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.

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 elderly emphysematous cholecystitis in 2019. The patient 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 1.5 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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Cholecystocutaneous Fistula in CLL

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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.

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].

Figure 3: Intraoperative photo depicting a Kocher incision into the abdominal wall abscess and fistulization to the gallbladder fundus.
Table 1: Management of Previous Reported Cholecystocutaneous Fistula Cases.

| Report          | Patient | Presentation                        | Time from onset of symptoms | Treatment                                                                 | Outcome                                                                 |
|-----------------|---------|-------------------------------------|-----------------------------|---------------------------------------------------------------------------|--------------------------------------------------------------------------|
| Davies et al. [5] | 84 y/o male | Draining sinus tract                | 3 months                   | Incision and drainage, Foley placed into gallbladder for drainage          | Patient biliary system on delayed cholecystogram, successful fistula tract closed after drain removal |
| Cruz et al. [1]  | 81 y/o male | Right subcostal abscess             | 28 days                    | Midline laparotomy, cholecystectomy, external drainage of abscess          | Death from massive pulmonary embolism, POD3                              |
| Malik et al. [12] | 76 y/o female | Abdominal wall abscess              | N/A                        | Laparoscopic cholecystectomy                                              | Recovery and discharge                                                   |
| Pezzilli et al. [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 et al. [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 et al. [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 et al. [15] | 85 y/o female | RUQ abscess and drainage            | 3 months                   | ERCP with stone removal and sphincterotomy                                | Progressive fistula healing                                              |
| Khan et al. [10]  | 76 y/o male | RUQ pain, swelling                  | N/A                        | Open cholecystectomy                                                     | N/A                                                                      |
| Ozdemir et al. [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 et al. [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 et al. [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.

REFERENCES

1. Jr RJC, Nahas J, Figueiredo LF (2006) Spontaneous cholecystocutaneous fistula: a rare complication of gallbladder disease. Sao Paulo Med J 124: 234-236. [Crossref]
2. Vasanth A, Siddiqui A, O’Donnell K (2004) Spontaneous cholecystocutaneous fistula. South Med J 97: 183-185. [Crossref]
3. Nicholson T, Born MW, Garber E (1999) Spontaneous cholecystocutaneous fistula presenting in the gluteal region. J Clin Gastroenterol 28: 276-277. [Crossref]
4. Batty L, Freeman L, Dubrava Z (2011) A spontaneous cholecystocutaneous fistula. ANZ J Surg 81: 847. [Crossref]
5. Davies MG, Tadros E, Gaine S, McEntee GP, Gorey TF et al. (1989) Combined internal and external biliary fistulae treated by percutaneous cholecystolithotomy. Br J Surg 76: 1258. [Crossref]
6. Hoffman L, Beaton H, Wanz G (1982) Spontaneous cholecystocutaneous fistula: a complication of neglected biliary tract disease. J Am Geriatr Soc 30: 632-634. [Crossref]
7. Kapoor Y, Singh G, Khokhar M (2013) Spontaneous cholecystocutaneous fistula—not an old time story. Indian J Surg 75: 188-191. [Crossref]
8. Ozdemir Y, Yucel E, Sucullu I, Filiz I, Gulec B et al. (2012) Spontaneous cholecystocutaneous fistula as a rare complication of gallstones. Brit J Lek Listy 113: 445-447. [Crossref]
9. Yüceyar S, Ertürk S, Karabiçak I, Onur E, Aydoğan F (2005) Spontaneous cholecystocutaneous fistula presenting with an abscess containing multiple gallstones: a case report. Mt Sinai J Med 72: 402-404. [Crossref]
10. Khan A, Rajendran S, Baban C, Murphy M, O’Hanlon D (2011) Spontaneous cholecystocutaneous fistula. BMJ Case Rep 2011: bcr0520114176. [Crossref]
11. Kumar SS (1998) Laparoscopic management of a cholecystocutaneous abscess. Am Surg 64: 1192-1194. [Crossref]
12. Malik AH, Nadeem M, Ockrim J (2007) Complete laparoscopic management of cholecystocutaneous fistula. Ulster Med J 76: 166-167. [Crossref]
13. Pol MM, Vyas S, Singh P, Rathore YS (2019) Spontaneous cholecystocutaneous fistula: empirically treated for a missed diagnosis, managed by laparoscopy. BMJ Case Rep 12: e228138. [Crossref]
14. Pezzilli R, Barakat B, Corinaldesi R, Cavazza M (2010) Spontaneous Cholecystocutaneous Fistula. Case Rep Gastroenterol 4: 356-360. [Crossref]
15. Sayed L, Sangal S, Finch G (2010) Spontaneous cholecystocutaneous fistula: a rare presentation of gallstones. J Surg Case Rep 2010: 5. [Crossref]