

Combination therapy based on SpyGlass-guided electrohydraulic lithotripsy through cholecystoduodenostomy by lumen-apposing metal stent (SLAMS) for Mirizzi syndrome

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Abstract

Mirizzi syndrome is a rare complication of chronic gallstone disease characterised by the compression of the common bile duct due to an impacted lithiasis in the infundibulum of the gallbladder or cystic duct. In this case study, we discuss an 85-year-old patient who presented with Mirizzi syndrome associated with septic shock. She was not eligible for cholecystectomy due to her advanced age and frailty, requiring a less invasive alternative. Gallbladder drainage was initiated by endoscopic ultrasound-guided cholecystoduodenostomy with a lumen-apposing metal stent (LAMS). Utilising this bulbo-cholecystic stent, SpyGlass-guided electrohydraulic lithotripsy (SGEHL) was then performed, resulting in successful extraction of multiple bile stones, including a 20 mm lithiasis that was lodged in the cystic infundibulum. Subsequently, serum bilirubin levels and inflammatory markers were significantly reduced, consistent with resolution of Mirizzi syndrome. The combination of SGEHL and LAMS, designated as SLAMS, represents a novel, minimally invasive intervention for this potentially life-threatening disease. (*Acta gastroenterol. belg.*, 2024, 87, 424-426).

Keywords : Mirizzi syndrome, cholecystoduodenostomy, lumen apposing metal stents, SpyGlass-guided electrohydraulic lithotripsy, case study.

Introduction

Characterised by the compression of the common bile duct due to an impacted bile stone in either the infundibulum of the gallbladder or cystic duct, Mirizzi syndrome is a rare complication of chronic gallstone disease (1). This compression leads to inflammation and cholestasis, which can result in complications such as fistulae, acute pancreatitis, cholangitis or septic shock if left untreated (2). Laparoscopic cholecystectomy has been used for the treatment of Mirizzi syndrome since 1987 (3). However, open cholecystectomy remains the standard of care for Mirizzi syndrome due to an elevated risk of bile duct injury and a high conversion rate when the laparoscopic approach is used (4,5). Nevertheless, surgery may not always be feasible, especially in elderly patients or those with multiple comorbidities. This is especially pertinent in a globally ageing population, therefore increasing the need for minimally invasive therapeutic strategies. Prior studies have highlighted the increasing role of lumen-apposing metal stents (LAMS) in managing gallbladder drainage when endoscopic retrograde cholangio-pancreatography (ERCP) is not feasible (6). Additionally, the clinical efficacy of

SpyGlass-guided lithotripsy has been demonstrated for the treatment of large common bile duct and cystic duct stones (7,8). To our knowledge, there are no prior literature reports combining these two techniques for the management of biliary obstruction. The aim of this study is to combine SGEHL and LAMS, abbreviated SLAMS, as a less invasive treatment option for inoperable patients with Mirizzi syndrome.

Case history

In this case study, we present an 85-year-old patient who was not eligible for surgery due to her advanced age and frailty. Written informed consent for publication of non-identifying images and non-identifying case report was obtained from the participating patient in accordance with the Declaration of Helsinki. She was admitted through the emergency department and presented with septic shock of biliary aetiology. Broad-spectrum antibiotics and IV hydration were promptly administered after onset of hypotension, jaundice and fever. The initial blood workup revealed elevated bilirubin levels, raised liver enzymes, increased gamma-glutamyl transferase (GGT) and alkaline phosphatase (ALP) levels. Additionally, there was an upsurge in inflammatory markers such as C-reactive protein (CRP), along with raised leukocyte and neutrophil counts (Table 1). A bile fluid culture returned positive for *Escherichia coli* and *Enterococcus faecalis*. Our initial attempt to improve the patient's clinical condition through ERCP with sphincterotomy, lithiasis extraction using a balloon catheter and placement of a plastic biliary stent in the common bile duct was only partially successful. As a consequence, jaundice decreased; however, abdominal pain in the right hypochondriac region and sepsis persisted. The lack of complete clinical resolution was attributed to an additional lithiasis of approximately 20 mm in diameter lodged in the cystic infundibulum, a characteristic feature of type I Mirizzi syndrome.

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Table 1. — Evolution of blood test results prior to and seven days after SLAMS procedure

	Normal range	Before SLAMS procedure	After SLAMS procedure
Total bilirubin (mg/dL)	0.2 - 1.1	3.93	0.34
Direct bilirubin (mg/dL)	≤ 0.3	3.12	0.23
GGT (U/L)	< 38	468	374
ALP (U/L)	46 - 116	209	250
ALT (U/L)	10 - 42	172	50
AST (U/L)	< 34	233	48
CRP (mg/L)	< 10	263.1	37.9
WBC count (x 10e3/μL)	3.9 - 11.1	22.29	7.32
Neutrophil count (/μL)	1800 - 7400	19 994	3 836

GGT: Gamma-glutamyl transferase, ALP: Alkaline phosphatase, ALT: Alanine aminotransferase, AST: Aspartate aminotransferase, CRP: C-reactive protein, WBC: White blood cell.



Figure 1. — X-ray image of SpyGlass-guided EHL (asterisk) passing through the bulbo-cholecystic LAMS (arrow) (SLAMS procedure).

As a result, we opted for an alternative approach, an endoscopic ultrasound-guided cholecystoduodenostomy by LAMS with a diameter of 15mm to initiate gallbladder drainage. As shown in Fig. 1, we then utilised this bulbo-cholecystic LAMS to fragment the isthmus bile stone by performing SGEHL passing through the aforementioned LAMS. We then extracted the remaining biliary debris by endoscopic forceps and loop. To visually capture the efficacy of this combined SLAMS procedure, images were taken during and after the intervention, shown in Fig. 2, demonstrating the removal of the biliary obstruction. In the first image, we present a SpyGlass image of the 20 mm bile stone lodged in the cystic infundibulum responsible for type I Mirizzi syndrome in our patient (Fig. 2A). We then proceeded with SLAMS, involving the fragmentation of the isthmus lithiasis using SGEHL through the LAMS (Fig. 2B). Fig. 2C and 2D collectively demonstrate the resolution of biliary build-

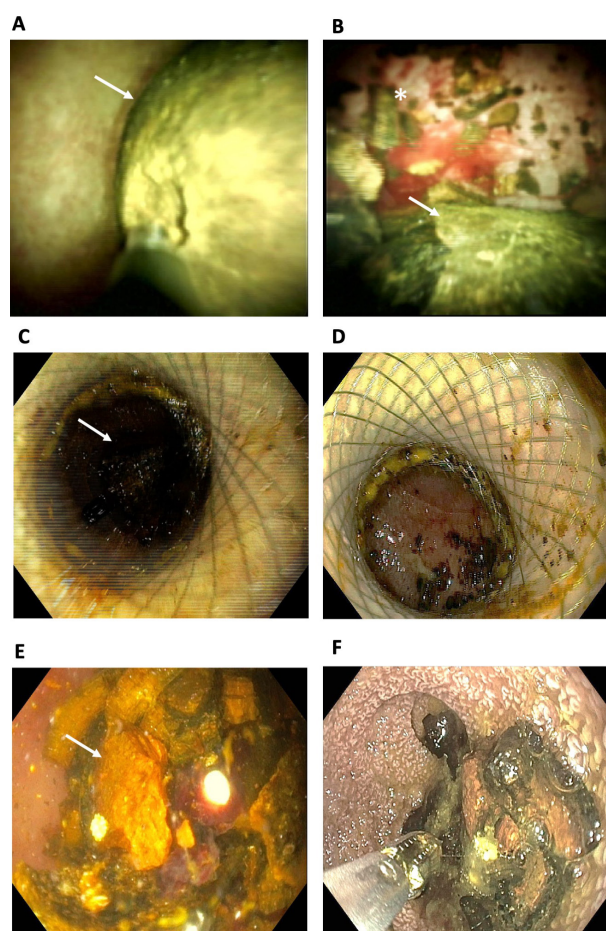


Figure 2. — Per-procedural comparison of SLAMS demonstrating removal of bile obstruction. (A) SpyGlass image of bile stone (arrow), lodged in the cystic infundibulum, responsible for type I Mirizzi syndrome; (B) SGEHL through LAMS of the bile stone (arrow), causing bile stone fragmentation (asterisk); (C) Perspective from the bulbo-cholecystic LAMS showing bile buildup (arrow) before SGEHL; (D) Perspective from the bulbo-cholecystic LAMS showing reduction of bile buildup after SGEHL; (E) Image captured before SLAMS within the gallbladder prior to extraction by endoscopic forceps and loop, revealing an abundance of bile stones (arrow); (F) Post-SLAMS gastro-duodenoscopy showing a significant reduction in bile stones compared to Fig. 2E.

up in the gallbladder after the procedure. Lastly, Fig. 2E and 2F illustrate the significant reduction in bile stones following the SLAMS procedure and subsequent bile extraction. Due to successful fragmentation of the isthmus lithiasis, no recurrence of cholestasis was observed after the removal of the plastic transpapillary stent. As illustrated in Table 1, most blood analyses returned to normal within seven days post-SLAMS, including bilirubin levels and leukocyte counts. We additionally noted a substantial reduction in alanine aminotransferase, aspartate aminotransferase and CRP levels post-SLAMS. GGT levels also showed improvement, but a minor increase in ALP was observed.

Discussion

We hereby establish proof of concept demonstrating the successful treatment of type I Mirizzi syndrome using a novel combination approach called SLAMS, which combines SGEHL and a cholecystoduodenostomy by LAMS. While previous case reports have shown success in treating Mirizzi syndrome by combining SGEHL and ERCP (8), to our knowledge, our study is the first to combine SGEHL and LAMS for this condition. This technique is particularly useful when retrograde access to the cystic duct is limited. In the context of endoscopic ultrasound-guided acute cholecystoenteric drainage using LAMS, a potential complication is stent obstruction caused by macrolithiasis lodging in the stent lumen (9). However, by utilising SLAMS, we can mitigate this risk, as EHL breaks down potentially obstructive macrolithiasis which enables better biliary drainage through the bulbo-cholecystic stent. Previous reports have documented other adverse events associated with LAMS in pancreatic pseudocyst drainage, such as gastrointestinal bleeding, stent dislodgement or buried LAMS syndrome (10). However, it is important to note that our case report focuses on gallbladder drainage, which typically involves a thicker and less fragile, non-necrotic wall compared to a pancreatic wall of necrosis. In certain patients, the external compression of the common bile duct can lead to fistulisation between the common hepatic duct and the gallbladder, causing type II Mirizzi syndrome. Further studies are needed to evaluate

the possibility of SLAMS for type II Mirizzi syndrome. The main advantage of SLAMS is that it allows for a minimally invasive intervention compared to surgical cholecystectomy, which represents the current standard of care for the management of Mirizzi syndrome. This study highlights the potential life-saving benefits of SLAMS in patients presenting with septic shock of biliary aetiology who are ineligible for surgery. Thus, SLAMS could be a valuable addition to our current therapeutic arsenal for managing inoperable patients presenting with Mirizzi syndrome. Further research is necessary to assess the safety and efficacy profiles of SLAMS for the treatment of cystic duct obstruction in larger patient cohorts.

Conflict of interest

All authors declare no conflict of interest.

References

1. KLEKOWSKI J., PIEKARSKA A., GORAL M., KOZULAM., CHABOWSKI M., The Current Approach to the Diagnosis and Classification of Mirizzi Syndrome. *Diagnostics*, 2021,11:1660.
2. ABOU-SAIF A., AL-KAWAS F.H., Complications of gallstone disease: Mirizzi syndrome, cholecystocholedochal fistula, and gallstone ileus. *American Journal of Gastroenterology*, 2002,97:249-254.
3. CUSCHIERIA., DUBOIS F., MOUIEL J., MOURET P., BECKER H., BUESS G., et al., The European experience with laparoscopic cholecystectomy. *American Journal of Surgery*, 1991,161:385-387.
4. CHEN H., SIWO E.A., KHU M., TIAN Y., Current trends in the management of Mirizzi Syndrome: A review of literature. *Medicine*, 2018,97:e9691.
5. KULKARNI S.S., HOTTA M., SHER L., SELBY R.R., PAREKH D., BUXBAUM J., et al., Complicated gallstone disease: diagnosis and management of Mirizzi syndrome, *Surgical Endoscopy*, 2017,31:2215- 2222.
6. MUSSETTO A., FUGAZZA A., FUCCIO L., TRIOSI O., REPICI A., ANDERLONIA., Current uses and outcomes of lumen-apposing metal stents. *Annals of Gastroenterology*, 2018,31:535-540.
7. LI G., PANG Q., ZHAI H., ZHANG X., DONG Y., LI J., et al., SpyGlass-guided laser lithotripsy versus laparoscopic common bile duct exploration for large common bile duct stones: a non-inferiority trial. *Surgical Endoscopy*, 2021,35:3723-3731.
8. ISSA H., BSEISO B., ALMOUSA F., AL-SALEM A.H., Successful Treatment of Mirizzi's Syndrome Using SpyGlass Guided Laser Lithotripsy. *Gastroenterology Res*, 2012,5:162-166.
9. BANG J.Y., HASSAN M., NAVANEETHAN U., HAWES R., VARADARAJULU S., Lumen-apposing metal stents (LAMS) for pancreatic fluid collection (PFC) drainage: may not be business as usual. *Gut*, 2017,66:2054-2056.
10. AHMED O., ROGERS A.C., BOLGER J.C., MASTROSIMONE A., LEE M.J., KEELING A.N., et al., Meta-analysis of outcomes of endoscopic ultrasound-guided gallbladder drainage versus percutaneous cholecystostomy for the management of acute cholecystitis. *Surgical Endoscopy*, 2018,32:1627-1635.