CASE SERIES PEER REVIEWED | OPEN ACCESS

Acute cholecystitis as a complication of diverting loop ileostomy: A case series and narrative review

Rama HG Mikhail, Marcel Chee, Siobhan C McKay, Konstantinos Syrrakos

ABSTRACT

Introduction: Diverting ileostomy is increasingly utilized in modern colorectal surgery for the management of colorectal cancer and inflammatory bowel disease (IBD) among other bowel pathologies. Cholelithiasis has known associations with ileostomy and IBD, potentially caused by changes to enterohepatic circulation and cholesterol absorption in the gallbladder. However, cases of cholecystitis following ileostomy have not been reported across the literature.

Case Series: We present three cases of cholecystitis following diverting loop ileostomy in a 56-year-old female, 74-year-old female, and 73-year-old female. A digital literature search was conducted on databases PubMed and EMBASE in February 2023 covering literature from 1974 to 2023 to identify any further cases; however, none were identified. To our knowledge, these are the first reported cases of cholecystitis following ileostomy.

Conclusion: We believe these three cases highlight that developing symptomatic gallstones following diverting ileostomy occurs, and patients should be carefully counseled of the risks preoperatively, and be educated of gallstone-related symptoms to enable early engagement of upper gastrointestinal surgeons for treatment. Additionally, we recommend preoperative gallbladder imaging and close monitoring prior to and after ileostomy formation for development of gallstone complications. Ultimately, this case series and narrative review lay the groundwork for further research investigating any causality between diverting loop ileostomy formation and development of complications of gallstone disease.

Keywords: Cholecystitis, Cholelithiasis, Consent, Diverting ileostomy

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INTRODUCTION

Diverting ileostomy is utilized in modern colorectal surgery for the management of colorectal cancer, diverticulitis, and inflammatory bowel disease (IBD) among other bowel pathologies. Cholelithiasis has been known to be associated with ileostomy and IBD through several hypothesized mechanisms related to impaired enterohepatic circulation. However, cases of cholecystitis following ileostomy have not been reported across the literature. We present the first reported cases of cholecystitis as a short-term complication of diverting ileostomy, as well as review of the evidence to date regarding hypothesized pathophysiological mechanisms and management.

CASE SERIES

We present three cases of cholecystitis following diverting loop ileostomy, not previously reported. A digital literature search on databases PubMed and EMBASE was conducted in February 2023 for English publications to assess for literature presence of any further cases. Medical subject headings were used where available with key words including "cholecystitis," "gallstones," "stoma," and "ileostomy" and the inclusion criteria of: (1) diverting loop ileostomy, (2) cholecystitis following ileostomy, (3) studies in humans, and (4) English language. No primary studies were identified from the digital search on PubMed and EMBASE that met the inclusion criteria. Additionally, a search was conducted to identify published ileostomy consenting documents to examine for inclusion of gallstone disease as a complication, none were identified. To our knowledge following our digital search, these are the only reported cases of cholecystitis following ileostomy.

Written consent has been obtained from the patients involved in this case series.

Case 1

A 56-year-old female who presented for surgical management of rectal cancer. She had no significant past medical history, and her surgical history was limited to a urethral sling. The patient's malignancy was diagnosed on colonoscopy and staged on computed tomography (CT) two weeks prior to her ileostomy, which showed an incidental finding of a gallstone, from which she was completely asymptomatic (Figure 1). She underwent laparoscopic ultra-low anterior resection and diverting loop ileostomy. She was discharged on postoperative day (POD) 5, during which she received antibiotic prophylaxis as per protocol as well as a drain which was removed on day of discharge. Histopathology showed a moderate differentiated adenocarcinoma with evidence of lymphovascular invasion. The patient presented to the emergency department 2 weeks from date of discharge with findings of gallstone cholecystitis (Figure 2). Cholecystectomy was offered, but the patient elected for conservative management. Two months following her presentation to the emergency department, the patient eventually underwent a robotic reversal of the stoma and cholecystectomy. Histopathology of the surgical specimens showed chronic cholecystitis and cholelithiasis. She was kept on a low-fat diet postoperatively and was ultimately discharged on POD 3. The patient remains well and has had no complications in the three months following her robotic stoma reversal and cholecystectomy.

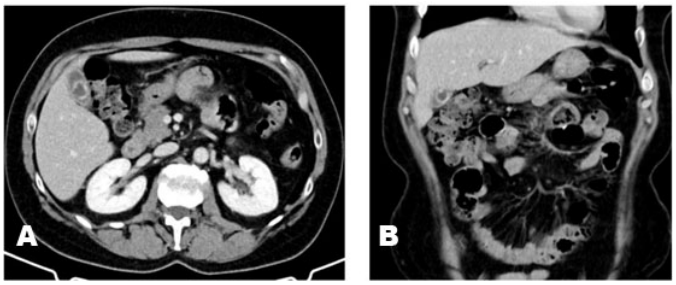

Figure 1: (A) Coronal and (B) axial slices of the preoperative CT of the abdomen performed in a 56F (Case 1) demonstrating a large gallstone within the gallbladder.

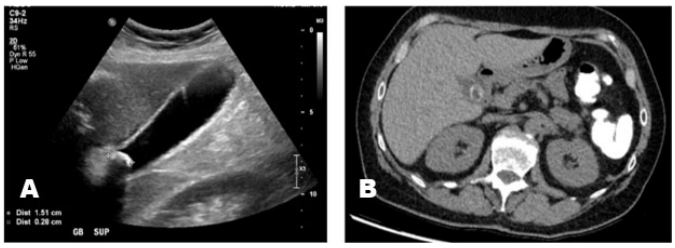

Figure 2: (A) Post-ileostomy gallbladder ultrasound and (B) axial CT abdomen slice in a 56F (Case 1) demonstrating evidence of acute cholecystitis with thickened gallbladder wall and trace pericholecystic fluid.

Case 2

A 74-year-old female who presented with a 15 cm fecal abscess involving the rectosigmoid colon. Her past medical history included type 2 diabetes, diverticulitis, hypertension, polycythemia rubra vera, gastro-oesophageal reflux disease, and a liver abscess (secondary to diverticulitis). Her surgical history included perforated diverticulitis requiring laparoscopic washout. As part of routine pre-ileostomy workup, the patient had a preparatory CT abdomen and pelvis (CTAP) which did not reveal any gallstones or gallbladder pathology (Figure 3). The patient underwent laparoscopic low anterior resection and diverting ileostomy and, after an uncomplicated recovery, was discharged on POD 5 with a plan for reversal in 12 months' time. Approximately one year following the ileostomy, a CT with rectal contrast was performed in anticipation of stoma reversal to assess the anastomosis and incidentally revealed evidence of acute cholecystitis (Figure 4). The patient was admitted and received conservative management for cholecystitis, from which she recovered and ultimately discharged. She underwent robotic reversal of the diverting ileostomy approximately one month later with a plan for elective cholecystectomy in six months' time.

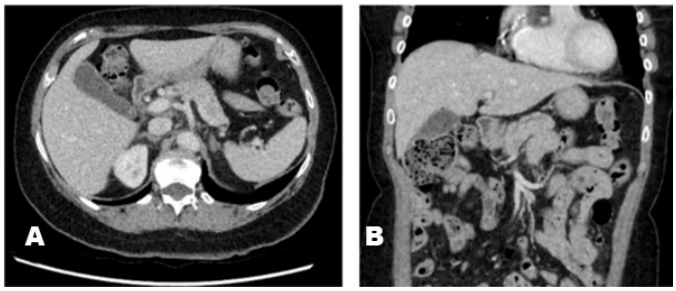

Figure 3: (A) Preoperative axial and (B) coronal CTAP slices in a 74F (Case 2) demonstrating an unremarkable gallbladder with no evidence of acute cholecystitis or cholelithiasis.

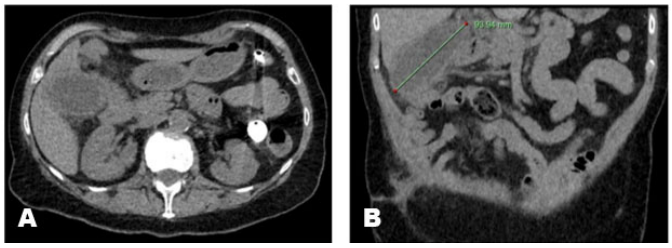

Figure 4: (A) Postoperative axial and (B) coronal CTAP slices in a 74F (Case 2) demonstrating comparative gallbladder distension (93.94 mm), gallbladder wall thickening and pericholecystic fluid.

Case 3

A 73-year-old female who presented for management of sigmoid colon strictures secondary to diverticulosis. Her past medical history includes hypertension, previous endometriosis, rheumatoid arthritis managed with biologics, and diverticulosis. Her surgical history included lumbar discectomy and hysterectomy. As part of routine pre-ileostomy workup, the patient had a preparatory CTAP which did not reveal any gallstones or gallbladder pathology (Figure 5). The patient underwent an ultra-low resection in the context of intraoperative findings of significant adhesions due to the hysterectomy as well as poor tissue health. Additionally, she was found to have a colovesical fistula intraoperatively. The patient was discharged on POD 4 after an uncomplicated postoperative course. Laparoscopic reversal of ileostomy was performed six months later and, as part of the investigative workup prior to the reversal, a preoperative CT with rectal contrast was performed for assessment of the anastomosis. The patient presented to the emergency department with acute cholecystitis five months after reversal (Figure 6). Of note, the CT performed prior to the patient's reversal did not show any evidence of gallbladder pathology. The patient managed conservatively initially and ultimately underwent elective laparoscopic cholecystectomy six months later, with an uneventful postoperative recovery. Histopathology showed cholelithiasis and chronic cholecystitis.

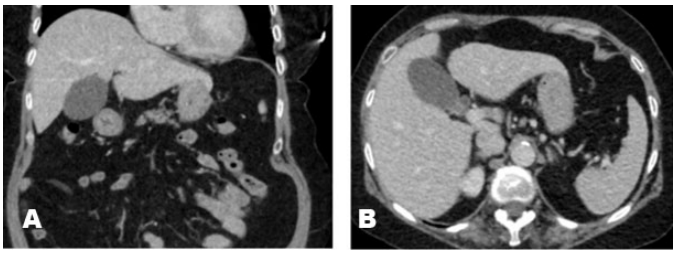

Figure 5: (A) Preoperative coronal and (B) axial CTAP slices in a 73F (Case 4) demonstrating an unremarkable gallbladder with no evidence of acute cholecystitis or cholelithiasis.

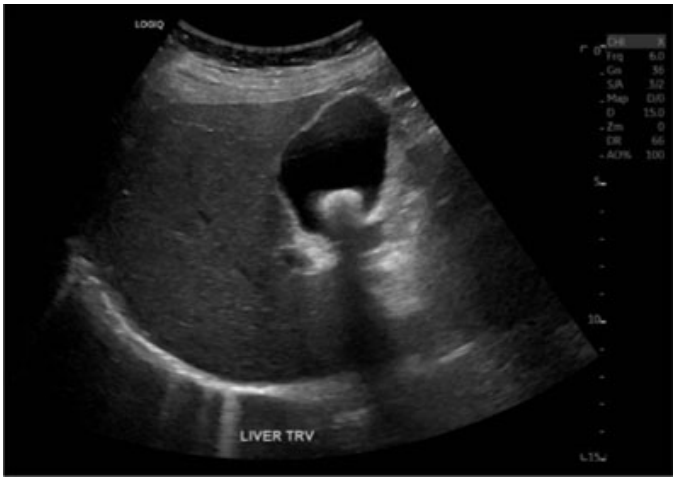

Figure 6: Postoperative gallbladder ultrasound in a 73F (Case 4) performed on the patient's presentation to the emergency department five months following reversal demonstrating evidence of large obstructing stone near the gallbladder neck.

DISCUSSION

Ileostomy is often performed following low anterior resection as a means of surgical management for advanced diverticulitis, IBD, and bowel cancer [1]. Diverting ileostomy has become an increasingly utilized technique in contemporary colorectal surgery to reduce the risk of the dire anastomotic leak in low and ultra-low bowel resections [1]. Over the years, the necessity and safety of fecal diversion through ileostomy has been disputed but a review published in 2015 ultimately found that in patients who had colorectal anastomosis <10 cm from the anal verge or coloanal anastomosis had resections that were technically difficult, were male or malnutritioned, that a diverting ileostomy should be strongly considered to reduce the risk of anastomotic leak [1]. In this paper, we present the cases of three patients who developed acute cholecystitis within a year of undergoing low and ultra-low anterior resections with diverting loop ileostomy. Notably, none of these patients reported any symptoms concerning gallstone disease (including but not limited to right upper quadrant or epigastric pain, reflux, nausea, or vomiting) prior to their ileostomies.

There are no cases describing cholecystitis as a complication of any form of ileostomy across the literature at the time of digital search in February 2023. However, the association between IBD, which is a common indication for ileostomy including in our case series, and the development of cholelithiasis is well covered in the literature. A retrospective study of over 150 patients with permanent ileostomy for IBD revealed that approximately 23% of patients developed cholelithiasis in the surveillance period following their ileostomy and recommended gallbladder sonography prior to ileostomy [2]. More recently, a large, longitudinal case-control study demonstrated that patients with IBD were significantly more likely

to develop subsequent cholelithiasis in comparison to non-IBD patients [3]. The study also demonstrated that patients with Crohn's disease were independently 2.78-fold more likely to develop common bile duct stones and intrahepatic stones when compared to the control non-Crohn's population, especially where the ileum was involved [3]. After statistical analysis of several variables (including age, sex, and comorbidities), the study ultimately demonstrated that IBD remains independently associated with gallstone disease [4].

Impact of bowel resection on cholelithiasis in inflammatory bowel disease

There is little across the literature examining the role of bowel resection in the development of cholelithiasis in IBD. In 1975, Hill et al. illustrated the positive association between cholelithiasis and the number of bowel resections and additionally demonstrated that ileal bowel resections of >10 cm were significantly more likely to develop cholelithiasis, which was later confirmed by a large case-control study by Parente et al. [5, 6]. The same cannot be said for ulcerative colitis (UC), which is likely due to reduced involvement of the ileum in its pathology. Another study demonstrated that bile in the gallbladders of Crohn's patients with ileal resection was more likely to be supersaturated with cholesterol in comparison to Crohn's patient without ileal resection; however, this study did not explicitly discuss the implication to cholelithiasis [7]. Ultimately, the literature is ambiguous, and though ileal bowel resection is associated with increased risk of cholelithiasis in Crohn's disease, it is difficult to infer if ileostomy independently carries a greater risk of cholelithiasis in this subgroup of patients. No literature exists specifically investigating any association between low or ultra-low bowel resection on cholelithiasis or cholecystitis.

Pathophysiology

Enterohepatic circulation (EHC) refers to the passage of bile acids (BA) from the liver, through the intestinal tract until they arrive back at the liver [8]. Approximately 1 liter of bile is produced by the liver daily, of which close to 95% undergoes rigorous reabsorption through the venous system; the remaining five perfect is lost in feces [8]. Significant reabsorption of BAs occurs through the ileal and colonic venous systems, with 98% of BA reabsorption occurring at the terminal ileum, and BAs ultimately arrive at the hepatocytes through the portal vein for reuptake [3].

While the general association between ileostomy and development of cholelithiasis has been demonstrated, the underlying pathology is unclear, and no literature exists to examine this. The pathogenesis of cholelithiasis in IBD, however, has been hypothesized across the literature. We have classified these hypotheses in the table below:

Biochemical EHC disruption

(1) The reabsorption of bile through the EHC is compromised in cases of IBD due to inflammation thereby impairing cholesterol absorption which increases the risk of cholelithiasis [9].
(2) The remaining unabsorbed BAs in the lumen keep unconjugated bilirubin in solution, resulting in the gallbladder formation of pigment stones due to the significantly greater presence of unconjugated bilirubin in bile [10].
(3) Inflammatory bowel disease is known to affect the gut microbiome leading to dysbiosis. Human studies investigating the effect of dysbiosis on metabolism of BAs demonstrated impaired metabolism of BAs in the lumen which is hypothesized to have an extrapolated increased risk of cholethiasis [11].

Mechanical EHC disruption

(1) Primary sclerosing cholangitis has a well-known association with ulcerative colitis. The mechanisms underlying this is hypothesized to be a combination of bile stasis because of reduced gallbladder motility and chronic inflammation [12].

It is reasonable to apply the above hypotheses when considering cholelithiasis following ileostomy: patients undergoing ileostomy in the context of IBD may exhibit both biochemical and mechanical impairments to EHC due to (a) loss of the terminal ileum due to resection as part of the ileostomy procedure and (b) the intrinsic consequences implicated by IBD theorized above.

It is exceptionally difficult to present the pathophysiology underlying the development of cholecystitis in ileostomy patients as there are no such cases across the literature. In this case series, we have demonstrated the acute development of cholecystitis in two patient groups: those with prior gallstone disease (case 1) and those with no previous history of gallstone disease (cases 2 and 3). Therefore, we postulate that ileostomy may independently elevate the risk of cholecystitis by impairing the reabsorption of bile through EHC through mechanisms hypothesized above, lowering bile concentration in the gallbladder. Additionally, the continuous flushing action of bile coupled with the bacteriostatic and bactericidal properties of bile salts, helps maintain sterility in the biliary tract and prevents ascending bacterial infection from the duodenum; a reduction in the intrahepatic bile salt concentration, therefore, may undermine this physiological defence, elevating the risk of cholecystitis [13, 14]. Further, severe dehydration, known to occur as a complication following ileostomy due to increased losses through the stoma, may also contribute to the development of cholecystitis by precipitating gallbladder stasis [15, 16].

Learning points

Cholecystitis should be regarded as a possible complication of ileostomy, particularly diverting ileostomy as is in our presented cases, and due preoperative consideration should be taken. At present, no accessible national guidelines on ileostomy nor any published ileostomy consenting protocols details cholecystitis as a potential complication of ileostomy. Based on our review of the literature in conjunction with our reported cases, we recommend counseling patients on the possible complication of cholecystitis as part of the consenting process for ileostomy, as well as performing early imaging of the gallbladder and biliary tree as part of the preoperative workup in IBD patients being considered for ileostomy. Ultimately, further research is needed to demonstrate the pathophysiology of cholelithiasis and cholecystitis in patients with ileostomy to allow for appropriate risk stratification and management protocols.

CONCLUSION

Cholelithiasis is well known to be associated in patients with IBD with a number of hypotheses presented across the literature, which we have classified into biochemical and mechanical EHC disruption. Cholecystitis, however, has not been reported in the literature as a potential complication of ileostomy and has certainly not been associated with low or ultra-low bowel resection and diverting ileostomy. We present the cases of three patients who developed cholecystitis shortly after diverting low ileostomy and explore the evidence to date regarding pathogenesis of this complication in this cohort of patients. As these are the first cases of cholecystitis following diverting ileostomy, further research is warranted investigating the pathogenesis, prevention, and management in order to guide preoperative workup for ileostomy.

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Author Contributions

Rama HG Mikhail – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Marcel Chee – Analysis of data, Interpretation of data, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Siobhan C McKay – Analysis of data, Interpretation of data, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy

or integrity of any part of the work are appropriately investigated and resolved

Konstantinos Syrrakos – Conception of the work, Acquisition of data, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

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Authors declare no conflict of interest.

Data Availability
All relevant data are within the paper and its Supporting Information files.