

# Asymptomatic cholecysto-enteric fistula and gallstone ileus: Two case reports

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## Abstract

Cholecysto-enteric fistula (CEF) is an uncommon complication that can be seen incidentally during elective surgery or as a cause of mechanical intestinal obstruction. It usually involves elder females having comorbid conditions. Stones that pass into the small intestine due to CEF present themselves with recurrent ileus attacks and the clinical picture becomes apparent in case of complete obstruction. Very small stones, however, can be eliminated from the digestive system without causing ileus. There is no predictor for the development of CEF either in patients unaware of their gallstone or in those aware of their gallstone but asymptomatic. Thus, the diagnosis is established based on tests performed in emergency conditions or accidentally during elective surgery. Herein, we present two cases; the one with gallstone ileus due to CEF and the other with CEF detected during elective surgery.

**Keywords:** Gallstone; Cholecysto-Enteric Fistula; Ileus; Rigler's Triad.

## INTRODUCTION

Mechanical intestinal obstruction due to gallstones is a rare cause of ileus mostly affecting old females and may be encountered during the course of acute and chronic cholecystitis (1,2). If gallstones are small, they may pass through the ileocecal valve and, in that case, may remain asymptomatic or may present itself with recurrent cholangitis and even with gallbladder cancer in later periods (1). However, there is no clear sign indicating the development of cholecysto-enteric fistula (CEF), which causes this situation. Diagnosis of CEF is usually established incidentally or based on analyses performed in emergency conditions. Herein, two cases, which were treated in the general surgery clinic and operated on for CEF, are presented together with the findings derived from their physical examination, laboratory analyses, and preoperative evaluation. Verbal and written informed consents of both patients were obtained.

## CASE REPORT

### Case 1

A 69-year-old female patient visited the emergency room with nausea, vomiting, and extensive abdominal pain existing for the last 3 days. She had no documented

comorbidity other than hypertension. With regard to her vital signs, heart rate was 96 pulse/minute, arterial blood pressure was 160/80 mmHg, and body temperature was 36.4°C. Her physical examination revealed dry tongue, slightly increased turgor, and distended abdomen. On palpation, there was rebound tenderness prominently in the right upper quadrant together with metallic bowel sounds. Plain abdominal radiograph revealed broad-based air-fluid levels. The results of laboratory analysis were as follows: white blood count (WBC): 11,100/μL, blood urea nitrogen (BUN): 101 mg/dL, creatinine: 1.59 mg/dL, potassium: 2.83 mmol/L, aspartate aminotransferase (AST): 74 U/L, alanine aminotransferase (ALT): 190 U/L, total bilirubin: 2.13 mg/dL, direct bilirubin: 0.92 mg/dL, gamma-glutamyl transferase (GGT): 109 U/L, C-reactive protein (CRP): 0.71 pg/mL, and high-sensitivity troponin: 18 pg/mL and 24.9 pg/mL after 4 hours. No change was observed in the consecutive electrocardiography (ECG).

Abdominal computed tomography (CT), which was performed to identify the etiology of ileus, demonstrated images suggestive of air in the bile ducts, dilated jejunal loops, and an image suggestive of a stone causing obstruction at the level of distal jejunum (Figure 1). Following the pre-operative cardiology consultation, the

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patient was considered to be at high-risk for emergency surgical procedure. The patient was operated on after fluid and electrolyte replacement with the pre-diagnoses of gallstone ileus (GSI). Her preoperative evaluation revealed an appearance suggestive of a stone (4x3x2 cm) causing an obstruction in the 140 cm distal to the Treitz ligament. A 6-cm of intestinal segment containing the stone was resected and then end-to-end anastomosis was performed (Figure 2). On the postoperative day 5, the patient was discharged from the hospital with improvements in clinical and laboratory findings. The patient having no complaints on follow-up refused to undergo cholecystectomy. She was followed for 16 months after the surgery without any problem.



**Figure 1.** Computed abdominal tomography displaying (A) an impacted stone at the level of distal jejunum-proximal ileum and (B) air in the intrahepatic biliary tract



**Figure 2.** During exploration, (A) trace of the stone in the intestinal loops and (B) the resected intestinal segment and the stone

### Case 2

A 67-year-old female patient, who had mild, recurrent dyspepsia complaints, visited the emergency room twice in a year with abdominal pain spreading over her low back. During the examination performed on her second admission, the patient was suggested surgery with the diagnosis of symptomatic cholelithiasis. Her findings of preoperative laboratory analyses were considered normal (AST: 22 U/L, ALT: 28 U/L, alkaline phosphatase: 94 IU/L, GGT: 32 U/L, total bilirubin: 0.67 mg/dL, direct bilirubin: 0.27 mg/dL, WBC: 8,700/ $\mu$ L, hemoglobin: 11.7 g/dL). Her abdominal ultrasound revealed images suggestive of multiple stones in the gallbladder. During laparoscopic cholecystectomy, it was observed that a fistula was formed between the fundus of gallbladder and the duodenum. Therefore, the surgical procedure was changed from laparoscopic to open surgery and the CEF was exposed (Figure 3A). The duodenal opening of the fistula was debrided, repaired primarily, and then gallbladder dissection was performed.

Mirizzi syndrome type 2 was detected during dissection of the Calot's triangle. Cholangiography was performed and no formation causing an obstruction in the choledoc duct was detected (Figure 3B). Thereafter, cholecystectomy was completed and the choledoc duct was repaired primarily. The patient was discharged from the hospital on day 5 with improvement. She developed no problem during 12-month follow-up period.



**Figure 3.** (A) Cholecysto-enteric fistula and (B) patent biliary ducts on cholangiography

### DISCUSSION

In the present report of two cases, the differences among the signs indicating the development of CEF and its therapeutic management were underlined. Various studies have reported that 15-25% of the patients with asymptomatic gallstone would become symptomatic at the end of 10-15 years of follow-up period (2-4). Moreover, surgical treatment for asymptomatic gallbladder stones is recommended in the presence of increased incidence of gallbladder cancer and hemolytic disorders and in patients undergoing gastric bypass for morbid obesity (5-7). There is no marker either indicating the development of CEF or estimating the time of development in asymptomatic patients (1-5). Although GSI is a rare cause of mechanical intestinal obstruction, it usually involves elder patients having critical comorbid conditions. Both of the present cases were over the age of 65 years. Mortality rate in patients treated for GSI has been reported to be 5-6.7% depending on the comorbid conditions they have (7,8).

In 1994, Reisner et al. (8) defined GSI as an uncommon complication seen in 1-4% of the cases with cholelithiasis; however, they stated that it accounted for 25% of non-strangulated small bowel obstructions over the age of 65 years. In 2014, this rate was reported to be 0.5% by Halabi et al. (10). In our unpublished series, GSI was determined in 2 of nearly 1200 patients operated on for gallstone. In one of these cases presented herein, cholecystectomy was performed in the same session and CEF was repaired.

Gallstone ileus may manifest with recurrent ileus attacks as a sign of the stone moving through the intestine due to CEF. However, the clinical picture of ileus becomes apparent in case of complete obstruction. While 20% of the patients with GSI have the signs of acute cholecystitis, jaundice is present in less than 15% (1,10). The mean duration of symptoms prior to hospitalization has been reported as 5 days in average (7,9). The stone rarely remains at the level of fistula and causes gastric outlet

syndrome (Bouveret's Syndrome) (7,9). Very small stones are excreted through the digestive system with recurrent abdominal pain without causing ileus (7,9,10). In the present case of GSI, high levels of bilirubin and enzyme suggested biliary tract obstruction. Nevertheless, when ileus was detected, the existing situation was considered CEF-related cholangitis picture.

Not detecting the gallstone on the presenting control abdominal ultrasound in patients with documented gallstone together with the presence of pneumobilia and ileus picture, which is known as Rigler's triad, raises the suspicion of GSI diagnosis. Abdominal CT is the preferred method for GSI. Detecting a gallstone that causes intestinal obstruction on abdominal tomography verifies the diagnosis of GSI (1,11,12). Although pneumobilia is seen in 30-60% of the patients with gallstone, it is a nonspecific sign (7).

The biliary tract's, which is considered sterile, becoming accessible for microorganisms of the intestinal system due to CEF facilitates the development of cholangitis (13). This may explain Charcot's triad-like picture in the present case with GSI. The same thing might not be valid for our second case; this might be originated from the cystic duct's being obstructed by the gallstone and thus forming a barrier between the infected gallbladder and the biliary tract (14).

Although surgical treatment is debatable in the literature, single-phase surgery in which ileus and CEF are treated together in the same session in stable patients not at high risk is the generally accepted approach. The surgical treatment of ileus includes the elimination of obstruction by removing the stone either together with the resection of relevant intestinal segment or alone via enterotomy. Segmental resection was preferred because of suspicion of stenosis which may develop after enterotomy and repair. High-risk patients may be followed for the second session. However, symptomatic gallbladder disease occurs in 15% of the patients not undergoing cholecystectomy (12,15,16).

Regarding our first case, dense intraabdominal adhesion was observed in the right upper quadrant intraoperatively. The patient, after cardiology consultation, was considered to be at high-risk for emergency surgical procedure. The treatment plan was made only for ileus because of the morbidity that might be resulted from the potential extension of surgery duration. Treatment of gallbladder disease was postponed to the second session; however, the patient refused to undergo this surgical procedure.

In their study published in 2010, Ravikumar et al. (15) reported the incidence of gallbladder cancer to be 0.82% in patients undergoing surgery for CEF. In their series, Clavien et al. (1) reported this incidence to be as high as 25% in 1990. Such a decrease in the incidence after two decades can be explained by increased rates of diagnosis and operability of gallbladder diseases (1,15).

In case CEF is detected in the elective surgery performed

for cholelithiasis, its repair at that session is more convenient. Imaging attempts made during the surgical procedure helps with exposing the gallstone disease, which is likely to be overlooked. Reviewing the medical history of the second case, we determined no additional sign such as obstructive jaundice or cholangitis attack, except for biliary colic attacks that raise the suspicion for both CEF and Mirizzi syndrome type 2. Considering both cases, one had a single large stone and the other had multiple small stones. Absence of CEF-related ileus picture in the second case was attributed to the small size of the stones, which suggested that a multifactorial origin for fistulation between the gallbladder and the adjacent organ due to eroded gallbladder wall (18).

With regard to the outcomes during follow-up of the patients with documented asymptomatic gallstone, it has been estimated that 15-25% of such patients become symptomatic at the end of 10-15 years of follow-up period and that complicated gallstone disease would occur in only 1-2% of the patients under follow-up (2-4). For this reason, monitoring is recommended for asymptomatic gallstones. In the literature, however, there is no sign that indicates development of CEF or requirement for its prevention, except for old and female patient groups (1-11). There are no determinative parameters for either the size of gallstone or the presence of inflammatory events during CEF. The indication for surgical treatment of gallbladder stones detected incidentally is limited to the increased probability of gallbladder cancer, hemolytic disorders, and those undergoing gastric bypass. For this reason, the diagnosis could be established based on the signs of CEF-related ileus or infection in patients with undocumented gallstones (17,18).

## CONCLUSION

In conclusion, signs of mechanical intestinal obstruction together with jaundice, subfebrile body temperature, and tenderness in the right upper quadrant detected on physical examination of patients with ileus picture may strongly suggest CEF-related GSI. Moreover, in the presence of cholangitis picture in patients suffering from recurrent biliary colic, CEF, in addition to choledocholithiasis, has to be one of the situations needs to be kept in mind.

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## REFERENCES

1. Clavien PA, Richon J, Burgan S, et al. Gallstone ileus. Br J Surg 1990;77:737-42.
2. Kirchmayr W, Mühlmann G, Zitt M, et al. A Gallstone ileus: rare and still controversial. ANZ J Surg 2005;75:234-8.
3. Capocaccia L, the GREPCO group. Clinical symptoms and gallstone disease: Lessons from a population study. In: Epidemiology and prevention of gallstone disease,

- Capocaccia L, Ricci G, Angelico F, Attili AF Edition, Lancaster MTP Press, 1984. p.153.
4. Shabanzadeh DM, Sørensen LT, Jørgensen T. A Prediction Rule for Risk Stratification of Incidentally Discovered Gallstones: Results From a Large Cohort Study. *Gastroenterology* 2016;150:167.
  5. Diehl AK, Sugarek NJ, Toddpa KH. Clinical evaluation for gallstone disease: usefulness of symptoms and signs in diagnosis. *Am J Med* 1990;89:29-33.
  6. Bates GC, Brown CH. Incidence of gallbladder disease in chronic hemolytic anemia (spherocytosis). *Gastroenterology* 1952;21:104-9.
  7. Shiffman ML, Sugerman HJ, Kellum JM, et al. Gallstone formation after rapid weight loss: a prospective study in patients undergoing gastric bypass surgery for treatment of morbid obesity. *Am J Gastroenterol* 1991;86:1000-5.
  8. Reisner RM, Cohen JR. Gallstone ileus: a review of 1001 reported cases. *Am Surg* 1994;60:441-6.
  9. Mallipeddi MK, Pappas TN, Shapiro ML, et al. Gallstone ileus: revisiting surgical outcomes using National Surgical Quality Improvement Program data. *J Surg Res* 2013;184:84-8.
  10. Halabi WJ, Kang CY, Ketana N, et al. Surgery for gallstone ileus: a nationwide comparison of trends and outcomes. *Ann Surg* 2013;259-329-35.
  11. Moss JF, Bloom AD, Mesleh GF, et al. Gallstone ileus. *Am Surg* 1987;53:424-8.
  12. Rigler LG, Borman CN, Noble JF. Gallstone obstruction: Pathogenesis and roentgen manifestations. *J Am Med Assoc* 1941;117:1753-9.
  13. Seal EC, Creagh MF, Finch PJ. Gallstone ileus: a new role for abdominal computed tomography. *Postgrad Med J* 1995;71:313-5.
  14. Sung JY, Costerton JW, Shaffer EA. Defense system in the biliary tract against bacterial infection. *Dig Dis Sci* 1992;37:689-96.
  15. Mirizzi PL. Síndrome del conducto hepático, *J int Surg* 1948;18:731.
  16. Rodríguez-Sanjuán JC, Casado F, Fernández MJ, et al. Cholecystectomy and fistula closure versus enterolithotomy alone in gallstone ileus. *Br J Surg* 1997;84:634-7.
  17. Ravikumar R, Williams JG. The operative management of gallstone ileus. *Ann R Coll Surg Engl* 2010;92:279-81.
  18. Beuran M, Ivanov I, Venter MD. Gallstone ileus clinical and therapeutic aspects. *J Med Life*. 2010;3:365-71.