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Less is more: an outcome assessment of patients operated for gallstone ileus without fistula treatment

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ABSTRACT

BACKGROUND: The treatment of gallstone ileus (GI) consists of surgical removal of the impacted bilestone with or without cholecystectomy and repair of the biliodigestive fistula. The objective of this study was to assess whether sparing patients a definitive biliary procedure adversely influenced the outcome.

MATERIALS AND METHODS: Patients with a diagnosis of GI were reviewed. Two groups were identified: patients who underwent a definitive biliary procedure with relieving the intestinal obstruction (group 1/G1) and those who did not have a definitive biliary procedure (group 2/G2). In G2, patients were evaluated on long-term follow-up for the risk of recurrent GI disease, cholecystitis, cholangitis and gallbladder cancer.

RESULTS: Among 1075 patients admitted for small bowel obstruction, 20 (1.9%) were diagnosed with gallstone ileus. 3 (15%) of these belong to G1, 17 (85%) to G2. The overall postoperative morbidity rate was 35% (7/20) with one complication exceeding grade II in each group. No deaths were reported. Mean follow-up was 50 months. During follow-up, one of G2 patients had recurrent disease. No biliary tract infections or gallbladder cancer were identified.

CONCLUSION: Enterolithotomy without fistula closure is confirmed to be safe and effective for the management of gallstone ileus both on a short- and long-term basis.

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1. Introduction

Gallstone ileus (GI) is defined as mechanical obstruction of the gastrointestinal tract caused by gallstones that enter the alimentary tract via a biliodigestive fistula. This rare clinical entity complicates less than 0.5% of cases of cholelithiasis and typically follows an attack of acute cholecystitis [1]. Most stones have an uneventful passage through the gastrointestinal tract. However, stones measuring 2.5 cm or greater in size could become lodged at various locations along the tract [2]. Contrast-enhanced CT is considered by far the most useful diagnostic modality as it can clearly pick up Rigler's triad (pneumobilia, intestinal obstruction, and an ectopic gallstone), demonstrate the condition of the gallbladder, and sometimes show the biliodigestive fistula [3–6]. Surgery remains the principal therapeutic modality for this condition. However, the most appropriate surgical approach remains controversial. Some advocate a definitive biliary procedure in the form

of an enterolithotomy combined with cholecystectomy and fistula closure as a single- or two-stage procedure [2,7–9]. Others support a more conservative surgical approach; simple enterolithotomy and preserving any additional surgery for patients with persistent and/or recurrent biliary symptoms [10–15]. At the authors' center, both surgical strategies were used between the years 2005 and 2010. The choice of surgery mainly depended on the surgeon's preference. Since 2010, the policy has changed to adopting a conservative surgical approach in all cases. The aim of the study was to assess the short and long-term outcomes in a series of patients who underwent enterolithotomy without cholecystectomy and fistula closure for GI. This work has been reported in line with the SCARE criteria [16].

2. Materials and methods

All patients with a diagnosis of bowel obstruction treated between January 2005 and November 2016 at the Department of General and Emergency Surgery (Cisanello University Hospital, Pisa, Italy) were identified from the administrative database using the keywords "Intestinal Obstruction"; "Bowel Occlusion" and "Acute Abdominal Pain". All retrieved patients' charts were

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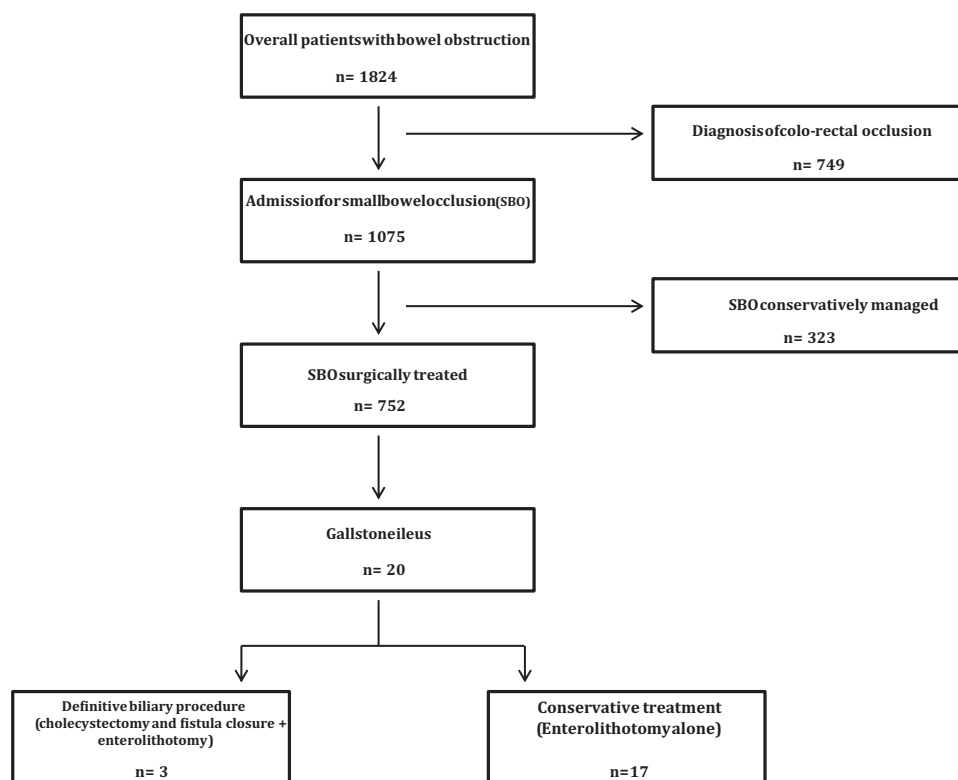


Fig. 1. Patient's flow chart.

reviewed for the final diagnosis and treatment strategy in order to properly identify those with a diagnosis of GI (Fig. 1). The charts of patients with a diagnosis of GI were reviewed for patient demographics and comorbidities; American Society of Anesthesiologists (ASA) physical status score; clinical presentation; diagnostic workup; the surgical strategy undertaken and its rationale; mean operative time; postoperative morbidity and mortality; and length of postoperative hospital stay. Postoperative morbidity was graded according to the Clavien and Dindo classification [17]. Based on the surgical strategy; patients were divided into two groups. Group 1 (G1) underwent a definitive biliary procedure in the form of a cholecystectomy and fistula closure in addition to enterolithotomy. Group 2 (G2) underwent a more conservative surgical approach by simply removing the stone obstructing the small bowel. The long-term outcome in G2 was evaluated focusing on the risk of developing unfavorable sequelae; and the need to undergo a repeat surgery. Potential adverse sequelae considered included: cholecystitis; cholangitis; recurrent gallstone ileus; and gallbladder carcinoma. Follow-up evaluation included irregular interval outpatient clinic visits and/or phone interviews. In some instances; the length of the follow-up exceeded ten years. This study has been worded in line with the PROCESS criteria [18] and with the SCARE guidelines [19]. Data collection and analysis were performed according to the institutional guidelines; and ethical standards of the Helsinki Declaration.

3. Results

During the study period, 1824 patients were admitted with a diagnosis of intestinal obstruction, 1075 of whom (58.9%) with a small bowel obstruction. Of these, 752 (69.9%) were surgically treated. Twenty patients (2.6%) were operated for GI (Fig. 1). Among these, 13 were females, with a female to male ratio of about 2:1. The average age at diagnosis of GI was 83.6 years (range: 73–104 years). The mean ASA score was 3 (range 2–4) and the mean BMI

was 27.7 kg/m² (range 22.6–33 kg/m²). Associated overall comorbidities are reported in Table 1. Five patients (25%) had a history of previous abdominal surgery and six patients had a known history of gallstone disease. Most patients (18/20, 90%) presented with clinical signs of complete intestinal obstruction (abdominal distension, vomiting, closure to feces and air) whereas two patients presented with abdominal pain and inability to pass only feces (Table 2). Plain abdominal x-rays, ultrasounds (US), and contrast-enhanced computed tomography (CT) scans were performed in all patients. Abdominal-US was mainly used to assess the condition of the gallbladder: it revealed an empty gallbladder in all cases. Based on the history, physical exam and imaging studies, a preoperative diagnosis of gallstone ileus was obtained in 17/20 (85%) of cases (Table 2). Rigler's triad was detected in 55% (11/20) of patients.

Surgery was warranted within 12 h from the admission and it was always performed via a laparotomy incision. A definitive biliary procedure was performed in 3 out of the 20 patients (15%), 2 of whom had the entrapped bilestone removed by enterolithotomy and one by segmental bowel resection (G1). In this group the cholecysto-duodenal fistula (3/3; 100%) was closed by cholecystectomy and suture repair of the duodenal wall defect; in one case (1/3), a cholecysto-colonic fistula was also present and was repaired by stapling. A more conservative surgery was offered to patients of G2 although a cholecysto-duodenal fistula was identified at laparotomy. This consisted of an enterolithotomy in 14 patients and a segmental resection in 3 patients. The need for a segmental resection was related to the presence of ischemic areas of the bowel wall at the site of stone impaction (two cases) and to GI complicating a case of Crohn's disease of the ileum in one case. In this series, the mean size of the lodged stones was 4 cm (range 2–5 cm). A single large stone was found in 80% (16/20) patients, and two large stones in the remaining four patients (20%) (Table 2). The level of obstruction was the terminal ileum in 65% (13/20) of patients, the jejunum in 15% (3/20) and was multiple in 20% (4/20). The mean operative

Table 1
Patients' general demography .

	Overall	G1	G2
Mean age (years)	83.6 (range: 73–104)	82.3 (74–88)	83.6 (73–104)
Female/Male	13/7	1/2	12/5
Mean BMI (Kg/m²)	27.7 (range: 22.6–33)	26.9 (24.5–28.9)	27.2 (22.6–32.80)
Mean ASA score	3	2.3	2.8
Overall co-morbidity:	16 (80%)	3 (100%)	13 (76%)
Cardiopathy	8 (40%)	1 (33%)	6 (35%)
Vasculopathy	8 (40%)	1 (33%)	7 (41%)
Arterial hypertension	9 (45%)	1 (33%)	8 (47%)
Pneumopathy	7 (35%)	1 (33%)	6 (35%)
Diabetes	4 (21%)	1 (33%)	3 (19%)
Crohn's disease	1 (5%)	1 (33%)	0
Previous abdominal surgery	5 (25%)	0	5 (29.4%)
Hysterectomy and bilateral salpingoopherectomy	2 (10%)	0	2 (12%)
Ileocolic resection	1 (5%)	0	1 (6%)
Caesarian section	1 (5%)	0	1 (6%)
Small bowel resection	1 (5%)	0	1 (6%)
Appendectomy	1 (5%)	0	1 (6%)
History of Gallbladder Disease			
Present	6 (30%)	1 (33%)	5 (29%)
Absent	14 (70%)	2 (67%)	12 (71%)

Table 2
Perioperative findings.

	Overall	G1	G2
Clinical presentation(n, %):			
Abdominal distension/complete intestinal obstruction	18 (90%)	2 (67%)	16 (94.1%)
Abdominal pain/partial intestinal obstruction	2 (10%)	1 (33%)	1 (6%)
Radiological Exams (RX, US, CT with contrast(n, %):	20	3	17
Gallstone Ileus identification	17 (85%)	3 (100%)	14 (82%)
Presence of Rigler's triad	11 (55%)	1 (33%)	10 (59%)
Biliodigestive fistula identification	0	0	0
Number of retrived/impacted stones(n, %)			
One	16 (80%)	2 (67%)	14 (82%)
Two	4 (20%)	1 (33%)	3 (18%)
Site of bowel obstruction (n, %)			
Terminal Ileum	13 (65%)	2 (67%)	11 (65%)
Jejunum	3 (15%)	0	3 (18%)
Two sites of obstruction	4 (20%)	1 (33%)	3 (18%)
Mean operative time (min)	110.06 (40–285)	228.33 (180–285)	93.5 (40–150)

time in G1 was 228 min (range: 180–285 min), and in G2 it was 93.5 min (range: 40–150 min).

The overall postoperative complication rate was 35% (7/20). The complication rates in G1 and G2 were 67% (2/3), and 29% (5/17), respectively. According to the Clavien – Dindo classification, only 1 patient in each group had a grade IVa complication (Table 3). The mean length of hospital stay was 10 days (range 5–18) in G1 and 8,8 days (range 5–20) in G2 with no 30-day mortality reported (Table 3).

The histologic examination of the resected gallbladders in G1 reported features consistent with chronic cholecystitis, and no evidence of malignancy. The mean follow-up period was 50 months (range: 5–132 months). 13 patients (76%) in G2 were followed up for more than one year (median follow-up time). One of them (6%) developed recurrent disease after nine months from the first episode and underwent enterolithotomy. No cholecystitis, cholangitis or features suggestive of gallbladder cancer were identified however.

4. Discussion

Gallstone ileus is a rare clinical entity, often with a non-specific clinical presentation. The main clinical presentation of GI is intestinal obstruction although a small number of patients (10% in

our series) could show different symptoms, like abdominal pain, potentially mimicking other acute abdominal disorders. Rarity and non-specific clinical signs may be responsible for a diagnosis that is often overlooked or delayed [20]. The literature has reported a 10%-44% accuracy rate of diagnosing the condition preoperatively [7,8,13], and a 3–5 day lag between the onset of symptoms and hospital admission [3,15,21]. A delayed diagnosis and the subsequent lack of timely surgical intervention are contributors to the considerable morbidity and mortality rates reported in patients with gallstone ileus [4,9].

The diagnosis of gallstone ileus can often be made by plain abdominal films that demonstrate Rigler's triad of intestinal obstruction, pneumobilia, and a radio-opaque shadow representing the ectopic gallstone. The prevalence rate of Rigler's triad in this study was 55% (11/20) and this is higher than the figures reported in the literature (4%–35%) [22]. Abdominal-US could be mainly used to assess the condition of the gallbladder: indeed, it may reveal an empty gallbladder and the presence of bowel distension. In our study, CT scan with contrast was able to identify the presence of a GI in a high percent of patients (85%).

The size of gallstone(s) plays a major role in the pathophysiology of gallstone ileus. It has been demonstrated that only larger stones (those greater than 2–2.5 cm) are likely to obstruct the digestive tract and that the level of obstruction is also dependent on the size

Table 3
Postoperative findings.

	Overall	G1	G2
Overall morbidity rate	7 (35%)	2 (67%)	5 (29,4%)
Type of postoperative complications:			
Surgical site infection (Grade II ^a)	4 (40%)	1 (33%)	3 (43%)
Paralytic ileus (Grade II ^a)	2 (20%)	1 (33%)	1 (19%)
Acute renal failure (Grade II ^a)	1 (10%)	0	1 (19%)
Atrial fibrillation (Grade II ^a)	1 (10%)	0	1 (19%)
Cerebrovascular accident (Grade IVa ^a)	1 (10%)	1 (33%)	0
Cardiac arrest (Grade IVa ^a)	1 (10%)	0	1 (19%)
Overall mean post-operative stay (days)	8,9 (range: 5–20)	10 (range 5–18)	8,8 days (range 5–20)

^a According to Clavien and Dindo classification [16].

of the stone [14,23]. It follows the general rule that states: the larger the stone the more proximal the obstruction [24]. In this series, the mean gallstone size was 4 cm (range: 2–5 cm), and the most common site of stone impaction was the terminal ileum (65%; 13/20). The jejunum was the site of obstruction in three patients (15%), while the remaining four patients (20%) had obstruction at more than one level. A cholecysto-duodenal fistula was identified in all patients (100%). In one patient (5%), a cholecysto-colonic fistula was also present.

The high morbidity and mortality associated with gallstone ileus are of major concern when considering the most appropriate management. Surgery is the cornerstone of gallstone ileus management. However, the surgical strategy itself remains a matter of controversy. Surgeons are mainly divided into those who advocate a definitive strategy or two-stage biliary procedure [2,7–9], and those who support a conservative one that only targets the obstructing factor [10–15]. The latter could be in the form of an enterolithotomy or segmental resection, according to the local condition of the obstructed bowel segment. Proponents of the former argue that definitive surgery spares patients potential future complications such as cholecystitis, cholangitis, and recurrent ileus. It also protects them against the risk of developing gallbladder carcinoma. On the other hand, proponents of a conservative surgical strategy argue that the likelihood of requiring additional surgery is minimal in the presence of a patent cystic duct and an empty gallbladder and that patients with gallstone ileus tend to be elderly patients with co-morbidities that could be adversely affected by lengthy and extensive interventions. They also consider the risk of developing gallbladder carcinoma associated with the presence of a biliodigestive fistula as a theoretical one that, to date, has not been supported by evidence [25,26].

All the procedures performed at the authors' center were via a laparotomy incision. The successfulness of laparoscopic enterolithotomy has been reported only in a few single cases in the literature [27–30]. The authors consider patients with gallstone ileus as a high-risk population of elderly patients with co-morbidities that are at risk of a delayed diagnosis; factors that collectively contribute to the significant morbidity and mortality rates associated with this condition. They also consider that additional iatrogenic morbidity and/or mortality could be avoided by sparing these patients lengthy and/or unnecessary procedures.

In the study, it has been noticed that patients who underwent conservative surgery (G2) had a better outcome in terms of postoperative morbidity compared to those who underwent definitive surgery (G1) (67% vs. 29%), suggesting that a shorter operative time and a less extensive surgery reduce postoperative complications. Moreover, in this series, it has been demonstrated that a more conservative surgery is related to a very low rate of recurrent gallstone ileus (6%) and to a lack of evidence of biliary disease like gallbladder cancer, arguments raised by proponents of definitive surgery [14,31–34].

5. Conclusion

Although limited by its retrospective nature and small sample size, this study supports the policy of not addressing the biliodigestive fistula during surgical management of GI. A more conservative surgical approach to GI seems to be non-inferior to definitive surgery in terms of therapeutic effectiveness and safety and with rare adverse long-term sequelae. Elderly patients and patients with comorbidities would benefit from a considerably shorter operative time and a less extensive surgery.

Conflicts of interest

No conflicts of interest to state.

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Ethical approval

Considering that this is a descriptive and retrospective study, no Ethical Approval has been requested.

Author contribution

Dario Tartaglia Study concept and design, data interpretation, drafting, critical revision, accountability for all aspects of the work.

Sohail Bakkar Study concept and design, data interpretation, article writing, critical revision, accountability for all aspects of the work.

Jessica Bronzoni Data collection and interpretation, final approval, accountability for all aspects of the work.

Lorenzo Piccini Data interpretation and analysis, final approval, accountability for all aspects of the work.

Luigi Cobuccio Data collection and interpretation, final approval, accountability for all aspects of the work.

Andrea Bertolucci Data collection and interpretation, final approval, accountability for all aspects of the work.

Christian Galatioto Study concept and design, critical revision, final approval, accountability for all aspects of the work.

Massimo Chiarugi Study concept and design, critical revision, final approval, accountability for all aspects of the work.

Guarantor

Dario Tartaglia, MD, University of Pisa, Italy.

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