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CASE REPORT

Bouveret's syndrome and cholecystogastric fistula: a case-report and review of the literature

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Abstract

An exceptionally rare cause of gastric outlet obstruction, Bouveret's syndrome results from proximal impaction of an ectopic gallstone, enabled by fistulization that aberrantly connects the biliary and luminal gastrointestinal tract, typically a cholecystoduodenal fistula. It occurs with a 2:1 female predominance, most often in the eighth decade of life. Endoscopic treatment is the preferred first-line strategy in management, followed by surgical intervention if unsuccessful. Endoscopy failed to retrieve the stone due to its size, despite attempted lithotripsy, which prompted laparoscopic retrieval. Bouveret's syndrome compels a high index of suspicion in proximal gastrointestinal obstruction even when presenting in a male a decade younger than the median age of diagnosis (74 years), with no preceding biliary symptoms particularly as early intervention can considerably reduce morbidity and mortality.

Keywords Bouveret's syndrome · Gastric outlet obstruction · Gallstones

Abbreviations

EGD Esophagogastroduodenoscopy
CT Computed tomography

Introduction

Luminal complications of errant gallstones occur in less than 1% of cholelithiasis and classically present as gallstone ileus [1, 2]. If obstruction does occur, it most frequently does so given the direction of peristalsis and the narrowed caliber of the distal intestine in the terminal ileum [3]. Gastric outlet obstruction from an ectopic gallstone, or Bouveret's syndrome, comprises less than 5% of these cases, with only around 300 observations reported in the literature to date [1, 2]. It is approximately twofold more common in females, with a median age at presentation of 74 years [4]. Early recognition of gallstone-related luminal disease is critical, as it confers a mortality of 12–30% [5, 6]. Management requires relieving the obstruction through removal or destruction of the impacted stone, which can be performed endoscopically or surgically. Endoscopic approach is the recommended first-line of treatment, considering the attendant co-morbidities in the affected demographic [7, 8].

Case report

A 63-year-old man with obstructive sleep apnea and prostate cancer status post radical prostatectomy 1 year prior presented to the Emergency Room following a 3-day history of epigastric pain and non-bilious, post-prandial vomiting. Physical exam was unremarkable apart from dry mucous membranes. Laboratory data showed leukocytosis of 14.2 ($\times 10^9$)/L and serum creatinine of 1.56 mg/dL.

An abdominal ultrasound was limited by bowel gas. Abdominal Computed Tomography (CT) demonstrated an air-filled gallbladder, with evidence of a cholecystogastric fistula (Fig. 1). The gallbladder wall was thickened with only mild cholecystic fat stranding. There was a small amount of air in the proximal common bile duct, extending into the left biliary duct. A calcified density was seen in the gastric antrum (Fig. 2).

A nasogastric tube was placed for gastric decompression. Esophagogastroduodenoscopy (EGD) demonstrated a large stone at the pylorus, with a substantial portion occupying the proximal duodenum (Fig. 3). The fistula could not be

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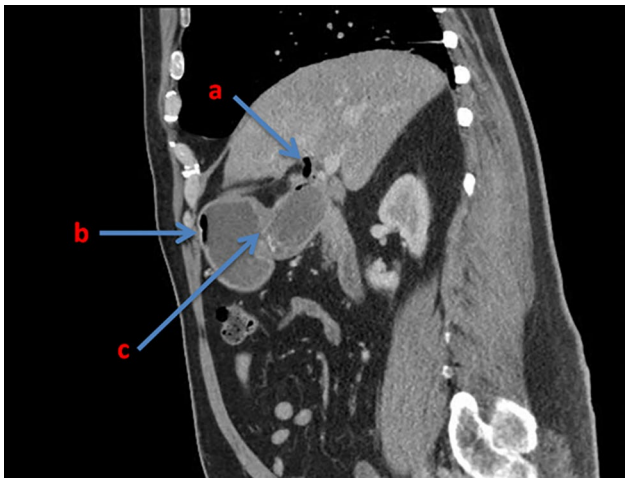

Fig. 1 Sagittal view abdominal computer tomography demonstrating pneumobilia (Arrow **a**), air in the stomach (Arrow **b**) and the cholecystogastric fistula (Arrow **c**)

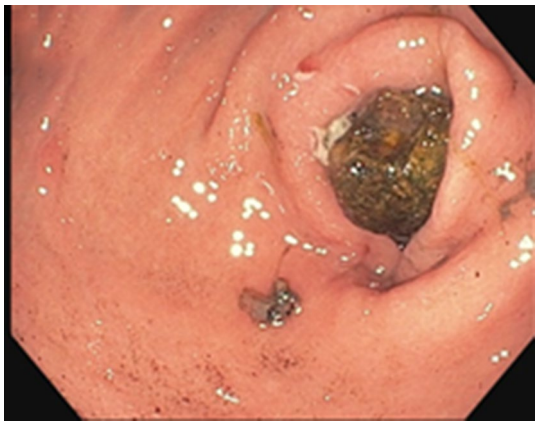

Fig. 3 Upper endoscopy revealing a large calcified body impacting the pyloric channel

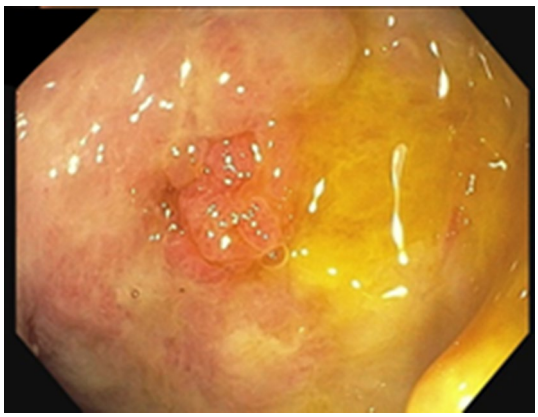

Fig. 4 Upper endoscopy showing a deep cratered ulcer at least 3 cm in diameter, with puckered mucosa underneath the stone

visualized, ostensibly due to the thickness of the gastric wall. Contrast dye infusion confirmed there was no leak or active fistula before attempting stone extraction. With intravenous glucagon and retrograde manipulation by a firm snare, the stone was dislodged from the pylorus. This revealed a deep, cratered ulcer at least 3 cm in diameter, with puckered mucosa, and further contrast dye did not reveal any extravasation from this source, either (Fig. 4). However, given the concern for pressure-induced ulceration from an impacted ectopic stone, which may result in perforation with further tissue manipulation, biopsy was not performed on the cratered ulcer at the time of attempted stone retrieval. With the stone in the gastric antrum, mechanical lithotripsy was unsuccessfully attempted with stiff wire snares and a Single-use Olympus LithoCrushV Mechanical Lithotriptor with a 30-mm basket. Electrical lithotripsy was also unsuccessful. En-block removal, despite implementing an esophageal overtube, was not attempted due to concern for

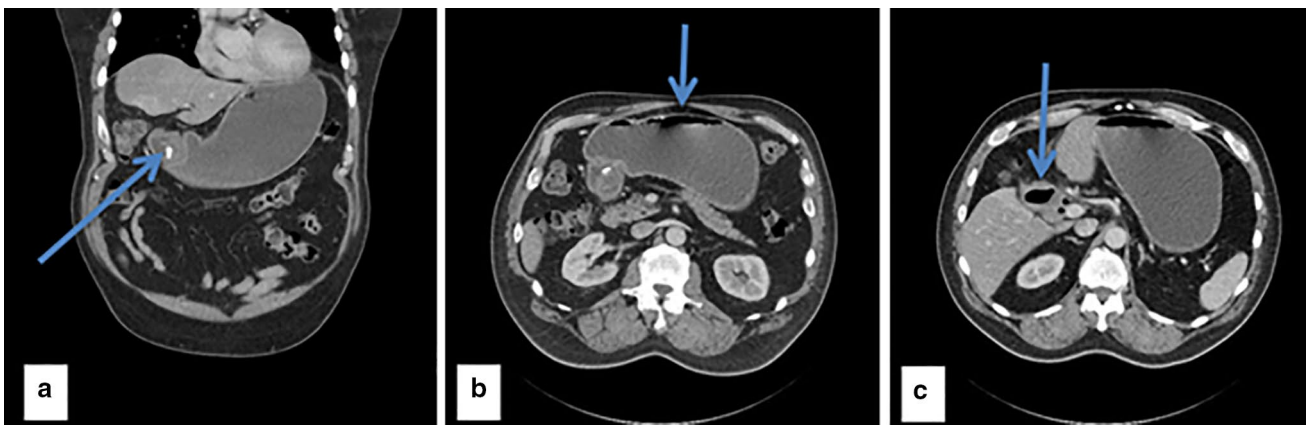

Fig. 2 Coronal view abdominal computed tomography showing ectopic stone in gastric antrum (Image **a**), air fluid level of the stomach (Image **b**), and air in the gallbladder lumen (Image **c**)

inducing esophageal perforation. The stone was left in the gastric fundus.

One day later, the patient underwent laparoscopic gastrotomy through the anterior gastric body, with stone removal via an Endo Catch™ Gold Specimen Retrieval Pouch (Fig. 5). Due to concerns about the inflammatory milieu and invasive nature of a cholecystogastric fistula take down, he did not undergo simultaneous cholecystectomy or fistulectomy. He was discharged on post-operative day four without issue. One month later, he denied any right upper quadrant pain, fever, chills, nausea, vomiting, anorexia, or jaundice. CT of the abdomen at that time noted the interval gastrotomy with removal of the obstructing gallstone, as well as residual soft tissue density tracking from the gastric antrum to a contracted gallbladder where there had previously been a cholecystogastric fistula. There was no evidence of biliary ductal dilatation, gastric distension, or duodenal obstruction. Given that the patient was asymptomatic, lacked menacing features on subsequent cross-sectional imaging and had no residual stones in the gallbladder, and that cholecystectomy would require a complex surgery for takedown of a cholecystogastric fistula, it was decided that an expectant approach would be pursued. The patient has remained asymptomatic on a follow-up visit 7 months later.

Discussion

French physician Leon Bouveret coined his eponymous syndrome in 1896 to describe the phenomenon of gastric outlet obstruction from an ectopic gallstone, permitted through bilioenteric fistulization [9]. Such fistulization is thought to occur from a sequence of luminal apposition, chronic inflammation, and subsequent ischemia, which may be the result of mechanical pressure or underlying malignant process, the latter of which compels thorough evaluation if suspected [10]. The two most commonly observed fistula types in ectopic gallstone disease are cholecystoduodenal and cholecystocolic which comprise 68% and 17% of the cases, respectively. Only 5% of the cases are observed to involve a cholecystogastric fistula, which was the presenting aberrant biliodigestive connection seen in our case, likely due to the relatively thicker tissue of the gastric wall compared to that of the proximal small bowel [1, 11].

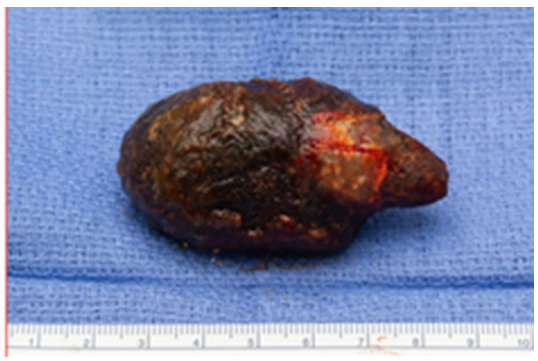

Fig. 5 Gross examination of the 6.2×3.4×3.1 cm gastric calculus

While the presence of a fistula can facilitate the introduction of gallstones into the intestinal lumen, it is thought that approximately 85% of such stones are eliminated through emesis or defecation, whereas those larger than 2–2.5 cm typically get impacted in the enteric lumen, often at the terminal ileum because of its narrow caliber [3]. Conversely, 1–4% of ectopic gallstones defy small bowel peristalsis and instead obstruct proximally at the gastric outlet, causing Bouveret's syndrome [12].

Bouveret's syndrome classically presents at a median age of 74.1 years and has a female predilection (65%) [4]. Unsurprisingly, common manifestations of Bouveret's syndrome include epigastric pain (71%) and nausea/vomiting (86%), though other features can include distention (27%), hematemesis (15%), and melena (6%). Physical exam can be unremarkable or can demonstrate abdominal tenderness (44%) and/or signs of dehydration (31%) [4, 13]. Approximately one-third of patients will lack clinically-apparent antecedent biliary disease, as was the case in our patient. This underscores the importance of a high index of suspicion for gallstone-associated luminal disease in patients presenting with obstructive symptoms [11].

The diagnosis can be made by abdominal X-ray, ultrasound, and CT. Ultrasound is often hampered by bowel gas, as it was here. CT is considered the most accurate test, conferring a specificity of 100%; however, 15–20% of gallstones is isoattenuating with surrounding soft tissue and would not be visualized on CT [4, 6, 14, 15]. Rigler's triad (small bowel obstruction, pneumobilia, and an ectopic gallstone) is found on CT in roughly 78% of cases of gallstone ileus [16].

EGD and surgery can aid in both diagnosis and treatment. Though it has been cited as successful in less than a third of cases of Bouveret's syndrome, endoscopy is still recommended by most authors as first-line treatment due to its safety profile [8]. Stone removal can be performed via endoscopic basket or net, though it typically fails with stones exceeding 2.5 cm, as occurred in our case, as removal must happen through the narrow lumen of the esophagus [6]. To facilitate retrieval, endoscopic lithotripsy can be performed through mechanical, electrohydraulic, and laser techniques, none of which have been compared in head-to-head trials for Bouveret's syndrome. Lithotripsy confers a risk of distal obstruction or ileus from stone fragments; thus, careful inspection of the bowel and extraction of stone fragments must be done following lithotripsy.

Failed management by endoscopy should be followed by surgical intervention. Nearly 73% of patients have undergone surgical intervention for Bouveret's syndrome, 61% of which

was compelled by failed endoscopic intervention [17]. Laparoscopic approach, which was first performed successfully for Bouveret's syndrome in 2005, now accounts for 6% of the cases overall [8, 18]. Surgical approach is typically by gastrostomy, as in our patient. If the stone is impacted in the distal duodenum or if the initial gastrolithotomy is unsuccessful, endoscopic translocation of the stone distally to the jejunum can allow for attempted jejunal enterolithotomy. Importantly, duodenotomy is not recommended due to the retroperitoneal orientation of the 2nd to 4th parts of the duodenum. In a unique case, Hanandeh et al. managed a case of Bouveret's syndrome by a duodenotomy in the lateral anti-mesenteric border of the 2nd part of the duodenum after failed trails of stone extraction via the stomach or the jejunum [19]. The defect at the enterotomy site is commonly sutured by primary closure. Bowel resection or Roux-en-Y duodenojejunostomy, in the case of a duodenotomy, is rarely needed for reconstruction [7]. Surgical management of Bouveret's syndrome should also include deliberate inspection of the entire intestine for other ectopic gallstones that would thwart recovery of bowel function post-operatively [6, 7, 11, 13].

There is on-going debate regarding cholecystectomy with fistula repair during or following surgical management of an ectopic stone. Proponents of the complete surgical management argue that spontaneous fistula closure is thought to promote a dysfunctional scleroatrophic gallbladder more prone to stone recurrence, infection, and cancer [7]. Pointing to gallstone ileus as a correlate, it has been shown that those patients who underwent enterolithotomy without cholecystectomy/fistulectomy had a 4.2–8.2% risk of recurrent stone-related complications, with an associated mortality of 12–20% [20, 21]. Though they may be instructive, these data are predicated on studies of gallstone ileus rather than Bouveret's syndrome explicitly.

Ultimately, the decision must be tailored to the patient. To that end, many authors agree that advanced age and serious medical comorbidities, which would not be unexpected in the demographics afflicted by Bouveret's syndrome, should have simple stone extraction and be observed, unless there is a high suspicion for underlying malignancy [7]. Surgery is considered strongly if the patient is young and healthy, if malignancy is strongly suspected, or if malignancy is present and amenable to surgical excision. This is most safely carried out in a two-staged procedure, with a delay of several weeks from relief of the obstruction from the impacted stone to allow for resolution of the inflammatory milieu [22]. Given our patient's age, the lack of symptoms post-operatively, and the lack of concern for malignancy, cholecystectomy and fistula-take down were not pursued by our surgical colleagues. Admittedly, while the duodenal ulcer seen on endoscopy was thought to be due to pressure-related changes from stone impaction, malignancy was not ruled out by tissue sampling, and the patient—reassured by his improvement after stone extraction and lack of changes on surveilling cross sectional imaging—declined further endoscopic diagnostic tests to that end.

In sum, we have presented a case of the rare Bouveret's syndrome with additional and notable atypical features: its occurrence in a male instead a female, at a younger age than median diagnosis and with a cholecystogastric instead of a cholecystoenteric fistula. Nevertheless, it serves to illustrate several important features of what can otherwise be a nefarious clinical picture of ectopic gallstone-associated luminal disease. Given the protean nature of gastric outlet obstruction and lack of obligatory antecedent gallbladder symptoms in Bouveret's syndrome, a high index of suspicion is necessary. Endoscopic approach should be the first-line of treatment considering that it usually presents in geriatric patients with multiple comorbidities. Surgical approach via a gastrolithotomy or an enterolithotomy would be indicated if endoscopic treatment failed. The decision to perform a cholecystectomy with fistula take-down must be tailored according to each patient's condition.

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Author contributions KO, DM and ATK performed the literature review and wrote the manuscript. ML reviewed and edited the manuscript. All authors approved of the submission.

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Compliance with ethical standards

Conflict of interest Karim Osman, Daniel Maselli, Ayse Tuba Kendi and Mark Larson declare that they have no conflict of interest.

Human and animal rights All procedures followed have been performed in accordance with the ethical standards laid down in the 1964 Declaration of Helsinki and its later amendments.

Informed consent Informed consent was obtained from all patients for being included in the study.

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