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Single-stage laparoscopic treatment of a cholecystoduodenal fistula with perforated small bowel ileus and fibrinous purulent peritonitis. A single case study

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Gallstone ileus is a rare complication of recurrent cholecystitis and one of the rarest causes of mechanical bowel obstruction, with an increasing incidence in the elderly. We present a case of multiple small bowel perforations of the jejunum due to gallstone obstruction and the incidental finding of high-grade appendiceal mucinous neoplasia of the appendix. To our knowledge, this is the first such case to be described and completely resolved by a laparoscopic approach.

An 83-year-old woman presented to our emergency department with severe vomiting, constipation, and deterioration of her general condition. A computed tomography scan revealed thickening of the gallbladder with a continuous aerobe to the duodenum, a mechanical obstruction of the small intestine at the transition to the ileum, and a thickened and calcified appendix. Due to a high suspicion of gallbladder perforation with the formation of a bilio-duodenal fistula, the patient underwent an emergency exploratory laparoscopy, which revealed a small bowel perforation in three segments with marked local fibrinous, purulent, and stercoral peritonitis of the left hemiabdomen. The gallstone was retrieved through the perforated small bowel, and a partial small bowel resection and a cecal wedge resection were performed laparoscopically. Small bowel continuity was restored with an anisoperistaltic side-to-side jejuno-jejunostomy. The patient was discharged on postoperative day eight.

Despite advances in imaging, gallstone ileus remains a diagnostic challenge. Because the disease occurs predominantly in elderly patients, gallstone ileus remains associated with high morbidity and mortality. It remains unclear from the literature whether the optimal surgical management of bilioenteric fistula is best resolved by a single-stage or a two-stage approach.

Keywords
cholecystolithiasis, cholecystoenteric fistula, gallstone ileus, small bowel perforation, Rigler's triad.

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Gallstone ileus is a rare complication of cholecystolithiasis and one of the rarest causes of mechanical ileus, with an increasing incidence in those over 65 years of age, with females predominating at a ratio of 4 : 1 to 16 : 1 [25, 32, 37]. The terminal ileum with 60 % and the jejunum with 16 % are described as the most common localization of the occluding calculus. Rarer stone locations are the stomach, duodenum (Bouveret syndrome), and colon [32]. Diagnosis is often delayed because symptoms can be intermittent, and supportive care can mask the disease. This, in addition to the fact that the disease often affects the elderly, is the reason for the increased morbidity and mortality, which are between 10 and 20 % [3, 29]. A national review from the United States by Halabi et al. of 3286 cases of gallstone ileus found an overall mortality rate of 6.67 %. In the case of partial small bowel resection, the mortality

rate was 12.87 %, almost three times higher than enterolithotomy alone (4.94 %) [3, 13, 29].

Gallstone ileus was first reported in 1654 by the Danish physician and anatomist Thomas Bartholin during autopsies. A systematic review was performed in 1890 by the Swiss surgeon Ludwig Georg Courvoisier, and the first detailed German-language collection of experienced case reports was published in 1902 by the German surgeon Dr. H. Karewski in the journal «Deutsche medizinische Wochenschrift» [9, 22, 35]. It is clear that this disease is not associated with a typical history or symptoms. The diagnosis is complex, and the indication for surgery is difficult to establish. Only the X-ray examination and the subsequent computed tomography (CT) diagnosis showed a high sensitivity and specificity for the presence of a gallstone ileus, which can be documented by Rigler's triad of aerobilia, small intestinal ileus, and ectopic gallstone [17, 18, 31, 39]. The need for surgery is easier to deduce from this, although the decision to treat the bilioenteric fistula at the same time is still under discussion [2, 13, 26, 28, 30].

Patient symptoms and clinical findings

An 83-year-old woman presented as an emergency because of torrential vomiting, constipation, and deterioration of her general condition. She reported no bowel movements and a loss of appetite for 5 days. She would only tolerate water and recurrent, sometimes foul-smelling vomiting. Last in the early morning hours. Abdominal pain was denied. Except for arterial hypertension, the initial diagnosis of diabetes mellitus (HbA1c 6.7 %) and medication with metoprolol, other diseases, and operations were denied.

On clinical examination, the abdomen was soft with slight tenderness in the left lower quadrant. There were no signs of guarding or peritonism. Auscultation revealed high-pitched bowel sounds.

Sonography showed the typical picture of a small bowel ileus with a dilated small bowel, the rope ladder phenomenon, and pendulum peristalsis. The wall of the gallbladder (GB) could not be completely delineated and was filled with concretions. The patient reported that GB stones had been known for years and had not caused any problems. Laboratory chemistry showed elevated C-reactive protein 147 mg/dl (< 5.0 mg/dL), and slightly elevated gamma glutamyl transferase 47 U/L (< 40 U/L) and alkaline phosphatase 142 U/L (35—104 U/L). Leukocytosis, hyperbilirubinemia, and elevated transaminases and lactate dehydrogenase were not observed. Elevated renal retention parameters were measured with creatinine of 1.37 mg/dL (< 0.9 mg/dL), urea of 140 mg/dL (< 50 mg/dL), and a reduced glomerular filtration rate of 36.9 ml/min (> 68 ml/min) consistent with chronic ileus.

A CT scan of the abdomen with intravenous contrast was performed because of a suspected mechanical ileus with elevated inflammatory blood parameters.

An inhomogeneously thickened, only partially filled GB with hypodense gallstones, presumably up to 2 cm in size, and intraluminal air collections («aerobilia») were found. A continuous transition to the duodenum was highly suspicious for a covered perforation with the formation of a bilio-duodenal fistula (Fig. 1). There was no free intra-abdominal air, no perivesical fluid collection.

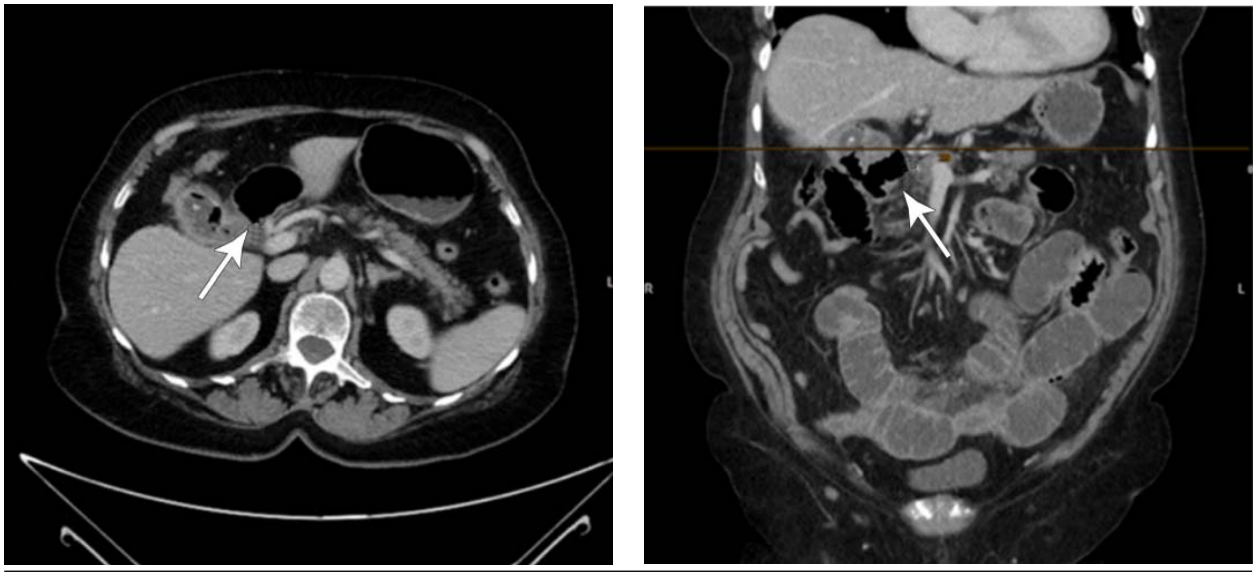

Figure 1. **Abdominal CT findings choleystoduodenal fistula**
(cystoduodenal fistula and aerobilia are indicated by arrows)

Furthermore, there was an obstruction of the small intestine with a long segment of small intestine up to 3 cm wide («small intestinal ileus»). In the right mid-abdomen, not far from the right lower pole of the kidney, there was a jump in the caliber of the ileum with subsequent starvation bowel and a collapsed colonic frame (Fig. 2). In combination with the suspected perforation of the duodenum, there was at least the suspicion that a gallstone had passed through and was blocking the intestinal passage at this point («missing ectopic gallstone»). Unfortunately, the gallstones in this patient were barely radiopaque. Furthermore, a tumorous, calcifying mass of the cecal pole or appendiceal base with a suspected mucocele differential diagnosis: cystadenocarcinoma was present (Fig. 3). An inflammatory correlate of the small intestine was not described. Thus, the CT image showed only two criteria of Rigler's triad; the ectopic GB stone could only be suspected indirectly. Despite the mild abdominal symptoms, we decided to perform exploratory laparoscopy due to increased inflammatory parameters and persistent inappetence with recurrent vomiting and lack of bowel movements. Antibiosis with ampicillin/sulbactam and metronidazole has already been initiated in the emergency room.

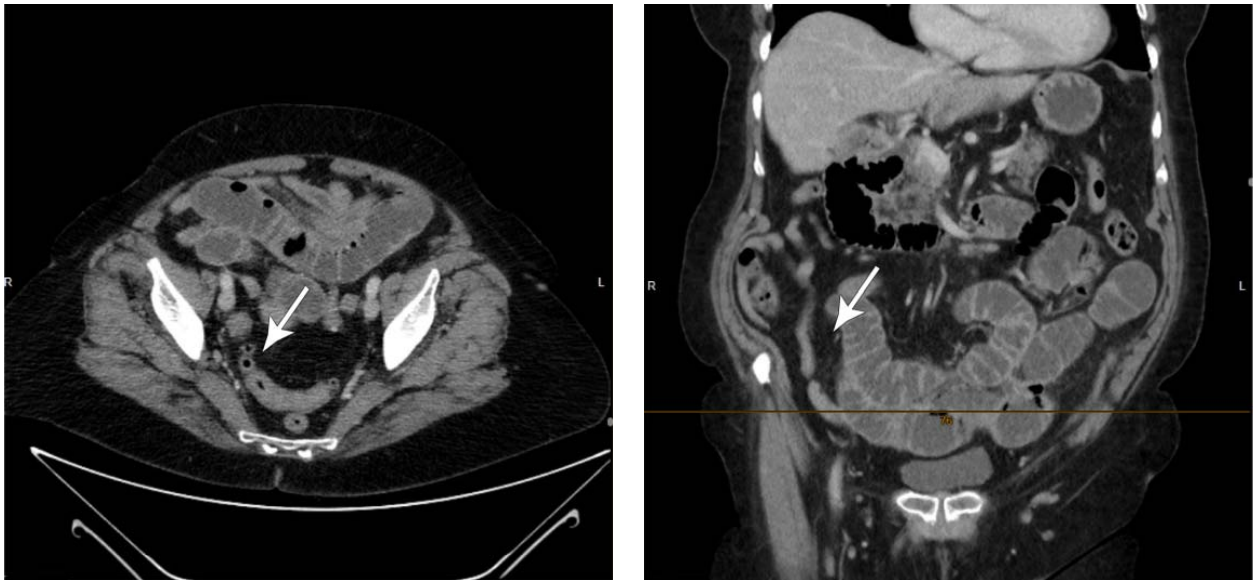

Figure 2. **Abdominal CT findings of small bowel ileus** (caliber leap of the proximal jejunum as indirect stone evidence, starvation bowel, small bowel ileus, and collapsed colon frame are indicated by arrows)

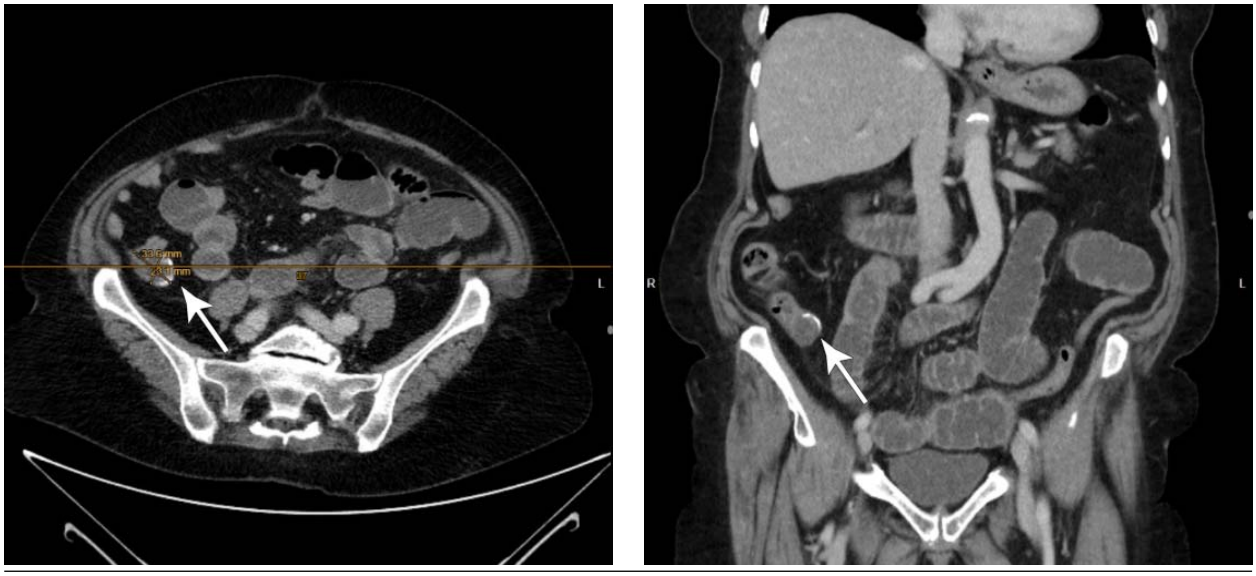

Figure 3. **Abdominal CT findings of suspicious appendiceal mass** (appendiceal mass are indicated by arrows)

Surgery and postoperative recovery

Intraoperatively, there was a small bowel perforation at three different sites within 15 cm with a marked localized fibrinous purulent incipient fecal peritonitis of the left hemiabdomen 160 cm from Treitz (Fig. 4—6). At first, no stone could be found. Only systematic instrumental palpation of the small intestine from the ileocoecum to the ligament of Treitz revealed an occlusion of the distal jejunum with prestenotic dilatation by a 2.0 × 2.5 cm GB stone 50 cm aboral to the devastating stone passage (Fig. 7). Resection of the coecum and extraction of the gallstone through the already perforated small bowel via minilaparotomy were followed by a partial small bowel resection of 30 cm of small bowel. Small bowel continuity was restored with an eanisoperistaltic side-to-side jejunojejunostomy. Extensive abdominal lavage and placement of a 24 Charrière drain in the left hemiabdomen were completed.

Figure 4. **Intraoperative finding of small bowel perforations** (surgical forceps in two perforation sites on the small intestine)

Figure 5. **Intraoperative finding small bowel perforation** (close-up perforation site is indicated by arrow)

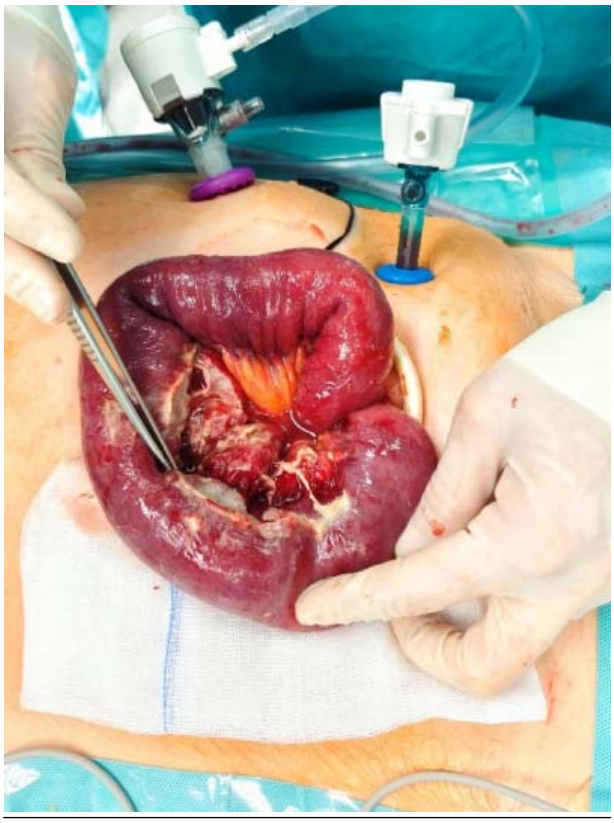

Figure 6. **Third perforation site on opposite side with fibrinous purulent peritonitis** (surgical forceps in third perforation of the small intestine

Figure 7. **Small ball with intraluminal gallstone 50 cm after the last perforation of the small bowel** (specimen small bowel with gallstone 50 cm)

Because of the ileus, the patient remained in the intensive care unit for two days for monitoring and supportive therapy. The antibiotic therapy was in accordance with the antibiogram of *Raoultella ornithinolytica*, *Enterobacter cloacae* complex, *Atopodium parvulum,* and *Streptococcus constellatus* cultured from the intra-abdominal specimen. The in-hospital esophagogastroduodenoscopy excluded a persistent enterobiliary fistula with normal mucosal findings up to the ligament of Treitz.

Pathology of the appendix revealed a high-grade appendiceal mucinous neoplasia pTis R0. The patient could be discharged on postoperative day 8 with an unremarkable course and a complete clinical recovery.

Discussion

The decision to perform a cholecystectomy should not be taken lightly, as the consequences of both having and not having the surgery can be severe and even fatal. In this particular case, the mortality rate is over 15 % due to the presence of ileus disease and a perforation of the small intestine resulting in purulent fibrinous peritonitis. Currently, health economics aims to reduce the number of cholecystectomies performed. However, pursuing this goal may lead us back to the 1990s, before the laparoscopic era, when procedures with fewer complications and shorter stays resulted in less hesitant indications for surgery. It is important to note that this approach carries a risk of significantly higher prevalence and associated morbidity and mortality from gallstone ileus, which can be triggered by recurrent, sometimes clinically occult, cholecystitis [4, 5, 16, 34, 36—38].

Gallstone ileus occurs in 0.15—1.5 % of cases with known cholecystolithiasis and is responsible for an obturation ileus in < 5 % of cases [13, 25, 29]

The recurrence rate is between 5—8 % [23] and occurs within 20—30 days in half of the cases [1, 10]. To our knowledge, only 13 cases with simultaneous small bowel perforation have been described in the literature [6, 12, 19—21, 24, 29, 33]. The enterolithotomy and repair of the perforation can rarely be solved laparoscopically. In our case, there was an unclear mass of the coecal pole and the base of the appendix, so an extensive median laparotomy would have been necessary. Therefore, in the case of overweight, we decided to first carry out the exploration laparoscopically, which is not undisputed, because a gallstone can easily be overlooked if there is no tactile possibility. Intraoperatively, there was devastating damage to the bowel due to the stone passage. Therefore, the decision to strive for a two-stage procedure was confirmed in order to get the patient out of the life-threatening situation as quickly as possible. The one-stage procedure is associated with increased morbidity and mortality and is based on experience from a gallstone ileus alone and without small bowel perforation [13]. A single center analysis of 29 cases of cholecystenteric fistula with and without gallstone ileus from 2010 to 2021 showed a gallstone impact on the intestinal system in 12 cases, which led to an ileus. A small bowel perforation was not described in any case. Five patients underwent laparoscopic procedures, with three undergoing a laparoscopic enterolithotomy and two requiring conversion to an open enterolithotomy. An open procedure was chosen for the other 7 patients, and a one-stage procedure was performed in 4 cases, which was associated with an increased operating time, length of stay, and morbidity. There is no mortality in the patient pool. In the case of a cholecystoenteric fistula and gallstone ileus, the authors recommend first relieving intestinal obstruction and avoiding fistula closure until the patient is in better condition [14]. The working group of Gonzalez-Uriquijo et al. came to the same conclusion; the significantly increased morbidity and mortality of the patients were decisive here [11]. The right procedure is still the subject of discussion today [15].

It is important to note that the persistence of a cholecystointestinal fistula carries the potential risk of retrograde cholecystitis and GB carcinoma [5, 7, 8, 38]. However, these are data from before the laparoscopic cholecystectomy era.

Nevertheless, in the majority of cases, there is a natural closure of the fistula opening, as was shown endoscopically in our case [27]. An increased carcinoma risk has not been described since then [1]. The relevant data was collected in the 1960s and 1970s. The number of cholecystectomies has increased significantly and therefore leads to such complications and malignant degeneration much less frequently. The question remains open as to whether the second treatment is actually necessary for our patient. Good surveillance of the patient and the high probability that she will survive the naturally healed fistula, along with an average current life expectancy of 83.4 years for women and another 9 years according to [1], the cohort life table in Germany, and the oldest documented patient being 76 years old when a recurrence occurred, most likely do not warrant repeat surgical intervention.

Declaration of interests

All authors declare that there is no conflict of interest and that they have no financial ties to disclose.

Ethics approval and written informed consents statements

Oral and written informed consent was obtained from the patient to publish the patient-related data in anonymized form.

Authors contributions

C.R.D. Demtröder, M. Murnik: surgery, treatment, literature research, literature review and draft of the manuscript; D. Dajchin, U. Giger-Pabst: literature research, literature evaluation and critical revision of important contents of the manuscript.

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Одноетапне лапароскопічне лікування холецистодуоденальної фістули з перфорацією тонкої кишки та фібринозно-гнійним перитонітом. Дослідження окремого випадку

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Жовчнокам'яна кишкова непрохідність є рідкісним ускладненням холециститу, що рецидивує, та однією з найрідкісніших причин механічної кишкової непрохідності, яка частіше трапляється в осіб похилого віку. Описано випадок множинних перфорацій порожньої кишки внаслідок обструкції жовчними каменями і випадкового виявлення апендикулярної муцинозної неоплазії апендикса високого ступеня. Наскільки нам відомо, це перший описаний випадок досягнення повного одужання за допомогою лапароскопічного підходу.

Жінка, 83 роки, звернулася до нашого відділення швидкої медичної допомоги із сильним блюванням, запором, погіршенням загального стану. На комп'ютерних томограмах виявлено потовщення жовчного міхура, ураження, спричинені аеробною інфекцією до дванадцятипалої кишки, механічну непрохідність тонкої кишки в місці переходу в клубову, потовщений і кальцифікований апендикс. У зв'язку з високою підозрою на перфорацію жовчного міхура з утворенням жовчно-дуоденальної нориці хворій проведено екстрену експлораторну лапароскопію, яка виявила перфорацію тонкої кишки в трьох відділах з виразним локальним фібринозно-гнійним і каловим перитонітом лівої половини живота. Жовчний камінь було вилучено крізь перфоровану тонку кишку. Лапароскопічно виконано часткову резекцію тонкої кишки та клинову резекцію сліпої кишки. Безперервність тонкої кишки відновлено за допомогою анізоперистальтичної бічної еюноеюностомії. Хвору виписано на восьму добу після операції.

Попри прогрес у візуалізації, жовчнокам'яна кишкова непрохідність залишається діагностичною проблемою. Оскільки захворювання трапляється переважно в пацієнтів літнього віку, жовчнокам'яна кишкова непрохідність залишається пов'язаною з високою захворюваністю та смертністю. З джерел літератури не зрозуміло, який підхід є оптимальним для хірургічного лікування біліоентеральної нориці — одноетапний чи двохетапний.

Ключові слова: холецистолітіаз, біліодигестивна фустула (нориця), жовчнокам'яна кишкова непрохідність, перфорація тонкої кишки, тріада Ріглера.

FOR CITATION

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