ACUTE POSTPARTUM INFLAMMATORY SACROILIITIS

A REPORT OF FOUR CASES

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We report four patients with unilateral postpartum sacroiliitis presenting with agonising unilateral pain, an elevated ESR, elevated alkaline phosphatase levels, leucocytosis and positive bone scans. The diagnosis of a non-infectious inflammatory cause was supported by the postpartum onset, the response to non-steroidal anti-inflammatory drugs, negative aspiration cultures in two cases and the lack of changes in the sacroiliac joints on long-term follow-up radiographs.

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Pain in the region of the sacroiliac joint is not uncommon, but the joint itself is seldom the source. The pain is usually referred from the neighbouring facet joints or intervertebral discs (Macnab and McCulloch 1990). The overlapping innervation of these structures and the proximity of the sacroiliac (SI) joint to the hip make clinical examination difficult and this has led many clinicians to doubt the existence of ‘real’ SI joint pain (Bernard and Cassidy 1991).

In pregnancy, however, SI joint pain is not uncommon (Fast et al 1987; Ostgaard and Andersson 1988). The SI joint and the pelvis undergo considerable changes in the early postpartum period and two conditions which may occur are sacroiliac strain and septic sacroiliitis.

We report four patients with postpartum sacroiliitis which started as agonising unilateral sacroiliac pain a few days after delivery accompanied by significant ‘inflammatory’ features. In none of the cases was there proof of a septic aetiology, and the pain subsided gradually and completely after treatment by non-steroidal anti-inflammatory drugs. Our cases illustrate the difficulties of diagnosis and treatment of this dramatic condition.

CASE REPORTS

All four of our patients were young women with no previous history of low back pain (Table I). The sacroiliac pain was unilateral, intense even at rest in bed, and aggravated by the smallest movement, even by tapping on the bed. Pain was exacerbated by side-to-side compression of the pelvis and by the Gaenslen test (Hoppenfeld 1976). All four had an elevated ESR, mild leucocytosis, mild elevation of alkaline phosphatase levels, and a positive bone scan (Fig. 1). The initial radiographs of the SI joints were normal, as were follow-up films taken at one year. Rheumatological blood tests, including HLA-B27 typing, were all normal. Cases 2 and 3 had aspiration of the SI joint under fluoroscopic control, yielding only a few drops of clear sterile fluid. In one patient (case 4) MRI during the acute episode of pain showed fluid in the right SI joint with bulging of its anterior capsule (Fig. 2).

Case 1 was given a course of gentamicin, ampicillin and metronidazole, and case 2 received ampicillin and metronidazole. All four patients were given non-steroidal anti-inflammatory drugs (indomethacin 25 mg three times
daily for cases 1 to 3 and naproxen 500 mg bid twice daily for case 4). In all cases recovery was slow, but by three months they could all perform the activities of daily living without pain.

Follow-up has continued for four to ten years, and all four women have had subsequent pregnancies without recurrence of any symptoms referable to the SI joint. Radiographs have remained normal.

**DISCUSSION**

The four cases which we describe occurred in a 15-year period among approximately 40 000 births, giving an incidence of about 1 in 10 000. The condition is very rare but has the unique features of agonising SI pain, elevated ESR and mild leucocytosis. There were never any definite signs of infection, and all the cases settled with no sequelae.

The normal SI joint has only a small range of motion (Frigerio, Stowe and Howe 1974), consisting of upward-downward and slight anteroposterior gliding motion (Bernard and Cassidy 1991). In the last trimester of pregnancy, hormonal changes cause the pelvic joints to relax. The hormone relaxin causes softening of the ligaments of the SI joint (Weiss et al 1977), preparing the pelvis for the passage of the fetus through the pelvic outlet. Trouseau (1873) gave the first description of SI joint strain during pregnancy and after delivery, describing severe pain in the sacroiliac region during the last trimester of pregnancy, and relating it to the increased motion in the SI joint. His patients had no accompanying fever and the pain disappeared after rest. Young (1940) gave the name ‘sacroiliac arthropathy’ to this clinical syndrome but described its onset usually in the sixth or seventh month of pregnancy.

Sashin (1930), describing the normal anatomy of the SI joint in postmortem studies, included two women who died soon after delivery and two who died in the last trimester of pregnancy. In these four cases, the capsule of the SI joint was loose and thin and it was possible to separate the joint surfaces by about 1 cm with light manual traction. Sashin also noted that slight separation
Table I. Details of four patients with postpartum inflammatory sacroiliitis

<table>
<thead>
<tr>
<th>Case</th>
<th>1</th>
<th>2</th>
<th>3</th>
<th>4</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age (yr)</td>
<td>24</td>
<td>24</td>
<td>27</td>
<td>30</td>
</tr>
<tr>
<td>Parity</td>
<td>1</td>
<td>1</td>
<td>1</td>
<td>1</td>
</tr>
<tr>
<td>Delivery</td>
<td>Normal</td>
<td>Caesarean</td>
<td>Normal</td>
<td>Normal</td>
</tr>
<tr>
<td>Onset of SI pain (days postpartum)</td>
<td>8</td>
<td>11</td>
<td>10</td>
<td>3</td>
</tr>
<tr>
<td>Side of pain</td>
<td>Left</td>
<td>Left</td>
<td>Left</td>
<td>Right</td>
</tr>
<tr>
<td>Duration of pain (days postpartum)</td>
<td>8 to 43</td>
<td>11 to 95</td>
<td>10 to 38</td>
<td>3 to 44</td>
</tr>
<tr>
<td>Duration of pyrexia (days)</td>
<td>2 to 12</td>
<td>2 to 16</td>
<td>None</td>
<td>None</td>
</tr>
<tr>
<td>WBC* (per mm³)</td>
<td>13 500</td>
<td>10 200</td>
<td>12 800</td>
<td>9 700</td>
</tr>
<tr>
<td>ESR 1st hour*</td>
<td>60</td>
<td>80</td>
<td>90</td>
<td>120</td>
</tr>
<tr>
<td>Alkaline phosphatase (IU)*</td>
<td>155</td>
<td>140</td>
<td>120</td>
<td>130</td>
</tr>
<tr>
<td>Possible infection focus</td>
<td>Endometritis (Proteus mirabilis)</td>
<td>None</td>
<td>None</td>
<td>None</td>
</tr>
<tr>
<td>Antibiotic treatment (days postpartum)</td>
<td>11 to 25</td>
<td>5 to 10</td>
<td>None</td>
<td>None</td>
</tr>
<tr>
<td>NSAID treatment (days postpartum)</td>
<td>14 to 50</td>
<td>11 to 95</td>
<td>10 to 38</td>
<td>3 to 44</td>
</tr>
<tr>
<td>Scintigraphy</td>
<td>Left positive</td>
<td>Left positive</td>
<td>Left positive</td>
<td>Right positive</td>
</tr>
<tr>
<td>Radiography</td>
<td>Loss of lumbar lordosis</td>
<td>Normal</td>
<td>Loss of lumbar lordosis</td>
<td>Normal</td>
</tr>
<tr>
<td>SI aspiration</td>
<td>Not done</td>
<td>Sterile</td>
<td>Sterile</td>
<td>Not done</td>
</tr>
</tbody>
</table>

* normal values: WBC, 4 to 10 000/mm³; ESR, 0 to 20 1st hour; alkaline phosphatase, 30 to 110 IU

at the pubis could tear the anterior sacroiliac ligaments, which are thin, weak structures. This was confirmed by Macnab and McCulloch (1990), who stated that the ligaments and capsule of the SI joints are susceptible to stretching and even tearing during parturition. Garagiola et al (1989) performed CT of the pelvis within 24 hours of delivery, and found gas in the SI joint in 42% of the women, bilaterally in 33%. This implies that the SI joint undergoes stretching during delivery which may create a vacuum effect. It may also cause bleeding or synovial effusion into the joint which could give rise to the dramatic clinical picture observed in our four patients.

The most important differential diagnosis is pyogenic sacroiliitis. This is rare, but may affect young men or women (Delbarre et al 1975; Sabato, Porat and Floman 1983). In female patients it is usually secondary to pelvic infections such as chorioamnionitis, postpartum endometritis, or an infected abortion. The clinical picture is similar to that of non-infectious sacroiliitis, with an acute onset of severe pain made worse by weight-bearing or movement at the sacroiliac joint, but it is accompanied by more definite systemic signs of infection. Typical radiological changes become apparent after a short period; these include joint erosions, sclerosis of the adjacent bone, and sometimes bony ankylosis (Gordon and Kabins 1980; Resnik and Resnick 1985).

Postpartum sacroiliac pain, termed 'inflammatory sacroiliitis', has been well described in the French literature (Gaucher et al 1968; Girodias 1970; Etienne, Vaudrey and Gougeon 1972), but is rarely mentioned in the English papers. The condition occurs a few days after delivery, and is usually associated with pyrexia, leucocytosis and a raised ESR. An infectious focus is often found elsewhere in the body (urinary tract, vagina, uterus and adnexae), but not in the SI joint itself. Etienne et al (1972) considered that the softened, oedematous postpartum capsule and ligaments of the SI joint were susceptible to infection through the lymphatics, the bloodstream (Batson 1940) or by direct extension. Gaucher et al (1968) questioned whether postpartum inflammatory sacroiliitis was actually an infectious process.

Our review of the literature found 17 cases of postpartum sacroiliitis reported since 1968 (Gaucher et al 1968; Girodias 1970; Etienne et al 1972) only one of which was shown to be due to infection. This was in a drug addict at one day postpartum; Staphylococcus aureus was cultured from the SI joint. Many of the other 16 patients were treated with antibiotics, but usually only after the pain had persisted for several days and without positive cultures. None of these cases showed the erosive changes in the SI joint that would be expected on follow-up radiographs after septic sacroiliitis.

Another possible differential diagnosis is osteitis condensans ili (Thompson 1954). This is most commonly seen in young multiparous women, and is often related to pregnancy and sacroiliac pain. This radiographic finding
is present in 1% of the general female population; 92% have experienced pregnancy and 83% have born children (Numaguchi 1971). The radiographic appearance is of bilateral increased radiodensity on the iliac side of the SI joints (Resnik and Resnick 1985).

The four cases which we report had all the clinical features of postpartum inflammatory sacroiliitis, with severe unilateral pain. Their bone scans showed increased sacroiliac activity on the affected side, although Ayres et al (1981) point out that false-positive scintigraphy of the SI joint is possible. The MRI in case 4 showed fluid in the joint, with bulging of the anterior capsule, but no evidence of a stress fracture or bone infection. This MR image was very different from that described by Wilbur, Langer and Spigos (1988) in a case of postpartum septic sacroiliitis. Our two sterile cultures, and the fact that two cases recovered without antibiotic treatment, suggest an inflammatory but non-infectious aetiology, as does the lack of changes in the long-term follow-up radiographs.

The infrequency of postpartum inflammatory sacroiliitis makes its differential diagnosis difficult. The agonising pain may lead the clinician to start antibiotic treatment, as in the first two cases in our series. The condition seems to be self-limiting, however, with no sequelae or risk of recurrence.

No benefits in any form have been received or will be received from a commercial party related directly or indirectly to the subject of this article.

REFERENCES


