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CASE REPORT | SMALL BOWEL

Cholecystoenteric Fistula Masquerading as a Bleeding Subepithelial Mass

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ABSTRACT

An 82-year-old man was referred for endoscopic ultrasound of an ulcerated subepithelial mass in the duodenal sweep. The mass was initially identified during upper endoscopy for coffee-ground emesis. During endoscopic ultrasound, a 21-mm hypoechoic ulcerated subepithelial mass with a duct-like structure was identified. During suction to appose the lesion against the tip of the echoendoscope, the ulceration opened into a fistulous tract with drainage of bile and stones. Subsequent abdominal imaging demonstrated that the mass-like duodenal lesion abutted the gallbladder, which had an air-fluid level. We report a cholecystoenteric fistula masquerading as a subepithelial duodenal mass.

INTRODUCTION

Cholecystoenteric fistulae are a rare sequela of cholelithiasis and gallbladder malignancy. These are seen predominantly in the setting of cholelithiasis but may also occur as a complication of operative hepatobiliary interventions. Large stones, recurrent cholangitis, female sex, and old age are risk factors for bilioenteric fistulae. The clinical presentation is variable, and a preoperative diagnosis may be achieved only in 8–17% of cases.[1]

CASE REPORT

An 82-year-old man with multiple comorbidities was initially evaluated for coffee-ground emesis associated with a 3-g/dL drop in hemoglobin. Upper endoscopy demonstrated a subepithelial mass with an overlying ulceration and an adherent clot in the duodenal bulb, with no histological evidence of a malignancy (Figure 1). Computed tomography (CT) obtained during the initial hospitalization demonstrated air within an inflamed gallbladder that contained stones and sludge, as well as a rounded, well-circumscribed mass in the duodenum (Figure 2). Liver enzymes at the time were notable for aspartate aminotransferase 182 IU/L, alanine aminotransferase 167 IU/L, alkaline phosphatase 176 IU/L, and total bilirubin 1.4 mg/dL. The lesion, which was the source of the bleeding, was presumed to be a gastrointestinal (GI) stromal tumor, and he was referred for an endoscopic ultrasound (EUS).

EUS revealed a 21-mm hypoechoic ulcerated subepithelial mass with a duct-like structure in the duodenal bulb. During suction to appose the lesion against the tip of the echoendoscope, the mass decompressed, and the areas of ulceration morphed into 2 small orifices draining bile and small stones (Video 1; Figure 3). Subsequent magnetic resonance imaging demonstrated resolution of the mass-like density in the duodenum (Figure 4). A small bile duct stone and subtle contrast opacification of the cholecystoduodenal fistula was also identified (Figure 4). The patient had multiple comorbid conditions including metastatic prostate cancer, chronic liver disease due to hepatitis C infection, congestive heart failure, ischemic stroke, and atrial fibrillation. He did not have a known history of biliary disorders. Therefore, the patient was managed conservatively because the bleeding had ceased and biliary

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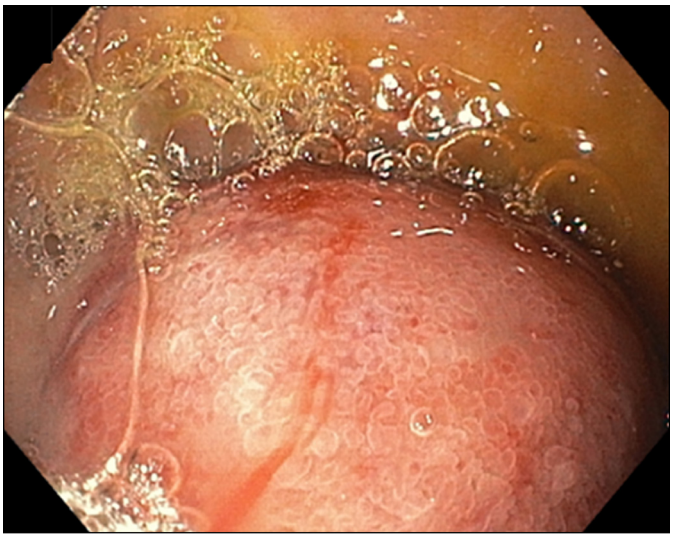

Figure 1. Endoscopy demonstrating a subepithelial mass with overlying ulceration and an adherent clot in the duodenal bulb.

drainage had been established through the fistula. The patient remained clinically stable without any bleeding, abdominal pain, or jaundice on follow-up. Subsequent endoscopy to assess healing demonstrated the persistence of the fistula orifice and clear decompression of the mass lesion (Figure 5).

DISCUSSION

Cholecystoenteric fistulae are a rare and late complication of gallstone disease, with a reported incidence of 0.005–0.9%.[2-5] Clinical presentation of such fistulae is variable, occurring predominantly in women around the age of 60 years, and

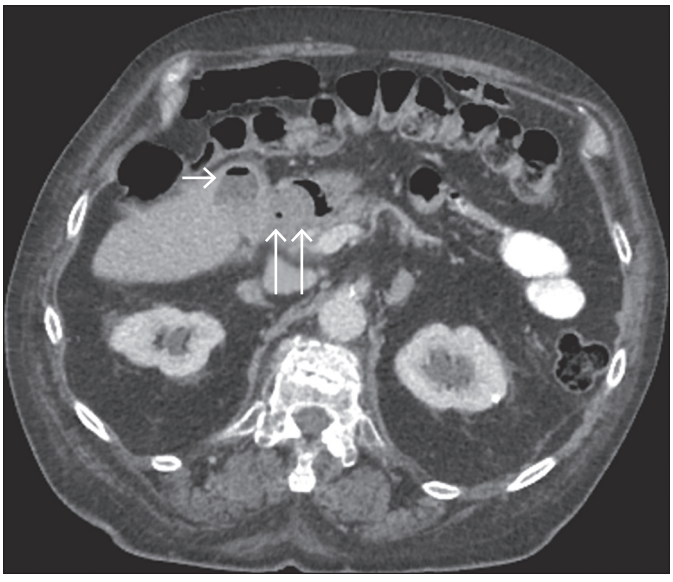

Figure 2. Computed tomography of the subepithelial duodenal mass (vertical arrows) showing air in the gallbladder (horizontal arrow).

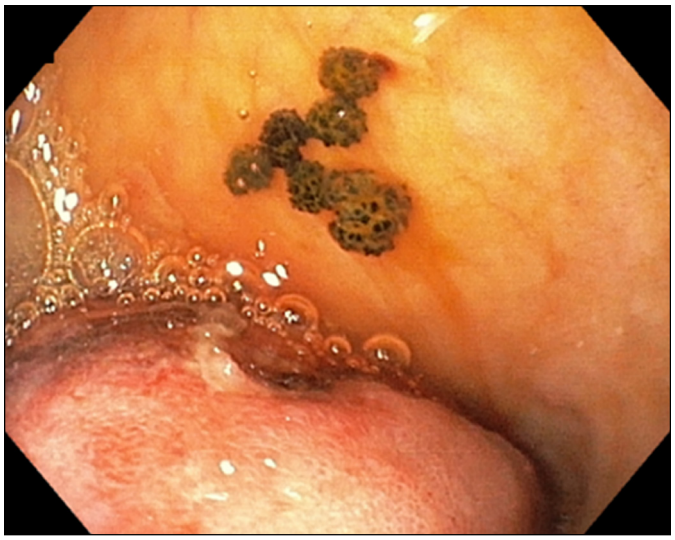

Figure 3. Endoscopy demonstrating a mass-like lesion with a small orifice draining bile and small stones.

Video 1. Decompression of the subepithelial mass and morphing of ulcerated areas into 2 small orifices draining bile and small stones. Watch the video: http://s3.gi.org/media/links/KohliVideo.mp4.

they are often diagnosed intraoperatively during interventions such as cholecystectomy.[4,6-9] The putative mechanism involves impaction of a gallstone and its subsequent erosion through the gallbladder and duodenal wall. The most common communication is between the gallbladder and the duodenum, although cholecystocolonic and cholecystogastric fistulae have been reported.[7] While gallstones are the most common cause of spontaneous bilioenteric fistula, peptic ulcers and malignancies can also lead to similar fistulization.[8] The presence of such fistulae can be associated with gallstone ileus, Bouveret syndrome, Mirizzi syndrome, and intraoperative adverse events.[6,9] There are isolated case reports of presentation with upper GI bleeding, often from a marginal ulcer located at the site of the fistula.[10,11]

Because these fistulae are often diagnosed intraoperatively, laparoscopic management of cholecystoenteric fistula is

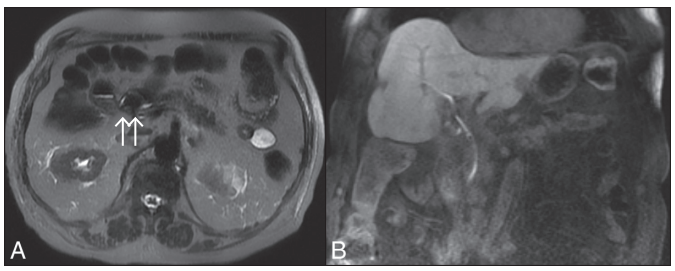

Figure 4. (A) Magnetic resonance imaging demonstrating the resolution of the mass-like density in the duodenum (arrows) and (B) the presence of choledocholithiasis along with the cholecystoenteric fistula.

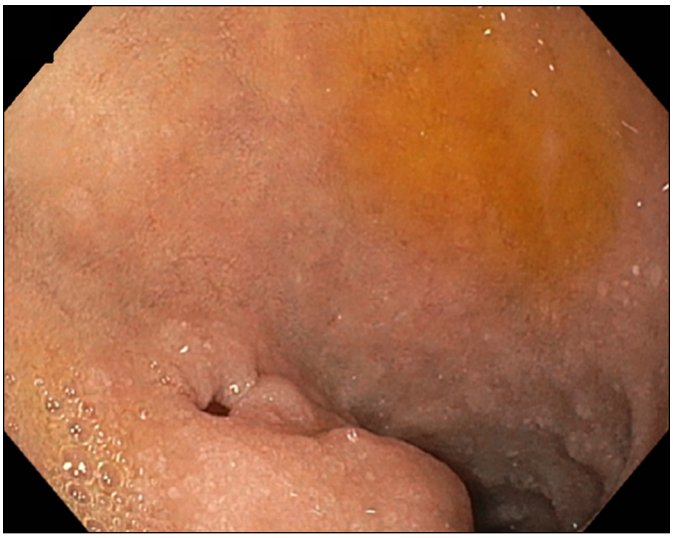

Figure 5. Follow-up endoscopy demonstrating decompression of the lesion with persistence of the fistula orifice.

feasible in well-equipped, high-volume centers.[12] Cholecystoenteric fistulae resulting from malignant masses have been reported in the scientific literature, but we did not find reports of cholecystoenteric fistulae presenting as a benign duodenal mass.[13]

Cholecystoenteric fistulae are relatively rare and may cause GI bleeding. A high index of suspicion is necessary, and an early diagnosis is crucial because these fistulae can have myriad clinical presentations and are associated with intraoperative adverse events. To our knowledge, this is the first description of a cholecystoduodenal fistula masquerading as a large subepithelial mass.

DISCLOSURES

Author contributions: DR Kohli searched the literature, wrote the manuscript, and is the article guarantor. M. Anis interpreted the radiological images. T. Shah edited the manuscript.

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Informed consent was obtained for this case report.

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