



Case Report

Mirizzi Syndrome Complicated With Transverse Colon Fistula Presenting as Colonic Tumor: A Case Report and Literature Review

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SUMMARY

An 88-year-old man was admitted because of right upper quadrant abdominal pain and diagnosed as having acute cholecystitis. He underwent endoscopic retrograde cholangiography for jaundice and showed common hepatic duct narrowing with external compression compatible with Mirizzi syndrome. Owing to his age and fragility, he received only internal drainage with a plastic stent instead of operation. The right upper quadrant abdominal pain recurred with bloody stool and decreased hemoglobin level. Colonoscopy revealed a 4- × 3-cm colonic tumor over the hepatic flexure, and colonoscopic biopsy revealed necrosis and inflammation. Abdominal computed tomography (CT) revealed ruptured acute cholecystitis and abscess formation. Surgical intervention was performed, and the operative finding showed a cholecystocolonic fistula. The patient was discharged after cholecystectomy and partial colectomy, and the period of secondary hospitalization was 46 days.

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1. Introduction

Acute cholecystitis is an inflammation of the gallbladder that occurs most commonly because of an obstruction of the cystic duct by gallstones. Elderly patients with acute cholecystitis may present with just vague symptoms. Without history and typical presentations, early diagnosis is not easy and the patient's condition may progress to complicated cholecystitis rapidly without warning. Mirizzi syndrome is a rare complication in which a gallstone becomes impacted in the cystic duct or neck of the gallbladder, causing compression of the common bile duct that results in obstructive jaundice. A fistula adjunct to a nearby organ can occur. According to the site of communication, cholecystoduodenal fistula (70%) is the most common, followed by cholecystocolonic fistula (10–20%)¹; only 0.13% were reported to cause symptoms. The most common symptoms were diarrhea, abdominal pain, jaundice, fever, nausea, vomiting, steatorrhea, and weight loss.^{1,2} Other rare origins

of spontaneous biliary-enteric fistula include cancer, amebic infections, diverticulitis, and peptic ulcers.^{1,3}

In this elderly case of cholecystitis and Mirizzi syndrome, the patient complained of bloody stool passage. The colonoscopic finding showed a colonic mass lesion over the hepatic flexure. Except for malignancy, the cholecystocolonic fistula should be taken into consideration, especially in Mirizzi syndrome.

2. Case report

An 88-year-old man with a history of diabetes mellitus and hypertension was admitted because of right upper quadrant abdominal pain for 3 days and diagnosed as having acute cholecystitis on abdominal CT. Percutaneous transhepatic gallbladder drainage failed because of thickening of the wall, small gallbladder cavity, and massive gallbladder stones. Owing to jaundice (total bilirubin level, 9.1 mg/dL), endoscopic retrograde cholangiography (ERCP) was performed and showed common hepatic duct narrowing with external compression, compatible with Mirizzi syndrome (Fig. 1). Then, an internal drainage with a 7-Fr 12-cm plastic stent was placed over the main bile duct with good drainage. The patient was discharged 8 days later after stent placement his condition stabilized without operation because his family was

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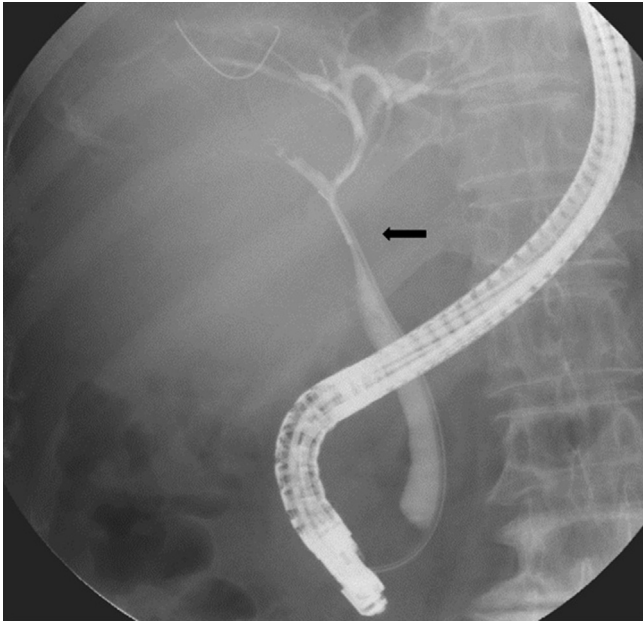


Fig. 1. An endoscopic retrograde cholangiography image showing common hepatic duct narrowing (arrow) with external compression, compatible with Mirizzi syndrome.

concerned about his age and weakness, which may increase the risk of surgical complications.

Intermittent right upper quadrant abdominal pain was observed 9 days later since discharge (17 days later after stent placement), and he was admitted again (21 days later after stent placement) because of progressive abdominal pain. On admission, bloody stool was observed (31 days later after stent placement) and his hemoglobin level declined from 8.9 to 6.8 g/dL. Esophagogastroduodenoscopy revealed no active bleeding. By using colonoscopy, a 4- × 3-cm colonic mass lesion was detected over the hepatic flexure (Fig. 2), and pathological examination from colonoscopic biopsy revealed necrosis and inflammation. Abdominal CT revealed acute

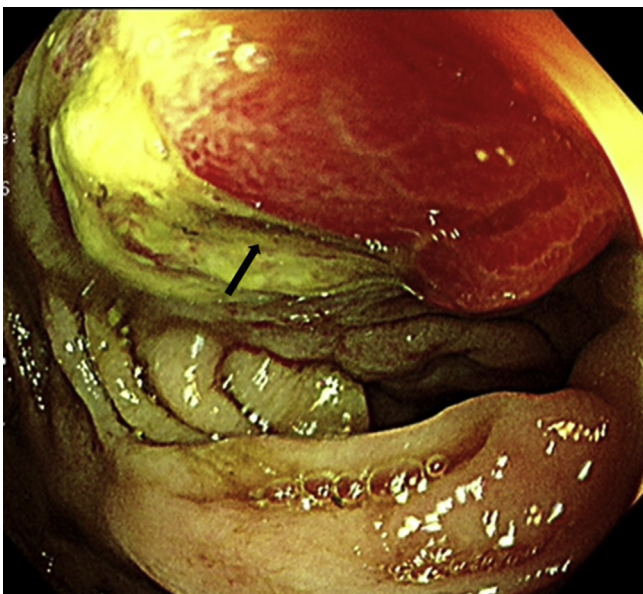


Fig. 2. Colonoscopy image showing a 4- × 3-cm colonic tumor (arrow) over the hepatic flexure.



Fig. 3. Abdominal computed tomography revealed acute cholecystitis with rupture and abscess formation (arrow).

cholecystitis with rupture and abscess formation (Fig. 3). Under the impression of cholecystitis with rupture and because colon tumor could not be ruled out, the patient was referred for treatment with surgical intervention and a tentative diagnosis of colonic abscess formation was made. Surgical intervention with cholecystectomy and partial colectomy were performed, and operative findings showed a cholecystocolonic fistula, which was confirmed on the basis of the pathological finding. The patient was discharged after further treatment with an antibiotic agent, and the time of secondary hospitalization was 46 days.

3. Discussion

Spontaneous biliary-enteric fistula is relatively rare. Most cases result from complications of gall stone disease, but other rare origins include cancers, amebic infections, diverticulitis, or peptic ulcers.^{1,3} During acute cholecystitis, the adjacent serosal surface has an inflammation and is adherent to the gall bladder. The gall bladder wall could become gangrenous over the ischemic area. Owing to the increased pressure within the gall bladder, its contents penetrate the necrotic wall to the adjacent colon, forming a cholecystocolonic fistula.⁴ Mirizzi syndrome has a good reason to form a cholecystocolonic fistula because when the stone impacts over the gall bladder neck or cystic duct, the pressure in the gall bladder is increased. The natural history of Mirizzi syndrome may not end with just a cholecystobiliary fistula. It may show continuous inflammation in the triangle of the Calot area that may result

in a complex fistula involving the biliary tract and adjacent viscera. Beltran retrospectively reviewed the records of all patients older than 18 years who underwent an emergency or elective cholecystectomy between 1995 and 2006. Of 5673 cholecystectomies performed during that period, 327 patients (5.7%) had Mirizzi syndrome and 105 (1.8%) had cholecystoenteric fistula. Ninety-four patients (89.5%) with cholecystoenteric fistula also had an associated Mirizzi syndrome ($p < 0.0001$).⁵

According to studies performed in a large series of more than 10,000 patients who underwent cholecystectomy, the incidence of the cholecystocolonic fistula during this procedure was reported to be 0.06–0.14%. The female-to-male ratio in the series was 2.47:1. The mean age of the patients at diagnosis was 68.9 years (range, 37–90 years), with 70.8 years reported in Western papers and 62.1 years reported in the Eastern ones.^{2,4,6,7}

No specific clinical symptoms or signs led to the diagnosis of a fistula. Only 0.13% of cases were reported to cause symptoms. Diarrhea is by far the most frequent symptom (71%)² due to the laxative effects of bile acids that bypass the distal ileum and reach the transverse colon unabsorbed. Other symptoms include abdominal pain, jaundice, fever, nausea, vomiting, steatorrhea, and weight loss.^{1,2} The causes of acute onset of symptoms in cholecystoenteric fistula are obstruction (biliary stone ileus), massive bleeding, and liver abscess (18%, 6%, and 1.7%, respectively).² In our patient, blood stool passage was a relative rare complication that is difficult to associate with the underlying disease. From the present case, we can know that fistula formation could occur in a short period (about 1 month) from the last ERCP finding (no evidence of fistula) to symptom onset.

The diagnostic tools for cholecystoenteric fistula include abdominal plain film, ERCP, abdominal ultrasonography, barium studies, and biliary scintigraphy.¹ Cholecystoenteric fistula is still difficult to diagnose and usually diagnosed intraoperatively.⁶ Pneumobilia may be an indicator of cholecystoenteric fistula, especially if the gallbladder is atrophic and anatomically adjacent to another organ on computed tomography or ultrasonography.⁸ Savvidou et al proposed a diagnostic triad composed of pneumobilia, chronic diarrhea, and vitamin K malabsorption. However, this triad was not observed in all the patients, and no further studies proved the validation of this triad.⁹

The recommended definitive procedure includes cholecystectomy, excision of the fistula, common bile duct exploration, and operative cholangiography.⁴ Owing to the recent developments in laparoscopic surgery, the results of intraoperative and postoperative complications have shown no significant differences between traditional laparotomy and laparoscopic surgery. Laparoscopic surgery would be another option depending on the patient's condition.⁷ Though not frequent, gallbladder carcinoma and cholecystocolonic fistula may coexist in the same patient (incidence of approximately 0.3%). The finding of a hard-to-dissect,

fistulized-to-colon gallbladder should prompt the surgeon to take a frozen section of the specimen.² Prasad and Foley reported that a safety margin should be considered during colonic resection for cholecystocolonic fistula because gallbladder cancer could possibly coexist.¹⁰ Elective cholecystectomy following by endoscopic treatment such as sphincterotomy combined with stone extraction are highly suggested for the elderly patient had cholangitis with common bile duct stone. The prophylactic cholecystectomy could further prevent recurrent cholangitis.¹¹

Colonic carcinoma may be the prior considerable diagnosis in the elderly with a colonic mass with bleeding, alternating diarrhea and constipation, and weight loss. Without pathological confirmation, the differential diagnosis of colonic mass includes abscesses formation, ameboma of the colon, endometriosis in women, and diverticulitis in the elderly. When a patient has Mirizzi syndrome without definitive operation, it may result in a complex fistula with neighboring digestive organs (cholecystogastric, cholecystoduodenal, and cholecystocolonic fistulas). The most common symptom and sign are unexplained persistent diarrhea but may also present as bloody stool in the presence of cholecystocolonic fistulas.

Conflicts of interest

All authors declare no conflicts of interest.

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