operation in the present case jaundice was never severe enough to be clinically detectable. The pain was presumably due to transient distension of the biliary system by blood.

The diagnosis of mycotic aneurysm was suggested by the history of bacterial endocarditis, though an interval of 15 months elapsed between adequate treatment of the infection and onset of the abdominal symptoms. In the three previously reported cases of preoperative diagnosis of intrahepatic aneurysm the lesion was attributed to biliary infection (Cohen *et al.*, 1966), idiopathic medial necrosis (Weaver *et al.*, 1968), and bacterial endocarditis (Stack *et al.*, 1968). In two of these cases blood was found in the biliary tree at a previous operation (Cohen *et al.*, 1966; Stack *et al.*, 1968).

It is well recognized that hepatic artery aneurysms may complicate subacute bacterial endocarditis. Of 217 patients with mycotic aneurysms due to blood-borne infection 187 showed evidence of endocardial disease, and the hepatic artery and its branches were involved 19 times (Stengal and Wolferth, 1923). Local or systemic infection has been regarded as the commonest cause of hepatic artery aneurysm (Guida and Moore, 1966), but some authors believe that since the advent of antibiotics arteriosclerosis has become the most important factors (Rutter and Sikkenk, 1971).

Because of the risk of recurrent haemorrhage after ligation of a hepatic artery bearing an aneurysm, Cohen and his colleagues (1966) treated a patient with an intrahepatic aneurysm by lobectomy. In a case reported by Weaver *et al.*

(1968) the patient was similarly treated and it has recently been stated that lobectomy offers the only chance of cure of intrahepatic aneurysm (Rutter and Sikkenk, 1971). In the present case ligation of the right hepatic artery proved a successful treatment, and this experience has been shared by other workers (Stack *et al.*, 1968).

We are very grateful to Dr. H. T. Howat and Mr. H. B. Torrance for permission to report this case. We thank Dr. V. Cope for his help in the interpretation of the angiogram, and the department of medical illustration for the photograph. Mr. H. B. Torrance performed the laparotomy and Dr. N. Greenwood carried out the necropsy.

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Dysphagia, Abdominal Pain, and Sarcoid Granulomata

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Although granulomata have been found in the stomach and intestine of patients with generalized sarcoidosis (Palmer, 1958), involvement of the gastrointestinal tract rarely gives rise to symptoms. Scadding (1967) described two cases in which sarcoid infiltration of the abdominal lymph nodes appeared to be the cause of pain, but in a further review of the literature only one case could be found in which sarcoidosis involving the oesophagus had been recognized during life (Polachek and Matre, 1964). In the following case sarcoid granulomata were found in both the oesophagus and the abdominal lymph nodes, and symptoms were severe.

Case Report

In 1955 a 46-year-old symptomless Caucasian man was found on radiography to have a shadow in the right upper lung field. Sputum and laryngeal swabs were negative for acid-fast bacilli. He began to complain of huskiness of the voice, and in January 1957 a further chest radiograph showed definite enlargement of both hilar regions and a paratracheal shadow. The possibility of sarcoidosis was considered, though a Mantoux test at 10 tuberculin units was positive. Bronchoscopy was clear and a scalene node biopsy showed no evidence of disease. On direct laryngoscopy the vocal cords were thickened, though no histological abnormality was found on biopsy. The hilar and paratracheal shadows resolved spontaneously.

In 1958 he developed progressive difficulty in swallowing, and barium studies three years later showed severe loss of peristalsis. Oesophagoscopy in 1963 showed a swelling in the upper third of the oesophagus, and exploration of this showed a tumour to be present in the muscle wall. Patchy degeneration of muscle with non-cascating giant cell granulomata were found on histological examination (see illustration). No acid-fast bacilli or fungi were found on special staining. A muscle biopsy specimen from the left deltoid, electromyography, and an edrophonium test were all normal.

In 1969 the patient was admitted to hospital as an emergency with pain in the right lower quadrant of the abdomen. A soft mass was felt, and two weeks later appendicectomy was performed. The appendix was histologically normal, as were biopsy specimens of muscle and vessels from the abdominal wall. The pain returned three months later.

Histological section of oesophageal biopsy specimen.

When reinvestigated in January 1971 his general health was maintained despite great difficulty in swallowing, severe abdominal pain, and a husky voice. His haemoglobin level was 14.2 g/100 ml,

leucocyte count 5,600/mm³ (normal differential), and erythrocyte sedimentation rate 12 mm in one hour. Blood urea and electrolyte levels, liver function tests, and serum proteins (with electrophoresis) and immunoglobulins were repeatedly normal, as were his Wassermann reaction, and serum levels and urinary excretion of calcium and phosphate. No abnormal porphyrins, acid fast bacilli, or other bacteria were found at any time in his urine. Intravenous pyelogram, barium-meal examination, gastroscopy, small-bowel barium-meal and barium-enema examinations, sigmoidoscopy, and rectal biopsy gave normal results. Barium-swallow examination showed a grossly dilated oesophagus with minimal peristaltic activity. Radiographs of hands and feet showed no bone cysts, and his chest radiograph showed minimal bilateral apical shadowing but no hilar or paratracheal lymph node enlargement. The Mantoux test was positive at 100 tuberculin units and the Kveim test negative on biopsy on two occasions. Ophthalmic slit-lamp examination and Schirmer test were normal, and no acid-fast bacilli or fungi were found in multiple sputum specimens. Histoplasmin latex agglutination tests were negative.

Full laparotomy, performed because of persistent pain, showed no abnormality apart from enlarged, firm, dark-coloured lymph nodes surrounding the coeliac axis and behind the first part of the duodenum. Several were excised and were shown histologically to be largely replaced by monotonously regular non-caseating granulomata. Special stains showed no acid-fast bacilli or fungi. After the operation the patient's pain was relieved for four months.

Comment

In a clinical study of 160 cases of sarcoidosis Israel and Jones (1958) found no oesophageal involvement. In the only case in which diagnosis of involvement of the oesophagus during life has been reported (Polachek and Matre, 1964) the patient had no dysphagia, though barium-swallow examination showed irregularity of the oesophagus.

In the present case non-caseating granulomata of the sarcoid type were unequivocally present in the oesophagus and abdominal lymph nodes, and the symptoms were severe. Features favouring a diagnosis of sarcoidosis were the pro-

longed course, enlargement of hilar lymph nodes with spontaneous resolution, and failure to find acid-fast bacilli and fungi. Against the diagnosis was the positive Mantoux test, though at the high doses required up to 30% of patients with sarcoidosis give positive reactions (Israel and Jones, 1958). The Kveim test was also negative. Although Siltzbach (1961) showed that this test was positive in 80% of patients with histological evidence of the disease, recently the specificity and validity of Kveim testing has been questioned. Israel and Goldstein (1971) suggested that a positive result in sarcoidosis correlated only with the presence of hilar and mediastinal lymphadenopathy.

Crohn's disease can affect the oesophagus (Dyer *et al.*, 1969), and the histological appearances of the lymph nodes in this disease may be indistinguishable from that in sarcoidosis. The finding that the Kveim test may be positive in up to 50% of cases of Crohn's disease (Mitchell *et al.*, 1970) heightens interest in the relation between the two diseases. However, a diagnosis of Crohn's Disease in this patient was unlikely in view of the absence of any evidence, from tests of absorption, radiology, and careful laparotomy of intestinal disease.

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