Adult-onset Still's disease: an unusual presentation of rubella infection

S.H.K. HUANG, MD; W.E. DECOTEAU, MD

Still's disease in the adult is uncommon.[1-3] Recently Harth, Thompson and Ralph[4] described four adults with this disease. Two of the patients had elevated rubella antibody titres, and rubella virus was isolated from the urine of one of them. In an accompanying editorial[5] Petty noted the "provocative and possibly coincidental similarities" between rubella arthritis and Still's disease, and suggested further studies to clarify the relation between rubella infection and Still's disease. Therefore, we are reporting the case of a 29-year-old woman with typical adult-onset Still's disease who had a brisk rise in the rubella antibody titre.

Case report

A 29-year-old woman presented to her family physician with a sore throat, fever, rash, polyarthralgia and severe myalgia of the back and legs. Her fever was remittent, with a daily spike in temperature to 40°C associated with chills and profuse sweating. The rash consisted of small, mildly pruritic, salmon-pink macules over the legs and, later, the back and arms. Characteristically, the rash disappeared during the night and reappeared the next day with the fever. The joint pain was migratory, affecting the wrists, elbows, knees, ankles and, finally, the small joints of the hands and feet.

At the time of physical examination her temperature was 40°C and pulse rate 100 beats/min. A 1/6 systolic ejection murmur was detected along the left sternal border. There was swelling of both knees and tenderness of both ankles and several metacarpophalangeal joints. The leukocyte count was 25.75 × 10^9/l (78% neutrophils), the erythrocyte sedimentation rate (ESR) 91 mm/h (Wintrobe) and the C-reactive protein value 4+. She was treated with acetylsalicylic acid, 6 g/d and penicillin G, 500 mg four times a day by mouth for, tentatively, rheumatic fever.

When there was no improvement a week later the woman was admitted to University Hospital. By this time she had lost 16 kg. She looked ill, had a temperature of 39°C and was having chills. A salmon-pink macular rash was noted over the extremities and the trunk. A grade 3/6 systolic ejection murmur was heard at the left sternal border. The liver and spleen were palpable, but there was no synovitis.

The hemoglobin level was 12 g/dl, the leukocyte count 9.2 × 10^9/l, the ESR 105 mm/h (Wintrobe) and the serum albumin level 2.9 g/dl. The serum levels of the following were persistently elevated: alkaline phosphatase, at 110 to 234 (normal 30 to 85) IU/l; lactate dehydrogenase, at 272 to 386 (normal 100 to 270) IU/l; and glutamic oxaloacetic transaminase, at 57 to 109 (normal 10 to 50) IU/l. The following gave either negative or normal results: tests for antinuclear antibody, DNA binding and rheumatoid factor; measurement of the serum levels of immunoglobulins A, G and M and the third component of complement; multiple blood and urine cultures; and serologic tests for cytomegalovirus, measles and mumps viruses, *Mycoplasma*, *Salmonella*, hepatitis B antigen, *Brucella*, *Toxoplasma*, *Yersinia*, *Treponema pallidum*, heterophil antibody and antistreptolysin O. The titre of rubella hemagglutination-inhibiting antibody was 1/1280 at the time of admission, but it rose to 1/5120 1 week later and 1/10 240 6 weeks later. Roentgenograms of the joints were normal. A technetium 99m liver and spleen scan revealed hepatosplenomegaly, with an increase in the splenic span to 20 cm. An echocardiogram, gallium 67 total body scan, abdominal ultrasonogram, chest roentgenogram and electrocardiogram were all normal.

The patient's temperature continued to spike to 40°C each day in association with chills and profuse sweating. Typically the rash faded in the evening and recurred the next day with the fever.

Four days after admission acute monoarthritis of the left wrist was noted. Aspirated synovial fluid had a leukocyte count of 40 × 10^9/l (93% neutrophils); a routine culture was negative. The inflammation spontaneously subsided in 48 hours but was followed 4 days later by acute arthritis of the right knee. Shortly thereafter the left fifth proximal interphalangeal joint became involved. Therapy with indomethacin, 50 mg three times a day, was begun; almost immediately the daily temperature spike was reduced and the rash cleared.

The patient was discharged feeling well following a total stay in hospital of 16 days. Follow-up visits 1 and 2 months later revealed no signs or symptoms, and the laboratory findings were all normal except for the rubella antibody titre, which had remained at 1/10 240.

Discussion

The patient we have presented had typical clinical features of adult-onset Still's disease. In addition, there was strong evidence of recent rubella infection because of a brisk rise in the titre of rubella hemagglutination-inhibiting antibody.

The relation between the patient's Still's disease and rubella in-

From the rheumatic disease unit, department of medicine, University Hospital, Saskatoon

Reprint requests to: Dr S.H.K. Huang, Rheumatic disease unit, Department of medicine, University Hospital, Saskatoon, Sask. S7N 0X0

fection is unknown, but some of the possibilities are as follows. First, rubella infection can produce a variety of musculoskeletal disorders, including polyarthritis of the small joints, which closely mimics adult-onset rheumatoid arthritis,[6-8] an illness that resembles juvenile rheumatoid arthritis,[9,10] and, as in our case, a systemic illness that has the typical features of adult-onset Still's disease. Second, it is possible that the rubella antibody response in this patient was nonspecific, as patients with various inflammatory disorders can show nonspecific increases in the titres of various antibodies.[11] However, the eightfold rise in the rubella antibody titre and the lack of elevation of other antibody titres in our patient make this unlikely. Finally, it is possible, though unlikely, that the concomitant occurrence of adult-onset Still's disease and rubella infection in our patient was purely coincidental.

Unfortunately, no attempt was made to isolate rubella virus from the synovial aspirate, which might have clarified the association between these two entities. However, this report should alert clinicians to study their patients with adult-onset Still's disease for the presence of rubella and other infections. Like many other diseases, adult-onset Still's disease may be a syndrome of many causes.

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continued from page 1274

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continued on page 1283