

Case Report

Eosinophilic Cholecystitis Associated with Papillary Hyperplasia of Gall Bladder

Sant Prakash Kataria , Ashima Batra, Gajender Singh,
Ekta Boombak, Sanjay Kumar, Rajeev Sen

Dept. of Pathology, Pt B.D. Sharma University of Health Sciences, Rohtak, Haryana, India

ABSTRACT

Eosinophilic cholecystitis (EC) is a rare entity that presents in a manner comparable to acute cholecystitis. The diagnosis is based on classical symptoms of cholecystitis with the presence of eosinophils (>90%) within the gallbladder. EC has been reported alone (idiopathic EC) or in combination with manifestations such as eosinophilic cholangitis, hypereosinophilic syndromes, and parasitic infestations. Papillary hyperplasia of gallbladder occurs in the setting of cholelithiasis, inflammatory lesion of gallbladder, primary sclerosing cholangitis or ulcerative colitis. To the best of our knowledge of the literature reviewed, papillary hyperplasia has never been reported in a setting of eosinophilic cholecystitis. We report a case of 30 years old female presenting with idiopathic eosinophilic cholecystitis associated with papillary hyperplasia of gallbladder in the year 2011 at PGIMS Rohtak (India). Hereby we report coexistence of these two entities never described together.

Keywords: Papillary Hyperplasia, Eosinophilic Cholecystitis, India

Introduction

Eosinophilic cholecystitis is a rare entity, with an incidence of 0.5–6.4% in cholecystectomy specimens (1). Although more than 90% of transmural infiltrating inflammatory cells are eosinophils, the etiology of eosinophilic cholecystitis is not clearly understood and might be associated with hypersensitivity to antibiotics, other drugs, herbal medicines, hepat-

ic echinococcosis, or as a variant manifestation of eosinophilic gastroenteritis (1). Papillary hyperplasia of gallbladder can be primary or secondary occurring in the setting of cholelithiasis, cholecystitis and cholesterosis (2).

Case Report

A 30 year female presented with pain in right upper quadrant along with vomiting for last 2 days

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Address Communications to: Dr. Sant Prakash Kataria, Department of Pathology, Pt B.D. Sharma University of Health Sciences, Rohtak, Haryana, India.

Email: drashimabatra@gmail.com

in the year 2011 in out patient (OPD) of PGIMS Rohtak (India). This Case report is based on the clinical notes of an individual patient with the informed consent and does not require research ethics review. She did not have specific past medical history including allergic reactions or any illicit drug use. The patient had a temperature of 38°C, a heart rate of 114 beats/minute, a respiration rate of 20 breaths/minute, and a blood pressure of 136/82 mm of Hg. Physical examination demonstrated a severe right hypochondrial tenderness and positive Murphy's sign. She had a white blood cell count of 10,500/L (80% neutrophils, 15% lymphocytes, 3% monocytes and 2% eosinophils), a hemoglobin level of 10.0 g/dL, and

a platelet count of 2.5 lakh /L. Biochemical investigations revealed normal levels of bilirubin, alanine transaminase, aspartate transaminase and alkaline phosphatase. Ultrasonography of the gallbladder showed thickening of the gallbladder wall with no evidence of calculi.

The patient underwent an open cholecystectomy. A gallbladder with a thickened wall was removed. There was no evidence of stones or parasitic infestation. Gallbladder was pear shaped measuring 9×6.5×6 cm with marked wall thickening measuring 1-1.3cm. The serosa was smooth and the mucosa was studded with multiple grey white papillary projections. (Fig. 1)



Fig.1: Surgical specimen of resected gallbladder with mucosa revealing multiple grey white papillary projections.

Microscopic examination revealed mucosal hyperplasia with mucosal folds lying close to one another and taller than normal (Fig 2a). They were lined by normal-appearing columnar cells and pencil-like cells (Fig. 2b). The papillary projections of the common bile duct were lined only by columnar cells with prominent subnuclear vacuoles. Submucosa revealed infiltration by chronic inflammatory cells comprising >90% of eosinophils. Muscularis and serosal layer were

unremarkable. (Fig. 2c & d) A histopathological diagnosis of eosinophilic cholecystitis with papillary hyperplasia was rendered.

Her symptoms resolved completely after cholecystectomy. The postoperative course was uneventful, and the patient was discharged and periodically monitored in the outpatient department.

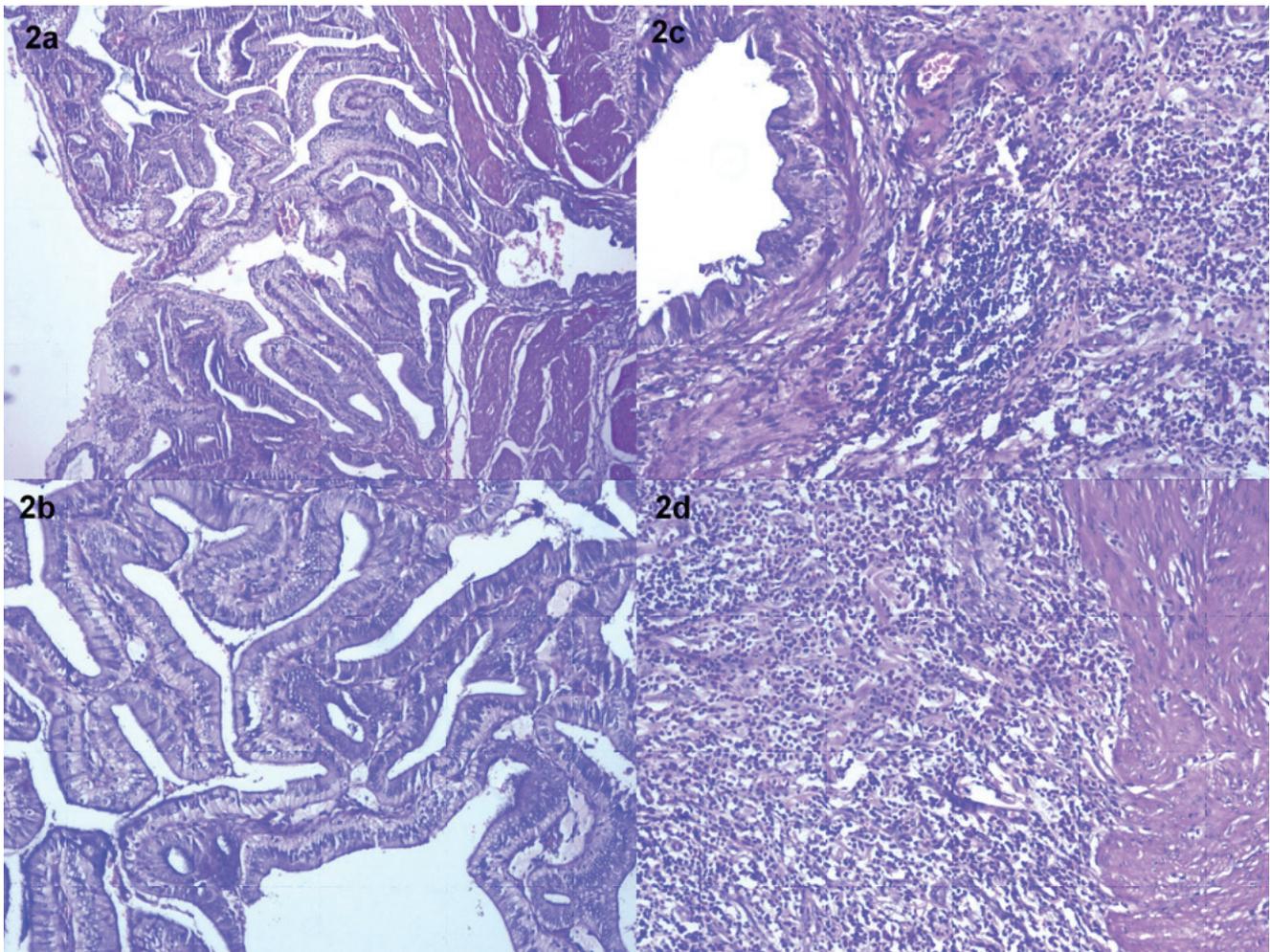


Fig.2: Histological examination revealed tall and closely placed mucosal folds (2a, H&E; $\times 40$) lined by columnar epithelium (2b, H&E; $\times 100$). The submucosa revealed infiltration by chronic inflammatory cells comprising $>90\%$ of eosinophils (2c & 2d, H&E; $\times 100$).

Discussion

Eosinophilic cholecystitis is an uncommon condition that was first described by Albot in 1949 (3). It has a clinical presentation similar to that of typical cholecystitis, with right upper quadrant pain and an elicited Murphy's sign (4). Although the etiology is still obscure, the postulated causes include allergies, parasites, hypereosinophilic syndrome, eosinophilic gastroenteritis, and local reaction to gallstones. It sometimes shows a marked peripheral blood eosinophilia. Eosinophilic infiltration can be associated with disorders of lung, heart, GIT, biliary tract and gallbladder. Peripheral eosinophilia indicates that it is likely to be a manifestation of a systemic hypereosinophilic disorder. The patient described

herein gave no indications of such previously suggested causes of EC. In the absence of any apparent precipitating etiology, the case described herein is considered one of idiopathic EC. In cases without peripheral eosinophilia, a local reaction to bile or gallstones may be responsible (5). Histologically, it is distinguished by a dense transmural infiltration of eosinophils comprising 90% or more of the leukocytes (4).

Gallbladder hyperplasia can be divided into primary and secondary hyperplasia on the basis current hypothesis (6). The term primary epithelial hyperplasia of the gallbladder was proposed by Elfving in 1960, restricted to cases not associated with cholelithiasis, inflammatory lesions of the gallbladder, primary sclerosing cholangitis or ulcerative colitis (7).

Though the exact cause of papillary hyperplasia cannot be ascertained but it is suggested that it could be induced by the increase of bile pressure in the extrahepatic bile ducts leading to morphological adaptation of the mucosa to permit a greater absorption of cholesterol from gallbladder lumen to the blood capillaries (8). The papillary hyperplasia in our case can be attributed to eosinophilic cholecystitis.

Conclusion

Idiopathic EC is significant because it is not apparent solely through laboratory tests. It is important to rule out causes associated with peripheral eosinophilia before the diagnosis is made with certainty. Papillary hyperplasia is a benign condition which has never been reported in a case of eosinophilic cholecystitis. This rare coexistence of the two entities in our case prompted us to bring it your knowledge.

Acknowledgements

The authors declare that there is no conflict of interests.

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