Chronic Cholecystitis with Cholecystodoudenal Fistula and Ectopic Galstone Resulting in Bouveret Syndrome, a Case Report

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ABSTRACT

Chronic cholecystitis is caused by prolonged gallbladder inflammation, which is most often associated with cholelithiasis, causing intermittent obstruction of the cystic duct. Cholelithiasis is associated with the condition in 95% of cases and is the most common finding besides non-specific gallbladder wall thickening. Chronic cholecystitis can occur with minimal symptoms such as epigastric abdominal pain and bloating predominantly after meals. We present a complex case demonstrating complications that can arise from chronic gallbladder inflammation, including cholecystodoudenal fistula with an ectopic cholelith in the duodenum resulting in Bouveret Syndrome. The patient was initially started on medical therapy but ultimately required surgery to correct the problem. This case focuses on the imaging findings as well as therapeutic interventions available for such patients with complicated chronic cholecystitis.

INTRODUCTION

It is estimated that gallstones are present in 10 to 15% of adult populations which equates to 20 to 25 million people in the United States1. This statistic is significant given the propensity for acute and chronic gallbladder disease to be related to the presence of gallstones. The presence of gallstones can intermittently or completely block the cystic duct resulting in acute cholecystitis. Chronic cholecystitis is thought to occur from chronic irritation of the gallbladder lining from interaction with choleliths or recurring infection2. Given that 95% of chronic cholecystitis is related to 3 choliths, it is important to know demographics with increased propensity for stones such as middle-aged females, estrogen supplementation, obesity, and drastic weight loss.[1,2]

Complications of chronic cholecystitis included fistula/abscess formation, malignant transformation, and perforation. This case further demonstrates the sequela of cholecystodoudenal fistula that led to Bouveret Syndrome. Bouveret Syndrome occurs when an ectopic gallstone lodges in the pylorus or proximal duodenum resulting in gastric outlet obstruction.[3]

Patient Information

The patient is an 87-year-old female who presented with 1 day of right-sided abdominal pain. The pain was rated an 8 out of 10 and was described as “crampy and intermittent” without any radiation throughout the abdomen. Before her pain, she was reportedly having nausea over the past 2 to 4 weeks that did not improve with Pepto-Bismol or Zofran. She also had associated constipation with no bowel movement for 4 to 5 days. There is no pertinent family or genetic history. Past medical history includes coronary artery disease, diabetes mellitus type 2, hypertension, and congestive heart failure. The only reported surgical history was cardiac stent placement, unknown knee/thyroid surgery, and pacemaker placement.

Clinical Findings

A physical exam in the emergency department noted epigastric tenderness to palpation although soft with no guarding, rebound, or peritoneal signs. The patient was found to have normal bowel sounds with no abnormalities. No significant findings were noted on the rest of the initial

examination. There was no progression or significant changes in her physical exam between the time of presentation and the initial surgical intervention.

Diagnostic Assessment

Initial abnormal labs included: WBC 8.5, hemoglobin 10.9, platelets 211, sodium 134, potassium 3.9, chloride 95, bicarb 29, BUN 16, creatinine 0.92, glucose 119, total bilirubin 0.5, AST 14, ALT less than 9, alkaline phosphatase 84, lipase 26.

A CT abdomen and pelvis with contrast was ordered in the emergency department. The scan demonstrated a distended gallbladder with cholecystoenteric fistula, a large peripherally calcified gallstone noted in the first segment of the duodenum, pneumobilia, and potential hepatic abscesses (Figure 1). The gallstone being located in the first segment of the duodenum resulted in dilation of the stomach and gastric outlet obstruction (Bouveret Syndrome).

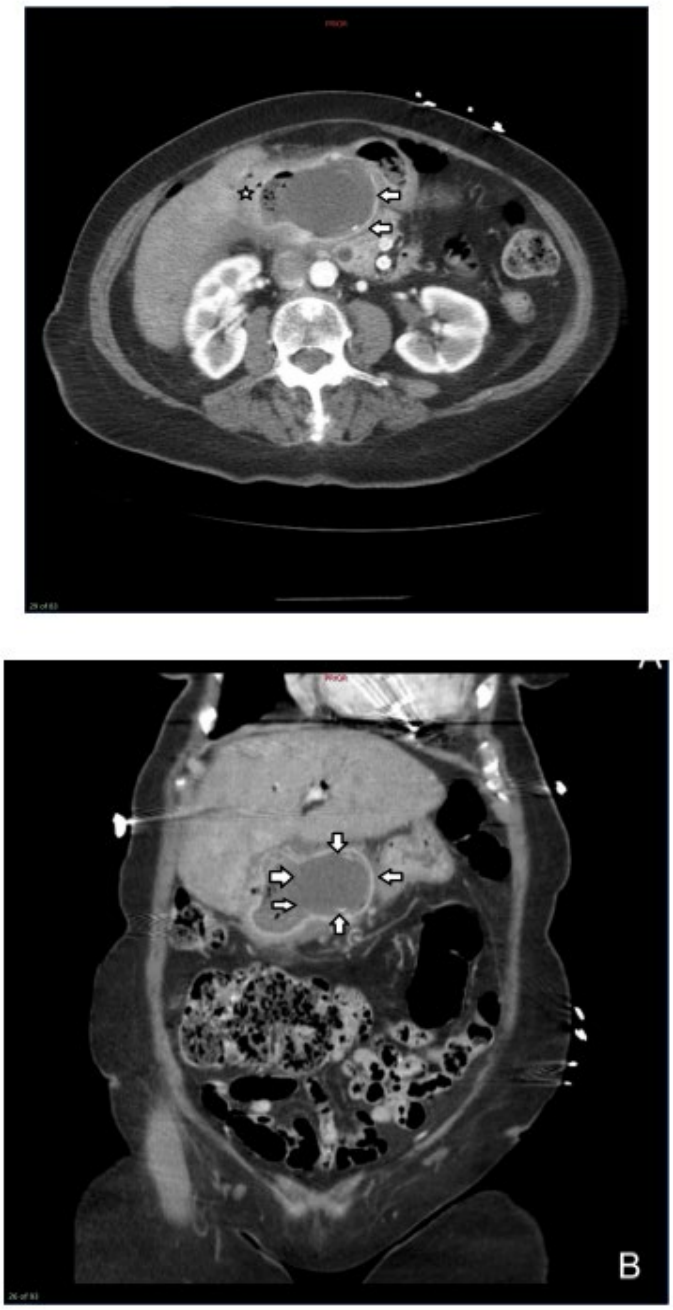

FIGURE 1a and 1b: Axial (a) and coronal (b) Images demonstrating the peripherally calcified and centrally hypodense gallstone in the proximal duodenum indicated with arrows. The cholecystodoudenal fistula is indicated with a star.

Therapeutic Intervention

General surgery was consulted with recommendations for the patient to have nothing by mouth (NPO), place a nasogastric tube set to low-intermittent suction, and Zosyn IV antibiotic for gram-negative enteric coverage. The patient declined to have the nasogastric tube placed due to discomfort.

On the second day of hospitalization, general surgery performed an esophagogastroduodenoscopy (EGD) with attempted morcellation and mobilization of the large duodenal gallstone. The procedure was unsuccessful as the 5cm stone was too large for many of the instruments to encompass or manipulate. The EGD was terminated to speak about additional treatment options, such as laser ablation or laparotomy. The patient and her family declined surgery at the time as she was a high risk due to her age. A second EGD was performed to use laser ablation to further morcellate or dislodge the large duodenal gallstone. There was limited visualization of the gallstone while attempting to use the working tools. The procedure was terminated due to concern for the potential of damaging surrounding structures outside the partial field of view with the laser.

A diagnostic laparotomy was then performed for definitive stone removal. During surgery, the stomach was easily located with the cholelith palpated in the first segment of the duodenum. The duodenum was unable to be separated from the surrounding mesentery due to all of the tissue inflammation and phlegmon. The decision was made to perform an anterior gastrostomy to remove the duodenal stone via the stomach. The stone was successfully removed, however, the fistula tract was unable to be ligated and repaired due to the significant amount of inflammation (Figure 2). The patient tolerated the surgery well although further staged definitive surgery was declined due to age and significant co-morbidities. Given the fistula tract was still patent, the patient was placed on Ursodiol 300mg twice daily to dissolve other choleliths and prevent future formation of them.

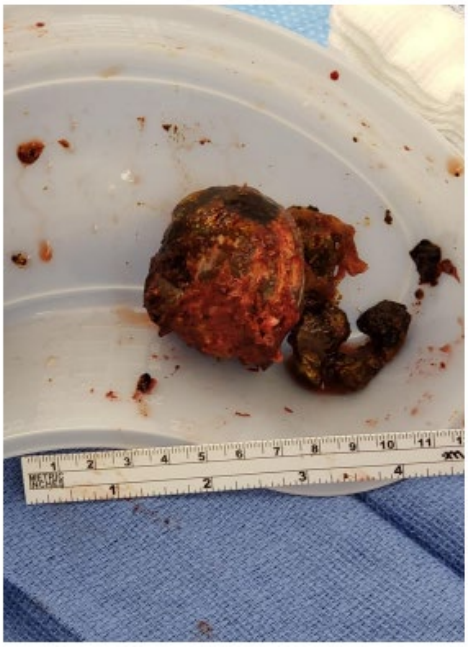

FIGURE. 2: Gross surgical specimen of the peripherally calcified gallstone that had passed through the cholecystodoudenal fistula into the proximal doudenum.

Follow-up and Outcomes

The patient tolerated surgery well and was discharged with home-health after a short observation period. She was seen in the outpatient general surgery clinic where there were complaints about the oral medication Urosodiol being expensive and resulting in gastrointestinal upset. Withholding the medication was trialed without any complications and was discontinued permanently given the low likelihood of such a significant stone recurring. The patient presented to additional outpatient appointments with no new complaints. A follow-up surgery for fistula removal and cholecystectomy was not performed secondary to the patient's age and co-morbidities. She eventually passed away from unrelated cardiac events.

Discussion

Acute cholecystitis is an acute inflammation of the gallbladder that affects 120,000 Americans yearly. Etiology typically arises from persistent gallbladder outlet obstruction leading to bile stasis and eventual mechanical, chemical, or infectious irritation of the gallbladder. Most cases arise from cholelithiasis, however, 10% of cases are precipitated by acalculous obstruction.[4] Acute cholecystitis can be prolonged with ongoing inflammation resulting in chronic cholecystitis and waxing/waning of symptoms. This case is an example of chronic cholecystitis leading to a fistula formation between the gallbladder and the duodenum, with the concomitant 9

passage of a gallstone into the proximal duodenum resulting in gastric outlet obstruction (Bouveret Syndrome).

Bouveret Syndrome is rare owing to the fact that 0.5-5% of gallstones develop fistulas and the majority simply pass through the digestive tract without incident. Approximately 2-3% of gallstone-related bowel obstructions are caused by Bouveret Syndrome re-emphasizing the rarity.[3,5] Given the infrequent diagnosis, there are no evidence-based treatment guidelines but instead progressively more invasive treatment options. Initial attempts to treat the obstructing gallstone are made from esophagogastroduodenoscopy (EGD) such as endoscopic nets, shockwave lithotripsy, laser, and mechanical fragmentation.[6] Only approximately 66% of gallstones are visible via EGD with a chance of being unsuccessful or advancing the stone/fragments resulting in a more distal obstruction.[6, 7]

Imaging is critical in the diagnosis and management of cholecystitis and its' complications. Treatment modalities differ in that acute cholecystitis requires emergent surgical removal of the gallbladder, whereas chronic cholecystitis treatment is elective removal.[8] Abdominal ultrasound serves as the initial imaging of choice in suspected cases of cholecystitis, however, it has limited value in evaluating complications of acute cholecystitis.[9] Computed tomography (CT) can be used to demonstrate surrounding complications of acute and chronic cholecystitis with increased sensitivity. Imaging findings with a high predictive value of acute cholecystitis include increased wall enhancement, increased wall thickness, mural striation, pericholecystic haziness or fluid, increased adjacent hepatic enhancement, and a focal wall defect with pericholecystic abscess. [10] Patients with renal injury or radiation concerns may undergo magnetic resonance imaging (MRI) to discern between acute and chronic cholecystitis. Hepatobiliary scintigraphy or HIDA scan uses a tracer to visualize delayed biliary transit time. The percentage of gallbladder emptying can be calculated to find a dyskinetic gallbladder. HIDA scans can confirm chronic cholecystitis but are unable to show complications of cholecystitis.

This study is limited primarily due to the fact that the patient was deceased at the time of its development which disallowed for informed consent or primary follow-up. In addition, it is a retrospective description of events with limited room to expand on information as it is being taken from clinician and nursing documentation.

Conclusion

Our case demonstrated a classic presentation of chronic cholecystitis that led to a heterotopic gallstone within the duodenum via a cholecystoenteric fistula. The gallstone became impacted in the duodenum and resulted in gastric outlet obstruction (Boiveret Syndrome). This patient's presentation was confounded by advanced age with multiple comorbidities requiring multiple treatment modalities. Definitive therapy was unable to be surgically performed secondary to inflammation and the patient being a poor surgical candidate on follow-up. Additional non-surgical treatments were used such as urosodiol with no further complications reported by the

patient for at least 2 years post-hospitalization. Multiple radiology modalities were available to provide a prompt diagnosis allowing early medical/surgical treatment to remove the gallstone.

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