

A Rare Case of Eosinophilic Cholecystitis Presenting in a Patient with Thalassemia Intermedia; a Case Report


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ABSTRACT

Eosinophilic cholecystitis is a rare inflammatory condition encountered in surgical cholecystectomy specimens. In terms of histopathology, it is defined by transmural infiltration of eosinophils composing more than 90% of leukocytes. We here report a case of 19-year-old male admitted with thalassemia intermedia and with severe left upper quadrant pain. The patient underwent open splenectomy and cholecystectomy. In his histopathologic examination eosinophilic infiltration was found in gallbladder wall.

Keywords: Thalassemia, Eosinophilic cholecystitis, Inflammation



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Introduction

Eosinophilic cholecystitis (EC) is a rare inflammatory condition of the gallbladder that was first mentioned in a study in 1949 (1). The prevalence of EC was reported from 0.1% to 6.4%. (2-4). Histologically, EC is confirmed when the transmural infiltration of the eosinophils composes more than 90% of leukocytes (2). The etiology of EC is obscure but numerous theories have been proposed including allergic reaction, parasitic infestation, eosinophilic gastroenteritis, hypereosinophilic syndrome (the eosinophils infiltrate tissues such as the brain, heart, lung, liver, and gallbladder) and the local reaction to gallstones (5-7). The diagnosis of eosinophilic cholecystitis is based on histopathologic findings because it has no specific clinical or laboratory feature. The main treatment in cases of typical cholecystitis is cholecystectomy. If there are any associated conditions treatment should also be followed.

Case Report

The patient was a 19-year-old male with a case of thalassemia intermedia who was admitted with severe left upper quadrant pain and was suspected for splenic crisis. At the time of admission, the patient's vital signs were

37.6°C temperature, 78 beats per minute heart rate, 20 breaths per minute respiration rate and 128/75 mm Hg blood pressure.

Past medical history revealed thalassemia intermedia with a history of right upper abdomen pain for the past 6 months and a loss of appetite due to chronic cholecystitis. An allergy to fava beans, known as favism, was also apparent. Medications consisted of folic acid tablets, periodic pneumococcal vaccinations, and at rare times a blood transfusion. The patient had not recently received courses of antibiotics or steroids and he denied any illicit drug or herbal supplement usage. He did not drink alcohol or smoke.

A physical examination was pertinent for his left upper quadrant tenderness and the enlargement of his spleen (Palpable 3 cm below the costal margin). Right upper quadrant tenderness was not detected during the examination and the Murphy sign was also not identified.

The initial laboratory tests are outlined in [Table 1](#), which notes a mildly decreased Hb level, no eosinophilia, and an elevated bilirubin level, which results in a hemolytic anemia diagnosis. An ultrasound & CT scan of

the abdomen demonstrated splenomegaly, increased spleen parenchymal echogenicity, and multiple gallstones.

The patient rejected additional imaging studies, such as Magnetic Resonance Imaging (MRI), due to his claustrophobia.

The patient was prescribed cephalosporin and penicillin during his hospital stay and underwent an open splenectomy due to his splenic crisis and a cholecystectomy due to his chronic cholecystitis. No intra or post-operative complications were encountered. The patient was discharged two days after surgery.

Histopathology findings showed a congested splenic tissue (Figure 1B) and eosinophilic cholecystitis (Figure 1A). On gross examination of specimen, the gallbladder was pear-shaped and measured 12 x7x4 cm and on opening gallbladder the maximal wall thickness was 0.3 cm. The internal surface had a velvety green mucosa which contained multiple stones with the maximum measurement of 1.5 cm. Microscopic views revealed a marked eosinophilic infiltration with few lymphoplasmic cell infiltrations, which confirmed the diagnosis of EC. There were no evidence of epithelial dysplasia, parasitic infestation, or identified malignancy.

Table 1. Laboratory test results

Blood test	Pre-operation Results	Post-operation Results	Normal Reference Interval
WBC	$7.4 \times 10^9 / L$	$24.1 \times 10^9 / L$ Neutrophil 88% Lymphocyte 6% Eosinophil 3% Monocyte 3%	$4-11 \times 10^9 / L$
RBC	$5.4 \times 10^{12} / L$	$5.61 \times 10^{12} / L$	$4 - 6 \times 10^{12} / L$
Hemoglobin	10.1 g / dl	10.4 g / dl	11.5 – 15.5 g/ dl
MCV	68.3 fL / rbc	68.8 fL / rbc	80 – 96 fL / rbc
Platelets	$289 \times 10^9 / L$	$304 \times 10^9 / L$	$150 - 400 \times 10^9 / L$
Total Bilirubin	6.9 mg / dl	-	0.1 – 1.2 mg / dl
Direct Bilirubin	0.4 mg / dl	-	0.3 mg / dl <
ALT	13 IU/L	-	0 – 30 IU / L
AST	20 IU/L	-	10 – 50 IU / L
ALP	198 IU / L	-	30 – 150 IU / L
Amylase	42 IU / L	-	100 IU / L<
Lipase	15 IU / L	-	60 IU / L<
BUN	8 mg / dl	12 mg / dl	12 – 20 mg / dl
Creatinine	0.7 mg / dl	0.7 mg / dl	0.6 – 1.2 mg / dl
Na	138 mg / dl	134 mg / dl	131- 146 mg / dl
K	3.9 mg / dl	3.6 mg / dl	3.6-5.1 mg / dl
BS	83mg / dl	125mg / dl	<200 mg/dl
PT	12.7 sec	12	
INR	1.1	1	
PTT	30 sec	46	25-45 sec

WBC: White Blood Cell, RBC: Red Blood Cell, MCV: Mean Corpuscular Volume, ALT: Alanine transaminase, AST: Aspartate transaminase, ALP: Alkaline Phosphatase, BUN: Blood Urea Nitrogen, PT: Prothrombin Time, PTT: Partial Thromboplastin time, INR: International Normalized Ratio

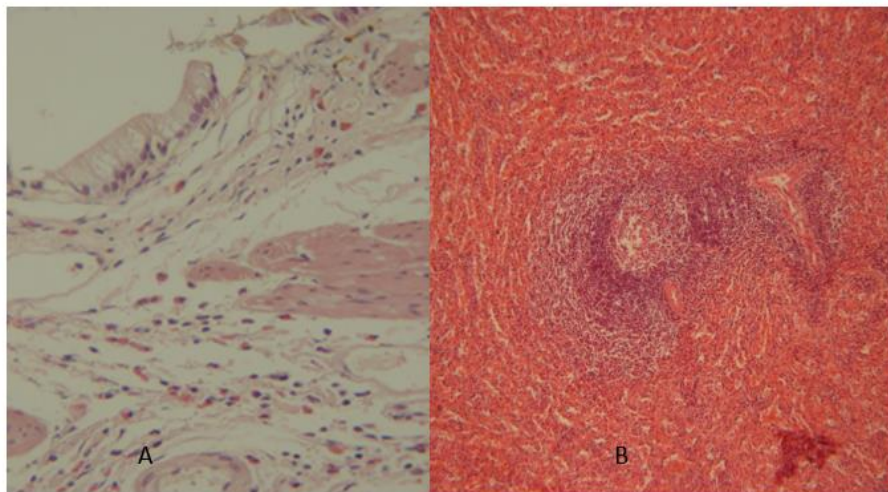


Figure 1. B. Congestive splenic tissue (40x, H&E stain) A. Eosinophilic cholecystitis (40x, H&E stain)

Discussion

EC is an infrequent inflammatory condition of the gallbladder with a prevalence of 0.25% to 6.4% in all patients with cholecystitis (5,8). The mean age of presentation occurs at about 40 years with no detected incidences in a specific gender or race [9,10].

Clinically, EC cannot be distinguished from causes of cholecystitis and its diagnosis is based on classical symptoms of cholecystitis such as right upper quadrant (RUQ) pain and Murphy's sign (patient firmly places his or her hand at the costal margin in the RUQ and with deep inspiration will have extreme pain). Thus, the diagnosis is based on the histopathological findings of a cholecystectomy specimen.

Although no definitive etiology can be drawn for EC, it has been reported as idiopathic or in combination with autoimmune reactions, allergies, hypereosinophilic syndromes (HES), parasites (Hepatic echinococcosis), eosinophilic gastroenteritis, and hypersensitivity to drugs and herbal medicines (5,6,11). In histopathology, when eosinophils account for 90% of inflammatory cells, it will be called eosinophilic cholecystitis but when the infiltrate consists of 50% to 75% eosinophils along with other inflammatory cells in the gallbladder wall, they are classified as Lymphoeosinophilic cholecystitis (12,13).

Calculi are associated in 40% of cases of both eosinophilic and Lymphoeosinophilic varieties (14). EC prevalence in acalculous cholecystitis is three times more than that of cholelithiasis (2). In literature acalculous cholecystitis is reported in obstructive jaundice which is more common in men (12).

In a conducted study of 7,494 patients who underwent cholecystectomies in a 14 year period, the prevalence of

EC was 0.16%, which is 1 case for every 625 cholecystectomies performed. All of them were idiopathic and had concomitant gallstones (15). After evaluating clinical examinations, history and investigations of this case report, the only noteworthy causes of EC were an allergic history, favism, and cephalosporin medications during hospital stay, which could be considered as predisposing etiologies.

The definitive treatment of EC is surgical cholecystectomy; however, medical management such as steroids may be useful in EC associated patients of eosinophilic gastroenteritis or HES, depending on the severity of disease (12,16). In our patient, cholecystectomy led to full recovery. The patient was also periodically monitored at the outpatient department and passed an uneventful postop course.

Conclusion

EC is an infrequent condition with unknown etiology. Gastroenterologists, pathologists, and surgeons need to consider EV because if a causative factor is discovered, follow up and treatment may lead to diagnosis of an important disease. The diagnosis of eosinophilic cholecystitis is based on histopathologic findings because it has no specific clinical or laboratory feature and cholecystectomy is the definitive treatment. As seen in our case, EC was associated with favism and thalassemia intermediate which in accordance to the findings of medical texts, no such case has been previously reported.

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Conflict of Interest

Authors declared no conflict of interests.

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