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Gall stone ileus: Unfamiliar cause of bowel obstruction. Case report and literature review

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ABSTRACT

INTRODUCTION: Gallstone ileus is a rare sequela of cholelithiasis. The pathology occurs as a result of bilioenteric fistula due to erosion by the offending gallbladder stone. It is most commonly encountered in elderly females and CT imaging is diagnostic in the majority of cases. Surgical intervention aims to promptly relieve the obstruction by removing the gallstone and dealing with the fistula. Morbidity and mortality are usually high since it usually occurs in elderly patients.

PRESENTATION OF CASE: An 88-year-old lady with multiple chronic medical problems and no history of biliary manifestation presented with acute small bowel obstruction. Abdominal CT imaging revealed a bilioenteric fistula and an impacted gallstone in the jejunum causing occlusion. Laparotomy was performed and the stone was removed via enterolithotomy. Manipulation of the cholecystoduodenal fistula was not attempted due to severe inflammatory adhesions. The patient had uneventful postoperative course and remained symptom free on one year follow-up.

DISCUSSION AND CONCLUSION: Management of gallstone ileus is mainly surgical. Delay in detection and treatment of gallstone ileus may result in significant morbidity and mortality. The choice of surgical option is influenced by the preoperative medical status of the patient. A literature review generally supports the employment of enterolithotomy in high-risk patients and reserving cholecystectomy and resection of the fistula for less comorbid patients with feasible anatomy.

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1. Introduction

Gallstone ileus is an unusual cause of intestinal obstruction. The underlying pathology is inflammation of the gallbladder and formation of a biliary enteric fistula. It typically occurs in the elderly female population [1]. Patients present with symptoms and signs of bowel obstruction. The diagnosis is made by using radiographic investigations, with computed tomography (CT) scan being the most useful modality. The treatment is surgical removal of the gallstone with or without excision of the biliary-enteric fistula during the same procedure depending on the patient general condition [2]. We describe a case of an 88-year-old female with multiple medical comorbidities that presented with intestinal obstruction due to gallstone ileus and was managed by open enterolithotomy.

2. Case presentation

We report the case of an 88-year-old female with the following chronic medical conditions: Diabetes mellitus, hypertension, dyslipidemia, end stage kidney disease on regular hemodialysis, atrial fibrillation, pulmonary hypertension, hypothyroidism and obstructive sleep apnea. She presented to our emergency department with a four day history of progressively worsening diffuse colicky abdominal pain that is associated with repetitive vomiting of food particles and bilious material. Additionally, she reported a two day history of constipation and obstipation. She denied any previous history of biliary symptoms. Her vital signs showed a heart rate of 120 beats per minute, blood pressure of 80/50 mmHg, respiratory rate of 35/min and a temperature of 37 C. Focused physical examination of the abdomen revealed a moderate abdominal distention, slight epigastric tenderness with mild rebound and no guarding was noted. Laboratory investigations revealed a deranged renal function and leukocytosis (16,000/mL). Her INR was 2.32 and liver function tests were unremarkable.

The patient was initially resuscitated with intravenous fluid and had nasogastric tube inserted for decompression. An enhanced computed tomography scan of the abdomen was obtained that revealed dilatation of the biliary system including the intrahepatic

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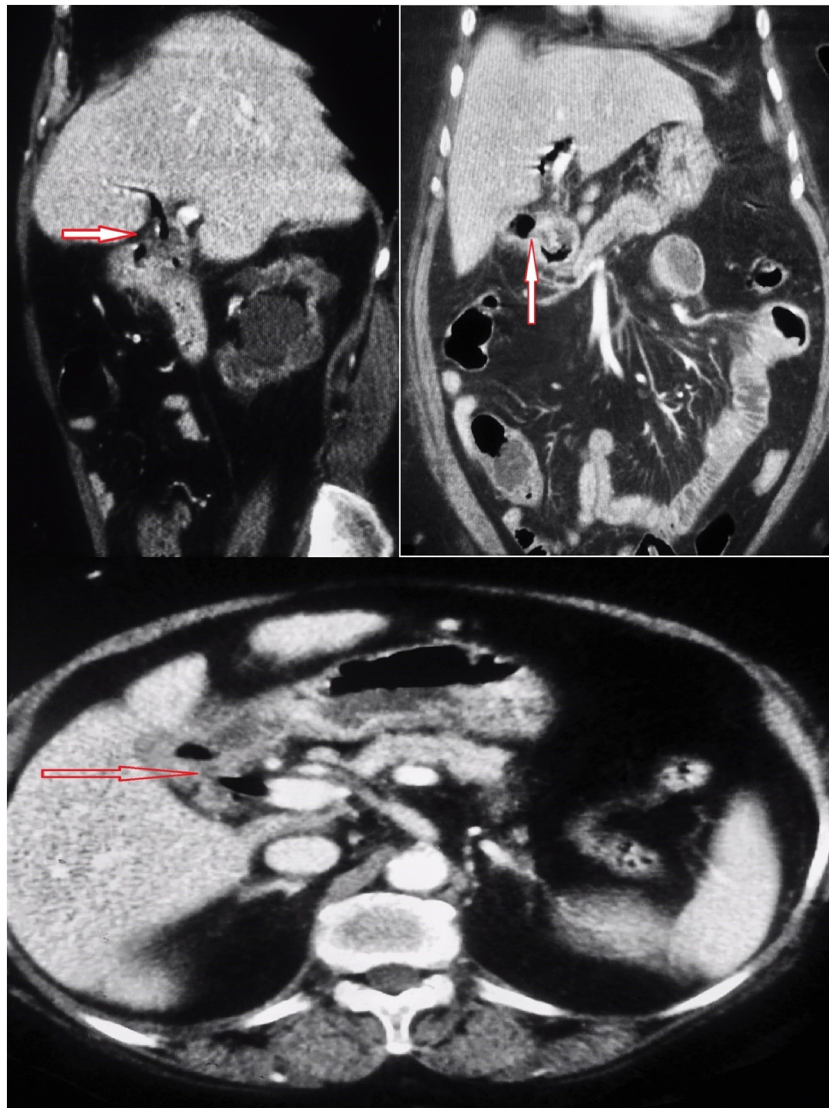


Fig. 1. Dilatation of the biliary system including the intrahepatic ducts with coexisting pneumobilia, cholecystoduodenal fistulization can be appreciated with contracted irregular gallbladder outline containing air locules extending to the juxtapositioned duodenal lumen with a heterogenous enhancement at the site of the abnormal communication.

ducts with coexisting pneumobilia, cholecystoduodenal fistulization with contracted irregular gallbladder outline containing air locules extending to the juxtapositioned duodenal lumen with a heterogenous enhancement at the site of the abnormal communication (Fig. 1). Ectopically positioned hyperdense luminal structure, measuring about 3.5×3 cm in caliber at the sentinel jejunal segment suggestive of a gallstone resulting in distended small bowel dilatation with adjacent collapsed small and large bowel loops (Fig. 2). The decision was made to proceed for an urgent exploratory laparotomy to relieve the obstruction.

The patient's condition was optimized prior to the intervention as she underwent a session of hemodialysis and her INR was corrected with fresh frozen plasma. The intraoperative findings revealed an impacted stone roughly 40 cm from the ligament of Treitz. An enterolithotomy was performed over a healthy jejunal segment few centimeters proximal to the transition zone demonstrating an oval shaped stone measuring roughly 3.5×3 cm in size was removed via a longitudinal incision and the defect was closed primarily in a transverse fashion using an absorbable suture (Fig. 3). Manual inspection of the entire small and large bowel revealed no other stones. The right upper quadrant was examined revealing

presence of very dense adhesions between the gallbladder and the duodenum and absence of any retained stone and therefore, the gallbladder was not manipulated and the fistula was not excised to avoid causing bleeding or injuries. The patient stayed postoperatively for 7 days and her hospital stay was uneventful. She was followed up in the outpatient clinic for 12 months and she remained symptoms free.

3. Discussion and conclusion

Mechanical obstruction of the gastrointestinal tract secondary to gallstones is a rare manifestation of biliary disease occurring in about 0.3–1.5% of cases of cholelithiasis [1]. This phenomenon is estimated to account for 1–4% of all mechanical bowel obstruction. However, this rate tends to increase roughly up to 25% in patients over the age of 65 [3]. A more recent retrospective review, published in 2014, of the Nationwide Inpatient Sample posits that the incidence may actually be much lower than previously held and quite steady accounting for roughly 0.095% of all mechanical bowel obstructions per year in the United States [1]. The typical patient afflicted with this disease is an elderly female. Females account for

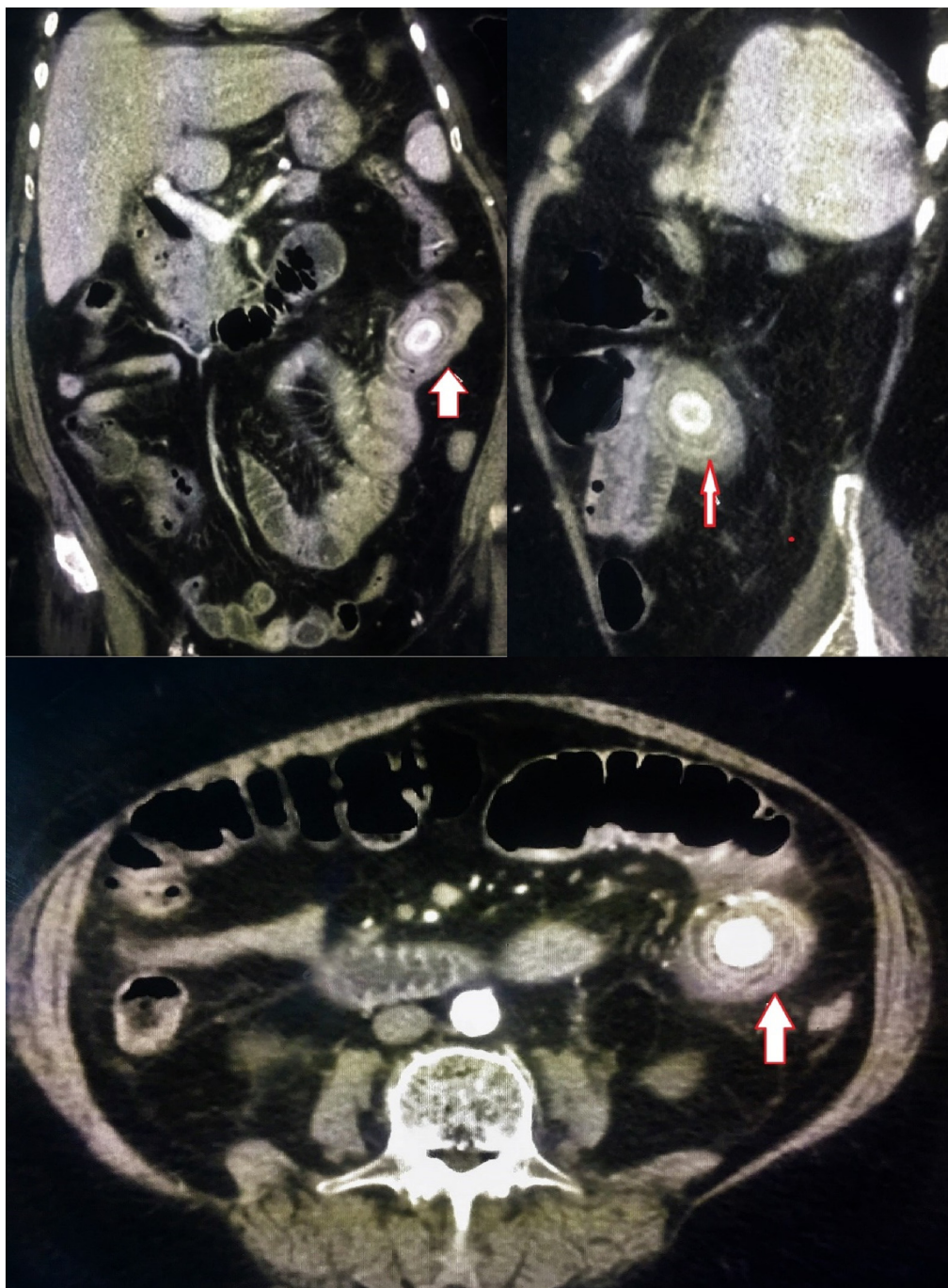


Fig. 2. Ectopically positioned hyperdense luminal structure, measuring about 3.5×3 cm in caliber at the sentinel jejunal segment suggestive of a gallstone resulting in distended small bowel dilataion with adjacent collapsed small and large bowel loops.

>70% of cases [1,4,5]. With the female to male ratio estimated as at least (3:1) [6,7].

The typical age group is in patients older than 70 year old [1,5]. There is usually a preceding history of cholelithiasis, however, this is not an absolute necessary historical factor as only 25–72% were known to have cholelithiasis prior to the diagnosis of gallstone ileus [1]. Table 1, summarises the patients' characteristics and comorbidities adapted from the NIS database [1].

Gallstone ileus occurs most commonly as a result of an aberrant fistula between the biliary tract and the gastrointestinal tract. Biliary enteric fistulas occur as a result of an impacted gallstone

that results in cholecystitis and subsequent adhesions formation. Persistent erosion and necrosis of the gallbladder wall result in inflammation that then spreads to the adherent bowel segment that is in close proximity to the site of the impacted gallstone; resulting in formation of a fistula [6,8–10]. This pathophysiological process also describes the Mirizzi syndrome [6] and there is an intimate relationship between Mirizzi syndrome and biliary enteric fistulas. In one study conducted by Marcelo et al. about ninety percent (90%) of patients who had biliary enteric fistulas had concomitant Mirizzi syndrome [11]. Typically, stones get impacted when their size exceeds the calibre of the bowel lumen, and the minimum size

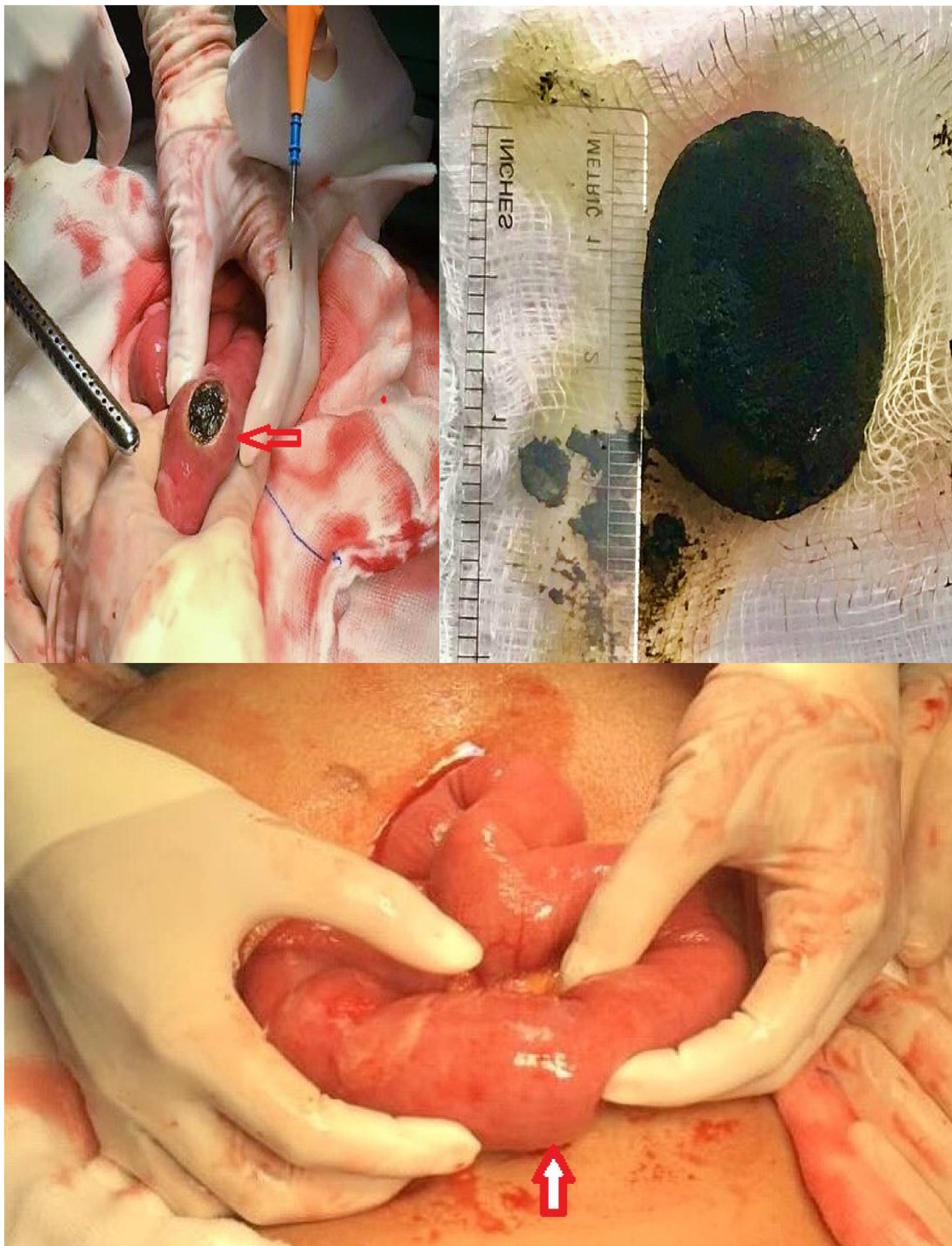


Fig. 3. Gallbladder stone impacted in a jejunal loop measuring about 3.5 × 3 cm.

that is most often quoted is two centimeters [6]. The most common site of impaction is the distal or terminal ileum (75%) [12]. Other potential, but less common, locations include the duodenum (Bouveret Syndrome), stomach, proximal ileum, jejunum, or colon [3,6,7]. Table 2, illustrates the incidence of the common sites of gallstone impaction according to Nuno-Guzman et al [4]. Gallstone ileus most commonly manifests itself in form of bowel obstruction and its cardinal features: constipation/obstipation, nausea, vomiting, abdominal pain, abdominal distension, features of dehydration and electrolyte imbalances [4,6,8,12]. Bowel obstruction

may be intermittent in nature, referring to a gallstone that results in obstruction that spontaneously resolves, the “tumbling phenomenon” by either spontaneous relief of impaction or progression of the gallstone through the gastrointestinal tract [1,4,6]. This may contribute to the delayed presentation sometimes seen in gallstone ileus. The average lag time between symptom onset and presentation is frequently reported as 4–8 days [6,7,10,12].

Although a history of biliary symptoms is common it is not a prerequisite. Rates of previous biliary complaints are frequently quoted at roughly 50% [2,4,12]. Concomitant acute cholecystitis in

Table 1
Demographics and comorbidities of patients from the NIS database. 409 patients that were treated with enterolithotomy are represented in this table. Adapted and modified from Halabi et al. [1].

Age	75 (67–83)
Male	29%
Female	71%
Congestive Heart Failure	14%
Valvular Heart Disease	6%
Chronic Pulmonary Disease	14%
Chronic Liver Disease	1.5%
Chronic Kidney Disease	10%
Anemia	12%
Diabetes	24%
Hypertension	57%
Obesity	8%
Peripheral Vascular Disease	5%
Weight Loss	13%

Table 2
Sites of gastrointestinal obstruction in patients with gallstone ileus. Adapted from Nuño-Guzmán et al. [4].

Duodenum	0–10.5%
Stomach	0–20%
Jejunum	0–50%
Jejunum/Proximal Ileum	0–50%
Ileum	0.89–5%
Colon	0–8.1%
Undetermined	0–25%

the index presentation is found in up to 30% of patients [4]. The modalities most frequently used to arrive at the diagnosis of gallstone ileus include: Plain abdominal radiograph, ultrasound and computed tomography (CT) scan of the abdomen. The classic triad reported by Rigler et al. in 1941 [15] describes the radiographic features that may suggest gallstone ileus: small bowel obstruction with pneumobilia and an ectopic gallstone in the gastrointestinal tract that changes location on serial films. Presence of two out of three of the criteria is pathognomonic for gallstone ileus. This classic triad occurs at a reported rate of (15–50%) in plain abdominal radiographs [4,6,14,15]. Ultrasonographic examination can help refine the clinical picture by detecting the presence of gallstones, fistula tract [14]. It is more sensitive for the detection of pneumobilia than abdominal radiographs [4]. CT scanning is the most sensitive and specific modality to examine for gallstone ileus [4,14,17,18]. Sensitivity and specificity for diagnosing gallstone ileus are high: 93% and 100% respectively [18]. An added benefit of CT scanning with IV contrast is determining the viability of the affected segment of bowel preoperatively as this can help in decision making and guide the therapeutic approach [18]. Despite the availability of the described imaging modalities, the correct preoperative diagnosis is made only in 50% of cases [19]. In our patient, Rigler’s triad was clearly demonstrated in the urgently done enhanced CT scan. The definitive management of this disease is in the form of relief of the bowel obstruction and removal of the impacted stone. Considerable thought must be given to the patient’s preoperative status and therefore patient’s condition must be optimised well before surgery as preoperative status impacts the surgical method to be employed [1].

According to the literature, there are three approaches described for the treatment of gallstone ileus: Enterolithotomy alone, Enterolithotomy coupled with cholecystectomy and fistula closure (one-stage procedure), Enterolithotomy then delayed cholecystectomy at a later time, typically 4–6 weeks (two-stage procedure) [4,6,7].

1 Enterolithotomy involves identification of the stone within the gastrointestinal tract and making a longitudinal incision into the

Table 3
Mortality rates of different operative approaches. Adapted and modified from Kirchmayr et al. [13].

Author	One Stage		Enterotomy	
	Number	Mortality Rate (%)	Number	Mortality Rate (%)
Bloom	5	0	12	3
Braun	2	0	5	4
Clavien	8	25	29	–
Day	3	0	26	5
Deitz	4	0	18	2
Hesselfeldt	2	0	31	6
Heumann	–	–	19	1
Kasahara	16	18	50	43
Keck	11	0	3	–
Lausen	1	0	31	3
Rodriguez	9	33	16	–
Reisner	113	17	801	87
Sandbichler	2	50	16	1
Zuegel	13	8	20	1

bowel at a healthy segment just proximal to the impaction site. The stone is then extracted through the incision site. The remainder of the bowel is then assessed for the presence of other stones. The incision is then sutured in a transverse fashion [3,4,7,20]. This approach is associated with an overall mortality rate of 4.94% [1,5]. However it must be noted that because the fistula tract is left intact, there is a risk of repeated impaction from further stones (5%–33%) [21,22] that would typically occur within 6 months of the initial presentation [23]. Despite these risks, spontaneous fistula closure rate occurs in 50% [12]. To alleviate the risk of repeat obstruction and to aid spontaneous fistula closure, the gallbladder must be assessed for the presence of residual stones as must the entirety of the gastrointestinal tract [1–3]. Table 3, demonstrates the mortality rates of different operative approaches. Adapted and modified from Kirchmayr et al. [13].

2 The one stage procedure has been traditionally reserved for patients who present with less severe disease or have been adequately optimized preoperatively, have less comorbid conditions and are relatively younger [3–5,12]. This approach is associated with longer operative times and may be complicated due to extensive adhesions over the inflamed segment of the biliary tract that requires careful dissection [10,23]. Despite the potential operative difficulty and poor patient status, occasionally the status of the gallbladder (e.g. gangrenous) and presence of gallstones may necessitate index cholecystectomy and fistula closure [25]. Table 2, summarises a review of the literature comparing the mortality rates between one stage procedures and enterolithotomy alone. More recently, the rates of mortality have been reported as 7% [1]. This decrease in mortality rate likely reflects the earlier diagnosis and the improvements in the surgical management and the perioperative care [1,24]. Primary fistula closure is an independent risk factor associated with greater mortality rates and longer lengths of postoperative stay [1].

3 The two-stage procedure is often recommended in more physically fit patients who suffer from persistent biliary symptoms secondary to residual gallstones or the biliary fistula e.g. biliary colic, acute cholecystitis, weight loss and malabsorption [1,6,10,23]. This management modality is infrequently implemented due to low rates of recurrence and unsuitability for further operations in the patient population encountered [26]. The reported in-hospital mortality rate for this procedure was 2.94% [1]. As mentioned previously, we elected to perform an enterolithotomy alone in the patient described in this report. The choice to select this prompt procedure was influenced by two factors: 1) the poor preoperative status of the patient and the presence of multiple comorbidities provided by the consis-

tent support in the literature that minimizes operative time and provides prompt relief of the obstruction, 2) and by the findings of inflammation and dense adhesions that may increase the risks of iatrogenic injuries and increase the operative time as well.

Laparoscopic and laparoscopic assisted enterolithotomy may play a role in the treatment of gallstone ileus and may have added benefits over open surgery in selected patients. The laparoscopic approach essentially emulates the open enterolithotomy approach. The potential advantages are shorter hospital stay and reduced morbidity associated with laparotomy [27]. The prerequisites when attempting laparoscopic surgery are that the surgeon is experienced with the laparoscopic technique and is capable of intracorporeal stitching. There are many precautions that must be taken into consideration such as the effect of pneumoperitoneum and the risks associated with high insufflation pressures, bowel edema making stitching of the incision site difficult, and bowel distension precluding safe port entry. Intraoperatively, the bowel must be thoroughly examined, just as with the open technique [28]. It is also imperative that the bowel is clamped proximally and distally to the site of obstruction to avoid spillage of the bowel contents [28]. Laparoscopic assisted extracorporeal enterolithotomy may act as a bridge to avoid some of the drawbacks associated with the total laparoscopic approach, particularly if expertise with intracorporeal stitching is not available [29].

Moberg et al retrospectively compared the outcomes of laparoscopic assisted and open enterolithotomy in their case series of 32 patients. They reported that those treated laparoscopically suffered only minor complications with similar operative times. The patients included in the laparoscopic group had no history of abdominal surgery [30]. However, only 10% of cases were managed laparoscopically in the review published by Halabi et al. [1]. High conversion rates were also reported particularly when a one-stage procedure was attempted [1]. Therefore, laparoscopic treatment of gallstone ileus may play a role in the treatment of carefully selected patients when an experienced laparoscopic surgeon is available and may result in improved and expedited postoperative recovery.

In conclusion, management of gallstone ileus is mainly surgical and the choice of surgical option depends on the preoperative medical status of the patient, the intraoperative findings and the skill set of the operating surgeon. Knowledge of this entity and familiarity of its clinical manifestation and radiological findings can lead to early diagnosis and better outcome.

Conflict of interest

The authors declare no conflict of interest.

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Ethical approval

The case report was approved by the local ethic committee of the Al-Amiri teaching hospital. LEC-project number 22-2018.

Consent

Written informed consent was obtained from the patient and is available upon request. No patient identifying material was used in this manuscript.

Author contribution

Data collection: Jaffar Hussain, Abdulla D. Behbehani.

Analysis and case management: Asmaa M. Alrashed, Sarah Wood.

Writing and revising of the final version of the manuscript: Salah Termos, Talal Alkhadher.

Registration of research studies

Not applicable.

Guarantor

Salah Termos.

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