



Spontaneous cholecystocutaneous fistula secondary to xanthogranulomatous cholecystitis presenting as a right upper quadrant abscess

Brennan Bowker MHS, PA-C, CPAAPA^{1,2} Alyssa Tuozzoli, MPAS, PA-C,¹ Jenna Brophy, LAT, ATC, PA-S,² Casey Fusco, LAT, ATC, PA-S,² Nicole Modell, R.T (R)CT, PA-S,² Alec Williams, PA-S,² Tyler J. Jones, MD,^{1,3} Dirk C. Johnson, MD^{1,3}

¹Yale New Haven Hospital, Department of Surgery, New Haven, CT
²Quinnipiac University, Department of Physician Assistant Studies, Hamden, CT
³Yale School of Medicine, Department of Surgery, New Haven, CT



Quinnipiac
School of Health Sciences
Department of Physician Assistant Studies

Introduction

- Cholecystocutaneous fistula (CCF) is an abnormal connection between the gallbladder and the skin of the abdomen.¹
- CCF is an extremely rare complication of untreated biliary disease with fewer than 100 reported cases; most commonly in elderly females.²
- Etiology derives from chronic obstruction of the cystic duct which increases intraluminal gallbladder pressures leading to necrosis and perforation.³
- Typical presentation includes local features of fluctuant mass, pain, swelling, and discharge, in addition to systemic symptoms.²
- Differential diagnoses include abdominal wall abscess, malignancy, herniation of bowel, tuberculoma, and rib osteomyelitis.^{4,5}
- Xanthogranulomatous cholecystitis (XGC) occurs due to extravasation of bile into intramural tissues leading to reactive inflammatory changes.⁶
- Chronic inflammatory changes lead to proliferative fibrosis of the gallbladder wall resulting in XGC.⁶
- XGC is present in less than 1.5% of specimens and is two times more common in males.⁷

Figure 1: Skin Lesion



Case Description

- A 41-year-old male with a history of hepatitis C presented to the emergency department (ED) for evaluation of a "lump" in his right upper quadrant (RUQ) which had been progressively increasing in size and pain intensity over several years.
- Prior to the formation of the mass, he does not report abdominal pain or history of gallbladder disease.
- He had received care in several states prior to settling in Connecticut

Medical History

PMH: Hepatitis C,
Medications: None
Surgical Hx: Denies
Social Hx: Former tobacco smoker, rare crystal meth, recently moved to Connecticut

Physical Examination

Gen: Alert, NAD
Abd: Soft, tenderness over erythematous, fluctuant mass (figure 1). Unable to assess Murphy's sign
Skin: anicteric
Eyes: anicteric

Laboratory Analysis

WBC: 10.5 mm³
Tbili: 0.2 mg/dL
AST: 39 U/L
ALT: 15 U/L
AlkPhos: 105 U/L
Lipase: 41 U/L

Hospital Course

- CT scan of the abdomen and pelvis was obtained (figure 2)
 - Focal lesion in the right upper quadrant subcutaneous fat, suggestive of abscess.
 - Circumferential gallbladder wall thickening with embedded calculi, and pericholecystic fat stranding, mostly related to overlying anterior abdominal wall abscess over acute cholecystitis or other process.
- Patient underwent incision and drainage of the lesion
 - Moderate amount of frank purulence was evacuated and sent for culture
- He was discharged to home with outpatient follow up
- During outpatient follow-up, the patient reported improved incisional pain but the when the dressing was changed, stones were noted on the dressing (figure 3) and emanating from an external fistulous tract (figure 4)

Figure 2: CT Scan



Figure 3: Stones on Dressing



Figure 4: Fistula Opening



Surgical Intervention

- The patient was taken to the operating room for laparoscopic cholecystectomy
- A fistula tract was encountered between the gallbladder and the skin
- A large stone was delivered through the fistulous tract in the skin
- RUQ inflammation was tenacious and necessitated conversion to open
- Pathology returned with chronic XGC and cholelithiasis with prominent mural fibrosis

Discussion

- Cholecystocutaneous fistulae are rarely reported most likely due to the availability of antibiotics and prompt surgical treatment of symptomatic biliary disease.^{5,8}
- The formation of CCF has been associated with XGC due to fluid extraversion, chronic inflammation, and fibrosis of the gallbladder wall.²
- Additionally, XGC is associated with a higher rate of complication including perforation and internal fistulization.⁹
- CT imaging with fistulogram is most specific for CCF diagnosis and suggested for pre-operative planning.^{2,10}
- Given its rarity, there is no standard for the management of CCF; however, standard practice is drainage of any abdominal wall abscess followed by cholecystectomy.^{3,10}
- Definitive treatment of both XGC with CCF is cholecystectomy.^{2,6}
- About 50% of patients undergoing laparoscopic approach will require open surgical conversion.⁶

Conclusion

- Cholecystocutaneous fistulae and xanthogranulomatous cholecystitis are both rare entities affecting the biliary tree. Although uncommon, providers must be aware of these pathologies as both require prompt intervention to minimize morbidity and mortality.

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