

https://doi.org/10.71599/bhr.v4i1.126

# Case report



# Gallstone migration out of circuit: A case report of an intestinal perforation from an ectopic gallstone

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#### Abstract

**Introduction**: Entero-biliary fistula is a rare and difficult-to-diagnose complication of cholelithiasis. While spontaneous evacuation may occur with small stones, surgical intervention is required in most cases. This condition presents significant challenges for the surgeon, as complete clearance of the digestive tract, along with cholecystectomy, may be fraught with difficulty. In this case, we report an instance of intestinal perforation caused by a migrating gallstone.

**Case Presentation:** An 80-year-old woman presented to our surgery department with abdominal pain. Several diagnostic modalities were employed to reach a definitive diagnosis. Abdominal ultrasound confirmed the diagnosis of acute cholecystitis. The patient underwent urgent surgery after brief resuscitation. During exploration of the abdominal cavity, a punctiform perforation of an intestinal loop was identified. Upon bidigital palpation, a mass was felt that rolled over the fingers. The patient underwent segmental resection, double stoma creation, and feeding jejunostomy. A 2 cm gallstone was expressed from the specimen.

**Discussion**: One of the rare complications of cholelithiasis is the development of an entero-biliary fistula. Small bowel perforation caused by a migrating gallstone is considered uncommon. The surgical challenge lies in managing both the perforation and the causative fistula, as cholecystectomy itself can be risky. Thorough examination of the small intestine during surgery is crucial to check for any additional intraluminal stones.

**Conclusion**: Despite its rare prevalence and atypical presentations, a high index of suspicion for entero-biliary fistula is essential, particularly in patients with risk factors. With prompt diagnosis and timely intervention, patients can successfully recover from this clinical condition.

#### Keywords:

Received: August 17, 2024; Accepted: December 25, 2024

## 1. Introduction

Non-traumatic small intestine perforation is a devastating condition that can endanger the patient. Apart from treating the perforation, special attention must be devoted to identifying its cause to prevent further perforations. Given the range of possible causal agents (including toxic, inflammatory, autoimmune, infectious, malformative, vascular, tumoral, or foreign bodies [1]), it is crucial to precisely identify the culprit. In this case, we describe an unusual cause of endogenous perforation, represented by an impacted gallbladder stone. We aim to raise awareness of the medical community about this condition, which is diagnosed intra-operatively in half of cases [2], to better equip healthcare professionals.

# 2. Case report

An 80-year-old woman, who had been experiencing recurrent biliary colic for 5 years prior to her current consultation, presented with right upper quadrant pain and chills. She denied any significant medical history or unhealthy habits.

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Upon examination, the patient exhibited a fever of 38.7°C, an accelerated pulse of 114 beats per minute, and localized guarding in the right hypochondrium with tenderness in the remaining quadrants, but no jaundice. Laboratory tests showed elevated inflammatory markers without signs of cholestasis. Ultrasound imaging revealed a thickened gallbladder measuring 5 mm, with gallstones, and subhepatic and Douglas effusion.

After a brief resuscitation, she was rushed for exploratory surgery under the suspicion of acute perforated cholecystitis. The surgical approach began with a right subcostal incision. Upon inspection, a generalized purulent effusion and dense adhesions in the hepatic area were noted, complicating the dissection. A slit on the left side allowed for the widening of the operative incision. Following adhesiolysis and thorough washout of the intraperitoneal spillage, an inflammatory intestinal-mesenteric organization was identified, compartmentalizing a punctiform perforation at the antemesenteric edge, with sphacelous edges and an oedematous intestinal wall around the perforation (Fig. 1). Bidigital palpation revealed a mass that rolled over the fingers. The gallbladder was embedded in the liver parenchyma. A segmental resection was performed with transformation into a double enterostomy, along with a feeding jejunostomy, to address the disorders associated with the proximal enterostomy, given the perforation's location 40 cm from the ligament of Treitz. A 2 cm gallstone was expelled from the specimen (Fig. 2).

Her hospital stay was uneventful, though several days were required for her to resume daily living activities due to her frailty and senility. Histopathological examination of the resected specimen revealed an isolated jejunal perforation with ischemic edges, covered with fibrous coating, but without signs of local inflammation or arteriosclerosis at the perforation site. No malignancy was detected. Unfortunately, the patient passed away 2 months later due to significant ionic disturbances.



Fig.1. Intraoperative view of the intestinal loop covered with loose fibrin, a punciform perforation at the antemesenteric edge with sphacelous edges, oedematous intestinal wall around the perforation.



Fig.2. Gross examination of the resected specimen with the expressed gallstone.

# 3. Discussion

Small intestine perforations are considered rare, especially when compared to gastroduodenal and colonic perforations [3]. The causes of these perforations exhibit a geographical pattern: in Western countries, perforations of the lower digestive tract are more common, while in developed countries, perforations of the upper tract predominate [4]. The primary causes of small intestine perforations include ischemic, occlusive, inflammatory (e.g., Meckel's diverticulitis), infectious (e.g., tuberculosis, typhoid fever), and traumatic (e.g., instrumentation, foreign bodies) factors [3]. In developing countries, digestive infections, particularly typhoid fever and tuberculosis, are frequent causes of perforation, whereas in developed countries, conditions such as Crohn's disease and malignancies are more prevalent [5]. Rare causes include lymphoma, malignant small intestine tumors, Crohn's disease, and internal hernias, which together account for only 5.4% of cases, according to a retrospective study conducted by a Turkish team aiming to track down all causes of small bowel perforation [6]. Gallstones, not included in this study, are the subject of the present case.

In a Tunisian surgery, in a small central city, crude prevalence of cholelithiasis was 4% (5.4% in women and 1% in men) [7]. This frequent affection carries a nonnegligible rate of complication. After a mean follow-up of 14.3 years, a United States-based study found that patients with gallstone disease (either ultrasound finding of cholelithiasis or cholecystectomy) had higher all-cause mortality in age-adjusted (hazard ratio (HR), 1.3; 95% confidence interval (CI), 1.2-1.5) and multivariate-adjusted analysis (HR, 1.3; 95% CI, 1.1–1.5). A similar increase was observed for cardiovascular disease mortality (multivariateadjusted HR, 1.4; 95% CI, 1.2-1.7) and cancer mortality (multivariate-adjusted HR, 1.3; 95% CI, 0.98-1.8) [8]. Moreover, in a cohort study of mortality trends related to gallstone disease, the annual incidence of death was flat in a 10-year period [9]. Among the infrequent complications that can be observed in this setting is entero-biliary fistula. It represents 0.15% and 8% of biliary tract surgery [10]. It is in the path of vesicular macrocalculi that exert spontaneously resolving acute cholecystitis attacks. Over time, the vesicular wall thickens due to the parietal inflammatory modulation with accentuation of fibrosis. During this transformation, it can attract an adhesion with the neighboring structures, particularly the duodenum (ranging from 32.5% to 96.5%) [11], due to its favorable anatomical position. In a cohort study, published in 2021, of 31 patients with biliary ileus, fistula encountered intraoperatively was cholecystoenteric in 23 cases (74.19%), cholecystogastric in 7 cases (22.58%), and cholecystocolonic in 1 case (3.22%) [12]. In 2022, a Chinese team reported 29 cases of cholecystoenteric fistula, most patients having a history of gallstones for less than 5 years (55.2%) [13].

The impact of the stone generates localized pressure necrosis up to parietal ulceration and then perforation, thus allowing the stone to be delivered into the digestive lumen to discharge the calculus into the digestive tract. The calculus will follow the peristaltic activity along its course, and it could be impacted against the anatomical stricture zones, generating occlusion. However, intestinal perforation is a rare event. This case illustrates this rare eventuality. Perforation generally occurs in the antimesenteric border. which is explained by its poor blood supply, making it more susceptible to pressure necrosis from gallbladder stones [14]. Moreover, sequestration of fluid and electrolytes within the intestinal lumen due to the obstructive foreign body, coupled with vomiting induced by excessive vagal stimulation, results in dehydration. The stretch of the intestinal wall induced by the foreign body increases parasympathetic activity, which stimulates mucosal secretion of a large amount of water and electrolytes. Hypotension and dehydration stimulate the sympathetic nervous system as a compensatory mechanism to maintain adequate tissue perfusion. Sympathetic activation can override the normal homeostatic parasympathetic activity, decreasing the production of gastrointestinal secretions to reduce hypovolemia. Although this is beneficial in reducing luminal distention, sympathetic activation is detrimental to the maintenance of the integrity of the intestinal mucosa. Decreased secretion of alkaline mucus by the Brunner's glands exposes the mucosal epithelium to gastric hydrochloric acid and proteolytic enzymes. This results in disruption of the mucosal tight junctions that normally exclude bacterial penetration of the mucosa, and shunting of blood away from the intestinal microvasculature impairs turnover of mucosal epithelial cells, resulting in subsequent villous blunting [15].

The most frequent location is the ileum, because of its smaller caliber and less active peristalsis [16]. It accounted for 64.5% of cases in a series collected over a decade [17]. However, the site of impaction depends on the gallstone's size, the fistula's site, and the diameter of the bowel lumen [18]. In a recent review, gallstones larger than 2.5 cm in size were considered prone to impaction, but gallstones as small as 2 cm were reported to cause obstruction, and those as large as 5 cm were found to pass freely through the bowel loops [19]. While Shi-fei Huang et al. reported a median diameter of the impacted gallstone of 3.8 cm, with a range from 2 to 6 cm [13].

The management covers the fundamental and elementary gestures: aspiration of pus, retrograde emptying of the small intestine, peritoneal cleansing with warm saline, and removal of loose fibrin. The operative challenge resides in the handling of the perforation and the causative fistula. Intestinal sutures are avoided, and intestinal bypass is necessary with a resection extension of 10 centimeters on both sides of the perforations, because the ulceration of the mucosa and of the surrounding submucosa is almost constant [20]. Anastomosis failure is more likely in case of documentation of shock at presentation, the presence of two or more perforations, and intraoperative contaminant volume of more than one liter. The choice of ileostomy over resection/anastomosis is then the keystone of surgical treatment [21]. We advocate resection because it additionally provides insight into the cause of the perforation, whose therapeutic pursuit is steered by the histological result. Withholding anastomosis is justified by the risk of leakage. It is proclaimed that infection or bacterial contamination leads to collagen lysis by stimulating the colon submucosa collagenase [22]. Dunphy [23] demonstrated that bacterial growth in the anastomosis stimulates collagen lytic activity, breaking the suture tension. The execution of a biliary gesture or its execution in a second operative course increases the morbidity and remains for most authors useless in the absence of ulterior symptoms since the fistula ceases spontaneously in more than 50% of cases and the recurrence rate is less than 5% [24]. By taking this information into consideration, we agreed to suspend the biliary procedure and shorten the operating time. The latest operative step must be devoted to examining the small intestine thoroughly for the presence of other intraluminal stones. Clavien et al. reported multiple ectopic gallstones in 16% of cases [25]. Despite meticulous postoperative care, our patient expired due to high stoma output. In fact, delayed stoma takedown was justified by the high risk of complications when not considering the appropriate timing. In 2021, a prospective longitudinal study compared early takedown by 2 weeks to late closure by 10 weeks, with 30 patients in each group. Their results concluded that, in the early setting, it is a more technically difficult procedure (adhesions rate: 26 vs. 12; p=0.004), leading to increased operative time (126.1667±27.5895 vs. 86.0000±34.2506 minutes; p<0.001), postoperative complications, and increased length of hospital stay (17.9667±6.9851 vs. 11.2000±4.0548 days; p<0.001) [26].

## 4. Conclusion

The clinical manifestations of gallstones and their associated complications can vary widely, ranging from asymptomatic cases to severe conditions. This highlights the importance of a thorough understanding of gallstone-related diseases for effective patient management in the modern era. Surgeons must be prepared for the possibility of small intestine perforation caused by gallstones to perform the appropriate surgical interventions. Based on our findings, future surgeons should be well-equipped to make accurate diagnoses and implement the necessary therapeutic measures.

# **Consent of patient**

Patient's written informed consent was obtained to publish this manuscript.

## **Consent for publication**

Written informed consent was obtained from the patient for publication on this case report and any accompanying images.

## **Ethical considerations**

Ethical approval was obtained from the Research Ethics Committee of Rabta University Hospital, Tunis, Tunisia.

#### Funding

None.

### **Conflict of interest**

None.

#### Authors' contribution

The authors participated equally.

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Cite this article as: Mseddi MA, Mokhtar A, Nouri T, Sassi K, Ben Slima M. Gallstone migration out of circuit: A case report of an intestinal perforation from an ectopic gallstone. Biomedicine Healthcare Res. 2025:4:26-9. https://doi.org/10.71599/bhr. v4i1.126