

Gallstone ileus associated with impaction at Meckel's diverticulum: Case report and literature review

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Abstract

Gallstone ileus due to erosion of one or more gallstones into the gastrointestinal tract is an uncommon cause of small bowel obstruction. The site of impaction is usually distal ileum, and less commonly the jejunum, colon, duodenum, or stomach. We report a rare case of gallstone ileus with impaction at the proximal small bowel and at a Meckel's diverticulum (MD) in a 64-year-old woman managed with laparoscopic converted to open small bowel resections. Patient was discharged home in stable condition and remained asymptomatic at 6-mo follow up. We review the current literature on surgical approaches to MD and gallstone ileus. Diverticulectomy or segmental resection is preferred for complicated MD. For gallstone ileus, simple enterolithotomy or segmental resection are the most the most favored especially in older co-morbid patients due to lower mortality rates and the rarity of recurrent gallstone ileus. In addition, laparoscopy has been increasingly reported as a safe approach to manage gallstone ileus.

Key words: Gallstone ileus; Meckel's diverticulum; Small bowel obstruction; Laparoscopy; Cholecystoenteric fistula; Laparoscopy

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Core tip: Gallstone ileus is an uncommon cause of small bowel obstruction in the population at large but is responsible for up to a quarter of mechanical bowel obstructions in the elderly in the United States. We report a rare case of gallstone ileus with impaction at the jejunum and at a Meckel's diverticulum in a 64-year-old female managed by laparoscopic converted to

open segmental bowel resections. We review current literature comparing surgical procedures for Meckel's diverticulum and gallstone ileus.

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INTRODUCTION

We are reporting a case of gallstone ileus involving multiple gallstones managed by laparoscopic converted to open small bowel resections of impacted stones at the jejunum and at a Meckel's diverticulum (MD). MD is the most common congenital anomaly of the gastrointestinal tract and is caused by the incomplete obliteration of the omphalomesenteric duct^[1]. It can be found in nearly 2 percent of the population but in most cases it remains asymptomatic^[2]. Most reported complications include bleeding, infection and obstruction^[3]. Gallstone ileus is an uncommon cause of obstructions in the general population but is responsible for 25% of mechanical bowel obstructions in the elderly^[4-6]. We found only 3 other case reports with mention of gallstone impaction at MD^[7-9].

CASE REPORT

A 64-year-old morbidly obese woman with a history of diabetes, hypertension, and atrial fibrillation presented to an outside hospital (OSH) for PO intolerance with nausea, bilious emesis, and post-prandial abdominal pain. Computerized tomography (CT) of abdomen and pelvis demonstrated small bowel obstruction. Hepatobiliary iminodiacetic acid scan was performed and demonstrated probable mass in the second portion of the duodenum. She was transferred to our institution for escalation of care where she was found to be tachypneic, tachycardic and in atrial fibrillation. On abdominal exam she was distended and mildly tender without rebound or guarding. Her white blood cell count was 14.9 without left shift. She was admitted to the medical intensive care unit for respiratory failure and metabolic acidosis in the setting of frequent bilious emesis.

Repeat CT of the abdomen and pelvis at our institution demonstrated pneumobilia, small bowel dilation and intraluminal small bowel filling defects consistent with cholecystoduodenal fistula with gallstone ileus. Three gallstones were identified, one in the jejunum and two in the ileum along with a mechanical small bowel obstruction with a transition point near the distal culci in the distal jejunum/proximal ileum (Figure 1). She was taken to the operating room for laparoscopic small bowel resection.

During laparoscopy, bleeding from the deep inferior epigastric vessels necessitated conversion to laparotomy. The small bowel was then run and a large gallstone was found to be obstructing the distal jejunum. We also identified a MD impacted with two smaller stones. A longitudinal incision was made in the jejunum to remove the stone and perform an enterolithotomy. However due to significant edema and inability to milk the stone distally, a small bowel resection was performed.

Once this was complete, we turned our attention to the MD and performed a small bowel resection to include the MD with approximately 5 cm of adjacent small bowel. This resection was performed in lieu of a diverticulectomy due to concern about narrowing of the small bowel lumen. At the end of the surgery the patient required pressor support. She was kept intubated and transferred to the surgical intensive care unit.

Patient recovered bowel function on postoperative day 4 but her postoperative course was remarkable for a midline incision hematoma secondary to treatment with therapeutic Lovenox for previous history of atrial fibrillation. She required wound opening, evacuation, and packing. Patient subsequently remained stable on Lovenox without further bleeding episodes and was eventually discharged in stable condition to a skilled nursing facility. Upon follow up, 6 mo later, patient was asymptomatic.

Pathological examination of the surgical specimen demonstrated mucosal ulceration and transmural inflammation of both of the resected bowel segments. The stone found in the jejunum was identified as a mixed type gallstone measuring 4.7 cm × 3.2 cm × 3.2 cm and the stones found at the MD were identified as mixed type gallstones measuring 4.0 cm × 2.7 cm × 2.7 cm and 2.5 cm × 2.0 cm × 1.4 cm (Figure 2).

DISCUSSION

Gallstone ileus is an uncommon complication, occurring in 0.3% to 0.5% of all cases of cholelithiasis, and accounting for 1% to 4% of mechanical small bowel obstructions. However, while gallstone ileus is rare in the general population, it accounts for 25% of mechanical bowel obstructions in patients over 65 years of age in the United States^[4-6]. Because of the advanced age at presentation, patients often have multiple comorbidities, which contribute to the high morbidity and mortality associated with gallstone ileus. The pathophysiology of gallstone ileus involves the erosion of one or more gallstones from a chronically inflamed gallbladder into the gastrointestinal tract, creating a cholecystenteric fistula. Gallstones less than 2 to 2.5 cm generally pass into the intestine without causing obstruction while stones 5 cm or larger are more likely to impact usually at the distal ileum, the narrowest part of the small bowel^[10]. Other reported sites of impaction include proximal ileum, jejunum, colon, and rarely the duodenum or stomach (Bouveret's syndrome)^[11]. In our case, a large,

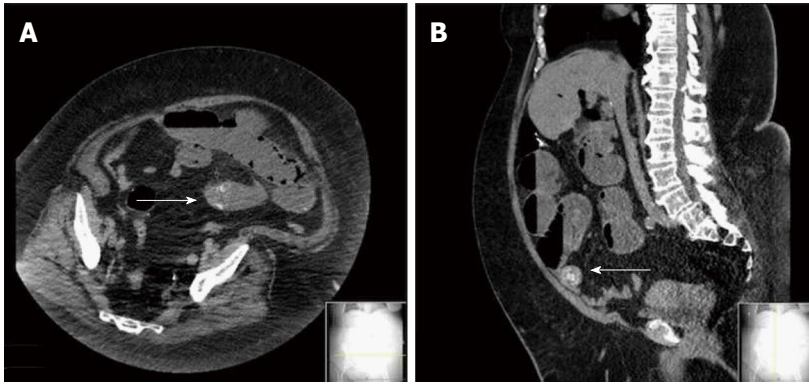


Figure 1 Computed tomography of abdomen pelvis without contrast. A: Axial section demonstrating 31.8 mm gallstone (arrow) in jejunum; B: Sagittal section demonstrating transition point and smaller gallstone (arrow) at proximal ileum.



Figure 2 Gross specimen of Meckel's diverticulum impacted with two gallstones.

approximately 5 cm, gallstone was found impacted at the jejunum while two smaller stones were found impacted at a MD. Clinical presentation of gallstone ileus is variable and often insidious. Patients can have painless intervals due to “tumbling” or incomplete small bowel obstruction in which the impacted stone intermittently passes and lodges in the intestinal lumen, until the stone either passes through the gastrointestinal tract or is impacted. It is possible this pattern of remitting symptoms may have contributed to the delay in diagnosis for our patient after her initial presentation to the OSH.

MD is a common congenital anomaly of the small intestine occurring in up to 3% of the population, typically 55 cm from the ileocecal valve^[8]. It is usually asymptomatic, with a low lifetime risk of developing complications, with most occurring later in adulthood. Most reported complications include obstruction, hemorrhage, perforation, and neoplasia^[8]. Small bowel obstruction can be caused by intussusception or volvulus of MD. In rare cases enteroliths, can form within a MD and cause an obstruction from impaction within the small bowel. Stasis in the diverticulum in combination with the alkalotic environment of the small bowel can promote precipitation of calcium and is

thought to contribute to formation of an MD enterolith^[7]. In our case the patient had an impaction of a gallstone in the MD. This is exceptionally rare with only 3 cases having been reported in the literature.

Impaction of a gallstone at MD can cause intermittent abdominal pain, bleeding, diverticulitis, perforation or small bowel obstruction (SBO). The presence of stones in a MD predispose to SBO by promoting local inflammation of the diverticulum and intussusception or by impaction of the stone in the bowel following its extrusion from the diverticulum. Clinical differentiation of MD enterolith from gallstone ileus can be difficult as both present similarly with bowel obstruction with prolonged indolent course. Pneumobilia on abdominal plain film suggests gallstone ileus but abdominal film has very low sensitivity for detecting gallstone ileus or MD enterolith^[12]. CT is the ideal diagnostic modality with sensitivity, specificity and diagnostic accuracy of over 93%^[13]. However, gallstone ileus is often diagnosed only at the time of laparotomy in up to 25% to 50% of cases^[5,14,15].

The surgical management of an incidentally found MD in the adult population remains controversial. Most surgeons would advise removing an asymptomatic diverticulum when found incidentally at laparotomy in pediatric patients and young adults secondary to the seemingly significant risk of developing complications. However, the literature is less decisive regarding prophylactic resection in adults. For example, Peoples *et al.*^[16] found a high morbidity and mortality rate associated with resection but low lifetime risk (6.2%) of developing symptoms from a MD, with the majority of complications occurring during the first 2 decades of life. Cullen *et al.*^[17] demonstrated operative morbidity and mortality for elective MD resection (2% and 1%) was significantly lower as compared to non-elective resection (12% and 2%)^[17]. However, Zani *et al.*^[18] pointed out 758 resections would have to be performed to prevent one death when consideration is given to the overall low number of patients affected by MD. Therefore, incidental diverticulectomy is generally discouraged, with exception in cases of narrow base,

long length, and palpable heterotopic tissue, where operative management is given special consideration.

On the other hand, it is more uniformly accepted that complicated and symptomatic MD requires operative management. The operative management options for MD with gallstone impaction include gallstone fragmentation and milking into the proximal colon, or gallstone removal through an enterotomy. The diverticulum itself should also be resected to prevent recurrent stone formation and further complications. The decision to perform a diverticulectomy or segmental resection remains contested. The diverticulum can be easily resected with a stapler without entering the bowel's lumen^[19]. However, bowel resection with primary anastomosis is indicated in cases of inflammation, perforation, and necrotic bowel. As in our case, if the small bowel lumen is in danger of being narrowed or the neck of the diverticulum is wide, a segmental resection is favored over a simple diverticulectomy^[8]. As far as we know, there are no studies directly comparing diverticulectomy with segmental resection.

The operative management of gallstone impaction of the small bowel are: (1) enterolithotomy, cholecystectomy and fistula repair (single-stage surgery); (2) enterolithotomy with delayed cholecystectomy and fistula closure (two-stage surgery) and (3) simple enterolithotomy (most reported surgical procedure). Proponents of the single-stage procedure cite recurrence and increased risk of developing cholangitis or gallbladder carcinoma as reasons for performing concurrent cholecystectomy and fistula closure^[20]. Those who support the two-stage procedure or simple enterolithotomy point to high mortality rates of single-stage procedures and low rates of recurrence and gallbladder carcinoma as reasons for deferring cholecystectomy or avoiding it altogether. Furthermore, fistulas have been shown to close spontaneously once the distal obstruction is removed^[20].

No randomized trial has been performed to address the question of appropriate treatment due to the ethical implausibility of randomizing patients to one group over the other. The literature on gallstone ileus is largely limited to retrospective studies or case series (Table 1). In one of the largest studies comparing outcomes between single stage and simple enterolithotomy, Reisner *et al*^[5] reviewed 1001 cases and found that a single-stage procedure had a higher mortality rate at 16.9% compared to 11.7% with simple enterolithotomy ($P < 0.17$). Moreover, recurrence rates, from retained stones missed during initial surgery or formation of new gallstones, were the same in both groups.

Thus, the one-stage procedure, while associated with higher mortality rates, did not reduce recurrence rates as its proponents have predicted. Confirming these findings is a review by Halabi *et al*^[21] which now exceeds Reisner and Cohen as the largest review of gallstone ileus. They used the Nationwide Inpatient Sample from 2004 to 2009 to compare data for 3268

Table 1 Mortality rates of two main surgical approaches in treating gallstone ileus, one-stage and two-stage procedure

Ref.	With cholecystectomy (single-stage)		Without cholecystectomy (two-stage)		Total
	Total	Mortality	Total	Mortality	
Kasahara <i>et al</i> ^[15]	105	19%	7	0%	112
Reisner <i>et al</i> ^[5]	113	16.8%	801	11.7%	1001
Doko <i>et al</i> ^[36]	19	10.5%	11	9.1%	30
Tan <i>et al</i> ^[37]	12	0%	7	0%	19
Mallipeddi <i>et al</i> ^[23]	14	7.1%	113	5.3%	127
Halabi <i>et al</i> ^[21]	741	7.3%	2527	6.5%	3268

patients. They found fistula repair and bowel resection to be independently associated with higher mortality rates and longer hospital stays when compared to simple enterolithotomy repair with an odds ratio of 2.86 (95%CI: 1.16-7.07) and 3.68 (95%CI: 1.59-5.76), respectively. In support of these studies are reviews by several other authors showing a preference for reserving the higher risk single-stage procedure for patients with lower ASA classifications^[14,22,23].

Just as with initial presentation, management options for recurrent gallstone ileus include simple enterolithotomy, single-stage, and two-stage surgery. In a systematic literature review, Mir *et al*^[24] compared treatment options in patients with recurrent gallstone ileus over the last 25 years and found a significantly lower mortality rate of simple enterolithotomy when compared to single-stage surgery (4.8% vs 22.2%). Several case reports of recurrent gallstone ileus successfully managed by repeat enterolithotomies lend support to this approach^[25-27].

A new development in the surgical management of gallstone ileus is the use of laparoscopy. There is to date one retrospective review of laparoscopic assisted vs open enterolithotomy of gallstone ileus by Moberg *et al*^[28]. Both groups had a similar duration of operation (60 min vs 58 min), similar median hospital stay (10 d vs 7 d), and similar complication rates (6 vs 5), and no deaths. In support of this is the publication of several recent case reports demonstrating the successful use of laparoscopic assisted surgery for gallstone management^[29-31]. Moreover, many case reports have been published on the efficacy of a totally laparoscopic approach in the management of gallstone ileus^[32-37]. However, in these reports only a single stone was involved. Our attempt at laparoscopic enterolithotomy was limited by bleeding from injury to the epigastric artery during port insertion and was complicated by involvement of multiple stones.

While mortality rates remain high for patients with gallstone ileus they are overall lower in more recent literature, which is likely a reflection of improved modern surgical and peri-operative care. A laparoscopic approach may be suited for the uncomplicated patient with a single stone who can tolerate the longer operative time required to close the enterolithotomy. However, com-

plicated cases such as impaction at a Meckel's diverticulum in a morbidly obese high-risk patient will benefit from an open approach. In such patients, diverticulectomy or segmental bowel resection of the should be strongly considered. These patients then have the option of undergoing an elective cholecystectomy at a later time. Patients who develop recurrence can be managed similarly. A single-stage procedure is rarely performed, typically in lower risk patients, or those with conditions requiring urgent attention to the gallbladder. It is associated with high mortality and morbidity and the decision to perform a single-stage procedure should be weighed carefully against the perceived benefits.

COMMENTS

Case characteristics

A 64-year-old female morbidly obese female with a history of diabetes, hypertension, an atrial fibrillation presented with post-prandial abdominal pain and bilious emesis.

Clinical diagnosis

Gallstone ileus with impaction in small bowel and at Meckel's diverticulum (MD).

Differential diagnosis

The differential diagnosis in this patient involves other causes of small bowel obstruction and mesenteric ischemia, which includes.

Laboratory diagnosis

Leukocytosis with left shift and metabolic acidosis were found in setting of frequent bilious emesis.

Imaging diagnosis

Hepatobiliary iminodiacetic acid scan demonstrating probable mass in small bowel and CT of abdomen and pelvis demonstrating pneumobilia, small bowel dilation and intraluminal small bowel filling defects were consistent with gallstone ileus.

Pathological diagnosis

Surgical small bowel specimen demonstrated distal jejunum with 4.7 cm stone and MD with two gallstones measuring 4 and 2.7 cm.

Treatment

Operative management with laparotomy, enter lithotomy of impacted jejunum, and small bowel resection of impacted MD.

Related reports

Other case reports of gallstone ileus associated with impacted MD have been very rarely presented in the literature. To our knowledge only three cases (two in English and one in Danish) have been published with varying presentations and varying treatment modalities.

Experiences and lessons

Gallstone ileus, while a rare occurrence over all is a more common cause of small bowel obstruction in the elderly and carries a high rate of morbidity and mortality making early clinical suspicion and intervention very important.

Peer-review

This is an important presentation of a rare finding that explores the appropriate management of gallstone ileus in an elderly co-morbid patient with an incidental finding of MD. It is accompanied by a very thorough and well written literature review of management of both symptomatic Meckel's diverticulum and gallstone.

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