We describe two patients with osteogenesis imperfecta who developed transient osteoporosis in both hips sequentially.

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Transient osteoporosis of the hip (TOH) is well-recognised, although uncommon. Its aetiology is still unclear. We describe two patients with osteogenesis imperfecta (OI) who developed this condition. We consider that a stress fracture was the triggering factor.

Case Reports

Case 1. A 37-year-old man was seen in 1994 with a painful right hip six weeks after playing table-tennis. He did not recall an obvious injury. He had type-I OI, as had his father and both of his sons, and had sustained several fractures as a child.

When seen he was using a walking-stick and could not fully bear weight. All movements of the right hip were decreased. He had typical blue sclerae, a ‘Tam-o’-Shanter’ skull and conductive hearing loss.

Radiographs of the right hip showed regional osteoporosis of the femoral head and a small stress fracture on the lateral cortex of the neck of the femur. A bone scan revealed increased uptake in the hip (Fig. 1).

He was admitted to hospital for pain control and bed rest, followed by mobilisation with protected weight-bearing as tolerated. Over a period of seven months, he improved steadily at which time his hip had recalcified to a normal appearance and his symptoms had disappeared.

He returned in 1996 having developed the same condition in his left hip (Fig. 2). His range of movement was slightly decreased. He was able to mobilise on crutches and improved with conservative management.

Case 2. A 30-year-old man was seen in 1992 with a painful left hip after a minor accident at soccer six weeks previously. He had OI, with blue sclerae and ligamentous laxity, and was of small stature. He had suffered several fractures in childhood. Movements of the left hip were reduced and painful; radiographs showed considerable osteoporosis of the head of the femur and the femoral cortex extending almost to the knee (Fig. 3).

A bone scan showed increased uptake in the left hip, both mastoid regions, the costochondral junctions and the left tibial plateau. CT of the proximal femur showed osteoporosis (Fig. 4). He was treated conservatively with protected weight-bearing. By one year he had returned to full activity and radiographs were normal.

In 1996 he again developed pain in the right hip. Radiographs showed a stress fracture of the neck of femur which was treated by internal fixation with a dynamic hip screw and one cannulated screw. He has recovered without complications.

Discussion

Transient osteoporosis is an uncommon cause of hip pain. It affects mostly healthy middle-aged men, and women in the third trimester of pregnancy. It is self-limiting and resolves symptomatically and radiologically within some months of presentation. If not recognised, however, the presentation can be alarming both clinically and radiologically.1

The aetiology is unknown. In 25% of reported cases no cause was found. In the remainder, trauma, either minor or major, had occurred in half. Other suggested triggering factors are neuralgia, herpes zoster, hemiplegia and vascular disturbances. It has been proposed that TOH is a form of reflex sympathetic dystrophy because of its diffuse pattern around the affected joint and the migratory involvement of other joints.2 This view has been disputed due to the absence of trophic changes in the superficial tissues and the benign clinical course and outcome.3,4

There are three stages in the clinical presentation of TOH. The first lasts for one to two months with a rapid increase in pain and functional disability and the second for a further two to three months with symptoms and signs

C. B. Karagkevrekis, FRCS, Orthopaedic Registrar
D. A. P. Ainscow, FRCS, Consultant Orthopaedic Surgeon
Cheltenham General Hospital, Sandford Road, Cheltenham, Gloucestershire GL53 7AN, UK.

Correspondence should be sent to Mr C. B. Karagkevrekis at the Orthopaedic Department, Alexandra Hospital, Woodrow Drive, Redditch, Worcestershire B98 7UB, UK.

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reaching a plateau; radiographs show diffuse demineralisation without loss of joint space. The third stage occurs up to six months after the onset of the condition with gradual resolution of the symptoms, although the radiological changes may persist for longer.

The differential diagnosis includes septic arthritis, malignancy and avascular necrosis. The diagnosis of septic arthritis should be straightforward, and radiography should exclude bone tumours. The distinction between TOH and osteonecrosis can be difficult, but MRI has been shown to differentiate between them at an early stage, therefore negating unnecessary investigations. The most common MRI change is oedema in the upper femur. The association of TOH with OI is extremely rare. In the only other case reported the condition occurred during pregnancy. The sequential involvement of both hips in our patients suggests that pathological fractures may have been the triggering factor.

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References