Pneumobilia Resulting From Choledochoduodenal Fistula Secondary to Metastatic Colon Adenocarcinoma

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Abstract

Pneumobilia, or air within the biliary tree, is a poor prognostic indicator in a patient without prior biliary sphincterotomy. Differential diagnosis includes infection with gas-forming organisms, choledochoenteric fistula in the setting of gallstones or penetrating ulcer disease, malignant invasion from a primary liver or biliary tract tumor, or metastatic disease. Treatment depends on etiology and patient factors, but often requires surgical intervention. We report a patient with gastrointestinal bleeding in whom pneumobilia was incidentally noted on abdominal plain film. Computed tomography and endoscopy revealed the biliary-enteric fistula to be caused by metastatic colon adenocarcinoma invading the biliary tree.

Introduction

The finding of gas within the biliary tree indicates pneumobilia. The etiology may be secondary to penetrating trauma, surgical intervention, endoscopy, gas-forming infection, or fistulization. Biliary-enteric fistulae (BEF) often arise in the setting of cholelithiasis, choledocholithiasis, or duodenal ulceration with treatment directed at the specific cause. Malignancy is a known cause of BEF, and typically is of pancreaticobiliary origin.

Case Report

A 76-year-old woman with stage 4 invasive colon adenocarcinoma, status post chemotherapy and right hemicolectomy with metastatic spread to the liver and lung, presented with abdominal pain, melena, and coffee-ground emesis. On examination, the patient was ill-appearing, lethargic, tachycardic, and hypotensive. Her abdomen was soft and non-tender to palpation. Rectal exam revealed melena. Laboratory data revealed WBC 14 k/ul, hemoglobin 4.8 g/dL (from baseline 9 g/dL), MCV 80 fl, platelets 437 k/ul, BUN 41 mg/dL, creatinine 1.2 mg/dL, and INR 2.8. She was admitted to the intensive care unit, where she was resuscitated with intravenous fluids and red blood cell transfusions. She also received an intravenous proton pump inhibitor.

Abdominal radiograph upon admission due to concern for small bowel obstruction revealed pneumobilia (Figure 1). Abdominal/pelvic computed tomography (CT) demonstrated mild biliary ductal dilatation with pneumobilia and progression of metastatic disease, including a new soft tissue mass encasing the gastric antrum and extending into the porta hepatis (Figure 2). Esophagogastroduodenoscopy (EGD) revealed LA grade D erosive esophagitis from the mid- to distal esophagus. Severe narrowing of the duodenum was noted just past the bulb, and both the true lumen of the duodenum and a choledochoduodenal fistula were visualized distal to this narrowing. The patient remained hemodynamically stable, and a duodenal stent was placed for palliation of symptoms. Eventually, the patient was discharged with home hospice services.
**Discussion**

Biliary-enteric fistulae can be categorized based on the specific communicating parts of the biliary system and intestinal tract. Stagnitti et al reported cholecystoduodenal fistulae as the most common subtype (68% or 55/81 cases), while Zong et al reported choledochoduodenal fistulae (CDF) as the most common subtype.\(^1,2\)

Most often, CDF arise in the setting of cholelithiasis, choledocholithiasis, or duodenal ulceration. Fistulae may also be observed in the post-procedural setting following pancreaticoduodenectomy, sphincterotomy, or biliary stent placement. Less commonly, neoplasms of the biliary system and surrounding structures may result in CDF. Typically, these malignancies include gallbladder carcinoma, ampullary carcinoma, cholangiocarcinoma, and pancreatic cancers. The literature includes only 1 report of primary adenocarcinoma of the duodenum resulting in choledochoduodenal fistula.\(^3\) It is also uncommon to have duodenal metastases arise from primary colon cancer.\(^4,5\) To our knowledge, this is the first reported case of choledochoduodenal fistula caused by metastatic adenocarcinoma of the colon.

Symptoms related to biliary-enteric fistulae are generally nonspecific and may include abdominal pain and vomiting.

Imaging of the biliary tree using CT or magnetic resonance cholangiopancreatography (MRCP) may be useful, though cases of pneumobilia have been diagnosed with abdominal ultrasound.\(^6\) Ultimately, EGD with or without endoscopic retrograde cholangiopancreatography (ERCP) may be necessary for diagnosis or therapeutic intervention.

Treatment of biliary-enteric fistulae is patient specific, and depends primarily on the underlying etiology of the fistula. For example, fistulae caused by ulcer disease may be treated with proton pump inhibitors, vagotomy, gastrectomy, or gastrojejunostomy. Surgical intervention is often necessary when cholelithiasis or choledocholithiasis is present, and sometimes this involves surgical anastomosis or drainage. In general, when the fistula involves the gallbladder, cholecystectomy and repair of the fistula is required.\(^7\) When the fistula involves the biliary tree, surgery may be avoided as long as there is no biliary obstruction or dilatation.\(^2\) Medical management with antibiotics may be considered in patients with mild symptoms. More recently, case reports have described the use of the over-the-scope-clip (OTSC) system to endoscopically close CDF.\(^8\) Treatment of CDF caused by metastatic disease is not well established in the literature, given the low incidence and lack of clinical trials. Management should be guided by patient-specific overall goals of care.

**Disclosures**

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