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Gallstone Ileus: Report of Two Cases and Review of the Literature

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Abstract: **Review Article**

Gallstone ileus is a rare and serious complication of gallstone disease, presenting as a mechanical intestinal obstruction caused by an ectopic gallstone. It mainly affects elderly individuals and carries a high mortality rate because of the advanced age of patients and the associated comorbidities. It results from the formation of a bilio-enteric fistula. A large gallstone may then migrate into the intestine and become impacted, most often in the terminal ileum. Symptoms are often misleading and intermittent, which may delay diagnosis. Computed tomography (CT) is the imaging modality of choice. Despite some limitations, it allows confirmation of the diagnosis by visualizing Rigler's triad, consisting of pneumobilia, intestinal obstruction, and an impacted stone at the transition zone. Treatment is mainly surgical and is essentially aimed at relieving the obstruction. Several strategies are debated, with enterolithotomy alone occupying a predominant place. A rapid diagnosis, facilitated by CT, and appropriate surgical intervention, most often a simple enterolithotomy, are essential to improve patient prognosis. We report two cases of gallstone ileus. The first concerned an 81-year-old woman with hypertension and diabetes, followed for gout, who presented with epigastric pain and vomiting evolving over 5 days. Ultrasound was performed and showed fluid-filled dilated small-bowel loops without wall thickening or visible obstruction, and a contracted gallbladder with no dilatation of the intrahepatic bile ducts. Abdominal computed tomography (CT) was requested and confirmed obstruction proximal to a stone in the last ileal loop, associated with thickening of the gallbladder wall and pneumobilia, suggesting chronic cholecystitis with a biliary-enteric fistula. The patient underwent laparotomy with a one-stage procedure; exploration did not demonstrate a stone in the ileum, and cholecystectomy with fistula repair was performed. It was concluded that the stone had passed into the colon. The patient was subsequently monitored, with a favorable course and notably no sign of recurrence. The second case concerned a 65-year-old man who presented to the emergency department with a picture of intestinal obstruction; emergency abdominal CT was performed and the diagnosis of gallstone ileus was established. He underwent laparotomy with a one-stage procedure and extraction of the stone by enterolithotomy. The postoperative course was favorable. The literature on gallstone ileus consists mainly of small series and case reports. We performed a literature review focusing on the relevant points of diagnosis and management of this condition.

Keywords: Gallstone Ileus, Rigler's Triad, Pneumobilia, Gallstone, Intestinal Obstruction, Cholecystoduodenal Fistula, One-Stage Procedure, Enterolithotomy.

INTRODUCTION

Gallstone ileus constitutes a particular clinical entity, both rare and potentially severe, corresponding to a mechanical intestinal obstruction secondary to the migration of a gallstone into the digestive lumen. Although this pathology represents only a small proportion of intestinal obstructions in the general population, estimated between 1 and 4% [1], and 0.3–0.5% of gallbladder stones [1], its incidence increases significantly in elderly subjects, reaching up to one quarter of small-bowel obstructions after the age of 65. This condition is characterized by a non-

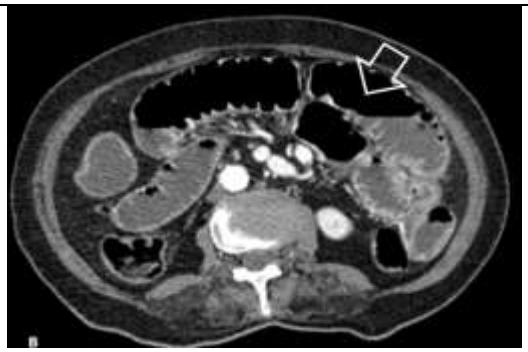
negligible mortality, largely attributed to the frailty of the patients, but also to frequent diagnostic delay and comorbidities. Imaging plays a decisive role. It is not limited to confirming the diagnosis, but actively contributes to understanding the mechanism, localizing the obstruction, and planning treatment [1-3].

CASE REPORTS

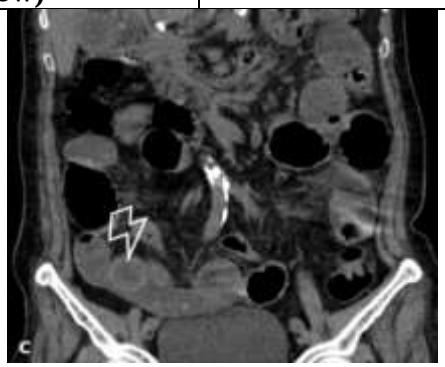
Case 1

This was an 81-year-old woman, known to have hypertension and diabetes under treatment, followed for gout, who presented with epigastric pain and vomiting evolving for 5 days. Ultrasound was performed and showed fluid-filled dilated small-bowel loops without wall thickening or visible obstruction. There was also a contracted gallbladder with no dilatation of the intrahepatic bile ducts. Abdominal computed tomography (CT) was requested and showed distension of the small-bowel loops with a few air-fluid levels measuring 30 mm in maximal diameter, proximal to a distal ileal stone associated with thickening of the gallbladder wall, pneumobilia, and duodenal infiltration, suggesting chronic cholecystitis with a biliary-enteric fistula. The patient underwent laparotomy with a one-stage procedure, but exploration did not reveal a stone in the ileum; cholecystectomy with fistula repair was performed. It was concluded that the stone had been eliminated into the colon. The patient was subsequently monitored, with a favorable course, notably with no bowel transit disorder and no sign of recurrence.

A: Axial portal venous phase CT image: Pneumobilia with suspicion of a cholecysto-duodenal fistula (arrow)



B: Axial portal venous phase CT image Distension of the small-bowel loops containing air-fluid levels (arrow)



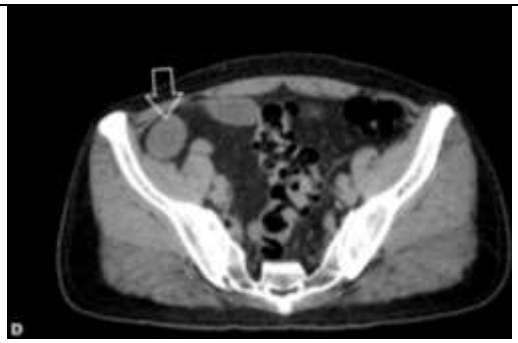
C: Coronal unenhanced CT image: Gallstone impacted in the distal ileum: note its slightly stratified appearance (arrow)

Case 2:

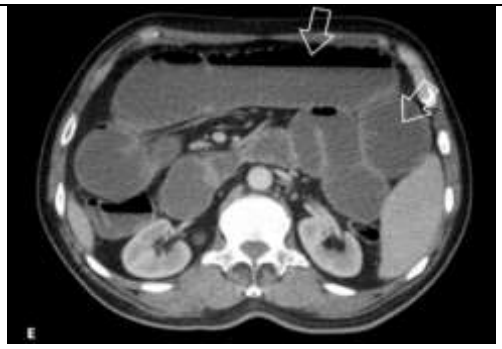
This was a 65-year-old man who presented to the emergency department with a picture of intestinal obstruction of recent worsening over two days, consisting of diffuse abdominal pain, abdominal distension, and cessation of stool and gas passage. In addition, the patient had previously reported chronic

right hypochondrial pain. Emergency abdominal CT was performed and the diagnosis of gallstone ileus was established on the basis of gastroduodenal distension and distension of the small-bowel loops measuring 50 mm in maximal diameter, containing air-fluid levels, proximal to a low-density ectopic intraluminal stone visible in the right iliac fossa measuring 29 × 24 mm. This was associated with thickening of the jejunal loop proximal to the stone, measuring 6 mm in maximal thickness, showing target enhancement after contrast administration.

He subsequently underwent laparotomy with a one-stage procedure and extraction of the stone by enterolithotomy. The postoperative course was favorable.



D: Gallstone impacted in the ileum at the level of the right iliac fossa



E: Dilated small-bowel loops with air-fluid levels

Note the stratified appearance of the stone (arrow)

F: Pneumobilia (arrow)

Comments on the cases:

The site of impaction was the distal ileum for the first patient and the mid-ileum for the second, corresponding perfectly to the transition zones between collapsed and dilated loops. The mean densities of the stones, excluding the hyperattenuating peripheral rim, were 15 HU and 60 HU respectively, which explains the difficulty in identifying low-density stones or stones with a density close to that of intestinal contents, as in the first case. Both stones shared a slightly stratified appearance with relatively dense concentric layers and measured 26 × 23 mm and 29 × 24 mm respectively. Concerning pneumobilia, in the first case it was visible in the gallbladder (pneumocholecyst) and in the common bile duct, and in the second case it was more obvious and diffuse in the intra- and extrahepatic bile ducts and in the gallbladder. The latter was contracted in both cases, somewhat more markedly in the second case. In both cases, there was a very strong suspicion of a cholecysto-duodenal fistula (second portion of the duodenum). Small-bowel obstruction was confirmed in both cases, with intestinal distension measuring 30 mm and 50 mm in maximal diameter respectively. As for signs of obstructive complications, there were no pathognomonic

signs of small-bowel wall ischemia. Nevertheless, in the first case there was mild mesenteric fat stranding adjacent to the afferent loop in the right flank with hyperemia and a small amount of peritoneal effusion, and in the second case there was slight thickening of the upstream loop with enhancement after contrast injection. Intraoperative assessment did not reveal established ischemia.

DISCUSSION

Gallstone ileus is defined as a mechanical intestinal obstruction due to the impaction of one or more gallstones migrated from the gallbladder or bile ducts, most often through a biliary-enteric fistula [3]. It is the most frequent acute small-bowel obstruction due to endoluminal obstruction and the leading cause of small-bowel obstruction in elderly women [2]. Indeed, it accounts for 25% of small-bowel obstructions in those over 70 years of age [1]. It is a benign condition, but its presentation is variable and often misleading, which is responsible for diagnostic delay and may worsen the prognosis. From a diagnostic point of view, CT has replaced plain abdominal radiography and gastrointestinal contrast studies despite some limitations, notably for low-density stones. Management remains controversial, with no consensus and a predominant role for enterolithotomy alone [3-5].

From a pathophysiological point of view, this condition is falsely called ileus because it is indeed a mechanical obstruction, due to the endoluminal passage into the duodenum, jejuno-ileum, or more rarely the colon of one or more large gallbladder stones, generally greater than 25 mm in diameter in order to cause small-bowel obstruction. It is mainly due to a biliary-enteric fistula complicating chronic cholecystitis with migration of gallbladder stones into the digestive lumen. However, it is possible for a common bile duct stone to enter the intestine via compliant and temporarily overdistended bile ducts. The gallstone erodes the inflammatory wall of the gallbladder, which is adherent to the digestive wall, leading to its perforation and the creation of a biliary-enteric fistula, usually located in the duodenum; the stone moves over a more or less long distance in the intestinal tract and may increase in size during this movement, finally leading to intestinal obstruction after becoming impacted. Sometimes it may present as a delayed complication (up to 2 months) of endoscopic retrograde cholangiopancreatography (ERCP) [3-7].

Clinically, it is a more or less classic picture of small-bowel obstruction with intermittent acute colicky pain, acute abdominal pain (20–30%), nausea, vomiting, fever, abdominal distension, and constipation. However, the clinical picture is frequently insidious, misleading, and incomplete (self-resolving subocclusions), which leads to diagnostic delay, hence the considerable mortality rate of gallstone ileus (7 to 50%). It should be considered in an elderly woman presenting with recurrent right hypochondrial pain and prolonged vomiting [3, 8-10]. For some authors, the chronic form is called Karewsky syndrome and is characterized by recurrent episodes of pain related to the passage of gallstones into the intestine, alternating with asymptomatic periods, progressing toward complete obstruction in several stages [11-15]. On the other hand, symptomatology may vary according to the site of obstruction, notably with nonspecific high obstruction in duodenal obstruction (Bouveret syndrome) [3, 16, 17]. Patients may also report a history of intermittent right hypochondrial pain, and acute cholecystitis is present in 10 to 30% of patients at the time of gallstone ileus [18].

The positive imaging diagnosis typically demonstrates Rigler’s or Riegler’s triad, associating small-bowel obstruction, ectopic gallstone(s), and pneumobilia [3, 6]. The preferred sites of obstruction are, in descending order of frequency: the ileocecal valve, proximal ileum, distal jejunum, sometimes the duodenum, and even the sigmoid colon [2, 3, 15]. Riegler’s triad was described in 1941 on plain abdominal films

showing intestinal obstruction with air-fluid levels, pneumobilia, and ectopic gallstone(s). But none of these elements is constant, and the formal diagnosis of gallstone ileus is made in only 30 to 39% of cases on plain radiography [3, 6]. In addition, radiolucent or poorly radiopaque stones may go unnoticed, and if the stone remains for a long time in the small bowel, it becomes coated with a fairly large quantity of calcium salts and may then become visible [2, 3, 6, 15, 19]. Formerly, gastrointestinal contrast studies were performed, showing the classic "huge serpent with a clear head," corresponding to the dilated loop proximal to the stone. It has limited diagnostic capacity [3, 6]. Computed tomography is the examination of choice, with specificity and sensitivity far superior to radiography. The first complete CT Rigler triad was published in 1985. The CT forms observed often combine a sufficient number of suggestive diagnostic elements, but they are not necessarily all constant [2, 3]. Pneumobilia is a cardinal diagnostic element indicating the existence of a biliary-enteric fistula, but it may be absent if the biliary-enteric fistula tract collapses. It may involve the intrahepatic bile ducts and must then be differentiated from portal venous gas. The presence of air bubbles in the gallbladder (pneumocholecyst) is easy to diagnose when the morphology of the gallbladder allows its identification, which is not the case when it is sclero-atrophic [2, 3, 6]. The gallstone migrated into the intestinal lumen becomes impacted in a digestive loop at the transition zone between a dilated and a collapsed segment. The stone may be almost exclusively cholesterol and therefore difficult to visualize on CT, with a density close to that of water. In this case, some morphologic features may guide the diagnosis: the presence of a core with regularly circumferential layers of variable density. One may also have a clear center with a calcified shell in a peripheral crown (radiolucent cholesterol center with a calcified shell of calcium bilirubinates). Therefore, it is recommended to carefully analyze the intestinal luminal contents and to narrow the display window in order to better see the stone. In addition, it is imperative to check all the small-bowel loops on CT images for multiple stones in order to avoid recurrence [2, 3, 15]. Intestinal obstruction may be insidious and misleading, but it can also present dramatically. Sub-occlusive episodes follow one another, resolving when the stone progresses. Impaction of the stone is linked either to its progression toward a digestive segment of narrowed caliber (typically the terminal ileum), or when the stone becomes larger by collecting sediment from the intestinal contents [2, 3, 15, 19]. We noted that the term "Barnard syndrome" is currently little used in the literature to designate a gallstone of the ileum, terminal ileum, or ileocecal valve, as in the review by Ploneda-Valencia *et al*., [15] and the publication by Williams *et al*., [20].

CT also makes it possible to look for the biliary-enteric fistula, signs of bowel compromise, and to optimize the site and length of the laparotomy: limiting parietal damage as well as manipulation of distended loops (which worsens postoperative morbidity) [2, 3, 19]. Apart from the problem of purely cholesterol stones, it has some limitations such as resolution of the obstruction at the time of imaging, multiple stones, and a collapsed biliary-enteric fistula without pneumobilia [3]. Ultrasound makes it possible to study the gallbladder state, notably the signs of chronic cholecystitis, a sclero-atrophic gallbladder, the presence of pneumobilia with comet-tail artifact, and can even detect a calcified or non-calcified gallstone: a rounded hyperechoic image with posterior acoustic shadowing, with the presence of distended small-bowel loops upstream. However, it remains an operator-dependent examination and is limited by overlying bowel gas [2, 19, 21]. MRI is rarely prescribed in this setting, but it presents the following advantages: better characterization of biliary-enteric fistulas, better visualization of the biliary tree and its contents, and it is s the imaging modality of choice for detection of radiolucent and isoattenuating stones, often not visible on standard radiography and CT. It makes it possible to localize the stone in

difficult cases and may show Rigler's triad in almost 100% of cases [57]. However, it remains little used in the acute diagnostic setting because of its disadvantages, notably the longer acquisition time, poor tolerance (elderly subject), and lack of availability. On the other hand, it could be useful for estimating the risk or tendency of a chronic lithiasic gallbladder to form a fistula into the intestine [2, 19, 22]. Finally, spectral CT may solve the problem of purely cholesterol gallstones that are indistinguishable from luminal contents on conventional CT; thus, in the publication by Peng Liu and Xian-zheng, dual-energy CT using Rho/Z reconstruction clearly demonstrated the impacted intraluminal digestive gallstone that was completely invisible on conventional CT.[23]

Three clinical forms of gallstone ileus deserve to be discussed: duodenal gallstone ileus, colonic gallstone ileus, and post-cholecystectomy gallstone ileus. Duodenal gallstone ileus is a particular form of gallstone ileus in which the stone is located in the duodenum. It is responsible for nonspecific high obstruction or an acute picture of pyloroduodenal obstruction defining Bouveret syndrome. This syndrome was described in 1896 by Léon Bouveret under the term "intermittent epigastric hardening." Frequently unrecognized, it may secondarily progress to "classic" gallstone ileus. Acute epigastric pain is accompanied by vomiting that are rather alimentary when the endoluminal obstruction is supravaterian, or bilious vomiting when the obstruction is subvaterian. A minority of patients may present with hematemesis. The biliary-enteric fistula is generally cholecysto-duodenal, more rarely choledocho-duodenal, and exceptionally cholecystogastric. On CT, the images may be striking, combining a bulky endoluminal stone in a duodenum with thickened and remodeled walls with pneumobilia involving the gallbladder and/or the common bile duct and the bile ducts [3, 16, 17, 24].

The colon is the least frequent location, representing 4.1% of cases [25]. Colonic gallstone ileus is a mechanical obstruction of the colon by a gallstone coming from the gallbladder, generally via a cholecysto-colonic fistula. Another cause is a stone that has passed through a cholecysto-duodenal fistula, traversed the small intestine, then crossed the ileocecal valve to reach the colon [26-29]. The most frequent site of impaction is the sigmoid colon (>85%); the ascending colon is the rarest location. The main symptoms are those of partial colonic obstruction. A stone may remain "silent" in the colon before manifesting as obstruction and may also "grow" in the colon by deposition of dehydrated fecal material on its surface. On the other hand, the literature contains a few cases of gallstone ileus diagnosed on imaging that resolved spontaneously with natural passage of the stone, especially in colonic locations [30, 31].

Finally, a previous cholecystectomy does not rule out the diagnosis of gallstone ileus; indeed, about 46 cases of post-cholecystectomy gallstone ileus were published between 1939 and 2019 [32].

The average delay between cholecystectomy and the occurrence of gallstone ileus was 12.4 years (range: 10 days–50 years) [32]. Various mechanisms of lithiasic intestinal obstruction in patients without a gallbladder have been proposed: incomplete cholecystectomy with a large stone at the neck causing stump cholecystitis and the formation of a cholecysto-enteric fistula, cholecysto-enteric fistulas not diagnosed during cholecystectomy, stones retained in intestinal diverticula, a primary stone formed in the bile duct then entering a persistent cholecysto-enteric fistula, stones trapped in the common bile duct leading to gallstone ileus after endoscopic sphincterotomy, a lost stone during cholecystectomy eroding the intestinal wall, stones displaced into the intestine and then increasing in size over time [32, 33].

From the point of view of differential diagnosis, the distinction between gallstone ileus and coproliths or enteroliths may be really difficult, particularly in the case of

incomplete Rigler's triad, and for some authors, gallstone ileus is considered a secondary enterolith (formed outside the digestive lumen). These enteroliths form in a large diverticulum (Meckel's diverticulum, other duodenal or jejunal diverticula) and secondarily migrate into the distal ileum, where they cause recurrent obstructive episodes identical to those of gallstone ileus. Among the signs in favor of gallstone ileus one may cite: the existence of pneumobilia (in particular a pneumocholecyst in a sclero-atrophic gallbladder) and the cholesterol-pigment nature of the stone by its morphology [3, 4, 34, 35]. In second place one may cite distal intestinal obstruction syndrome, particularly in the case of an obvious context with a clinical history of cystic fibrosis, history of meconium ileus, and pancreatic and pulmonary involvement. It is defined by the accumulation of viscid fecal material in the intestinal lumen, associated with sticky mucoid intestinal content adhering to the intestinal wall of the terminal ileum and cecum. Symptoms are intermittent, with a picture of acute or more frequently subacute intestinal obstruction. Differentiation between the two entities is crucial because the management of distal intestinal obstruction syndrome is strictly medical. Constipation is characterized by chronic progression and fecal material widely distributed throughout the colon [5, 36].

The therapeutic management of gallstone ileus remains controversial. While open surgery has long been the treatment modality of choice, other approaches have recently been used, notably laparoscopic surgery and lithotripsy, and for some authors endoscopic treatment may be considered, especially for duodenal and colonic obstructions. Enterotomy with extraction of the stone relieves the intestinal obstruction, but leaves the patient exposed to a risk of recurrence in case of residual stones, persistent symptoms related to an inflamed gallbladder, and possibly an increased risk of gallbladder cancer. For these reasons, other strategies include enterolithotomy associated with cholecystectomy and fistula repair in a one-stage procedure, or enterolithotomy followed by delayed cholecystectomy with fistula repair after recovery from the acute episode, especially since biliary-enteric fistulas may close spontaneously after resolution of the obstruction, and complications related to their persistence are rare. The literature devoted to gallstone ileus is based exclusively on retrospective studies involving small numbers of cases accumulated over long periods. nterolithotomy alone remains the preferred approach in most published series for the majority of patients. In addition, in high-risk patients, enterolithotomy alone remains preferred because of its lower morbidity, reduced mortality, and shorter operative time. Conversely, in patients in better general condition, cholecystectomy associated with fistula repair may offer long-term protection against recurrence, at the price of increased morbidity and more prolonged recovery. However, the question of delayed biliary surgery remains unresolved [37, 38].

CONCLUSION

Gallstone ileus is a nosological entity that should not be missed in the setting of obstructive syndromes in elderly subjects. Computed tomography is essential for the diagnosis of gallstone ileus despite some limitations. This is explained by its high accuracy and its overall ability to visualize the characteristic signs of this pathology. Magnetic resonance imaging offers better resolution, particularly for the study of the biliary tree and radiolucent stones, but it generally remains less suitable in acute emergency situations. Therapeutic management remains controversial, with a predominant place for enterolithotomy.

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