Gallstone ileus: a review

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ABSTRACT
Background Gallstone ileus is an important complication of cholelithiasis. In general, surgery is the treatment of choice for such cases, but clinicians face difficulty in the selection of an appropriate approach. Closure of a cholecystoenteric fistula can be achieved through one-stage or two-stage operation. Two-stage operation has a lower mortality rate than one-stage procedure, but persistence of the cholecystoenteric fistula is associated with the risk of carcinogenesis and recurrence of gallstone ileus.

Objective This study reviews the different surgical approaches according to the impaction site of the gallstone, using data of previous studies by our group and clinical reports in the literature.

Conclusions First, for cases involving impaction at the duodenum, the cholecystoenteric fistula can be repaired in the same surgical field, and one-stage operation obtains favourable outcome; hence, one-stage operation is considered as treatment of choice. Second, for cases involving impaction at the small intestine, natural closure of the cholecystoenteric fistula or low mortality is expected; hence, two-stage operation may be performed, possibly using minimally invasive laparoscopy. Third, for cases involving impaction at the colon, natural closure of the cholecystocolonic fistula is unlikely, and patients have a high risk of reflux cholangitis due to faecal fluid; hence, one-stage operation is considered as treatment of choice.

INTRODUCTION
Gallstone ileus is a relatively rare complication of cholelithiasis and a type of mechanical ileus involving impaction of gallstone in the intestinal tract. Owing to the high prevalence of gallstones, such cases may be presented in routine medical practice, and clinicians should consider gallstone ileus in the differential diagnosis of acute abdomen. However, selection of appropriate treatment is difficult due to poor general condition of the patients with ileus, and physicians need to make a decision on the choice of immediate one-stage or two-stage closure of the cholecystointestinal fistula or waiting for natural closure.

My previous studies reported the experience with cases of gallstone ileus with different sites of impaction, including the disease treatment. These studies indicate that in each patient, the choice of surgical approach should be based on the site of gallstone impaction. This paper describes a treatment approach for gallstone ileus based on findings of current reports.

EPIDEMIOLOGY
Gallstone ileus occurs in 0.15%–1.5% of cholelithiasis cases and <0.1% of ileus cases overall, and the recurrence rate is 5%–8%.

PATHOGENESIS
In patients with gallstone ileus, the gallstones are usually eliminated via an internal biliary fistula. In majority of cases (75%–83%), the elimination route is from the gallbladder to the duodenum. However, elimination may proceed from the gallbladder to the colon or stomach, or in extremely rare cases, naturally via the bile duct, involving passage from the gallbladder to the common bile duct and subsequently to the papilla of Vater. Based on these facts, this study focused only on internal biliary fistula classified as cholecystointestinal fistula. In many cases, an internal biliary fistula forms due to vascular insufficiency or chronic inflammation caused by compression of the gallbladder wall during calculus formation, which allows elimination of the calculus.

The most common impaction site of the gallstone is the ileum (50.0%–60.5%), jejunum (16.1%–26.9%), duodenum (3.5%–14.6%) and colon (3.0%–4.1%), in order. Bouveret syndrome, named after the physician who first discovered the condition, refers to impaction in the duodenal bulb that causes gastric outlet obstruction. The impacted gallstones are mainly of the cholesterol type and approximately 4 cm in size, which is within the ≥2.5 cm range considered as impaction prone.

DIAGNOSIS
Gallstone ileus has no distinctive symptoms, but tumbling gallstone advancement that results in alternating aggravation and resolution of ileus is a relatively common feature. Many patients with gallstone ileus are elderly, present with comorbidities, have
poor general condition and delayed diagnosis that leads to dehydration, shock, sepsis, or peritonitis.12

In a review of 176 cases (49 male and 127 female patients; age, 24–91 years) of gallstone ileus, Nakao et al9 reported abdominal pain at presentation in majority of the patients (91.5%) with accompanied abdominal distension, vomiting and fever in 84.7%, 59.7% and 40.9% of all patients, respectively.

In a review of 128 cases of Bouveret syndrome, Cappell and Davis13 reported the mean age of patients of 74.1 years, a larger proportion of female patients (female-to-male ratio of 1.86), and clinical symptoms of nausea and vomiting (87%), abdominal pain (71%), haematemesis (15%), weight loss (14%) and poor appetite (13%).

In a systematic review of 38 cases of sigmoid gallstone ileus, Farkas et al14 reported the age range of patients of 65–94 years, mean age of 81.1 years, and nearly threefold higher proportion of female patients than of male patients; regarding clinical symptoms, 74% of the patients presented abdominal pain, whereas 93% of the patients presented other obstructive symptoms of constipation (61%), vomiting (50%) and abdominal distension (26%).

Rigler et al15 reported that pneumobilia and intestinal tract dilatation and specularity, calculus within the intestinal tract, and calculus movement observed on plain abdominal radiography are useful for diagnosis of the condition; however, the combined signs only appeared in <50% of cases.5 9

In another review, Cappell and Davis13 reported the frequency of the plain radiographic findings as follows: pneumobilia, 25%; gastric dilatation, 15%; calcified mass or calculus around the gallbladder or in the upper right abdominal area, 24%; moreover, those findings were confirmed on CT scans at frequency of 60%, 20% and 90%, respectively, which indicates that advances in CT could significantly improve the diagnosis rate.

The imaging findings of intestinal tract dilatation showed similar frequency in both abdominal radiography and CT (88% vs 92%); however, CT achieved more than twofold higher rate of diagnosis of ectopic gallstone than radiography (33% vs 81%),16 which suggests that CT is useful for visualisation of pneumobilia and intestinal calculi. Several case reports have indicated that contrast-enhanced CT achieves high sensitivity (95%), specificity (100%), and accuracy (99%) in the diagnosis of gallstone ileus.17 The presence of a fistula can be diagnosed by drip infusion cholangiographic CT to confirm leakage of contrast medium from the biliary system into the gastrointestinal tract. In addition, abdominal ultrasonography can visualise impacted calculus and presence of cholelithiasis. Upper gastrointestinal endoscopy can be used to confirm the diagnosis of impacted calculus and internal biliary fistulas such as cholecystointestinal fistulas.

The differential diagnosis includes adhesive ileus in patients with history of abdominal surgery or abdominal radiotherapy, dietary ileus, ileus accompanying gastric calculus, ileus due to ingestion of a foreign object undetectable through radiography, and ileus due to malignant tumour.

**TREATMENT**

**Principles of treatment**

In general, since natural stone exclusion occurs at a low rate of approximately 1.3%,9 surgery is indicated for cases of gallstone ileus involving both stone extraction to resolve the ileus and closure of the fistula. However, whether surgery should be performed as a one-stage or two-stage operation remains unclear. Based on a study in 1994, Reisner and Cohen1 indicated that despite mortality rate in simple stone extraction of 11.7%, one-stage surgery shows higher mortality rate of 16.9% with a higher level of invasiveness. Based on a study in 2014, Halabi et al17 reported significantly increased length of hospital stay and mortality rates in patients who underwent one-stage internal biliary fistula closure; for patients with ileus, one-stage surgery is excessively invasive, and associated postoperative complications are common. Nevertheless, those studies assessed overall cases of gallstone ileus regardless of the impaction site, and majority of the cases included showed impaction at the small intestine.

With regard to two-stage surgery for gallstone ileus, the initial procedure involves simple stone extraction, followed by fistula closure performed separately. However, two-stage surgery has a disadvantage of associated risk of recurrence, retrograde cholecystitis, and cancer development due to remaining cholecystitis, fistula. Surgeons should consider three related issues as follows.

**Effects of persistent cholecystointestinal fistula**

Persistence of cholecystointestinal fistula is a potential causal factor for retrograde cholecystitis or gallbladder cancer. The cholecystointestinal fistula remained untreated in 86.7% of cases of recurrent gallstone ileus, while cholecystectomy is necessary indication in patients with symptomatic gallstone. A total proportion of 11% of cholecystoduodenal fistula and 60% of cholecystocolonic fistula resulted in cholangitis,18 19 and 15% of cases of fistula showed complication of gallbladder cancer.20

**Natural closure of cholecystointestinal fistula as a possible outcome**

Kaneda et al21 reported that natural fistula closure without treatment of the biliary tracts occurred in 61.5% of cases, which suggests that when planning two-stage surgery, natural closure of the fistula after lithotomy should be expected. However, there are no established guidelines for the waiting time until natural closure. Ráf and Spangen22 proposed radical surgery for cases without natural closure 3–6 months after simple lithotomy. Mir et al23 reviewed recurrent cases of ileus and reported that 85% of cases manifest some symptoms of recurrent gallstone ileus within 6 months. Considering the follow-up period required for recurrence assessment, radical treatment is indicated in patients for who natural closure is not observed 6 months postoperatively. However, one case report indicated
absence of fistulous opening under endoscopy at postoperative 2 months, which suggests natural closure, but confirmed patency of the opening through endoscopic retrograde cholangiography at postoperative 4 months. Based on these results, in patients with expected natural closure under conservative treatment, clinicians should perform careful follow-up evaluation and provide sufficient explanation to the patients.

Lithotomy with extracorporeal shock wave lithotripsy and endoscopy

Recently, simple lithotomy is increasingly performed in two-stage surgery involving endoscopic lithotomy and lithotripsy for duodenal impaction. Endoscopic extraction is commonly performed in patients with impaction of the duodenum or colon, but achieves a low success rate. Lithotripsy is classified into various types as follows: mechanical, electrohydraulic, extracorporeal shock wave lithotripsy (ESWL), and that using Yttrium Aluminum Garnet (YAG) laser. Lithotomy with ESWL showed poor performance in patients with obesity and gas-containing bowel loops. Endoscopic treatment approach may not be effective, based on reports of reimpaction of fragments of the calculus in patients who underwent lithotripsy. These collective findings indicate that although the treatment is less invasive than surgery, it is less reliable. Nevertheless, endoscopic modality is considered as a treatment option in elderly patients or those judged as unable to withstand surgery.

Based on these facts, clinicians should consider the importance of planning the treatment strategy according to the site of gallstone impaction; each specific surgical strategy is highlighted in the following section.

Involvement of impaction at the duodenum

In cases of duodenal impaction, natural stone exclusion is rare, and almost all cases are treated surgically; one-stage surgery is commonly performed in these patients, due to the fact that lithotomy can be performed through the resection site of the fistulous opening, and cholecyst extraction and fistula closure can be performed at the same surgical field after lithotomy, which enables relatively simple technical procedure of one-stage surgery. A previous study including 23 cases of duodenal impaction in Japanese patients indicated that favourable postoperative outcomes are achieved by one-stage surgery.

In particular, in cases of Bouveret syndrome involving impaction of the duodenal bulb, the surgeon attempts lithotomy from the fistulous opening resection site; for impaction at the third portion of the duodenum, if the stone can be manually moved to the small intestine, stone extraction through the resection site of the small intestine is associated with a lower risk of postoperative complications and is considered to be less invasive than duodenotomy. For cases of cholecystoduodenal fistula treated with lithotomy from a site other than the fistulous opening, one-stage closure may have fatal outcome due to postoperative complications such as duodenal anastomotic leakage depending on the patient’s general state and underlying disease; therefore, surgeons should consider performing lithotomy alone only in exceptional circumstances. If the duodenum is resected, additional surgery involving an omental patch or small intestine serosal patch may be required.

Involvement of impaction at the small intestine

Based on a study in 2003, Doko et al reported the findings of 30 cases of gallstone ileus including one case of duodenal impaction case, categorised into two treatment groups of enterolithotomy alone or one-stage surgery; their results indicated a morbidity of 27.3% and 61.1% for the enterolithotomy alone group and one-stage surgery group, respectively, and mortality of 9% and 10.5% for those respective groups. Moreover, both groups attained similar scores in the American Society of Anesthesiologists grading system, but the one-stage surgery group showed significantly longer operating time.

Urgent fistula repair is associated with significant postoperative complications. A study of Halabi et al in 2014 including a large database of 3268 cases of gallstone ileus revealed significantly longer hospital stay and higher mortality rate for patients who underwent the one-stage procedure for internal biliary fistula closure. Based on those findings, two-stage surgery is considered as the standard method for cases of gallstone ileus and those involving impaction at the small intestine, since two-stage surgery achieves lower mortality rate than one-stage surgery. In two-stage surgery, the patient’s general condition can be improved by first releasing the ileus by lithotomy, and the biliary system can also be investigated in detail. In addition, two-stage surgery is a relatively less invasive approach for lithotomy via small intestinal resection. Currently, lithotomy is performed under laparoscopic assistance, and for cases without confirmed natural closure of the fistula subsequently, two-stage cholecystointestinal fistula closure may be performed. In cases of cholecystoduodenal fistula, the probability of natural closure is high, and surgery involving lithotomy alone is considered as method of choice; however, regular patient follow-up is required due to the risk of remaining cholecystointestinal fistula.

Based on a study in 2007, Moberg and Montgomery reported that for cases of gallstone ileus involving impaction of the small intestine that underwent enterolithotomy, laparoscopic surgery achieves shorter hospital stay than laparotomy, and both procedures showed no cases of mortality.

A previous study reported my experience of cases treated with two-stage laparoscopic procedure comprising both simple lithotomy and cholecystointestinal fistula closure; the results indicate that the approach allows lower level of invasion than that associated with laparotomy.

Involvement of impaction at the colon

For cases with impaction of the colon, selection of non-surgical treatment may be indicated, but 74% of cases with non-surgical treatment show failure. One review indicated that if surgery is selected, one-stage surgery is performed in majority of the cases. The sigmoid colon
is the most common impaction site, mainly due to presence of intestinal stenosis caused by diverticulitis. However, some studies reported cases caused by gynecological diseases and inguinal hernia. In such cases, cholecystocolonic fistula is the most common predicted route. A previous report provided evidence-based recommendation for one-stage surgery in cases of gallstone ileus accompanied by cholecystocolonic fistula. This is supported by the lower natural closure rate of 10% for cholecystocolonic fistula versus the other types of cholecystoenteral fistula and the high risk of retrograde cholecystitis due to reverse flow of faecal fluid from the cholecystocolonic fistula.

CONCLUSIONS

In the treatment approach for cases of gallstone ileus, selection of surgical method should be based on the impaction site. Two-stage surgery is recommended for cases with impaction at the level of the small intestine, while one-stage surgery is recommended for cases with impaction at other sites. However, surgeons should judge the feasibility of one-stage surgery on the basis of the patient’s general condition.

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