



Broncho-biliary fistula: a rare complication of subphrenic abscess due to perforated gallbladder

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Background

- A 2016 to 2020 retrospective cohort study found a 2.7% incidence of gallbladder perforations prior to surgery. Risk factors include male gender, diabetes, hypertension, and hyperlipidemia. The most common site of perforation was the fundus (42.86%). The median length of hospital stay was 8.5 days, compared to 4 days for patients diagnosed with acute cholecystitis without gallbladder perforations.¹
- Objective findings include tachycardia and elevated white blood cell (WBC) count, total and direct bilirubin, and gamma-glutamyl transferase (GGT) compared to non-perforated gallbladders undergoing cholecystectomy.¹
- Niemeier in 1934 was the first to describe and categorize gallbladder perforations. Type 1 perforations are acute and spread into the peritoneal cavity. Type 2 perforations, the most common type, are subacute and develop a surrounding abscess walled off by peritoneal adhesions. Type 3 perforations develop chronically and eventually lead to a fistulous connection between the gallbladder and other viscera, such as the bowel.² Interestingly, there was no difference in mortality amongst the different types.³
- As expected, in 86.6% of patients, perforations were associated with cholelithiasis.³
- An extremely rare complication of gallbladder perforations is a broncho-biliary fistula. Compared to gallbladder perforations, there is a lower incidence and lack of consensus around the management of broncho-biliary fistulas, making this a fascinating topic for further study.

Case Description

Brief History

- 62-year-old Caucasian male presents with two weeks of persistent generalized abdominal pain and decreased appetite with associated nausea/vomiting/diarrhea (N/V/D), chills, and weakness. Also reports chest pain, shortness of breath, and dizziness. Accompanying him, his sister states that his skin and eyes have changed color.
- Reports taking 1,000 milligrams acetaminophen every 4-6 hours x2 weeks without relief.
- Past Medical History: Gastroesophageal reflux disease, obstructive sleep apnea, hyperlipidemia, hypertension, degenerative disc disease
- Health maintenance: Last colonoscopy January 2021 with polypectomy (pathology report unavailable)
- Social History: Drinks 10 to 12 beers daily; Smokes 10 to 12 cigarettes daily.

Pertinent Diagnostic Findings

WBC 23.7 k/uL (H)
 Absolute Neutrophils 20.2 k/uL (H)
 Lactic Acid 2.3 mmol/L (H)
 Prothrombin time 19.9 seconds (H)
 INR 1.7 (H)
 Albumin 3.0 g/dL (L)
 Total bilirubin 2.2 mg/dL (H)
 Direct bilirubin 1.1 mg/dL (H)
 Alk phos 215 U/L (H)
 AST 101 U/L (H)
 ALT 168 U/L (H)
 Lipase 8 U/L (L)

Assessment and Plan

RUQ Peritoneal Drain into Subphrenic Abscess

- Left in place, feculent foul-smelling drainage throughout the hospital stay
- Contrast demonstrated fistula between right subphrenic cavity and gallbladder lumen/cystic duct

Right Pleural Chest Tube and Intrapleural thrombolysis

- Left in place, bilious drainage throughout the hospital stay

Confusion secondary to alcohol intoxication

- CIWA protocol, thiamine, and folic acid

Day #6

- Cultures grew Enterococcus, E. coli, Klebsiella, Bacteroides, alpha Strep, Candida.
- Piperacillin/tazobactam and fluconazole initiated.
- Second right pleural chest tube inserted.

Day #11

- Video-assisted thoracoscopic surgery (VATS) with decortication of the right lung.
- Procedure revealed a diaphragmatic defect.

Day #12-17

- Received low-dose vasopressors and fluids. Extubated.
- Acute on chronic anemia treated with packed red blood cells.

Day #18

- Endoscopic retrograde cholangiopancreatography (ERCP)
- Sphincterotomy with balloon sweep and metal stents placed.

Day #23-30

- All drains removed. Tolerated regular diet.
- Moderate left pleural effusion requiring thoracentesis.

Objective Findings

Blood pressure: 94/54 mmHg
 Heart rate: 112 beats per minute
 Temperature: 98.1 degrees Fahrenheit
 Respiratory rate: 17 breaths per minute
 Pulse oximetry: 96% on room air
 Body mass index: 28.98 kg/m²

- Patient was **ill-appearing** but not in acute distress. Lethargic.
- Scleral icterus, generalized jaundice, and pale skin present.** No ecchymoses or rashes. **Mucus membranes were dry.**

- No signs of respiratory distress. **Expiratory wheezing and diminished breath sounds bilaterally.**

- Normal rate and regular rhythm. No murmurs, rubs, gallops.

- Abdomen soft and **distended.** Normative bowel sounds. Tympanic to percussion. **Generalized abdominal tenderness that intensified in the epigastric and right upper quadrant (RUQ) regions**

- No guarding or rebound. No CVA tenderness bilaterally. Negative psoas, negative obturator.

- Tremors of the hands bilaterally with movement.** A/Ox4. No focal deficits. No peripheral ataxia, asterixis, or spasticity.

- Remainder of the exam unremarkable.

Discussion

- This case report demonstrates a **type 2 Niemeier perforation** complicated by a right broncho-biliary fistula.
- CT was an appropriate diagnostic modality. Confirmed and suspected perforations are more often found on CT rather than abdominal ultrasound in those with acute gallbladder perforations.¹ Additionally, case reports of broncho-biliary fistulas show that complete blood count, comprehensive metabolic panel, CT, and ERCP were appropriate at making the diagnosis.⁴
- The management was consistent with Figure 1, an algorithm for type 2 perforations proposed by a systematic review.³
- There is not a standard treatment for broncho-biliary fistulas. However, the patient in this case study was treated successfully with non-surgical drainage procedures, which is consistent with the following studies.
- A February 2021 study discusses two case reports of broncho-biliary fistulas. The first was an 80-year-old male with a history of diabetes, alcohol use, and hepatocellular carcinoma. He was treated non-surgically with drainage procedures and survived. The second patient was a 68-year-old female with a history of cirrhosis, hepatitis C, and hepatocellular carcinoma. In addition to drainage, she was treated surgically with bronchial occlusion and percutaneous transhepatic portal vein embolization but eventually died.⁴
- In the future, the treatment of broncho-biliary fistulas may include material to block fistulous drainage into the lungs, as described in a November 2023 case report. A broncho-biliary fistula found in a 63-year-old male with a history of metastatic colon cancer was treated with the Amplatzer Vascular Plug inserted into the bronchus. There was successful control of biliary leakage into the lung.⁵

Conclusion

- Clinicians should suspect gallbladder perforations in ill-appearing patients presenting with epigastric/RUQ abdominal pain, fevers, N/V/D, and jaundice.
- Broncho-biliary fistulas are a rare complication of gallbladder perforations. In the few cases reported, treatment always includes drainage procedures.
- Despite the successful outcome of this case report, future research is necessary to reduce complications and hospital stays in patients with gallbladder perforations.

References

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Outcome

- Upon discharge, patient had an elevated white count (14.3), low hemoglobin (7.7), and low hematocrit (24.8).
- The patient was discharged to skilled nursing facility with four-week course of amoxicillin-clavulanate 875-125 mg twice daily and fluconazole 400mg once daily.
- He had multiple follow-up visits with thoracic surgery, general surgery, infectious disease, and gastroenterology. Awaiting cholecystectomy followed by biliary stent removal in 2-3 months.



Image 1: Computed Tomography (CT) Abdomen Pelvis With Intravenous (IV) Contrast.

- Gas-filled gallbladder with the stones remaining present in the gallbladder lumen.
- Discontinuity of the gallbladder wall communicating with a large perihepatic collection containing gas and fluid consistent with abscess.

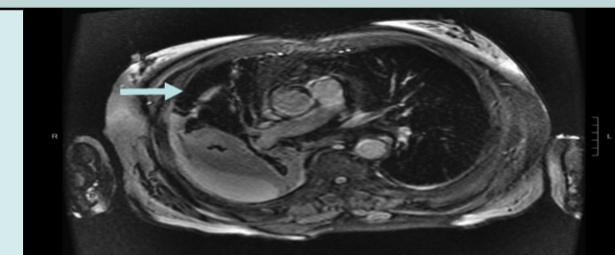


Image 2: Magnetic Resonance Imaging (MRI) Abdomen with and without IV contrast

- Cholelithiasis, Choledocholithiasis
- Gas in the gallbladder with fistulous connection to the patient's right subphrenic air/fluid collection.
- Right pleural effusion & atelectasis. Slight left pleural effusion.

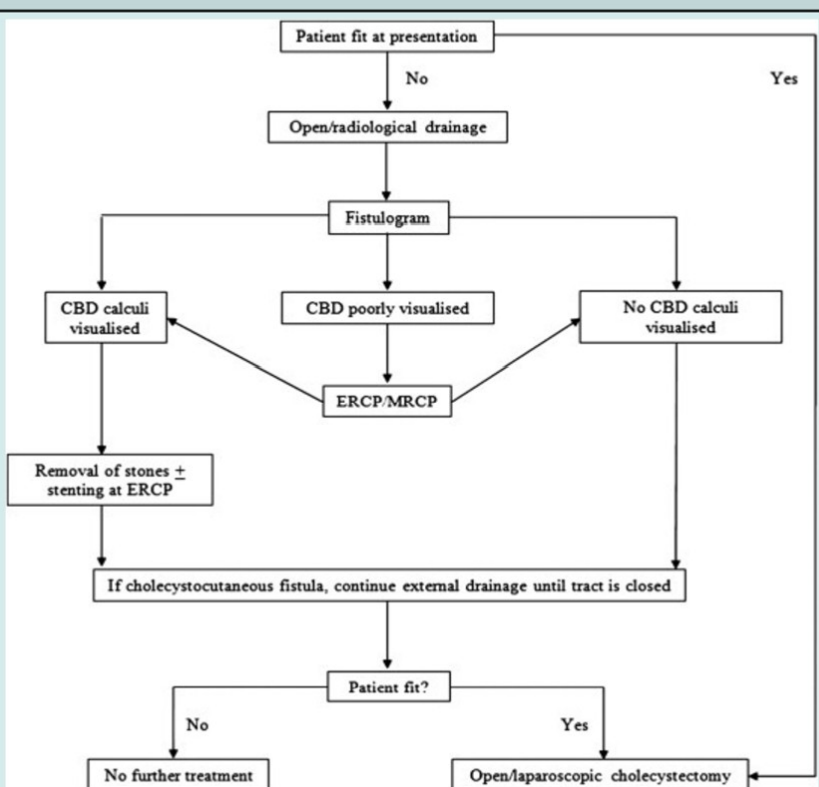


Figure 1: Proposed algorithm for managing type II gallbladder perforations Adapted from [Gallbladder perforation: case series and systematic review]³