

Bladder Networking: A Unique Case of Cholecystovesicular Fistula

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ABSTRACT

We present a case of a 60-year-old woman with chronic lower abdominal pain and green urine. Further workup revealed a cholecystovesicular fistula (CVF), a newly coined term to indicate a fistula between the gallbladder and the urinary bladder. The CVF was treated surgically. The pathophysiology of CVF is thought to result from gallbladder perforation into the liver. Over time, a tract forms inferiorly until it meets another organ, in this case, the urinary bladder. This later complication of the gallbladder disease joins the broader spectrum of cholecystic fistulas. To our knowledge, a CVF has never been reported in the literature.

INTRODUCTION

The most common type of internal biliary fistula is a cholecystoduodenal fistula, which accounts for 75% of all cholecystoenteric fistulas.^{1,2} Cholecystocolonic fistula is the second most common type (10%–20%).^{1,3,4} The patient exhibited a cholecystovesicular fistula (CVF), a newly coined term to indicate a fistula between the gallbladder and the urinary bladder. The diagnosis was made clinically, and the patient presented with frequent urinary tract infections (UTIs), green urine, and a computed tomography (CT) scan suspicious for CVF. This diagnosis was confirmed intraoperatively with gross visualization and cholangiogram. We managed the CVF surgically with a cholecystectomy and bladder repair.

CASE REPORT

A 60-year-old woman with a history of chronic back pain, anxiety, and gastritis presented to our institution with green-tinged urine and lower abdominal pain, but no nausea, vomiting, fever, or chills. The patient stated that she had 1 year of lower abdominal pain, and for the past 2 months, she developed recurrent UTIs unresponsive to antibiotics. She also reported an unintentional 40 lbs weight loss over the past 3 to 4 months. One week before presentation, she noticed green streaks in her urine. The patient's surgical history included back surgery and tonsillectomy. No allergies were indicated. Home medications included oxycodone, baclofen, diazepam, and lansoprazole. The patient reported smoking half a pack of cigarettes a day for 40 years and denied alcohol or drug use. Her mother has a history of kidney cancer.

On admission, her abdomen was mildly tender to palpation in the right lower quadrant with no rebound or guarding. The patient's laboratory data showed white blood cell count 10.5×10^9 cells/L, hemoglobin 12.6 gm/dL, hematocrit 37.8%, platelets 466×10^9 cells/L, sodium 133 mEq/L, potassium 4.6 mEq/L, blood urea nitrogen 7 mg/dL, creatinine 0.7 mg/dL, elevated alkaline phosphatase 156 IU/L, alanine aminotransferase 45 IU/L, aspartate aminotransferase 28 IU/L, albumin 3.4 g/dL, and total bilirubin 0.2 mg/dL. She saw a urologist who ordered a CT scan, which showed a perforated gallbladder, a pelvic abscess abutting the urinary bladder, and a fistulous tract connecting them both (Figure 1).

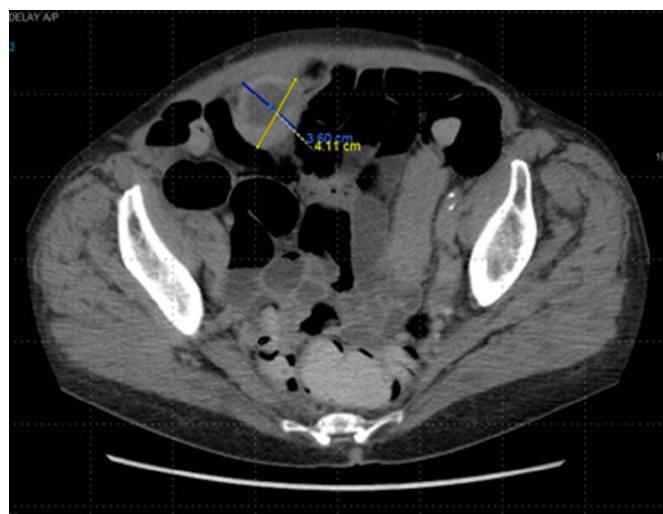


Figure 1. Computed tomography scan showing pelvic abscess abutting the bladder.

A magnetic resonance cholangiopancreatography showed a dilated common bile duct measuring 13 mm with distal tapering, no filling defects, gallbladder sludge, no gallstones, and a fistulous tract from the inferior aspect of the gallbladder through the liver into a pelvic fluid collection superior to and abutting the bladder.

The patient was admitted, started on intravenous antibiotics, and prepared for surgery. Once in the operating room, our urology colleagues began with a cystoscopy, which showed an internal opening near the dome of the bladder. Because of the complex nature of the case and our comfort level with laparoscopy, given the potential degree of intra-abdominal inflammation, we felt that an open approach was safer. A midline laparotomy was made, which revealed a contracted gallbladder. There was chronically inflamed tissue that originated from the gallbladder and traveled inferiorly toward the bladder. This was consistent with a fistula that extended through segment 5 of the liver with a palpable fistula tract, which led directly into an abscess abutting the urinary bladder.

At this point, we turned our attention back to the gallbladder. The triangle of Calot was dissected, and the critical view of safety was obtained without difficulty. We opted to perform a cholangiogram to help delineate the fistula tract. The cholangiogram clearly outlined the gallbladder, cystic duct, common hepatic duct, right and left hepatic ducts, and common bile duct, as well as the fistula and abscess immediately on injection of contrast (Figure 2). We had confirmed our preoperative diagnosis of CVF. The gallbladder was dissected out, and a small portion of segment 5 of the liver containing the tract was also removed. The tract was dissected free from omentum and colon until it was only attached to the bladder by the abutting abscess (Figure 3). The abscess was separated from the bladder, and the specimen was removed and sent to pathology. The bladder was closed over a suprapubic tube (Bard Medical, Covington, GA),

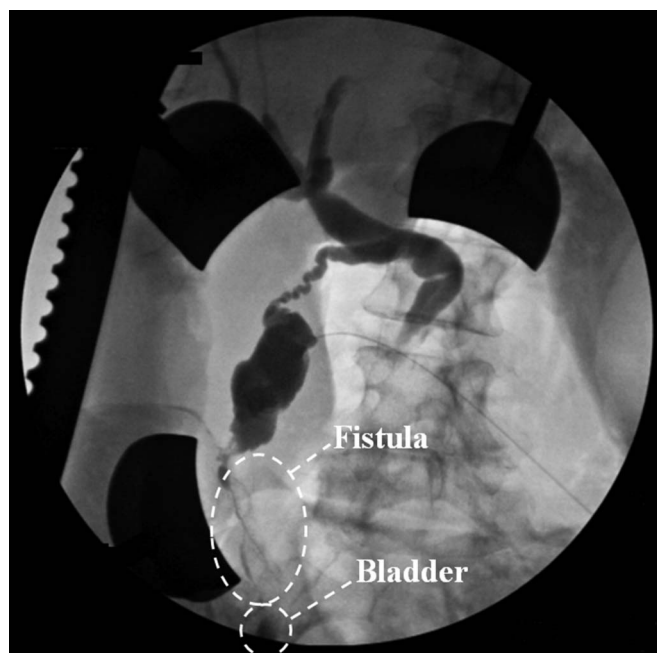


Figure 2. An intraoperative cholangiogram delineating the right and left hepatic ducts, common hepatic duct, common bile duct, cystic duct, gallbladder, and fistulous tract from the inferior aspect of the gallbladder through the liver to the bladder.

and a Jackson-Pratt drain (Bard Medical) was left over the bladder suture line. The patient was closed in standard fashion, extubated, and brought to the recovery room in stable condition.

On the floor, the patient did well and discharged on post-operative day 8 with her suprapubic tube in place. The tube was

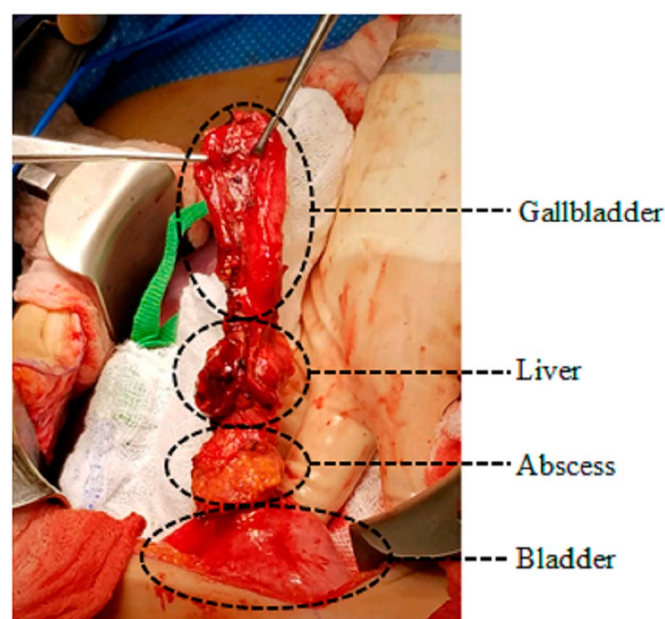


Figure 3. An intraoperative view of the gallbladder, liver, abscess, and bladder from superior to inferior before removing the specimen.

removed by urology as an outpatient. She followed up with us in the office after her tube removal and was doing well with no postoperative issues. Pathology showed chronic cholecystitis, serosal material consistent with bile, marked chronic histiocytic and foreign-body giant cell reaction, fibrosis, abscess formation, and adherent liver tissue, consistent with ruptured gallbladder with fistula and abscess formation with bile-stained urothelial mucosa and muscularis propria consistent with gallbladder to bladder fistula.

DISCUSSION

The report highlights the first documented CVF, a term devised from this case. The patient presented with abdominal pain, which is also a symptom of other cholecystic fistulas,^{2,3} but also had frequent UTIs and biliuria which seem to be unique to this case. Interestingly, the diagnosis of a CVF was clear preoperatively, based on the CT scan, magnetic resonance cholangiopancreatography, and clinical presentation. However, this is not common for other cholecystic fistulas. Most cholecystic fistulas are diagnosed intraoperatively,⁵ with studies showing a preoperative diagnosis rate of 7.9% for cholecystocolonic fistulas,³ and a preoperative diagnosis rate of 31% for cholecystoenteric fistulas.⁶ More women than men have been cited to have cholecystoenteric and cholecystocolonic fistulas, and the average age of these patients commonly falls between 60 and 70 years.^{3,6}

Although the case is rare and not written about in the literature, it begs the question, why did this occur? The etiology of these cholecystic fistulas is thought to be because of adhesion of the gallbladder to the viscera (such as small bowel) after a gallstone obstructs the cystic duct. This obstruction, along with inflammation, can cause the walls of the gallbladder and the neighboring viscus to degrade, allowing for fistulas to propagate.^{3,5} Others note that the pressure of the gallstone can also erode into the surrounding tissues and contribute to fistula formation.⁵ Another possibility is that the pathophysiology is similar to that of Crohn's inflammation mediated by unrestrained T-helper cells and their cytokines such as interleukin-12 and tumor necrosis factor- α stimulating the inflammatory response.⁷

Because this is the first published case of CVF, the pathophysiology is unclear. Our hypothesis based on this case and the intraoperative findings is related to long-standing chronic cholecystitis, leading to intrahepatic perforation. Over time, before presentation, this may have manifested as abdominal pain and subjective fevers. The patient had a very long liver,

extending inferiorly and nearly touching the bladder. This may have acted as a bridge for the fistula tract to connect the gallbladder to the urinary bladder. We believe that the perfect arrangement of time, inflammation, and convenient anatomy resulted in a CVF and biliuria. This presentation of a unique fistula provides insight on a new type of cholecystic fistula and its surgical management.

DISCLOSURES

Author contributions: AM Nouri, ZH Nemeth, GM Cavallo, AN Lazar, K. Kong, and KA Bickenbach wrote the manuscript. AM Nouri, ZH Nemeth, GM Cavallo, and K. Kong edited the manuscript. ZH Nemeth is the article guarantor.

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REFERENCES

1. Ha GW, Lee MR, Kim JH. Cholecystocolic fistula caused by gallbladder carcinoma: Preoperatively misdiagnosed as hepatic colon carcinoma. *World J Gastroenterol*. 2015;21(15):4765–9.
2. Agrawal V, Joshi U, Manandhar S. Spontaneous cholecystocolic fistula: An uncommon complication of chronic cholecystitis. *Clin Case Rep*. 2017; 5(11):1878–81.
3. Costi R, Randone B, Violi V, et al. Cholecystocolonic fistula: Facts and myths. A review of the 231 published cases. *J Hepatobiliary Pancreat Surg*. 2009;16(1):8–18.
4. Balent E, Plackett TP, Lin-Hurtubise K. Cholecystocolonic fistula. *Hawaii J Med Public Health*. 2012;71(6):155–7.
5. Chowbey PK, Bandyopadhyay SK, Sharma A, Khullar R, Soni V, Baijal M. Laparoscopic management of cholecystoenteric fistulas. *J Laparoendosc Adv Surg Tech A*. 2006;16(5):467–72.
6. Li XY, Zhao X, Zheng P, Kao XM, Xiang XS, Ji W. Laparoscopic management of cholecystoenteric fistula: A single-center experience. *J Int Med Res*. 2017;45(3):1090–7.
7. Ghazi LJ. Pathophysiology. Medscape. Crohn disease website. (<https://emedicine.medscape.com/article/172940-overview#a3>) (2018). Accessed on November 3, 2018.

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