Running Into a Wall: A Case of Bouveret Syndrome

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Introduction

- Bouveret Syndrome is characterized by gastric outlet or duodenal obstruction secondary to stone impaction in the pylorus or duodenum in the setting of cholecystogastric or more often cholecystoduodenal fistula.
- It is a very rare complication of cholethiasis, seen in 0.3-0.5% of gallstone cases.[1]
- Risk factors include history of cholethiasis, stones greater than 2-8cm, female gender, and age greater than 60 years.
- Due to its disproportionate presentation in the elderly, complexity, and non-specific presentation, in combination with no set guidance in terms of workup and management, it has high morbidity with mortality rate estimates ranging from 12-30%.
- It often presents after an episode of cholecystitis (more rarely malignancy), with fistula developing secondary to gallbladder inflammation and adhesion to the GI tract in combination with gallstone pressure on the gallbladder wall. The development of symptoms may depend on factors including gastrointestinal anatomical distortion and the size of the stone.[1]

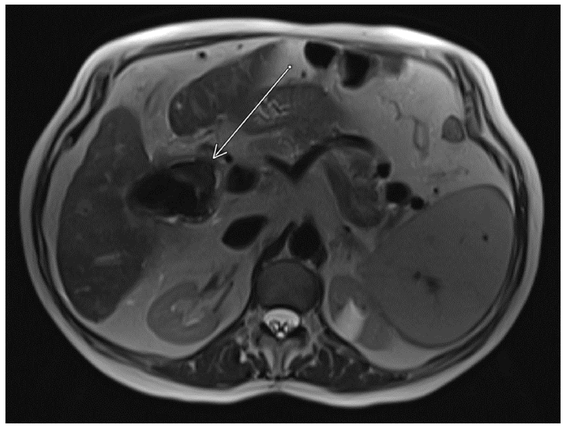

Figure 1. MRI Demonstrating Indistinct Wall Between Gallbladder Neck and Common Bile Duct with Pneumobilia

Case Presentation

A 75-year-old female with a history of decompensated cryptogenic cirrhosis being followed up in our transplant hepatology clinic underwent an MRI as part of routine screening for hepatocellular carcinoma. Following completion of the MRI, critical findings (Figure 1) were identified and communicated. These findings included inflammatory changes of the gallbladder concerning for acute cholecystitis, pneumobilia, an indistinct wall between the gallbladder neck and common bile duct, and presumed gas in gastrointestinal lumen suspicious for fistula to the descending duodenum. The patient was contacted and reported recurrent post-prandial episodes of nausea and emesis that occurred several times the week before the MRI. The patient was advised to present to the ED, whereupon she was admitted to the hospital with labs demonstrating mildly elevated liver enzymes, as well as a CT abdomen/pelvis (Figure 2) disclosing a fistula between the gallbladder and duodenum. Her physical exam was unremarkable and vital signs were within normal limits. She was started empirically on empiric antibiotics due to concern for acute cholecystitis. After discussion with the transplant surgeon, the decision was made to perform a diagnostic esophagogastroduodenoscopy (EGD) to rule out an underlying mass, which demonstrated a large non-obstructive gallstone in the duodenal bulb (Figure 3). No therapeutic intervention was performed during EGD due to resolving symptoms as well as lack of the presence of expertise in stone removal techniques in our center. A surgical intervention was deemed not to be appropriate at that time due to high risk in the setting of decompensated cirrhosis. The patient was discharged on a 14-day course of antibiotics and remained asymptomatic. A liver transplant evaluation was initiated to potentially allay the surgical risk. As of writing, the decision has been made to pursue TIPS placement, not only to optimize surgical risk for planned cholecystectomy but also to prevent worsening of her varices, ascites, and renal function.

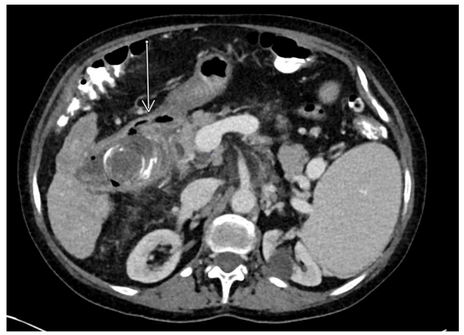

Figure 2. CT Demonstrating Fistulization of Gallbladder Neck Anteriorly into Duodenum

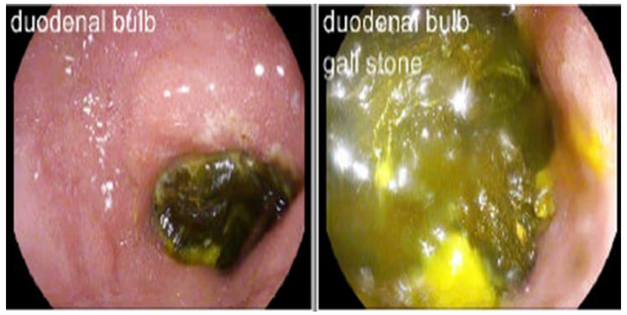

Figure 3. EGD Demonstrating Gallstone in Duodenal Bulb; Note Fistula on Screen Right

Discussion: Diagnosis

The diagnosis of Bouveret Syndrome can be challenging due to its non-specific symptoms and rarity. MRI or CT are helpful, and can disclose Rigler's triad (dilated stomach, pneumobilia, and a radiopaque shadow consistent with an enteric gallstone). Imaging can also identify fistula,[2] as seen in our patient. EGD is very often performed after the initial suspicion is present. It is used for both diagnostic and, especially given the surgically high-risk patient population that this syndrome often presents with, therapeutic purposes.

Discussion: Management

Options for endoscopic removal include:

- Endoscopic removal with baskets or nets
- Mechanical lithotripsy with stone fragmentation via crushing
- Specialized procedures including electrohydraulic lithotripsy and laser lithotripsy, both of which involve energy-focused stone fragmentation and removal.[2]
- Extracorporeal shockwave lithotripsy to induce fragmentation, followed by EGD to remove the gallstone pieces.[2]

EGD can also be used as an adjunct to surgery, if needed.[2] Surgical options are considered in the setting of lack of technical expertise for EGD-driven removal, or EGD failure. Surgical approaches include:

- Open duodenotomy, pyloromyotomy, or gastrotomy
- Enterolithotomy under laparoscopy

Open approaches have a higher morbidity and mortality compared to laparoscopic approaches.[2] Cholecystectomy with fistula repair in these cases remains controversial. Often, as in the patient in this case, Bouveret syndrome patients are elderly with comorbidities and as such poor surgical candidates; in many of these patients cholecystectomy is commonly not performed due to significant risk.

- However, this does leave these patients at risk for recurrent gallstone episodes, as well as with an increased risk of malignancy due to the unremoved fistula.[2]

Conclusion

This rare and interesting case was initially discovered incidentally, via routine HCC MRI screening in a cirrhotic patient. The postprandial symptoms she describes prior to MRI likely represent intermittent obstructions, relieved by transit of the gallstone.

Due the dearth of data regarding outcomes in this rare condition, decisions regarding management must be tailored to the individual patient. In our patient's case, her underlying decompensated cirrhosis represented a significant hurdle in terms of surgical risk, and ultimately necessitated potential cholecystectomy being deferred pending further management of her cirrhosis either through transplant, or as is currently the plan, TIPS. This represents one of the common challenges faced in managing Bouveret Syndrome - significant patient comorbidity, which makes the creation of definitive guidelines difficult and tends to push management towards an individually-tailored approach.

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