Discussion

Primary arteritis of the aorta is a distinct entity. Earlier descriptions of Takayasu's disease,¹ brachial arteritis,² and aortic arch syndrome³ cover only one facet, and may be misleading. More recent reports^{4, 5} based on necropsy or angiographic findings suggest that occlusive changes may affect any part of the aorta and its branches.

This present case presented with renovascular hypertension. Patchy narrowing of the abdominal aorta with its accompanying calcification and partial occlusion of the left renal artery with non-specific histological findings strongly suggest the diagnosis of primary aortitis. Removal of the stenosed vessel and affected kidney restored the blood pressure to near normal levels.

The aetiology of this apparently rare condition remains obscure. The disease may well represent the end result of a variety of different initiating agents, the anatomical site of disease being modified by other unknown variables.

¹ Takayasu, M, *Acta Societatis Ophthalmologicae Japonicae*, 1908, **12**, 554.

² Koszewski, B J, *Angiology*, 1958, **9**, 180.

³ Judge, R D, *et al*, *American Journal of Medicine*, 1962, **32**, 379.

⁴ Schrire, V, and Asherson, R A, *Quarterly Journal of Medicine*, 1964, **33**, 439.

⁵ Munoz, N, and Correa, P, *American Heart Journal*, 1970, **80**, 319.

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Tracheo-oesophageal fistula associated with hormonal contraception during pregnancy

Progestogen/oestrogen preparations have been suspected as potentially teratogenic drugs for several years but reports are conflicting. It is therefore important to document all cases where congenital abnormality is associated with the ingestion of the drugs during pregnancy.¹ I report here such a case.

Case report

A 23-year-old primigravida was booked for hospital delivery at about the 20th week of pregnancy. Her last menstrual period had been on 22 March 1975, thus making her expected date of delivery 29 December 1975. Nevertheless, she had taken the oral contraceptive Ovral (Di-norgestrel 0.5 mg, and ethinyl oestradiol 0.05 mg) regularly until June 1975. At this time a vaginal examination performed to elicit the cause of 12 weeks' amenorrhoea confirmed a 12 weeks' size pregnancy. The patient attended antenatal clinic regularly for a total of 14 visits. The size of the uterus was initially small for dates and later large for dates owing to hydramnios. The result of a glucose tolerance test was normal and an x-ray film showed a normal singleton fetus. The hydramnios, therefore, was of unknown origin. On 20 January 1976, when the pregnancy was post term by dates, there was a slight rise in blood pressure and induction of labour was considered. Clinically the fetus was of good size and the amniotic fluid gave a positive "bubble" test and contained 20% fat staining cells with Nile blue sulphate reagent. Induction of labour was planned, and was performed on 23 January 1976. Hind-water and fore-water rupture of membranes resulted in the drainage of over 4000 ml of clear liquor. A syntocinon infusion at 1 μ unit/minute was begun and the dose doubled every 30 minutes to 4 μ unit/minute, when labour was established. The latent phase of the first stage of labour lasted four hours and the active phase six hours. A delayed second stage was terminated by an easy vacuum extraction and the third stage of three minutes was uneventful. The puerperium was normal apart from a slight rise of blood-pressure for the first 24 hours after delivery.

The baby girl weighed 3000 g and the Apgar score at one and five minutes was 8. A nasogastric tube could not be passed and an oesophageal atresia with a fistula between the distal segment and the trachea was confirmed on x-ray examination. A repair was performed on the same day. Her postoperative progress was stormy. A gastrostomy was performed on 29 January 1976 and closure of fistulous track on 19 February 1976. After the latter her general condition improved, but she died from bronchopneumonia on 28 March 1976.

Discussion

This patient inadvertently took Ovral for the first three months of her pregnancy and gave birth to an infant with a tracheo-oesophageal fistula, an abnormality whose normal occurrence is 1 in 3000. This association was also reported by Oakley *et al*² in a letter to the *Lancet* on the relation between hormonal pregnancy tests and congenital malformation.

Until it is proved that there is no relation between these preparations and congenital abnormality it is prudent to ensure that no patient is prescribed oral contraceptives unless pregnancy has been definitely excluded.³

¹ Nora, J L, and Nora, A H, *New England Journal of Medicine*, 1974, **291**, 732.

² Oakley, G P, Flynt, W J, and Falck, A, *Lancet*, 1973, **2**, 256.

³ *British Medical Journal*, 1974, **4**, 485.

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Neurological manifestations in a patient with filariasis

Filariasis, in particular infections with *Loa loa* and *Depetalonema perstans*, may occasionally develop some neurological complications.^{1, 2} However, these are neither common nor well known. We believe it important to call attention to them because they can be mistaken for a neurological disease of different aetiology. For that reason we report following case.

Case report

A 28-year-old Negro male born in Rio Muni, West Africa, was admitted to hospital because 10 days previously he had started to complain of vomiting, unsteadiness, loss of hearing on the left side, and paraesthesiae on the left side of the face. Clinical examination disclosed hepatosplenomegaly, nystagmus, sensorineural hypacusis, and signs of left facial paresis. Blood analysis showed eosinophilia of 18% and numerous microfilariae, later identified as *Loa loa* and *D perstans*. No plasmodia were seen. Blood chemistry, urine analysis, stool examination, and serological tests for *Echinococcus granulosus* and toxoplasma were all normal. In a bone marrow aspiration a high eosinophil count was the only abnormality. An electrocardiogram showed T wave inversions on V1 to V5. Lumbar puncture yielded clear fluid with 45 mg/100 ml (0.45 g/l) protein, 76 mg/100 ml (4.2 mmol/l) glucose, and 45 cells. The presumptive diagnosis of cerebellopontine angle tumour or acoustic neuroma was made. Cerebral angiographic, pneumoencephalographic, and echoencephalographic studies were all normal.

One month after symptoms began spontaneous recovery set in, and all the symptoms completely disappeared without treatment in two more weeks.

Discussion

The clinical picture in this case suggested a lesion in the cerebellopontine angle which subsided spontaneously, leaving no sequel about two months later. A careful clinical examination showed no specific cause for it. The only abnormal finding was a severe infection of the blood with *Loa loa* and *D perstans* microfilariae. Both species and *Meningonema peruzzi* have been implicated in the production of cerebral symptoms. Some authors^{1, 3, 4} have reported cases of fatal meningoencephalitis or cases with focal symptoms that subsided spontaneously.⁵

We were unable to find microfilariae in the CFS as has been reported in other cases. We cannot be certain that the patient's symptoms were due to filariasis, and we cannot rule out a viral infection or even a minor cerebrovascular accident. Nevertheless, it is possible that filariasis was responsible for neurological symptoms, and we emphasise the need to consider this possibility in the diagnosis of any neurological syndrome in a patient coming from countries where these helminthiasis are endemic.

The pathogenesis is uncertain. It may be local oedema due to migration of an adult worm, or a vascular lesion, or even vascular block due to the presence of large number of microfilariae in cerebral vessels.

¹ van Bogaert, L, *et al*, *Journal of Neurology, Neurosurgery, and Psychiatry*, 1955, **18**, 103.
² Collomb, H, *et al*, *Bulletin de la Société de Pathologie Exotique et de ses Filiales*, 1969, **62**, 907.
³ Orihel, T C, *American Journal of Tropical Medicine and Hygiene*, 1973, **22**, 596.
⁴ Kivits, M, *Annales de la Société Belge de Médecine Tropicale*, 1952, **32**, 235.
⁵ Browne, S G, *Journal of Tropical Medicine and Hygiene*, 1954, **57**, 229.

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Treatment of palindromic rheumatism with D-penicillamine

Palindromic rheumatism is uncommon but important, firstly because it may lead to chronic rheumatoid arthritis, and, secondly, as a cause of disability if attacks are frequent. It is regarded as a variant of rheumatoid arthritis, and this is supported by the finding of positive tests for rheumatoid factor and other immunological abnormalities.¹ Mattingly² reported that gold treatment appeared to halt the attacks, often within a few weeks, though relapse was frequent when treatment was stopped or the dose reduced. The similarity of the effects of gold and D-penicillamine in chronic rheumatoid arthritis³ suggested the possibility of using D-penicillamine for palindromic rheumatism.

Patients, methods, and results

Details of the patients treated are shown in the table. All had a typical history of recurrent acute attacks of joint pain and swelling affecting various joints, each lasting for two or three days. They were kept under observation for a sufficient period of time to record the frequency of the attacks before treatment was started with D-penicillamine in a dose of 250 mg. daily. All have been followed up for at least a year while receiving the treatment. In four patients no further attacks occurred after starting D-penicillamine and dosage was not increased. One patient subsequently reduced the dose of D-penicillamine progressively and had a further attack while receiving 500 mg weekly; she has again been free of attacks on 250 mg daily. No side effects occurred in these four patients. One patient (case 3) continued to have attacks of arthritis and the dose was increased at fortnightly intervals up to 750 mg daily. Treatment was interrupted by a rash, which developed after four weeks; the drug was restarted when the rash disappeared and it did not recur. In this patient the frequency and severity of attacks were reduced.

Details of patients with palindromic rheumatism treated with D-penicillamine, including the number of attacks of arthritis during the period of observation before the start of treatment and for one year after

Case number	Age	Sex	Duration of disease (years)	Latex titre	Daily maintenance dose (mg)	Number of attacks	
						Before treatment	After treatment
1	45	F	8	neg	250	16 in four months	None for one year
2	33	F	7	neg	250	12 in 10 months	None for one year
3	65	F	5	1/80	750	25 in six months	Four in one year
4	19	F	3	1/20	250	8 in two years	None for one year
5	52	M	4	neg	250	17 in three years	None for one year

Discussion

Like gold, D-penicillamine appears to be a useful therapeutic agent in palindromic rheumatism. Though the course of palindromic rheumatism is unpredictable, it is most unlikely that such a sudden and spectacular change in the frequency of attacks would have occurred by chance or continued for so long. It is a considerable advantage that D-penicillamine can be used in a small dose which is unlikely to cause side effects. The rarity of the disease would make controlled trials difficult, but it would be desirable to confirm the effectiveness of D-penicillamine and to investigate the possibility that this drug might prevent the development of chronic rheumatoid arthritis. D-penicillamine treatment should certainly be considered in patients with frequent attacks of palindromic rheumatism.

¹ Williams, M H, *et al*, *Annals of the Rheumatic Diseases*, 1971, **30**, 375.
² Mattingly, S, *Annals of the Rheumatic Diseases*, 1966, **25**, 307.
³ Huskisson, E C, *et al*, *Annals of the Rheumatic Diseases*, 1974, **33**, 532.

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Megaloblastic anaemia due to pyridoxine deficiency associated with prolonged ingestion of an oestrogen-containing oral contraceptive

Pyridoxine deficiency is an uncommon cause of hypochromic and megaloblastic anaemias. I report here a case of macrocytic hypochromic megaloblastic anaemia responding to pyridoxine in a patient taking an oestrogen-containing oral contraceptive agent, in the absence of any other apparent cause for pyridoxine deficiency.

Case report

A 46-year-old Caucasian woman presented with tiredness and mild depression. There was nothing significant in her past medical history and the only drug she was taking was the oral contraceptive agent Gynovlar (ethinyl-oestradiol 50 µg and norethisterone acetate 3 mg), which she had used continuously for the previous seven years. The only abnormal finding on examination was clinical anaemia. Investigations showed haemoglobin 7.6 g/dl, a hypochromic macrocytic film, a reticulocyte count of less than 1 %, and a normal white cell count. The bone marrow showed "appearances typical of a frankly megaloblastic anaemia" and special stains failed to show any free iron. Serum folate was low at 1.0 µg/l, but red cell folate was normal at 200 µg/l. Urea, electrolytes, glucose, calcium, phosphate, alkaline phosphatase, and thyroid function tests were all normal. Stools were negative for occult blood. Tests for antinuclear factor, intrinsic factor antibody, and parietal cell antibody were all negative, and a chest x-ray was normal. Barium meal and follow-through showed "gastric rugae, probably flatter than the average, and compatible with pernicious anaemia. The small