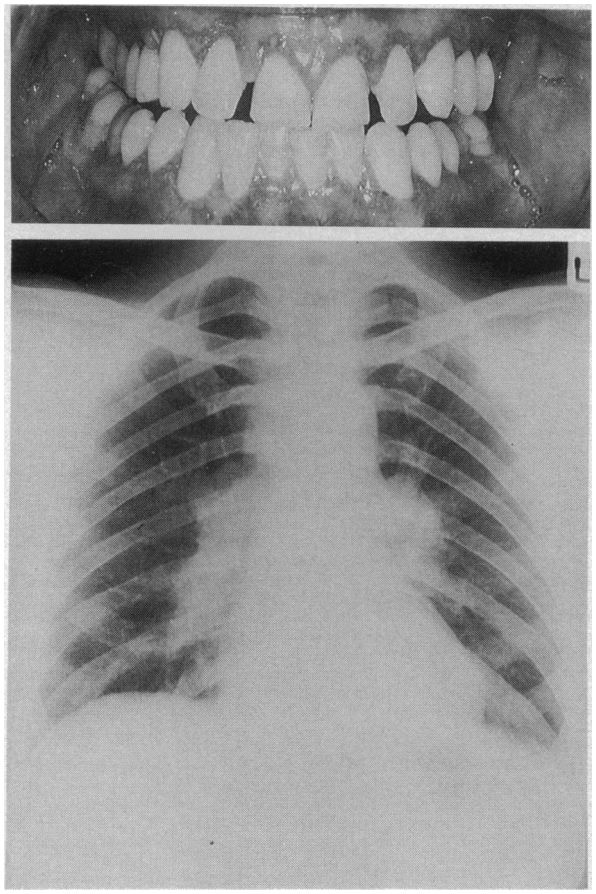
SHORT REPORTS

Sarcoidosis presenting as gingivitis

Sarcoidosis is a systemic granulomatous disease of unknown cause that may affect any tissue in the body. In the head and neck the most common signs, together with widespread systemic disease, are cervical lymphadenopathy and swelling of the parotid glands.[1] Gingival symptoms at any stage are rare,[2] and patients who have them usually present with other clinical features of the disease[2,3] or with gross gingival hyperplasia.[4,5] We describe a patient whose sole presenting complaint was persistent soreness and ulceration of the gums and who was subsequently found to have systemic sarcoidosis.

Case report

A 34 year old man was referred by his dentist with a two month history of soreness and ulceration of the gums; he had no other symptoms. There had been no improvement after scaling and courses of metronidazole and penicillin. He was taking no drugs although he had taken phenytoin and phenobarbitone for childhood epilepsy until he was 18. He had no history of allergy. No abnormality was found on general physical examination; in particular there was no lymphadenopathy or visceromegaly. On examination of his mouth the attached gums looked granulomatous with superficial ulceration and bled readily when probed.



Granulomatous appearance of attached gums on presentation and initial chest radiograph.

Histological examination of a gingival biopsy specimen showed a granulomatous chronic inflammatory infiltrate with numerous giant cells, many associated with birefringent material. No central caseation was seen, and Ziehl-Neelsen staining showed no acid fast bacilli. These findings were consistent with a diagnosis of sarcoidosis or Crohn's disease. A chest radiograph showed enlarged bilateral hilar and right paratracheal shadows consistent with adenopathy, and a Kveim test was reported positive. The full blood count; serum urea, electrolyte and calcium concentrations; and simple respiratory function values were all within normal ranges. Sarcoidosis was diagnosed.

After discussion with a physician the patient was reviewed regularly. After four months the gingival lesions had become less florid and serial chest radiographs showed reduced hilar shadowing. After nine months the oral and radiological signs had resolved. The patient remained well three years after presentation.

Comment

Sarcoidosis may include oral symptoms and clinically normal oral tissues may provide histological evidence of systemic sarcoidosis. It is rare, however, for oral symptoms alone to lead to the diagnosis.[1,2] Persistently sore, ulcerated gums that do not respond to local treatment and antimicrobial drugs suggest an underlying systemic disorder. The possibilities include erosive lichen planus, mucous membrane pemphigoid, Wegener's granulomatosis, blood dyscrasias, and Crohn's disease. This report of gingival symptoms as the sole clinical sign of the underlying disease confirms the place of sarcoidosis among the differential diagnoses.

1 Gold RS. Sager E. Oral sarcoidosis: review of the literature. *Journal of Oral Surgery* 1976;34:237-44.
2 Altman K, Robinson PD. Sarcoidosis with oral involvement. *Br Dent J* 1984;157:310-1.
3 Woodward RTM. Lofgrens syndrome. *Br Dent J* 1983;154:143-4.
4 Watts KD. Sarcoid of the gingivae: a case report. *British Journal of Oral Surgery* 1968;6:108-13.
5 Sloan PJM, O'Neil TCA, Smith CJ, Holdsworth CD. Multisystem sarcoid presenting with gingival hyperplasia. *British Journal of Oral Surgery* 1983;21:31-5.

(Accepted 23 February 1988)

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Long term follow-up of factitious anaemia

In Münchhausen's syndrome,[1] or chronic factitious disorder with physical symptoms,[2] some patients' factitious anaemia is due to self induced bleeding,[1] which is difficult to diagnose.[3] We report the long term follow up of four such patients.

Case reports

Case 1—This woman was a laboratory technician who had had iron deficiency anaemia with amenorrhoea and mild anorexia nervosa when aged 24. The anaemia's cause had not been established at first, but, eventually, devices for phlebotomy were discovered among her personal belongings and antecubital venepuncture sites found. She never admitted to bleeding herself. She has tolerated severe chronic anaemia, with a haemoglobin concentration usually below 50 g/l for 11 years, being able to swim and ski regularly and work part time. A long trial of psychotherapy for her borderline personality disorder was unsuccessful.

Case 2—This woman was 28 when iron deficiency anaemia and hypokalaemia were diagnosed but unexplained. In discussions she admitted to drawing blood from her antecubital veins daily or weekly and to taking diuretics and laxatives; she eventually developed anorexia nervosa. She had had severe emotional problems since puberty, including deep depression, anxiety, promiscuity, and drug abuse. She had regular psychotherapy but never stopped bleeding herself; her anorexia nervosa and bulimia also did not improve. In seven years of follow up her haemoglobin concentration was usually 40-80 g/l. She worked temporarily in a general practitioner's surgery but not recently. A borderline personality with autodestructive and antisocial behaviour was diagnosed.

Case 3—This patient was a student nurse of 21 when she was found to be anaemic and have fresh puncture sites in the antecubital fossae. As she tolerated haemoglobin concentrations of 50-60 g/l with few symptoms her anaemia was presumed to be long standing. The cause remained unknown until she admitted to bleeding herself. Later she began to take heroin; she left nursing and was lost to follow up. A psychiatrist treating her for opiate withdrawal described her as very introverted and diagnosed a severe borderline neurosis. Six years after factitious anaemia had been diagnosed she agreed to see one of us again. She was working and looked anaemic, but she refused to say whether she was still bleeding herself and to be examined or have a blood sample taken.

Case 4—This patient was 23 when she was admitted to hospital with iron deficiency anaemia. Extensive investigations failed to find the cause until she admitted to bleeding herself. After reluctantly accepting psychiatric help she stopped bleeding herself for six months but then relapsed. She was physically and professionally active while being chronically anaemic with a haemoglobin concentration of 45-60 g/l. At school she had looked anorectic (and still did), was

antisocial, and had intellectual difficulties. A psychotherapist she saw regularly diagnosed a narcissistic personality disorder. After four years of self bleeding she stopped for one year, and her haemoglobin concentration became 120-130 g/l.

Comment

Our patients had typical features of factitious disease,[1 2] three of them developing anorexia nervosa. They had been bleeding themselves for four to 11 years and were well adapted to severe chronic anaemia with haemoglobin concentrations rarely above 60 g/l. Their physical fitness was remarkable, and, notably, they tailored their bleeding such that it did not jeopardise an active life.

Although our patients showed little overt psychopathological behaviour, all had a borderline personality disorder.[2] Psychotherapy seems to be of little value in such disorders, and only one patient with factitious anaemia has been successfully treated with psychotherapy.[4] Nevertheless, such patients should be offered regular medical and psychiatric care; psychiatric consultation at an early stage in somatisation disorders reduces the costs of health care considerably, although it does not improve mental, physical, or social health.[5] Similarly, our patients appreciated regular meetings with doctors or psychotherapists to discuss their problems, although such meetings did not result in a cure.

1 Asher R. Münchhausen's syndrome. *Lancet* 1951;i:339-41.
2 Task Force on Nomenclature and Statistics. *Diagnostic and statistical manual of mental disorders*. 3rd ed. Washington, DC: American Psychiatric Association, 1980.
3 Bernard J, Najean Y, Alby N, Rain JD. Les anémies hypochromes dues à des hémorragies volontairement provoquées. Syndrome de Lasthénie de Ferjol. *Presse Méd* 1967;75:2087-90.
4 Yassa R. Münchhausen syndrome: a successfully treated case. *Psychosomatics* 1978;**19**:242-3.
5 Smith GR, Monson RA, Ray DC. Psychiatric consultation in somatization disorder. A randomized controlled study. *N Engl J Med* 1986;**314**:1407-13.

(Accepted 27 January 1988)

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Fatal cardiac failure due to myocardial microthrombi in systemic lupus erythematosus

Systemic lupus erythematosus has a variable clinical presentation and a high mortality. We report on a patient with the condition whose death was caused by acute circulatory failure resulting from diffuse thrombotic occlusion of the myocardial microcirculation.

Case report

A 22 year old woman with a six month history of "poor circulation" and exertional dyspnoea was admitted to hospital because of digital ischaemia. Examination showed lip ulceration, cervical lymphadenopathy, and vasculitic lesions on her hands and feet. She had a sinus tachycardia of 110 beats/minute, blood pressure of 130/100 mm Hg, and an apical mid-diastolic murmur.

Investigations showed that the haemoglobin concentration was 121 g/l, white cell count $24{\cdot}2\times10^9$/l, platelets 77×10^9/l, blood urea 13·8 mmol/l, and serum creatinine 135 μmol/l. Dipstick testing of urine for blood and protein yielded positive results, and red cell casts were seen on microscopy. An electrocardiogram showed generalised S-T segment depression of 1 mm, and a chest *x* ray film showed slight cardiomegaly with no pulmonary congestion. Echocardiography confirmed a moderately severe mitral stenosis. The left atrium, right atrium, and right ventricle were enlarged, but the left ventricle was of normal size. Serum titres of antinuclear antibody were 1/640 (IgG) and 1/80 (IgM), and the concentration of antibody to double stranded DNA was 80 units/ml (normal range 0-25). Serum was strongly positive for IgG and weakly positive for IgM anticardiolipin antibodies on enzyme linked immunosorbent assay (ELISA) with Loizou's method. Serum C3 concentration was 0·58 g/l (normal range 0·75-1·50). A coagulation screen was within normal limits; the test for lupus anticoagulant was not done.

Her condition deteriorated with confusion, oliguria, and rising blood urea concentrations. Aggressive systemic lupus erythematosus was diagnosed and methylprednisolone and cyclophosphamide started, the intention being to start plasma exchange and dialysis the next day. Within 24 hours, however, she developed profound hypotension and peripheral circulatory failure and died.

At necropsy the kidneys showed focal glomerulosclerosis, proliferative glomerulonephritis, and a superimposed necrotising glomerulitis. The glomerulitis was related to afferent arteriole microthrombi and to fibrinoid vasculitis, which was also present in the lungs, liver, and pancreas. No abnormality of the brain was seen and no source of infection found. There was moderately severe mitral stenosis with fusion of the valve cusps due to warty, erythematous endocardial nodules. Aggregates of fibrin, inflammatory cells, and DNA debris were attached to the valve surface and adjacent endocardium. The main coronary arteries were mildly atheromatous. Many of the intramyocardial arteries contained occlusive thrombi unaccompanied by vasculitis but surrounded by extensive recent myocardial necrosis.

The findings at necropsy were consistent with systemic lupus erythematosus but also showed mitral stenosis caused by Libman-Sacks endocarditis. Death resulted from diffuse acute myocardial necrosis due to extensive thrombi in the microvasculature.

Comment

Though renal failure is one of the major causes of death in systemic lupus erythematosus and our patient had severe lupus nephritis, the degree of uraemia would not explain the fatal circulatory failure. There was no evidence of overwhelming infection, of lupus in the cerebrum, or of appreciable atheroma in the large vessels. Libman-Sacks endocarditis rarely causes severe valvular dysfunction, although isolated reports have documented mitral regurgitation and combined stenosis and regurgitation similar to that in our patient.[2] In our case, however, there was no clinical or radiological evidence of pulmonary oedema or central cyanosis, and the mitral stenosis is thus unlikely to have led to the fatal circulatory failure.

Death from fulminant cardiac necrosis resulting from thrombotic occlusion of the myocardial microcirculation has not been described previously.[1 3] Circulating antiphospholipid antibody may have played a part, as the lupus anticoagulant has been associated with arterial and venous thromboses.[4] Thus thrombolytic or fibrinolytic treatment, or both, might be beneficial. Kant has reported resolution of microvascular thrombosis in patients with systemic lupus erythematosus treated by ancrod, the defibrinogenating enzyme obtained from the venom of the Malayan pit viper.[5]

1 Loizou S, McCrea JD, Rudge AC, Reynolds R, Boyce CG, Harris EN. Measurement of anti-cardiolipin antibodies by an enzyme-linked immunosorbent assay (ELISA): standardisation and quantitation of results. *Clin Exp Immunol* 1985;**62**:738-45.
2 Evans DTP, Sloman JG. Mitral stenosis and mitral incompetence due to Libman-Sacks endocarditis with mitral valve replacement. *Aust NZ J Med* 1981;**11**:562-8.
3 Correia P, Cameron JS, Lian JS, *et al*. Why do patients with lupus nephritis die? *Br Med J* 1985;**290**:126-31.
4 Hughes GRV. Thrombosis, abortion, cerebral disease, and the lupus anticoagulant. *Br Med J* 1983;**287**:1088-9.
5 Kant KS, Pollak VE, Dosekun A, *et al*. Lupus nephritis with thrombosis and abnormal fibrinolysis: effect of ancrod. *J Lab Clin Med* 1985;**105**:77-88.

(Accepted 2 February 1988)

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Effect of low dose heparin on blood loss at caesarean section

There is good evidence for the efficacy of low dose heparin in preventing venous thrombosis and pulmonary embolism after surgery,[1] but prophylactic heparin is not usually given before caesarean section. The main reason is the fear of increasing the amount of bleeding in an operation which has a high operative blood loss, but there is also the difficulty of predicting the effect of heparin in pregnancy, where the coagulation mechanism may already be disturbed. We decided to measure the effect of heparin on blood loss during caesarean section.

Patients, methods, and results

Fifty patients were given a subcutaneous injection of either 5000 units of sodium heparin or isotonic saline one hour before the operation and similar injections twice daily for five days after the operation. There were no significant differences in age, parity, blood pressure, height, weight, and gestation between the two groups. Patients were excluded from the study if they had a placenta praevia, multiple pregnancy, pregnancy induced hypertension, antepartum