



**СРПСКИ АРХИВ**  
ЗА ЦЕЛОКУПНО ЛЕКАРСТВО  
**SERBIAN ARCHIVES**  
OF MEDICINE

Address: 1 Kraljice Natalije Street, Belgrade 11000, Serbia

+381 11 4092 776, Fax: +381 11 3348 653

E-mail: [office@srpskiarhiv.rs](mailto:office@srpskiarhiv.rs), Web address: [www.srpskiarhiv.rs](http://www.srpskiarhiv.rs)

**Paper Accepted\***

**ISSN Online 2406-0895**

**Case Report / Приказ болесника**

Dušan Šaponjski<sup>1,†</sup>, Aleksandra Đurić-Stefanović<sup>1,2</sup>, Aleksandra Janković<sup>1</sup>, Milica Mitrović-Jovanović<sup>1</sup>, Stefan Kmezić<sup>3</sup>, Slađana Mihajlović<sup>2,4</sup>, Ivan Popović<sup>5</sup>, Đorđije Šaranović<sup>6,7</sup>

**Spontaneous cholecystoduodenal fistula – spectrum of complications**

Спонтана холецисто-дуоденална фистула – спектар компликација

<sup>1</sup>Clinical Center of Serbia, First Surgical Clinic, Department of Abdominal Radiology, Belgrade, Serbia;

<sup>2</sup>University of Belgrade, Faculty of Medicine, Belgrade, Serbia;

<sup>3</sup>Clinical Center of Serbia, First Surgical Clinic, Clinic for Abdominal Surgery, Belgrade, Serbia;

<sup>4</sup>Dragiša Mišović Clinical Hospital Centre, Belgrade, Serbia;

<sup>5</sup>Stefan Visoki General Hospital, Smederevska Palanka, Serbia;

<sup>6</sup>Medigroup General Hospital, Belgrade, Serbia;

<sup>7</sup>University of Banja Luka. Faculty of Medicine, Banja Luka, Republic of Srpska, Bosnia and Herzegovina;

**Received: April 8, 2019**

**Accepted: June 10, 2019**

**Online First: June 20, 2019**

**DOI: <https://doi.org/10.2298/SARH190408072S>**

\***Accepted papers** are articles in press that have gone through due peer review process and have been accepted for publication by the Editorial Board of the *Serbian Archives of Medicine*. They have not yet been copy-edited and/or formatted in the publication house style, and the text may be changed before the final publication.

Although accepted papers do not yet have all the accompanying bibliographic details available, they can already be cited using the year of online publication and the DOI, as follows: the author's last name and initial of the first name, article title, journal title, online first publication month and year, and the DOI; e.g.: Petrović P, Jovanović J. The title of the article. *Srp Arh Celok Lek*. Online First, February 2017.

When the final article is assigned to volumes/issues of the journal, the Article in Press version will be removed and the final version will appear in the associated published volumes/issues of the journal. The date the article was made available online first will be carried over.

**†Correspondence to:**

Dusan ŠAPONJSKI

Clinical Center of Serbia, Pasterova 2, 11000 Belgrade, Serbia

E-mail: [saponjski.d@gmail.com](mailto:saponjski.d@gmail.com)

## Spontaneous cholecystoduodenal fistula – spectrum of complications

### Спонтана холецисто-дуоденална фистула – спектар компликација

#### SUMMARY

**Introduction** Spontaneous cholecystoduodenal fistula is a rare complication of the gallbladder calculosis. 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 duodenum, and it is called syndrome Bouveret. The rarest complication is floating non-obstructing gallstone trapped in a stomach.

**Outline of cases** We had three elderly female patients with persistent abdominal pain and known gallbladder calculosis in the patient's history. Plain radiography of thorax and abdomen and ultrasound were made as first choice and afterwards contrast-enhanced computer tomography (CT). In the first patient, CT and MRI showed signs of pneumobilia, cholecystoduodenal fistula and presence of the gallstone in the stomach. The iodine contrast X-ray swallow study revealed cholecysto-duodenal bulb fistula and floating calculus in the stomach, confirmed by endoscopy. In second patient with persistent abdominal pain, CT and barium swallow study 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 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 MRCP, as well as the contrast X-ray swallow study 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

#### САЖЕТАК

**Увод** Спонтана холецисто-дуоденална 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 је патогномична за билијарни илеус. У 1–3% пацијената се опструкција танког црева ектопичним калкулусом из жучне кесе нађе у дуоденуму и то се назива Боуверев синдром. Најређа компликација је неинклавирани камен који се налази у желуцу.

**Прикази болесника** Имали смо три старије пацијенткиње са константним абдоминалним болом и познатом калкулозом жучне кесе. Као први дијагностички избор је урађен нативни снимак абдомена и ултразвук, а након тога компјутеризована томографија. Код прве пацијенткиње компјутеризована томографија и магнетна резонанца су показали знакове пнеумобилије, холецисто-дуоденалне фистуле и присуство камена у желуцу. Гастроуденоскопија је показала фистулу између булбуса дуоденума и жучне кесе, као и присуство неинклавираниог камена у желуцу, потврђеног ендоскопијом. Код друге пацијенткиње компјутеризована томографија и гастроуденоскопија баријумом су показали знакове пнеумобилије, холецисто-дуоденалну фистулу и два ектопична калкулуса који опструирају дуоденум – Боуверев синдром. Код трећег случаја имамо знакове Риглерове тријаде – типични знаци билијарног илеуса.

**Закључак** Спонтана холецисто-дуоденална фистула је ретко стање са могућим компликацијама као што су Боуверев синдром, билијарни илеус или плутајући, неопструирајући камен у желуцу као најређа компликација описана у литератури. Компјутеризована томографија, магнетна резонанца, као и гастроуденографија могу бити од велике помоћи у детекцији билијарно-цревних фистула и камења из жучне кесе на ектопичним локализацијама.

**Кључне речи:** холецисто-дуоденална фистула, Боуверев синдром, Риглерова тријада, компликације

## INTRODUCTION

Spontaneous cholecystoduodenal fistula is a rare complication of the gallbladder calculosis, 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 elderly and it 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 it is known as the syndrome Bouveret[1,6,7]. The rarest complication is a floating non-obstructing gallstone trapped in a stomach, which can also lead to gastric outlet, persistent emesis and esophageal rupture (Boerhaave syndrome) as reported in a study of 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 calculosis 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 10mm in size hypodense ovoid lesion in the stomach with hyperdense rim - calculus (Figure 2a). Magnetic resonance (MR) examination with the MR cholangiopancreatography (MRCP) confirmed cholecystoduodenal fistula (Figure 3a, b) and 12 mm-sized calculus in the stomach (Figure 3c). Afterwards, an upper gastrointestinal X-ray study using the iodine contrast was performed, which revealed cholecysto-duodenal bulb fistula (Figure 4a) and 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 age, clinical condition and following comorbidities, it was decided only to follow-up the patient.

**Patient 2.** A woman, age 74, was admitted to the hospital with a 10-days history of abdominal pain and recurrent vomiting. Comorbidities included *diabetes mellitus*, cardiomyopathy and hypothyroidism. Previous abdominal ultrasound study showed stones in the gallbladder. Blood analyses showed no signs of leukocytosis and abnormal liver function, therefore endoscopic examination was performed.

**Patient 3.** An 83-year-old female was admitted in our department with repetitive vomiting, abdominal pain and distention. Gallstones were diagnosed five years earlier however, it was decided not to be operated due to the poor general and cardiac condition. Plain radiography of thorax and abdomen were 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 demonstrated distended small bowel loops with ectopic isodense, with hyperdense rim, 27-mm gallstone 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 happens in approximately 1% of all patients with the 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 basically the gallbladder “hangs,” which is why it is in a direct contact with 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 gender disparities in some parts of cardiovascular system anatomy has been already revealed [9].

The most common is cholecystoduodenal fistula (60%) and also described cholecystocolic, cholecystogastric and choledochoduodenal fistulas [10]. 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 could also migrate to all parts of the gastrointestinal tract without causing any symptoms. The most commonly obstructed is 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 half of the patients usually seen are 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 studies, CT, MRI with MRCP as well as ERCP and hepato-biliary scintigraphy could be used for diagnosis of cholecystoduodenal fistulas [16]. In our cases the patients had knowledge of cholelithiasis and were not operated. CT showed signs of pneumobilia, which indicated the existence of pathological communication between biliary tree and gastrointestinal tract [13, 17, 18]. Pneumobilia, gas in the gallbladder and cholecystoduodenal fistula were also confirmed by MRI and MRCP examination. MRI with MRCP is useful in detection of isoattenuating gallstones and in patients with the intolerance to oral contrast medium. Negi et al. successfully demonstrated that gallstones, pneumobilia, as well as cholecystoduodenal fistula can be visualized by this imaging modality [19].

As demonstrated in our cases, an oral contrast swallow study can be very helpful in confirming the presence of fistula, obstructing gallstone in 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 cholecystoduodenal fistula, 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 an extraction of the gallstones using a basket or 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 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 migrating fragments to distal parts of the small bowel leading to new obstruction. Therefore, surgery remains the choice of treatment [22]. A review of 1001 cases concluded that simple enterolithotomy was both safe and effective in managing a patient with gallstone ileus [23]. It is still controversial whether cholecystectomy and repair of fistula should be performed, due to spontaneous closure of fistulas in some cases[6, 15].

In case of patient number three, after unsuccessful try 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, bad clinical condition of the patient and comorbidities.

In conclusion, one should bear in mind that spontaneous cholecystoduodenal fistula is a rare condition with possible 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 study 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:** All authors have declared that no financial support was received from any organization for the submitted work, as well as that there are no other relationships or activities that could appear to have influenced the submitted work. All authors declared that all figures of our manuscript are original. The report fulfills the ethical guidelines of the most recent declaration of Helsinki (Edinburgh, 2000) and has received approval from the local ethics committee.

**REFERENCES**

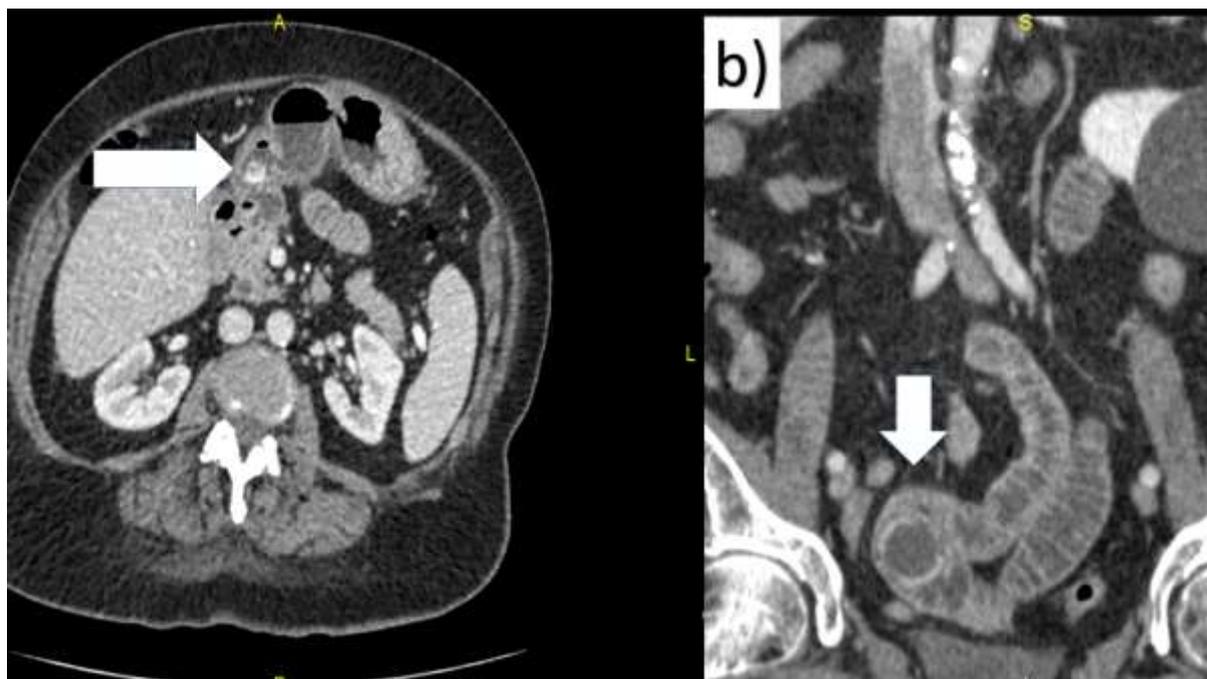
1. Gan S, Roy-Choudhury S, Agrawal S, Kumar H, Pallan A, Super P, et al. More than Meets the Eye: Subtle but Important CT Findings in Bouveret's Syndrome. *Am J Roentgenol* [Internet]. 2008 Jul;191(1):182–5. doi: 10.2214/AJR.07.3418. PMID 18562743
2. Nuño-Guzmán CM. Gallstone ileus, clinical presentation, diagnostic and treatment approach. *World J Gastrointest Surg* [Internet]. 2016;8(1):65. doi: 10.4240/wjgs.v8.i1.65 PMID 26843914
3. Roothans D, Anguille S. Rigler triad in gallstone ileus. *Can Med Assoc J* [Internet]. 2013 Oct 1;185(14):E690–E690. doi: 10.1503/cmaj.121432 PMID 23422449
4. Takahashi K, Kashimura H, Konno N, Nakagawa M, Kawahara Y, Munakata A, et al. Gallstone ileus with spontaneous evacuation: A case report. *J Gen Fam Med* [Internet]. 2018 Sep;19(5):173–5. doi: 10.1002/jgf2.196 PMID 30186731
5. Chang L, Chang M, Chang HM, Chang AI, Chang F. (2018, April 1). Clinical and radiological diagnosis of gallstone ileus: a mini review. *Emergency Radiology*. Springer New York LLC. PMID: 29147883 DOI: 10.1007/s10140-017-1568-5
6. Haddad FG, Mansour W, Deeb L. Bouveret's Syndrome: Literature Review. *Cureus* [Internet]. 2018 Mar 10; doi: 10.7759/cureus.2299 PMID 29755896
7. Tindell N, Holmes K, Marotta D. (2017). Gallstone ileus of the duodenum: An unexpected presentation of Bouveret's syndrome. *BMJ Case Reports*, 2017. PMID: 28790094 DOI: 10.1136/bcr-2017-220324
8. Modi BP, Owens C, Ashley SW, Colson YL. Bouveret Meets Boerhaave. *Ann Thorac Surg* [Internet]. 2006 Apr;81(4):1493–5. PMID 16564302
9. Teofilovski-Parapid G, Jankovic R, Kanjuh V, Virmani R, Danchin N, Prates N, Simic DV, Parapid B. Myocardial bridges, neither rare nor isolated-Autopsy study. *Ann Anat*. 2017 Mar;210:25-31. doi: 10.1016/j.aanat.2016.09.007. Epub 2016 Oct 21.
10. Luu MB, Deziel DJ. Unusual Complications of Gallstones. *Surg Clin North Am* [Internet]. 2014 Apr;94(2):377–94. doi: 10.1016/j.suc.2014.01.002 PMID 24679427
11. Wang L, Dong P, Zhang Y, Tian B. Gallstone ileus displaying the typical Rigler triad and an occult second ectopic stone. *Medicine (Baltimore)* [Internet]. 2017 Nov;96(45):e8541. doi: 10.1097/MD.00000000000008541 PMID 29137063
12. Masannat YA. A rare complication of a common disease: Bouveret syndrome, a case report. *World J Gastroenterol* [Internet]. 2006;12(16):2620. PMID 16688813
13. Vehbi H, Agirgun C. Abdominal CT Findings of Cholecystogastric Fistula. *SM J Gastroenterol Hepatol*. 2017;3(type I):1–2.
14. Watson RS, Folkers TE, Van Every MJ. A Multidisciplinary Approach to Management of Bouveret Syndrome. *Clin Med Res*. 2018;16(3-4):73–75. PMID: 30587561 DOI: 10.3121/cmr.2018.1430
15. Mavroidis VK, Matthioudakis DI, Economou NK, Karanikas ID. Bouveret syndrome-the rarest variant of gallstone ileus: a case report and literature review. *Case Rep Surg* [Internet]. 2013;2013:839370. doi: 10.1155/2013/839370. PMID 23864977
16. Chuah PS, Curtis J, Misra N, Hikmat D, Chawla S. (2017, April 1). Pictorial review: the pearls and pitfalls of the radiological manifestations of gallstone ileus. *Abdominal Radiology*. Springer New York LLC. PMID: 27896385 DOI 10.1007/s00261-016-0996-0
17. Chou CK (2016). Computed tomography demonstration of cholecystogastric fistula. *Radiology Case Reports*, 11(2), 70–73. DOI: 10.3121/cmr.2018.1430 PMID:30587561
18. Wong C, Crotty J, Naqvi S. Pneumobilia: A case report and literature review on its surgical approaches. *J Surg Tech Case Rep* [Internet]. 2013;5(1):27. doi: 10.4103/2006-8808.118616 PMID 24470847
19. Negi R, Chandra M, Kapur R. Bouveret syndrome: Primary demonstration of cholecystoduodenal fistula on MR and MRCP study. *Indian J Radiol Imaging* [Internet]. 2015;25(1):31. doi: 10.4103/0971-3026.150136 PMID 25709163
20. Chang KC, Chen WM, Wei KL (2016, November 1). Endoscopic treatment of Bouveret's syndrome in an extremely elderly patient with Holmium: YAG laser. *Annals of Saudi Medicine*, 36(6), 436–439. PMID:27920418 DOI:10.5144/0256-4947.2016.436

21. Pezzoli A, Maimone A, Fusetti N, Pizzo E. (2015). Gallstone ileus treated with non-surgical conservative methods: A case report. *Journal of Medical Case Reports*, 9(1). PMID:25971789 DOI:10.1186/1752-1947-9-15
22. Portincasa P, Di Ciaula A, De Bari O, Garruti G, Palmieri VO, Wang DQ H. (2016, January 1). Management of gallstones and its related complications. *Expert Review of Gastroenterology and Hepatology*. Taylor and Francis Ltd. PMID: 26560258 DOI: 10.1586/17474124.2016.1109445
23. Reisner RM, Cohen JR. Gallstone ileus: a review of 1001 reported cases. *Am Surg* [Internet]. 1994 Jun;60(6):441–6. PMID 8198337

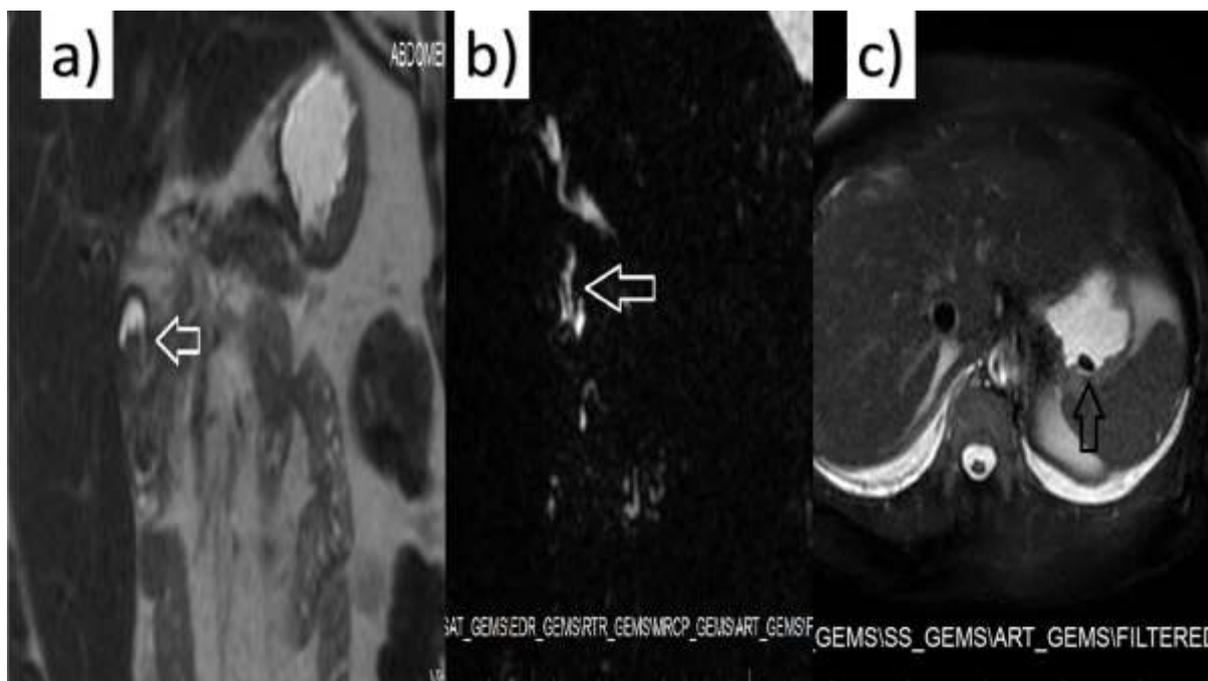
Paper accepted



**Figure 1.** a) Cholecystoduodenal fistula in patient 1 – arrow; b) a 4cm calculus in the duodenal cap and a 3 cm one in a thick-walled gallbladder – thin arrows, as well as a fistulous formation between duodenum and 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 T2 sequence on MRI – arrow; b) cholecystoduodenal fistula on MRCP – arrow; b) calculus in the stomach on T2 sequence on MRI – arrow