IDIOPATHIC TRANSIENT OSTEOPOROSIS OF THE HIP

L. Z. SHIFRIN, N. D. REIS, H. ZINMAN, M. I. BESSER

From the Rambam Medical Center, Haifa

We have reviewed 11 patients with idiopathic transient osteoporosis of the hip; the six who were women all developed the condition during pregnancy. Both simultaneous and sequential bilateral involvement were seen, but biochemical studies were consistently normal and one synovial biopsy showed only non-specific inflammation. Radioisotope bone scans and CT scans were useful to aid diagnosis.

Treatment by limiting weight-bearing relieved symptoms, and spontaneous resolution was paralleled by radiographic remineralisation, usually within a few months. One patient developed a stress fracture of the hip and other areas of transient osteoporosis. A hip involved by the condition should be protected from overloading until bone density has recovered.

In 1959 Curtiss and Kincaid reported three women who developed a painful hip in the third trimester of pregnancy and showed periarticular osteoporosis on radiographs. Both the symptoms and the radiographic changes improved spontaneously and no cause was found. This condition, idiopathic transient osteoporosis of the hip, was shown to affect other groups of patients by De Marchi, Santacroce and Solarino (1966) who reported five men and one woman with the same clinical picture.

Additional reports of cases have since appeared and have further defined the clinical signs and symptoms (Hunder and Kelly 1968; Lequesne 1968; Rosen 1970; Swezy 1970; Longstreth, Malinak and Hill 1973; Pantazopoulous, Exarchou and Hartofilikidis-Garofalidis 1973; Beaulieu, Razzano and Levine 1976; Kaplan and Stegman 1985). Failure to recognise this unusual cause of pain in the hip can result in delayed or mistaken diagnosis and lead to unnecessary diagnostic and therapeutic measures.

We present our experience in the management of 11 patients with idiopathic transient osteoporosis of the hip in order to discuss the spectrum of clinical manifestations and the outcome.

Table I. Details of 11 patients with idiopathic transient osteoporosis of the hip

<table>
<thead>
<tr>
<th>Case number</th>
<th>Age (years)</th>
<th>Sex</th>
<th>Pregnancy</th>
<th>Side</th>
<th>Duration of symptoms (months)</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>33</td>
<td>M</td>
<td></td>
<td>Left</td>
<td>5</td>
</tr>
<tr>
<td>2*</td>
<td>22</td>
<td>F</td>
<td>No</td>
<td>Left</td>
<td>3</td>
</tr>
<tr>
<td></td>
<td>23</td>
<td></td>
<td>Yes</td>
<td>Right</td>
<td>9</td>
</tr>
<tr>
<td>3</td>
<td>33</td>
<td>F</td>
<td>Yes</td>
<td>Both</td>
<td>3</td>
</tr>
<tr>
<td>4</td>
<td>45</td>
<td>M</td>
<td></td>
<td>Right</td>
<td>6</td>
</tr>
<tr>
<td>5*</td>
<td>32</td>
<td>F</td>
<td>Yes</td>
<td>Left</td>
<td>4</td>
</tr>
<tr>
<td></td>
<td>36</td>
<td></td>
<td>Yes</td>
<td>Right</td>
<td>3</td>
</tr>
<tr>
<td>6</td>
<td>61</td>
<td>M</td>
<td></td>
<td>Left</td>
<td>3</td>
</tr>
<tr>
<td>7</td>
<td>45</td>
<td>M</td>
<td></td>
<td>Left</td>
<td>3</td>
</tr>
<tr>
<td>8</td>
<td>31</td>
<td>M</td>
<td></td>
<td>Left</td>
<td>4</td>
</tr>
<tr>
<td>9</td>
<td>34</td>
<td>F</td>
<td>Yes</td>
<td>Left</td>
<td>3</td>
</tr>
<tr>
<td>10*</td>
<td>23</td>
<td>F</td>
<td>Yes</td>
<td>Both</td>
<td>10</td>
</tr>
<tr>
<td></td>
<td>25</td>
<td></td>
<td>Yes</td>
<td>Left</td>
<td>6</td>
</tr>
<tr>
<td>11</td>
<td>33</td>
<td>F</td>
<td>Yes</td>
<td>Left</td>
<td>3</td>
</tr>
</tbody>
</table>

* Patients affected more than once

MATERIAL AND METHODS

During the years from 1974 to 1984, a total of 11 patients with transient osteoporosis of the hip were seen by the authors at the Rambam Medical Center, Haifa, and the Rebaeca Sieff Hospital, Safed and are listed in Table I. All patients complained of pain in the hip, particularly on weight-bearing, with no history of recent injury or
ILLUSTRATIVE CASE REPORTS

Case 1. A healthy 23-year-old man developed spontaneous pain in his left groin and thigh. Two months later he was unable to bear weight on this leg. Hip movement was moderately painful and restricted. His blood count, sedimentation rate, rheumatoid serology, brucellar antigen level and tuberculin skin test were all normal and he was afebrile. Radiographs showed marked demineralisation of the femoral head (Fig. 1). The diagnosis was in doubt and open biopsy of the hip was performed. The synovium and articular cartilage appeared normal and histology revealed a low-grade, non-specific chronic synovitis (Fig. 2). Bacterial cultures were negative. After three months of walking on crutches he became painfree, and a radiograph taken 10 months after the onset of symptoms showed partial remineralisation of the femoral head (Fig. 3).

Case 2. A 22-year-old woman presented with a three-week history of pain in the left groin; she denied having had any injury. Physical

Radiograph of a 23-year-old man two months after the onset of pain in the left hip. There is decreased bone density in the proximal femur and the acetabulum, with indistinct margins to the femoral head. Figure 2 - Histological section of synovium from the left hip showing moderate proliferation and chronic inflammatory cell reaction (H & E × 125). Figure 3 - Radiograph taken 10 months after the onset of pain. The patient was asymptomatic; bone density about the left hip has improved, and the borders of the femoral head are now distinct.

illness. Most had very little discomfort at rest and there was usually only minimal restriction of movement. Trophic changes in the affected limb were absent. Radiographs showed differing degrees of demineralisation of the femoral head and a normal joint space. All biochemical studies were normal, although the extent of this examination varied from a simple blood count and sedimentation rate in some cases to complete serological and metabolic investigation in others.

Of the 11 patients, six were women and five men. At the time of diagnosis the mean age of the women was 29 years (range 22 to 36 years) and of the men 43 years (range 31 to 61 years). These 11 patients had in all 14 episodes of transient osteoporosis affecting 17 hips. On two occasions there was simultaneous bilateral hip involvement and three patients had a recurrence after intervals of one, two, and four years. The left hip was affected in 11 instances and the right hip in six. All six of the women developed the condition during pregnancy, although one had also had an episode of transient osteoporosis a year before becoming pregnant. Four of the eight pregnancies in the series were first pregnancies. Three of these women subsequently had a second pregnancy and two then suffered recurrence of transient osteoporosis.

In the three cases in which radioisotope bone scanning with 99mTc-MDP was performed, each showed increased uptake about the femoral head. A single CT study confirmed a homogeneous decrease in bone density of the femoral head and acetabulum. One patient had a normal myelogram performed at another hospital. In one case an open biopsy of the hip revealed chronic, non-specific synovitis.

Treatment in all cases was by restricting weight-bearing according to the degree of pain. When transient osteoporosis occurred simultaneously in both hips, a walker or wheelchair was required; otherwise crutches were sufficient. A few patients used mild analgesics without significant influence on their symptoms. The mean time interval from the onset of symptoms to clinical recovery was 4.7 months (range 3 to 10 months). Radiographic evidence of remineralisation paralleled the reduction of pain. The only significant complications were seen in one patient who sustained a subcapital stress fracture and developed transient multifocal osteoporosis of the knee, ankle, foot and spine.
IDIOPATHIC TRANSIENT OSTEOPOROSIS OF THE HIP

Case 2. Figure 4 – Radiograph of a 22-year-old woman with pain in the left hip. Bone density is decreased about the left hip joint and the edge of the femoral head is indistinct, but the joint space is normal. Figure 5 – A radiograph taken of the right hip after delivery and one year after Figure 4 shows patchy rarefaction of bone throughout the proximal femur. A healing subcapital fracture is seen, but the joint space appears normal.

Examination was unremarkable other than revealing an antalgic gait. Radiographs showed demineralisation of the left femoral head (Fig. 4); the blood count, sedimentation rate and rheumatoid serology were all normal. After three months of partial weight-bearing her symptoms disappeared.

One year later she returned during the seventh month of her first pregnancy complaining of pain in the right groin for several months. This had recently become worse, to the point that she was unable to walk. On examination her right hip was painful and showed marked limitation of movement. Biochemical studies were again normal. Her obstetrician refused permission for radiography. Strict bed rest brought marked relief of her hip pain, but soon she developed discomfort and slight local warmth and swelling in her right knee, ankle, and foot. Radiographs of these showed marked periarticular rarefaction of bone with normal joint spaces.

After an uncomplicated delivery, a radiograph revealed a healing subcapital fracture with minimal displacement and patchy rarefaction (Fig. 5). Two months after delivery she was able to walk unaided and without pain, but four months later she returned complaining of backache and a stooping posture. Radiographs then showed demineralisation of the thoracolumbar vertebrae, with biconcave intervertebral disc spaces, and decreased vertebral height. She was able to remain active with the aid of a hyperextension brace and became asymptomatic six months later. Follow-up radiographs showed remineralisation but no change in the dimensions of the vertebral bodies. Three years later she completed her second pregnancy without recurrence of symptoms.

Case 4. A previously healthy 45-year-old farmer complained of pain in the right thigh when walking, but could recall no injury. Pain became worse and he was confined to bed for a month, after which he was able to walk with a cane. Three months later he had a slightly antalgic gait and some pain at the extremes of a normal range of movement. Radiographs taken two months after the onset of pain showed demineralisation of the femoral head and a normal joint space. Blood count, sedimentation rate, uric acid level, rheumatoid serology, and LE factor were normal. Radioisotope imaging with $^{99m}$Tc-MDP showed increased uptake in the region of the hip and a CT scan demonstrated homogeneous decrease of bone density about the hip. Follow-up radiographs showed an improvement in bone density and at six months' follow-up the patient was asymptomatic.

DISCUSSION

The clinical picture of idiopathic transient osteoporosis of the hip is non-specific (Lequesne 1968; Pantazopoulos et al. 1973). Pain usually evolves to maximum intensity over a few weeks and is first described as a dull ache felt in the groin and anterior thigh, occasionally in the buttock. This pain is worse on weight-bearing and causes an antalgic limp. At rest, pain is relieved and there is minimal restriction of movement with pain only at the extremes of range. Thigh atrophy is seen occasionally but is never marked and recovers as pain lessens. The discrepancy between the disability and the minimal abnormalities on examination is characteristic (Lequesne 1968) and provides help in the differential diagnosis.

In pregnant women the condition is often first diagnosed as symphysiolysis of the pubis, but there is no tenderness over the symphysis or adductor muscle origins, and abduction is not limited by adductor spasm. Typically, transient osteoporosis appears in the last trimester of pregnancy and resolves during the first two postpartum months (Curtiss and Kincaid 1959; Beaulieu et al. 1976).

The radiographic features of the condition have
been well described by Lequesne (1968). Rarefaction appears within three to six weeks of the onset of pain, and is best seen by comparison with a radiograph of the asymptomatic hip. The cortical outline of the femoral head thins, with blurring of its margins and mottling. There may be periosteal reaction along the calcar (Fig. 6). Curtiss and Kincaid (1959) reported a healed stress fracture which, unlike that in Case 2 of our series, caused no acute symptoms. The joint space is always preserved and subchondral cavitation is not seen. Lequesne et al. (1977) have reported a well-defined local area of demineralisation within the femoral head which eventually developed into more general changes. To exclude synovial chondromatosis, arthrography has been recommended (Lequesne 1968; Murphy, Siegal and Gilula 1977), while radioisotope uptake has been shown to increase even before radiographic changes are seen (O’Mara and Pinals 1970; Gaucher et al. 1978; Kaplan and Stegman 1985). Since an isotope scan lacks specificity, CT may be used to confirm a homogeneous decrease in density and help to exclude other disease processes (Case 4).

Hunder and Kelly (1968) performed open biopsy in all their cases to exclude neoplasm or sepsis. Increased synovial fluid and macroscopically abnormal synovium were common, while microscopy showed minimal non-specific synovial inflammation and a varied pattern of bone resorption, formation, and necrosis. Pregnant women with transient osteoporosis may have an elevated sedimentation rate and urinary hydroxyprolene output, but these are normal in late pregnancy (Longstreth et al. 1973; Beaulieu et al. 1976). The differential diagnosis at initial presentation will include many common and rare causes of hip pain in adults (Lequesne 1968; Swezey 1970; Arnstein 1972).

Treatment is based on the premise that the condition is benign and self-limiting, requiring only pain relief by restricting weight-bearing. Drugs seem to have little influence on the symptoms. Corticosteroids (O’Mara and Pinals 1970), phenylbutazone (Dieghelm, Cadalbert and Huggler 1980), and calcitonin (Scheinberg, Aristides and Svartman 1978) have been tried with unproved effect; all carry a risk of side-effects.

Most patients recover within three to six months, but in our series three patients had recurrence in the same or the opposite hip, and other authors (De Marchi et al. 1966; Lequesne 1968; Pantazopoulous et al. 1973; Kaplan and Stegman 1985) have reported recurrence in the contralateral hip after intervals of nine months to more than 10 years. Stress fracture is rare, Curtiss and Kincaid (1959) having reported the only other case in an asymptomatic patient.

One of our patients (Case 2) had involvement of the ipsilateral knee, ankle and foot and later, of the spine; involvement of other joints has been reported by De Marchi et al. (1966), Hunder and Kelly (1968), Rosen (1970) and by Swezey (1970). Beaulieu et al. (1976) reported one case where caesarean section was necessary because painful hip movement prevented vaginal delivery, but none of our patients had this problem.

The aetiology of idiopathic transient osteoporosis of the hip remains obscure. Curtiss and Kincaid (1959) considered the possibility of mechanical compression of the obturator nerve in pregnant women, but they failed to produce hip osteoporosis by obturator nerve injury in dogs. Impairment of venous return has been suggested (Rosen 1970), while Hunder and Kelly (1968) have pointed out that the bone demineralisation is unlikely to be due to disuse since normal density is restored despite restricted weight-bearing. Lequesne (1968) proposed a non-traumatic form of Sudeck’s atrophy but patients with transient osteoporosis do not show the characteristic trophic changes seen in post-traumatic reflex dystrophy, and all our patients were well-motivated individuals with high pain thresholds. Doury, Dirheimer and Pattin (1981) have discussed the many reactive or reflex pain syndromes and use the term “primary algodystrophy of the hip” for the condition described here. There is a lack of evidence for the primary role of synovial inflammation.

The similarity of idiopathic transient osteoporosis of the hip and regional osteoporosis elsewhere in the skeleton (Duncan et al. 1967; Arnstein 1972) suggests a common aetiology, and Frost (1977) has classified the latter as a disorder of increased bone turnover. It is proposed that, in idiopathic transient osteoporosis, an unknown stimulus activates a large number of bone turnover foci in the femoral head, initiating intensive osteoclastic resorption. Later in the cycle, osteoid is laid down, mineralised and remodelled, but during the hiatus between resorption and formation, significant loss of bone tissue is manifested by decreased radiographic density. During this interval the bone is weak and vulnerable to the microfractures which we consider to be the cause of pain on weight-bearing. The periosteal reaction occasionally observed along the femoral neck is further evidence, and if healing of microfractures is inadequate a stress fracture may occur. This is the reason for recommending continued protected weight-bearing until bone density is restored. The association between transient osteoporosis and pregnancy seems more than coincidental, and in view of our two pregnant patients with recurrence, we are guarded in our advice to women about the risk of involvement in future pregnancies.

In conclusion, we emphasise that the diagnosis of idiopathic transient osteoporosis of the hip is by the exclusion of other diseases in addition to the typical findings that we have described. This means that the diagnosis cannot be absolutely certain until the patient has made a complete recovery.
REFERENCES


IDIOPATHIC TRANSIENT OSTEOPOROSIS OF THE HIP: BRIEF REPORT

G. W. KEYS, J. WALTERS

Idiopathic transient osteoporosis of the hip occurs mostly in middle-aged men, but sometimes in women, usually in late pregnancy. There is increasing pain and a limp, with some local muscle wasting. An abnormal bone scan may precede radiographic osteoporosis of the femoral head and neck (Gaucher et al. 1979). Symptoms reach a plateau then resolve, and bone density returns to normal.

Biochemical, haematological, bacteriological and serological studies are normal. The EMG and nerve conduction studies done on our patients were also normal.

**Case 1.** A 36-year-old man had spontaneous onset of pain in his left hip, which increased in severity over six weeks, being made worse by activity. He had some wasting and stiffness and an antalgic gait. Radiographs showed marked rarefaction of the whole femoral head and proximal femur (Fig. 1). Although ghostlike, the outline of the head was clear in all views and there was no narrowing of the joint space. Tomography revealed no subchondral or articular defect. Routine blood counts and chemistry were normal, the sedimentation rate was 2 mm per hour, the Mantoux test was negative and his chest radiograph was normal. The serum complement level was normal and rheumatoid serology negative. A