

The Masquerading Biliary Cystadenoma

Daniel Stenberg, DO¹, Mohammed Alkhero, MD¹, Mazen M. Jamal, MD², and Michael Tran, MD³

¹UHS Southern California Medical Education Consortium, Temecula, CA

²Oceana Gastroenterology Associates, Corona, CA

³Mission Surgical Clinic, Corona, CA

CASE REPORT

Cystic diseases of the liver occur in about 5%–10% of the population.¹ Biliary cystic tumors (BCTs), such as biliary cystadenoma (BCA) and cystadenocarcinoma, comprise less than 5% of all liver cysts.² BCA occurs predominantly in women (90%). Hueter first reported BCA in 1887, and Keen reported the first BCA resection 5 years later.^{3,4} Because of the rarity of BCT, many clinicians may be unfamiliar with the diagnostic features, therapeutic management, and natural history of these liver neoplasms. Although 10% of reported BCTs originate in the extrahepatic biliary tree, the overwhelming majority arise from the intrahepatic biliary system. In our clinical image report, we demonstrate a distant and rare site for BCA in the gastric fundus, away from the biliary tree.

We present an 80-year-old woman with a history of atrial fibrillation on eliquis and hypertension, who came to the clinic after a referral from her primary care physician for abdominal pain with nausea. The patient had no changes in bowel habits—no hematochezia or melena. She reported mild heartburn with spicy foods, although she did not take any antacid medications. She denied any nonsteroidal antiinflammatory drugs use. There was no history of stomach or colon cancer.

A noncontrasted abdominal computed tomography demonstrated a stable, complex cystic mass measuring 4.9×4.8 cm arising from the greater curvature of the gastric fundus and stable, small liver cysts (Figure 1). Subsequent endoscopy was unremarkable. Endoscopic ultrasound showed a large, simple cyst by gastric fundus measuring 50×44 mm without nodules. A fine needle aspiration was not attempted because of the risk of peritonitis (Figure 2). The patient was referred to a general surgeon for ex-lap and cystectomy. Surgery had planned a wedge resection at the outset of the operation; however, intraoperative exploration revealed the mass to have a thin fibrous connection to the liver. The pathology report confirmed BCA, a single layer of cuboidal or conciliated columnar epithelium resting on a basement membrane (Figures 3 and 4). The epithelium forms multiple papillary projections.⁵ We conclude that BCA can be listed in the differential diagnosis for any cystic mass involving the biliary tree, whether intrahepatic or extrahepatic. We would also like to emphasize that, historically, BCA was treated by marsupialization, internal Roux-en-Y drainage, aspiration, sclerosis, or partial resection. However, those modalities are associated with high complication rates, including sepsis,



Figure 1. (A) Coronal and (B) transverse view of abdominal computed tomography showing a stable complex cystic mass 4.9×4.8 cm arising from the greater curvature of the fundus of the stomach. It is also showing a thin fibrous connection between the liver and the mass crossing the midline around the stomach.

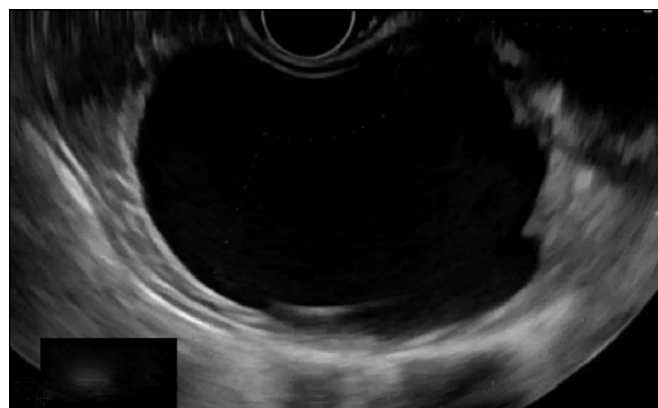


Figure 2. Endoscopic ultrasound image showing a large cyst by gastric fundus measuring 50×44 mm, simple, no nodules.



Figure 3. Gross surgical specimen for the biliary cystadenoma 5 × 5 cm.

continued growth, and progression to malignancy.⁵ Complete surgical excision or enucleation provides the best therapeutic approach with a good long-term prognosis.

DISCLOSURES

Author contribution: D. Stenberg wrote the article, provided the images, revised the article for intellectual content, approved the final article, and is the article guarantor. M. Alkheri wrote the article, revised the article for intellectual content, and approved the final article. MM Jamal and M. Tran edited the article, revised the article for intellectual content, and approved the final article.

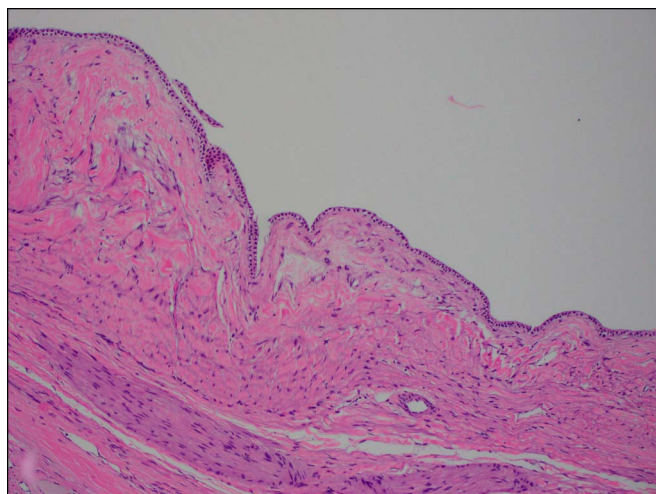


Figure 4. Gross histology showing cuboidal cell lining of the cystadenoma.

Financial disclosure: None to report.

Informed consent was obtained for this case report.

Received September 22, 2020; Accepted February 22, 2021

REFERENCES

1. Caremani M, Vincenti A, Benci A, et al. Ecographic epidemiology of non-parasitic hepatic cysts. *J Clin Ultrasound*. 1993;21(2):115–8.
2. Walt AJ. Cysts and benign tumors of the liver. *Surg Clin North Am*. 1977; 57(2):449–64.
3. Short WF, Nedwich A, Levy HA, Howard JM. Biliary cystadenoma. Report of a case and review of the literature. *Arch Surg*. 1971;102(1):78–80.
4. Lewis WD, Jenkins RL, Rossi RL, et al. Surgical treatment of biliary cystadenoma. A report of 15 cases. *Arch Surg*. 1988;123(5):563–8.
5. Pillai SA, Velayutham V, Perumal S, et al. Biliary cystadenomas: A case for complete resection. *HPB Surg*. 2012;2012:6.

Copyright: © 2021 The Author(s). Published by Wolters Kluwer Health, Inc. on behalf of The American College of Gastroenterology. This is an open access article distributed under the terms of the Creative Commons Attribution-Non Commercial-No Derivatives License 4.0 (CCBY-NC-ND), where it is permissible to download and share the work provided it is properly cited. The work cannot be changed in any way or used commercially without permission from the journal.