Quantitative sacroiliac scintigraphy in ankylosing spondylitis and Crohn's disease: a single family study

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SUMMARY A single family is presented in which 2 of the 3 sibs and 3 of 9 first degree relatives had Crohn's disease. Two subjects with Crohn's disease had classical ankylosing spondylitis (AS), and HLA typing revealed HLA B27 in all 5 members of the immediate family. The value of quantitative sacroiliac scintigraphy (QSS) in clinical practice is demonstrated. Both patients with AS had evidence of currently active sacroiliac disease in association with radiological change. One symptomatic sib had QSS evidence of sacroiliitis without radiological abnormality.

Quantitative sacroiliac scintigraphy (QSS) has been shown to be a useful investigation in the evaluation of sacroiliac disease (Lentle *et al.*, 1977a). Increased uptake of radionuclide in the sacroiliac joints correlates well with radiological changes of sacroiliitis in patients with ankylosing spondylitis (Russell *et al.*, 1975). In more recent studies it has been shown that sacroiliac scintigraphy may detect inflammatory disease of the sacroiliac joints in patients suffering from both low back pain and Crohn's disease in whom radiological evidence of sacroiliitis is absent (Lentle *et al.*, 1977b; Davis *et al.* 1978). This technique has also been shown to be useful in the assessment of the sacroiliitis associated with psoriasis (Barraclough *et al.*, 1977), Reiter's syndrome (Russell *et al.*, 1977), and anterior uveitis (Russell *et al.*, 1976). The value of this technique in the clinical management of patients with low back pain can be seen from the following case report and associated family study.

New York criteria (Bennett and Burch, 1967) and the diagnosis of Crohn's disease established by standard clinical and pathological criteria (Leonard-Jones *et al.*, 1968).

THE PATIENT

Miss A, aged 22, presented with a 2 year history of low back pain suggestive of inflammatory disease in the sacroiliac joints. There was associated morning stiffness and involvement of costochondrial joints although no peripheral arthropathy. Associated with a recent exacerbation in her back symptoms had been the development of diarrhoea. Clinical evaluation failed to reveal any abnormal physical signs. X-rays of the lumbosacral spine, the pelvis, and sacroiliac joints failed to show any abnormality. Quantitative sacroiliac scintigraphy showed significantly increased uptake at both sacroiliac joints, with subjective evidence of increased uptake throughout the axial skeleton. Tissue typing revealed the presence of HLA B27. Subsequent investigation of the diarrhoea confirmed the diagnosis of Crohn's disease.

THE FAMILY

The family tree is shown in Fig. 1. Brief clinical details are provided for the patient's parents and sibs and are all summarised in Table 1.

Mr B (father), aged 54, had had low back pain since his late 20s which had been associated with stiffness but no peripheral joint disease. He had been diagnosed as suffering from ankylosing spondylitis for 19 years. X-rays of his lumbar spine showed squaring of the lower dorsal and upper lumbar

Patients and methods

Each patient in this family study was investigated by routine radiology of the pelvis and lumbosacral spine. Quantitative sacroiliac scintigraphy was performed by a technique previously described (Lentle *et al.*, 1976), and each patient had his HLA antigens characterised (Mittal *et al.*, 1968). The diagnosis of ankylosing spondylitis was made by the

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Fig. 1 The family tree.

Table 1 Details of patient and family

	Age	X-rays	Crohn's disease	QSS	Tissue typing
A (patient)	22	NAD	+	1.79/1.62	A2, A28, BW44, B27
B (father)	54	AS	+	1.50/1.48	A28, A29, B13, B27
C (mother)	49	NAD	—	1.22/1.10	A2, BW44, B27
D (Sib)	24	AS	+	1.44/1.30	A2, A29, B13, B27
E (Sib)	13	NAD	—	—	A2, A29, B13, B27
Normal				<1.30 bilaterally	

QSS=quantitative sacroiliac scintigraphy. NAD=nothing abnormal detected. AS=ankylosing spondylitis.

vertebrae with paravertebral ossification. X-rays of the sacroiliac joints confirmed the presence of stage III sacroiliitis. Quantitative sacroiliac scintigraphy revealed the presence of markedly increased uptake at both sacroiliac joints, with evidence of increased uptake at both manubriosternal and acromioclavicular joints. Tissue typing revealed the presence of HLA B27. Subsequent investigation of abdominal pain led to the diagnosis of Crohn's disease, some 30 years after the onset of his spondylitic symptoms. His own parents and 1 brother had no history of either ankylosing spondylitis or Crohn's disease.

Mrs C (mother), aged 49, was asymptomatic. X-rays of the lumbrosacral spine and pelvis failed to show any significant change. Quantitative sacroiliac scintigraphy was normal. Tissue typing revealed the presence of HLA B27. Her own mother and one sister suffered from Crohn's disease.

Mrs D (sister), aged 24, had been diagnosed 2 years previously as suffering from Crohn's disease.

At that time associated low back pain had been thought to be related to her inflammatory bowel disease. X-rays at that time were normal. Seen recently, x-rays of the lumbar spine were normal but x-rays of the sacroiliac joints showed stage III sacroiliitis. Quantitative sacroiliac scintigraphy showed abnormal uptake at both sacroiliac joints. Tissue typing revealed the presence of HLA B27.

Miss E (sister), aged 13, was completely asymptomatic. X-rays of the lumbar spine and sacroiliac joints were normal. Bone scintiscanning was not performed, as quantitative evaluation cannot be assessed in the immature skeleton. Tissue typing revealed the presence of HLA B27.

Discussion

This patient and her family demonstrate the value of quantitative sacroiliac scintigraphy in the evaluation of sacroiliac disease. In the case of the patient, the diagnosis of ankylosing spondylitis could not be confirmed owing to the absence of radiological change. However, quantitative sacroiliac scintigraphy confirmed the presence of inflammatory disease at the sacroiliac joints, and this was seen in association with both Crohn's disease and with the presence of HLA B27. HLA B27 is not more prevalent in patients with Crohn's disease than control populations (Hyla *et al.*, 1976) but is seen in patients with Crohn's disease and associated ankylosing spondylitis (Russell *et al.*, 1974). Long-term follow-up will be of interest to see whether radiological change will develop as has been observed in previous patients from this unit with positive QSS but negative radiology (Chalmers *et al.*, 1978).

The eldest sib and the father exemplify the association previously reported between radiological change and quantitative sacroiliac scintigraphy in patients with classical ankylosing spondylitis. Both father and daughter possessed HLA B27. They are also diagnosed as suffering from Crohn's disease. On the mother's side, although she herself was not affected with either ankylosing spondylitis or Crohn's disease, there was a strong family history of Crohn's disease. The youngest sib was apparently well, with no radiological change and no symptoms. However, in view of the presence of HLA B27 and the strong family history of both ankylosing spondylitis and Crohn's disease she will be carefully followed up. Although both parents possessed HLA B27, none of the sibs were homozygous at the B locus. There was therefore no evidence to suggest that in this family homozygous B27 patients have more severe disease. Nor was it possible to relate the genetic predisposition to the development of either disease in the sibs to the inheritance of B27 from either parent.

The association between Crohn's disease and ankylosing spondylitis is well recognised. In addition sacroiliitis may be a feature of Crohn's disease, but it would appear that only those patients with classical ankylosing spondylitis and Crohn's disease possess the tissue antigen HLA B27 (Hyla *et al.*, 1976; Davis *et al.*, 1978). The presence of an abnormal scan in patient A, strongly suggests the presence of sacroiliitis in the absence of radiological change. In the father, B, and eldest sib, D, the presence of the abnormal scan suggests currently active sacroiliitis in the presence of radiological change and classical ankylosing spondylitis. These findings suggest that the patient, A, may well develop classical ankylosing spondylitis at some time in the future.

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