SHORT REPORTS

Oral tuberculosis—an unusual presentation

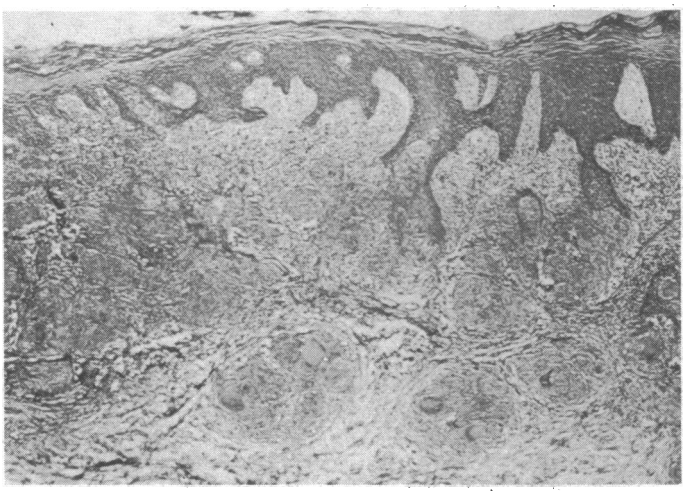
Descriptions appear occasionally of oral tuberculous lesions which may be either apparently solitary or, more commonly, associated with more widespread disease. The lesions are generally described as proliferative, granulomatous, or ulcerated. The case reported here presented in a quite different and unusual form.

Case report

A 56-year-old woman complained of soreness of the right cheek for two months. On examination there was a faint and diffuse lesion some 2 cm in diameter on the buccal mucosa. The appearance was of a lightly indurated reddened patch with some intrinsic faint white areas but no ulceration. No other significant active lesions were found on clinical or radiographic examination, although chest radiographs showed calcified hilar foci. Blood examination showed WBC $7{\cdot}1\times10^9$/l (7100/mm^3); RBC $4{\cdot}7\times10^{12}$/l (4 700 000/mm^3); Hb 13·3 g/dl; ESR 26 mm in 1 h. There was no relevant personal medical history but the patient's father and her daughter had had pulmonary tuberculosis respectively 17 years and 15 years previously.

A biopsy specimen of the lesion showed a subepithelial tuberculoid granuloma with epithelioid cells, Langhans's giant cells, and very limited caseation. The overlying epithelium showed non-neoplastic hyperplasia. The histological appearances in general resembled that of lupus vulgaris (figure) but Ziehl-Neelsen staining of multiple sections showed no acid-fast organisms. Swabs and smears from the area were also negative, as were cultures of faeces and urine. On further biopsy, however, *M tuberculosis* were recovered directly from fresh tissue by culture and guinea-pig inoculation. Within one month of starting treatment with rifampicin and isoniazid the lesion had begun to regress. By four months only a slight scar-like lesion remained. By eight months the buccal mucosa appeared normal and remained so. Treatment was continued for a total of one year.

During treatment the patient complained of abdominal pain. A barium meal and gastroscopy 11 months after the diagnosis of tuberculosis showed some abnormality of the gastric mucosa. At operation carcinoma of the pyloric antrum was found and subtotal gastrectomy was carried out. Eventually the patient died from multiple metastatic growths.



Section of lesion showing resemblance to lupus vulgaris. (H and E.)

Comment

This case has several notable features. The clinical presentation of the condition was very unusual. Oral tuberculous lesions may vary widely[1] but virtually all have been described as granulomatous or ulcerated. This lesion presented as a simple patch not unlike oral lichen planus or one of the other "white patches" of the oral mucosa. Facial lupus vulgaris may extend to the oral mucosa but, again, granulating, vegetative, or ulcerating lesions are described.[2] The fact that tubercle bacilli could not be cultured from swabs of the surface of the lesion and that acid-fast organisms could not be found on Ziehl-Neelsen staining of the original biopsy material seems not to fulfil the usual criteria for diagnosing tuberculous lesions. But I think that direct culture or inoculation of fresh biopsy material, as in this case, provides the more reliable evidence. Some lesions described as tuberculides because of the absence of acid-fast organisms on staining might have been otherwise diagnosed had attempts been made to recover organisms in this way.[3]

There is no positive evidence that the lesion in this case was the result of a reactivation of latent organisms and that this was potentiated in some way by the early gastric neoplasm. Nevertheless, the calcified foci in the hila suggest a previous tuberculous infection, possibly acquired at the same time as that of the father or daughter. On the other hand, the further infection might have been coincidence, as might the cancer developing at about the same time as the infection.

1 Laws, I M, *British Medical Journal*, 1976, **2**, 45.
2 Wilkinson, D S, in *Textbook of Dermatology*, ed A J Rook, D S Wilkinson, and F J G Ebling, 2nd edn, p 622. Oxford, Blackwell, 1973.
3 Rao, T V, *et al*, *Journal of Oral Surgery*, 1977, **35**, 595.

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Indian childhood cirrhosis presenting in Britain with orcein-positive deposits in liver and kidney

Indian childhood cirrhosis, a rapidly progressive disorder of early childhood, is almost entirely limited to the Asian subcontinent, where it is an important cause of death. This first report of a patient presenting in Britain is of interest as orcein-staining deposits were found in hepatocytes, as we have recently described in Indian childhood cirrhosis.[1] These deposits, which are likely to represent increased copper-binding protein, were also present in renal tubular cells. Functional renal tubular abnormalities occur in Indian childhood cirrhosis[2 3] as they do characteristically in Wilson's disease.

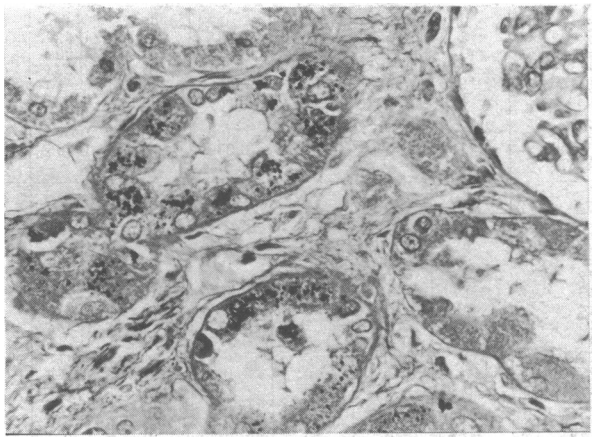
Case history

A 10-month-old boy, an Asian immigrant, was admitted with a one-month history of jaundice. The first child of healthy, unrelated Hindu parents, he had been born in Gujarat, India, and had lived there until 9 months of age. Hepatomegaly had been noticed at 2 months with subsequent intermittent fever and vomiting and, from the age of 6 months, abdominal distension, irritability, poor feeding, and weight loss.

He weighed 6·8 kg and was a thin, wasted child with jaundice, anaemia, hepatosplenomegaly, ascites, and distended abdominal veins. Investigations showed severe hepatic dysfunction, with urinary abnormalities consistent with proximal tubular malfunction. The concentration of total serum bilirubin was 390 μmol/l (22·8 mg/100 ml) (normal <20 μmol/l (1·2 mg/100 ml)), conjugated bilirubin 242 μmol/l (14·2 mg/100 ml), serum aspartate transaminase 107 IU/l (normal 10-14), and serum albumin 22 g/l; prothrombin time was 32 seconds (prolonged) and partial thromboplastin time 102 seconds (control 38). There was appreciable generalised aminoaciduria, with normal serum amino-acids. The urine contained lactose, sucrose, glucose, and fructose, but was free of albumin. Plasma creatinine concentration was normal. Haemoglobin concentration was 5·9 g/dl, reticulocyte count 3·2-4·6%, WBC $18{\cdot}9\times10^9$/l, and platelets 120×10^9/l; serum iron and B_{12} concentrations were normal. The bone marrow showed greatly increased cellularity, and erythropoiesis was very active, dyserythroblastic, and macronormoblastic. Both the patient and his mother had thalassaemia trait on haemoglobin electrophoresis.

He died four days after admission. The macroscopic appearances of the liver were of a micronodular cirrhosis and histological examination showed all the features of Indian childhood cirrhosis—namely, generalised hepato-

cyte degeneration with ballooning, hyaline inclusion bodies in many hepatocytes, extensive intralobular pericellular fibrosis, and neutrophil infiltration. Orcein staining showed coarse dark brown or black inclusions in all hepatocytes. The renal histological findings were normal with haematoxylin and eosin staining, but with orcein staining granules were seen in the cells of about half of the convoluted tubules (figure).



Section of kidney taken post mortem showing extensive orcein-positive deposits within the proximal convoluted tubules (magnification × 400).

Comment

The aetiology of Indian childhood cirrhosis is unknown, although our recent finding, in a series of children from India, of prolific orcein-staining material in the hepatocyte cytoplasm has afforded a new approach. In Wilson's disease and in prolonged cholestasis similar, but less massive, orcein staining is due to a copper-binding protein,[4] raising the possibility that increased copper accumulation may be causally implicated in Indian childhood cirrhosis.

It was therefore of great interest to observe similar orcein-staining material not only in the liver but also in some of the renal tubular cells. As in Wilson's disease, proximal tubular dysfunction occurs in Indian childhood cirrhosis, and was seen in this case. Saini *et al*[2] showed an increased renal clearance of several amino-acids, while Dhatt *et al*[3] recorded the presence of mono- and disaccharides in the urine.

A further feature of Wilson's disease is haemolytic anaemia, attributed to the acute toxic effect of copper. While this infant's anaemia could not be thoroughly investigated, the laboratory data were suggestive of haemolysis. Anaemia is a recognised feature of Indian childhood cirrhosis,[5] but it has not been reported to be haemolytic. The suggestion that copper may be implicated in Indian childhood cirrhosis has important therapeutic implications, while assay of hepatic copper will provide the final proof. We hope that this report may enable further cases to be recognised in immigrant Asian children.

We thank Dr C G D Brook for referring this case and giving permission to report it; Helen Brown for technical help; and Linda Rimmer for editorial help. M S Tanner was supported by a grant from Action Research for the Crippled Child.

[1] Portmann, B, *et al*, *Lancet*, 1978, **1**, 1338.
[2] Saini, A S, *et al*, *Indian Pediatrics*, 1976, **13**, 713.
[3] Dhatt, P S, *et al*, *Indian Journal of Medical Research*, 1968, **59**, 39.
[4] Salaspuro, M, and Sipponen, P, *Gut*, 1976, **17**, 787.
[5] Gupta, S, *et al*, *Indian Pediatrics*, 1975, **12**, 1217.

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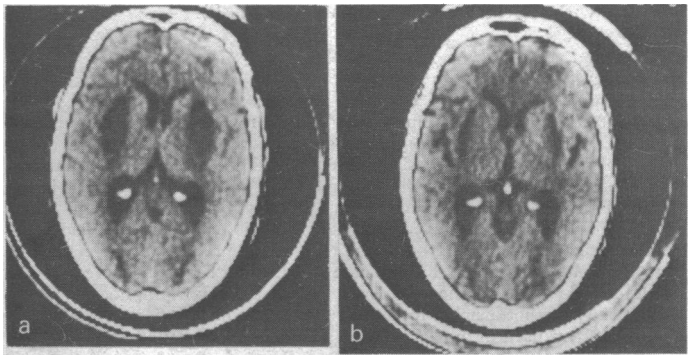
Computerised tomography in severe methanol intoxication

The death rate is high in untreated methanol intoxication, and early diagnosis is of fundamental importance for effective treatment. Especially when confusional states or decreased consciousness dominate the clinical picture, diagnosis may be based entirely on the results of laboratory investigations. We report a patient with methanol intoxication who initially presented diagnostic difficulties and in whom the use of computerised tomography (CAT-scan) was important in making the diagnosis.

Case report

A 62-year-old plumber was admitted during the night to a provincial hospital. According to his wife he had been healthy and had not abused alcohol. The day before he had complained of headache and visual disturbances ("objects appeared yellow") and that night he had developed difficulties in breathing. He was cyanotic and confused. His pupils were constricted and reacted poorly to light. Laboratory examinations disclosed a severe acidosis (pH 6·8, base-excess −34·9 mmol(mEq)/l, P_{CO_2} 3·3 kPa (24·8 mm Hg)). Blood-glucose concentration was normal. Methanol was reported not to be present in the blood. Treatment by artificial respiration and intravenous infusions of sodium bicarbonate was initiated and the acidosis was overcome within a few hours. Pulmonary embolism was suspected as the primary diagnosis and the patient was initially treated with heparin. Pulmonary scintigraphy, *x*-ray films of the lungs, and tests for fibrinogen-degradation products, however, gave negative results.

When extubated one day later the patient reacted to verbal command but was unable to speak and swallow. On examination two days later impaired vision was noted: the pupils did not react to light and he had oedema of both optic discs. A slight facial weakness, as well as extensor plantar response on the right side, was found. A left carotid angiogram was normal, and EEG showed no focal abnormalities. CSF spectrophotometry aroused suspicion of intracranial haemorrhage, and the patient was transferred to Uppsala University Hospital. On admission to the department of neurology (six days after his first symptoms) the patient was responsive to pain but not to verbal command. The pupils did not respond to light. The optic discs were pale and oedematous with slight protrusion and flame-shaped haemorrhages on the left disc, and mild peripapillary oedema was also present. These findings suggested necrosis of the papillary part of the optic nerves. The presence of the other earlier clinical signs was confirmed. On the day of arrival CAT-scan showed areas of decreased attenuation symmetrically localised, especially to the putamens (figure *a*). Frozen plasma samples taken on the day of admission to the provincial hospital, analysed by a selective gas-chromatographic procedure, were found to contain 1 g/l of methanol.



a. First CAT-scan, showing bilateral, symmetrical areas of low attenuation (20 Hounsfield Units) in the putamens. *b*. Second CAT-scan (five weeks later), showing reduction in size of areas of low attenuation.

The patient improved gradually and six weeks later was alert, orientated, and without gross memory defects. He was, however, completely blind and had signs of a moderate polyneuropathy. He now admitted to drinking home-made liquor a few days before he had been taken ill. A second CAT-scan showed diminution in size of the low-density areas described earlier (figure *b*).

Comment

This report illustrates the utmost importance of reliable laboratory data for a specific diagnosis in a patient with decreased consciousness. It was later established that the negative result of the initial determination of methanol in blood was due to a laboratory error.