Case reports


Eosinophilic granuloma of the ileum

A. MYERS
M.B., M.R.C.P.

DAPHNE M. HUMPHREYS
M.B., M.R.C.P.

R. C. N. WILLIAMSON
F.R.C.S.

Department of Medicine, Royal Berkshire Hospital, Reading

Summary
A case of eosinophilic granuloma of the ileum is described in association with a high (50%) eosinophil count. A review of published suggested classifications, aetiology and therapy is made.

Case report
Mr R.B. aged 61 years was admitted as an emergency to the Royal Berkshire Hospital, Reading, in October 1972. He had been in good health until he returned from holiday in Ibiza 2 months before admission. Since then he had complained of daily frontal headaches, lower abdominal pain unrelated to food, and occasional vomiting. There had been no change in bowel habit although the stools had been lighter than usual. He was anorexic and had lost 12 lb in weight. There was no known contact with infective hepatitis and no injections or administration of drugs. There was a past history of bacillary dysentery in Beirut in 1916.

On examination he was ill, thin and sallow, with early clubbing and slight epigastric tenderness. His temperature was 37.5°C, pulse 85/min and blood pressure 100/80 mmHg.

He was extensively investigated. The only abnormal findings were a marked peripheral eosinophilia (7000 eosinophils/mm³) and a weakly positive (1:32) fluorescent amoebic antibody test.

The following investigations were normal or negative. Haemoglobin; ESR; prothrombin time; urea and electrolytes; blood sugar; serum calcium, serum proteins; alkaline phosphatase; LDH, SGOT; bilirubin and urine bile salts; serum immunoglobulins; Paul Bunnell; LE cells; Australia antigen; antibody titres for leptospirosis, brucellosis, salmonellosis, toxoplasmosis, psittacosis and a wide variety of viruses, Mycoplasma pneumoniae and Rickettsia burnetii; urine and stool culture. No ova or cysts were seen in the stools and occult bloods were negative. A liver biopsy was normal except for trapped eosinophils in the liver sinususes. Liver and pancreatic scans and ECG were also normal.

He was treated with consecutive 5-day courses of metoclopramide 10 mg t.d.s. and metronidazole 400 mg t.d.s., and then paromomycin 500,000 u q.d.s., with marked improvement in his symptoms. Following discharge he gained weight, still suffered from occasional lower abdominal pains and was slightly constipated, but continued to improve. The eosinophilia persisted (Fig. 1). All other biochemical tests were normal.

He was readmitted on 26 January to the Surgical Department, Royal Berkshire Hospital, with a 24-hr history of severe constant pain in the right iliac fossa
of gradual onset, not associated with nausea or vomiting. There had been no bowel action. There was 6.35 kg weight loss since the first onset of symptoms.

On examination he was confused and dehydrated but a pyrexial. There was a tender 4 cm palpable mass in the right iliac fossa, associated with marked guarding and rebound tenderness. Rectal examination was normal apart from rather pale stool, and plain X-ray of the abdomen showed gaseous distension of both small and large bowel. A total white cell count of 2700/mm³ was obtained and later shown to contain 56% eosinophils, 20% neutrophils, 18% lymphocytes and 5% monocytes. A diagnosis of appendix abscess was made, but on examination under anaesthetic the mass was found to be mobile. He was rehydrated, and emergency laparotomy performed through a gridiron incision. A large tumour was found arising from mid-ileum, the appendix being attached by its tip, together with a separate loop of terminal ileum. Large fleshy glands extended into the root of the mesentery. The adhesions were freed and the appendix removed, together with 3 ft of ileum bearing the tumour and a deep wedge of mesentery to include the majority of the glands; end-to-end anastomosis was performed. The early postoperative period was complicated by chest infection and bacteraemic shock, but the patient made a complete recovery.

The small bowel specimen (Fig. 2) was 60 cm long; the central portion was obstructed by sloughing of the mucosa and cobblestone hypertrophy. The gut wall at this point was grossly thickened and permeated with rather firm, yellow material. Histological section showed extensive ulceration with marked underlying infiltration with eosinophils, reticulum cells, neutrophils and plasma cells (Fig. 3). Eosinophils were dominant and infiltrated the whole wall including the serosa. The grossly enlarged
mesenteric lymph nodes only showed sinus catarrh. No parasites were found. Two days postoperatively the eosinophil count had fallen to 6% and 4 days later to zero (Fig. 1). A repeat fluorescent amoebic antibody test was positive. The patient regained weight and 5 months postoperatively was well, the peripheral blood showing a 2% eosinophil count.

Discussion
A number of classifications of this condition have been proposed (Ureles, Alscibaja and Lodico, 1952; Higgins, Lamm and Yutzy, 1966; O'Neil, 1970). Ureles classified seventy-nine cases from the world literature into two classes: Class I: diffuse eosinophilic gastroenteritis; Class II: circumscribed eosinophilic infiltrated granuloma. O'Neil divided the cases into only two groups—Group I with peripheral eosinophilia and Group II without peripheral eosinophilia.

The differences between eosinophilic, infiltrated and granulomatous lesions have recently been summarized by Morson and Dawson (1972).

*Eosinophilic enteritis* comprises single or multiple diffuse infiltrating and ulcerated lesions in the small or large bowel and may present with diarrhoea or obstruction. There may be malabsorption, protein-losing enteropathy (Kaplan, 1970) or asthma and, usually, a peripheral eosinophilia. Microscopic examination shows marked eosinophilic infiltration away from the bowel lumen. Response to steroids is generally good and recurrences are uncommon (Salmon, 1967).

*Eosinophilic granulomatous polyp*. The lesion is usually a localized nodule or polyp consisting of vascular granulation tissue, and eosinophilic infiltration is most marked in the submucosa layer. An allergic history is generally absent and there is no associated peripheral eosinophilia.

Waldman *et al.* (1967) described an allergic enteropathy with anaemic, protein-losing enteropathy and eosinophilia with no macroscopic local lesion.

Our case was unlike any previously reported case. Cases reported by Pitchumoni *et al.* (1971) and O'Neil (1970) had no peripheral eosinophilia and none of Ureles’ Class II patients had an eosinophilia exceeding 20%. In all probability the disease exhibits continuous variations from a polyp containing a few eosinophils and no peripheral eosinophilia to a diffuse gastroenteritis with widespread eosinophil infiltration and a peripheral eosinophilia.

Aetiology of the condition is confused, but the consensus of opinion favours an allergic response to an ingested agent. Kaijser (1937) reported that one of his patients was allergic to onions, and another two to neo-arsonamines. Various foods have been incriminated; O'Neil (1970) suggested an allergy to beef, Orr, Miller and Russel (1954) reported a case with allergy to glyceryl trinitrate and chocolate, and Moran and Sherman (1954) suggested that zinc chloride may be a causative factor. Ruzic, Dorsey...
and Huber (1952) thought that eosinophilic granuloma may be related to Loeffler’s syndrome. A number of reports have incriminated parasites; Kuipers et al. (1960) reported the remains of the herring parasite Eustoma rotundatum in the small bowel in two cases. Larvae of the nematode Anisakis were found in four patients with gastric eosinophilic granuloma by Asami, Watanuki and Sakai (1965). Afendoulis (1954) reported five cases with acute allergic gastritis with amoebiasis of the colon, and Stemmerman (1961) found Strongyloides stercoralis in the faeces of six of ten patients with eosinophilic granuloma of the appendix. Skin tests for allergens are seldom convincing, but other allergic manifestations such as asthma, hay fever and eczema have frequently been reported.

Our patient denied any specific food intolerance and there was no atopic history. Initially there was a weakly positive amoebic antibody fixation test and the patient was treated with anti-amoebic therapy. At the time of operation, 2 months after the anti-amoebic therapy, the test was positive but only at a low titre. Three months after operation the test became negative. The significance of these tests is uncertain, but the positivity may have been a cross-reaction to another allergen. It is possible, but unlikely, that the granuloma was related to amoebic infection. A more tenable theory is that the patient became infected with an unknown agent, possibly a parasite, while in Ibiza, although no other evidence of an intestinal parasite was found.

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References


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A. Myers, Daphne M. Humphreys and R. C. N. Williamson

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