Case report

A case of Reiter's disease complicated by fulminating colitis

P. HICKLING,¹ M. F. R. MARTIN,¹ AND S. BROOKS²

From the Departments of ¹Rheumatology and ²Surgery, St James's University Hospital, Leeds

SUMMARY We describe a patient who, 6 months after the onset of Reiter's disease associated with a destructive peripheral arthritis and keratoderma blenorrhagica, developed fulminating colitis. The possible relationship between Reiter's disease and ulcerative colitis is discussed, and the need for further family studies to assess its validity is stressed.

There is considerable interest in the interrelationship of a number of diseases forming the seronegative spondarthritides, and the subject is discussed in detail in the latest edition of *Copeman's Textbook of the Rheumatic Diseases*.¹

The association of Reiter's disease with ankylosing spondylitis and psoriasis has been confirmed by several family studies. Its association with inflammatory bowel disease is less well documented. We describe here a case of severe Reiter's disease in a patient who subsequently developed fulminating ulcerative colitis.

Case report

A 40-year-old man presented in April 1980 with a 5-week history of polyarthritis, initially involving his knees and later his ankles, right shoulder, and right second metacarpophalangeal joint. He was being successfully treated for essential hypertension with hydralazine and atenolol. A full blood count was normal, ESR (Westergren) 53 mm/h, antinuclear factor negative, and differential agglutination test for rheumatoid factor negative.

Hydralazine was stopped and prazosin was substituted. A few days later he complained of dysuria and developed buccal ulceration and a scaly rash on his palms and soles. A diagnosis of Reiter's disease was made and he began erythromycin 250 mg 4 times a day. Over the next 2 days his arthritis deteriorated, he became unable to walk, and was admitted to hospital.

On examination an active synovitis was noted in both knees and ankles, with effusions. The second right metacarpophalangeal joint was painful and swollen. Examination of other joints including the spine showed them to be normal. Keratoderma blenorrhagica involved his hands and feet, and this subsequently spread to the arms and trunk, being associated with the development of marked nail dystrophy. A circinate balanitis was also present. At no time were there any signs or symptoms of ocular involvement, but there were erosive buccal and lingual mucosal lesions.

Investigations on admission. Full blood count normal, ESR 104 mm/h, HLA B27 positive, urethral swab sterile. A more vigorous swab, taken for isolation of chlamydia in McCoy cells, was negative after 48 hours' incubation. Midstream specimen of urine showed 50 polymorphs per high-powered field but was sterile on culture. Gonococcal fixation test negative, C3 2.56 g/l (0.55–1.2 g/l), C4 0.83 g/l (0.20–0.50 g/l) (normal range in parentheses). X-rays of his hands, knees, feet and sacroiliac joints were normal except for some soft tissue swelling in his knees.

Erythromycin was continued, and in addition he was given indomethacin and gentle physiotherapy for his joints. Topical clobetasol propionate was given for his keratoderma, 1% hydrocortisone cream was applied to his circinate balanitis, and he was discharged home. The keratoderma, however, became more extensive, resulting in nail shedding; his arthritis deteriorated, and he was readmitted to hospital. Haemoglobin was 10.5 g/dl with evidence of iron deficiency; white blood cell count was normal. Tar pomade was applied to his skin and potassium per-

Accepted for publication 6 April 1981.

Correspondence to Dr P. Hickling, St James's University Hospital, Beckett Street, Leeds LS9 7TF.

manganate soaks to his feet. Azathioprine 50 mg bd was begun, and his keratoderma slowly resolved.

In September 1980 he was readmitted with a week's history of increasing bowel frequency, bloody diarrhoea, slime, and tenesmus associated with lower abdominal pain. Immediately preceding his diarrhoea he had become constipated, a notable change, as his normal bowel habit was 4 times a day, and he developed an anal fistula. On examination he looked ill, anaemic, and had lost considerable weight. There was lower abdominal tenderness with guarding, and digital rectal examination was impossible owing to severe pain from the small anal fistula. He was noted to have a boutonnière deformity of his right index finger and flexion deformities of both ring fingers. Spinal examination was normal. Haemoglobin 3.6 g/dl, white blood count $3.8 \times 10^9/l$ (neutrophils 44%, lymphocytes 45%), platelets $269 \times 10^9/l$. ESR 120 mm/h. Blood urea and electrolytes were normal. Upper gastrointestinal endoscopy showed mild gastritis, but no blood, erosion, or ulceration in the upper gastrointestinal tract were noted. Azathioprine and indomethacin were discontinued, and a 6-unit blood transfusion was given.

A later sigmoidoscopy to 25 cm revealed petechial haemorrhages and congestion in the mucosa with some loss of vessel pattern. There was no frank pus or contact bleeding. Mucosal and fistula biopsies were taken; the fistula was laid open and packed. No specific histological features were seen in the biopsy of the fistula but the histology of the mucosa showed

an acute inflammatory cell infiltrate. He became pyrexial soon after this procedure and 3 days later developed profuse liquid diarrhoea with blood and slime and became severely ill. Abdominal x-ray revealed marked dilation of the transverse colon, and his white cell count rose to $15\,600 \times 10^9/l$, with a toxic neutrophilia. No pathogens were isolated from the faeces, and a diagnosis of an acute toxic colitis was made. He was started on a modified Truelove regimen² including salazopyrine 1 g tds. After a somewhat stormy course, during which the salazopyrine had to be stopped because of thrombocytopenia, his bowel symptoms gradually settled. A barium enema on 23 October 1980 showed an inflammatory colitis extending from the sigmoid colon to the caecum. The appearances were those of ulcerative colitis. A small-bowel barium meal was normal. There was no relapse of his Reiter's disease in this period, and he was eventually discharged home taking prednisolone 10 mg bd, piroxicam 20 mg daily, and naproxen 500 mg bd.

At follow-up 6 weeks later his colitis was quiescent and his keratoderma had almost cleared; his joints, however, had deteriorated, but there were no signs of active synovitis. A photograph of his hands at this time is shown in Fig. 1. X-rays of his hands showed evidence of an asymmetrical inflammatory polyarthritis involving some of the metacarpophalangeal joints, the left carpus, and many of the interphalangeal joints associated with juxta-articular osteoporosis, cartilage loss, and scattered bony erosions.

Fig. 1 The hands 6 months after onset of Reiter's disease. Note marked nail dystrophy, swelling of MCP and PIP joints, with flexion deformities of the ring fingers and a boutonnière deformity of the right index finger.

Discussion

To our knowledge there has been only 1 previous report of Reiter's disease associated with ulcerative colitis,³ though the association has been discussed in the French literature.^{4,5} Despite this dearth of case reports the possible association has been reported in the recent British literature⁶ and is alluded to in a standard rheumatology textbook,¹ though the reference quoted to substantiate this claim may well have been misinterpreted.⁷ Family studies have failed to reveal any increase in the prevalence of inflammatory bowel disease in the blood relatives of patients with Reiter's disease, though there is a significantly increased prevalence of psoriasis, sacroiliitis, and spondylitis.⁸

Of the seronegative spondarthritides ankylosing spondylitis is the only condition to show a definite association with ulcerative colitis. In these cases the spondylitis frequently antedates the colitis. There is also an increased prevalence of psoriasis and/or ankylosing spondylitis in patients with Crohn's disease and their first-degree relatives. Our patient had no family history of these associated diseases, though it is interesting to note that he is B27 positive. 65% of patients with Reiter's syndrome are B27 positive, and the frequency approaches 100% in those patients developing sacroiliitis, uveitis, or circinate balanitis. The most recent sacroiliac joint x-ray in our patient is suggestive of an early, asymptomatic left sacroiliitis.

Although our patient did not fulfil the classical triad of Reiter's disease, since he lacked conjunctivitis, the onset of large-joint arthritis, non-specific urethritis followed by severe keratoderma blenorrhagica, circinate balanitis, and oral ulceration is well recognised as an 'incomplete' form of the disease. Conjunctivitis is the most variable component of the triad and often goes undetected. In this case the arthritis was noticeable for its aggressive course, resulting in early joint deformities in the hands, and it is generally accepted that severe arthritis is often associated with severe mucocutaneous manifestations of the disease.

The radiological appearance of the large bowel was in keeping with an ulcerative colitis rather than with Crohn's disease despite the development of an anal fistula, which is a complication now generally regarded as common to both conditions.⁹ The possibility that the colitis was precipitated by azathioprine and/or indomethacin was considered, but the fact that the main exacerbation occurred 10 days after withdrawal of these drugs make it less likely. A coincidental colitis, of course, cannot be ruled out nor can a possible pre-existing low-grade inflammatory bowel disorder. The previous bowel frequency of 4

motions per day was considered normal by the patient, but the episode of constipation just prior to the acute colitis was a definite change in bowel habit. There had never been any previous history of the passage of blood or mucus.

We have described a B27 positive patient with incomplete Reiter's syndrome and severe keratoderma blenorrhagica who developed fulminating colitis during the initial presentation of his Reiter's disease. No firm conclusions can be drawn from this single case nor from the previous reports, but it would seem that in addition to the already well documented interrelationships between the seronegative spondarthritides, there may be a relationship between Reiter's disease and ulcerative colitis. The confirmation of this relationship must await further extensive family studies of patients with Reiter's disease. Conversely, in several large studies of patients with ulcerative colitis there has been no report of an increased incidence of Reiter's disease. This may be explained by the fact that in the majority of patients with Reiter's disease the symptoms are often self-limiting, the signs of urethritis and conjunctivitis can be overlooked, and the patients show some diffidence in giving such a history. The arthritis is less easily missed, though in a population of patients with ulcerative colitis it may be diagnosed as an enteropathic arthritis rather than being part of a Reiter's syndrome.

We are grateful to Dr J. R. Golding, Dr M. Waugh, and Mr T. G. Brennan for their permission to report this case.

References

- 1 Moll J M H, Haslock I, Wright V. In: Scott J T, ed. *Copeman's Textbook of the Rheumatic Diseases*. 5th ed. London: Churchill Livingstone, 1978: 578-91.
- 2 Truelove S C, Jewell D P. Intensive intravenous regimen for severe attacks of ulcerative colitis. *Lancet* 1974; ii: 1067-70.
- 3 Bourell M, Pawlowsky Y, Lenoir P, Simon M. Rectocolite ulcero-hémorragique et syndrome de Fiessinger-LeRoy-Reiter avec cellules à inclusions. *Sem Hop Paris* 1968; 44: 1005-8.
- 4 Cachin M, Antebi L. Colite ulcéreuse, érythème noueux et syndrome de Fiessinger-LeRoy-Reiter. *Sem Hop Paris* 1961; 37: 2724-8.
- 5 Hillemand P, Gilbrin E, Bensaude A, Delavière Ph. A propos des rectocolites hémorragiques avec manifestations articulaires. Leur rapports avec le syndrome de Fiessinger-LeRoy-Reiter. *Arch Fr Mal App Dig* 1960; 49: 613-21.
- 6 Editorial: Reiter's syndrome. *Lancet* 1979; ii: 567.
- 7 Billson F A, de Dombal F T, Watkinson G, Goligher J C. Ocular complications of ulcerative colitis. *Gut* 1967; 8: 102-6.
- 8 Lawrence J S. Family survey of Reiter's disease. *Br J Vener Dis* 1974; 50: 140-5.
- 9 De Dombal F T, Watts J McK, Watkinson G, Goligher J C. Incidence and management of anorectal abscess, fistula and fissure in patients with ulcerative colitis. *Dis Colon Rectum* 1966; 9: 201-6.