



Case report

Gallstone ileus associated with cholecystogastric fistula: Case report, diagnosis and surgical treatment

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ABSTRACT

Introduction: Gallstone Ileus is a rare complication of cholelithiasis, associated with multiple episodes of cholecystitis, with the formation of adhesions and fistulas between the gallbladder and adjacent organs. Its diagnosis is difficult, requiring complementary imaging tests such as computed tomography or radiography.

Presentation of case: Female patient, with intestinal obstruction for 7 days, associated with abdominal pain and previous episodes of pain in the right hypochondrium for 3 months. Abdominal CT scan identified aerobilia, gallstone impacted in the ileocecal valve and small loop dilatation, in addition to a probable cholecystogastric fistula. Opted for exploratory laparotomy, enterolithotomy and fistula correction in one surgical time.

Discussion: Gallstone ileus is rare among the complications of cholelithiasis, in addition to the fact that cholecystogastric fistula is associated with gastric pylorus obstruction and not impaction on the ileocecal valve. Imaging tests are useful to complement the diagnosis, and if Rigler's triad is present, the suspicion of gallstone ileus is increased. The presence of fistula between the gallbladder and stomach presents a frequency between 0 and 13.3%. There is no gold standard treatment for gallstone ileus, but surgery options for each type of patient and severity level.

Conclusion: There is no definitive protocol for optimal surgical treatment for biliary ileus, but the possibility of enterolithotomy associated with cholecystectomy and fistula correction can be evaluated in selected patients.

1. Introduction

Gallstone ileus is a rare complication of cholelithiasis, occurring in 0.3-0.5% of patients with the disease, occurring due to chronic inflammation after multiple episodes of cholecystitis and subsequent fistula formation with adjacent organs, such as the duodenum, colon and stomach [1]. It is defined as an obstruction of the gastrointestinal transit due to gallstones.

Despite the suggestive clinical suspicion due to the history of multiple attacks of cholecystitis and acute obstructive abdomen, the diagnosis is difficult and needs to be complemented with radiological exams. Its treatment is surgical, with options of approaches for each type of patient [2].

This case follows 2020 SCARE guidelines for reporting of cases in

surgery [3].

2. Presentation of case

A 41-year-old female patient was admitted to the emergency department due to a complaint of diffuse abdominal pain for 15 days, evolving to stopping of feces and flatus elimination during seven days. The patient had associated nausea and vomiting since the onset of symptoms, with progression of pain intensity. The patient also reported pain crises in the upper abdomen, mostly located in the right hypochondrium and epigastrium in the last three months, but not demanding analgesia or urgent and emergency care services, without any previous history of obstruction or difficult to difficulty in evacuation. On physical examination the patient was hemodynamically stable, with globe

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abdomen, diffusely painful to palpation, without peritoneal irritation. Laboratory and imaging tests were requested.

Laboratory tests showed hemoglobin 15.11 g/dL, leukocytes 23,510, 78% neutrophils. There were no alterations in total or fractional bilirubin, alkaline phosphatase or gamma-GT. Computed tomography initially showed distension of the small bowel loops throughout its length, associated with the presence of air in the intrahepatic bile ducts and gallbladder, in addition to a possible fistula between the gallbladder and the gastric antrum. In ileocecal, we identified a rounded image in valve topography, with defined contours, compatible with gallstones (Fig. 1).

It was decided to start initial clinical measures for the intestinal obstruction, such as passage of a nasogastric tube for abdominal decompression, followed by exploratory laparotomy. Gallbladder with thin walls was identified with firm adherence to the gastric antrum (showing cholecystogastric fistula) and gallstone impaction point near the ileocecal valve (Fig. 2), followed by longitudinal ileolithotomy in the antimesenteric edge of the distal ileum with transverse milking maneuver for gallstone removal, of approximately 3.1 cm in the largest diameter (Fig. 3), following by transverse enterorrhaphy in a single plane. Due to favorable clinical conditions, an option was made for cholecystectomy and gastrorrhaphy with freshened margin at the same surgical time using polypropylene 4-0, both procedures performed without complications.

After surgery, the patient was referred to the ward, evolving with improvement in postoperative pain and no other complaints and was discharged from the hospital after 10 days, referred to the outpatient surgery service for follow-up. After 3 months, the biopsy evidenced only chronic cholecystitis.

3. Discussion

Gallstone ileus is considered a rare complication caused by consecutive episodes of acute cholecystitis, causing a fistula between the gallbladder and the main gastrointestinal tract, with subsequent migration of stones through it, although at the time of diagnosis, only 27% patients had a history of previous acute cholecystitis [2]. This occurs after the formation of adhesions after inflammatory episodes, with multiple erosions and local ischemia, later evolving to a fistula, among the most common: cholecystoduodenal, colonic and gastric [4]. It has a mortality rate of 7-30% and is associated with unfavorable clinical conditions, such as advanced age, comorbidities or obesity [5,6].

After gallstone migration, its point of impact depends on the location of the fistula with the gastrointestinal tract. The most common fistula is the cholecystoduodenal fistula, in which the gallstone migrates in favor of peristalsis and impacts the ileocecal valve [7]. In case of cholecystocolonic fistula, the gallstone usually obstructs the sigmoid colon [8]. Fistulas with stomach mostly obstruct the gastric pylorus, causing Bouveret's syndrome, and it can also occur in caeculi from the duodenum that migrates against peristalsis [9]. In very rare and poorly documented cases, the gallstone that migrates from a cholecystogastric fistula surpasses the pylorus with few or no symptoms, obstructing in another point of the intestinal transit, as in the case reported [2,10]. It is associated with the fact that this fistula presents only 0-13.3% among all variations of fistulas between the biliary tract and the gastrointestinal tract [11].

The clinical signs presented by the patient are variable. As the pathophysiology of the ileum consists of multiple episodes of cholecystitis, it is expected that the patient will present pain in the right hypochondrium as a symptom prior to the obstruction. However, in

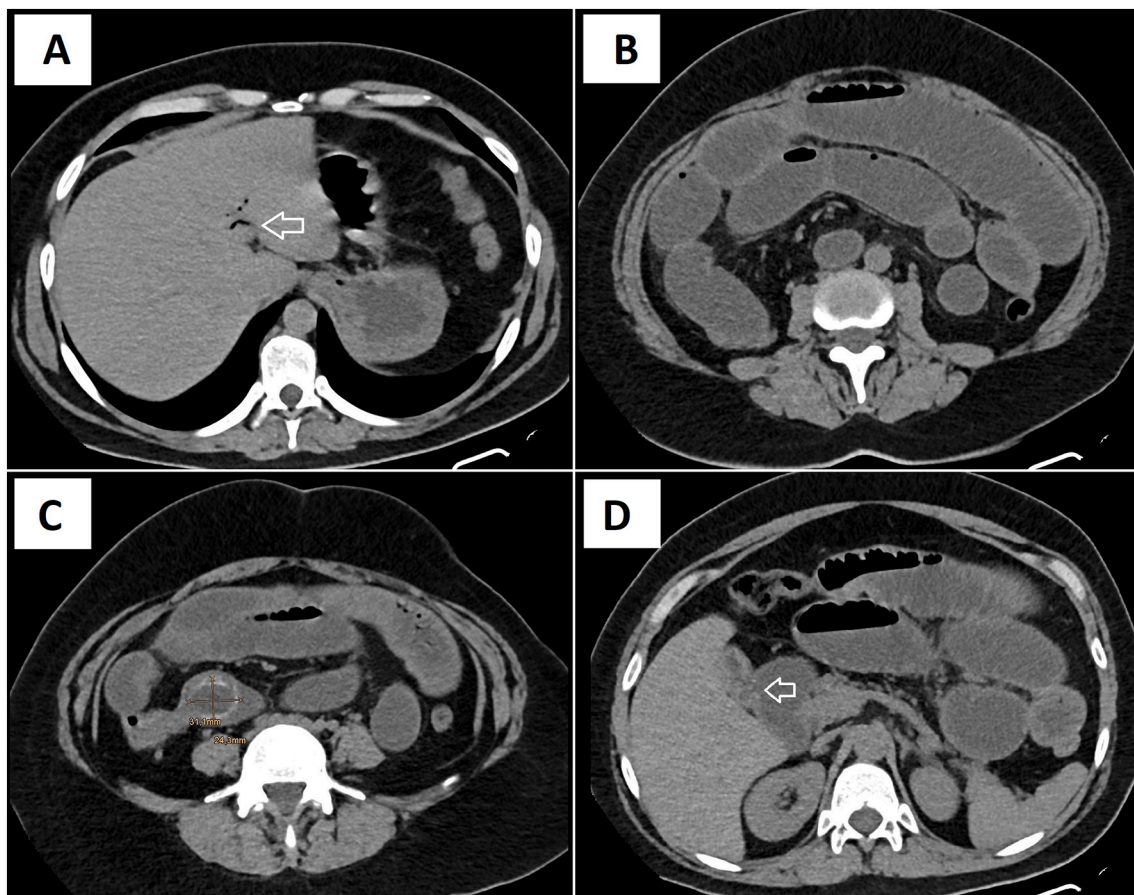


Fig. 1. Abdominal CT scan showing the Rigler's Triad: aerobilia (A), dilation of intestinal loops (B) and ectopic gallstone measuring approximately 31.1 × 24.3 mm (C). There is also the presence of a fistula between the gallbladder with thickened walls and the gastric antrum (D).

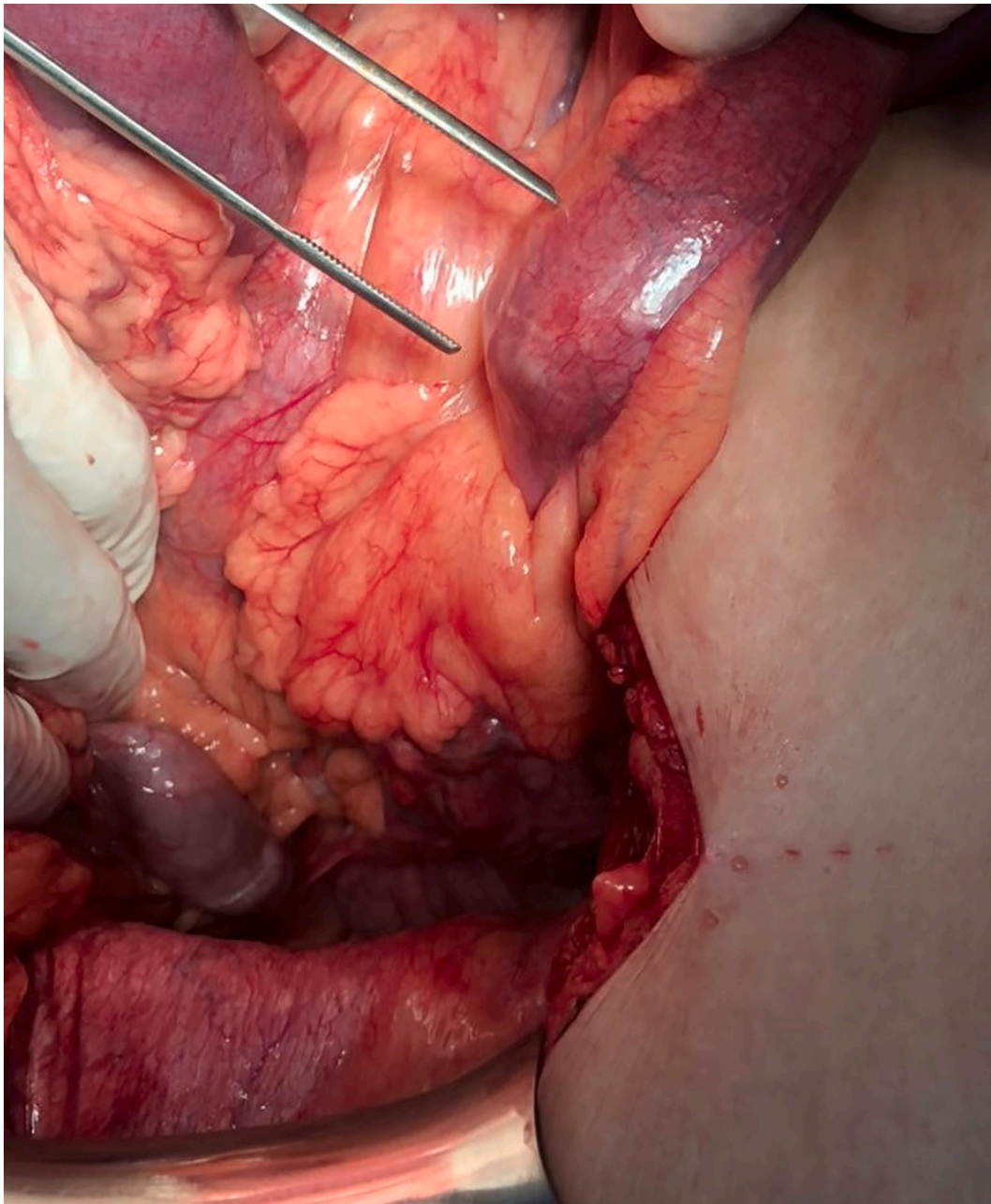


Fig. 2. Exploratory laparotomy view showing impacted gallstone in ileocecal valve.

retrospective surveys, only 27% of patients with gallstone ileus have a history of cholecystitis or a confirmed diagnosis of cholelithiasis [2]. Symptoms in urgency are limited to nausea, vomiting, abdominal distension and pain, usually diffuse, with the possibility of being intermittent due to movement of the ectopic gallstone in the gastrointestinal tract [12]. For definitive impact and total obstruction, the gallstone must be greater than 2-2.5 cm in diameter [5].

Complementary laboratory tests can help, but are not specific, presenting leukocytosis due to obstruction. Imaging exams are necessary to elucidate cases where the clinic is doubtful. Abdominal radiography may show specific signs of gallstone ileus, but CT is considered the gold standard for diagnosis [5]. On examination, 3 main findings are sought: aerobilia (due to the presence of air in the gastrointestinal tract through the fistula with the gallbladder), loop distension due to the obstruction, and the ectopic gallstone [13]. There are divergent values regarding the finding of all elements of Rigler's triad in patients, ranging from 11.11%

to 77.78% [5,14]. Cholangioresonance can be useful to assess the fistula and biliary tract, however, due to the clinical status of the patient, this exam is not the most appropriate, even more than the fistula, even though it can be seen in other exams, it does not always follow the biliary ileus [15].

There are three treatment options, depending on the patient: conservative, urgent and one-time definitive treatment. Conservative treatments are not largely studied and are used when the patient is in a condition of partial obstruction and with a stone smaller than 2.5 cm [6]. Treatment is surgical in most cases. In cases where the patient presents hemodynamic instability, over 70 years of age, low respiratory reserve, ASA III or IV status or any other signs of severity, we can use urgent surgery initially [16]. Longitudinal enterolithotomy is used close to the impactation site for excision, followed by transverse rafia [17]. Laparoscopic access is preferable, although in cases where the equipment is not available or where the technique is contraindicated, the



Fig. 3. Gallstone after removal, measuring approximately 3 cm in diameter.

laparotomic route is accepted [18]. It is noteworthy that it is not always easy to identify the place of obstruction [6]. However, removing the patient from the state of obstruction does not eliminate the origin of the obstruction, and new episodes of gallstone ileus may occur, suggesting a new surgery to correct the fistula and cholecystectomy [19].

The other option for surgical treatment is definitive one-step surgery, in which enterolithotomy is performed for clearance, cholecystectomy and fistula correction [20]. However, this treatment should be chosen in patients who present a favorable clinical condition for a more extensive surgery, as in the case reported here. Currently, there is no definition or consensus as to which is the best option for treating gallstone ileus, which must be evaluated on a case-by-case basis.

4. Conclusion

Gallstone ileus, despite being rare, should be investigated as a possible cause of obstructive abdomen, since its diagnosis is difficult, requiring additional tests for a definitive diagnosis. Cholecystogastric fistula, stone migration through the pylorus with few symptoms and its

impact in the distal ileum, although rare, is shown to be possible and should be considered as a possibility during the case evaluation. A thorough evaluation of the clinical status of the patient is necessary to choose the appropriate treatment, which may be just a surgery to remove the patient from an emergency situation, as well as a definitive treatment for the fistula.

Informed 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.

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We do not have any funding source, this manuscript is just a case report, not a research.

Ethical approval

As the manuscript is not a research study, we only have the patient consent for writing and others forms of publication. Also, the ethical approval for this case reports has been exempted by our institution.

Author contribution

Josias Torres, Ana Elisa and Victor Ripardo made contributions to conception and design, collected the patient details and wrote the paper. Juan Rodriguez, Magnum Pereira made contributions to patient management. Juan Rodriguez and Decius Cunha critically revised the article. All authors read and approved the final manuscript.

Research registration

N/A.

Guarantor

Juan Eduardo Rios Rodriguez.

Declaration of competing interest

We do not have any conflicts of interests.

References

- [1] P.A. Clavien, J. Richon, S. Burgan, A. Rohner, Gallstone ileus, *Br. J. Surg.* 77 (1990) 737–742, <https://doi.org/10.1002/bjs.1800770707>.
- [2] A.A. Ayantunde, A. Agrawal, Gallstone ileus: diagnosis and management, *World J. Surg.* 31 (2007) 1292–1297, <https://doi.org/10.1007/s00268-007-9011-9>.
- [3] for the SCARE Group, R.A. Agha, T. Franchi, C. Sohrabi, G. Mathew, The SCARE 2020 guideline: updating consensus Surgical CAse REport (SCARE) guidelines, *Int. J. Surg.* 84 (2020) 226–230, <https://doi.org/10.1016/j.ijssu.2020.10.034>.
- [4] S.-U.-N. Abbasi, D.B. Khan, K. Khandwala, R. Raza, W.A. Memon, Cholecystocolonic fistula, *Cureus* 11 (2019), e4874, <https://doi.org/10.7759/cureus.4874>.
- [5] L. Chang, M. Chang, H.M. Chang, A.I. Chang, F. Chang, Clinical and radiological diagnosis of gallstone ileus: a mini review, *Emerg. Radiol.* 25 (2018) 189–196, <https://doi.org/10.1007/s10140-017-1568-5>.
- [6] M. Jakubauskas, R. Luksaite, A. Sileikis, K. Strupas, T. Poskus, Gallstone ileus: management and clinical outcomes, *Medicina (Kaunas)* 55 (2019), <https://doi.org/10.3390/medicina55090598>.
- [7] S.G. Bruni, M. Pickup, D. Thorpe, Bouveret's syndrome—a rare form of gallstone ileus causing death: appearance on post-mortem CT and MRI, *BJR Case Rep.* 3 (2017) 20170032, <https://doi.org/10.1259/bjrcr.20170032>.
- [8] G.-U. Mauricio, H.-G. David Eugenio, Q.-F. Enrique, Gallstone ileus of the sigmoid colon caused by cholecystocolonic fistula: a case report, *Ann. Med. Surg.* 31 (2018) 25–28, <https://doi.org/10.1016/j.amsu.2018.06.001>.
- [9] E. Alibegovic, A. Kurtcehajic, A. Hujdurovic, S. Mujagic, J. Alibegovic, D. Kurtcehajic, Bouveret syndrome or gallstone ileus, *Am. J. Med.* 131 (2018), e175, <https://doi.org/10.1016/j.amjmed.2017.10.044>.
- [10] E. Nessler, F. Stoss, J. Walsler, G. Wohlgenannt, L. Riedler, The cholecystogastric fistula, *Surg. Endosc.* 5 (1991) 46–47, <https://doi.org/10.1007/BF00591387>.
- [11] C.M. Nuño-Guzmán, M.E. Marín-Contreras, M. Figueroa-Sánchez, J.L. Corona, Gallstone ileus, clinical presentation, diagnostic and treatment approach, *World J. Gastrointest. Surg.* 8 (2016) 65–76, <https://doi.org/10.4240/wjgs.v8.i1.65>.
- [12] F. Alemi, N. Seiser, S. Ayloo, Gallstone disease: cholecystitis, mirizzi syndrome, bouveret syndrome, gallstone ileus, *Surg. Clin. North Am.* 99 (2019) 231–244, <https://doi.org/10.1016/j.suc.2018.12.006>.
- [13] L.E.O.G. Rigler, C.N. Borman, J.F. Noble, Gallstone obstruction: pathogenesis and roentgen manifestations, *J. Am. Med. Assoc.* 117 (1941) 1753–1759, <https://doi.org/10.1001/jama.1941.02820470001001>.
- [14] F. Lassandro, N. Gagliardi, M. Scuderi, A. Pinto, G. Gatta, R. Mazzeo, Gallstone ileus analysis of radiological findings in 27 patients, *Eur. J. Radiol.* 50 (2004) 23–29, <https://doi.org/10.1016/j.ejrad.2003.11.011>.
- [15] M. Gonzalez-Urquijo, M. Rodarte-Shade, G. Lozano-Balderas, G. Gil-Galindo, Cholecystoenteric fistula with and without gallstone ileus: a case series, *Hepatobiliary Pancreat. Dis. Int.* 19 (2020) 36–40, <https://doi.org/10.1016/j.hbpd.2019.12.004>.
- [16] M.I. Salazar-Jiménez, J. Alvarado-Durán, M.R. Fermín-Contreras, F. Rivero-Yáñez, A.I. Lupian-Angulo, A. Herrera-González, Gallstone ileus, surgical management review, *Cir. Cir.* 86 (2018) 182–186, <https://doi.org/10.24875/CIRU.M18000032>.
- [17] W.J. Halabi, C.Y. Kang, N. Ketana, K.J. Lafaro, V.Q. Nguyen, M.J. Stamos, D. K. Imagawa, A.N. Demirjian, Surgery for gallstone ileus: a nationwide comparison of trends and outcomes, *Ann. Surg.* 259 (2014) 329–335, <https://doi.org/10.1097/SLA.0b013e31827eefed>.
- [18] G. Singh, N. Merali, S. Shirol, P. Drymoussis, S. Singh, D. Veeramootoo, A case report and review of the literature of Bouveret syndrome, *Ann. R. Coll. Surg. Engl.* 102 (2020) e15–e19, <https://doi.org/10.1308/rcsann.2019.0161>.
- [19] L. Dunphy, I. Al-Shoek, Gallstone ileus managed with enterolithotomy, *BMJ Case Rep.* 12 (2019), <https://doi.org/10.1136/bcr-2019-231581>.
- [20] C.F. Ploneda-Valencia, M. Gallo-Morales, C. Rinchon, E. Navarro-Muñoz, C. A. Bautista-López, L.F. de la Cerda-Trujillo, L.A. Rea-Azpeitia, C.R. López-Lizarraga, El íleo biliar: una revisión de la literatura médica, *Rev. Gastroenterol. México.* 82 (2017) 248–254, <https://doi.org/10.1016/j.rgmx.2016.07.006>.