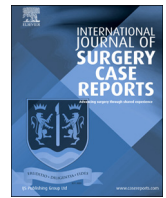


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Lymphoeosinophilic cholecystitis: A rare cause of acalculous cholecystitis in immunocompetent patients – A case report

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ABSTRACT

INTRODUCTION: Eosinophilic and lymphoeosinophilic cholecystitis are uncommonly encountered causes of acalculous cholecystitis characterised by a clinical presentation of acute cholecystitis with eosinophilic infiltration of the gallbladder. Acalculous cholecystitis is a disease that is traditionally associated with patients who are critically unwell and immunosuppressed.

PRESENTATION OF CASE: A fit and well 37-year-old man presented to the emergency department with a 12-h history of constant upper abdominal pain radiating through to his back. Abdominal examination revealed tenderness in the right upper quadrant with a positive Murphy's sign. An abdominal ultrasound was performed, revealing a thickened gallbladder wall with probe tenderness, but no gallstones. He proceeded to an uneventful emergency laparoscopic cholecystectomy. Histological examination of the gallbladder revealed mucosal and transmural inflammation comprising of lymphocytes and more than 50% eosinophils. No gallstones were found. A diagnosis of lymphoeosinophilic cholecystitis was made. The patient had improvement in his symptoms and was discharged home. He was well at follow-up.

DISCUSSION: There is a small subset of immunocompetent patients who are not critically unwell who present with acalculous cholecystitis. There is significant hesitancy in offering a cholecystectomy to these patients without radiological evidence of gallstones or sludge preoperatively. Cholecystectomy should be offered to these patients if the clinical picture fits acute cholecystitis.

CONCLUSION: Eosinophilic and lymphoeosinophilic cholecystitis are important causes of acalculous cholecystitis that can occur in immunocompetent patients. The decision to offer the patient a cholecystectomy should be based on clinical presentation and examination, rather than the absence or presence of gallstones.

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1. Introduction

Eosinophilic cholecystitis is a rare condition that was originally described in 1949 [1]. Lymphoeosinophilic cholecystitis exists on a spectrum with eosinophilic cholecystitis rather than existing as a separate clinical entity. They are histopathological diagnoses with the same clinical presentation. Lymphoeosinophilic cholecystitis is diagnosed histologically when the inflammatory cell population is comprised of 50–89% eosinophils, while eosinophilic cholecystitis is characterised by more than 90% eosinophils [2]. Acalculous cholecystitis is traditionally thought of as a disease process occurring in patients who are critically unwell and immunocompromised [3]. There is emerging evidence to suggest that acalculous cholecystitis also occurs in young, middle-aged healthy individuals [4]. Lymphoeosinophilic cholecystitis is an important cause of acal-

culous cholecystitis to consider, as it can occur in clinically well patients.

We present a rare case of lymphoeosinophilic cholecystitis occurring in an otherwise fit and well patient to emphasise the importance of clinical judgement in offering cholecystectomy, rather than presence or absence of gallstones. This case report has been prepared and reported in line with the SCARE guidelines [5].

2. Case report

A fit and well 37-year-old man presented to the emergency department with a 12-h history of constant upper abdominal pain radiating through to his back. He had associated nausea, anorexia, and had felt feverish with chills. His past medical history was only significant for a previous open appendicectomy; there was no history of asthma. He was not taking any medications and denied any allergies. The patient was haemodynamically stable and afebrile on initial review. Abdominal examination revealed tenderness in the right upper quadrant with a positive Murphy's sign.

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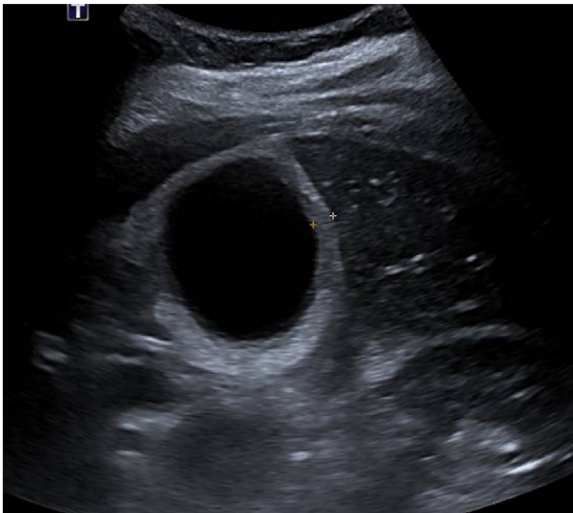


Fig. 1. Transverse axis ultrasound view of the gallbladder demonstrating a thickened gallbladder wall measuring 5 mm without sludge or gallstones.

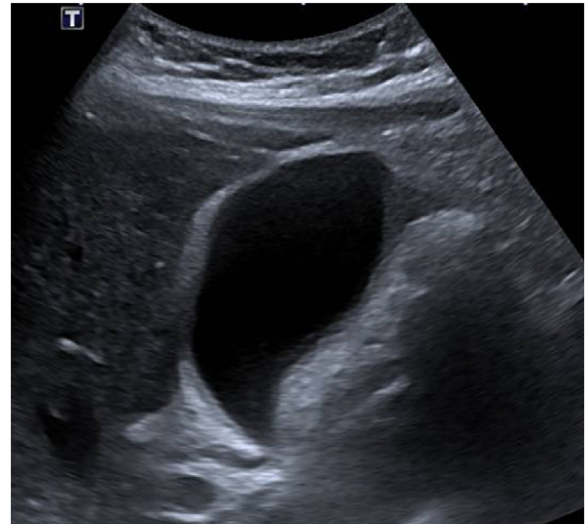


Fig. 2. Long axis ultrasound view of the gallbladder demonstrating a thickened gallbladder wall measuring 5 mm without sludge or gallstones.

Initial laboratory investigations revealed a raised white cell count of $19.91 \times 10^9/L$ and a C-reactive protein of 1.7 mg/L. His serum absolute eosinophil count was $0.00 \times 10^9/L$. He had normal liver function tests and a normal lipase level. An abdominal ultrasound was performed, which revealed a thickened gallbladder wall at 5 mm with associated probe tenderness; however, there was no sonographic evidence of gallstones or sludge (Figs. 1 and 2).

Due to the absence of stones and sludge, there was initial hesitancy about the need for cholecystectomy. As the patient had ongoing pain consistent with cholecystitis over the next 12 h, he proceeded to an uneventful emergency laparoscopic cholecystectomy. Intraoperative cholangiogram was performed and was unremarkable. Findings at the time of laparoscopy were a distended and oedematous gallbladder, with serous fluid but no pus. Histological examination of the gallbladder revealed mucosal and transmural inflammation comprising of lymphocytes and more

than 50 % eosinophils (Figs. 3 and 4). A reactive lymph node was identified in the neck of the gallbladder. No gallstones were found. A diagnosis of lymphoeosinophilic cholecystitis was made. The patient had improvement in his symptoms and was discharged home. He was well at time of follow up. Throughout his admission, serial blood tests never revealed a serum eosinophilia.

3. Discussion

Eosinophilic and lymphoeosinophilic cholecystitis are rare entities. Early retrospective studies of cholecystectomy specimens report a prevalence of 6.4 % for both eosinophilic and lymphoeosinophilic cholecystitis [2]. Subsequent studies in some populations have found the prevalence of eosinophilic cholecystitis to be as low as 0.16 % [6]. The aetiology and pathophysiology of

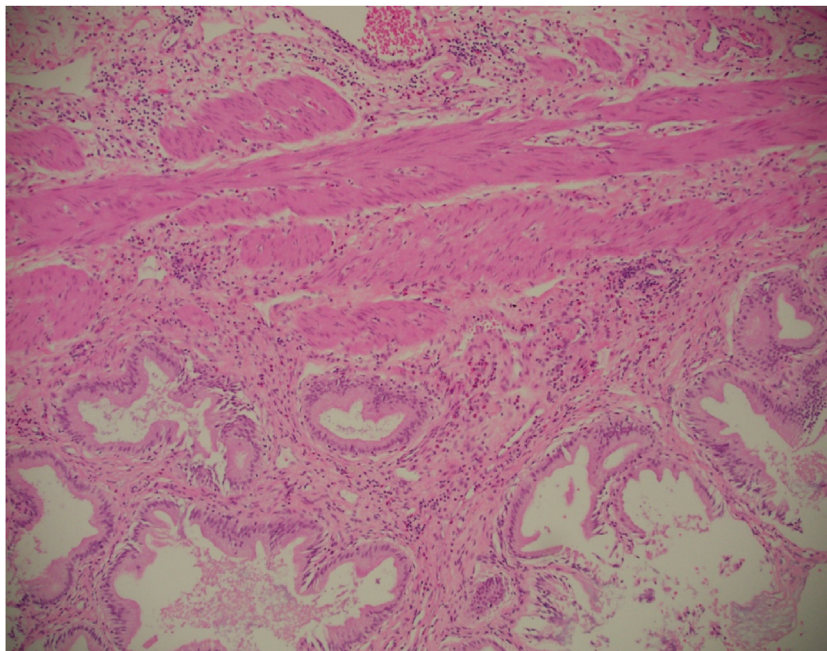


Fig. 3. H&E stain at 10× magnification demonstrating transmural inflammation and fibromuscular thickening with lymphoeosinophilic infiltration comprising >50 % eosinophils.

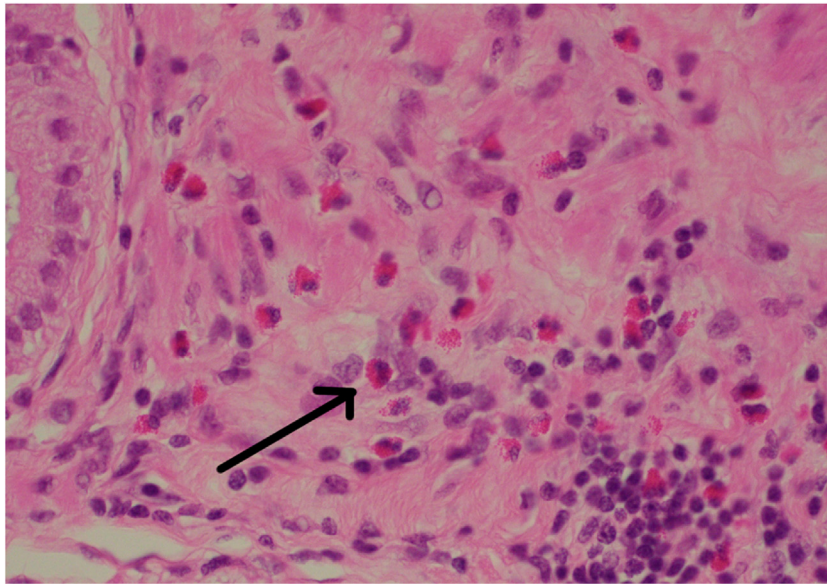


Fig. 4. H&E stain at 40× magnification with the black arrow demonstrating an eosinophil in amongst lymphocytes.

these conditions are poorly understood. Previous case reports and studies have suggested associations with various conditions including the following: parasitic infections (e.g. *Ascaris spp*, *Clonorchis spp*, *Echinococcus spp*) [7–9], gastroenteritis [10], eosinophilic airway inflammation [11], allergies [12], hypereosinophilic syndromes [13], cephalosporins [14], and use of herbal medicines (e.g. 1-tryptophan) [15]. Many times, however, no obvious cause is identifiable. It has been hypothesised by some authors that idiopathic eosinophilic cholecystitis may be driven by a local reaction to bile or gallstones [16]. Eosinophilic cholecystitis can be calculous or acalculous [2,17]. It may also be associated with eosinophilic cholangiopathy, sometimes involving bile duct strictures [18].

Clinically, presentations of lympho-eosinophilic and eosinophilic cholecystitis are often indistinguishable from other forms of acute cholecystitis. Eosinophilia in peripheral blood is usually not present, as in our case; it may be found in 10–20% of patients with eosinophilic cholecystitis [19]. Whilst it is an entity that may be suspected, the diagnosis remains histological. The definitive treatment of eosinophilic and lympho-eosinophilic cholecystitis is cholecystectomy, given the nature of its presentation. Once a diagnosis has been achieved, steroids as a postoperative adjunctive treatment may be indicated in patients who have a concurrent allergy syndrome [20].

4. Conclusion

Eosinophilic and lympho-eosinophilic cholecystitis are important causes of acalculous cholecystitis that can occur in immunocompetent patients, without a history of allergy or parasitic infection. Cholecystectomy should be offered to patients based on clinical symptoms and examination that fit acute cholecystitis, rather than the absence or presence of gallstones.

Declaration of Competing Interest

The authors have no competing interests to declare.

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No funding was obtained for this case report.

Ethical approval

No ethics approval was required.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution

Dr Steven Tran: Writing – original draft and writing of the paper.
 Dr Izhar-ul Haque: Writing – reviewing and editing of the paper.
 Dr Deepak Dhatrak: Interpretation and procurement of histopathology.
 Dr Paul Dolan: Writing – reviewing and editing of the paper.

Registration of research studies

Not applicable.

Guarantor

Dr Steven Tran.

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