


**BOWEL OBSTRUCTION DUE TO GALLSTONE ILEUS AS A RARE CAUSE OF ACUTE ABDOMEN: CASE REPORT AND LITERATURE REVIEW**

**ÍLEO BILIAR COMO CAUSA RARA DE ABDOME AGUDO OBSTRUTIVO: RELATO DE CASO E REVISÃO DA LITERATURA**

**ÍLEO BILIAR COMO CAUSA RARA DE ABDOMEN AGUDO OBSTRUCTIVO: REPORTE DE CASO Y REVISIÓN DE LA LITERATURA**

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**ABSTRACT**

Gallstone ileus as a cause of acute intestinal obstruction is a rare condition resulting from a complication of a common disease, cholelithiasis. It is caused by the impaction of a gallstone in the gastrointestinal tract, posing a potential life-threatening risk in cases of delayed diagnosis and management. We present the case of a 65-year-old man with gallstone ileus causing intestinal obstruction, treated with a longitudinal enterotomy and removal of the stone from the intestinal loop, followed by an urgent transverse enterorrhaphy, in which the surgical team opted not to manipulate the gallbladder region during the same procedure. Postoperative evolution was satisfactory, and the patient was discharged eight days after admission. This report aims to describe an uncommon event that should be included in the diagnostic repertoire of obstructive abdomen, especially in the current context of increasing cholelithiasis cases.

**Keywords:** Gallstone Ileus. Obstructive Acute Abdomen. Complicated Cholelithiasis. Cholecystoduodenal Fistula. Small Bowel Obstruction.

**RESUMO**

O íleo biliar como causa de obstrução intestinal aguda é uma entidade rara, decorrente de uma complicação de uma doença comum, a colelitíase. É causado pela impactação do cálculo biliar no trato gastrointestinal, com potencial ameaça a vida no caso de atraso diagnóstico e em sua resolução. Apresentamos um caso de um homem, 65 anos, com íleo biliar gerando obstrução intestinal, tratado com enterotomia longitudinal e retirada do cálculo da alça intestinal com posterior enterorrafia transversal de urgência, onde foi optado por não manipular topografia de vesícula biliar no mesmo tempo cirúrgico. A evolução no pós-operatório foi satisfatória com alta hospitalar após 8 dias da admissão. Esse relato tem como objetivo reportar um evento incomum que deve fazer parte do repertório diagnóstico de abdome obstrutivo no cenário atual de aumento de casos de colelitíase.

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**Palavras-chave:** Íleo Biliar. Abdome Agudo Obstrutivo. Colelitíase Complicada. Fístula Colecistoduodenal. Obstrução de Intestino Delgado.

## **RESUMEN**

El íleo biliar como causa de obstrucción intestinal aguda es una entidad rara, resultante de una complicación de una enfermedad común, la colelitiasis. Es causado por la impactación de un cálculo biliar en el tracto gastrointestinal, con un riesgo potencial para la vida en caso de retraso en el diagnóstico y en su resolución. Presentamos el caso de un hombre de 65 años con íleo biliar que generó obstrucción intestinal, tratado con una enterotomía longitudinal y extracción del cálculo de la asa intestinal, seguida de una enterorrafia transversal de urgencia, en la cual se optó por no manipular la región de la vesícula biliar en el mismo acto quirúrgico. La evolución posoperatoria fue satisfactoria, con alta hospitalaria ocho días después del ingreso. Este reporte tiene como objetivo describir un evento poco común que debe formar parte del repertorio diagnóstico del abdomen obstrutivo en el escenario actual de aumento de casos de colelitiasis.

**Palabras clave:** Íleo Biliar. Abdomen Agudo Obstrutivo. Colelitiasis Complicada. Fístula Colecistoduodenal. Obstrucción del Intestino Delgado.

## 1 INTRODUCTION

Biliary ileum is a rare cause of intestinal obstruction, accounting for about 1-4% of cases (W.Kirchmayr et al, 2005). It is defined as a complication of a common problem, cholelithiasis, capable of generating a mechanical obstruction due to the presence of one or more gallstones in the gastrointestinal tract (A. Ayantunde, A. Agrawal, 2007), most commonly through a cholecystoduodenal fistula. Most cases affect females (male ratio of 1.9-2.5:1), between the sixth and seventh decade of life (Gonzalez-Urquijo M et al, 2020).

The fistula is formed during successive episodes of cholecystitis, where the inflammation causes an adhesion between the duodenum and the gallbladder, which associated with the pressure exerted by the stone, generates communication between the two organs. It is possible that there are other types of communications such as cholecystocolonic (10-20%), cholecystogastric (5-10%) and choledocoduodenal (<5%) communications, among all are less common (Costi R, 2009; Kohl's DR, 2017).

The exit of the gallstone through the fistula results in obstruction of the terminal ileum in up to 89% of cases (Nuño-Guzmán CM et al, 2016), due to conditions such as the disproportion of the stone diameter and the narrow lumen hair, in addition to reduced peristalsis when compared to other intestinal segments (M.B Luu, D.J. Deziel, 2014). Other sites of impact are: jejunum/proximal ileum (0-50%), jejunum (0-50%), indeterminate (0-25%), duodenum (0-10.5%), and colon (0-8.1%) (Nuño-Guzmán CM et al, 2016). In cases where the stone passes via fistula and obstructs the proximal portion of the duodenum or even the pylorus, resulting in gastric obstruction, the presentation is known as "Bouveret Syndrome" (Caldwell KM et al, 2018). It is possible for another condition to occur, also associated with the presence of the fistula, the "Mirizzi Syndrome", which is characterized by the presence of a large gallstone in the distal portion of the gallbladder or in the cystic duct, causing a narrowing of the common hepatic duct (Esparza Monzavi CA et al, 2019).

In addition to cholelithiasis, intestinal obstruction is also a common pathology, responsible for high rates of hospital admission and morbidity (Nuño-Guzmán CM et al, 2016; Caldwell KM et al, 2018; G. Miller et al, 2000), the main etiology being obstruction caused by adhesions (> 70% of cases). Other less common causes are: hernias, malignancies, inflammatory bowel diseases, intestinal volvulus, intussusception, and biliary ileus (Nuño-Guzmán CM et al, 2016; B.T. Fevang, 2000).

We present the case of a patient with cholelithiasis, with signs of chronic cholecystitis and acute obstructive abdomen due to biliary ileum, requiring urgent surgical treatment. This

report aims to highlight the need to actively consider the etiology, with a high level of diagnostic suspicion, through a successful example of immediate surgical treatment.

## 2 CASE REPORT

Patient J.B., male, 65 years old, with type 2 Diabetes Mellitus, admitted to the Emergency Room of the Hospital de Caridade São Vicente de Paulo complaining of abdominal pain in hypogastro, right iliac fossa and left iliac fossa with onset one month before associated with bilious nausea and vomiting. It also referred to the cessation of evacuations and the cessation of the elimination of flatus for a week.

On physical examination, he was anicteric, dehydrated, tachycardic, and had a blood pressure of 90/50 mmHg, with filiform peripheral pulses. Palpation of the abdomen showed pain in the hypogastrium, with tympanism on percussion and increased auscultation of bowel sounds. The digital rectal examination was without blood or melena, with gloved finger feces.

With the diagnostic hypothesis of acute obstructive abdomen, clinical treatment was initiated with hydration and nasogastric tube (NGS) insertion for gastric decompression. At admission, a radiological series of the acute abdomen was requested – X-ray of the standing and lying abdomen – which showed significant distension of the small bowel loops with air-fluid levels (Figure 01). Laboratory tests showed leukocytosis without deviation and changes in urea, creatinine, and increased C-reactive protein levels.

Following the investigation, a contrast-enhanced tomography scan of the upper abdomen and pelvis was performed, which showed a poorly filled gallbladder, with discretely thickened walls associated with aerobilia (Figure 02) and an image suggestive of calculus, inside the distal ileal loop in the right iliac fossa (Figure 03). In the evaluation of the intestinal loops, it showed moderate distension of the small bowel loops and upstream air-fluid level of the loops. The findings of this imaging study were compatible with Rigler's triad, suggestive of biliary ileum, which was associated with cholecystoduodenal fistula.

The patient underwent exploratory laparotomy, which showed great distension of the small bow loops diffusely and hyperemia, with no signs of ischemia, with gallstones of about four centimeters in diameter, impacted 90 centimeters from the ileocecal valve (Figure 04). There was also an intense blockade in the topography region of the bile duct, and it was decided not to undo the local blockade. It was decided to perform enterotomy 150 centimeters from the ileocecal valve, vertically with removal of calculus from the intestinal loop and posterior horizontal enterorrhaphy (Figure 05).

After surgery, the patient remained for six days in a closed sector of a hospital unit, using combined antibiotic therapy of ciprofloxacin and metronidazole, evolving clinically stable without the use of blood products or vasoactive drugs, with return of flatus elimination and normalization of laboratory test results. Initially, a liquid diet was introduced, which, on the sixth postoperative day, was progressed to a pasty diet and later a bland diet. Patient is discharged from hospital 8 days after hospital admission and 7 days after enterectomy surgery by laparotomy with elective surgical programming for gallbladder wall defect correction.

### Figure 1

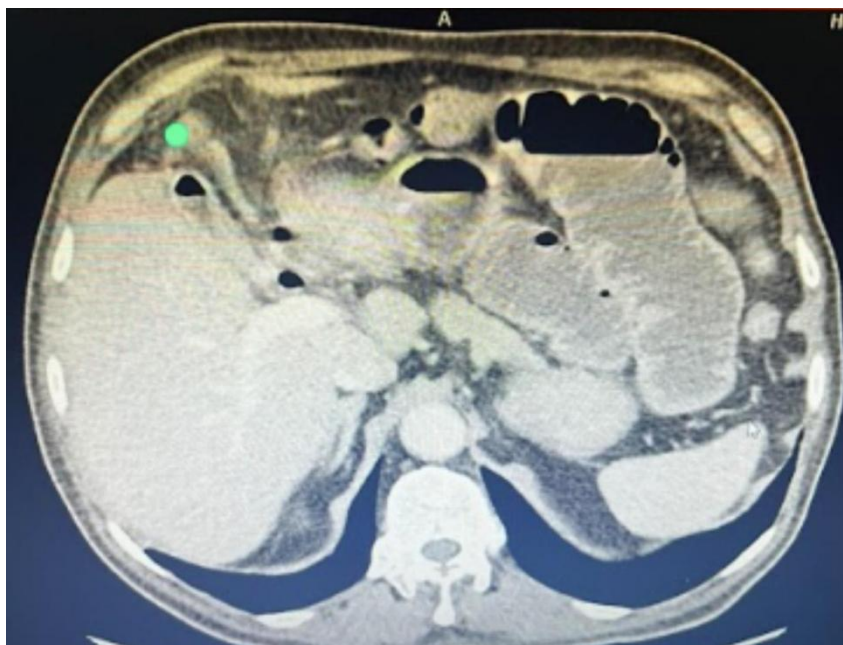
X-ray of the acute abdomen showing distension of the small bowel loops with air-fluid levels inside



Source: Patient record in the São Vicente de Paula Hospital system

**Figure 2**

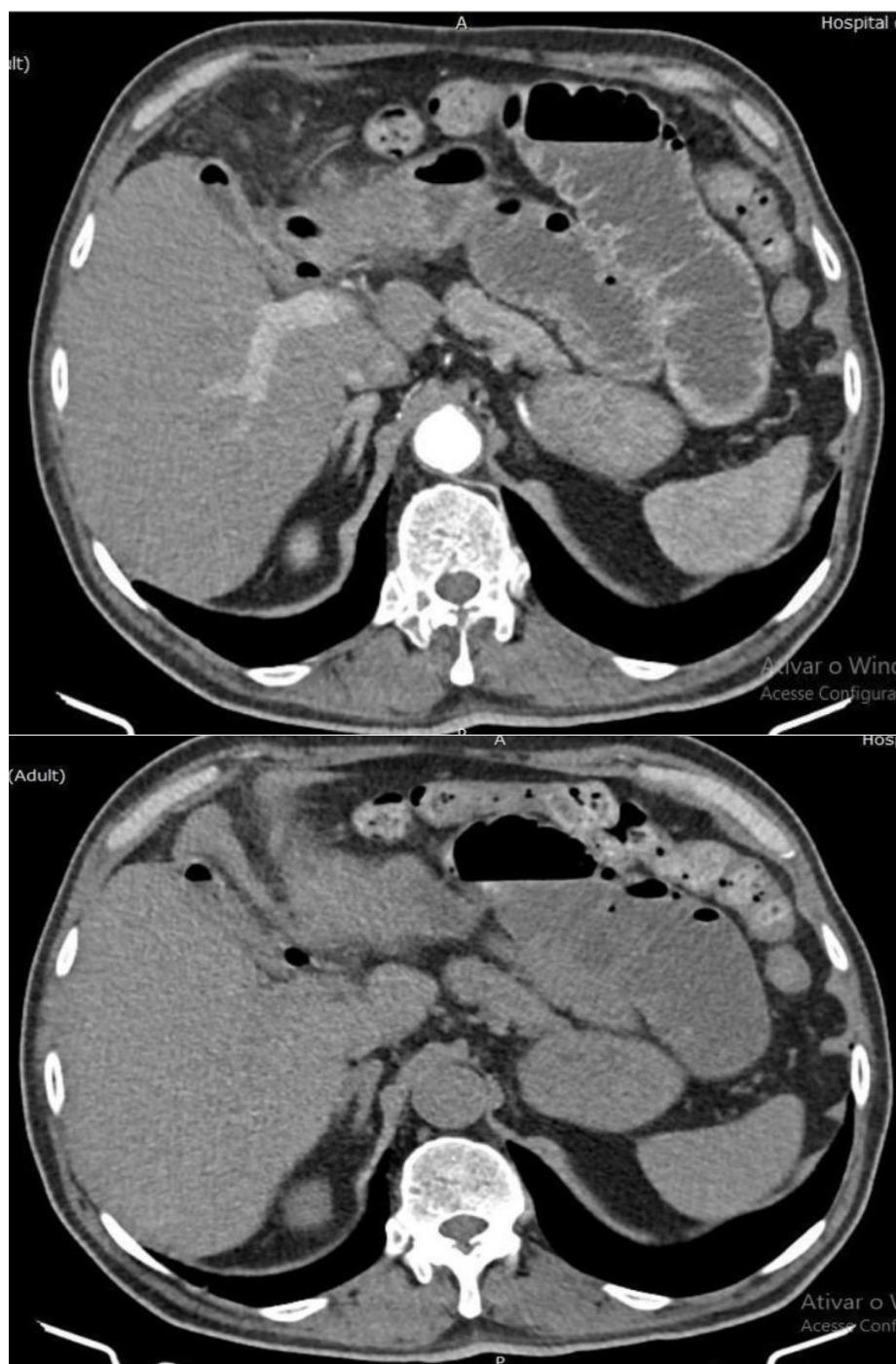
*CT slicing showing distension of the bowel loops at the air-fluid level, and aerobilia (green dot)*



Source: Patient record in the São Vicente de Paula Hospital system

**Figure 3**

*CT slices with an image suggestive of an inflammatory process where the cholecystoenteric fistula occurred*



Source: Patient record in the São Vicente de Paula Hospital system



**Figure 4**

*Intraoperative image showing a stone impaction site with significant distension of the upstream loops*



Source: Photo taken by the authors themselves

**Figure 5**

*Image of the gallstone responsible for the biliary ileum presented by the patient*



Source: Photo taken by the authors themselves



### 3 DISCUSSION

Biliary ileum, a rare complication of cholelithiasis, is the cause of 1-3% of intestinal mechanical obstructions, and cholelithiasis affects approximately 65% of the population over 65 years of age (W. Kirchmayr et al, 2005; Halabi WJ et al, 2014). Due to the high prevalence of gallstones and the biliary ileum being a complication of these, these cases may present routine cases in medical practice, which justifies the need to think of this pathology as a differential diagnosis in cases of acute abdomen. Mortality for biliary ileus cases can reach 18% in elderly patients, especially when considering the associated comorbidities in this population and the high prevalence of insidious presentation (Halabi WJ et al, 2014). Early diagnosis and timing of surgical approach are considered the two most important predictors of good prognosis (W. Kirchmayr et al, 2005).

The clinical findings of this pathology are variable and nonspecific, depending on the site of obstruction. The most common symptom at presentation is abdominal pain (91.5% of patients), accompanied by abdominal distension (84.7%), vomiting (59.7%), and fever (40.9%) (Nakao A et al, 2008). The presentation may be an acute, intermittent, or chronic bowel obstruction; with variable mortality of 7-20% that increases from the presence of risk factors such as advanced age, presence of multiple comorbidities and late presentation of symptoms (Nuño-Guzmán CM et al, 2016; Chang L et al, 2018; Inukai K, 2019).

The diagnosis is made through clinical presentation with the aid of imaging tests, and contrast-enhanced computed tomography (CT) is the modality with the highest diagnostic sensitivity (Lassandro F et al, 2004).

On abdominal radiography, the presence of Rigler's Triad is a pathognomonic sign, however, it is present in < 50% of cases (Ravikumar R, Williams JG, 2010), composed of small bowel obstruction with distension, pneumobilia, and the presence of ectopic gallstones (Rigler LG, Borman CN, Noble JF, 1941). Two out of three radiological signs are sufficient for the diagnosis of ileus. However, CT is the modality of choice in the emergency setting, aiming at a rapid and clarifying diagnosis of the etiology of acute obstructive abdomen (Halabi WJ et al, 2014; Chang L et al, 2018; Rigler LG, Borman CN, Noble JF, 1941; Yu C-Y et al, 2005; Ravikumar R, Williams JG, 2010; Doko M et al 2003). Two other signs were added to Rigler's triad: the change in the location of the stone on subsequent examination, and the presence of various levels of fluids adjacent to the right upper quadrant (Yu C-Y et al, 2005).

CT is described as having high sensitivity (93%), specificity (100%), and accuracy (99%) in the diagnosis of ileus (Yu C-Y et al, 2005).

Laboratory tests are typically non-specific, and may show: hyperbilirubinemia, increased liver transaminase enzymes, leukocytosis, hydroelectrolyte disturbances, acid-base disturbances, and acute renal failure. These changes will depend on the time of onset of the condition, the intensity of the inflammatory response generated, and previous comorbid conditions (Nuño-Guzmán CM et al, 2016; Halabi WJ et al, 2014).

The treatment of this condition is the surgical removal of the stone causing the obstruction, via proximal enterotomy, with or without excision of the bilioenteric fistula during the same procedure, depending on the patient's performance status and overall condition (Halabi WJ et al, 2014; Ravikumar R, Williams JG, 2010). The 2-step surgical procedure is the most commonly applied technique in the management of these cases, with elective laparoscopic cholecystectomy performed a few weeks after the first procedure.

In general, spontaneous stone expulsion occurs in a minimal portion of cases (1.3%) (Ravikumar R, Williams JG, 2010), and the procedure is indicated in one or two surgical procedures. Morbidity in cases of ileus is higher in the one-step surgical approach (61.1%) compared to two-step resolution (27.3%), with a mortality rate of 9% in the two-intervention group and 10.5% in the single-intervention group (Doko M et al 2003). In addition, there are data that indicate an increase in hospital stay and mortality levels in patients who underwent resolution of the condition in one surgical time (Halabi WJ et al, 2014), considered an excessively invasive procedure, with a greater association with postoperative complications.

The two-step procedure involves an initial step, with the removal of the stone, followed by closure of the fistula in a subsequent procedure. However, this procedure has the disadvantage of having associated a higher risk of recurrence, retrograde cholecystitis, and the development of neoplasms (Cooperman AM, Dickson ER, ReMine WH, 1968; Bossart PA, Patterson AH, Zintel HA, 1962).

It has been reported that there is a natural closure of the fistula in 61.5% of cases, suggesting that planning two-step resolution can promote spontaneous fistula closure (Kaneda H et al, 2007).

Therefore, the two-step procedure is considered the gold standard method for cases of ileus

#### **4 CONCLUSION**

Cholecystoenteric fistulas are rare entities that remain a diagnostic and therapeutic challenge, due to their nonspecific nature in the presentation of the condition. Therefore,

biliary etiology should be suspected in all patients with acute or chronic abdominal pain, chronic diarrhea, gastric or small bowel obstruction, especially in patients with chronic biliary pathologies and multiple comorbidities. All surgeons should be aware of this differential diagnosis, since it requires a high index of suspicion, even decades after cholecystectomy, which can be confirmed by contrast-enhanced abdominal CT demonstrating Rigler's triad. The management of the ileum is essentially surgical, and the choice of procedure depends on the patient's baseline surgical condition, the intraoperative findings, and the surgeon's expertise. The increase in the rate of biliary pathologies in recent years should draw even more attention to this diagnostic possibility.

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