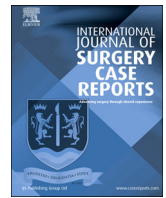




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Gallstone ileus after recent cholecystectomy. Case report and review of the literature

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ABSTRACT

INTRODUCTION AND IMPORTANCE: Gallstone ileus in cholecystectomized patients is very infrequent and when it happens shortly after surgery is even rarer. We report the case of a patient who presented Gallstone ileus few days after open cholecystectomy which has not been reported before in literature.

CASE PRESENTATION: A 52-year-old male with a history of recent open cholecystectomy was referred to our center due to a presumable surgical complication. During his hospitalization while trying to restart the oral route he presented abdominal pain and nausea. He evolved toward a bowel obstruction. We suspected gallstone ileus based on medical history as well as preoperative image study. We confirmed the diagnosis using a Computed Tomography. Surgical management was performed and a large gallstone was extracted from the bowel. The patient progressed favorably and was discharged. He was asymptomatic during the follow-up.

CLINICAL DISCUSSION: Cholecystectomized patients who have been reported with Gallstone ileus demonstrate different pathophysiological mechanisms or extraordinary presentations. This case describes a unique presentation illustrating relevant aspects of this pathology such as showing that acute cholecystitis can be its clinical manifestation or that it could happen after a cholecystoenteric fistula is found during a cholecystectomy.

CONCLUSION: Gallstone ileus in cholecystectomized patients is very rare. Clinical suspicion remains the cornerstone of diagnosis.

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1. Introduction

Gallstone ileus (GI) is a complication in 0.3% to 0.5% of patients with cholelithiasis [1,2]. It is a mechanical intestinal obstruction due to the impaction of one or more gallstones within the gastrointestinal tract [3]. In most cases, gallstones enter the intestinal lumen through cholecystoenteric fistulas [2–4].

In patients with a history of cholecystectomy this pathology is extremely rare [5,6].

This work is reported in line with SCARE criteria [7].

2. Presentation of case

A 52-year-old male, single, with elementary schooling only, denied chronic degenerative diseases. No other significant medical history.

Four days earlier, he underwent emergency surgery for acute cholecystitis in a private hospital, open cholecystectomy and intestinal repair were performed with findings of gallbladder empyema and 2 × 1 cm cholecystoduodenal fistula and thereafter he was transferred to a public hospital where continued to postsurgical care. He had biliary output through the drain placed in surgery, so there was suspicion of Bile Duct Injury and he was referred to us for diagnosis and management.

The patient was admitted at our hospital presenting stable vital signs, body mass index 33.3 Kg/m². Awake, not jaundiced. Abdomen with right subcostal 8 cm wound and a surgical drain type Saratoga with biliary output of approximately 900 mL per day since its placement; no peritoneal irritation. Blood tests only showed 15,100 leukocytes / mL; Neutrophils 90%; Albumin 2.5 g / dl, normal bilirubin and transaminases levels.

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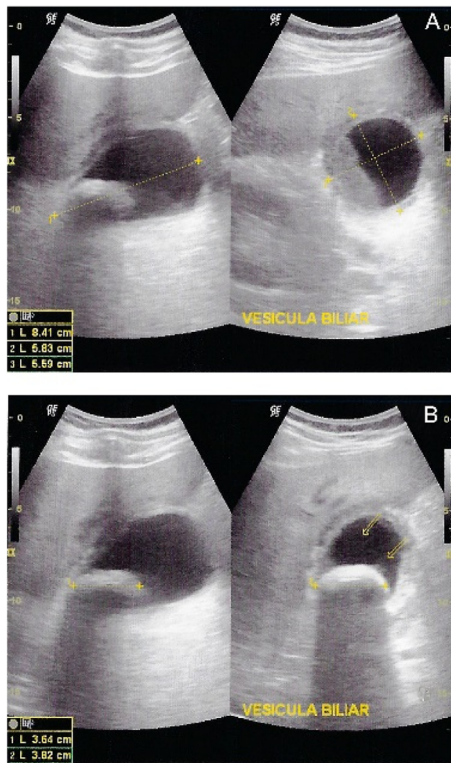


Fig. 1. (A) Ultrasound images showing gallbladder dimensions, the presence of a gallstone and biliary sludge. (B) Gallstone's dimensions and its acoustic shadow.

We begin treatment with fasting, intravenous hydration and antibiotic. Total Parenteral Nutrition (TPN) were added on the second day. Magnetic Resonance Cholangiopancreatography (MRCP) was performed on the fourth day and reported a bile duct without anatomical alterations and no other findings. Saratoga's output progressively decreased and stopped on the fifth day. On the sixth day with clinical improvement the oral route was restarted and he presented nausea, so he had to continue fasting. Two days later the oral route was restarted again and after eight hours he presented distension and abdominal pain, two episodes of gastrobiliary vomiting and failure to pass flatus.

We re-evaluated the case. He had presented recurrent acute cholecystitis for about 18 months. Three weeks before cholecystectomy, he presented pain in his right hypochondrium of moderate to severe intensity, so he went to a private medical service where an abdominal ultrasound was performed diagnosing acute cholecystitis. He was offered surgical management which he did not accept. He self-medicated himself with analgesics such as acetaminophen and diclofenac to provide temporary pain relief. Subsequently, there was an increase in abdominal pain, as well as fever and several episodes of gastrobiliary emesis, so he sought medical attention again and had a cholecystectomy.

We asked his family for the referred ultrasound and in it, we could observe the presence of a large gallstone (Fig. 1).

We consider post-surgical adhesions and gallstone ileus as probable diagnoses. Supported by a Computed Tomography we confirmed that it was Gallstone Ileus (Fig. 2).

We requested a surgical time and found a 4 × 3 cm gallstone in his ileum (Fig. 3A–C).

Intentionally, we went back to review the MRCP done days before and a presence of a gallstone could already be observed (Fig. 4).

In the postoperative, he was managed with fasting, antibiotics and TPN for seven days, after which the oral route was restarted

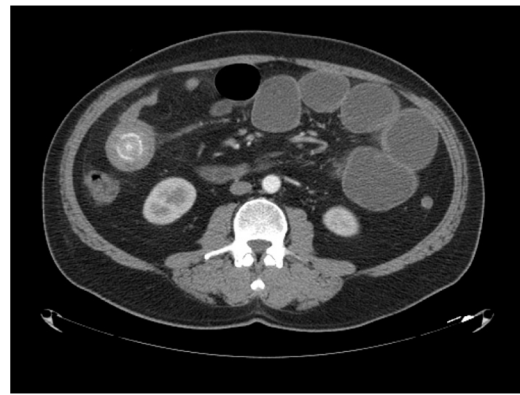


Fig. 2. Preoperative Computed Tomography demonstrating a lost gallstone with calcified edges in the right hemiabdomen.

without problems. Due to favorable clinical progress, he was discharged on the eleventh day. During follow-up on the 7th day and five weeks after his discharge, he was asymptomatic.

3. Discussion

GI causes small bowel obstruction in 1%–4% of patients under 65 years of age [4]. Mainly affecting women and elderly patients [3,4,8]. Mortality rate is from 6.7%–22.7% [9].

Pathophysiologically, recurrent inflammation episodes in a gallbladder with stones causes adhesions with the surrounding structures mainly the Gastrointestinal (GI) tract, gallstones originate an increase pressure within gallbladder causing decreased arterial and venous flow, in addition to mucosa inflammation, ischemia and erosion, first of the gallbladder wall, and then spreading towards the adhered intestinal segment, giving rise a fistula formation [3,4,10]

In more than 90% of cases, the stones gain access to the gastrointestinal tract through cholecystoenteric fistulas [11] and the cholecystoduodenal one is the most frequent [12]. Cholecystectomized patients who have been reported with GI, demonstrate different pathophysiological mechanisms or extraordinary presentations (Table 1).

It is important to note that not all patients with cholecystoenteric fistulas are accompanied by GI. González Urquija et al. [23] report in their series (n = 15) that occurs in 33.3% while Tandon et al. [24] report 8.6% (n = 35).

The size of the stone and the luminal diameter are the two main factors determining whether the stone will become impacted [25]. Most stones less than 2 cm could pass spontaneously through a normal gastrointestinal tract unless the patient has history of any alteration that causes narrowing of the intestinal lumen [3]. Stones ≥ 2.5 cm are most likely to be impacted [12]. The average size of obstructing gallstones is 4 cm [2,12] and 69% is caused by a unique stone [26]. Cylindrical or faceted stones causing obstruction increases the possibility of recurrence [10,27]. The most common impaction site of the gallstone is the ileum (50.0%–60.5%), jejunum (16.1%–26.9%), duodenum (3.5%–14.6%) and colon (3.0%–4.1%), in order [12]. Bouveret's syndrome is a rare variant which occurs by the impaction of a gallstone in the proximal duodenum or in the distal stomach causing an obstruction of the gastric outlet [28].

Finding cholecystoenteric fistula during cholecystectomy - an event reported in 1.4% [24] - should warn the possibility that GI happens, especially if there is evidence that a stone exists as occurred in this case.

The typical clinical manifestation is gastrointestinal obstruction characterized by abdominal pain and distension, anorexia, nausea, vomiting, obstipation, and hydro-electrolyte imbalance with

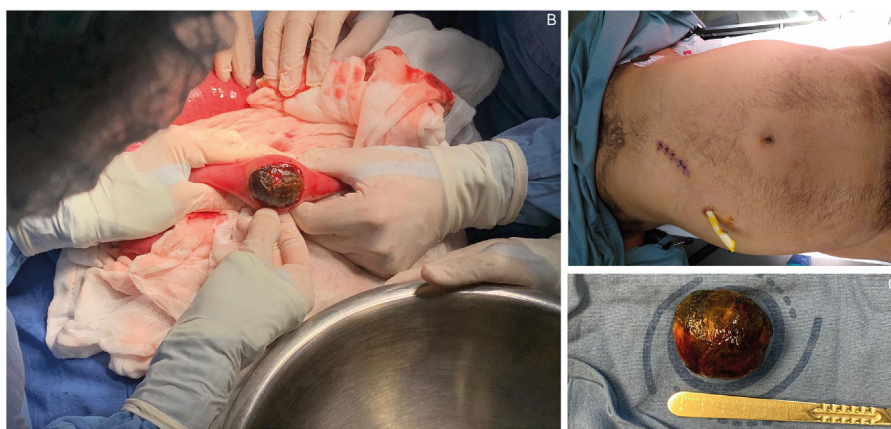


Fig. 3. (A) Recent open cholecystectomy. (B) Gallstones is removed by enterolithotomy. (C) Gallstone measured.

Table 1
Cases of gallstone ileus in colecistectomized patients.

Possible mechanism or unusual presentation	Authors	Observations
Gallstone passes through the Vater papilla after an endoscopic procedure (ERCP and sphincterotomy)	Fedele et al [5]. Halter et al [13]. Narasimhan et al [14]. Ivanov et al [15].	OC 30 years before. T: 7 months. C 7 years before. T: 7 days. LC 7 years before. T: 2 years.
Gallstones spilled into the peritoneal cavity during cholecystectomy that are not retrieved cause inflammation and erosion of the intestinal wall until they gain access to the intestinal lumen.	Zamora et al [16]. Draganic et al [17]. Habib [18]	LC 6 months before. A stone partially lodged in the transverse colon. LC 1 month before. LC 2 months before. The stone eroded the jejunum. LC 8 years before. A stone gain access into the intestinal lumen through a Meckel's Diverticulum.
Gallstone lodged in a diverticulum that then moved to cause obstruction.	Saedon et al. [19]	OC 24 years before.
Patient with history of CS where the remnant contained a large stone, which later caused stump cholecystitis and formation of a fistula.	Teelucksingh et al [20].	OC 1 year before.
Clinical and radiological presentation suggestive of intussusception.	Prasad et al [21].	OC 30 years before.
Enlargement of a stone in the site of an ancient anastomosis.	Papavramidis et al [22].	C 3 years before. A 44 year old man.
Fistula between the bile ducts and intestine.	Mansson [6].	C 40 years before.

C: Cholecystectomy; LC: Laparoscopic Cholecystectomy; OC: Open Cholecystectomy; SC: Subtotal Cholecystectomy; ERCP: Endoscopic Retrograde Cholangiopancreatography; T: Time from endoscopic procedure to ileus.

a sudden or intermittent onset which is characterized by abdominal pain that alternates with asymptomatic periods until complete obstruction occurs [3,4,10,12].

In this case, it is possible that the clinical presentation of acute cholecystitis that was the indication to cholecystectomy it might have actually been one of the GI manifestations, due to that in 10%–30% of cases acute cholecystitis could occur at the time of bowel obstruction, as described by Nuno Guzman et al. [3]. In one series, biliary colic is reported as initial manifestation what motivated to surgery in one of its patients, who was re-operated shortly after and it was found to be gallstone ileus [26].

Physical examination and laboratory tests are not specific [3,4]. The Mordor triad (history of lithiasis, signs of acute cholecystitis and sudden onset of intestinal occlusion) in an elderly female patient should arouse the diagnostic suspicion [4,10]. Preoperative diagnosis is made in 30%–75% of the cases [3].

The sensitivity of plain abdominal radiography for the diagnosis is between 40% and 70%. Rigler's triad (pneumobilia, ectopic radiopaque gallstones and intestinal loop dilation) is pathognomonic, however, it occurs only in 14%–53% of cases [29,30].

Computed Tomography (CT) has a sensitivity of 93% and specificity of 100% [30,31]. Diagnostic criteria are: small bowel obstruction, ectopic gallstones with calcified edges or fully calci-

fied, and an abnormal gallbladder [30]. Non-contrast enhanced CT is a good option in patients who have renal impairments or risk of contrast allergy, and has the benefit of easier location of the ectopic stone regardless of the degree of calcification [29,30].

Magnetic resonance imaging does not play a role in the acute setting as it is more time consuming and less readily available than CT [29]. In this case, the diagnosis could have even been made earlier if we had noticed the gallstone before (Fig. 4) in MRCP, however, the truth was that we did not suspect it.

Once the diagnosis is made, the patient's hydroelectrolytic and metabolic disorders must first be corrected [2,32]. The treatment is the surgical approach and the options are usually three. The one-stage procedure consists of enterolithotomy, cholecystectomy, and closure of the fistula, all during a single surgical time, entails greater morbidity [33] and it is reserved for younger and clinically stable patients [34]. Mallipedi et al. compare 113 cases vs. 14 cases of patients with and without cholecystectomy respectively, and suggest that the one-stage procedure is safer than what has been historically reported [35]. The two-stage procedure consists of performing an enterolithotomy first, and after 4–6 weeks, cholecystectomy and closure of the fistula are performed [33]. Enterolithotomy at the antimesenteric border is currently the most widely used procedure [33,34]. The main drawback of solely

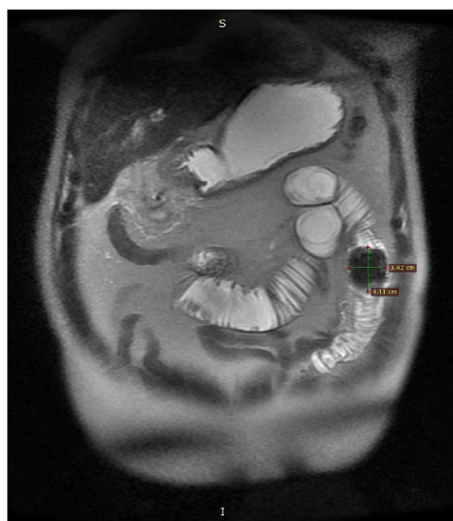


Fig. 4. Magnetic Resonance Cholangiopancreatography image showing the presence of gallstone.

doing enterolithotomy is that fistula persistence may cause recurrent cholangitis, sepsis, or recurrent gallstone, the latter occurring between 4.7% and 17%, and in these cases 57% occurs within the first six months of the original surgery [36]. Spontaneous closure of cholecystoenteric fistulas that persist after management with enterolithotomy, has been reported [37].

4. Conclusion

Gallstone ileus is a rare disease and its presentation in patients with a history of cholecystectomy is even rarer. Preoperative diagnosis is challenging and suspicion is the key. Unusual presentations such as the presented case provide more knowledge about the subject and it is important to know about them.

Declaration of Competing Interest

The authors report no declarations of interests.

Funding

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Ethical approval

There was no need for ethical approval.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution

María Paulina Sesman Bernal: performed surgery, data analysis, and manuscript revision.

Andrea Carolina Pérez Burguete: Data collection, data analysis, and manuscript revision.

Antonio Pérez Sánchez: performed surgery, data analysis, and manuscript revision.

Uriel Martínez Segundo: Conceptualization, Investigation, writing- original draft, writing-review an editing.

Registration of research studies

Not applicable.

Guarantor

Uriel Martínez Segundo and Antonio Pérez Sánchez.

Provenance and peer review

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