INTRODUCTION

Cholelithiasis is one of the most common surgical diseases. Its incidence is age-related, with increasing prevalence in the elderly population. In its evolution, approximately 2% of patients develop symptoms or complications, such as acute cholecystitis, pancreatitis and choledocholithiasis. In rare cases acholecysto-enteric fistula may occur, which can lead to intestinal obstruction after migration and impaction of gallstones, creating a condition known as biliary ileus. We report a case of a 55 year old female who presented with small bowel obstruction owing to large gallstone in ileum.

CASE REPORT

Patient female, 55 years. Previous history of hypertension and total hysterectomy for benign disease. Reported intense pain in the right hypochondrium for 6 days associated to vomit. On the 3rd day of evolution, the pain started to have diffuse pattern, associated with feculent vomit, abdominal distension and stop elimination of gas and faeces.

On physical examination the patient was hemodynamically stable, anicteric, with mild signs of dehydration, the abdomen was distended with very active bowel sounds and painful diffusely. Leukogram pointed slight elevation, liver and canalicular enzymes (total bilirubin, direct and indirect bilirubin, alkaline phosphatase, gamma glutamyl transferase, transaminase glutamic oxaloacetic and transaminase glutamic pyruvic) within normal parameters.

Computed tomography indicated aerobilia, important distension of the small bowel, calcified and impacted foreign body in the distal ileum, suggesting biliary ileus (Figures 1 and 2). Given the findings, the patient underwent upper digestive endoscopy, which
identified the existence of wide cholecysto-duodenal fistula, allowing internal visualization of gallbladder, discarding the presence of remaining gallstones (Figures 3 and 4).

Based on the clinical and laboratory tests, the patient was taken to the operating room. Intraoperatively, was observed significant distension of the small bowel up to the region of the distal ileum, which had an intraluminal foreign body, about 60 cm from the ileocecal valve. The gallbladder was partially viewed, and was well adhered to the first duodenal portion, forming a single block inflammatory which did not compromise the structures of the hepatic hilum. The enterotomy was performed on the antimesenteric border of the obstruction, with subsequent extraction of the 5 cm gallstone and enteroraphy (Figures 5 and 6). We chose not to address the gallbladder. Postoperatively, the patient recovered uneventfully and was discharged from the hospital on the fourth day for outpatient treatment.

Exploratory laparotomy was done. Intra-operatively, a gallstone of size 5x3 cm was found obstructing the proximal ileal lumen, causing dilatation of proximal bowel loops and distal collapse. The gallbladder was partially viewed, and was well adhered to the first duodenal portion, forming a single block inflammatory which did not compromise the structures of the hepatic hilum. Proximal longitudinal enterolithotomy was done, with subsequent decompression after gallstone extraction (Figures 5 and 6). We choose not to perform the cholecystectomy and the fistula repair. Postoperatively the patient recovered uneventfully and was discharged on the fourth day for outpatient treatment.

DISCUSSION

Biliary ileus was first described by Bartholin in 1645 during an autopsy (1). It is a rare condition, being unusual before 50 years and more frequent in women, with an average rate of 3.5: 1 (2). Happens in 1 to 3% of small bowel obstruction in patients up to 65 years of age, incidence which rises to 25% in patients over this age (3). It is a disease presents in only 0.06% of patients with cholelithiasis. It results from repeated
indicating the presence of cholecisto-duodenal fistula.

Figure 3. Cross-sectional and sagittal computed tomography showing foreign body impacted in the ileum.

The gallbladder was proximal ileal lumen, causing dilatation of proximal inflammatory conditions in the gallbladder, leading to the involvement of surrounding structures, formation of fistulous orifices and further migration of gallstones to the gastrointestinal tract (1). The literature describes three main possible fistulas between the biliary tract and digestive tract: cholecysto-duodenal (65-77%), cholecysto-colics (10-25%) and cholecysto-gastrics (5%) (5). Consequently, the symptoms become varied, as the gallstone may lead to obstruction of any segment of the digestive tract, proximal and distal.

Interestingly, one third of the patients are absent of prior symptoms of biliary colic and up to 50% have no history of gallbladder disease. The occurrence of severe pain located mainly in the right upper quadrant may be the harbinger of the biliary-digestive fistula (6). After the formation of the fistula and gallstones migration, many symptoms tend to alleviate until complete disappearance. If the gallstone is small, can be eliminated by faeces, leading to a lack of knowledge of the disease (7). However, relief of symptoms can be only momentary (a few hours or a few days), with subsequent migration of the pain due to the "tumbling phenomenon": migration and impaction of the gallstone into a most distal segment of the digestive tract (8).

The distal ileum, due to the small size and weak peristalsis, is the most frequent site of obstruction (9). The symptoms in this case are typical of small bowel obstruction, including nausea, vomiting, distension, dehydration and abdominal pain. The Riegler’s triad (aerobilia, distended loops and ectopic gallstone), set of highly suggestive biliary ileus signals, is observed in only 15% of the radiographs, which leads many patients to surgery without a predetermined diagnosis. Aiming to a more accurate diagnosis, computed tomography (CT) can demonstrate Riegler’s triad up to 80% of cases (10). Therefore, the image provided by CT is crucial not only for the diagnosis of biliary ileus, but also to helping in the therapeutic decision (11).

Figure 5. Gallstone extraction through enterotomy in antimesenteric.

Another rare complication of cholelithiasis is Mirizzi’s syndrome, characterized by extrinsic compression of the bile duct by the infundibulum of the gallstone or cystic duct (12), which may culminate in cholecysto-biliary fistula. By involving different stages of the same disease, when identified cholecysto-enteric fistula, the possibility of association with Mirizzi’s syndrome should be considered. In a study (13), where patients underwent to a cholecystectomy in a 11 - year period, 29% had cholecysto-enteric fistula associated with Mirizzi’s syndrome.

Figure 6. Gallstone measuring 3 cm x 5 cm.

Extracorporeal lithotripsy shock waves for fragmenting the stones, mechanical dilation of stenotic segments and endoscopic removal, have been described as alternatives forms of non-surgical treatment (14). But, these treatments take into account the location of the obstruction, according as the gallstone can be removed by endoscopy or colonoscopy if they are in a favorable position (15). Furthermore, the availability and cost of materials have to be considered too.

The treatment is most often surgical and can be divided into two steps: one is mandatory and for the relief of intestinal obstruction (enterolitotomy); and the other, that is not always possible and often not recommended, is for the cholecystectomy and fistula closure. A more complex and slow operation may result in higher complications rates. Thus, cholecystectomy and fistula repair should be reserved only for selected patients with absolute indications, including acute cholecystitis, gangrene of the gallbladder or residual gallstones. Furthermore, recurrent episodes of biliary ileus are rare, reported in only 5% of patients, and attributed to the residual gallstones previously undetected. When there are no residual gallstones, most fistulas will close spontaneously (16). Therefore, considering the rare recurrence of biliary ileus and excellent tolerability of biliodigestive fistula that does not close spontaneously, just enterolitotomy proves advantageous for most patients with biliary ileus (17).
Being an uncommon cause of small bowel obstruction, the diagnosis of this pathology is usually delayed, sometimes being performed only at the surgery\(^7\). The late diagnosis worsens the prognosis, with a mortality ranging from 12 to 27%. However, the worst outcome is mainly due to advanced age and their associated comorbidities, which increase the surgical risk. Therefore, it is extremely important to choose the most advantageous and the safe surgical approach. A longer operation time associated with an additional surgical trauma caused by the fistula repair cannot be beneficial for the majority of these patients\(^18\).

In this case, the treatment showed excellent results without associated complications. The fistula closure was not done, following the guidelines described in the literature that demonstrate high rate of complications and small benefit in fistula repair. Moreover, endoscopic visualization of the gallbladder, discarding the presence of residual gallstones, eliminated the possibility of recurrence of biliary ileus in this patient.

**Conflicts of Interest:** None.

**BIBLIOGRAPHIC REFERENCES**


**Correspondence:**
Eversøn L. A. Artifon
Rua Guimarães Passos, 260/apto 121 Sao Paulo. ZC: 04107-030; Brazil
E-mail: eartifon@hotmail.com