Gastric Outlet Obstruction arising from Adhesions Secondary to Chronic Calculous Cholecystitis with Cholecystoduodenal Fistula Formation in an Immunocompetent Male: A Case Report

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Abstract

Background: Gastric outlet obstruction (GOO) results from intrinsic and extrinsic obstruction of the pyloric channel or the duodenum. Here we present a rare case of GOO attributed to dense adhesions between the gallbladder and duodenum secondary to chronic cholecystitis with choledococystoduodenal fistula formation. Previous reports identified elderly females with comorbidities as a predisposing factor; however, our patient was an immunocompetent adult male.

Case: A 43-year-old male with no comorbidities consulted for recurrent epigastric pain, vomiting and weight loss. On contrast enhanced abdominal CT scan, a lamellated cholelithiasis with pneumobilia and an irregular thickening at the proximal duodenum with subsequent GOO was identified. A choledococystoduodenal fistula was considered. Exploratory laparotomy revealed extensive fibrosis and cholecystitis with dense adhesions to surrounding structures. Dissection revealed a gallstone impacted in and adherent to the wall of the gallbladder and a fistula opening into the duodenum. However, there was no definite evidence of impacted gallstone in the duodenum. The dense adhesions secondary to chronic cholecystitis caused duodenal narrowing and subsequent GOO. He eventually underwent antrectomy, pancreatic sparing, total duodenectomy, cholecystectomy, with loop gastrojejunostomy, cholecystojejunostomy and pancreaticojejunostomy. Biopsy specimens taken were negative for malignancy. He was discharged subsequently. However, he was readmitted after five months due to acute abdomen secondary to small bowel rupture, likely from a marginal ulcer.

Summary: This case highlights that preoperative and intraoperative differential diagnosis of GOO is a challenge. Chronic calculous cholecystitis through severe inflammation can present as a rare cause of GOO. Optimal treatment plan should take into consideration the underlying etiology of the GOO.

Keywords: Gastric Outlet Obstruction, Cholecystitis, Cholecystoduodenal Fistula, Complications

Introduction:

Gastric outlet obstruction (GOO) is a clinical syndrome characterized by epigastric abdominal pain and postprandial vomiting due to mechanical obstruction. It may result both from intrinsic and extrinsic obstruction of the pyloric channel or the duodenum. Intrinsic processes such as those of peptic ulcer disease, *Crohn's disease* or caustic ingestion may cause inflammation, edema, stricture and chronic scarring of the pyloric channel or the duodenum. *Bouveret* syndrome is a rare variant of gallstone ileus occurring mainly in elderly females with multiple comorbidities. It is characterized by migration of a gallstone into the intestinal lumen via a bilio-enteric fistula causing GOO.

Meanwhile, extrinsic inflammatory processes in the pyloric region can be attributed to acute pancreatitis or pancreatic fluid collections causing significant edema and inflammation around the area causing compression and displacement of structures. Malignancy can also cause obstruction via local invasion and/or mass effect.[1] Pancreatic adenocarcinoma with extension to the duodenum or stomach is a common cause of GOO. Gastric lymphoma, proximal duodenal neoplasms and advanced cholangiocarcinoma as well can cause GOO.[2] The precise incidence of GOO is unknown and has

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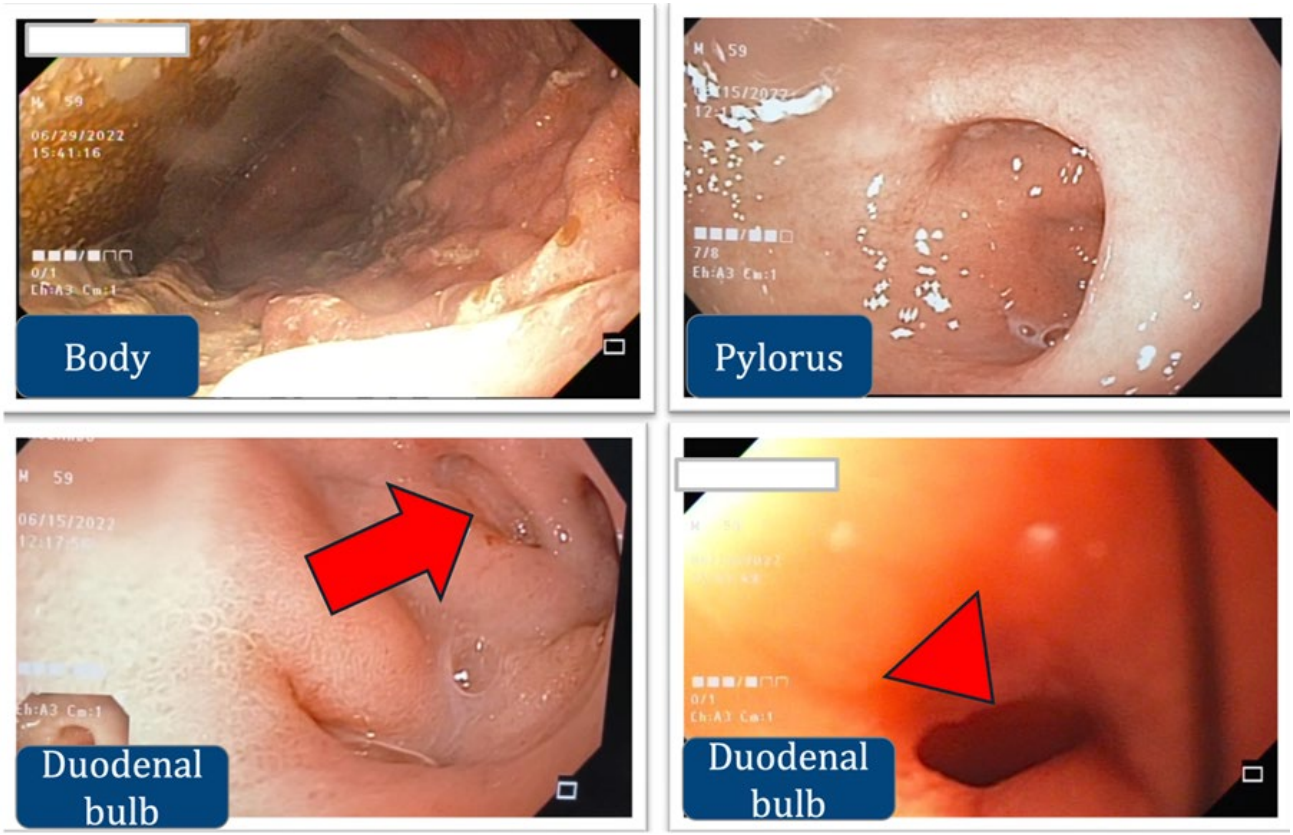

Figure 1. Endoscopic findings. (Body) Gastric body with retained food particles (Pylorus) Pyloroduodenal area (Duodenal Bulb) Duodenal diverticulosis (Duodenal Bulb) Luminal narrowing going into the 2nd portion of duodenum.

changed over the past decades due to effective treatment of peptic ulcer disease which was the leading cause of GOO previously. Currently, approximately 50-80% of GOO is now attributed to malignancy.[1,3,4] Few case reports have also attributed GOO to acute cholecystitis in elderly females with multiple comorbidities and underlying malignancy.[1,5]

Here we present a rare case of GOO attributed to dense adhesions between the gallbladder and duodenum secondary to chronic cholecystitis with subsequent choledochoduodenal fistula formation in an adult immunocompetent male. The exact incidence of cholecystitis causing GOO is unknown. Few case reports identified its occurrence mainly in elderly females with multiple comorbidities.[1] None have mentioned it in immunocompetent patients.

Case Presentation

This is a case of a 43-year-old male retired policeman who consulted due to a recurrent history of burning epigastric pain noted one year prior to consult. He is not known to be hypertensive, diabetic, nor asthmatic. He denies illicit drug use nor is he a smoker or alcoholic beverage drinker. He has no history of recurrent use of non-steroidal anti- inflammatory drugs nor antiplatelet medication and has no previous abdominal surgeries.

A brief review of the patient's history revealed intermittent burning epigastric pain with nausea and intermittent vomiting one year prior to consultation. CT scan of the whole abdomen showed mild thickening at the pyloric region. An upper gastrointestinal endoscopy showed a clean based ulcer at the pyloric area. Test result for *Helicobacter pylori* was unrecalled, and the patient was given Pantoprazole 40 mg daily for 3 months with good compliance and with relief of symptoms. Three months prior to admission, the patient had recurrence of symptoms and this time there was associated anorexia and a 5 kg weight loss. An upper abdominal ultrasound showed a gallbladder polyp with a 1.6 cm gallstone associated with reactive wall thickening of the gallbladder wall. The patient was advised surgery, however he refused. The persistence of symptoms along with postprandial vomiting, bloating, further weight loss (approximately 10 kgs) and early satiety prompted consultation at our institution.

On physical examination, he was underweight with a body mass index of 17. The abdomen was scaphoid with normoactive bowel sounds, tympanitic with no tenderness nor succussion splash or presence of Murphy's sign. Laboratory test results revealed mild normocytic, normochromic anemia (Hgb: 11.6, Hct: 34.6, MCV: 88.5 and MCH: 29.7) along with pre-renal acute

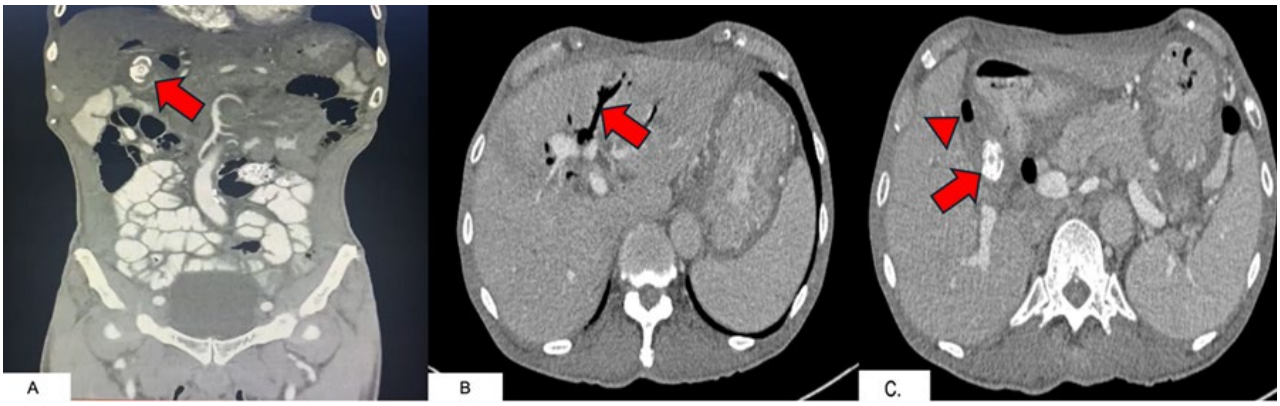

Figure 2. Contrast enhanced CT scan of the whole abdomen. (A) Lamellated cholelithiasis 1.9 x 1.7 cm in the gallbladder (B) Axial view at the superior hepatic level with the arrow pointing to the presence of pneumobilia (C) Axial view of the inferior liver at the gallbladder fossa level with the gallbladder walls thickened and indistinct. The arrowhead points to a pocket of air within the gallbladder fundus. The arrow indicates the lamellated cholelithiasis presumably at the gallbladder body. Loss of the normal intervening fat plane between the gallbladder and duodenum.

kidney injury (Creatinine: 1.3 mmol/li, EGFR: 70 ml/min and BUN of 25 mg/dL) and hypoalbuminemia (albumin: 2.9 g/dL). The rest of the liver function tests and tumor markers were unremarkable. On endoscopy as shown in *Figure 1*, a moderate amount of retained food particles was seen upon entry into the stomach. Narrowing of the pyloric opening along with a diverticulum in the anterior wall of the duodenal bulb was observed. Attempts to bypass the first portion of the duodenum were unsuccessful due to a narrowing at the duodenal bulb going into the second portion of the duodenum. Mucosal specimen of the duodenal narrowing was taken for biopsy. The test for *Helicobacter pylori* was positive.

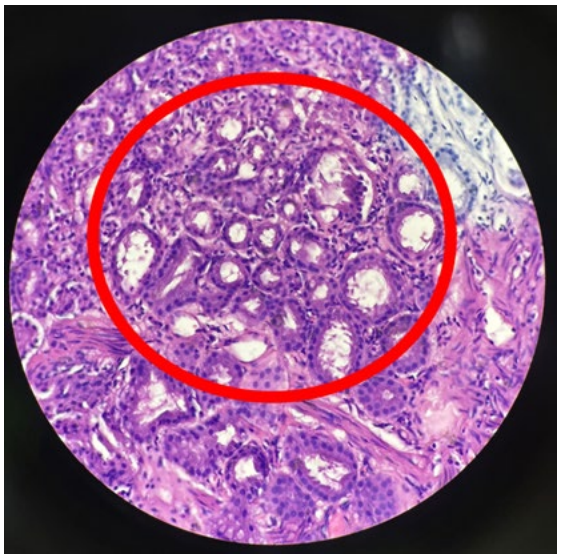

Figure 3. Histopathologic slide of the duodenal stricture showing simple columnar cells with the lamina propria exhibiting mild to moderate infiltration of lymphoplasmacytic cells. Red circle indicates the focal area exhibiting nests of oxyntic glands. No atypical cells seen.

Figure 2 shows the contrast enhanced CT scan of the whole abdomen which showed an irregular thickening at the proximal duodenum/duodenal bulb approximately 7 cm in length causing partial GOO. A consideration of duodenal ulcer versus malignancy was made. Likewise, gastritis along with a large non-obstructing lamellated cholelithiasis 1.9 x 1.7 cm, 599 HU with adjacent pericholecystic fluid and pneumobilia was seen and there was suspicious communication between the proximal duodenum and gallbladder. Although a fistulous tract was not identified, the above findings were consistent with a cholecystoduodenal fistula.

Figure 3 shows the histopathologic findings of the biopsy specimen of the duodenal narrowing taken during endoscopy which revealed chronic nonspecific duodenitis with focal cell nests suspicious of ectopic gastric oxyntic type glands vs. neuroendocrine hyperplasia. The patient was initially managed as a case of GOO secondary to a duodenal stricture probably malignant with a cholelithiasis and a possible cholecystoduodenal fistula. He was placed on nothing per orem and Cefoxitin was started prophylactically. He underwent exploratory laparotomy, antrectomy, pancreatic sparing total duodenectomy, cholecystectomy, with loop gastrojejunostomy, cholecystojejunostomy and pancreaticojejunostomy.

Operative findings revealed extensive fibrosis and acute on chronic inflammation of the gallbladder with dense adhesions to surrounding structures, requiring dissection of the liver, duodenum, and omentum (*Figure 4*). Dissection further revealed a gallstone impacted in and adherent to the wall of the gallbladder and a fistula opening into the duodenum.

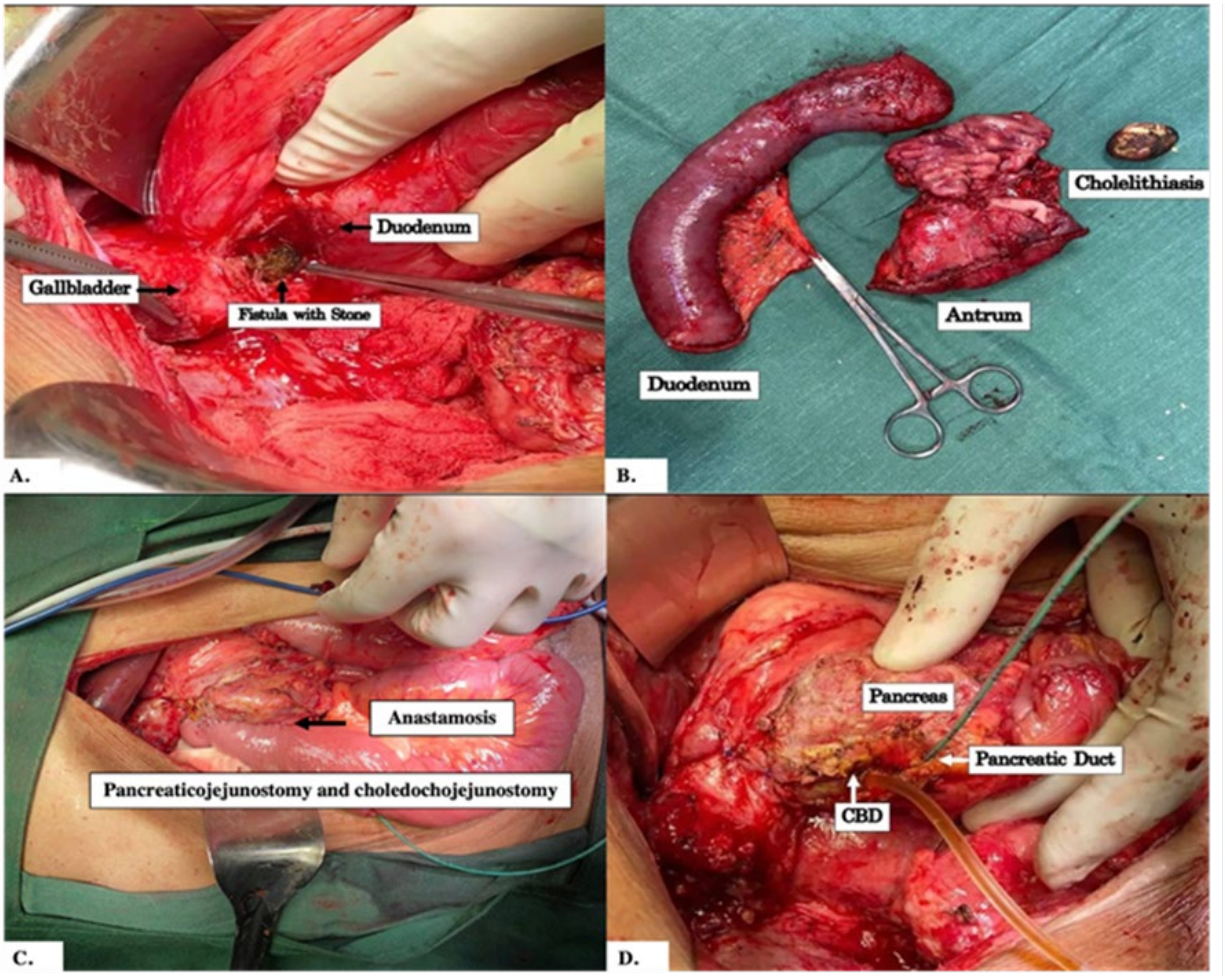

Figure 4. Operative findings. (A) Extensive fibrosis and acute on chronic inflammation of the gallbladder with dense adhesions to the surrounding liver, duodenum, and omentum. Impacted gallstone in and adherent to the wall of the gallbladder and a fistula opening into the duodenum. (B) Surgically removed duodenum, antrum with a 1.9 cm gallstone (C) Arrow indicating the anastomosis between the pancreas and jejunum as well as common bile duct and jejunum (C and D).

Postoperatively the patient remained stable and was able to gradually tolerate progression of diet. Antibiotics were continued with the addition of Metronidazole and the proton pump inhibitor was continued. The patient was started on triple therapy for *H. pylori* consisting of Amoxicillin 1g twice a day, Clarithromycin 500 mg 1 tab twice a day and Pantoprazole 40 mg twice a day for 14 days. He was then discharged on the 14th hospital day.

Final histopathology results of the surgical specimen revealed peptic ulcer disease on the segment of the antrum (*Figure 5*). The duodenum showed benign duodenal tissue with periduodenal lymph nodes exhibiting nonspecific lymphoid hyperplasia. The mesenteric and peripancreatic lymph nodes had nonspecific lymphoid hyperplasia and the gallbladder exhibited chronic cholecystitis.

Four months after discharge, the patient was admitted at another institution due to episodes of melena and anemia. Although the patient had improved appetite and gained 5 kgs since discharge, he was non-compliant with the prescribed proton pump inhibitors. A repeat endoscopy was done which showed a marginal ulcer near the anastomotic site. Sclerotherapy was done. Repeat *H. pylori* test was negative, and the patient was discharged with a proton pump inhibitor with no recurrence of melena.

Five months after discharge the patient was again admitted due to diffuse abdominal pain. Contrast enhanced CT scan of the whole abdomen showed bowel wall perforation with reactive enteritis located approximately 3.5 cm from the gastrojejunostomy anastomotic site. Unfortunately, the patient expired two hours before surgery.

Discussion

Inflammation of the gallbladder rarely directly involves surrounding structures. The most common sequelae for patients with chronic cholecystitis presenting with GOO

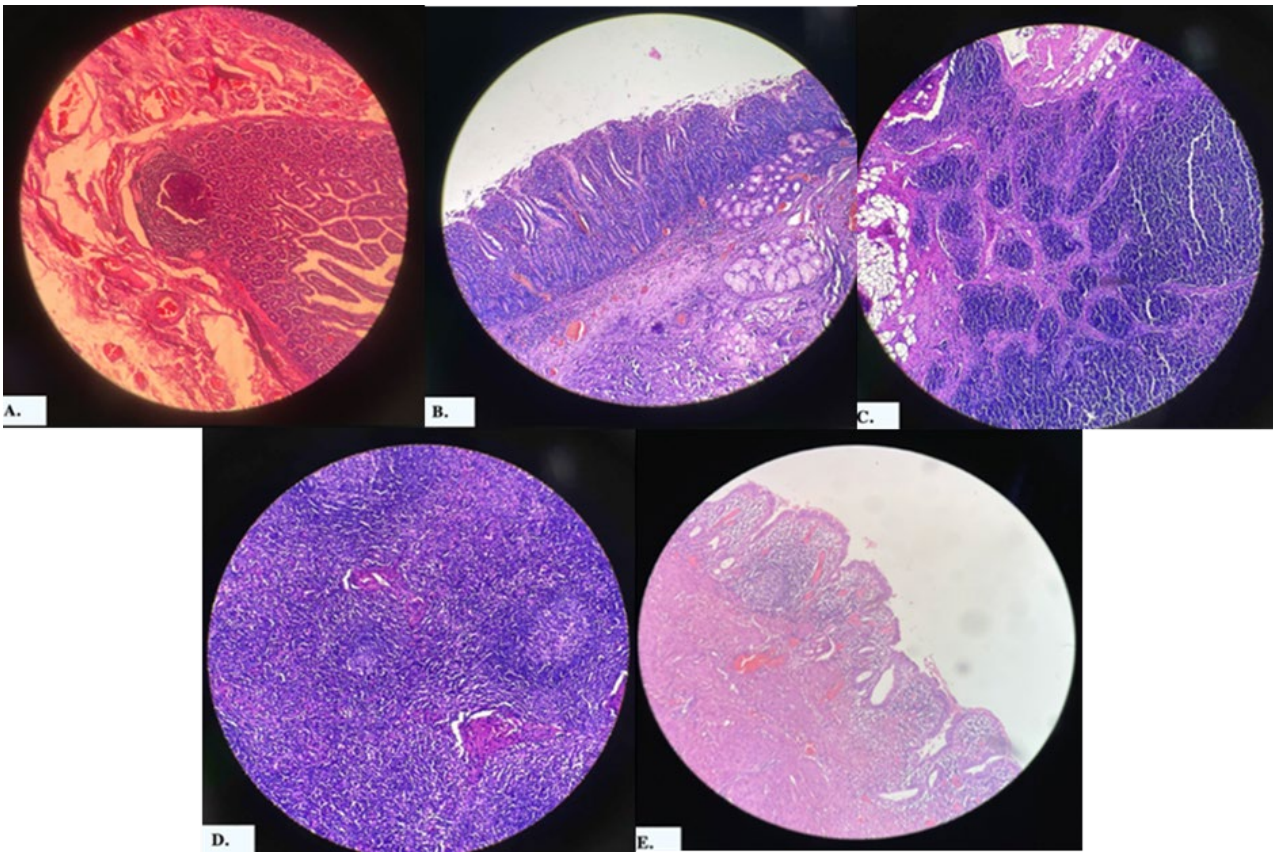

Figure 5. Histopathologic slides of surgical specimens. (A) Section of benign duodenal tissue with 1 lymph node attached exhibiting enlarged mucosal lymphoid particles with a germinal center. No tumor cells seen with no signs of chronic inflammation. (B) Section of gastric tissue with the epithelium eroded and replaced by heavy infiltration of both mononuclear and polymorphonuclear leukocytes. (C and D) Section of lymphoid tissue- mesenteric and peripancreatic exhibiting enlarged mucosal lymphoid follicles with widened germinal centers with no tumor cells seen. (E) Section of the gallbladder lined by villous mucosa from which Rokitansky-Aschoff sinuses are formed. The lamina propria and muscular layer exhibit vascular congestion and moderate infiltration of mononuclear cells consistent with chronic cholecystitis.

includes perforation of the gallbladder wall with abscess formation, creation of fistulous tracts, and extension of the inflammatory process to the surrounding structures.[5] In these cases, dense adhesions secondary to chronic inflammation of the gallbladder can cause narrowing of the pylorus and/or the duodenum causing extrinsic compression with resultant GOO. The exact incidence of this case is unknown with very few case reports published. In one report, a 54-year-old man presented with GOO arising from Xanthogranulomatous Cholecystitis, accompanied by cholecystoduodenal fistula and cholecystocolonic fistula, but without impacted gallstones. He was initially conservatively managed and eventually underwent exploratory laparotomy. Intraoperatively, the patient had edematous gallbladder with diffuse wall thickening, accompanied by an adhesion to the adjacent omentum, the first portion of duodenum and the proximal transverse colon. The patient underwent cholecystectomy and primary closure of cholecystoduodenal fistula and cholecystocolonic fistula and was discharged after 28 days.[6]

Spontaneous biliary fistula such as cholecystoenteric fistula formation is a rare complication of gallstone disease with an incidence of 0.1-0.5%. Spontaneous biliary fistula is usually caused by chronic disease of the biliary tract, either through chronic calculous cholecystitis or choledocholithiasis. It is commonly seen in female patients with advancing age and comorbidities. The most common type of spontaneous internal biliary fistulas are cholecystoduodenal (70%), cholecystocolonic (14%) and cholecystogastric (6%).[7]

Various imaging modalities are helpful in identifying the cholecystoenteric fistula. Conventional abdominal radiographic findings may show pneumobilia, ectopic radiopaque gallstones and dilation of the small bowels, the so-called Rigler's triad. CT scan may show similar findings delineating the site of the obstruction, the size of the stone and the location of fistula. It has a sensitivity of 93% and specificity of 100% in diagnosing gallstone ileus.[7] Further evaluation with upper endoscopy or even exploratory laparotomy may be performed if imaging findings are inconclusive such as in this case.

Once a fistulous tract is established and if sufficiently large, stones may pass, and patients may present with mechanical obstruction in the form of gallstone ileus whereas other patients can present with cholangitis or non-specific symptoms in the background of calculous biliary disease. Thus, patients with spontaneous biliary fistulas can present as obstructive or nonobstructive types.[7]

In the obstructive type such as gallstone ileus, the stone that passes through the fistula most commonly passes distally. If the stone is more than 2.5 cm, it usually gets impacted in the terminal ileum, less commonly in the jejunum or colon. Very rarely does it pass proximally causing duodenal obstruction or the so-called *Bouveret syndrome*. As the stone migrates, the patient may have intermittent obstruction. Enterolithotomy alone and enterolithotomy plus cholecystectomy with excision of the fistulous tract are the two surgical strategies employed to manage a patient with gallstone ileus. There is no high-level evidence regarding which strategy is the best.[7] In cases where *Bouveret syndrome* is considered, surgery is an option since it removes the impacted stone, repairs he fistula, and removes the gallbladder. However, endoscopic removal and lithotripsy of the obstructing stone can also be done.[9]

In the non-obstructive type of spontaneous biliary fistula, the patient usually presents with nonspecific signs and symptoms and is usually identified intraoperatively during exploration for calculous biliary disease.[7] Patients commonly present with ascending cholangitis and managed with open cholecystectomy and fistula repair.

In our case, the patient presented with a one-year history of recurrent epigastric pain with gradual worsening along with the presence of postprandial vomiting, early satiety, and bloating. On repeat contrast enhanced CT scan of the whole abdomen, the presence of cholelithiasis along with pneumobilia and possible cholecystoduodenal fistula could not be totally ruled out; *Bouveret syndrome* as a cause for the obstruction was not ruled out. Based on his symptomatology on admission he was managed as a case of GOO probably secondary to *Bouveret syndrome*.

Endoscopy for this case revealed a duodenal narrowing at the first to the second portion of the duodenum. However, the cause of such narrowing is unknown and malignancy could not be ruled out thus the decision to refer the patient for surgical evaluation. Intraoperatively, dense adhesions between an inflamed gallbladder and duodenum with a gallstone impacted in and adherent to the wall of the gallbladder and a fistula opening into the duodenum. In this case, the patient had no definite evidence of impacted gallstone in the duodenum however he did have a gallstone accompanied by a cholecystoduodenal fistula. An extensive surgical procedure was done and although initially successful, he still developed complications (marginal ulcer and perforation) and the patient eventually expired.

Summary

This case report highlights the fact that preoperative and intraoperative differential diagnosis of GOO is a challenge when it is associated with inflammatory changes involving surrounding tissues and structures. Chronic calculous cholecystitis through intense inflammation can present as one of the rare causes of GOO. In this case, the degree of GOO was dependent on the degree of inflammation from cholecystitis causing severe intraluminal narrowing. Optimal treatment plan should take into consideration the underlying etiology of the GOO.

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