

## SACRO-ILIITIS IN STILL'S DISEASE\*

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Sacro-iliitis is not generally regarded as of common occurrence in adult rheumatoid arthritis, and was not a known or expected feature of the juvenile form. Because its symptoms seldom obtrude its presence is seldom excluded. In published series of cases of juvenile rheumatoid arthritis, few instances are reported, but the number of sacro-iliac x rays available is small. Thus Sairanen (1958), investigating particularly the radiological characteristics, reports only two cases with sacro-iliitis, but in only sixteen of his 100 cases had this region been x-rayed.

In this Unit, 256 cases of Still's disease have been seen between 1947 and January 1, 1961, who fulfil the diagnostic criteria, and these have been followed at least annually on a well-defined plan. As material on these cases accumulated, our attention was drawn to the number of apparently abnormal sacro-iliac joints in pelvic films taken because of hip damage.

Investigation of sacro-iliitis might contribute valuable information not only for the more complete description of Still's disease, but also on the other variants or special forms of the adult disease, that is ankylosing spondylitis, psoriatic arthropathy, and ulcerative colitis with arthritis, since few cases have been described in adolescents. In addition, because of familial association (Ansell, Bywaters, and Lawrence, unpublished), we have paid particular attention to the evaluation of sacro-iliac joint change in estimating the factors which in the adult differentiate ankylosing spondylitis from rheumatoid arthritis. To this end, pelvic films were taken of all previously-seen cases coming for follow-up and of all new cases.

Before attempting to assess the incidence of abnormal sacro-iliac joints in our series, a detailed study of the normal radiological appearance of sacro-iliac joints in juveniles appeared necessary. There are few descriptions in the literature of these joints in the growing period, and the difficulty of their interpretation is stressed.

A limited study was also planned to investigate

the anatomical features, both gross and microscopic, of juvenile sacro-iliac joints, in the *post mortem* specimens available. This work is published separately (Carter and Loewi, 1962).

### Control Series of Pelvic Radiographs

**Material.**—One pelvic film in antero-posterior projection (with gonadal protection for males) was obtained from each of sixty patients not suffering from any form of chronic joint disease, equally divided between the sexes among three age groups: 0 to 9, 10 to 14, and 15 to 19 years.

The diseases from which these patients were suffering are given in Table I. The majority were convalescent from a first attack of rheumatic fever or chorea, the diagnosis of which had been confirmed by standard criteria and by regular follow-up studies.

TABLE I  
DIAGNOSIS OF CONTROL CASES

Diagnosis	No. of Cases
Rheumatic Fever and Chorea .. ..	50
Schönlein-Henoch Purpura and Erythema Nodosum .. .. .	6
Primary Tubercle, Anaemia, Chest Pain, and Bronchitis .. .. .	4
Total .. .. .	60

Rest in bed had been maintained in the majority for less than 3 months (Table II).

TABLE II  
PERIODS OF IMMOBILIZATION (REST IN BED) OF CONTROL SERIES, BY AGE GROUP

Age Group (yrs)	Rest in Bed (wks)				
	<6	6-11	12-23	24-51	
0-9	11	6	3	—	
10-14	9	3	5	3	
15-19	8	6	4	2	
Total	No. ..	28	15	12	5
	Per cent. . .	47.0	25.0	20.0	8.0

\* This article is based on a thesis submitted for the degree of Doctor of Medicine of the University of London.

The sixty control *x* rays were scrutinized by two observers (Prof. E. G. L. Bywaters and the author), and no abnormal sacro-iliac joint changes were seen. A representative selection of seven of the best quality films from each group was made, and these were set up as standards against which to compare the Still's disease films (Figs 1 to 7, opposite).

**Normal Radiological Appearance of Sacro-iliac Joints in Three Age Groups.**—There is a tendency for the joint space to be wider in males. In the 0 to 9-year age group, the anterior and posterior spaces are usually visible, sometimes throughout the whole extent of the joint.

Both the anterior and posterior spaces are wider in the older groups, the posterior (seen medially) always appearing narrower than the anterior (seen laterally). The margins of the joint are intact and fairly distinct, and there is usually an area of greater density along the iliac border of the anterior space, which should not be confused with the faint line of subchondral density which appears in adult joints. No epiphyses are seen in this age group.

In the 10 to 14-year age group both spaces are still visible, although often not seen throughout the extent of the joint. The anterior (laterally seen) space is less clearly defined than in the younger group and appears closer to the posterior.

In the 15 to 19-year age group there is not much change from the previous group, except that the subchondral density is beginning to appear in the form of a fine radio-opaque line along the iliac margin. Young (1951) described this as a typical appearance in the adult sacro-iliac joint. In this group, lateral sacral epiphyses are sometimes visible, and show better in the female pelvis; epiphyses are also commonly seen along the iliac crest. The presence of sacral epiphyses often makes the interpretation of the sacro-iliac joints more difficult.

Rogers and Cleaves (1935) took 6-monthly stereoscopic plates of normal adolescents to observe the epiphyseal appearance and fusion. They stated that sacral epiphyses appeared at an average age of 14 to 15 years, and fused by 18 years. Not only did epiphyses complicate the radiological interpretation of sacro-iliac joints, but the presence of accessory joints also added confusion.

50 per cent. of 485 *post mortem* specimens examined by Trotter (1937) had one or more accessory facets, most often seen at the level of the second posterior sacral foramina. The incidence increased with age. Schunke (1938) found that 34 per cent. of 47 pelvis had accessory joints. No radiographs of these specimens were shown to illustrate the position of the accessory joints. A number of sacro-iliac *x* rays of patients aged 16 to 18 years or more show a particularly "convoluted" sacro-iliac articulation. The sacrum seems to be more deeply grooved and complex than in others, and it may be that these have accessory facets; this type of *x* ray is always more difficult to interpret (Fig. 8, opposite).

#### Clinical Survey of Still's Cases

The main purpose of this study is to investigate the incidence of sacro-iliitis in juvenile rheumatoid

arthritis and to determine if possible the nature of the change in these joints, that is, whether it is due to:

- (a) Rheumatoid sacro-iliitis.
- (b) Pre-spondylitic stage of ankylosing spondylitis, psoriatic arthropathy, or ulcerative colitis.
- (c) Other possible factors (e.g. immobilization).

**Material.**—216 patients with Still's disease were seen in this Unit up to December 31, 1958. The present series consists of 202 of those 216 patients who have had at least one pelvic *x* ray. We were unable to obtain films from the remaining fourteen patients, five of whom are dead, one is lost, and eight live too far away or are unwilling to co-operate.

**Diagnosis.**—These 202 patients all fulfil the diagnostic criteria for Still's disease used in this Unit, which are as follows: rheumatoid arthritis starting before the age of 16 years, with evidence of at least four joints involved for a minimum of 3 months, or of one joint involved for a minimum of 3 months with biopsy evidence. Involvement of a joint is defined as either pain and swelling, pain and limitation, or limitation and swelling. Patients with evidence of other disease either initially or at follow-up (such as rheumatic fever, disseminated lupus erythematosus, polyarteritis nodosa, psoriasis, ulcerative colitis, and ankylosing spondylitis) are excluded.

About half the patients had pelvic *x* rays because of suspected hip involvement, and the rest were taken routinely when this survey was contemplated, with the exception of a few which were obtained indirectly through other radiological investigations, such as intravenous pyelography.

**Sex, Onset, and Follow-up.**—There are 120 females and 82 males in the series: 97 seen within one year from the onset of their illness will be referred to as "early" cases, and 105 seen more than one year after onset as "late" cases. Follow-up ranges from less than one year to 12 years, and 74 per cent. were followed for 4 years or more.

**Functional State.**—This is graded from I to V.

- I Helpless
- II Chair/bed existence
- III Moderate limitation of function
- IV Slight limitation of function
- V No limitation of function

**Family History.**—This is regarded as positive when a history of rheumatoid arthritis is recorded in parents, siblings, aunts and uncles, grandparents, or great aunts and uncles.

**History of Trauma.**—This is based on injury to a joint following a specified accident within one month before the onset of symptoms in the injured joint. Accidents range from minor episodes, such as a kick or blow, to major incidents, such as a car or bicycle accident.

**Immobilization.**—This is recorded for patients who have been rested in bed, with or without traction, plasters, etc., often in other hospitals, for periods of 2 months or more.



Fig. 1.—Male, aged 5 years (Control).

Fig. 2.—Male, aged 7 years (Control).

Fig. 3.—Female, aged 9 years (Control).

Fig. 4.—Female, aged 13 years (Control).

Fig. 5.—Female, aged 15 years (Control).

Fig. 6.—Male, aged 18 years (Control).

Fig. 7.—Female, aged 21 years (Control).

Fig. 8.—Female, aged 20 years.

**Classification of Patients by Radiological Sacro-iliac Appearances.**—The series is divided by radiological appearance into two main groups, normal and abnormal, the latter group being sub-divided into definitely and probably abnormal.

The pelvic *x* rays were all scrutinized on two separate occasions by two observers (Prof. E. G. L. Bywaters and the author), and compared with the standard control *x* rays. Three grades of severity of involvement were distinguished:

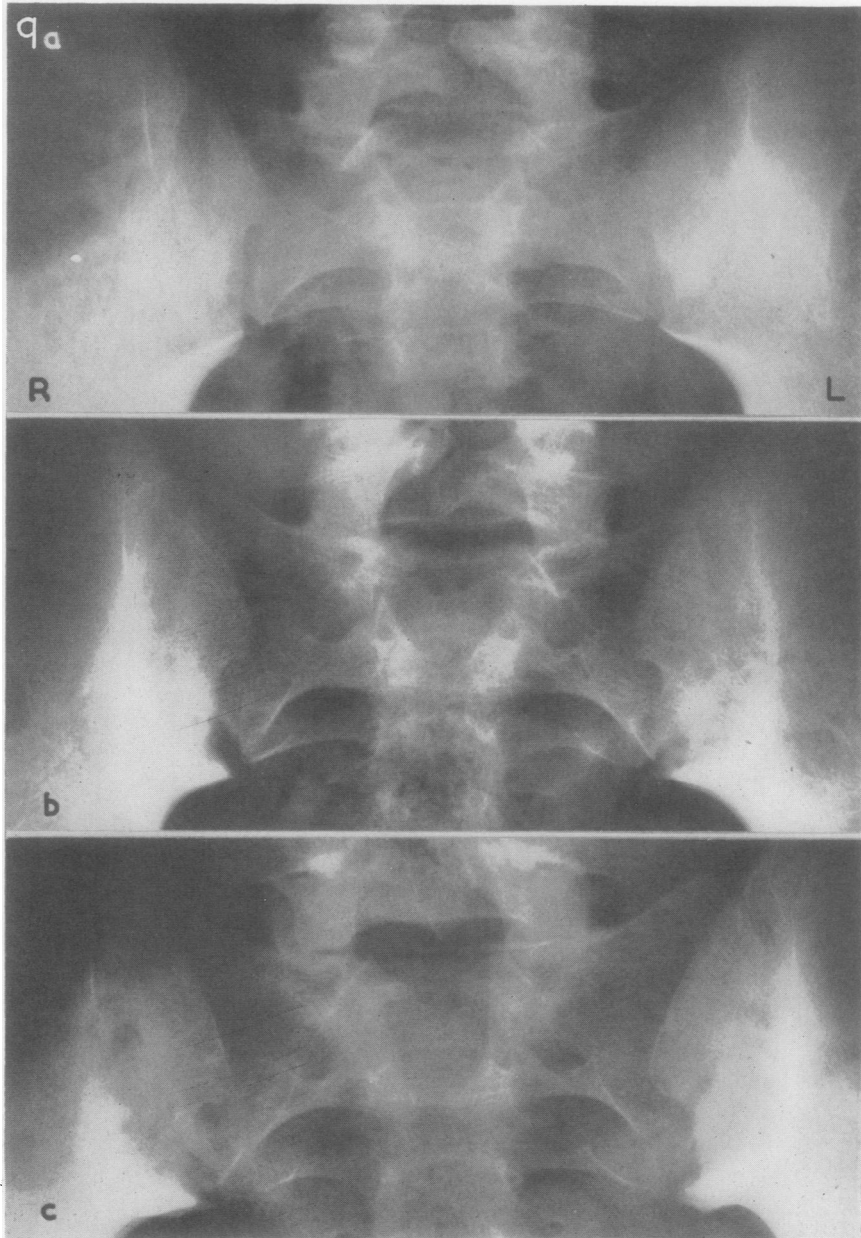


Fig. 9.—Three sacro-iliac *x* rays from a male patient with Still's disease, showing change from normal to abnormal over a period of 5 years

- (a) At age 12, probably normal. Doubtful early erosions on right.  
 (b) At age 15 years. Both joints show marginal sclerosis and erosions.  
 (c) At age 17 years. Both joints show gross marginal sclerosis and erosions.

*Grade I (Mild):* Blurring of joint spaces, slight irregularity of margins, decalcification and/or marginal sclerosis only.

*Grade II (Moderate):* Early erosions (irregular margins), with or without Grade I features.

*Grade III (Severe):* Fusion of joints, or sclerosis and marked erosions.

Examples from the definite and probable abnormal groups are illustrated in Figs 9 to 11, and these should be compared with the normal standards shown in Figs 1 to 7.

The routine view of sacro-iliac joints used in this Unit is that in the antero-posterior projection, and oblique views are rarely taken. Wilkinson and Bywaters (1958) aver that early changes in sacro-iliac joints are better shown by the antero-posterior projection than by oblique views; indeed, oblique

views may be misleading, an opinion which is borne out in this survey, and which Grainger (1960) confirms.

#### Results of Analysis of Still's Survey

Table III (overleaf) shows the classification by radiological appearance of x rays. Of the 202 cases, 48 (23·7 per cent.) are abnormal (35 definitely and 13 probably). There is a preponderance of females in the total series, but the proportion of females to males is 1:1 in the abnormal group. There is no difference in the proportion of "early" to "late" cases in either group, but in the abnormal group the mean duration of follow-up is rather shorter (5·9 compared with 7·0 years), and the mean duration from onset of disease to end of follow-up is also



Fig. 10.—Probable normal sacro-iliac x ray from a male patient with Still's disease aged 16 years.



Fig. 11.—Probable abnormal sacro-iliac x ray from a female patient with Still's disease aged 15 years, showing loss of definition of anterior aspect of joint spaces, with bridging of left posterior space.

TABLE III

SEX INCIDENCE; MEAN DURATION OF FOLLOW-UP; INTERVAL BETWEEN ONSET AND FIRST EXAMINATION AND BETWEEN ONSET AND END OF FOLLOW-UP

Sacro-iliac Status	No. of Cases	Sex		Onset		Mean Duration		No. of Cases with 4-year Follow-up	Mean Duration between Onset and End of Follow-up (yrs)	
		Male	Female	Late (> 1 yr)	Early (< 1 yr)	Before First Examination (yrs)	Follow-up (yrs)		"Early" Cases	"All" Cases
Normal . . . . .	154	58	96	79	75	2.1	7.0	116	5.6	9.1
*Abnormal and *Probably Abnormal	48 $\left\{ \begin{array}{l} 35A* \\ 13P* \end{array} \right.$	24	24	26	22	2.9	5.9	33	5.2	8.8
Total . . . . .	202	82	120	105	97	2.3	6.7	149 (73.8 per cent.)	5.5	9.0

shorter (5.2 compared with 5.6 years). 73.8 per cent. of the total series were followed for 4 years or more.

The mean age at onset of illness (7.3 years) is shown for the total series in Table IV, divided into three age groups, 0 to 4, 5 to 9, and 10 to 15 years; there are more children whose illness started in the later category in the abnormal series, whereas they are almost evenly distributed in the normal series.

The mean age at which sacro-iliac abnormality was first seen is 12.9 years, and the mean duration of illness from onset before this abnormality was seen is 5.6 years. This last figure is probably not a true representation of the age at onset of sacro-iliac changes, as some pelvic x rays were taken because of hip involvement, and some were taken routinely at follow-up visits when this survey was started—hence abnormalities may have been present earlier in some cases.

Expanding the information given in Table IV, nine of the 22 "early" abnormal cases were x-rayed

within one year from the onset of the disease and seven of these showed abnormal sacro-iliac joints at that time. The other two had normal x rays within one year of onset at 12 and 16 years respectively, and they became abnormal at 15 and 17 years respectively. Of the thirteen "early" cases who were not x-rayed within one year of onset of illness, eleven were abnormal at the first x ray, and two became abnormal later. Patient K.J., age at onset 5 years, showed normal films at 7, 8, and 9 years, and probably abnormal at 11 years. Patient R.B., age at onset 15 years, had a normal first film taken at 19 years and changed to probably abnormal at 22 years.

The smaller group of thirteen sacro-iliac x rays designated "probably abnormal" show earlier and less marked changes, but differ from the normal. This group is compared with the definitely abnormal group of 35, and the two are compared with the normal group of 154. Nine of the thirteen cases are "early" (69.2 per cent.), the mean age at onset

TABLE IV

COMPARISON OF NORMAL, DEFINITELY ABNORMAL AND PROBABLY ABNORMAL GROUPS  
MEAN DURATION OF FOLLOW-UP; FROM ONSET TO END OF FOLLOW-UP; AND AGE AT ONSET

Sacro-iliac Status	No. of Cases	"Early" Cases		Mean Age at End of Follow-up (yrs)	Mean Duration of Follow-up (yrs)	Mean Duration from Onset to End of Follow-up (yrs)	Mean Age when Abnormality First Seen (yrs)	Mean Duration from Onset to Time Abnormality First Seen (yrs)	Age at Onset (yrs)				
		No.	Per cent.						0-4	5-9	10-15	Mean	
Normal	154	75	48	16.3	7.0	9.1			49	52	53	7.2	
Abnormal	Definite	35	13	37	16.9	6.4	8.5	14.5 all 14.9 early	6.2 all 3.9 early	9	9	17	8.4
	Probable	13	9	69	11.9	4.5	6.3	9.4 all 9.1 early	3.8 all 2.4 early	5	5	3	5.6
Total . . . . .	202	97	48	16.3	6.7	9.0	12.9 all	5.6 all 3.3 early	63	66	73	7.3	

is 5.6 years (as against 8.4 for the definitely abnormal group and 7.2 for the normal group), and their mean age at the end of follow-up is 11.9 years (compared with 16.9 for the definitely abnormal group and 16.3 for the normal group). The mean duration of follow-up is shorter (4.5 years compared with 6.4 for the definitely abnormal group and 7.0 for the normal group), and the mean duration of the disease is also shorter. Also, the mean age at which this probable abnormality is first seen is 9.4 years compared with 14.5 in the definite cases, and the changes were seen earlier, that is within 3.8 years of onset (mean) compared with 6.2 (mean), although these latter figures may be affected by the reasons for which they were x-rayed, as previously stated.

The thirteen "probables" are younger patients, 69.2 per cent. of whom were seen early in their illness, and it is likely that the sacro-iliac joints will progress to show more marked changes. There was no case in the definitely abnormal group in which an x ray was considered abnormal or doubtful at an early stage and normal at a later stage, nor were any patients placed in the normal category who had earlier shown an abnormal or doubtful x ray which then reverted to normal. Scarcely any patient had symptoms referable to the sacro-iliac joints, although, before this survey started, this was not specifically sought. We have little information on the relationship of onset of radiological sacro-iliac change to the overt onset of the disease, since in only two "early" cases out of nine x-rayed within one year of the onset of the disease was the change from normal to abnormal seen.

**Correlation with D.A.T. Results.**—The D.A.T. (differential agglutination titre) performed routinely in this Unit is a modification of the Rose-Waaler technique and the method used was described by Bywaters and Scott (1960).

Table V shows the distribution of D.A.T. results in the series of 202 patients. A total of 1,481 tests was performed on all patients, giving a mean of 7.3 per patient. A titre of 1:16 or more is regarded as positive, and 33 per cent. of patients showed at least one positive result, or 12.9 per cent. showed a positive result at first test. 41.6 per cent. of the abnormal group showed a positive D.A.T. at some time compared with 30.5 per cent. of the normal group (not a significant difference). However, 31.6 per cent. of the "abnormals" gave two or more positive results at 1:16 or more compared with 11.7 per cent. of the normal group, and this is a highly significant difference ( $0.01 > p > 0.001$ ).

**Incidence of Iritis, Rash, Nodules, Endocarditis, Family History, History of Injury, and Immobilization.**—Attempts were made to correlate other clinical features with abnormal sacro-iliac joints. Neither iritis, nodules, rash, endocarditis, a history of rheumatoid disease in the family, nor a history of trauma preceding onset of illness were significantly associated (Table VI, overleaf), although iritis was slightly more frequent and rash slightly less frequent in the abnormal group.

In this series, we have seen only one child with endocarditis (aortic). She also had abnormal sacro-iliac joints and iritis, as well as extensive peripheral joint disease and involvement of the cervical spine typical of Still's disease, but no other spinal limitation or x-ray change, and no nodules. She had two out of fifteen positive D.A.T.s at 1:16.

**Effects of Immobilization.**—Periods of immobilization were considered as far as possible in dealing with a large number of cases in retrospect. Forty (25.9 per cent.) of the patients with normal sacro-iliac joints had been rested in bed for periods of 2 months or more and 25 (52 per cent.) of those with abnormal sacro-iliac joints had been subjected

TABLE V  
D.A.T. RESULTS IN TOTAL SERIES, BY SACRO-ILIAC STATUS

Sacro-iliac Status	No. of Cases Tested	Total Tests Done	Mean No. Tests per Patient	Results of D.A.T.					
				At Least One Positive		At Least Two Positive		First Positive	
				No.	Per cent.	No.	Per cent.	No.	Per cent.
Normal .. ..	154	1,094	7.3	47	30.5	18	11.7	17	11
Abnormal and Probably Abnormal .. ..	48	387	8.1	20	41.6	15* (Definite 12 Probable 3)	31.6 (34.3 23.1)	9	18.8
Total .. ..	202	1,481	7.3	67	33.2	33	16.3	26	12.9

\*  $\chi^2 = 8.8$   $0.01 > P > 0.001$

TABLE VI

INCIDENCE OF IRITIS, NODULES, RASH, ENDOCARDITIS, FAMILY HISTORY, AND TRAUMATIC HISTORY

Sacro-iliac Status	No. of Cases	No. with Iritis	No. with Nodules	No. with Still's Rash	No. with Endocarditis	No. with Positive Family History	No. with History of Injury	No. Immobilized > 2 mths
Normal .. .. .	154	7	16	37	0	12	24	40
Abnormal and Probably Abnormal .. .. .	48	4	6	6	1	6	6	25*
Total .. .. .	202	11	22	43	1	18	30	65

\*  $0.01 > P > 0.001$ 

to similar periods of immobilization (usually at other centres). This is a highly significant difference ( $0.01 > p > 0.001$ ).

Only eleven of our patients were completely immobilized for more than a year. Seven of these had abnormal sacro-iliac joints. Fig. 12 illustrates the sacro-iliac joints of a girl aged 15 years with Grade II changes. She had been immobilized for

2 years.

Table VII divides the Still's series of patients according to periods of immobilization, and shows that, of the forty immobilized patients with normal sacro-iliac joints, only eight had been immobilized for more than 6 months, whereas fourteen of the 25 immobilized patients in the abnormal group had been immobilized for more than 6 months.



Fig. 12.—Sacro-iliac x ray from a female patient with Still's disease aged 12 years, who was immobilized for 2 years, showing sclerosis and patchy obliteration of joint space on right, and blurring of anterior space on left.

TABLE VII

NUMBER OF PATIENTS IN SERIES IMMOBILIZED FOR THREE PERIODS OF TIME BY SACRO-ILIAC STATUS

Sacro-iliac Status	No. of Cases	Duration of Immobilization							
		> 2 mths		2-5 mths		6-11 mths		> 12 mths	
		No.	Per cent.	No.	Per cent.	No.	Per cent.	No.	Per cent.
Normal .. .. .	154	40	25.9	32	20.7	4	2.6	4	2.6
Abnormal and Probably Abnormal .. .. .	48	25	52.0	11	25.0	7	14.6	7	14.6
Total .. .. .	202	65	32.2	43	21.3	11	5.4	11	5.4



The extent and severity of joint involvement was compared in the immobilized and non-immobilized children. Table VIII shows that the mean number of joints involved is greater in the immobilized cases with sacro-iliac involvement than in those without sacro-iliac involvement, and also greater than in the number of non-immobilized cases. Also, those immobilized longest had the greatest number of joints involved, and were therefore the most severely ill.

Table IX divides the 48 patients with sacro-iliac involvement into three grades of severity of involvement of these joints, and shows that five patients immobilized for more than a year had Grade III changes, compared with two in the less severe grades, whereas of the patients immobilized for less than a year, the numbers in the three grades of severity were equal. However, the numbers are small, and not too much significance can be placed on them.

Previously published work on the effect of immobilization on sacro-iliac joints (Abramson and Kamberg, 1949; Abel, 1950) referred to adult patients only. It was therefore necessary to examine the pelvic x rays of a number of juvenile patients not suffering from rheumatoid arthritis or its variants who had been immobilized for periods of time comparable with those of our cases, to see if changes occurred subsequent to immobilization. Mr. G. P.

Arden kindly gave permission to study the case notes and x rays of his juvenile patients from Heatherwood Orthopaedic Hospital. Unfortunately, all but three of these patients had severe hip disease (congenital dislocation, Perthé's disease, or tuberculosis) and for that reason they were not an ideal choice. However, the sacro-iliac x rays of 72 juveniles under the age of 16 years were reviewed, all having been immobilized for at least 4 months. Of the 72 children, only seven showed any radiological sacro-iliac abnormality, and four of these were doubtful. In all cases the sacro-iliac changes were present before immobilization started. The quality of most of the films reviewed was not adequate for assessment, since they were centred on one or both hips, and some were taken through plaster casts. The changes seen comprised irregular and blurred joint margins, without obvious erosion. Although this series of cases was not satisfactory for the purpose, it is clear that immobilization had not affected the sacro-iliac joints of these juvenile orthopaedic patients.

**Peripheral Joint Involvement.**—Table X (overleaf) shows that the mean number of non-spinal joints involved at any time in all patients was 8.5, with slightly more (9.4 mean) in the group with sacro-iliac involvement than in the "normal" group (7.6 mean). No patient showed root joint involvement only, and

TABLE VIII

CORRELATION OF MEAN JOINT INVOLVEMENT AND IMMOBILIZATION IN SERIES BY SACRO-ILIAC STATUS

Sacro-iliac Status	No. of Cases	Duration of Immobilization					
		< 2 mths		> 2 mths		> 6 mths	
		No. of Cases	Joints Involved (Mean)	No. of Cases	Joints Involved (Mean)	No. of Cases	Joints Involved (Mean)
Normal .. .. .	154	114	7.0	40	7.5	8	8.9
Abnormal and Probably Abnormal .. .. .	48	23	8.3	25	10.4	14	12.2
Total .. .. .	202	137	7.2	65	8.6	22	10.9

TABLE IX

48 PATIENTS IN THREE GRADES OF SEVERITY OF SACRO-ILIITIS CORRELATED WITH THREE PERIODS OF IMMOBILIZATION, AND WITH HIP AND NECK INVOLVEMENT

Grade of Severity of Sacro-iliitis	No. of Cases	Percentage Male	No. of Patients Immobilized			No. of Patients with Hip Involvement	No. of Patients with Neck Involvement
			2-5 mths	6-11 mths	> 12 mths		
I	13	54	3	3	1	6	7
II	14	29	4	2	1	8	9
III	21	62	4	2	5	13	7
Total .. .	48	25	11	7	7	27	23

TABLE X  
INCIDENCE AND DISTRIBUTION OF PERIPHERAL AND ROOT JOINT INVOLVEMENT  
BY SACRO-ILIAC STATUS

Sacro-iliac Status	No. of Cases	Mean No. of Joints Involved per Patient	Root Joints only	Knees, Ankles, and/or Feet Only		Neck Involvement		Hip Involvement	
				No.	Per cent.	No.	Per cent.	No.	Per cent.
Normal .. .. .	154	7.6	0	25	16.2	60	41.6	54	34.4
Abnormal and Probably Abnormal ..	48	9.4	0	5	10.4	23	47.9	27*	56.3
Total .. .. .	202	8.5	0	30	14.9	83	43.0	81	39.6

\* 0.05 > P > 0.02

a higher percentage (16.2 compared with 10.4) of the normal subjects had arthritis confined to the knees, ankles, or feet only. There was no significant difference in neck involvement between the two groups. It is difficult to assess the influence of hip involvement on sacro-iliac changes, but a significantly greater number of patients with abnormal sacro-iliacs showed hip involvement (0.05 > p > 0.02).

The "abnormal" group of 48 children was divided into three grades of severity of involvement, and various possible clinical characteristics for each grade were sought. Table XI shows that those with the severest change (Grade III) were the oldest at onset of the disease, and those with the least change (Grade I), the probably abnormal group, were the youngest patients. Grade III patients had fewer joints involved (8.7 mean) than those in Grades I and II. No patient had root joint involvement only. Grade II, those with definite but moderate changes in the sacro-iliac joints, had the most hip involvement and the least neck involvement, and also had the most nodules, the greatest number of positive sero-agglutination tests, and the most extensive joint involvement including the cervical spine.

**Prognosis Based on Functional Status.** 71 "early" patients, followed for at least 4 years, were allocated

to five grades of functional state at the first examination and at their fourth annual follow-up examination. Those with sacro-iliac involvement and those with normal sacro-iliac joints show equal improvement (Table XII). Bywaters, Carter, and Scott (1959) showed that young patients with rheumatoid arthritis do better than adults, 88 per cent. of juveniles showing improvement over a 4-year period compared with 50 per cent. of adults, probably because children often reach a lower functional grade during the early acute stage of their illness and have therefore more room for improvement.

TABLE XII  
PROGNOSIS IN 71 EARLY CASES FOLLOWED MORE THAN 4 YEARS, JUDGED ON MEAN FUNCTIONAL STATUS AT FIRST EXAMINATION AND FOURTH FOLLOW-UP

Sacro-iliac Status	No. of Cases	Mean Functional Status	
		At First Examination	At Fourth Follow-up
Normal .. .. .	57	2.8	4.8
Abnormal and Probably Abnormal ..	14	2.7	4.7
Total .. .. .	71	2.8	4.8

### Discussion

Sacro-iliac x rays were obtained from 202 out of 216 patients with Still's disease, but the time at

TABLE XI  
48 PATIENTS IN THREE GRADES OF SEVERITY OF SACRO-ILIITIS CORRELATED WITH EXTENT AND DISTRIBUTION OF JOINT INVOLVEMENT, SERO-POSITIVITY, AND PRESENCE OF NODULES

Grade of Severity of Sacro-iliitis	No. of Cases	Mean Age at Onset (yrs)	Mean No. of Joints Involved per Patient	Knees, Ankles, and/or Feet Only	At Least Two Positive D.A.T.s	Nodules
I	13	5.6	10.2	1	3	1
II	14	7.3	10.3	3	6	4
III	21	9.1	8.7	1	6	1
Total ..	48	7.6	9.4	5	15	6

which they were taken in relation to the course of the disease was very variable. None had symptoms referable to the sacro-iliac joints, so that films were taken either because of hip joint involvement, where we often obtained serial pictures, or more recently for the purpose of this study as a routine procedure, whether hip joint involvement was present or not, when there was usually one film only. It is therefore impossible to assess accurately on this data the relationship between the onset of peripheral joint disease and the onset of sacro-iliitis.

The incidence of sacro-iliac joint involvement in our 202 cases of Still's disease is 23·7 per cent. (probable and definite). Grokoest, Snyder, and Ragan (1957) reported unequivocal radiological evidence of sacro-iliac abnormality in 17 per cent. of their series of 110 patients suffering from juvenile rheumatoid arthritis. Barkin, Stillman, and Potter (1955) described 71 cases of juvenile rheumatoid arthritis, of whom 25 (35·2 per cent.) showed sacro-iliitis, but they did not state whether sacro-iliitis was predominant in the 54 per cent. of cases with dorsal and/or lumbar spine involvement. It is probable that some of their cases, particularly the 54 per cent. with dorsal and lumbar spine involvement, had ankylosing spondylitis; they referred to their patients as suffering from "rheumatoid spondylitis", and it is likely that their series of 71 cases included cases of both juvenile rheumatoid arthritis with typical cervical spine involvement (80 per cent. showed radiological evidence of neck involvement) and ankylosing spondylitis with lumbar and dorsal spine involvement. We, on the other hand, have excluded from the present series two patients with ankylosing spondylitis.

Sairanen (1958) reported only two cases with sacro-iliitis in his 100 cases of Still's disease, but in only sixteen cases were pelvic *x* rays taken.

In the series of 161 cases of juvenile rheumatoid arthritis reported by Edström (1958), only one patient was found to show sacro-iliac change, but the number *x*-rayed in the pelvic region is not stated. Sury (1952) with 151 cases of Still's disease, Colver (1937) with 69 cases, Bille (1948) with 65 cases, Coss and Boots (1946) with 56 cases, and Lockie and Norcross (1948) with 28 cases, make no reference to sacro-iliitis. Pickard (1947) found only one instance of sacro-iliitis among her 35 cases of juvenile rheumatoid arthritis. The data from these series are therefore insufficient to compare the incidence of sacro-iliac involvement.

Although Still (1897) believed that the condition he first described differed from adult rheumatoid arthritis, data accumulated in this Unit have led to the belief that Still's disease (juvenile rheumatoid

arthritis) and the adult form are not separate entities, since all features are common to both, though present in different proportions in the two age groups. On a comparison of clinical, pathological, and serological tests, Bywaters and others (1959) concluded that the differences between the juvenile and adult disease were due to age only. McEwen, Ziff, Carmel, DiTata, and Tanner (1958) also took this view, on the grounds of serological observations, and the opinion is now widely held.

Controversy continues on the relationship between ankylosing spondylitis and rheumatoid arthritis. It is accepted that rheumatoid arthritis may affect the spine, although spinal involvement is rarely a prominent clinical feature. Sharp (1957) found evidence of spinal involvement of 50 per cent. of 100 patients, but in contrast to ankylosing spondylitis the cervical region was more commonly affected, being seen in four-fifths of those with spinal involvement, the dorsal and lumbar region being affected in less than one-third. In over half those with spinal involvement, only the cervical region was affected. Sharp noted that sacro-iliac change in rheumatoid arthritis was "not uncommon" but gave no figures.

Our present series of cases of Still's disease is the largest reported. The first suggestion that change in the sacro-iliac joints is due to rheumatoid disease rather than due to ankylosing spondylitis is supported by the fact that each has peripheral joint involvement and none has radiological evidence of spinal involvement of the ankylosing spondylitis type.

Those patients who have abnormal sacro-iliac joints do not show any difference in the distribution of peripheral joint involvement from those without sacro-iliitis, whereas it was shown by Wilkinson and Bywaters (1958) in adult ankylosing spondylitis that 39 per cent. of 222 cases had root joint involvement, and only 24 per cent. had peripheral arthritis, other than of the root joints. 50 per cent. of their patients in whom the disease began before the age of 20 years showed root joint involvement compared with 36 per cent. of those with a later onset. At follow-up (mean 6·2 years, standard deviation 3·8), only sixteen of their total of 222 patients had disease confined to the sacro-iliac joints only, without spinal involvement.

Although a slightly higher proportion of our "abnormal" group had iritis, this is not a significant difference from the normal group, and does not compare with the 25 per cent. incidence found by Wilkinson and Bywaters in their ankylosing spondylitis series, but a time factor is probably involved.

The occurrence of nodules in the "abnormal" group favours the diagnosis of rheumatoid arthritis, since these are not seen in ankylosing spondylitis.

The D.A.T. test is not a useful diagnostic aid in juvenile rheumatoid arthritis. In a series of 142 of the same patients with Still's disease as are surveyed here, who were followed for 4 years or more, Bywaters and others (1959) showed that the test was positive in only 13.4 per cent. of juveniles compared with 50 per cent. of adults, taking only the first test into consideration, and that 19.7 per cent. of juveniles had two or more tests positive at titres of 1:16 or more. Our present series of 202 patients gave 12.9 per cent. positive results at the first test, but there was a significant association between sero-positivity and abnormal sacro-iliac joints, taking the D.A.T. results on patients with two or more positive tests. Since cases of ankylosing spondylitis, psoriatic arthritis, Reiter's syndrome, and ulcerative colitis are usually seronegative (Alexander and de Forest, 1954; Ziff, Brown, Badin, and McEwen, 1954; McEwen and others, 1958; Dresner and Trombly, 1959), the marked association of sero-positivity with abnormal sacro-iliac joints presents a strong argument against a diagnosis of ankylosing spondylitis or other "variants" of rheumatoid arthritis, and favours the view that these patients have severe Still's disease.

The possibility that those with abnormal sacro-iliac joints may be in the pre-spondylitic stage of ankylosing spondylitis must be considered. Newton (1957) remarked that there was an extraordinary lack of awareness of the early manifestations of ankylosing spondylitis outside Departments of Physical Medicine and Hospitals for Rheumatic Disorders, and that the average time taken to reach such a diagnosis was 6 years. It may be that our cases, because they have presented with peripheral arthritis, have been discovered early before sacro-iliac symptoms or spondylitis could appear.

Lucchesi and Lucchesi (1949), reporting a series of 584 cases of ankylosing spondylitis (the diagnosis being confirmed radiologically), emphasized the silent onset and progress of the disease as well as the wide range of age at onset, irrespective of sex. The onset of symptoms occurred before the age of 16 years in 54 of their patients, 22 of whom were males; ten of these children, four of whom were males, had symptoms before the age of 5 years. These authors suggest that many cases are missed, particularly in childhood, through lack of knowledge of pre-spondylitic symptoms and lack of awareness that the disease can occur in childhood. Unfortunately they do not describe the early symptoms obtained by them from these children, nor do they illustrate with pelvic *x* rays taken at the pre-spondylitic stage of the illness. There is no follow-up record of the lapse of time between the

onset of sacro-iliitis and that of spondylitis.

The proportion of males to females in our series of sacro-iliac abnormalities is 1:1 compared with 1:1.5 for the "normal" group; the generally accepted male:female sex ratio for adult ankylosing spondylitis varies from about 4:1 to 10:1. The difficulty in reaching a conclusion is due to the absence of any spinal changes characteristic of ankylosing spondylitis either clinically or radiologically. Of all the patients admitted here and diagnosed as cases of Still's disease, only one, who has been followed for 10 years, has developed the characteristics of ankylosing spondylitis. He is not included in this survey for that reason. The only other patient seen here with onset of illness before the age of 16 years in whom spinal change has developed is a female, with onset of illness at 11 years. She was admitted here at age 15 years with ankylosing spondylitis, when signs of sacro-iliac and spinal changes were already present.

Polley and Slocumb (1947) give the age range at onset in their series of 98 cases of ankylosing spondylitis as 15 to 35 years (mean 26.7). In 23 per cent. of their series the ankylosing spondylitis started with peripheral joint involvement, and Boland and Present (1945) noted that involvement of the peripheral joints antedated the spinal symptoms in 13 per cent. of cases. Parr, White, and Shipton (1951) reported involvement of the peripheral joints as an initial symptom in only 6 per cent. of cases. All our cases showed peripheral joint involvement at onset.

A significant difference was shown between those immobilized for more than 2 months in the abnormal group compared with the normal subjects, and an even greater significance in the smaller group immobilized for more than 6 months. However, it seems improbable that these sacro-iliac changes are due solely to immobilization, and none of 72 similarly immobilized young orthopaedic patients developed sacro-iliitis after a strict regimen of rest in bed. The mean number of joints involved is greater in the immobilized cases with sacro-iliac involvement than in those without sacro-iliac involvement, and also greater than in the non-immobilized cases. Also, those immobilized longest had the greatest number of joints involved, and were therefore the most severely affected children.

Two cases of Still's disease described in the anatomical study in the succeeding paper (Carter and Loewi, 1962) show two distinct forms of change in the sacro-iliac joints. Case 7 shows fusion of cartilage without inflammatory change in one sacro-iliac joint, and Case 8 shows erosion of cartilage and bone with much chronic inflammation,

as is seen in rheumatoid arthritis. The former showed probable abnormality in the radiograph of her right sacro-iliac joint, and the latter had a normal pelvic radiograph.

Hip disease may affect the sacro-iliac joints; eighty out of our 202 cases had clinical hip involvement, with or without radiological change. There was a significant correlation between hip joint and sacro-iliac joint involvement, but there is no means of assessing the influence of hip disease in the aetiology of sacro-iliitis in these children.

A possible explanation for sacro-iliitis in our series is the "adolescent sacro-iliac syndrome" described by Rogers and Cleaves (1935) if, indeed, this is a separate entity; it is characterized by stiffness, low back pain, and tenderness over the sacro-iliac joint. As Solonen (1957) remarked, the cases described included a more or less definite history of strain to the sacro-iliac joint. Rogers and Cleaves stressed the difficulty of interpreting the epiphyseal growth area in the region of the sacro-iliac joints, and described cloudy irregular areas similar to the appearances of some of our *x* rays. They suggested that the lesion was an epiphysitis rather than an arthritis, but their patients were otherwise healthy adolescents. The mean age at onset of our whole series is 7.3 years, but the mean age at which sacro-iliac abnormality was first seen is about 13 years, *i.e.* in the adolescent stage, although in many of our cases it must have been present well before the first *x* rays were taken. It is therefore possible that some of the sacro-iliac joint changes may be due to epiphysitis caused by excessive strain imposed by other arthritic joints.

Rare conditions in which peripheral arthritis

occurs with sacro-iliitis must be considered, but it is unlikely that many of the patients with Still's disease will develop either ulcerative colitis or psoriasis. However, four patients originally diagnosed as cases of juvenile rheumatoid arthritis developed ulcerative colitis and four developed psoriasis, and these cases were therefore excluded from the Still's disease series. None developed Reiter's syndrome. It is not possible to differentiate between the radiological appearances of the sacro-iliac joints in these conditions, so that no prophesies can be made until the full clinical picture develops. In the four patients now diagnosed as cases of ulcerative colitis with arthritis, the arthritis was the presenting symptom, and the bowel symptoms appeared from a few weeks to 4 years after onset; there is a selection of material in such cases because such patients are usually referred to this Unit because of arthritis. A fifth patient who presented with bloody diarrhoea was referred to us from the general medical unit at this hospital; he developed arthritis 3 months later. These five cases demonstrate that some patients presenting with arthritis may develop the symptoms of ulcerative colitis as much as 4 years later. The four cases who presented with arthritis all had sacro-iliac involvement (Fig. 13), but the fifth presenting with colitis did not.

The same observations apply to psoriasis with arthritis in our small series of four cases. They presented with arthritis, the psoriasis appearing from 3 to 9 years later. Another child referred to us late in the illness (2 years after the onset of arthritis) had had psoriasis for 7 years, and in another psoriasis appeared one year before arthritis. The



Fig. 13.—Sacro-iliac *x* ray from a male patient aged 16 years, with ulcerative colitis and arthritis, showing decalcification and erosions of both joints.

pelvic *x* rays of four of these patients included three definitely abnormal (Fig. 14) and one probably abnormal. The D.A.T. test does not distinguish between these variants. The tests were all negative in the eleven patients described above, except for one positive titre of 1:32 in a boy with ulcerative colitis.

By grading sacro-iliitis into three levels of severity, and correlating features associated with each grade, Grade I is the early, mildly affected group, with "in-between" associations, Grade II comprises the most severely-affected patients of classical rheumatoid type, and Grade III, showing fusion or marked erosions, contains those in whom the disease is of later onset; these last may represent a different category, namely those who may later develop the additional signs of the "variants" of rheumatoid arthritis. The sex incidence in these three groups also supports this theory, since there is a preponderance of males in Grade III, and of females in Grade II, the sexes being equally distributed in the indeterminate Grade I. (There was no difference in the proportion immobilized in the three grades of severity of involvement.) If there is a distinct degenerative change due to immobilization (as suggested in the *post mortem* findings of Case 7\*), then some of this type will be seen in the three groups, and presumably some cases combine rheumatoid and degenerative change.

#### Conclusions and Summary

A series of pelvic *x* rays from sixty patients not suffering from chronic joint disease provided normal

\* See Carter and Loewi (1962), p. 132 and 133 below.

standards for different age groups with which to compare the findings in 202 cases of Still's disease.

Sacro-iliac joint changes occurred in 24 per cent. of this series of 202 cases; they were equally distributed among males and females, and were seen more frequently in those in whom the disease started after the age of 10 years. The changes were more common in patients with a greater number of joints involved and in those who were sero-positive. Those who had been immobilized for long periods and those who had hip involvement were more often affected, despite which the prognosis in the abnormal and normal groups was equally good.

The distribution of joint involvement did not resemble that seen in ankylosing spondylitis, with root joints or lower limb joints predominantly affected.

Iritis, rash, nodules, family history, and trauma preceding onset were not significantly related to the occurrence of sacro-iliitis.

The group with less severe sacro-iliac changes (Grade I) comprises the patients who were youngest at the time of onset of the disease, and shows no specific clinical associations. Grade II comprises those with moderately severe sacro-iliac change, who appear to be patients with classical, severe Still's disease, with nodules, sero-positivity, and the most extensive joint involvement, including that of the cervical spine. There is a preponderance of females in this grade. Grade III comprises those with fusion or extensive erosions of the sacro-iliac joints. In these patients the disease starts later, and fewer joints are involved; they also have the most hip involvement and the least neck involvement, and males predominate.

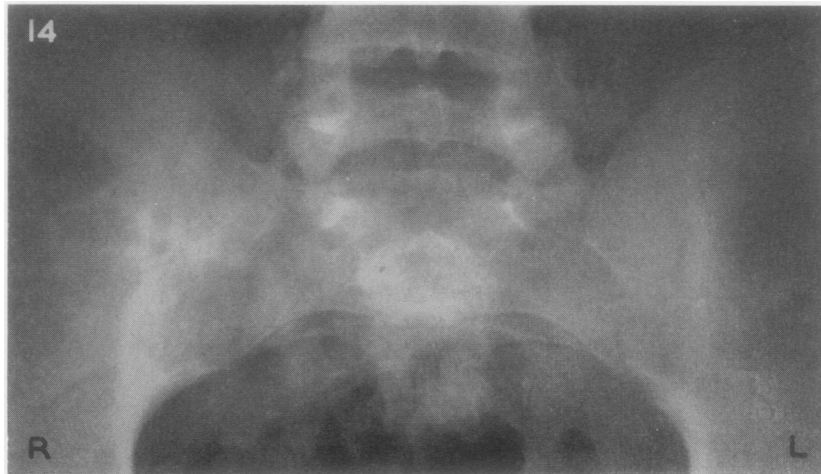


Fig. 14.--Sacro-iliac *x* ray from a male patient aged 16 years, with psoriasis and arthritis, showing sclerosis and partial obliteration of joint spaces.

Nine patients who were diagnosed on admission as cases of Still's disease developed either ulcerative colitis (four), psoriatic arthropathy (four), or ankylosing spondylitis (one), and were therefore excluded from the series. These few cases did not differ sufficiently from the remainder in the early stages for one to be able to foretell the onset of the symptoms of the "variant" diseases, that is diarrhoea, rash, or spondylitis.

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## REFERENCES

- Abel, M. S. (1950). *Radiology*, 55, 235.
- Abramson, D. J., and Kamberg, S. (1949). *J. Bone Jt Surg.*, 31A, 275.
- Alexander, R., and de Forest, G. K. (1954). *Amer. J. Med.*, 16, 191.
- Barkin, R. E., Stillman, J. S., and Potter, T. A. (1955). *New Engl. J. Med.*, 253, 1107.
- Bille, S. V. (1948). *Nord. Med.*, 37, 307.
- Boland, E. W., and Present, A. J. (1945). *J. Amer. med. Ass.*, 129, 843.
- Bywaters, E. G. L., Carter, M. E., and Scott, F. E. T. (1959). *Ann. rheum. Dis.*, 18, 233.
- and Scott, F. E. T. (1960). In "Recent Advances in Clinical Pathology", Series 3, p. 278. Ed. S. C. Dyke. Churchill, London.
- Carter, Mary E., and Loewi, G. (1962). *Ann. rheum. Dis.*, 21, 121.
- Colver, T. (1937). *Arch. Dis. Childh.*, 12, 253.
- Coss, J. A., and Boots, R. H. (1946). *J. Pediat.*, 29, 143.
- Dresner, E., and Trombly, P. (1959). *New Engl. J. Med.*, 261, 981.
- Edström, G. (1958). *Arthr. and Rheum.*, 1, 497.
- Grainger, R. G. (1960). *Rheumatism*, 16 (No. 1), p. 3.
- Grokoest, A. W., Snyder, A. I., and Ragan, C. (1957). *Bull. rheum. Dis.*, 8, 147.
- Lockie, L. M., and Norcross, B. M. (1948). *Pediatrics*, 2, 694.
- Lucchesi, M., and Lucchesi, O. (1949). *Rev. Rhum.*, 16, 618.
- McEwen, C., Ziff, M., Carmel, P., DiTata, D., and Tanner, M. (1958). *Arthr. and Rheum.*, 1, 481.
- Newton, D. R. L. (1957). *Proc. roy. Soc. Med.*, 50, 850.
- Parr, L. J. A., White, P., and Shipton, E. (1951). *Med. J. Aust.*, 1, 544.
- Pickard, N. S. (1947). *A.M.A. Arch. intern. Med.*, 80, 771.
- Polley, H. F., and Slocumb, C. H. (1947). *Ann. intern. Med.*, 26, 240.
- Rogers, M. H., and Cleaves, E. N. (1935). *J. Bone Jt Surg.*, 17, 759.
- Sairanen, E. (1958). *Acta rheum. scand.*, Suppl. 2.
- Schunke, G. B. (1938). *Anat. Rec.*, 72, 313.
- Sharp, J. (1957). *Brit. med. J.*, 1, 975.
- Solonen, K. A. (1957). *Acta orthop. scand.*, Suppl. 17.
- Still, G. F. (1897). *Med.-chir. Trans.*, 80, 47.
- Sury, B. (1952). "Rheumatoid Arthritis in Children." Munksgaard, Copenhagen.
- Trotter, M. (1937). *Amer. J. phys. Anthropol.*, 22, 247.
- Wilkinson, M., and Bywaters, E. G. L. (1958). *Ann. rheum. Dis.*, 17, 209.
- Young, J. H. (1951). *Med. J. Aust.*, 2, 761.
- Ziff, M., Brown, P., Badin, J., and McEwen, C. (1954). *Bull. rheum. Dis.*, 5, 75.

## Atteinte sacro-iliaque dans la maladie de Still

## RÉSUMÉ

Une série de radiographies du bassin de soixante sujets n'atteints d'aucune maladie articulaire chronique permet de déterminer des valeurs normales pour de différents groupes selon l'âge et de comparer à ces valeurs les résultats obtenus dans 202 cas de maladie de Still.

Les altérations dans l'articulation sacro-iliaque furent rencontrées dans 24 pour cent de cette série de 202 cas. Ces lésions furent également distribuées entre les deux sexes et on les trouva plus souvent chez des patients dont la maladie avait débuté après l'âge de 10 ans. Elles furent plus communes dans des cas où de nombreuses articulations se trouvaient atteintes et chez les séro-positifs. Ceux qui avaient subi une immobilisation prolongée ou dont la hanche était déjà impliquée, étaient plus souvent affectés; malgré cela le pronostic était aussi bon dans le groupe normal que dans l'anormal.

La distribution de l'atteinte articulaire ne ressemblait pas celle de la spondylarthrite ankylosante.

L'irite, l'exanthème, les nodules, les antécédents de famille et de traumatisme avant le début de la maladie étaient sans influence appréciable sur l'atteinte sacro-iliaque.

Le groupe avec des lésions sacro-iliaques moins graves (Grade I) comprend des malades ayant eu un début précoce et n'accusant pas d'associations cliniques spécifiques. Le Grade II groupe ceux accusant des lésions sacro-iliaques d'intensité modérée et comprend des sujets atteints de maladie de Still grave et classique, avec des nodules, séropositivité et une atteinte articulaire multiple impliquant aussi la colonne cervicale. Dans ce grade le sexe féminin prédomine. Le Grade III comprend les cas de fusion et de forte érosion de l'articulation sacro-iliaque. Chez eux, la maladie avait débuté plus tard et moins d'articulations se trouvaient atteintes; la hanche était impliquée le plus et la nuque le moins souvent. La plupart de ces malades étaient du sexe masculin.

Neuf sujets, diagnostiqués après l'admission comme cas de maladie de Still, développèrent à la suite une colite ulcéraive (4), arthropatie psoriasique (4) ou spondyl arthrite ankylosante (1), et furent, par conséquent, exclus de cette série. Au début, ces cas ne se distinguaient suffisamment du reste pour qu'on puisse prévoir les symptômes de la maladie "variante", tels que diarrhée, exanthème ou spondylite.

#### Sacro-iliitis en la enfermedad de Still

##### SUMARIO

Una serie de radiografías de la pelvis de sesenta sujetos que no sufrían de enfermedad crónica articular proporcionó los valores normales para los diferentes grupos según la edad, con el fin de comparar los hallazgos en 202 casos de enfermedad de Still.

Los cambios en la articulación sacro-iliaca ocurrieron en un 24 por ciento de esta serie de 202 casos, encontrándose igualmente distribuidos entre los sexos y observándose más frecuentemente en aquellos en quienes la enfermedad había aparecido después de los 10 años de edad. Las lesiones fueron más habituales en enfermos con un gran número de articulaciones afectas y en los sero-positivos. Los que estuvieron inmovilizados por largos períodos y los con una cadera ya implicada fueron los más comúnmente afectados, a pesar de lo cual el pronóstico en el grupo normal y en el anormal fué igualmente bueno.

La distribución de la afectación articular no recuerda a la vista en la espondilartritis anquilosante.

Iritis, erupciones, nódulos, antecedentes familiares y de trauma antes del comienzo de la enfermedad no fueron significantes en relación con la frecuencia de sacro-iliitis.

El grupo con menos acusadas lesiones sacro-iliacas (Grado I) comprendió a los pacientes que fueron atacados a edades más tempranas por la enfermedad y que no mostraron específicas asociaciones clínicas. El Grado II incluye aquellos con alteraciones sacro-iliacas de moderada intensidad y comprende pacientes con acentuada y clásica enfermedad de Still, con nódulos, seropositividad y una amplia afectación articular incluyendo la columna cervical. Existe un predominio del sexo femenino en este grado. El Grado III comprende aquellos con soldadura o erosiones extensas de la articulación sacro-iliaca. En estos pacientes la aparición de la enfermedad fué más tardía y menos articulaciones estaban atacadas. En ellos, sin embargo, la cadera fué la más, y la nuca la menos frecuentemente afectada. Aquí predominan los varones.

Nueve sujetos, diagnosticados en su admisión como casos de enfermedad de Still, desarrollaron colitis ulcerativa (4), artropatia psoriásica (4) o espondilartritis anquilosante (1), y fueron por lo tanto excluidos de la serie. Estos pocos casos no diferían al principio suficientemente de los demás para poder anticipar la aparición de los síntomas de la enfermedad "variante", tales como diarrea, erupción o espondilitis.