CASE REPORT

Gallstone ileus of the duodenum: an unexpected presentation of Bouveret's syndrome

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SUMMARY

This report describes a patient who presented with a large gallstone obstructing the duodenal bulb, with the chief complaint of acute on chronic abdominal pain. Classically, this is known as Bouveret's syndrome or a gallstone ileus of the duodenum. Our patient's current health status presented a challenge, with the presence of several comorbidities, particularly a large abdominal aortic aneurysm. We chose an open procedure for this reason. The stone was removed through a laparotomy, and the cholecystoduodenal fistula that the stone used to pass into the small bowel was repaired. With our patient's future medical needs in mind, only the necessary interventions were performed to regain functionality of the bowel. This came in the form of a diverting gastrojejunostomy, with a distal jejunostomy and feeding tube inserted. The patient tolerated the procedure well, only remaining intubated postoperative due to her chronic obstructive pulmonary disease (COPD). She achieved a complete recovery and transitioned home.

under the regular care of a primary care physician. An abdominal CT scan demonstrated both a large, non-enhancing 3 cm mass within the lumen of the duodenum (figures 1–3) and gangrenous cholecystitis. In addition, the CT scan also showed an abdominal aortic aneurysm (AAA) measuring 5 cm × 5 cm. Although the patient's CT showed worrisome findings, the patient was stable and reported only moderate discomfort. An elevated white count was the only abnormality within her panel of laboratories. As the patency of the common bile duct (CBD) was unknown, and her bilirubin levels were not elevated, it was hypothesised that a fistula was allowing drainage of bile from the gallbladder. This fistula would also provide an explanation for the gallstone passing into the duodenum. Magnetic resonance cholangiopancreatography (MRCP) was performed to assess the CBD, which demonstrated several non-obstructing stones within the CBD. As the largest gallstone was the most acute pathology, it was the focus of our intervention.

BACKGROUND

Incidence reports demonstrate that 0.3%–4% of cholecystitis results in the complication of a gallstone ileus. Furthermore, approximately 1%–3% of cases gallstone ileus present with the presence of a gallstone ileus at the duodenum, making Bouveret's syndrome a rare condition to diagnose.^{1–3} This finding was first described in 1770 by Beaussier and first reported by Leon Bouveret in 1896, where a gallstone was found to be impacted at the duodenal bulb, causing a gastric outlet obstruction (GOO).^{4–7} Our patient presents with this exact finding, demonstrated by both CT and MRI. In addition to the rarity of the condition, there have been numerous reported techniques for addressing the removal of the gallstone. As there is no consensus regarding which is superior, it is important that each case is described. Our method represents a straightforward approach in surgical management that resulted in a successful removal of the stone and repair of the duodenum.

CASE PRESENTATION

This is the case of a 69-year-old woman, who presented through the emergency department with an episode of acute on chronic abdominal pain. Her medical history was significant only for chronic obstructive pulmonary disease (COPD) due to a history of smoking. The patient was not

TREATMENT

Our surgical approach used a laparotomy, performed through a midline incision. As the peritoneal cavity was entered, omental adhesions and small abscesses within the omental fat were encountered in the epigastric region overlying the duodenum. Within these adhesions, we encountered the remaining tissue of the gangrenous gallbladder, which was positioned anterior and superiorly to the duodenum. The gallstone was confirmed to be within the duodenal bulb by palpation, located proximal to a cholecystoduodenal fistula, through which the stone migrated. This was taken down, leaving an opening into the small bowel, which was extended longitudinally towards the pylorus, allowing for the removal of the stone. The stone was noted to be bilious in composition and approximately 3 cm in diameter, as described in the radiology report (figure 4). The extended opening was then closed in a transverse, two-layered fashion to prevent stricture of the duodenal lumen. Next, the inflammatory tissue surrounding the gallbladder was dissected to perform a cholecystectomy. The gallbladder was largely obliterated, with only a small portion being discernible as tissue from the organ. As the critical view of safety was impossible to isolate from the surrounding inflammatory tissue, we performed the cholecystectomy in a top-down manner, avoiding damage to any surrounding structures that were obscured due to the above noted pathology.⁸ Considering this, a 45 mm thick endovascular staple

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Figure 1 CT scans demonstrating our patient's duodenal gallstone ileus. A 30.47 mm gallstone is positioned just distal to the pylorus, within the duodenum.

Figure 2 Coronal view of previous scan, giving a better view of the stone's positioning. Additionally, note the enhancement surrounding the duodenum and cystic ductwork, demonstrating the significant inflammation encountered during surgery.

was placed across the neck of the gallbladder, leaving the cystic duct intact, while achieving ligation of the cystic artery. On visual inspection, numerous 2–3 mm bilious calculi were observed in the gallbladder on its removal. As a protective measure, a gastro-jejunostomy (GJ) was created with the fundus of the stomach and the jejunum approximately 15 cm distal to the ligament of Treitz. This was performed using a 75 mm thick tissue stapler, in a side-to-side fashion, without exclusion of the duodenum. Finally, a jejunostomy was performed distally to the GJ, using a Witzel technique, to provide supplemental feeding as the patient recovered postoperation.

Figure 3 T2-weighted MRI of upper abdomen in our patient demonstrating the presence of a large, hypodense mass appearing to be situated distal to the pyloric region within the duodenal lumen. Also of note, the significant inflammation surrounding the biliary ductwork and gallbladder.

Figure 4 The gallstone after removal.

OUTCOME AND FOLLOW-UP

The surgery went without complication, but the patient's postoperative recovery was initially plagued by low oxygen saturations. This was a predicted issue due to her history of smoking and COPD. Because of the concern over her ability to maintain adequate ventilation, the patient remained intubated for several days. The patient was extubated by postoperative day 7 and kept on oxygen through nasal cannula from that point on. Pulmonary supplement feedings were initiated on postoperative day 3. By postoperative day 14, the patient was tolerating solids and liquids by mouth during the day and was supplemented with tube feedings at night. One month postoperatively, the patient progressed significantly, regaining independent ambulation. By this point, the feeding tube was discontinued and is planned to be removed in 3–4 months. This is to ensure proper adherence of the bowel to abdominal wall. The patient continued to recover in the transitional care unit until discharged home approximately 2 months postoperatively. She maintains independence in her day-to-day activities and has declined intervention for repair of her AAA. Her recovery 5 months postoperative has been excellent. She remains asymptomatic regarding biliary and/or abdominal symptoms. We will continue to follow her as she continues her recovery.

DISCUSSION

Patients with Bouveret's syndrome classically present with symptoms of a GOO, such as epigastric abdominal pain, abdominal distention, nausea, vomiting and early satiety. To our surprise, our patient presented solely with an acute worsening of her chronic, colicky abdominal pain. In this patient's case, the CT clearly demonstrated the findings of Rigler's triad: bowel obstruction, pneumobilia and a large ectopic gallstone.⁹ This was in addition to the preoperative suspicion of a fistula between the gallbladder and the duodenum. Bilioenteric fistulas are a common finding in the setting of a gallstone ileus, of which Bouveret's syndrome is a rare variant. Cholecystoduodenal fistulas represent the most common variant of fistula in reported cases of a gallstone ileus, occurring in approximately 60% of the reported cases. As stated previously, Bouveret's syndrome represents approximately 3% of gallstone ileus cases.^{10 11} We find this interesting that even though cholecystoduodenal fistulas are the most common type of fistula present in intestinal obstructions by a gallstone, the obstructing stone only migrates retrograde to duodenal bulb in an extremely small subset of these cases. Specific factors that

contribute to the development of Bouveret's syndrome have yet to be elucidated; however, it is likely that the antegrade peristalsis of the small bowel is protective against the development of Bouveret's syndrome.

For this case, our initial step was to evaluate the best approach for the removal of the ectopic gallstone, followed by our addressing the gangrenous cholecystitis described on imaging. Of interest, was the presence of this finding in the presence of a patent cholecystoduodenal fistula. We believe that this was present because our patient was likely suffering from subclinical cholecystitis for an extended period of time. Once the stone migrated through the fistula into the duodenum, necrosis of the gallbladder had begun. This was unable to resolve in the setting of the extensive inflammation noted in our surgical exploration above. There have been several techniques reported in the literature aimed at treating Bouveret's syndrome, including the traditional open methodology we used. These include endoscopic removal, net extraction, electrohydraulic removal, intracorporeal laser lithotripsy, extracorporeal shock wave lithotripsy or even mechanical lithotripsy.^{4-7 12-18} Due to the large size of the gallstone and the large amount of inflammation surrounding the duodenum and gallbladder, we felt that an open surgical technique would provide the level of access needed to manage the full extent of our patient's pathology. On entering the abdomen, we discovered the gallbladder was necrotic and encased within inflammatory tissue and omental fat, confirming the findings on CT scan. Although the bilioenteric fistula was allowing bile to drain into the duodenum, the gallbladder's position puts it directly in our path to extract the gallstone. This, in addition to the gangrenous state of the gallbladder, led us to the decision to remove the stone, close the fistula and perform a cholecystectomy in a one-step fashion. It has been described in the literature that it is more efficacious to remove the gallstone and then perform a cholecystectomy in a separate two-step sequence; however, given the comorbidities our patient possessed, and the presence of an obliterated gallbladder, we felt that a one-step procedure was necessary for the patient to recover fully.^{19 20} Considering the location of the fistula's entry into the duodenum, a pyloric exclusion (PEX) was initially considered because of the trauma created by the resulting repair of the duodenal mucosa. PEX has been used for the diversion of gastric contents around repaired duodenal trauma; however, it has been demonstrated that its effectiveness may not be statistically significant compared with leaving the pylorus intact, and in this case, forming a diverting GJ with favourable results.²¹ Because the gallstone was distal to the pylorus, neither a pyloromyotomy nor a gastrectomy was necessary in this case. Additionally, the fistula provided an excellent point of entry into the lumen of the duodenum, through which we could extract the stone without further insult to the bowel.

As described above, several non-obstructing stones were present in the CBD seen on MRCP. Initially, an ERCP was considered to break-up the obstructing stone, and remove any other stones from the common duct. We consulted our gastroenterology for discussion on this, and we concluded that this plan was not ideal in our patient for two reasons: the stone formed a complete obstruction at the duodenal bulb, and it was deemed to be a necessary procedure with the stones not obstructing the duct. Furthermore, because of the friability of the gallbladder and biliary ductwork, along with the patient's concomitant AAA, we decided against an intraoperative choledochostomy to remove the stones within the CBD, with subsequent T-tube placement for drainage. As discussed with our patient, our plan is to address these if the patient becomes symptomatic in the future. With mortality rates ranging at approximately 19%–24% for Bouveret's syndrome, we feel

Learning points

- ▶ Patients presenting with the clinical and radiographic evidence of Bouveret's syndrome can be managed surgically through a multitude of approaches.
- ▶ Laparotomy is a viable approach for surgical treatment of Bouveret's syndrome in a broad range of patients.
- ▶ Even in the presence of duodenal trauma, forgoing a pyloric exclusion can result in the successful resolution of an obstruction without increased risk of leakage of gastric contents through the repaired bowel.

the above management was successful, and that the patient response was positive.³

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Contributors DM was the attending physician in charge of the medical and surgical management of the patient's care, in addition to initiating the request to write-up this case. NT was on DM's service as a medical student, assisted in the medical and surgical management of the patient, and conducted the initial literature review and drafting of this case report. KH assisted in the surgical management of the surgery. Each of the authors were extensively involved in the data gathering, editing and the final approval of this case report.

Competing interests None declared.

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