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A Rare Case of Cholecystocolonic Fistula Secondary to Choledocolithiasis

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A Rare Case of Cholecystocolonic Fistula Secondary to Choledocolithiasis
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ABSTRACT
Choledocholithiasis usually presents with signs and symptoms of cholangitis. Rarely, patients with choledocholithiasis present with biliary enteric fistulas (cholecystoduodenal, cholecystocolonic and cholecystogastric). Women in their sixth and seventh decade of life are most commonly affected. While symptoms of cholecystocolonic fistula are nonspecific, diarrhea is present in more than 70 percent of cases. Careful evaluation of common bile duct should be performed in all cases of cholecystocolonic fistula, as removal of stone leads to regression of fistula in the majority of cases. We present a case of an elderly woman referred for evaluation of fecal incontinence, weight loss, and massive pneumobilia.

INTRODUCTION
The first biliary fistula was described by Courvoisier in the 1890s. Since then cholecystoenteric fistulas, their associations, and complications have been described in the literature several times. Because of a lack of common presenting symptoms and low incidence, it usually kept last in the differential for hepatobiliary disease. Despite advanced diagnostic imaging techniques, fistulous biliary disease is mostly diagnosed during intraoperative cholangiogram. Lack of specific symptoms and diagnostic techniques makes the diagnosis of cholecystoenteric fistulas challenging. In this paper, we report a rare cause of cholecystocolonic fistula with multiple common bile duct stones. This case report also illustrates that major surgeries are not always required, and stone removal can lead to resolution of fistula along with relief of symptoms.

CASE REPORT:
An 88-year-old woman without any previous hepatobiliary disease or biliary intervention was referred to our facility for the evaluation of unexplained pneumobilia, weight loss, fecal incontinence and elevated alkaline phosphatase. The patient had been having diarrhea with overflow incontinence and weight loss of approximately 40 pounds over one year. She denied having fever, jaundice or abdominal pain. An abdominal Computerized Tomographic (CT) scan to determine the cause of weight loss showed extensive pneumobilia and dilatation of the intra and extra hepatic bile ducts. Laboratory revealed INR (International Normalized Ratio) and alkaline phosphatase levels of 1.6 and 615 U/L, respectively.

Endoscopic retrograde cholangiopancreatography (ERCP) was performed, and cholangiogram revealed a large filling defect extending from the proximal common hepatic duct down to the mid bile duct and occupying the entire width of the bile duct. The cystic duct was patent. A contracted gallbladder communicating through a fistula to the right colon was seen, as shown in Figure 1.

Biliary sphincterotomy was performed with removal of sludge. It was unclear whether this large filling defect could represent a stone or a mass. A 10-French, 12-cm long biliary stent was placed into the intrahepatic system (Figure 2).

Two weeks later, repeat ERCP was performed. The previously placed biliary stent was exiting the papilla, and it was occluded by sludge debris. Previously placed stent was removed, and cholangiogram was performed, which showed changed configuration of the previously seen large filling defect, suggesting a bile duct stone rather than a mass. Using a combination of mechanical lithotripsy, stone removal balloons, and large-diameter sphincterotomy and intra-ductal electro-hydraulic lithotripsy, the stone was nearly completely extracted. Complete removal was achieved at the time of final ERCP six weeks later. The patient’s diarrhea has resolved, and she is regaining weight.

DISCUSSION
Choledocholithiasis refers to the presence of one or more stones within the common bile duct (CBD). Biliary-enteric fistulas are very rare, comprising less than two percent of all biliary tract diseases.\(^1\)

Cholecystocolonic fistula (CCF) results forms a communication between the gallbladder and the right side of colon. CCF is usually a late complication of chronic gallstone disease and is the second most common cholecystoenteric fistula after the cholecystoduodenal fistula.\(^2\) Women are more commonly affected than men, and it usually identified in sixth and seventh decade of life.\(^1,2\)

Chronic inflammation secondary to gallstones is the most common etiologic factor. Other conditions shown to contribute to fistula formation includes previous gastric surgery, cholecystostomy, abdominal trauma and rarely peptic ulcer.\(^1-3\) Although most patients present with nonspecific symptoms, diarrhea resulting from laxative effect of bile acid bypass is the most common presenting symptom and is present in more than 70 percent of patients with CCF.\(^2\) It has been stated that the triad of pneumobilia, chronic diarrhea, and vitamin K malabsorption is a pathognomonic for cholecystocolonic fistulas; however, it is not present in all cases and has not been validated by any major studies.\(^1,3\)

The Mirizzi syndrome, a rare cause of obstructive jaundice, is also associated with fistulous biliary disease. McSherry and colleagues originally classified the Mirizzi syndrome into type 1 and type 2 in 1982, and then subsequently reclassified a couple of times into four types. Presence of cholecystobiliary fistula was classified as type 2 Mirizzi syndrome in original classification.\(^4,5\)

Because of unusual presentation and nonspecific symptoms, pre-operative diagnostic modalities often fail and diagnosis is usually made intraoperative. Several imaging techniques have been shown to identify CCF from X-ray and CT scan to an invasive procedure such as colonoscopy and ERCP, but it has been reported that only 7.9 percent of cases of CCF can be diagnosed preoperatively.\(^2\)

It has been reported in literature that CBD exploration with cholangiography should always be performed first due to an association between CBD stone and CCF. It has also been reported that extraction of CBD stone alone leads to regression of CCF, and major surgeries could be avoided. In other cases a surgical approach with either laparoscopy or laparotomy is required.\(^2,6\) In our case, complete extraction of CBD stone lead to resolution of patient’s symptoms.

**CONCLUSION**
This case illustrates that patients with otherwise unexplained pneumobilia often harbor serious underlying biliary abnormalities. Careful evaluation of CBD for the presence of stones should be performed in all cases of CCF.

**References:**
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Figure Legends:

Figure1_cholycystoclonic fistula.TIF
Figure 1. Cholangiogram revealing large biliary filling defect and a contracted gallbladder which communicated through a fistula to the right colon.

Figure2_cholycystoclonic fistula.TIF
Figure 2. ERCP: A 10-French, 12-cm long biliary stent was placed into the biliary system.