Sacroiliitis detected by bone scintiscanning: a clinical, radiological, and scintigraphic follow-up study

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SUMMARY Twenty-four patients had abnormal sacroiliac joints detected by quantitative sacroiliac scintigraphy but no radiological evidence of sacroiliitis on original investigation. We studied them again after intervals of 12 to 36 months. Four patients developed radiological change. Two young HLA B27-positive men had undoubted ankylosing spondylitis, and a young woman had possible ankylosing spondylitis. A middle-aged man had changes that could be attributed to post-traumatic osteoarthritis. Of the remaining 20 cases 15 had symptoms and signs suggestive of inflammatory disease of the axial skeleton (and peripheral arthropathy in 5 cases). The sexes were equal (8 females, 7 males), and only 2 of the 15 were B27-positive. The response to anti-inflammatory medication was generally good to excellent, and scintiscans tended to improve. Of the remaining patients, 3 had mechanical or traumatic problems, and in 2 there was no explanation for the abnormal sacroiliac scintiscan. We conclude that quantitative sacroiliac scintigraphy may detect ankylosing spondylitis prior to the development of radiological change and that it can identify an organic basis for backache in patients with a spondylitis-like syndrome. The clinical circumstances must be taken into account, as scintigraphic abnormalities are not diagnostic of any specific disease entity.

Bone scanning with $^{99m}\text{Te}$-technetium-labelled stannous pyrophosphate is a sensitive means of detecting disease of the axial skeleton (Lentle et al., 1976). The technique is particularly applicable to the sacroiliac joints, where the uptake of radiotracer can be accurately quantitated in relation to the uptake by the central sacrum (Lentle et al., 1977a; Namey et al., 1977). The resultant sacroiliac joint to sacrum uptake ratios are clearly raised in a number of diseases associated with sacroiliitis (Russell et al., 1975; Russell et al., 1976a; Russell et al., 1976b; Lentle et al., 1977b; Barraclough et al., 1977; Russell et al., 1977; Davis et al., in press), and, while most of the patients studied had radiological changes in their sacroiliac joints, an appreciable number did not. The finding of scintigraphic evidence of sacroiliitis in patients with appropriate clinical symptoms and signs but who did not have radiological abnormality suggests that the scintiscan may be a valuable tool for the early diagnosis of ankylosing spondylitis and similar conditions. To test this postulate we have followed up a number of patients with scintigraphic sacroiliitis and radiologically normal sacroiliac joints, and we report our findings in this paper.

Patients and methods

Our cases were selected from a series of 172 patients seen prior to 1976 who had abnormal sacroiliac joints on bone scanning. We excluded all those with radiological abnormalities and all who had a diagnosis of psoriatic spondylitis or Reiter's syndrome and were left with 37 names. From these we traced 27 patients who were willing to participate in this project. Three were found on review not to fulfil the criteria for inclusion: 2 had scintiscans that were probably normal for the patients' degree of skeletal immaturity, and 1 had radiological abnormalities evident on the original films.

The final study population of 24 patients comprised 12 men and 12 women with a mean age of 39 years (range 19 to 59). The median interval between the original investigation and follow-up was 22 months (range 12 to 36).
Each patient's records were reviewed and relevant clinical and laboratory data noted. The patients were then examined clinically with particular attention to the axial skeleton. Measurements were made of the expansion of a 10 cm segment of lumbar spine (normal >5 cm) and of the finger-floor distance on forward flexion (normal = 0) and of chest expansion (normal > 2.5 cm). Other lumbar movements and cervical spine mobility were assessed subjectively. Tenderness was detected by direct pressure and stressing of the sacroiliac joints. Physical signs detected by these manoeuvres may be the result of a variety of pathological processes. There is unfortunately no reliable clinical method for specifically identifying inflammatory disease, so one has to rely on these techniques in conjunction with the patient's symptoms.

In each case radiographs of pelvis and lumbar spine were obtained and a bone scan, including measurement of sacroiliac joint to sacrum uptake ratios (SI:S ratios), was made as was previously described (Russell et al., 1975; Lentle et al., 1977a); new and old radiographs were read 'blind' and in random order by 2 independent observers experienced in the radiological assessment of sacroiliac joints. The appearances were graded according to the New York criteria (Bennett and Wood, 1968). Differences in interpretation were resolved by a combined review by the 2 observers who were unaware of the nature of the discrepancy.

Tissue typing by standard techniques (Russell et al., 1974) was carried out on all patients.

Results

We have classified the patients into 2 groups on the basis of the results of the radiological investigation. Group I comprises 4 patients who developed new radiological abnormalities. In the 20 patients in group II, no radiological changes were found. In describing SI:S ratios, we have used a value of 1·32 as the upper limit of normal. This figure is based on the readings obtained from a previously studied group of subjects without articular disease (Lentle et al., 1977a) and supersedes the initially recommended figure of 1·20 (Russell et al., 1975).

**Group I: New Radiological Abnormality**

The scintiscan and radiological data on the 4 patients in this group are listed in Table 1. Clinical details are as follows.

**Case 1.** This 34-year-old man was originally seen with a 14-month history of low backache and morning stiffness following a minor injury. Back movements were markedly restricted. Radiological studies included a myelogram which was normal. His sedimentation rate was 29 mm/h (Wintrobe), and tissue typing revealed the presence of the HLA B27 antigen. At follow-up 18 months later he was entirely asymptomatic on treatment with fenoprofen. He had a full range of movement of his lumbar spine and his chest expansion was 8 cm.

**Case 2.** This 25-year-old man presented with a 4-year history of intermittent pain in the hips. For the month prior to being seen he had experienced pain and stiffness of the left hip and lower back. Several months earlier he had had his first attack of acute iritis. Back movements were markedly reduced and there was a finger-floor distance of 30 cm at maximal forward flexion. Chest expansion was 4 cm. The sedimentation rate was 14 mm/h, and serological tests for rheumatoid arthritis and lupus erythematosus were negative. At follow-up 32 months later he had minimal pain but still experienced prolonged morning stiffness despite phenylbutazone therapy. Back movements and chest expansion were unchanged. There was a mild degree of painful restriction of hip movements. There was no tenderness of the sacroiliac joints or lumbar spine. He had had several attacks of iritis, and was just recovering from the most recent of these. Tissue typing performed at follow-up showed the presence of the HLA B27 antigen. This patient has a brother who is B27-positive and has ankylosing spondylitis.

**Case 3.** This 33-year-old woman presented with a 2-year history of low back pain and morning stiffness.
The spine was normal on clinical examination. The sedimentation rate was 6 mm/h. Tissue typing was negative for the HLA B27 antigen. At follow-up 18 months later she complained of intermittent low backache and pain in the neck associated with morning stiffness of up to 1 hour's duration. She had not taken any anti-inflammatory medication since experiencing an 'allergic' reaction to indomethacin some months previously. Both sacroiliac joints were tender to palpation, the right more than the left. Movements of the lumbar spine were slightly limited. Chest expansion was 6 cm. Cervical spine movements were normal.

Case 4. After a motor accident many years previously this 56-year-old man developed pain in the left knee, right ankle, and right elbow. There were no back symptoms. Radiologically there were degenerative changes in the clinically involved joints. During the year following initial investigation the patient experienced severe backache, but this disappeared promptly after a total knee replacement. When seen at follow-up 26 months after the original studies he was entirely asymptomatic, with a clinically normal axial skeleton. This patient does not have the HLA B27 antigen.

GROUP II: RADIOLOGICALLY NORMAL SACROILIAC JOINTS

We have further classified this group of 20 patients into 3 subgroups on the basis of their clinical presentation.

A: Presentation with backache. Ten patients presented with low backache associated with early morning stiffness or exacerbation. All but 2 had limited lumbar spine mobility. One of the latter patients had pain in the dorsal spine and reduced chest expansion. Nine of the patients had taken nonsteroidal anti-inflammatory drugs with good to excellent relief in 8 and persistent severe symptoms in 1 case. At the time of follow-up 4 of the asymptomatic patients had normal spinal movements. Two had discontinued their anti-inflammatory medication. One young woman was on prednisone for Crohn's disease of the small bowel, and she too had no back symptoms at follow-up. Further data on these patients are given in Table 2.

Table 2  Scintiscan and other data on patients in group IIA

<table>
<thead>
<tr>
<th>Case</th>
<th>Sex</th>
<th>Age (yr)</th>
<th>Duration of symptoms at original investigation</th>
<th>Scintiscan findings</th>
<th>Follow-up</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
<td>L SI:S ratio</td>
<td>R SI:S ratio</td>
</tr>
<tr>
<td>5</td>
<td>M</td>
<td>19</td>
<td>9 mo</td>
<td>1.39 1.50</td>
<td></td>
</tr>
<tr>
<td>6</td>
<td>F</td>
<td>22</td>
<td>2 yr</td>
<td>1.47 1.41</td>
<td></td>
</tr>
<tr>
<td>7</td>
<td>F</td>
<td>26</td>
<td>3 yr</td>
<td>1.34 1.33</td>
<td></td>
</tr>
<tr>
<td>8</td>
<td>F</td>
<td>28</td>
<td>5 yr</td>
<td>1.44 1.29</td>
<td></td>
</tr>
<tr>
<td>9</td>
<td>M</td>
<td>31</td>
<td>9 mo</td>
<td>1.42 1.49</td>
<td></td>
</tr>
<tr>
<td>10</td>
<td>F</td>
<td>34</td>
<td>2 yr</td>
<td>1.40 1.35</td>
<td></td>
</tr>
<tr>
<td>11</td>
<td>M</td>
<td>40</td>
<td>4 mo</td>
<td>1.40 1.45</td>
<td></td>
</tr>
<tr>
<td>12</td>
<td>F</td>
<td>41</td>
<td>1 yr</td>
<td>1.53 1.36</td>
<td></td>
</tr>
<tr>
<td>13</td>
<td>M</td>
<td>43</td>
<td>2 yr</td>
<td>1.48 1.48</td>
<td></td>
</tr>
<tr>
<td>14</td>
<td>M</td>
<td>53</td>
<td>6 mo</td>
<td>1.36 1.43</td>
<td></td>
</tr>
</tbody>
</table>
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B: Presentation with peripheral joint pain. Five patients presented with pain and early morning stiffness mainly affecting large peripheral joints. There was no evident joint swelling or deformity. Three of the patients complained of backache and all 5 had limitation of lumbar movement. One had no measurable chest expansion. All had taken non-steroidal anti-inflammatory drugs but were still mildly symptomatic at follow-up. Three still had abnormal back movements. None had symptoms to suggest eye, skin, genitourinary, or chronic inflammatory bowel disease. All had normal sedimentation rates and negative latex tests for rheumatoid factor. Further data are presented in Table 3.

C: Presentation with symptoms thought not to be inflammatory in origin. The remaining 5 patients presented with back or peripheral joint pain thought in each case to be caused by some process other than chronic inflammation. Their clinical and scintiscan data are summarised in Table 4.

Table 3 Scintiscan and other data on patients in group IIB

<table>
<thead>
<tr>
<th>Case, sex, age (yr), duration of symptoms at original investigation</th>
<th>Sites of symptomatic involvement</th>
<th>Scintiscan findings</th>
<th>Other sites of uptake</th>
<th>Other features</th>
</tr>
</thead>
<tbody>
<tr>
<td>15 F 1 mo</td>
<td>Hips, upper arms, L knee</td>
<td>L SI:S ratio 1.41 R SI:S ratio 1.29</td>
<td>Shoulders, hips, knees</td>
<td></td>
</tr>
<tr>
<td>31 M 2 yr</td>
<td>Hips, knees, low back</td>
<td>L SI:S ratio 1.35 R SI:S ratio 1.27</td>
<td>Shoulders, hips, knees, ankles, tarsi</td>
<td>Nil</td>
</tr>
<tr>
<td>16 M 1 mo</td>
<td>Wrists, hands, low back</td>
<td>L SI:S ratio 1.36 R SI:S ratio 1.32</td>
<td>Nil</td>
<td></td>
</tr>
<tr>
<td>35 F</td>
<td>Wrists, hands, low back</td>
<td>L SI:S ratio 1.25 R SI:S ratio 1.12</td>
<td>Nil</td>
<td></td>
</tr>
<tr>
<td>44 F 1 mo</td>
<td>Temporomandibular joints R wrist</td>
<td>L SI:S ratio 1.23 R SI:S ratio 1.34</td>
<td>R hip, manubriosternal joint, temporomandibular joints</td>
<td>R hip, 'Several large joints'</td>
</tr>
<tr>
<td>18 F Many yr</td>
<td>Large peripheral joints, low back</td>
<td>L SI:S ratio 1.52 R SI:S ratio 1.37</td>
<td>Nil</td>
<td>Anti-inflammatory medication discontinued some weeks before follow-up visit</td>
</tr>
<tr>
<td>49 M</td>
<td>Low back</td>
<td>L SI:S ratio 1.63 R SI:S ratio 1.50</td>
<td>Shoulders, elbows, wrists, knees, ankles, feet</td>
<td></td>
</tr>
<tr>
<td>19 M Many yr</td>
<td>Large peripheral joints low back</td>
<td>L SI:S ratio 1.82 R SI:S ratio 1.57</td>
<td>Knees, elbows, mid-dorsal spine</td>
<td>Chest expansion 0</td>
</tr>
<tr>
<td>59 Many yr</td>
<td>Large peripheral joints, low back</td>
<td>L SI:S ratio 1.41 R SI:S ratio 1.35</td>
<td>Knees</td>
<td>B27-positive</td>
</tr>
</tbody>
</table>

Table 4 Clinical and scintiscan data on patients in group IIC

<table>
<thead>
<tr>
<th>Case, sex, age (yr), duration of symptoms at original investigation</th>
<th>Clinical features</th>
<th>Scintiscan findings</th>
<th>Other sites of uptake</th>
</tr>
</thead>
<tbody>
<tr>
<td>34 F</td>
<td></td>
<td>Follow-up</td>
<td>L SI:S ratio 1.52 R SI:S ratio 1.55</td>
</tr>
<tr>
<td>21 F 1 yr</td>
<td>Low backache. No morning stiffness. Marked increase in lumbar lordosis</td>
<td>Original</td>
<td>L SI:S ratio 1.58 R SI:S ratio 1.46</td>
</tr>
<tr>
<td>35 F</td>
<td></td>
<td>Follow-up</td>
<td>L SI:S ratio 1.47 R SI:S ratio 1.37</td>
</tr>
<tr>
<td>22 M Many yr</td>
<td>Crush fractures of dorsal spine with marked kyphosis. Low backache. No morning stiffness</td>
<td>Original</td>
<td>L SI:S ratio 1.40 R SI:S ratio 1.37</td>
</tr>
<tr>
<td>50 M 4 yr</td>
<td></td>
<td>Follow-up</td>
<td>L SI:S ratio 1.29 R SI:S ratio 1.26</td>
</tr>
<tr>
<td>23 F</td>
<td>Calcific periarthritis of shoulders. No back symptoms. Slight reduction in lumbar flexion</td>
<td>Original</td>
<td>L SI:S ratio 1.37 R SI:S ratio 1.41</td>
</tr>
<tr>
<td>54 F</td>
<td></td>
<td>Follow-up</td>
<td>L SI:S ratio 1.45 R SI:S ratio 1.31</td>
</tr>
<tr>
<td>58 M</td>
<td></td>
<td>Follow-up</td>
<td>L SI:S ratio 1.14 R SI:S ratio 1.13</td>
</tr>
</tbody>
</table>
Discussion

In this investigation we have demonstrated that in some patients scintiscan abnormalities may fore-
shadow the development of radiological changes in
the sacroiliac joints. There can be little doubt that
cases 1 and 2 described above have ankylosing spondylitis. Case 1 fulfils the New York criteria
(Russell et al., 1977) for the diagnosis of definite
ankylosing spondylitis. This is not so for case 2, as
he has only unilateral grade 2 changes. However,
he has other attributes (for example, possession of the
HLA B27 antigen and a positive family history)
which add support to the diagnosis. These 2 cases,
therefore, show that an abnormal sacroiliac bone
scan may be a precursor to undoubted ankylosing
spondylitis. Like case 2, case 3 had radiological
involvement of only one sacroiliac joint at follow-
up but she lacked the additional features of case 2.
She can therefore be regarded as having no more than
possible ankylosing spondylitis, but she does
show that scintiscan apheresomalities may precede
radiological changes in the sacroiliac joints.

The patients in groups II A and II B (cases 5 to 19)
are of some interest. Since none of them developed
radiological changes they cannot be regarded as
having ankylosing spondylitis, but their illnesses
do appear to represent a fairly distinct clinical
syndrome. All had back pain, with or without
peripheral arthralgia, at some time, and most had
measurable changes in spinal movement. An
inflammatory cause for the pain was suggested by the
presence of morning stiffness and the prompt relief
of pain in most cases by nonsteroidal anti-
inflammatory drugs. Of patients who had discontinued
their medication 1 was symptomatic while owners
were in sustained remission. No patient had a
history or physical signs to suggest a mechanical
cause for their pain. Sedimentation rates for the
most part were normal, as they may be in anky-
losing spondylitis (Ogryzlo, 1972), and other lab-
oratory tests were negative. Apart from one instance
(case 7, with Crohn's disease) no patient had eye,
skin, genitourinary, or bowel symptoms. Radi-
ologically their sacroiliac joints were persistently
normal despite symptoms dating back many years
in some cases. The identification of an organic basis
for the patients' symptoms rested on the scintiscan
findings.

As a group these patients showed some differences
from the usual pattern seen in ankylosing spondylitis.
With 7 men and 8 women there was no male pre-
ponderance, and some of the patients were rather
older than the average newly diagnosed spondylitic.
Only 2 of the 15 patients were positive for HLA B27,
a figure no greater than could be expected by chance
(normal prevalence for our population 9·2 %)
(Russell et al., 1974). Because of these differences
it may be that these patients have a rheumatic disease
distinct from ankylosing spondylitis. However,
they could equally well be argued that they have thelatter condition but that they occupy positions in the
mildest extremity of the spondylitic spectrum.
The paucity of B27-positive individuals might be kept
with the association of B27-negativity with less severe
disease (Russell et al., 1976a), although this association has been disputed (Khan
et al., 1977).

The greater than expected number of women could be regarded as supporting the recent sug-
gestion that ankylosing spondylitis is underdiagnosed
in women, possibly because it is milder (Calin and
Fries, 1975). Resolution of this problem will require
detailed family studies of patients with scintigraphic
evidence only of sacroiliitis. Family histories in the
present series were generally negative, although one
patient (case 13) has a son with ankylosing spondyl-
itis. The possibility also exists that the sacroiliitis
in these patients may be the herald of other disease, for example Crohn's disease, as in case 7. The
young woman's diagnosis had been made coinci-
dently at laparotomy following a motor accident
and only later did she develop first backache and
then bowel symptoms.

In comparing the original SI:S ratios with those
obtained at follow-up for all the patients whom we
believe to have inflammatory back disease (that is,
cases 1, 2, 3, and 5 to 19) we found that the majority
showed improvement. Of the 18 patients 13 showed
improvement bilaterally (to complete normality in
3), 3 showed improvement in 1 joint, and 2 had
deteriorated. One of the last-mentioned had failed to
respond to anti-inflammatory drugs and the other
was symptomatic and taking no treatment. The
mean SI:S ratio originally was 1·46 ± 0·17
(standard deviation) while that at follow-up was
1·33 ± 0·16. The difference is statistically signi-
ficant (n=18, t=3·14, P < 0·005). It has been shown
that in the short term anti-inflammatory therapy
may induce a return towards normal in the sacroiliac
scan of ankylosing spondylitis (Namey et al., 1972)
and our results suggest that such improvement may
persist for long periods. A similar phenomenon has
been observed in acute Reiter's syndrome
(Russell et al., 1977).

While quantitative scintigraphy is a valuable
technique for establishing the presence of sacro-
iliitis, the finding of raised SI:S ratios is clearly not
specific for this condition. This fact is best illus-
trated by case 4, who had 'undoubted' degenerative
spondylitis. This patient is of further interest in that he developed radiological changes that were interpreted as sacro-

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References


