

Single Case

Mirizzi-Induced Bouveret's Syndrome: Revelations of Timely Surgical Intervention

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Keywords

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Abstract

Bouveret's syndrome is an unusual clinical presentation of gastric-outlet obstruction and is the most infrequent variant of gallstone ileus with just over 300 cases in the literature. A 73-year-old female presented with innocuous constitutional symptoms and was found to have Mirizzi type Vb, a cholecystoduodenal fistula with obstruction. Esophago-gastroduodenoscopy-attempted dislodgement was unsuccessful. A gastric-jejunal bypass was the only option due to friability of the tissue. On post-op day 5, the patient developed acute abdominal pain and was found to have gallstone ileus. This case emphasizes the importance of early surgical intervention in cases of acute on chronic cholecystitis.

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Introduction

We report a highly atypical case of gallstone ileus as a late adverse event of surgically resolved, Mirizzi-induced Bouveret's syndrome. Gallstone ileus is rare, at about 30–35/1,000,000 admissions over a 45-year period [1] and carries a mortality rate five to ten times higher than those of mechanical small-bowel obstructions [2–4]. While gallstone ileus represents 0.3–0.5% of all mechanical small-bowel obstructions, Bouveret's syndrome represents approximately 1–3%

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within those gallstone ileus cases. The stages of Mirizzi syndrome are sequela of a single, large, or many small gallstones impacted in Hartman's pouch or the gallbladder infundibulum and cystic duct. When chronic or unaddressed, this pathology progresses insidiously until presentation.

Case Report

A 73-year-old female presented to the ED with a 3-week history of right-upper quadrant pain. A subsequent workup revealed chronic cholecystitis with cholelithiasis. She was stabilized and instructed to follow-up shortly with a surgeon for elective cholecystectomy; however, she never presented to follow-up. A year later, she presented again with severe abdominal pain with emesis. Computed tomography (CT) scan showed a large stone causing gastric-outlet obstruction with the presence of a cholecystoduodenal fistula shown in Figure 1. An emergent esophago-gastroduodenoscopy was performed in attempt to remove the stone; however, the gastroenterologist was unable to dislodge the stone despite two separate attempts. Consequently, she underwent exploratory surgery where a gastro-jejunostomy was performed due to the severely friable state of gastric tissue surrounding the impacted stone. The patient recovered well when on the fifth day post-op, she developed significant abdominal distention and pain; the CT scan revealed a terminal ileum gallstone ileus as shown in Figure 2. She successfully underwent another hand-assisted laparotomy where a small enterotomy was performed with resolution of her symptomatology.

Discussion

In 1896, Leon Bouveret described the first 2 cases of this unique syndrome. It remains difficult to detect as patients typically present with nonspecific symptoms. A large review in 2006 of 128 cases found that greater than 85% of patients diagnosed presented with nausea and vomiting, followed by 70% with abdominal pain [5]. A high index of suspicion is crucial, particularly in older females with a history of cholelithiasis. Most cases of Bouveret's syndrome are preceded by acute cholecystitis. Inflammatory cytokines and activated neutrophils lead to the formation of adhesions. Inflammation and pressure of the culprit gallstone result in erosion through the gallbladder wall, leading to a fistula between the gallbladder and the now-adhered portion of the GI tract [6, 7]. Adopted after the work of Mirizzi in 1948, the similarly named syndrome is a phenomenon whereby extrinsic compression of the bile duct via pressure applied indirectly from an impacted stone in the gallbladder results in chronic inflammation and ulceration. This results in a spectrum of cholecystobiliary or cholecystoenteric fistulas, as in the case of our patient [8]. A cholecystoduodenal fistula is the most common

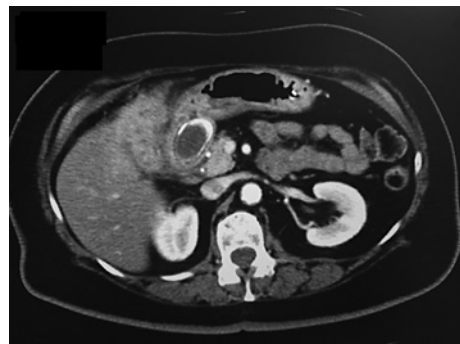


Fig. 1. CT scan showed a large stone causing gastric-outlet obstruction with the presence of a cholecystoduodenal fistula.

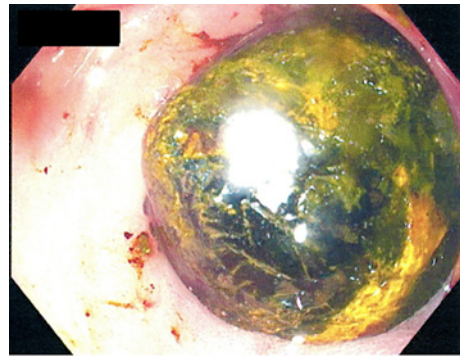


Fig. 2. CT scan revealed a terminal ileum gallstone ileus.



Fig. 3. Direct visualization of obstruction stone on EGD. EGD, esophago-gastroduodenoscopy.

biliary-enteric fistula, between 32.5 and 96.5%, and the least common are cholecysto-jejunal/ileal [9]. Diagnoses may be multimodal; however, CT is considered superior to either abdominal film or ultrasound with sensitivity of up to 93% [10]; however, abdominal films are not imperative. Established in 1941, Rigler et al. [12] identified four signs of obstruction seen on radiograph: the first was partial or complete obstruction; the second was pneumobilia or contrast material in the biliary tree; the third was a gallstone; and the fourth was a stone position change in subsequent films as it tumbles. Two of the first three are considered pathognomonic for diagnoses and are found in 20–50% of cases [12]. In 1978, a fifth sign was added which consisted of the presence of two air-fluid levels in the RUQ, and this finding is present in 24% of patients on admission [13].

Most patients will undergo an esophago-gastroduodenoscopy; yet, while nearly all endoscopies show the presence of obstruction, actual visualization, as occurred in our case, only occurs in roughly two-thirds of cases as shown in Figure 3 [5]. As therapies advance, endoscopic removal of impacted stones is increasing. Nets/baskets and mechanical, electrohydraulic, laser, and extracorporeal shockwave lithotripsy have been successfully documented in facilities where the equipment is available. In our patient, several attempts were made to dislodge the stone endoscopically but were unsuccessful. Surgical intervention was then required.

While ultrasound is a reliable first-line method of investigating biliary disease and can reveal evidence of Mirizzi syndrome, its sensitivity is highly variable. CT scans are useful for determining the location and impetus of biliary obstruction; yet, there are no specific radiographically evident features of Mirizzi. Endoscopic retrograde cholangiopancreatography (ERCP), despite its invasiveness is the gold standard with a sensitivity upward of 76.2% [14]. ERCP yields the most direct evidence of Mirizzi via visualization of the extrahepatic bile ducts and classic extrinsic compression by impacted stones in the CBD. Most importantly, it can show fistulas and biliary obstruction and is a treatment modality as endoscopists can perform

sphincterotomy and biliary stent placement. Unfortunately, the patient was not a candidate due to Bouveret's syndrome, preventing an ERCP from being performed, and thus necessitating surgery.

The significant delay in seeking care led to a precarious intra-abdominal situation for the surgeon. Had the patient undergone a cholecystectomy at initial presentation, the gallstone would not have had time to migrate via cholecystoduodenal fistula formation into the gastric-outlet, resulting in significant tissue ulceration. Any manipulation of the friable tissue would have surely led to sloughing and destruction of the anatomy, including her gallbladder. A gastro-jejunostomy was performed, bypassing the site of the Bouveret's obstruction, leaving the gallbladder untreated. On post-op day 5, she complained of abdominal pain. Gallstones are notoriously difficult to identify on AXR, typically the first-line imaging given the low cost, low risk, and speed of the exam. Typically, only 10–20% of stones have enough calcium to be radio-opaque. While Rigler's pentad is useful, a high degree of suspicion is necessary to continue with diagnostic imaging in the setting of unremarkable imaging [15]. Thus, a CT scan was done revealing a distal gallstone ileus, a rarity of mechanical obstructions, with incidence of <5%. Following the bypass, inflammation had decreased to a level whereby the stone dislodged and tumbled until resting in the narrow, terminal ileum. Following a repeat hand port-assisted laparoscopy, a small enterotomy was required to remove the stone with resolution of obstructive symptomatology. The patient was then safely discharged home.

The patient developed advanced and exceptionally rare intra-abdominal pathology due to delayed surgical intervention. The chronicity of her cholecystitis led to an advanced stage of Mirizzi syndrome that evolved into a gastric-outlet obstruction. Had the patient received specialty consultation at her initial encounter, the potentially disastrous and financially burdensome chain-of-events may have been avoided.

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Statement of Ethics

Informed written consent was obtained from the patient for the publication of their information and imaging in this journal. Consent is available to the editor upon request. This study protocol was reviewed, and the need for approval was waived by Riverside Community Hospital Ethics Committee.

Conflict of Interest Statement

COI: Daniel Stenberg, Massimo Arcerito, Harpreet Kaur, and Mazen Jamal each state they do not have any conflict of interest or any financial support behind this work.

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Author Contributions

Dr. Stenberg wrote the entire manuscript, reviewed the data, and provided and edited the pictures. He drafted the work for important intellectual content and provided the extra-final approval of the revision to be published. He agreed to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. Dr. Arcerito cowrote the manuscript, reviewed the data, provided and contributed photos, and edited for content. He drafted the work for important intellectual content and provided the final approval of the revision to be published. He agreed to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. Dr. Kaur reviewed the manuscript and provided substantial contribution to the conception or design of the work, and she revised the manuscript critically for important intellectual content; she gave the final approval of the final version to be published. She agreed to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. Dr. Jamal reviewed the manuscript, provided photos, and provided substantial contribution to the conception or design of the work; he revised the manuscript critically for important intellectual content, and he gave the final approval of the final version to be published. He agreed to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

Data Availability Statement

All data generated or analyzed during this study are included in this article. Further inquiries can be directed to the corresponding author.

References

- 1 Kurtz RJ, Heimann TM, Beck AR, Kurtz AB. Patterns of treatment of gallstone ileus over a 45-year period. *Am J Gastroenterol*. 1985;80(2):95–8.
- 2 Ayantunde AA, Agrawal A. Gallstone ileus: diagnosis and management. *World J Surg*. 2007;31(6):1292–7.
- 3 Reisner RM, Cohen JR. Gallstone ileus: a review of 1,001 reported cases. *Am Surg*. 1994;60(6):441–6.
- 4 Mallipeddi MK, Pappas TN, Shapiro ML, Scarborough JE. Gallstone ileus: revisiting surgical outcomes using national surgical quality improvement program data. *J Surg Res*. 2013;184(1):84–8.
- 5 Chen H, Siwo EA, Khu M, Tian Y. Current trends in the management of Mirizzi Syndrome: a review of literature. *Medicine*. 2018;97(4):9691.
- 6 Fox PF. Planning the operation for cholecystoenteric fistula with gallstone ileus. *Surg Clin North Am*. 1970;50(1):93–102.
- 7 VanLandingham SB, Broders CW. Gallstone ileus. *Surg Clin North Am*. 1982;62(2):241.
- 8 Mohan PR, Kumar M, Pacharu R. Mirizzi syndrome. *Med J Armed Forces India*. 2011;67(3):280–1.
- 9 Nuño-Guzmán CM, Marín-Contreras ME, Figueroa-Sánchez M, Corona JL. Gallstone ileus, clinical presentation, diagnostic and treatment approach. *World J Gastrointest Surg*. 2016;8(1):65–76.
- 10 Deitz DM, Standage BA, Pinson CW, McConnell DM, Krippaehne WW. Improving the Outcome in Gallstone Ileus. *Am J Surg*. 1986;151(5):572–6.
- 11 Yu CY, Lin CC, Shyu RY, Hseih CB, Wu HS, Tyan YS. Value of CT in the diagnosis and management of gallstone ileus. *World J Gastroenterology*. 2005;11(14):2142–7.
- 12 Rigler LG, Borman CN, Noble JF. Gallstone obstruction: pathogenesis and roentgen manifestations. *J Am Med Assoc*. 1941;117(14):1753.
- 13 Luu MB, Deziel DJ. Unusual complications of gallstones. *Surg Clin North Am*. 2014;94(2):377–94.
- 14 Cappell MS, Davis M. Characterization of Bouveret's syndrome: a comprehensive review of 128 cases. *Am J Gastroenterol*. 2006;101(9):2139–46.
- 15 Chang L, Chang M, Chang HM, Chang AI, Chang F. Clinical and radiological diagnosis of gallstone ileus: a mini review. *Emerg Radiol*. 2018;25(2):189–96.