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Case Report

The Cholecystocutaneous Fistula: A Rare Manifestation of Neglected Cholecystitis

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ABSTRACT

Introduction: Cholecystocutaneous fistula is a rare complication of acute or chronic cholecystitis, most often seen in the elderly in whom diagnosis is delayed. Management can be difficult due to its late presentation, patient comorbidities, and poor candidacy for a major surgical intervention. We report the management of a case of cholecystocutaneous fistula in a patient with chronic lymphocytic leukemia who presented in acute sepsis.

Case Report: An 89-year-old female presented with several days of abdominal pain, fever, and a right upper quadrant abdominal wall abscess with surrounding cellulitis. A CT demonstrated fistulization of the gallbladder to the anterior abdominal wall. The patient was stabilized in our ICU and external drainage of the abscess and decompression of the gallbladder was performed for source control. Physicians should be aware of the cholecystocutaneous fistula as a late complication of undiagnosed cholecystitis in the elderly patient population as well as the surgical options available for treatment.

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Introduction

The complications related to cholecystitis are well documented including cholangitis, perforation, peritonitis, and sepsis. An infrequent and insidious complication of cholecystitis is the cholecystocutaneous fistula. The overarching theme of predisposition to developing a cholecystocutaneous fistula includes a prolonged course of chronic cholecystitis. However, the patterns and circumstances surrounding the development of this complication are still ill-defined. We present the case of an 89-year-old female of Lebanese descent to further elucidate potential causes contributing to its evolution.

Case Presentation

Our patient was an 89-year-old Lebanese Arabic-speaking female with a medical history significant for chronic lymphocytic leukemia (CLL), chronic kidney disease (CKD), congestive heart failure (CHF), coronary artery disease (CAD), hypertension, hyperlipidemia, chronic pain, and

dementia, who presented in transfer to the medical intensive care unit of our hospital from an outside facility after cross-sectional imaging demonstrated ruptured necrotic cholecystitis with fistulization to the anterior abdominal wall containing abscess filled with air and fluid (Figure 1). Of note, the patient had a prior CT available from seven months prior to her presentation that showed close proximity of the gallbladder to the anterior abdominal wall (Figure 2). On review of her history, she had experienced 5 days of abdominal pain on a history of chronic cholecystitis. At the time of surgical consultation, she was critically ill, requiring vasoactive support and intravenous antibiotics. Her laboratory values showed a white cell count of 387,000 (lymphocyte-predominant in a background of CLL) with hemoglobin of 9.2g/dl, platelet count of 213 k/uL, international normalized ratio (INR) of 1.8, a lactic acid of 3.1 mEq/L, and an acute on chronic kidney injury with a creatinine of 2.57 mg/dl (baseline 1.3-1.5 mg/dl). She was in septic shock on a norepinephrine infusion for vasoactive support. She had a cardiac ejection fraction of 55% by recent echocardiogram. After extensive discussions with her family, she was brought to the operating room for source control.

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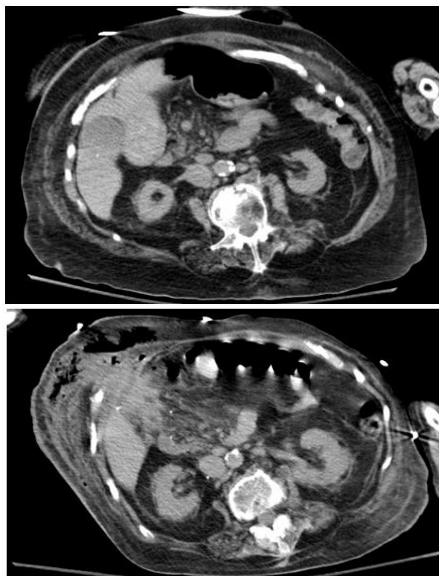


Figure 1: Representative cross-sectional images showing gallstones and a ruptured cholecystitis contiguous with a large air- and fluid-filled abdominal wall abscess.

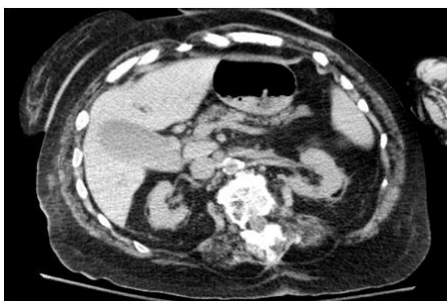


Figure 2: A CT scan of our patient 7 months prior, highlighting the presence of cholelithiasis and proximity of the gallbladder fundus to the abdominal wall.

After general anaesthesia was safely initiated, we performed incision and drainage of the subcutaneous abdominal wall abscess through a Kocher incision in the right upper quadrant (Figure 3). Intraoperatively, purulent fluid was evacuated and sent for culture. The abscess cavity was then irrigated and noted to extend into the gallbladder fundus. Completion cholecystectomy was not performed given her acute sepsis, with plans to later return to the operating room for definitive surgical management once she recovered and hemodynamically stable. The abscess cavity was packed with sterile moist gauze and she remained intubated and was transferred to the surgical intensive care unit for continued management. She remained critically ill over the next several days with increasing vasopressor requirements. Her intraoperative cultures were polymicrobial, growing *Klebsiella*, *Streptococcus viridans*, *Candida albicans*, and *Staphylococcus* species; she remained on broad spectrum antimicrobial coverage. Unfortunately, she continued to deteriorate into multiple system organ failure due to her superimposed chronic medical conditions and went into progressive nonischemic cardiac failure. The family wished for no further escalation in care and elected for palliative extubation. The patient expired shortly after withdrawal of ventilatory and vasoplegic support.



Figure 3: Intraoperative photo depicting a Kocher incision into the abdominal wall abscess and fistulization to the gallbladder fundus.

Discussion

Spontaneous cholecystocutaneous fistula is a particularly rare complication of cholecystitis, with fewer than approximately 20 cases reported in the literature in the past 50 years [1]. Modern diagnostic methods, antibiotics, and early effective surgical treatment for cholecystitis have all likely contributed to the low incidence of this particularly morbid complication. Generally, the patient presents after a long duration of symptoms suspicious for cholecystitis, with an abscess or fistula in the right upper quadrant, though less common areas such as the epigastrium, umbilicus, and right groin have been described [2, 3]. The majority of patients are over the age of 60 and have multiple medical comorbidities which can lead to a delay in presentation and diagnosis [1, 4-8].

Several approaches for management of this unusual complication have been proposed (Table 1), dependent on the acuity of the presentation. Generally, most authors have put forward a two-step surgical approach. Initially, external drainage of the abscess for biliary decompression and antibiotics can be effective in controlling sepsis on presentation and is the preferred approach for patients presenting in poor clinical condition. This can generally serve to be sufficient treatment in the short-term as the cystic duct is usually obstructed and a true fistulous tract to the biliary system can be avoided [5]. Following stabilization, an open cholecystectomy with or without excision of the fistula tract may be performed [8, 9]. For stable patients who may present with relatively few symptoms other than an abscess or draining fistula tract, a single stage definitive operation with external drainage and open cholecystectomy has been considered the treatment of choice [1, 7, 10]. A laparoscopic approach to this single-stage operation has been reported with success by several authors, though generally this is reserved for patients who have a more subacute presentation [11-13]. In patients too frail or unstable to tolerate an operation, treatment with antibiotics, supportive care, and source control with bedside incision and drainage, percutaneous removal of stones and biliary decompression by ERCP, or CT-guided drain placement have been advocated [5, 14, 15].

Table 1: Management of Previous Reported Cholecystocutaneous Fistula Cases.

Report	Patient	Presentation	Time from onset of symptoms	Treatment	Outcome
Davies <i>et al.</i> [5]	84 y/o male	Draining sinus tract	3 months	Incision and drainage, foley placed into gallbladder for drainage	Patent biliary system on delayed cholecystogram, successful fistula tract closed after drain removal
Cruz <i>et al.</i> [1]	81 y/o male	Right subcostal abscess	28 days	Midline laparotomy, cholecystectomy, external drainage of abscess	Death from massive pulmonary embolism, POD3
Malik <i>et al.</i> [12]	76 y/o female	Abdominal wall abscess	N/A	Laparoscopic cholecystectomy	Recovery and discharge
Pezzilli <i>et al.</i> [14]	90 y/o female	Low grade fevers, diarrhea, erythema along right abdominal quadrant	4 days	CT-guided drainage of abscess, IV antibiotics, supportive care	Death on HD3
Yuceyar <i>et al.</i> [9]	70 y/o female	RUQ pain, abdominal wall swelling	8 weeks	Incision and drainage, IV antibiotics, delayed open cholecystectomy and excision of fistula tract (HD12)	Uneventful postoperative recovery
Kumar <i>et al.</i> [11]	72 y/o male	Fever, leukocytosis, RUQ subcutaneous abscess	N/A	Laparoscopic cholecystectomy, open drainage of abdominal wall abscess	Recovery, no reported complication
Sayed <i>et al.</i> [15]	85 y/o female	RUQ abscess and drainage	3 months	ERCP with stone removal and sphincterotomy	Progressive fistula healing
Khan <i>et al.</i> [10]	76 y/o male	RUQ pain, swelling	N/A	Open cholecystectomy	N/A
Ozdemir <i>et al.</i> [8]	89 y/o female	RUQ pain, abdominal wall abscess	3 days	IV antibiotics, delayed open cholecystectomy and excision of fistula tract, CBD exploration and removal of stone (HD24)	Uneventful postoperative recovery
Kapoor <i>et al.</i> [7]	45 y/o male	RUQ and biliary discharge from previous scar	1.5 months	Fistula exploration, open cholecystectomy, excision of fistula tract	Uneventful recovery in both patients
	65 y/o male	Chronic draining sinus of the RUQ	2.5 years	Open cholecystectomy, excision of fistula tract	
Pol <i>et al.</i> [13]	70 y/o female	RUQ pain, swelling, draining sinus tract	2 years	Laparoscopic cholecystectomy and excision of fistula tract	Discharge on POD3, fistula tract healed at POD21

¹HD=Hospital Day, ²POD=Postoperative Day, ³N/A=Information not available, ⁴RUQ=Right upper quadrant of abdomen.

Conclusion

Cholecystocutaneous fistula has become a rare phenomenon in an age of early diagnosis and timely intervention for acute cholecystitis. The complication is most often seen in elderly patients with multiple comorbidities who frequently present after an insidious course of acute cholecystitis and are poor surgical candidates. Here we have presented a case of this increasingly uncommon entity and review the literature for possible approaches to treatment. Physicians should be aware of the cholecystocutaneous fistula as a late complication of undiagnosed cholecystitis in this patient population as well as surgical options for

treatment, though the literature does not conclude upon one gold standard approach for all cases.

Disclosure

None.

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Author Contributions

Conceptualization: Davek Sharma, Jacob Katsnelson, James Butz; Writing: Davek Sharma, Jacob Katsnelson, James Butz; Editing: Davek Sharma, Jacob Katsnelson, James Butz, Jeffery Kolff; Davek Sharma: Primary author, researcher, and resident surgeon for case and post-op care; Jacob Katsnelson, James Butz: Researchers and resident surgeons for ICU post-op care; Jeffrey Kolff: Attending surgeon for case; Guarantors: Davek Sharma, Jeffrey Kolff.

Conflicts of Interest

None.

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Ethical Approval

The authors did not seek IRB approval for this case report, which contains only retrospective, deidentified patient information. The writing or publication of this case report did not affect this patient's treatment or outcomes in any way. There are no ethical dilemmas with this case.

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