CASE REPORT

Eosinophilic cholecystitis with common bile duct stricture: a rare disease

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SUMMARY

Although the most common cause of cholecystitis is gallstones, other conditions may present as acute cholecystitis. We describe a case of eosinophilic cholecystitis with common bile duct stricture. A 36-year-old woman initially had generalised abdominal pain and peripheral eosinophilia. Diagnostic laparoscopy showed eosinophilic ascites and necrotic nodules on the posterior abdominal wall. She was treated with anthelmintics on presumption of toxocara infection based on borderline positivity of serological tests. She later presented with acute cholecystitis and had a cholecystectomy and choledochotomy. Day 9 T-tube cholangiogram showed irregular narrowing of the distal common bile duct. The patient's symptoms were improved with steroids and the T-tube was subsequently removed.

BACKGROUND

Although the most common cause of cholecystitis is gallstones, other conditions may present as acute cholecystitis. We describe a case of eosinophilic cholecystitis (EC) with common bile duct stricture.

CASE PRESENTATION

A 36-year-old woman presented with biliary colic. Her medical history included a termination of pregnancy at the age of 15 years, laparoscopy, dilation and curettage, a massive pulmonary embolus (treated with thrombolytics) as well as recurrent episodes of abdominal pain.

She had originally presented in late 1994, with recurrent episodes of abdominal pain and had been investigated.

INVESTIGATIONS

Blood tests showed leukocytosis with prominent eosinophilia. Investigations included a laparoscopy, which yielded 1.5 L of ascitic fluid showing a marked predominance of eosinophils. Biopsy of a nodule in the posterior abdominal wall was performed, showing fat necrosis. Other investigations showed a borderline positivity for *Toxocara canis* infestation. The patient was treated with mebendazole with the presumption of dog hookworm infestation, then treated with thiabendazole on the presumption of toxocara infection, which seemed to settle her symptoms. She presented to our institution with biliary colic and, after an ultrasound showing gallstones with no duct dilation, was taken to theatre for laparoscopic cholecystectomy.

TREATMENT

Operative findings included 2 L of turbid fluid, oedematous and hyperaemic small bowel and yellow plaques at the mesenteric border of the small bowel, which were biopsied. Operative cholangiogram could not demonstrate flow of contrast into the duodenum so the operation was converted to an open procedure. A supraduodenal choledochotomy was performed and a choledochoscope was introduced. No obstruction could be identified and a 14-French T-tube was inserted.

The patient made a very slow postoperative recovery because of continuing abdominal pain and high output of bile from the T-tube (400 mL/day). Further investigations included abdominal ultrasound and CT scan, which showed a moderate amount of free fluid. A T-tube cholangiogram performed on day 9 showed irregular narrowing of the distal common bile duct with flow of contrast into the duodenum. A gastroscopy was also performed, showing gastritis and duodenitis. Stomach biopsy showed gastritis without marked predominance of eosinophils. Histopathology of the gallbladder showed cholelithiasis, chronic cholecystitis and cholesterosis with large numbers of eosinophils present (figures 1 and 2). Small bowel biopsy showed numerous eosinophils within the serosa and muscularis propria. Blood results postoperatively continued to show marked leukocytosis of up to 20×10^9 (reference range, $4\text{--}11 \times 10^9/\text{L}$) in addition to eosinophilia of up to 10 (reference range, <0.44). After consultation with the medical team, an oral dose of 50 mg of prednisone was started with almost immediate improvement of symptoms. Within 24 hours of treatment, the eosinophil count dropped from 7.26×10^9 to 0.21×10^9 with a reduction in white cell count from 18.2 to 10.4.

OUTCOME AND FOLLOW-UP

The patient was discharged on day 18, with a tapering dose of oral steroids. She was again

Figure 1 Biopsy of the gallbladder.

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Figure 2 Biopsy of the gallbladder (continued).

admitted 5 days later for analgesia due to severe abdominal pain experienced when the T-tube was removed in the outpatient department. When last seen in the outpatient clinic, the patient had improved clinically, and the steroid was ceased. Consideration was being given to restart her oral anticoagulation.

DISCUSSION

EC is a rare form of cholecystitis indistinguishable clinically from common calculous cholecystitis. First described by Albot *et al* in 1949,¹ it is characterised histologically by a dense, transmural leukocyte infiltration composed of more than 90% eosinophils.

Eosinophil infiltration to some extent is not an uncommon finding in the gallbladder. In a study of 217 gallbladders, 48 (22.2%) contained eosinophils to some degree. EC was diagnosed in 14 (6.4%) specimens and another 14 (6.4%) specimens demonstrated substantial numbers of eosinophils but with other cell types in abundance.² In another study of 625 surgically removed gallbladders, the incidence of EC was found to be 0.05%.³

Since being described in 1949, there have been 17 cases reported in the literature, 15 of which were reviewed in a recent case report.⁴ Of these 15 cases of histologically proven EC, the average age was 37 years (range 17–58), with the maximal incidence being in the third and fourth decades. The group consisted of nine females and six males. The aetiology was considered to be idiopathic except two cases where *Ecchinococcus* and paraquat were implicated. Unlike eosinophilic gastroenteritis, the majority of patients (13 of 15) had no history of allergy. Peripheral eosinophilia was present only in four cases. Gallstones were found in 5 of the 15 cases and found exclusively in women. The six cases of EC with obstructive jaundice were found predominantly in men (M/F=5:1), and without associated cholelithiasis, leading some authors to suggest differing disease processes.⁵

In a recent case report and review of the literature, Tenner *et al* suggested that the term 'eosinophilic cholangiopathy' be used to describe eosinophilic disease of the gallbladder and biliary tree.⁶ A 38-year-old woman with EC and common bile duct stricture treated with cholecystectomy and stenting was described. After being treated with steroids, the stent was removed with no residual stricturing. The self-limiting nature of eosinophilic cholangitis and its response to steroids was also described by several other authors.^{7–9} Eosinophilic cholangiopathy may mimic acute calculous cholecystitis or sclerosing cholangitis/cholangiocarcinoma both clinically and on imaging with ultrasound and endoscopic retrograde cholangiopancreatography, although a recent report suggested that MRI may be useful.¹⁰

The aetiology of EC remains obscure. Suggested possibilities include a local eosinophilic inflammatory reaction to gallstones or parasites, a manifestation of eosinophilic gastroenteritis, a systemic hypereosinophilic syndrome (HES), or an allergic hypersensitivity response to immunogens. When a peripheral eosinophilia is present, it has been suggested that EC is likely to be a manifestation of a systemic hypereosinophilic disorder.¹¹

The treatment for EC is cholecystectomy, whereas the treatment for eosinophilic cholangitis is usually conservative.

The idiopathic hypereosinophilic syndrome (IHES) groups together a number of conditions in which eosinophilia occurs for apparently no identifiable cause. First described in 1968, by Hardy and Anderson,¹² Chusid *et al* subsequently established criteria to more adequately define the syndrome. These criteria include a persistent eosinophilia of $1.5 \times 10^9/L$ for longer than 6 months, no apparent aetiology for the eosinophilia and signs of end organ involvement.¹³ The last criterion excludes patients who have eosinophilia that is clinically benign and that possibly remains asymptomatic for decades.

An increase in the blood eosinophil count may occur in a number of disease states including allergies, parasitic infections, vascular diseases and as a reaction to the presence of malignant tumours. It is only once these causes have been excluded that a firm diagnosis of IHES can be made.¹⁴

IHES is more common in men than in women (9:1), and tends to occur between the ages of 20 and 50 years. It usually presents insidiously over months or years, with various symptoms.¹⁵ In a series of 50 patients, 12% of patients with HES had their eosinophilia detected incidentally.¹⁶ Other presenting symptoms included tiredness (26%), cough (24%), breathlessness (16%), muscle pains or angioedema (14%), rash or fever (12%) and retinal lesions (10%).

Patients with IHES do not exhibit a propensity to bacterial or other infections and are not anergic.¹⁷ Weight loss or cachexia is not seen unless there is secondary malnutrition or end-stage congestive cardiac failure. Some patients experience alcohol intolerance with abdominal pain, flushing, nausea, weakness or diarrhoea.¹⁸

Organ systems affected include haematological (eosinophilia, neutrophilia, anaemia, thrombocytopenia, thrombocytosis, splenic infarction causing pain), cardiac (eosinophilic endocardial fibrosis, dilated cardiomyopathy), neurological (thromboembolic events, primary central nervous system dysfunction, peripheral neuropathy), cutaneous, pulmonary (fibrosis, infiltrates) and rheumatological.

Gastrointestinal manifestations may be wide-ranging with 20% of patients having diarrhoea at some time.¹⁹

In a series of 50 patients, Fauci *et al* found organ system involvement to be haematological (100%), cardiovascular (54%), cutaneous (56%), neurological (64%), pulmonary

Learning points

- ▶ Eosinophilic cholecystitis is one of the rare causes of cholecystitis.
- ▶ Eosinophilic cholangiopathy may mimic acute calculous cholecystitis or sclerosing cholangitis and ideally should be diagnosed early as the treatment is conservative.
- ▶ It is advisable for gastrointestinal and general surgeons to be familiar with the symptoms and manifestations of idiopathic hypereosinophilic syndrome in patients with acute abdomen.

(40%), splenic (46%), hepatic (32%), ocular (18%) and gastrointestinal (14%).

In regard to gastrointestinal manifestations, eosinophilic gastroenteritis, colitis, pancreatitis, and sclerosing cholangitis and cholecystitis may occur.^{20–22}

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