Acute mastitis; a novel presentation of relapsing polychondritis

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Summary: A 30 year old female with previous Crohn's disease presented with recurrent cutaneous vasculitis and polyarthrititis. She subsequently developed recurrent transient bilateral mastitis with auricular and laryngotracheal chondritis typical of relapsing polychondritis. Acute mastitis is a previously unrecognized association of this disorder.

Introduction

Relapsing polychondritis is a rare condition characterized by inflammation of cartilage at any site, often associated with vasculitis and autoimmune disorders.¹ We report a hitherto unrecognized association between relapsing polychondritis and acute mastitis.

Case report

A 30 year old unmarried woman was admitted with a rash and arthritis. She had been well until 2 months before admission when she had developed transient, recurrent painful swelling of her calves accompanied by widespread tender swelling of the joints.

In the past, she had been treated for Crohn's disease for 2 years from the age of 18. She had had no bowel symptoms since that time. There was no history of sexually transmitted disease or ocular symptoms. She had been taking the oral contraceptive pill until the onset of her illness. On admission her only medication was occasional aspirin. A sister had active Crohn's disease.

On examination there was a symmetrical, purpuric, macular, non-tender rash on the palms, soles and lower limbs below the knees. There was arthritis affecting the right metacarpo-phalangeal joints and knee and tenderness of the right achilles tendon.

Investigations revealed a haemoglobin of 11.6 g/dl with a normal white cell count and differential and normal platelets. The erythrocyte sedimentation rate

was 16 mm/h, C-reactive protein was 44.7 g/l (normal range < 10 g/l). Routine biochemistry was normal and the urinary sediment unremarkable. Urine, blood and throat cultures were sterile. Viral and ASO titres were negative as was syphilis serology. X-rays of the chest and affected joints were normal. Anti-nuclear antibodies were weakly positive with a diffuse pattern, other autoantibodies, including rheumatoid factor, were negative. Two separate skin biopsies of the cutaneous lesions showed acute capillaritis. Immunofluorescent staining was positive for complement but negative for immunoglobulins. Rectal biopsy suggested mild Crohn's disease.

Despite investigation, the nature of the patient's illness remained obscure. The rash faded and recurred every few days, eventually spreading to involve the buttocks and thighs. During admission, the patient noted a rapid onset of generalized tender swelling and erythema of the right breast. There was no discrete mass, no localized venous thrombophlebitis and no nipple discharge. The swelling resolved spontaneously within 24 hours.

The patient was discharged from hospital but was readmitted 2 weeks later with tender breast swelling and painful ears. On examination, there was bilateral diffuse mastitis, swelling of the external auricles with a zone of demarcation above the lobes and tenderness of the laryngeal cartilage. There was widespread arthritis and an extensive vasculitic rash. Laryngeal tomography and lung function testing were normal. Anti-type II collagen antibodies were negative.

A diagnosis of relapsing polychondritis was made and therapy started with oral prednisolone and azathioprine to which she made an excellent response. Eight months after starting therapy she remains well without recurrence of her symptoms.

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Discussion

Relapsing polychondritis is a rare disorder of obscure cause, although the demonstration of antibodies to cartilage and type II collagen² and the association with autoimmune disease suggests an immune-mediated aetiology. The diagnosis is based on the presence of 3 or more of the most common clinical features¹ which include: recurrent bilateral auricular chondritis, non-erosive seronegative inflammatory polyarthritis, nasal chondritis, ocular inflammation, respiratory tract chondritis and audiovestibular damage.

Although there is some debate about the stringency of application of the diagnostic criteria, the case presented above clearly fulfils the requirements for diagnosis, without necessity for histological confirmation.³

Skin involvement in relapsing polychondritis is fairly common, with cutaneous vasculitis, erythema nodosum-like lesions and other eruptions being reported in approximately one sixth of cases.¹ Association of relapsing polychondritis with immune-mediated and inflammatory diseases, including ulcerative colitis, is well recognized,¹ although Crohn's disease has not previously been described with relapsing

polychondritis. The associated disorder usually precedes the onset of relapsing polychondritis, as seen in this case.

Acute mastitis is usually associated with lactation or with viral illnesses, particularly mumps. Transient unilateral symptoms may be due to Mondor's disease,⁴ (superficial mammary thrombophlebitis). Acute recurrent mastitis has not been described in relapsing polychondritis, nor is it a recognized association of other autoimmune disorders. Although vasculitis, including thrombophlebitis, may occur in relapsing polychondritis, and cutaneous capillaritis was a prominent feature of this case, there was no clinical evidence of mammary thrombophlebitis. Thus the aetiology of the mastitis must remain obscure.

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