



## CASE REPORT / ПРИКАЗ БОЛЕСНИКА

# Spontaneous cholecystoduodenal fistula – spectrum of complications

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## SUMMARY

**Introduction** Spontaneous cholecystoduodenal fistula is a rare complication of the gallbladder calculus. Bowel obstruction is the complication in less than 1% of these patients. The pathognomonic triad (Rigler triad) of pneumobilia, small-bowel distention, and ectopic gallstones is typical for gallstone ileus. In only 1–3% of the patients with bowel obstruction by ectopic gallstone the localization of obstruction is in the duodenum, and it is called Bouveret syndrome. The rarest complication is a floating non-obstructing gallstone trapped in the stomach.

**Outline of cases** We present three elderly female patients with persistent abdominal pain and known gallbladder calculus in the patients' histories. Plain radiography of the thorax and abdomen and ultrasound were performed as the first choice and contrast-enhanced computer tomography (CT) was done subsequently. In the first patient, CT and magnetic resonance imaging (MRI) showed signs of pneumobilia, cholecystoduodenal fistula, and the presence of the gallstone in the stomach. The iodine contrast X-ray swallow test revealed a cholecysto-duodenal bulb fistula and floating calculus in the stomach, confirmed by endoscopy. In the second patient with persistent abdominal pain, CT and barium swallow test showed signs of pneumobilia, cholecystoduodenal fistula, and two ectopic gallstones obstructing duodenum – Bouveret syndrome. The third case showed signs of the Rigler triad – typical signs of gallstone ileus.

**Conclusion** Spontaneous cholecystoduodenal fistula is a rare condition with possible complications such as Bouveret syndrome, gallstone ileus and floating, non-obstructive gallstones in the stomach, as the rarest possible complication. CT, MRI with magnetic resonance cholangiopancreatography, as well as the contrast X-ray swallow test can be very helpful in the detection of the bilio-enteric fistula and ectopic gallstones.

**Keywords:** cholecystoduodenal fistula; Bouveret syndrome; Rigler triad; complications; ectopic gallstone.

## INTRODUCTION

Spontaneous cholecystoduodenal fistula is a rare complication of the gallbladder calculus, which can cause asymptomatic migrating gallstones within the bowel. Also, in less than 1% of the patients, a bowel obstruction can occur. This entity is more common in the elderly and is manifested with a number of nonspecific signs and symptoms. The pathognomonic triad (Rigler triad) of pneumobilia, small-bowel distention, and ectopic gallstones on conventional abdominal radiographs is seen in 30–35% of all cases with gallstone ileus [1–5]. Duodenal obstruction caused by an ectopic gallstone happens in only 1–3% of the patients and is known as the Bouveret syndrome [1, 6, 7]. The rarest complication is a floating non-obstructing gallstone trapped in the stomach, which can also lead to gastric outlet, persistent emesis, and esophageal rupture (Boerhaave syndrome), as reported in the study by Modi et al. [8].

## CASE REPORTS

### Patient 1

A 77-year-old female with persistent abdominal pain and continuous subfebrile condition was admitted to the Clinic for infectious diseases. Gallbladder calculus was reported in the patient's history. Blood analyses showed normal leucocyte number and liver function. Abdominal contrast-enhanced computer tomography (CT) was done the following day. CT scan of the thorax and abdomen showed pneumobilia, highly suspected cholecystoduodenal fistula (Figure 1a – arrow) and a 10-mm hypodense ovoid lesion in the stomach with hyperdense rim – calculus (Figure 2a). Magnetic resonance imaging (MRI) examination with magnetic resonance cholangiopancreatography (MRCP) confirmed a cholecystoduodenal fistula (Figures 3a and 3b) and a 12-mm calculus in the stomach (Figure 3c). Subsequently, an upper gastrointestinal

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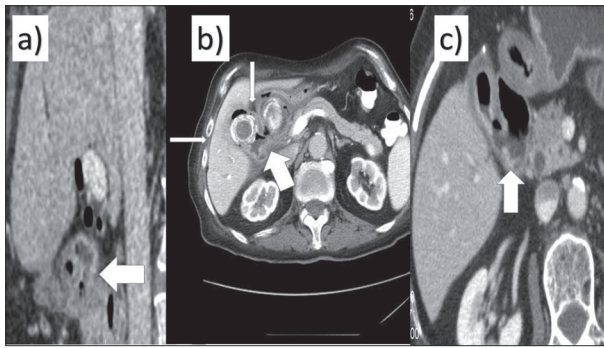
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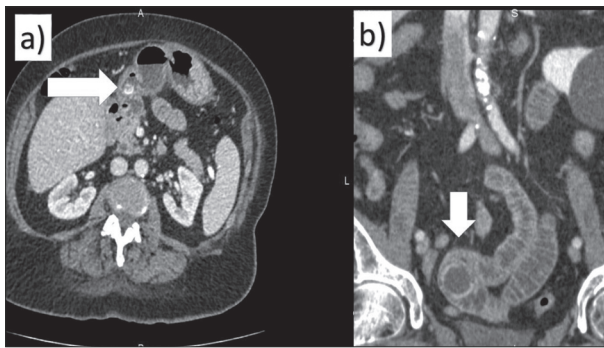
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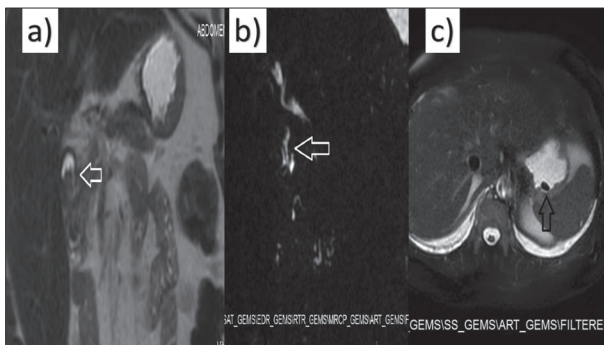
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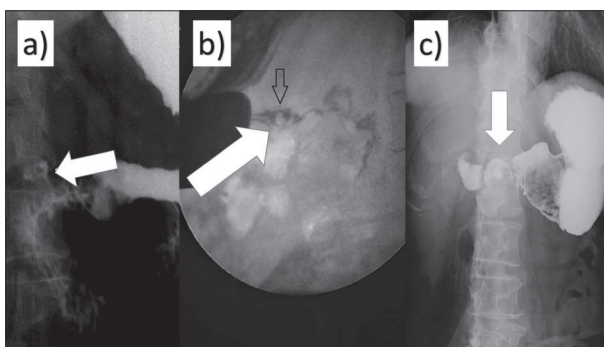
**Figure 1.** a) Cholecystoduodenal fistula in patient 1 – arrow; b) a 4-cm calculus in the duodenal cap and a 3-cm one in a thick-walled gallbladder (thin arrow), as well as a fistulous formation between the duodenum and the gallbladder (thick arrow); c) cholecystoduodenal fistulization – arrow



**Figure 2.** a) Hypodense ovoid lesion in the stomach with hyperdense rim calculus – arrow; b) hyperdense rim, 27-mm gallstone obstructing the ileum – arrow



**Figure 3.** a) Cholecystoduodenal fistula on the T2 sequence on magnetic resonance imaging (MRI) – arrow; b) cholecystoduodenal fistula on magnetic resonance cholangiopancreatography – arrow; c) calculus in the stomach on the T2 sequence on MRI – arrow



**Figure 4.** a) Cholecystoduodenal fistula on the X-ray barium test – arrow; b) floating calculus in the stomach on the X-ray barium test – arrow; c) the barium test showed a large filling defect in the duodenal cap (arrow) with extraluminal passage of contrast from the duodenal bulb

X-ray examination using iodine contrast was performed, which revealed a cholecysto-duodenal bulb fistula (Figure 4a) and a floating calculus in the stomach (Figure 4b). The existence of the gallstone in the stomach was confirmed by endoscopy, but it could not be removed using the basket. Considering the age, clinical condition and the comorbidities, it was decided to only follow-up the patient.

### Patient 2

A woman, age 74, was admitted to the hospital with a 10-day history of abdominal pain and recurrent vomiting. Comorbidities included diabetes mellitus, cardiomyopathy, and hypothyroidism. A previous abdominal ultrasound examination showed stones in the gallbladder. Blood analyses showed no signs of leukocytosis and abnormal liver function – therefore, an endoscopic examination was performed.

### Patient 3

An 83-year-old female was admitted to our department with repetitive vomiting, abdominal pain, and distention. Gallstones were diagnosed five years earlier; however, it was decided not to perform a surgical procedure due to the poor general and cardiac condition of the patient. Plain radiography of the thorax and abdomen was made. Plain radiography of the abdomen showed small bowel air-fluid levels. The patient was treated only with intravenous fluids and antibiotics. Later, clinical exacerbation was investigated with abdominal CT, which showed signs of pneumobilia, cholecystoduodenal fistulization (Figure 1c – arrow) and contracted irregular gallbladder outline containing air. CT also revealed distended small bowel loops with an ectopic, isodense 27-mm gallstone with hyperdense rim obstructing the ileum (Figure 2b). The gallstone was extracted by means of a longitudinal enterotomy, closed transversally. No attempt was made to remove the gallbladder or to repair the cholecystoduodenal fistula. The patient recovered well.

## DISCUSSION

Bilio-enteric fistula is a rare complication which occurs in approximately 1% of all patients with gallstones. Most commonly it happens in older women, which is probably due to the female constitution, as there is regularly elongation and ptosis of hypotonic gallbladder and less visceral abdominal fat tissue around it, so the gallbladder essentially “hangs,” which is why it is in direct contact with the duodenal wall. Adding decubitus of the chronically pressured ischemic gallbladder walls (elderly ischemia) with the stones, the cholecystoduodenal fistula is created. Finally, one should be aware of the fact that the presence of sex disparities in some parts of the cardiovascular system anatomy has already been revealed [9].

The most common type is cholecystoduodenal fistula (60%), while cholecystocolic, cholecystogastric, and

choledochoduodenal fistulas are also described [10]. The diagnosis of complications is challenging since clinical signs and symptoms are unclear and unspecific. Patients could suffer from numerous symptoms such as dyspepsia, abdominal pain, malabsorption, melena, and diarrhea. Migrating through the fistula, large gallstones could cause intestinal obstruction, but they could also migrate to any part of the gastrointestinal tract without causing any symptoms. The most commonly obstructed part is the terminal ileum, which leads to the gallstone ileus [2, 4, 10]. Classic abdominal radiographic signs of pneumobilia, mechanical bowel obstruction, and ectopic gallstone were first described by Rigler, but in around one half of the patients there are usually two out of three signs [1, 3, 11]. It rarely happens that patients vomit gallstones passed through fistula to the stomach. There may be a wide range of complications due to an existing fistula such as cholangitis, peritonitis, intestinal obstruction, and hemorrhage due to malignancy [12, 13]. One of the complications of the cholecystoduodenal fistula was presented in our second case – Bouveret syndrome, which implies proximal duodenal or distal stomach obstruction by biliary calculus. It is a rare syndrome that affects elderly women with previous history of biliary calculosis [6, 14, 15].

Oral contrast (iodine or barium) X-ray tests, CT, MRI with MRCP, as well as endoscopic retrograde cholangiopancreatography and hepato-biliary scintigraphy, can be used for diagnosing cholecystoduodenal fistulas [16]. In our cases, the patients had prior knowledge of cholelithiasis and were not operated on. CT showed signs of pneumobilia, which indicated the existence of pathological communication between the biliary tree and the gastrointestinal tract [13, 17, 18]. Pneumobilia, gas in the gallbladder and cholecystoduodenal fistula, were also confirmed by the MRI and MRCP examination. MRI with MRCP is useful in detecting isoattenuating gallstones and in patients with the intolerance to oral contrast medium. Negi et al. [19] successfully demonstrated that gallstones, pneumobilia, as well as cholecystoduodenal fistula, can be visualized by this imaging modality.

As demonstrated in our cases, an oral contrast swallow test can be very helpful in confirming the presence of a fistula, an obstructing gallstone in the duodenum as a filling

defect, as well as non-obstructing calculi in the stomach. It is a simple and low-cost examination which is highly recommended in cases when a cholecystoduodenal fistula, an ectopic gallstone, and Bouveret syndrome is suspected if the patient can tolerate oral contrast intake [12].

Endoscopy may be performed for both diagnostic and therapeutic purposes as a minimally invasive approach; however, utilization of endoscopy in extracting gallstones using a basket or a net is limited [20]. The primary goal is to eliminate the obstruction (if there is one) by removing the gallstone. Modern management focuses on the less invasive techniques, taking into consideration the patient's age, additional comorbidities, fistula and calculus size, as well as possible complications of more invasive methods such as surgery [21]. In cases of a large calculus, endoscopic removal is often not an option. Furthermore, fragmentation of gallstones with endoscopic graspers may result in fragments migrating to distal parts of the small bowel, leading to new obstruction. Therefore, surgery remains the treatment of choice [22]. A review of 1,001 cases concluded that simple enterolithotomy was both safe and effective in managing patients with gallstone ileus [23]. It is still a matter of debate whether cholecystectomy and repair of the fistula should be performed, due to spontaneous closure of fistulas in some cases [6, 15].

In case of the third patient, after an unsuccessful attempt to remove the floating gastric gallstone endoscopically, the clinicians decided to follow-up the patient having in mind small size of the non-obstructing calculus, poor clinical condition of the patient, and her comorbidities.

In conclusion, one should bear in mind that spontaneous cholecystoduodenal fistula is a rare condition, but that it can have complications such as Bouveret syndrome, gallstone ileus, and floating non-obstructive gallstones in the stomach. CT, MRI with MRCP, as well as the contrast X-ray swallow test can be very helpful in the detection of the bilio-enteric fistula and ectopic gallstones.

**Informed consent statement:** Consent was obtained from the patients for publication of this report and any accompanying images.

**Conflict of interest:** None declared.

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## Спонтана холецистодуоденална фистула – спектар компликација

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### САЖЕТАК

**Увод** Спонтана холецистодуоденална фистула је ретка компликација калкулозе жучне кесе. Опструкција црева је компликација која се деси у мање од 1% случајева. Риглерова тријада, коју чине пнеумобилија, дистензија танког црева и камен из жучне кесе инклавирани у цревној вијуги, патогномична је за билијарни илеус. Код 1–3% болесника се опструкција танког црева ектопичним калкулусом из жучне кесе нађе у дуоденуму и то се назива Бувереов синдром. Најређа компликација је неинклавирани камен који се налази у желуцу.

**Прикази болесника** Приказали смо три старије болеснице са константним абдоминалним болом и познатом калкулусом жучне кесе. Као први дијагностички избор урађен је нативни снимак абдомена и ултразвук, а после њих компјутеризована томографија (КТ). Код прве болеснице КТ и магнетна резонанца су показале знакове пнеумобилије, холецистодуоденалне фистуле и присуство камена у желуцу.

Гастродуоденоскопија је показала фистулу између булбуса дуоденума и жучне кесе, као и присуство неинклавираниог камена у желуцу, потврђеног ендоскопијом. Код друге болеснице КТ и гастродуоденоскопија баријумом показале су знакове пнеумобилије, холецистодуоденалну фистулу и два ектопична калкулуса који опструирају дуоденум – Бувереов синдром. Код трећег случаја имамо знакове Риглерове тријаде – типични знаци билијарног илеуса.

**Закључак** Спонтана холецистодуоденална фистула је ретко стање са могућим компликацијама, као што су Бувереов синдром, билијарни илеус или плутајући, неопструирајући камен у желуцу, као најређа компликација описана у литератури. КТ, магнетна резонанца и гастродуоденографија могу бити од велике помоћи у откривању билијарно-цревних фистула и камења из жучне кесе на ектопичним локализацијама.

**Кључне речи:** холецистодуоденална фистула; Бувереов синдром; Риглерова тријада; компликације