Idiopathic granulomatous hepatitis and abdominal pain

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Summary
Granulomatous hepatitis may present with abdominal pain. Four patients are described who had longstanding severe abdominal pain associated with this condition.

Introduction
‘Granulomatous hepatitis’ is not a disease but a pathological reaction that may be elicited by a large number of stimuli. After careful investigation an underlying cause is often found (Neville, Piyasena and James, 1975).

In the last 3 years, 4 women with idiopathic granulomatous hepatitis presented with similar right-sided abdominal pain. During intensive investigation for this pain, liver biopsies were performed; all of which showed multiple non-caseating granulomata.

Patients and results
Patient 1
A 72-year-old presented with a 3-year history of right-sided and central abdominal pain. This was worse at night, after food, and was sometimes associated with vomiting. Intensive investigation, including barium meal and enema, cholecystogram, intravenous pyelogram (IVP) and endoscopic retrograde cholangiopancreatogram (ERCP) did not reveal a cause. Her liver function tests were normal, but ultrasonography twice suggested an hepatic mass. At laparotomy no abnormality was found and the liver was biopsied. Two years later (1979) she remains well but the pain persists, being partially eased by simple analgesia.

Patient 2
A 38-year-old Jamaican was investigated for abdominal pain which had been present for 2 years. The pain was maximal in the right hypochondrium, occasionally causing her to roll in agony or to vomit. She appeared unwell and had lost weight. Her ESR was 62 in the 1st hr (Westergren), alkaline phosphatase 83 KAU. (normal 4–13), serum aspartate transaminase (SGOT) 115 i.u./l (25–65) and γ-glutamyl transeptidase (γ-GT) 86 i.u./l (0–18). A liver scan, barium meal, cholecystogram, IVP and ERCP were normal. Hepatic metastases were suspected and at laparoscopy diffuse white mottling was seen on the liver, which was biopsied; no other abnormality was present. Because of systemic upset she was treated with prednisone, 30 mg/day and improved dramatically. The liver function tests and ESR returned to normal but on maintenance treatment with 7 mg/day mild pain has returned.

Patient 3
A 79-year-old presented with a 6-month history of right hypochondrial pain and weight loss. The pain was aggravated by movement of food and sometimes radiated to her back and right shoulder. Tender hepatomegaly was present but liver function tests were normal apart from a γ-GT of 69 u./l. Gastroscopy and barium enema were normal but an ultrasound examination and Tc-sulphur-colloid scan were suggestive of hepatic metastases. Liver laparoscopy biopsies were taken from the liver which appeared enlarged but otherwise normal. The pain settled spontaneously and she has remained well for 2 years.

Patient 4
A 54-year-old had a past history of cholecystectomy and removal of a chromophobe adenoma. For 2 years she had suffered from severe intermittent right-sided abdominal pain. Her alkaline phosphatase was 22 KAU, SGOT 113 i.u./l and γ-GT 64 u./l. ERCP was normal and there was diminished hepatic uptake on a Tc-sulphur-colloid scan. A percutaneous liver biopsy was performed. She now (1979) gets less frequent pain but is distressed during the attacks.

Investigations
The following investigations were normal in every patient: full blood count, bilirubin, antimitochondrial antibodies, tuberculin skin test, chest X-ray and gastroscopy. The liver biopsies all showed multiple non-caseating epithelioid granulomata (Fig. 1). A Kveim test was not performed on patient 3, and was negative in the others.

Discussion
Multiple epithelioid granulomata are found in 3–10% of liver biopsies (Guckian and Perry, 1966). The aetiology is seldom obvious on histological grounds alone but may eventually be ascertained in...
most patients (Klatskin, 1977). The commonest causes are sarcoidosis, tuberculosis and primary biliary cirrhosis but there are many others. After extensive investigation no cause was found in the 4 patients described in this paper.

These patients all presented with pain, mainly in the right hypochondrium. It was frequently aggravated by food, and vomiting was common. Antacids were ineffective and simple analgesia gave the best relief. There was no associated arthralgia, fever, pruritus or bowel disturbance.

Neville et al. (1975) found an elevated alkaline phosphatase in 64% of their patients with idiopathic granulomatous hepatitis, and an elevated SGOT in 55%. These enzymes were increased in 50% of the patients described by Guckian and Perry (1966). In the present patients, alkaline phosphatase and SGOT were raised in 2 cases (nos. 2 and 4), with an elevated y-GT in 3 (nos 2, 3 and 4).

Many reviews do not mention abdominal pain as a feature of granulomatous hepatitis (Terplan, 1971; Fitzgerald, Fitzgerald and Towers, 1971; Israel and Goldstein, 1973; Klatskin, 1977). However, Neville et al. (1975) comment that 18% of their patients with liver granuloma had abdominal pain, more commonly (50%) in those with no cause found. Similarly Simon and Wolff (1973) mention that 6 out of 13 patients with pyrexia and granulomatous hepatitis had abdominal pain, predominantly affecting the right upper quadrant. Guckian and Perry (1966) in an analysis of cases noted abdominal pain in 33% of patients with granulomatous hepatitis due to sarcoidosis or tuberculosis, but in only 8% with the idiopathic variety—on later investigations of these, Guckian and Perry (1968) often found a cause.

The 4 patients described presented initially with severe abdominal pain. The granulomatous hepatitis was only diagnosed after extensive investigation. Abdominal pain of uncertain aetiology is a common clinical problem and in some patients may be due to unrecognized granulomatous hepatitis.

Acknowledgment

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References


