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CASE REPORT

Gallstone Ileus with Resolution of Bile Leak Post-Subtotal Cholecystectomy and Spontaneous Closure of Cholecystoduodenal Fistula: Case Report and Literature Review

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ABSTRACT

Gallstone ileus (GSI) is a mechanical intraluminal bowel obstruction caused by biliary calculi through the biliary-enteric fistula. This is a rare sequela of cholelithiasis occurring in 0.3 – 1.5% of patients with worrying mortality of 11.7 – 20%. This is a case of GSI in a 67-year-old woman who presented with small bowel obstruction secondary to impaction of biliary calculi at terminal ileum with underlying cholecystoduodenal fistula (CDF). Enterolithotomy with stone extraction (ES) was performed, followed by subtotal reconstituting cholecystectomy due to iatrogenic gallbladder perforation. The diagnosis of GSI is ascertained by the presence of the Rigler's triad on abdominal X-ray, while CDF was demonstrated by post-surgery CT images. Bile leak post-operation was managed conservatively based on the SNAP (Sepsis, Nutrition, Anatomy, Plan) approach, and spontaneous closure of CDF was observed. In a nutshell, GSI should always be kept in mind as a differential diagnosis of mechanical bowel obstruction, especially among elderly female patients. Radiological findings of Rigler's triad aid clinical diagnosis of GSI. Despite its rare incidence, early diagnosis is crucial as it is readily treatable with surgery. ES alone is the gold standard in the management of GSI.

INTRODUCTION

The burden of gallstone disease varies among nations ranging from 3 – 5% in the Asian population and doubles in the West (Kratzer et al., 1999). Generally, patients with cholelithiasis present with a spectrum of complications,

but only 0.3 – 1.5% develop gallstone ileus (Kurtz et al., 1985). Gallstone ileus (GSI) is an uncommon yet crucial aetiology of mechanical intraluminal bowel obstruction owing to its substantial morbidity and mortality (Beuran et al., 2010; Halabi et al., 2014; Reisner et al., 1994). This entity is commonly seen among older women with no tell-tale clinical presentation, thus often a diagnosis challenge. Computed Tomography (CT) helps diagnose GSI based on the classical Rigler's triad (Lassandro et al., 2004). The key treatment of GSI is primarily surgical – Enterolithotomy with Stone extraction (ES) (Halabi et al., 2014; Reisner et al., 1994). Here, we describe a case of GSI complicated with bile leak after ES and subtotal cholecystectomy; spontaneous resolution of both bile leak and CDH ensued.

CASE PRESENTATION

A 67-year-old Malay woman presented to the emergency department with generalized colicky abdominal pain, obstipation, vomiting, and reduced appetite for two days. There was no prodrome or constitutional symptoms prior. Other than her three uncomplicated caesarean sections, her functional status was good with well-controlled diabetes mellitus. Clinically, she was hemodynamically stable with a distended abdomen and localized tenderness over the upper abdomen. There was a well-healed lower midline laparotomy scar with no hernia. Bowel sounds were active. Apart from the mild azotemia, other parameters were within range. Her abdominal X-ray showed small bowel dilation. The provisional diagnosis was acute intestinal obstruction secondary to adhesion. She was subjected to a brief period of resuscitation while observing for spontaneous resolution.

She underwent a midline laparotomy as her symptoms did not improve. Intraoperatively, there was a huge intraluminal mass at the terminal ileum (20 cm from the ileocecal valve), causing proximal small bowel dilation. An enterotomy was done proximal to the mass, and a pigmented stone measuring 5 × 3.5 cm was extracted (Figures 1 and 2). The enterotomy was closed primarily. She was hemodynamically stable during the surgery, and cholecystectomy was attempted. An iatrogenic perforation occurred at the fundus of the contracted gallbladder during dissection due to surrounding adhesions, and a subtotal reconstituting cholecystectomy was performed with subhepatic drain placement. She recovered well in the immediate post-operative period.

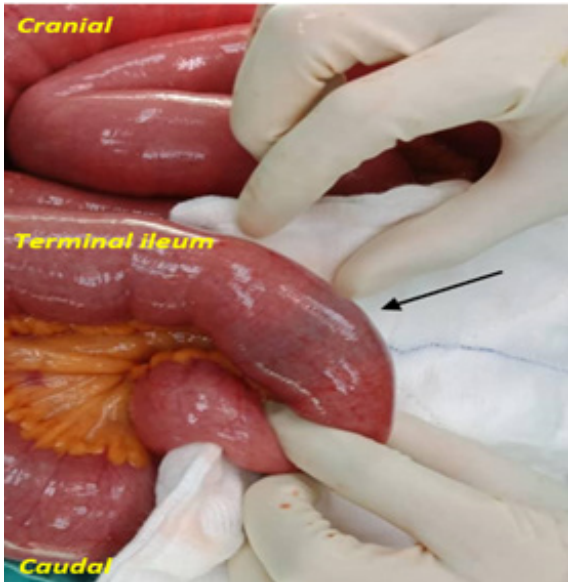

Figure 1 Gallstone (*black arrow*) impacted at terminal ileum (20 cm from ileocecal valve) causing proximal small bowel dilation. Enterotomy and stone extraction performed.

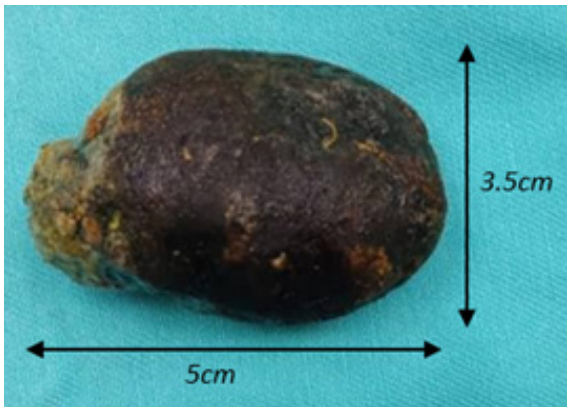

Figure 2 Single pigmented gallstone extracted measuring 3.5 cm × 5 cm.

On day five post-operation, the subhepatic drainage showed bilious content. CT abdomen performed demonstrated contrast leak lateral to 1st part of the duodenum into the gallbladder remnant complicated with a heterogenous intraabdominal collection. The tract outlined is the CDF with surrounding collection suggesting a leak from the gallbladder remnant (Figures 3 and 4).

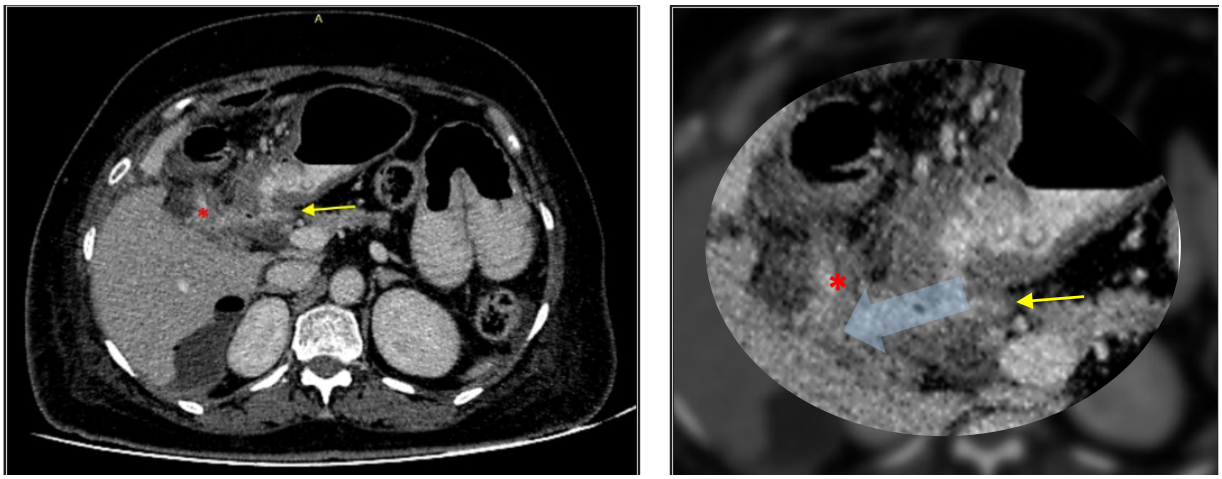

Figure 3 CECT Abdomen outlining the cholecystoduodenal fistula (*blue faded arrow*) demonstrated by contrast leak lateral to 1st part of duodenum (*yellow arrow*) to gallbladder remnant (*red asterisk*) with leak into peritoneal cavity.

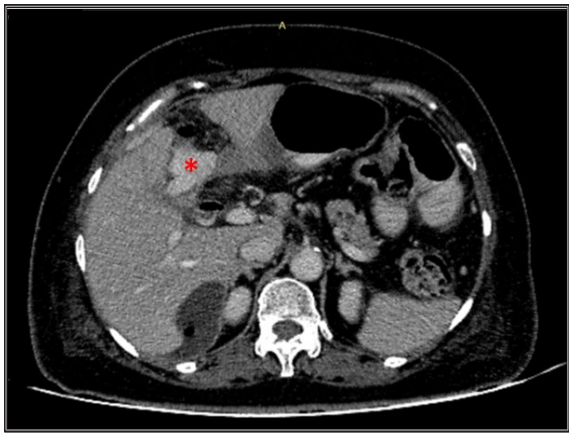

Figure 4 CECT Abdomen demonstrating pooling of contrast in the gallbladder remnant and leaking into peritoneal cavity (*red asterisk).*

A conservative approach was decided upon to manage gallbladder remnant leak and percutaneous drainage for the intraabdominal collection as she was clinically well with normal septic parameters. The patient was kept nil by mouth, and parenteral nutrition was commenced early to reduce bilious output. She has also been prescribed somatostatin analogue and proton pump inhibitors.

Both drainages attenuated gradually over the next ten days, and oral feeding was sequentially introduced. An oral contrast study performed two weeks post-operation showed no contrast leak from the duodenum inferencing CDF closure. The resolution of subhepatic collections was confirmed with an ultrasound done at week three post-surgery, and tubes were removed. During our follow-up on the patient in the outpatient clinic one week later, she was well, and histopathology of the gallbladder showed chronic inflammation without any evidence of malignancy. We monitored the patient until six months post-operation and found her to be asymptomatic.

DISCUSSION

In 1654, Dr Erasmus Bartholin reported the first GSI case examined during an autopsy, which laid a foundation for further exploring this intriguing entity (Deckoff, 1955). Gallstone ileus (GSI) constitutes 1-5% of mechanical bowel obstruction (Halabi et al., 2014). Generally, gallstones travel into the gastrointestinal

tract from the gallbladder or biliary tract via a biliary-enteric fistula. The most frequently encountered fistula is CDF accounting 60-80%. Most CDFs (90%) are gallstone-related (Lee et al., 2003; Nuno-Guzman et al., 2016). Pathophysiology of classical CDF is preceded by acute cholecystitis; existing gallstone induces pressure necrosis on the weakened gallbladder wall, thus creating an abnormal track into the adjacent bowel (Doycheva et al., 2009). Impaction of gallstone causing intestinal obstruction happens primarily in the narrowest bowel segment- terminal ileum (85-89.5%) (Ayantunde et al., 2007; Nuno-Guzman et al., 2016). Gallstones larger than 2cm usually result in intestinal obstruction, whereas smaller calculi tend to pass out spontaneously (Clavien et al., 1990; Kasahara et al., 1980).

There is no pathognomonic feature of GSI to date. The typical presentation of GSI is often described as wax and wane abdominal pain when the gallstone is propelled along the bowels before accentuating obstructive symptoms. CT shows the highest accuracy for the diagnosis of GSI (78%) compared to abdominal X-ray (15%) in exhibiting the classical radiological Rigler's triad- pneumobilia, gallstone in bowel and bowel dilatation (Rigler et al., 1941). A pre-operative CT should be carried out whenever possible as it provides information on the number, size, and locations of stones, assess for possible complications (ischaemia, necrosis, or perforation) as well as delineating the biliary-enteric fistula. Interestingly, this triad is present on a retrospective look on the abdominal X-ray of this patient (Figure 5).

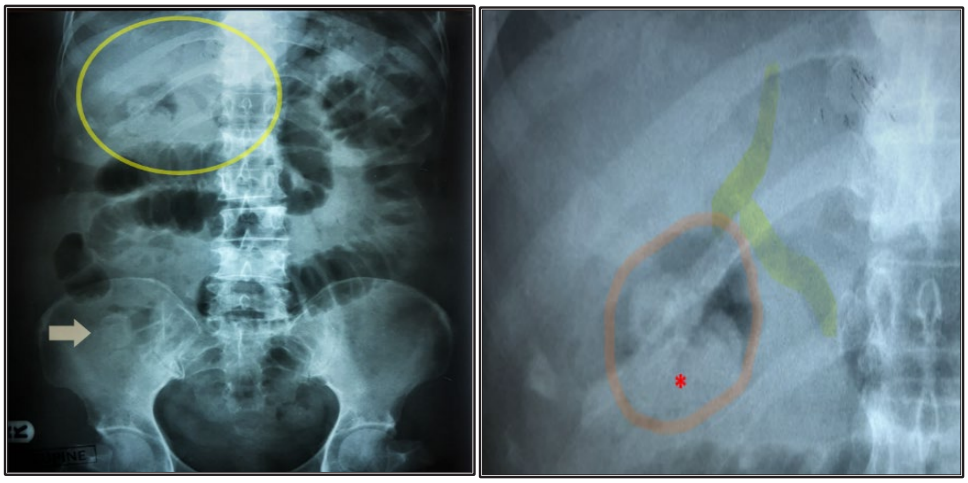

Figure 5 Rigler's triad in abdominal X-ray- aerobilia (*yellow highlight*), aberrant gallstone (*arrow*) and small bowel obstruction. Gallbladder with radiopaque stones (*red highlight and asterisk*)

Intestinal obstruction, biliary enteric fistula, and cholelithiasis are the three principles of surgical management of GSI. An enterotomy can relieve intestinal obstruction followed by stone extraction (ES) with or without bowel resection (Halabi et al., 2014). Biliary-enteric fistula and cholelithiasis are generally addressed as a combined procedure of cholecystectomy, and primary fistula closure (CF) is performed either during the index operation (single staged) or delayed (elective setting). This decision is based mainly on the patient's clinical status, the complexity of the gallbladder's anatomy, and the clinician's expertise.

Previous literature highlighted ES alone as the gold standard of GSI treatment, especially in high-risk patients (American Society of Anesthesiology; ASA III-IV) with a proven reduction in mortality rates and shorter hospital length of stay (Halabi et al., 2014; Reisner et al., 1994). On the contrary, the single staged procedure (ES+CF) can be opted in

carefully selected patients such as the younger age group with ASA I-II to reduce recurrent GSI, other biliary disease complications and prevent malabsorption gallbladder carcinoma from a persistent biliary-enteric fistula. A single-centre cohort study in Singapore reported zero mortality and no significant differences in morbidity with proper selection of patients for both procedures mentioned (Tan et al., 2004).

Strasberg et al. (2016) introduced the concept of subtotal cholecystectomy, addressing difficult operative conditions where the free personalised portion of the gallbladder is excised, and the lowest portion of the gallbladder remnant is left in-situ. The two subtypes of subtotal cholecystectomy described include Fenestrating (open gallbladder remnant with cystic duct closure) and Reconstituting (closed gallbladder remnant with patent cystic duct). 'Reconstituting' cholecystectomy reduces the incidence of postoperative bile fistula, while the 'Fenestrating' type has less chance of developing complications from a gallbladder remnant. However, there is no head-to-head comparison between both techniques to demonstrate superiority.

Bile leak post total cholecystectomy occurs in approximately 0.8-1.1% of patients. In this case, the bile leak originated from the remnant gallbladder. Low output bile leaks usually resolve without intervention; on the other hand, high output leaks can be treated via Endoscopic Retrograde Cholangiopancreatography (ERCP) methods-sphincterotomy alone, stenting alone or combination therapy or managed surgically (Rustagi et al., 2014).

A careful and systematic treatment approach (SNAP-Sepsis, Nutrition, Anatomy, Plan) can be adopted to address post-operative bile leaks. Prompt *sepsis* control is achieved by initiating antibiotics and drainage of intraabdominal collection (Levy et al., 2018). Next, nutrition optimization via collaborative care under the Nutritional Support Team promotes healing over the bile leak site and fistula. Parenteral nutrition is introduced in the initial phase to attenuate bile leakage, while enteral feeding is commenced once drainage reduces to stimulate gut trophic effects (Kuvshinoff et al., 1993; Lloyd et al., 2006). Long-acting somatostatin analogue (Octreotide) is beneficial in reducing bile leak, biliary fistula output, duration of parenteral nutrition required, and hasten CDF healing time (Nubiola et al., 1989; Pran et al., 1995). Proton pump inhibitors are used to decrease the volume and acidity of gastric output, which helps to reduce bile leaks (Hollington et al., 2004). Thirdly, the assessment of biliary anatomy can be done endoscopically via fluoroscopy or with CT before subsequent planning for definitive treatment. Based on several retrospective studies, 50-61.5% of patients experienced spontaneous biliary-enteric closure following successful ES (Inukai, 2019; Raf & Spangen, 1971). For patients with persistent CDF, the risk of recurrent GSI and symptomatic biliary disease is 4-8% and 15%, respectively; definitive biliary surgery can be offered 3-6 months after the index surgery in carefully selected individuals (Farkas et al., 2018; Mir et al., 2015).

CONCLUSION

GSI should always be considered a differential diagnosis of mechanical bowel obstruction, especially among the geriatric age group females. Rigler's triad is a potential guide radiologically to suggest the diagnosis of GSI. Despite its rare incidence, early diagnosis is crucial as it is readily treatable with surgery. Enterotomy with stone extraction (ES) alone is the gold standard in the management of GSI.

CONFLICT OF INTEREST

The authors declare that they have no competing interests in publishing this case.

CONSENTS

Written consent was obtained from the patient to publish the case. A copy of the written consent is available for review by the Chief Editor.

REFERENCES

Ayantunde, A. A., & Agrawal, A. (2007). Gallstone ileus: diagnosis and management. *World Journal of Surgery*, *31*(6), 1292–1297. https://doi.org/10.1007/s00268-007-9011-9

Beuran, M., Ivanov, I., & Venter, M. D. (2010). Gallstone ileus--clinical and therapeutic aspects. *Journal of Medicine and Life*, *3*(4), 365–371. Retrieved from https://www.ncbi.nlm.nih.gov/pmc/articles/PMC3019077/

Clavien, P. A., Richon, J., Burgan, S., & Rohner, A. (1990). Gallstone ileus. *The British Journal of Surgery*, *77*(7), 737–742. https://doi.org/10.1002/bjs.1800770707

Deckoff, S. L. (1955). Gallstone ileus; a report of 12 cases. *Annals of Surgery*, *142*(1), 52–65. https://doi.org/10.1097/00000658-195507000-00007

Doycheva, I., Limaye, A., Suman, A., Forsmark, C. E., & Sultan, S. (2009). Bouveret's syndrome: case report and review of the literature. *Gastroenterology Research and Practice*, *2009*, 914951. https://doi.org/10.1155/2009/914951

Farkas, N., Kaur, V., Shanmuganandan, A., Black, J., Redon, C., Frampton, A. E., & West, N. (2018). A systematic review of gallstone sigmoid ileus management. *Annals of Medicine and Surgery (2012)*, *27*, 32–39. https://doi.org/10.1016/j.amsu.2018.01.004

Halabi, W. J., Kang, C. Y., Ketana, N., Lafaro, K. J., Nguyen, V. Q., Stamos, M. J., Imagawa, D. K., & Demirjian, A. N. (2014). Surgery for gallstone ileus: a nationwide comparison of trends and outcomes. *Annals of Surgery*, *259*(2), 329–335. https://doi.org/10.1097/SLA. 0b013e31827eefed

Hollington, P., Mawdsley, J., Lim, W., Gabe, S. M., Forbes, A., & Windsor, A. J. (2004). An 11-year experience of enterocutaneous fistula. *The British Journal of Surgery*, *91*(12), 1646–1651. https://doi.org/10.1002/bjs.4788

Inukai K. (2019). Gallstone ileus: a review. *BMJ Open Gastroenterology*, *6*(1), e000344. https://doi.org/10.1136/bmjgast-2019-000344

Kasahara, Y., Umemura, H., Shiraha, S., Kuyama, T., Sakata, K., & Kubota, H. (1980). Gallstone ileus. Review of 112 patients in the Japanese literature. *American Journal of Surgery*, *140*(3), 437–440. https://doi.org/10.1016/0002-9610(80)90185-3

Kratzer, W., Mason, R. A., & Kächele, V. (1999). Prevalence of gallstones in sonographic surveys worldwide. *Journal of Clinical Ultrasound: JCU*, *27*(1), 1–7. https://doi.org/10.1002/(sici)1097-0096(199901)27:1<1::aid-jcu1>3.0.co;2-h

Kurtz, R. J., Heimann, T. M., Beck, A. R., & Kurtz, A. B. (1985). Patterns of treatment of gallstone ileus over a 45-year period. *The American Journal of Gastroenterology*, *80*(2), 95–98. Retrieved from https://pubmed.ncbi.nlm.nih.gov/3970007/

Kuvshinoff, B. W., Brodish, R. J., McFadden, D. W., & Fischer, J. E. (1993). Serum transferrin as a prognostic indicator of spontaneous closure and mortality in gastrointestinal cutaneous fistulas. *Annals of Surgery*, *217*(6), 615–623. https://doi.org/10.1097/00000658-199306000-00003

Lassandro, F., Gagliardi, N., Scuderi, M., Pinto, A., Gatta, G., & Mazzeo, R. (2004). Gallstone ileus analysis of radiological findings in 27 patients. *European Journal of Radiology*, *50*(1), 23–29. https://doi.org/10.1016/j.ejrad.2003.11.011

Lee, S. B., Ryu, K. H., Ryu, J. K., Kim, H. J., Lee, J. K., Jeong, H. S., & Bae, J. S. (2003). Acute acalculous cholecystitis associated with cholecystoduodenal fistula and duodenal bleeding. A case report. *The Korean Journal of Internal Medicine*, *18*(2), 109–114. https://doi.org/10.3904/kjim.2003.18.2.109

Levy, M. M., Evans, L. E., & Rhodes, A. (2018). The Surviving Sepsis Campaign Bundle: 2018 update. *Intensive Care Medicine*, *44*(6), 925–928. https://doi.org/10.1007/s00134-018-5085-0

Lloyd, D. A., Gabe, S. M., & Windsor, A. C. (2006). Nutrition and management of enterocutaneous fistula. *The British Journal of Surgery*, *93* (9), 1045 – 1055. https://doi.org/10.1002/bjs.5396

Mir, S. A., Hussain, Z., Davey, C. A., Miller, G. V., & Chintapatla, S. (2015). Management and outcome of recurrent gallstone ileus: A systematic review. *World Journal of Gastrointestinal Surgery*, *7* (8), 152 – 159. https://doi.org/10.4240/wjgs.v7.i8.152

Nubiola, P., Badia, J. M., Martinez-Rodenas, F., Gil, M. J., Segura, M., Sancho, J., & Sitges-Serra, A. (1989). Treatment of 27 post-operative enterocutaneous fistulas with the long half-life somatostatin analogue SMS 201-995. *Annals of Surgery*, *210* (1), 56 – 58. https://doi.org/10.1097/00000658-198907000-00009

Nuño-Guzmán, C. M., Marín-Contreras, M. E., Figueroa-Sánchez, M., & Corona, J. L. (2016). Gallstone ileus, clinical presentation, diagnostic and treatment approach. *World Journal of Gastrointestinal Surgery*, *8* (1), 65 – 76. https://doi.org/10.4240/wjgs.v8.i1.65

Paran, H., Neufeld, D., Kaplan, O., Klausner, J., & Freund, U. (1995). Octreotide for treatment of post-operative alimentary tract fistulas. *World Journal of Surgery*, *19* (3), 430 – 434. https://doi.org/10.1007/BF00299182

Räf, L., & Spangen, L. (1971). Gallstone ileus. *Acta chirurgica Scandinavica*, *137* (7), 665 – 675. Retrieved from https://pubmed.ncbi.nlm.nih.gov/5149154/

Reisner, R. M., & Cohen, J. R. (1994). Gallstone ileus: a review of 1001 reported cases. *The American Surgeon*, *60* (6), 441 – 446. Retrieved from https://pubmed.ncbi.nlm.nih.gov/8198337/

Rigler, L.G., Borman, C. N., & Noble, J. F. (1941). Gallstone obstruction: pathogenesis and roentgen manifestaions. *JAMA*, *117*, 1753 – 1759. https://doi.org/10.1001/jama.1941.02820470001001

Rustagi, T., & Aslanian, H. R. (2014). Endoscopic management of biliary leaks after laparoscopic cholecystectomy. *Journal of Clinical Gastroenterology*, *48* (8), 674 – 678. https://doi.org/10.1097/MCG.0000000000000044

Strasberg, S. M., Pucci, M. J., Brunt, L. M., & Deziel, D. J. (2016). Subtotal cholecystectomy-"Fenestrating" vs "Reconstituting" Subtypes and the Prevention of Bile Duct Injury: Definition of the Optimal Procedure in Difficult Operative Conditions. *Journal of the American College of Surgeons*, *222* (1), 89 – 96. https://doi.org/10.1016/j.jamcollsurg.2015.09.019

Tan, Y. M., Wong, W. K., & Ooi, L. L. (2004). A comparison of two surgical strategies for the emergency treatment of gallstone ileus. *Singapore Medical Journal*, *45* (2), 69 – 72. Retrieved from https://pubmed.ncbi.nlm.nih.gov/14985844/