic rapid bone destruction.

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