periodic acid Schiff (PAS)-positive foamy macrophages containing rod-shaped bacilliform bodies on electron microscopy. Similar appearances were seen in a muscle biopsy specimen.² Addition of oxytetracycline, 250 mg four times daily, induced complete symptomatic remission and the steroid dosage was slowly reduced. Eight weeks later a repeat biopsy specimen still showed PAS-positive macrophages but there was evidence of improvement on electron microscopy with replacement of the bacteria-like rods by intracellular membranous material.³ Three months after diagnosis, however, when taking 5 mg of prednisolone daily together with oxytetracycline, weakness and fever recurred, accompanied by severe headache and vomiting. He was drowsy and febrile (39°C) with neck stiffness but no focal neurological signs and normal fundi.

The results of investigations on admission were as follows: leucocyte count $12 \times 10^9/l$ (80% neutrophils); blood culture no growth; throat swab no growth; and cerebrospinal fluid (CSF)—pressure 360 mm, protein 0.9 g/l, microscopy 900 polymorphs/mm³, Gram stain no organisms, and no growth on culture. Countercurrent immunoelectrophoresis (CIE) indicated presence of *Neisseria meningitidis* type-Z antigen. Benzylpenicillin, 1 megaunit six-hourly, was substituted for the oxytetracycline and the prednisolone increased to 30 mg/day. Rapid recovery ensued, with return of CSF to normal. Two weeks later when the dose of prednisolone had been reduced to 10 mg daily meningism recurred, again responding to penicillin and an increased dose of prednisolone. In the next three weeks he had several further episodes of clinical meningitis. Brain scan, electroencephalography, and skull and sinus x-ray investigations were performed but failed to show evidence of any focal intracranial abnormality.

In view of these relapses cephaloridine was substituted for penicillin and metronidazole added to cover the possibility of anaerobic infection but a further recurrence occurred. No evidence of infection was found during any of these episodes and CIE failed to demonstrate further antigen. Cytological examination of CSF, however, showed the typical PAS-positive macrophages of Whipple's disease (figure). In the fifth attack of meningitis no antibiotic was given but the dose of prednisolone was raised and the effect of the increased dose observed. A rapid clinical response occurred within 24 hours with disappearance of the CSF and blood leucocytosis as before.

Prednisolone, 20 mg/day, with reintroduction of oxytetracycline prevented further symptoms until five months later, when another attempt at steroid reduction resulted in meningitis, which again resolved completely on returning the prednisolone dose to 20 mg/day without additional antibiotic.

Course of the disease showing changes in CSF and peripheral blood leucocytes with treatment.

Discussion

Widespread foci of PAS-positive macrophages have been found in the brain and meninges of patients with Whipple's disease at necropsy, even in the absence of clinical neurological abnormality.⁴

Our inability to show organisms in the CSF; the failure of antibiotics to influence the course of the illness; the complete clinical resolution on increased prednisolone dosage alone; and the finding of typical PAS-positive phagocytes in the CSF must strongly support a diagnosis of Whipple's meningitis. Whether this was induced by an

immune response to viable intracerebral Whipple's organisms or to intracellular PAS-positive material alone in the meninges is unknown.

The importance of discovering type-Z meningococcal antigen on one occasion is uncertain and may be a spurious result. It may, however, indicate the presence of an antigen common to type Z meningococcus and to the Whipple's organism, as has been noted with *Shigella*, *Streptococcus*, and *Escherichia coli* antigens.⁵

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¹ Schwartzova, K, Schwartz, A, and Marke, J, *Plzensky Lek Sborn*, 1967, **28**, 99.

² Swash, M, *et al*, *Gut*, 1977, **18**, 800.

³ Roberts, D M, *et al*, *Journal of Pathology*, 1970, **100**, 249.

⁴ Stoupe, N, *et al*, *Journal of Neurology, Neurosurgery, and Psychiatry*, 1969, **32**, 338.

⁵ Keren, D F, *et al*, *Johns Hopkins Medical Journal*, 1976, **139**, 51.

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Anorexia nervosa and Turner's syndrome

The concurrence of anorexia nervosa and Turner's syndrome (gonadal dysgenesis) was first reported in 1963.¹ Nine cases of XO gonadal dysgenesis and two of XO/XX (mosaic) gonadal dysgenesis have now been reported.² It is debatable whether this association occurs with a greater than chance frequency and, if so, what the relation between these disorders is. We describe the first reported British case of anorexia nervosa in association with Turner's syndrome.

Case report

A 23-year-old postgraduate student had developed a close relationship with her mother after her father died when she was 8. Turner's syndrome was diagnosed when she was a child, karyotyping showing a 45,XO chromosome pattern. She had the typical features of short stature (147 cm), webbing of the neck, low hairline, "shield-like" chest, absent breast development, sparse pubic hair, primary amenorrhoea, a rudimentary uterus, and skeletal abnormalities. At age 18 she began treatment with oestrogens to induce a more feminine appearance and to induce withdrawal bleeding. Two and a half years later the oestrogens were stopped because she developed jaundice.

She was overweight and was frequently teased by children at school about her appearance. At 18 years she weighed 57.5 kg, but in her first year at university she became a vegetarian and began to diet, largely eliminating fats and carbohydrates. Within six months she had lost 15 kg. This was followed by short periods of overeating, for which she had guilt feelings. Dieting continued for four years and she considered the weight loss as desirable and not excessive. On admission to hospital she weighed 35 kg. Self-induced vomiting to avoid weight gain was suspected. Anorexia nervosa was diagnosed according to Feighner's criteria.³

Comment

One theory of anorexia nervosa is that it results from a wish to regress to an asexual prepubertal state to avoid mature biological sexuality.⁴ This seems an unlikely explanation for a girl with Turner's syndrome, especially of the 45,XO variety, who remains largely prepubertal. Although patients with Turner's syndrome may pass through a rudimentary puberty, our patient could not remember either physical or psychological changes, indicating that adolescent sexual conflict was unlikely. But she developed anorexia nervosa while undergoing an artificial puberty in her late teens and stated that she was not enthusiastic about the changes induced by oestrogens. The regression theory might therefore still apply.

Alternatively, an association between Turner's syndrome and anorexia nervosa might result from the low self-esteem associated with the genetically determined physical defects. Our patient was self-conscious about her appearance, and her illness may have been

precipitated by leaving a protective home environment for university, where she faced major interpersonal difficulties as a consequence of her low self-esteem and appearance.

Should the association between anorexia nervosa and Turner's syndrome occur with greater than chance frequency, the latter may not be the only condition which shows this association. Anorexia nervosa may well be a manifestation of a wide range of both psychological and organic disturbances,⁵ and not a unitary syndrome with a common cause. Epidemiological studies of the association with other disorders seems desirable.

¹ Pitts, E N, and Guze, S B, *American Journal of Psychiatry*, 1963, **119**, 1100.

² Kron, J L, et al, *Archives of General Psychiatry*, 1977, **34**, 332.

³ Feighner, J P, et al, *Archives of General Psychiatry*, 1972, **26**, 57.

⁴ Crisp, A H, *Proceedings of the Royal Society of Medicine*, 1977, **70**, 464.

⁵ *British Medical Journal*, 1978, **1**, 5.

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Sexual intercourse and angina pectoris

With the incidence of ischaemic heart disease rising in the younger population the number of patients with this condition and sexual problems may be expected to increase. Hence doctors should be familiar with the physiological responses to sexual intercourse so that they can counsel patients accurately.

Patients, management, and outcome

A total of 35 patients aged 36-70 with angina were examined at monthly intervals. Each completed a questionnaire on his family life, including sexual activity. This showed that 29 had intercourse more than once a week, 19 developing angina on most occasions. Four had palpitations during intercourse, and six abstained.

Patients received basic advice on preparing themselves and their environment for intercourse. They were advised to warm the bedroom and sheets, to avoid intercourse soon after a meal or bath, and specific questions relating to each individual were then answered. Fourteen patients underwent 24-hour electrocardiographic recordings and indicated in diaries the time and duration of any activity, including intercourse. The recordings were analysed for heart rate and arrhythmias. All patients received beta-blockers and those who had had 24-hour recordings had repeat recordings. If pain occurred in spite of basic advice and beta-blockers patients were advised to take isosorbide dinitrate sublingually 10 minutes before intercourse.

The patients with symptoms became pain free during intercourse, though six required nitrates in addition to beta-blockade. Four of the six who had abstained resumed sexual activity. Two patients with palpitation developed supraventricular tachycardia and two sinus tachycardia. Beta-blockers abolished the supraventricular tachycardia and complaints of palpitation. The table shows that the sinus rate during intercourse was no more than that during normal daily activity and was appreciably reduced by beta-blockade.

Discussion

During sexual intercourse the heart rate, blood pressure, and respiratory rate rise.¹ An excessive increase in heart rate and blood pressure in a patient with coronary artery disease might induce angina, infarction, or dysrhythmia as myocardial oxygen demand

24-Hour maximal heart rates (mean $\pm$ SEM)

	Before beta-blockade	After beta-blockade	P
24-Hour maximal sinus rate excluding sexual intercourse	124.0 $\pm$ 7.2	82.1 $\pm$ 6.0	<0.001
Maximal heart rate during intercourse	122.2 $\pm$ 7.1	82.0 $\pm$ 2.8	<0.001

exceeds supply. Hellerstein and Friedman² showed that the average maximal heart rate in middle-aged married couples during intercourse was 117 beats/min, compared with 120 beats/min during other activity. This similar heart rate response has been confirmed in this study. As the blood pressure rises only moderately when intercourse takes place in the security of the home environment,³ there is little evidence to suggest that the stress to the myocardium during intercourse is any greater than that during normal daily activity. In spite of this, in a study of 100 patients after infarction, though 90 returned to work, only 40 resumed normal sexual activity.⁴ Sexual activity does not appear to be related to an increased mortality rate in patients with heart disease who enjoy intercourse with their spouses in their own home. The person most at risk is usually middle-aged, having extra-marital relations.⁵

Ignorance of the effects of intercourse on the heart and fear of death are overcome by time and informed comment. In this series of 35 patients only two were not enjoying angina-free intercourse after specific simple management. They mostly benefited by having the subject raised, discussion, and reassurance. In two patients 24-hour electrocardiographic tape recordings disclosed a dysrhythmia which responded to treatment enabling normal sexual activity to be maintained. Furthermore, the tape recordings after beta-blockade showed an appreciable reduction in sinus rate during intercourse. By preventing the rise in heart rate, and probably that in blood pressure, the incidence of angina during intercourse decreased. While sublingual nitrates effectively relieve angina their use removes the spontaneity of intercourse allowed by beta-blockade.

It should be routine policy to advise patients and their spouses on sexual activity, whether they have had an infarction or are regularly attending with angina pectoris. Most patients with ischaemic heart disease can enjoy normal sexual relations without risk.

¹ Bartlett, R G, *Journal of Applied Physiology*, 1956, **15**, 469.

² Hellerstein, H K, and Friedman, E H, *Archives of Internal Medicine*, 1970, **125**, 987.

³ Nemec, E D, Mansfield, L, and Kennedy, J W, *American Heart Journal*, 1976, **92**, 274.

⁴ Bloch, A, Maeder, J P, and Haissly, J-C, *American Heart Journal*, 1975, **90**, 536.

⁵ Ueno, M, *Japanese Journal of Legal Medicine*, 1963, **17**, 536.

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Proliferative retinopathy in a patient with diabetes mellitus and idiopathic haemochromatosis

The belief was once widely held that patients with diabetes mellitus and idiopathic haemochromatosis were not prone to develop diabetic microangiopathy.^{1,2} Recently there have been reports of glomerulosclerosis with typical Kimmelstiel-Wilson nodules³ and of diabetic retinopathy^{4,5} in such patients. In two studies retinopathy was noted in roughly 30% of diabetic patients with haemochromatosis.^{4,5} These reports suggest that the long survival of treated patients with haemochromatosis shows that the vascular complications are related to the duration of the diabetes in the same way as those associated with idiopathic diabetes. Nevertheless, a feature of these reports has been the mildness of the retinopathy, which was not considered to have produced visual disturbance. Proliferative retinopathy has not been reported by any of these workers. We report incapacitating proliferative retinopathy in a patient with diabetes mellitus and idiopathic haemochromatosis.

Case report

A Caucasian man was aged 26 years when diabetes was diagnosed in 1956. His father had mild diabetes but there was no family history of haemo-