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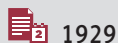
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A Rare Case of Cholecystoduodenal Fistula with Rapid Distal Gallstone Migration

Authors' Contribution:

Study Design A
Data Collection B
Statistical Analysis C
Data Interpretation D
Manuscript Preparation E
Literature Search F
Funds Collection GAEF 1 **Kayla K. Brown**
AEF 2 **Kyle J. Cunningham**
AEF 3 **Adam Howell**
AE 3 **John Edward Vance**1 College of Osteopathic Medicine, Nova Southeastern University Dr. Kiran C. Patel
College of Osteopathic Medicine, Davie, FL, U.S.A.
2 College of Osteopathic Medicine, Alabama College of Osteopathic Medicine,
Dothan, AL, U.S.A.
3 Department of General Surgery, Ascension Michigan Genesys Hospital,
Grand Blanc, MI, U.S.A.**Corresponding Author:** Kayla K. Brown, e-mail: Kb2020@mynsu.nova.edu**Conflict of interest:** None declared**Patient:** Female, 51-year-old
Final Diagnosis: Cholecystoduodenal fistula
Symptoms: Nausea • non-bilious emesis • right upper quadrant abdominal pain
Medication: —
Clinical Procedure: —
Specialty: Surgery**Objective:** Unusual clinical course**Background:** Abnormal communicating channels or fistulas between the gallbladder or common bile duct and the intestine are rare, but have potential to result in serious complications. Further complications can arise with migration of gallstones from the gallbladder to the intestines, causing distal obstruction in the ileum, intestinal hemorrhage, or intestinal perforation. High clinical suspicion is warranted for the diagnosis of Bouveret's syndrome, with anticipation of surgery to prevent distal gallstone migration that would otherwise result in unfavorable patient outcomes.**Case Report:** A 51-year-old woman presented with biliary colic and a computed tomography scan showed that a gallstone measuring approximately 3 cm was lodged in the first portion of the duodenum. The patient was diagnosed with cholecystoduodenal fistula with Bouveret's syndrome. Because of the acute presentation of symptoms, she underwent an exploratory laparotomy with disconnection of the cholecystoduodenal fistula, cholecystectomy with debridement of the duodenum, transduodenal gallstone removal, and primary duodenoplasty closure of D1.**Conclusions:** As the present case illustrates, distal migration of a gallstone through a cholecystoduodenal fistula can occur rapidly and without obvious symptoms. It also can occur spontaneously and not just secondary to fragmentation by laser lithotripsy. To prevent morbidity and mortality, a high degree of clinical suspicion is warranted when diagnosing patients in whom a gallstone ileus is seen on imaging.**Keywords:** Biliary Fistula • Gallstones • IleusFull-text PDF: <https://www.amjcaserep.com/abstract/index/idArt/929150>

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Background

Obstruction of the gastrointestinal (GI) tract is a rare but serious and potentially fatal complication of gallstone disease. Gastric outlet obstruction (GOO) is any process that causes impaired or blocked emptying of the stomach. Furthermore, GOO due to a gallstone that has passed into the GI tract is known as Bouveret's syndrome. Patients with acute GOO classically present with early satiety, abdominal pain, and postprandial vomiting. Patients with Bouveret's syndrome occasionally present with Mirizzi syndrome, wherein an impacted stone in the cystic duct or neck of the gallbladder obstructs the hepatic duct [1]. As in our case described here, stones blocking the gastric outlet can become dislodged, either spontaneously or iatrogenically, and transit through the intestines, potentially causing damage or obstruction further along the GI tract. Our patient presented to a community hospital for management. The aim of the present work, which meets the Surgical Case Report criteria, is to describe surgery used to manage this complex presentation with the goal of preventing further complications associated with the disease process [2].

Case Report

Our patient was a 51-year-old woman who presented to the Emergency Department (ED) with right upper-quadrant abdominal pain. She had a history of biliary colic and a computed tomography (CT) scan performed 4 months before her hospital visit showed a gallstone that measured approximately 3 cm. At the time the present report was written, imaging results and laboratory values, and information about the original location of the gallstone and presence of a fistula, were unavailable. Documentation was not available as to why surgery was not performed when the patient first presented with biliary colic.

During her visit to the ED, the patient reported symptoms of mild nausea and non-bilious vomiting and stated that she had been passing flatus and had a bowel movement that morning. A CT scan ordered in the ED showed evidence of a cholecystoduodenal fistula and a gallstone measuring approximately 3 cm lodged in the first portion of the duodenum. The patient then was diagnosed with cholecystoduodenal fistula with Bouveret's syndrome. She was immediately scheduled for an exploratory laparotomy with disconnection of the cholecystoduodenal fistula, cholecystectomy with debridement of duodenum, transduodenal gallstone removal, and primary duodenoplasty closure of D1. Specimens were to be collected during the procedure, including the gallbladder and gallstones.

During surgery, a hardened, highly inflamed, decompressed gallbladder was palpable beneath the caudad retracted colon. Metzzenbaum scissors were used to separate the gallbladder from the first portion of the duodenum and the cystic duct. The

cystic duct was clamped and tied off with silk suture. The hepatoduodenal ligament was visualized. There was a clear fistulous tract in the superior portion of the duodenum, and a Kocher maneuver was performed to mobilize the duodenum. The large gallstone that had been identified on CT was not palpable in the duodenum, so the small bowel was run from the first portion of the duodenum to the ligament of Treitz. The stone was identified then milked back proximally to the original D1 fistulous tract and extracted from the bowel lumen. Intraoperatively, there was discussion about creating a second enterotomy if the stone could not be safely milked in retrograde fashion. However, because the gallstone was sufficiently mobile within the intestine, we had no difficulty retrieving it atraumatically and without extensive labor. This was a unique case, in that, because gallstone retrieval was easy, an enterotomy was unnecessary and not performing it decreased the potential for further leaks and complications. Done with care, mobilization of the stone was atraumatic throughout its manipulation.

Metzenbaum scissors were used to debride all the devitalized tissue on the edges of the duodenal wound. We then used interrupted 3-0 silk suture to close the duodenal wound transversely in a single layer. Next, the anesthesiologist irrigated the stomach via the nasogastric (NG) tube and there was no evidence of saline leak from the closure sites. A Graham patch was placed over the top of the duodenal closure and a 15 F flat JP drain was placed into the gallbladder fossa. The patient's abdomen was thoroughly irrigated and there was no evidence of bleeding or succus before fascial closure. She tolerated the procedure well, with no intraoperative complications and was transferred to the floor. Since the procedure, the patient has remained stable with no surgical complications.

In retrospect, our tactical surgical plan carried greater risk than we had anticipated, but that was not evident until after we had taken down the cholecystoduodenal fistula. From the preoperative CT scan, the gallstone appeared to be lodged in the proximal duodenum, and upon initial palpation of the duodenum. It also appeared as though the gallstone was obstructed in this same location, but we were deceived by what turned out to be inflammatory tissue from the fistulization. Once we gained intraluminal access, we were not able to identify the gallstone proximally; subsequently, were able to locate it distally in the small bowel. Ideally, had we identified the gallstone earlier, we could have avoided additional manipulation of the fistula. Early identification was not possible, however, based on our clinical suspicion, results from the physical examination and imaging studies, and the patient's presentation, which underscores the rarity and uniqueness of this case presentation.

Unfortunately, no imaging studies, additional laboratory values, or illustrations were available at the time of this writing with which to outline the progression of the patient's course.

Pathology

Because additional images and illustrations were unavailable, detailed descriptions of the operative specimen, based on the information in the gross and microscopic pathology reports, are provided here.

Gross Description

The specimen was from a gallbladder gallstone and portion of the duodenum. It consisted of multiple ragged, disrupted fragments of gallbladder that could not be oriented; a separate, barrel-shaped calculus and a portion of possible bowel were loose in the container. The fragments of gallbladder were tan-brown, dusky, and rubbery and measured 6×5×2.0 cm in aggregate. The fragments contained multiple areas of defect, ranging from 0.2 cm up to 2 cm in the largest dimension. Definitive orientation could not be determined because the specimen was disrupted and ragged. No definitive cystic duct margin was identified on gross examination. The external surfaces of the fragments were cauterized, tan-brown, and disrupted; all had exposed tan-brown, dusky mucosa with a velvety appearance. Further sectioning of the larger fragment showed a wall thickness up to 0.4 cm. The calculus loose in the container, which was tan-brown to yellow, measured 3.5 cm long and 2.5 cm in diameter. The separate fragment of possible bowel in the container, which had exposed tan-brown, ragged, dusky mucosa, measured 2.5×1.2×1 cm.

Microscopic Description

The gallbladder wall was thickened and fibrotic, with areas of transmural xanthogranulomatous inflammation. Fibrosis of the mucosa was variable, as was the degree of acute and chronic inflammation. Much of the xanthogranulomatous inflammation involved the wall of the gallbladder and serosa. There were also fragments of duodenal tissue, including mucosa, submucosa, and muscularis propria. There were areas of mucosal ulceration, with associated reactive epithelial changes, hemorrhage, edema, and submucosal and muscularis propria fibrosis. In some areas, there was a transmural infiltrate of neutrophils and eosinophils.

Discussion

Abnormal communicating channels or fistulas between the gallbladder, main bile duct, or intestines are rare, but they can result in serious complications [3]. The prevalence of internal biliary fistula is less than 0.3% in patients with gallstone disease; 70% to 90% are cholecystoduodenal fistulas, making them the most common type [4]. Patients with a cholecystoduodenal fistula may also present with Mirizzi syndrome, which has

been described, in rare cases, in specific rural Chilean populations [1]. Most stones that pass through a cholecystoduodenal fistula do so uneventfully through the ileum or cause gallstone ileus, and just 2% to 4% of all gallstone-related obstructions result in Bouveret's syndrome. Thanks to improved detection and surgical techniques, the mortality rate for Bouveret's syndrome has dropped to <12% [5]. Endoscopic lithotripsy and stone retrieval is often the first-choice treatment in patients with symptomatic Bouveret's syndrome, followed by surgical lithotomy and simple enterotomy only when attempts at stone retrieval fail [6]. Endoscopic lithotripsy, however, often results in fragmentation of the gallstone into smaller pieces that are capable of causing distal obstruction in the ileum, intestinal hemorrhage, or intestinal perforation [5]. Regardless of where the primary obstructing stone is located, the remaining portion of the bowel should be examined to exclude other stones. In our case, the obstructing stone had migrated to the mid-jejunum within hours of patient presentation and administration of oral contrast, underscoring the necessity of examining the entire bowel. A single study previously described another unique case of stone migration within a few days of the onset of GOO symptoms [5]. Again, our patient had even faster transit in stone migration, which led to high clinical suspicion and a need for immediate intervention to prevent morbidity and mortality.

In our case, the patient presented with the classic symptoms of nausea, non-bilious vomiting, and abdominal pain. However, she did not meet the "emergent" criteria of obstruction, given her continued ability to pass flatus and a produce bowel movement earlier in the day. When a CT scan revealed a gallstone that measured approximately 3 cm in the first portion of the duodenum, surgery was warranted to prevent distal migration and resolve the patient's symptoms. During the procedure, which was performed within hours of her presentation and CT imaging, it was evident that the gallstone had migrated a significant distance distally to the mid-jejunum. This rapid migration was unexpected but did not create significant complications, such as perforation, distal obstruction, or intestinal hemorrhage. Had the distally migrated gallstone been recognized earlier in the surgery during initial inspection, a simple enterolithotomy could have been completed. This would have eliminated the need to disrupt the cholecystoenteric fistula and perform a cholecystectomy, thereby avoiding the potential for increased patient morbidity. Further, this type of approach would have allowed for expectant management following surgery, given the high probability of biliary-enteric fistula closure or spontaneous shrinkage. The standard of care for gallstone ileus management is highly controversial, ranging from enterolithotomy to cholecystectomy and fistula closure. For patients with gallstone ileus, however, the standard of care and most widely accepted and appropriate choice is simple enterolithotomy [7].

Conclusions

The presentations and complexities of GOO are multiple and varied in individuals who have complaints of early satiety, abdominal pain, and postprandial vomiting. Whether to manage medically and what type of surgery to perform must be decided early on in a patient's course to decrease the overall risk of future complications. One potentially serious cause of GOO is a gallstone that has passed through an aberrant tract or fistula from the gallbladder or bile duct into the lumen of the GI tract, such as a cholecystoduodenal fistula. Various imaging modalities can be used for early diagnosis of GOO, ranging from CT to magnetic resonance (MR) and MR cholangiopancreatography [8].

Current surgical management of symptomatic Bouveret's syndrome often begins with endoscopic lithotripsy and stone retrieval. Surgical lithotomy and simple enterotomy can only be used once stone fragment retrieval fails; that, in turn, can predispose the patient to future and more devastating complications [9]. Furthermore, these types of gallstones have the potential for spontaneous displacement distally within the bowel, rather than secondary to lithotripsy or other iatrogenic causes. For these reasons, we recommend that clinicians have a high index of clinical suspicion for Bouveret's syndrome during the diagnosis and evaluation of patients with GOO caused by passage of a gallstone through a cholecystoduodenal fistula. In such cases, the threshold for surgical intervention should be low to reduce patient morbidity and mortality.

Conflict of Interest

None.

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