

Case Report

Single stage totally laparoscopic management of Barnard's syndrome: the first such case report with review of literature

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ABSTRACT

Gall stone ileus is an uncommon however important complication of cholelithiasis. This condition is seen commonly in elderly females with comorbidities. It results from the passage of stone into the small bowel from the gall bladder through a cholecysto-duodenal fistula. Presenting symptoms include typical features of small bowel obstruction. Diagnosis can be made radiologically. Surgical management is the mainstay of treatment. The procedure can be done either in a single stage, where the stone retrieval is done through enterotomy followed by cholecystectomy and fistula repair; or in two stages, which comprise of enterolithotomy alone in stage 1 followed by interval cholecystectomy and fistula repair in stage 2. Laparoscopic single stage management enables definitive surgical therapy of the condition during a single hospital stay and under a single session of anaesthesia.

Keywords: Gall stone ileus, Cholecysto-duodenal fistula, Small intestine obstruction, Laparoscopic management

INTRODUCTION

Gallstone ileus (GSI) is an unusual complication of cholelithiasis and is defined as a mechanical intestinal obstruction due to impaction of one or more gallstones within the gastrointestinal tract. The term gall stone ileus is a misnomer, since 'ileus' refers to non-mechanical, adynamic, functional obstruction whereas GSI related obstruction is a true mechanical phenomenon.¹ The gall stone reaches small bowel usually after passing through biliary-enteric fistula typically formed between the gall bladder and duodenum.² GSI is observed with higher frequency among elderly and due to predominance of gall stone disease in females, majority of GSI patients happen to be females.^{1,3}

Surgery is the mainstay of treatment, without a clear consensus about the best surgical approach. Here, we present a case of an elderly lady with GSI, managed totally by laparoscopy, in a single stage procedure, which included enterolithotomy, cholecystectomy and fistula repair.

CASE REPORT

An 82 years old female presented to the emergency ward with acute colicky abdominal pain and distension, multiple episodes of vomiting, constipation since 3 days and obstipation since 1 day. She did not have any addictions, had been a homemaker and did not have any previous surgical scar on her abdomen. Her general examination revealed a pulse of 92 beats/minute, blood pressure of 130/90 mmHg and a respiratory rate of 12/minute. On systemic examination she was obese, with a hugely distended tympanic abdomen with diffuse generalised significant tenderness all over. She had hyperperistalsis in the lower abdomen. A per rectal exam revealed an empty rectum. She had a leukocytosis of 12500/cumm. Her serum creatinine was 0.9 mg/dl and liver profile was within normal limits. Her plain X-ray abdomen done in standing position showed multiple air-fluid levels in a classical 'step-ladder' pattern, suggesting small bowel obstruction (Figure 1 A). A contrast enhanced computed tomography (CECT) scan of the abdomen was then done. It revealed a 4x3 centimeters (cm) large oval lamellated stone in the terminal ileum

with dilated contrast filled small bowel noted proximally (Figure 1B and C). The CECT also revealed pneumobilia along with a cholecysto-duodenal fistula (Figure 1D and E). Thus, a diagnosis of GSI was reached. She was then planned for a one stage laparoscopic intervention.

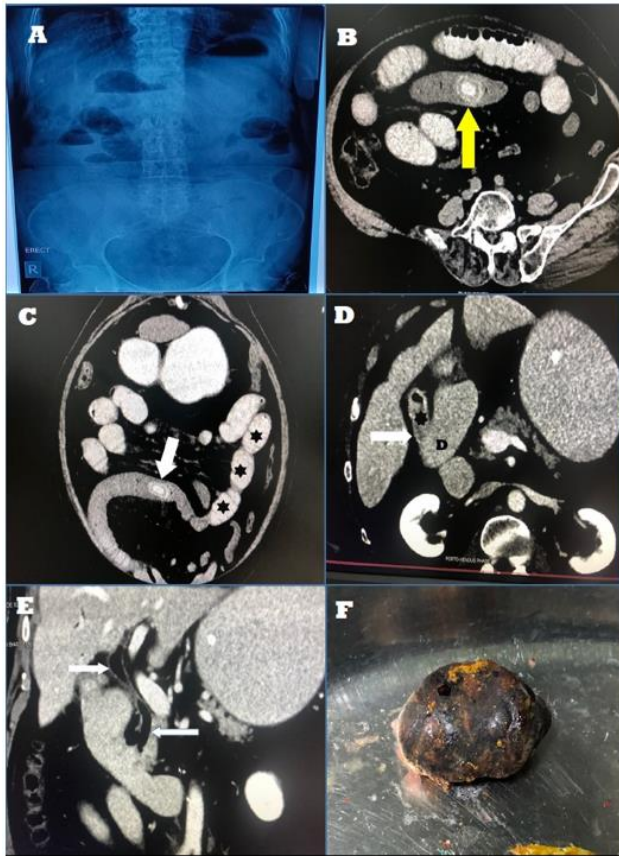


Figure 1: (A) Plain X-ray abdo shows multiple air-fluid levels in classical step-ladder pattern; (B) CECT abdo pics-axial view showing the large lamellated stone (yellow arrow) in the terminal ileum; (C) coronal view showing the obstructing gall stone in ileum (white arrow) with the dilated proximal contrast filled small bowel loops (black asterisks); (D) axial view showing the cholecysto-duodenal fistula (white arrow) betn the GB (black asterisk) and duodenum (black 'D'); (E) coronal view showing pneumobilia (white arrows); and (F) the retrieved culprit.

At laparoscopy, a small bowel 'walk' revealed the large gall stone at the ileo-cecal junction. It was then 'milked' proximally into the terminal ileum and delivered out through an enterotomy (Figure 2A-C and 1F), which was then suture closed in 2 layers (inner continuous and outer interrupted) using 3-0 Vicryl® (Figure 2D and E). The camera was now shifted from the supra-pubic to the umbilical 10 mm trocar. The cholecysto-duodenal fistula was located and excised (Figure 2F and 3A). The duodenal opening was suture closed using simple interrupted sutures using 2-0 Vicryl® (Figure 3B and C) and the sutureline was further buttressed with an omental

onlay patch. The cholecystectomy was then performed (Figure 3D). A peritoneal toilet was given and 2 Jackson-Pratt drains were inserted through the right lateral 5 mm trocars to be placed in the Morrison's pouch and the pelvis (Figure 3E). Thus, the operation was carried out in both the lower and the upper abdomen using 6 trocars (three 10 mm and three 5 mm), in total (Figure 3F). She had an uneventful postoperative recovery. She passed flatus on postoperative day (POD)-3 and was started on liquid feeds per orally on POD-4. Upon tolerating the same, she was started on oral semi-solid feeds on POD-5 and her abdominal drains as well as the per-urethral catheter were removed. She moved bowels on POD-6 and was discharged from the hospital on POD-7. On her POD-12 out patient department visit, all her wounds had healed well and she was asymptomatic.

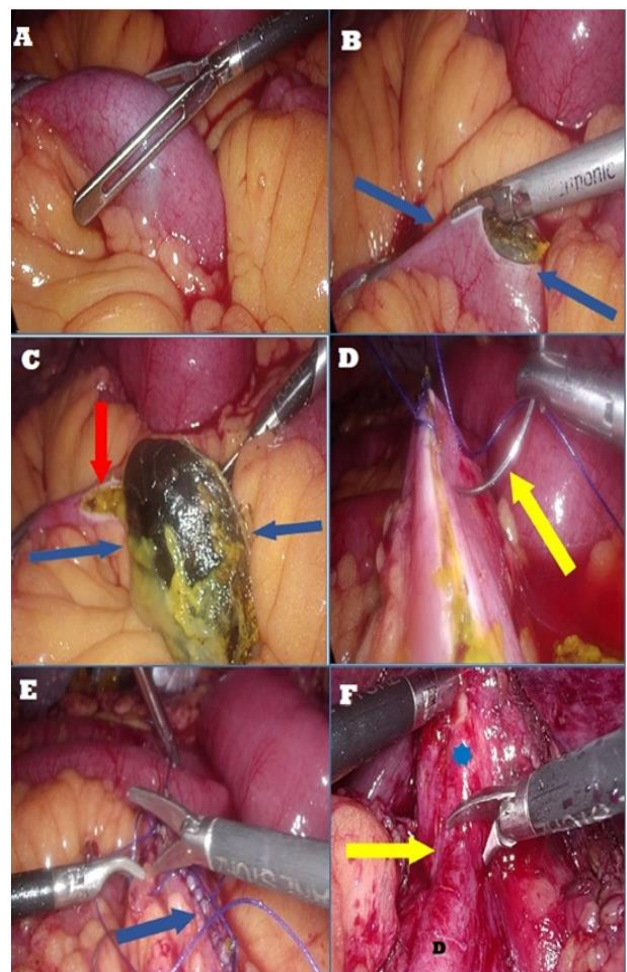


Figure 2: (A) Migrated obstructing gall stone being 'felt' and located during small bowel 'walk'; (B) enterotomy being performed (blue arrows); (C) the large obstructing gall stone (blue arrows) being delivered out of the enterotomy (red arrow); (D) enterotomy being suture-closed (yellow arrow); (E) completed enterotomy suture line (blue arrow); and (F) cholecysto-duodenal fistula betn GB (blue asterisk) and duodenum (black 'D') being divided (yellow arrow).

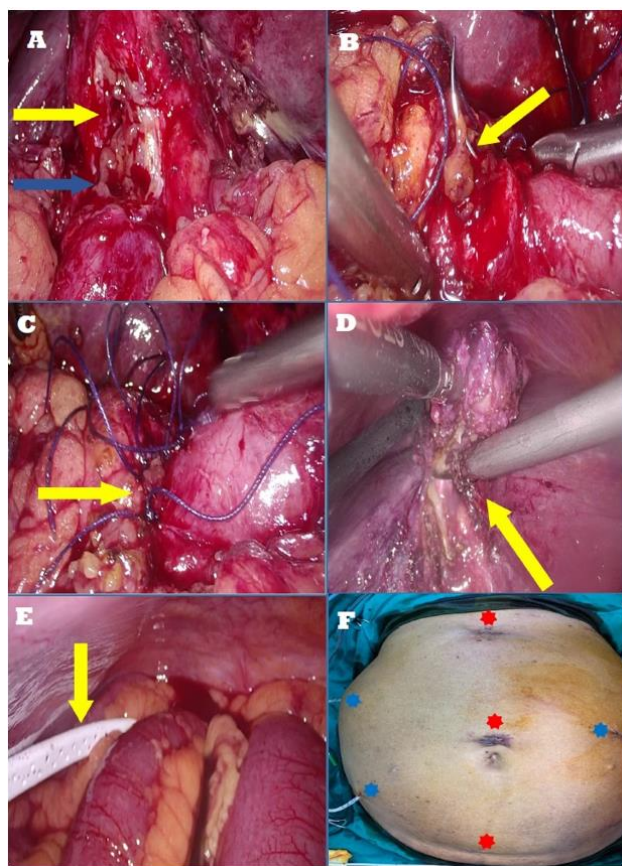


Figure 3: (A) GB (yellow arrow) and duodenal (blue arrow) openings after division of the cholecysto-duodenal fistula; (B) duodenal opening being suture-closed (yellow arrow); (C) completed duodenal suture line (yellow arrow); (D) cholecystectomy in progress (yellow arrow), drain in situ (yellow arrow); (E) endresult- three 10 mm trocars (red asterisks) and three 5 mm trocars (blue asterisks) used; and (F) 2 Jackson-Pratt drains inserted through the 2 right 5 mm trocar sites into the Morrison's pouch and the pelvis.

DISCUSSION

Gallstone ileus develops in 0.3-0.5% of patients with cholelithiasis.⁴ It constitutes the etiologic factor in less than 5% of cases of intestinal obstruction, but up to one quarter of non-strangulated small bowel obstructions in elderly patients.⁵ In accordance to the predominance of female patients in gallstone disease, the majority of gallstone ileus patients (72-90%) belong to the female gender.⁶

In 1654, Thomas Bartholin first described a cholecystointestinal fistula with a gallstone within the gastrointestinal tract in a study.⁷ In 1890, Courvoisier published the first series of 131 cases of gallstone ileus, with a mortality rate of 44%.⁸ In 1896, Bouveret described a syndrome (Bouveret's syndrome) of gastric outlet obstruction caused by an impacted gallstone in the duodenal bulb after its migration through a cholecysto- or

choledocho-duodenal or cholecysto-gastric fistula.⁹ In 1941, Rigler et al described the classical radiological diagnostic triad for gall stone ileus comprising of pneumobilia (Gotta-Mentschler sign), small bowel obstruction and ectopic gallstones.¹⁰ Before the era of computed tomography, Rigler's triad was only seen in 14-53% of patients in plain abdominal films. This increased to the 93% in the post computed tomography era.¹⁰

Gall stone ileus is always preceded by long standing cholelithiasis, which can cause acute inflammation of the gall bladder. Due to this inflammation, adhesions develop with surrounding structures. The pressure effect of the gall stone leads to erosion of the gall bladder wall and eventually leads to the formation of a fistula.² It is due to this communication, which is formed most commonly between the gall bladder and duodenum (85%), that passage of gall stones into the gastrointestinal tract is enabled.¹¹ Alternatively, the gallstone may pass through the common bile duct into the duodenum through the ampulla or during the manipulation of the gall bladder during cholecystectomy.^{11,12}

The impaction of the stone can occur anywhere in the gastrointestinal tract, starting with the stomach. However, the most common site for stone impaction is at the terminal ileum (60.5%).^{2,13} The obstruction caused by impaction of the gall stone at the ileo-cecal junction is known as Barnard's syndrome.¹⁴ Stones may also lodge in the jejunum (16.1%), stomach (14.2%) and less commonly the duodenum (3.5%).^{2,13} The stone must measure at least 2 cm to be able to cause obstruction.¹¹

The clinical picture is consistent with that of bowel obstruction. Dehydration, nausea, vomiting, abdominal distension, pain and high-pitched bowel sounds, are most commonly present.^{9,15} Patients may delay in presenting partly due to the 'tumbling phenomenon'. It describes the intermittent nature of symptoms secondary to temporary gallstone impaction followed by symptom relief when the stone dislodges, travels distally and impacts again.^{9,16}

Evidence of this phenomenon as change in stone position on X-ray, which when added to Rigler's triad, is known as the Rigler's tetrad.¹¹ Patients may also present with tachycardia, hypotension and fever-signs suggestive of sepsis from either cholecystitis or peritonitis due to impaction of the gallstone causing pressure to the bowel wall resulting in possible necrosis and perforation.² Biochemical markers show elevated WBC count and deranged electrolytes. Apart from X-ray, the features are also apparent on computed tomography scan abdomen, which, with sensitivity and specificity of 93% and 100%, respectively, has become the gold standard.¹⁷

The aim of therapy is to relieve the obstruction, which can be achieved with 3 different techniques-1) Enterolithotomy alone, 2) One stage surgery comprising enterolithotomy, cholecystectomy and

fistula repair and 3) Two staged procedure with enterolithotomy in stage 1 followed by cholecystectomy with fistula repair in stage 2, performed later, after about 4 weeks to 6 months. While there is consensus regarding the need for enterolithotomy to relieve the obstruction, there is controversy in the literature as to whether a cholecystectomy and fistula repair should be performed concurrently. Two staged surgery is preferred in patients with poor preoperative status, multiple comorbidities, dense adhesions and inflammation found intraoperatively which can increase the risk of iatrogenic injury.¹³ Proponents of enterolithotomy alone, argue that fistula closure is time consuming, technically demanding,

increases the morbidity, mortality rates and the duration of hospital stay.² Spontaneous fistula closure is also observed in a few patients.¹⁸ Proponents of one stage procedure claim that this procedure obliterates the need for reintervention and reduces incidence of complications related to fistula persistence, including recurrent gall stone ileus, cholecystitis, cholangitis.² Also, the risk of gall bladder malignancy associated with retained fistula reduces from 15% to 16%.^{11,19} Open surgery carries significant morbidity (20-57.5%) as well as the mortality (7-18%).²⁰ Laparoscopic surgery provides a faster recovery and shorter hospital stay as well as therefore is now a popular choice.

Table 1: A summary of case reports on laparoscopic management of GSI in the last 10 years.

Authors	Journal	Year of publication	Surgery done
Inukai et al ¹⁸	Journal of minimal Access surgery	2019	Stage 1-Lap assisted enterolithotomy Stage 2-Fistula closure + Chole
Gupta et al ²¹	Cureus	2020	Lap assisted enterolithotomy
Gari et al ²²	International journal of surgery	2018	Totally lap enterolithotomy
Orellana et al ²³	International journal of surgery	2021	Totally lap enterolithotomy
Bircan et al ²⁴	Clinical medicine insights: case reports	2014	Lap assisted enterolithotomy
Coisy et al ²⁵	Journal of gastrointestinal surgery	2015	Lap assisted enterolithotomy
Zygomalas et al ²⁶	Journal of laparoendoscopic and advanced surgical techniques	2012	Lap enterolithotomy

A literature search reveals few reports on laparoscopic management of gall stone ileus as shown in the Table 1. However, most of these cases are laparoscopy assisted, with small bowel delivered out through an extended trocar site and enterolithotomy done outside. Also, in all these said reports, only enterolithotomy was performed. To the best of our knowledge, this paper is the first and the only published report (till date) of a totally laparoscopic single stage therapy of the gall stone ileus i.e.; enterolithotomy with fistula closure and cholecystectomy in the same session, in the world literature.

CONCLUSION

As seen in this report, careful patient selection is the key to success in the therapy of GSI, like most other conditions. Totally laparoscopic single stage definitive therapy of GSI is safe, feasible and indeed beneficial to the patient, in an advanced laparoscopy setup at the hands of an experienced operator, when coupled with optimum patient selection.

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