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Bouveret's Syndrome: A Case Report and Review of the Literature

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Abstract

Bouveret syndrome is an exceedingly rare complication of cholelithiasis, characterized by gastric or intestinal obstruction due to the impaction of one or more gallstones in the proximal gastrointestinal tract. We present a case of an elderly woman with multiple comorbidities who presented with gastric outlet obstruction caused by a cholecystogastric fistula. The management and treatment of Bouveret syndrome present a challenge, as endoscopic retrieval is the first-line treatment strategy, but it is rarely successful. Most patients ultimately require surgery as was seen in our case

Keywords

Bouveret syndrome, gastric outlet obstruction, cholecystogastric fistula, and rare complication of cholelithiasis.

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CASE REPORT

Bouveret's Syndrome: A Case Report and Review of the Literature

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Abstract

Bouveret syndrome is an exceedingly rare complication of cholelithiasis, characterized by gastric or intestinal obstruction due to the impaction of one or more gallstones in the proximal gastrointestinal tract. We present a case of an elderly woman with multiple comorbidities who presented with gastric outlet obstruction caused by a cholecystogastric fistula. The management and treatment of Bouveret syndrome present a challenge, as endoscopic retrieval is the first-line treatment strategy, but it is rarely successful. Most patients ultimately require surgery as was seen in our case.

Keywords: Bouveret syndrome, Gastric outlet obstruction, Cholecystogastric fistula, Rare complication of cholelithiasis

1. Introduction

Bouveret's Syndrome is a rare condition where gallstones migrate into the gastric outlet or proximal duodenum, obstructing the bowel through a fistulous tract between the gallbladder and gut wall. It accounts for 1–3% of bowel obstructions caused by migrating gallstones.¹ Gallstones can travel from the biliary tract to the enteric tracts through various fistulas, with the cholecystoduodenal fistula being the most common and the cholecystogastric fistula being the least common, representing only 5% of cases. The terminal ileum is the most frequent site of gallstone lodgment, accounting for 60–70% of all gallstone-related bowel obstructions.²

2. Case presentation

An 84-year-old female with a significant past medical history of coronary artery disease, diastolic heart failure, obstructive sleep apnea, type 2 diabetes mellitus, stage 3 chronic kidney disease, gastroesophageal reflux disease, and hiatal hernia presented with abdominal pain, nausea, and vomiting. The patient complained of epigastric pain radiating

to her shoulders for 3 days, with multiple episodes of bilious vomiting. The patient was hemodynamically stable at the time of admission, however, physical examination was remarkable for diffuse abdominal tenderness and sluggish bowel sounds. Blood workup was significant only for leukocytosis, elevated CRP, and mild hyponatremia. CT abdomen revealed acute cholecystitis with a probable gallbladder-stomach fistula and gallstones as the suspected cause of gastric outlet obstruction (GOO). CT abdomen revealed cholecystitis and acute gastric outlet obstruction with a cholecysto-gastric fistula and impacted gallstones in the stomach. Imipenem was initiated, and an Esophagogastroduodenoscopy (EGD) was attempted at an outside hospital but failed to completely remove the gallstones. Subsequently, the patient was transferred to a tertiary center where she underwent a repeat EGD with a snare-on and attempted gallstone fragmentation. This was unsuccessful due to the size of the gallstone and she developed post-procedural nausea and vomiting and required a nasogastric tube for symptom relief. Eventually, an exploratory laparotomy was performed and small bowel resection with gallstone removal was performed, after which the patient's condition improved.

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3. Discussion

About 2–8% of all emergency department visits are due to acute intestinal obstruction.³ Gallstone ileus makes up only 1–4% of these cases and still rarer is Bouveret syndrome as a cause of gallstone ileus which makes up a mere 1–3% of cases.⁴ Bouveret syndrome derives its name from the French internist Leon Bouveret who reported two cases of gastric outlet obstruction caused by migrating gallstones in 1896 in the journal *Revue de Medicin*.⁵ Since then Bouveret syndrome has remained a rare clinical diagnosis with only 315 cases being reported from 1967 to 2016.⁶ Elderly females make up the most common demographic for this disease with a median age reported as 74 years. The female-to-male ratio for the diagnosis of Bouveret syndrome is reported as 1.9.⁷ The size of the stones seen obstructing the gastric outlet is mostly more than 2.5 cm, smaller stones get extruded from the gastrointestinal tract either in vomitus or feces.⁴

The most common type of connection between the biliary and the enteric tract is the cholecystoduodenal fistula, seen with a frequency of 77–90% followed by the cholecystocholonic fistula and the choledochoduodenal fistulas. Our case of Bouveret syndrome presented with a cholecystogastric fistula i.e. a communication between the gallbladder and the stomach wall. Cholecystogastric fistula is the rarest type of fistula as a cause of Bouveret syndrome, with the first case being reported in 1968.^{8,9}

The diagnosis of Bouveret syndrome can often be a challenge as the clinical presentation can be nonspecific and has been reported in literature ranging from acute pancreatitis to Boerhaave syndrome.^{10,11} The Rigler's triad of pneumobilia, intestinal obstruction, and presence of gallstones in the intestinal lumen has been regarded as pathognomic for the diagnosis of Bouveret syndrome but is classically seen in only 30–35% of cases on conventional x-rays.¹² CT scan remains the test of choice when it comes to diagnosing Bouveret syndrome. It has been reported to have a sensitivity of 93% and a specificity of 100%.¹³ This was the diagnostic tool used in our case as well and was successful in detecting the fistulous tract. Endoscopy has also been used as a diagnostic tool for visualizing the gallstone and has successfully been able to spot the stone in 69% of the cases.¹⁴ Endoscopic retrieval is almost always attempted as it is relatively noninvasive when compared to laparoscopy or laparotomy but unfortunately, the success rate falls to less than 10%.¹⁵ This low rate of success can be attributed to a number of factors, the most important being the large size of

the stone which can obstruct the esophagus if an attempt is made to remove it whole. Attempts to fracture the stone can result in distal gallstone ileus and aggressive use of lasers and fragmentation devices carry a risk of perforating or damaging the intestinal wall.⁵ In our patient endoscopy failed to extract the stone on two separate occasions and we ultimately had to resort to surgery for complete treatment. Cholecystectomy and fistula repair along with duodenotomy as a single operative procedure has shown a higher mortality rate of 20–30% compared to duodenotomy alone which has a 12% mortality.¹⁵ The benefit of cholecystectomy and fistula repair as a second elective operative procedure is debatable as the usual patients who suffer from Bouveret syndrome are elderly and have many underlying comorbidities. The advantage of elective cholecystectomy to prevent the risk of recurrence of gallstones often pales in comparison to the multiple risks of another surgical procedure under anesthesia. Cholecystectomy, however, is of benefit and is recommended if the patient is younger or if there is a risk of gallbladder malignancy.¹³

4. Conclusion

Bouveret syndrome remains a rare clinical entity and requires a significant degree of suspicion in order to make a timely diagnosis. If caught in a timely fashion many patients survive this disease despite being elderly and having a notable past medical history like the patient described in our case. No single treatment modality guarantees ultimate success and the management strategies differ from case to case based on the condition of the patient. It is seen however that laparotomy was needed in a majority of cases as a last resort and proved to be lifesaving for most of the patients. Despite the advances in endoscopic techniques, this is one diagnosis that still often requires resorting back to the basics.

Conflict of interest

The authors declare there are no conflicts of interest.

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