Mirizzi syndrome in a patient with partial gastrectomy with Billroth II anastomosis: A case report

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ABSTRACT

INTRODUCTION: Mirizzi Syndrome (MS) is a common bile duct (CBD) obstruction caused by extrinsic compression from an impacted stone in the cystic duct or infundibulum of the gallbladder [1,2]. It's not always recognized immediately at radiological evaluation, so it may be mistaken for common bile duct (CBD) stones in jaundiced patient. Moreover, confusion in diagnosis of this condition in jaundiced patients who underwent gastric surgery may cause a wrong management and a delayed treatment. In addition, patients who have altered anatomy of upper gastrointestinal (GI) tract are clearly challenging in case of endoscopic treatment, and difficulty usually depends on the type of gastric surgery previously performed. Endoscopic retrograde cholangiopancreatography (ERCP) remains a challenging procedure in patients with altered anatomy, such as sub-total gastrectomy (STG) with Billroth II (STG B–II) anastomosis [3–5], and its indication is usually the extraction of CBD stones [6,7].

Sub-total gastrectomy was commonly performed as treatment of choice in peptic ulcer starting from 1910 [8], even if nowadays it’s indicated just in case of perforated gastric ulcers larger than 2 cm or bleeding gastric ulcers (after failure of repeated endoscopy or angiographic embolization) [9] for the possibility of neoplasms, while it has obviously kept other indications as malignant and benign stomach disease [10]. STG may have a variety of options available to restore gastrointestinal continuity, the most common of which are the Billroth I (STG B–I), Billroth II (STG B–II), and Roux-en-Y reconstructions. Many studies analyzed patients with STG B–I or STG B–II anastomosis and total gastrectomy (TG) and their association with cholelithiasis, due to its incidence in these patients [11–15]. The incidence of cholelithiasis after gastric surgery, indeed, is between 10 and 25% [16]. STG B–II is associated with an increased risk of gallstones and biliary complications [17].

1. Background

Mirizzi Syndrome is a common bile duct obstruction caused by extrinsic compression from an impacted stone in the cystic duct or infundibulum of the gallbladder [1,2]. It’s not always recognized immediately at radiological evaluation, so it may be mistaken for common bile duct (CBD) stones in jaundiced patient. Moreover, confusion in diagnosis of this condition in jaundiced patients who underwent gastric surgery may cause a wrong management and a delayed treatment. In addition, patients who have altered anatomy of upper gastrointestinal (GI) tract are clearly challenging in case of endoscopic treatment, and difficulty usually depends on the type of gastric surgery previously performed. Endoscopic retrograde cholangiopancreatography (ERCP) remains a challenging procedure in patients with altered anatomy, such as sub-total gastrectomy (STG) with Billroth II (STG B–II) anastomosis [3–5], and its indication is usually the extraction of CBD stones [6,7].

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We present a rare case of a STG B–II patient with jaundice, who was affected by Mirizzi syndrome. We present this case report in line with the SCARE 2018 criteria [18].

2. Presentation of the case

A 69-year-old male in otherwise good health with a history of STG B–II 25 years prior for gastric ulcer arrived at the emergency department reporting severe thoracic and abdominal pain beginning after eating lunch. The pain was constant, localized to right thorax and upper-right quadrant of abdomen and up to a 7 out of a possible 10 in intensity. The pain was associated with nausea and mild fatigue. The patient denied fever, jaundice, change in the color of her urine, and change in the color of his stool. The patient’s past medical and surgical histories included angina around 20 years before and nothing else. He did not take any medication or herbal supplement, and he had no allergy. He denied a history of jaundice, tattoo, intravenous drug use, sexual contact with an individual known to have liver disease. He had no known family history of gastrointestinal, hepatic, biliary, or pancreatic disease.

Blood pressure was 125/80 mmHg, pulse was 75 beats/minute, respirations were 15 breaths/minute, and temperature was 36.7°C. The skin was jaundiced, but without telangiectasia or palmar erythema. An electrocardiogram and cardiologist consultation excluded a heart attack or some cardiological etiology of the pain.

Initial laboratory results revealed white blood cells 9.76 × 10⁹/jul (normal range, 4.0–10.0 × 10⁹/jul), 87.4% (37–80%) of which were segmented neutrophils, hemoglobin 11.0 g/dl (13–17.0 g/dl), platelets 241 × 10³/jul (150–450 × 10³/jul), international normalized ratio (INR) 0.85 (0.8–1.1), albumin 4.1 g/dl (3.5–5.0 g/dl), total bilirubin 3.24 mg/dl (0.3–1.0 mg/dl), direct bilirubin 3.2 mg/dl (0.0–0.2 mg/dl), gamma glutamyl transferase (γGT) 506 (8–61 U/L), alkaline phosphatase 120 U/L (40–129 U/L), aspartate aminotransferase (AST) 213 U/L (3–50 U/L), alanine aminotransferase (ALT) 299 U/L (3–50 U/L), lipase 10 U/L (0–90 U/L), amilase 5 U/L (8–53 U/L), and Reactive C protein 72.5 (0–5 mg/L).

An ultrasound of the abdomen identified a calcification of 2 cm in maximal dimension within the fifth hepatic segment and some sporadic biliary cyst. Intrahepatic bile ducts and common bile duct were normal. Gallbladder was noted to have biliary sludge in the infundibulum. Moreover, abdomen CT scan with contrast was performed, but it was inconclusive according to radiologist opinion, despite it has been useful to exclude hepatobiliary and pancreatic malignancies with higher specificity than ultrasound of abdomen.

Therefore a broad differential diagnosis of acute cholestatic hepatitis was entertained, including acute viral infectious hepatitis, initial ischemic hepatitis, and occult hepatotoxicity. Extrahepatic biliary obstruction was also considered. Acute alcoholic or autoimmune hepatitis was thought to be unexpected. Viral markers, including markers for hepatitis B virus (HBV) and hepatitis C virus (HCV), were negative. Cancer markers, as alpha-1 fetoprotein (AFP), CA 125, CA 19.9, carcinoembryonic antigen (CEA), were negative as well.

An MRCP (Magnetic Resonance Cholangiopancreatography) identified a mild dilatation of intrahepatic bile ducts, and a normal common bile duct (6 mm diameter) with a 4 mm biliary stone and sludge inside it (Fig. 1). Gallbladder was distended with biliary sludge. Considering the altered upper GI anatomy, laboratory tests and MRCP, which has higher sensibility and specificity than ultrasound to identify common bile duct stones, a careful ERCP was performed by an experienced endoscopist (more than 300 ERCP procedures per year). Patient was under conscious sedation with midazolam and rectal indomethacin suppository (100 mg) was administered before the procedure. As expected it was technically hard with duodenoscope to reach the duodenal stump, as is known [19], so that endoscopist had to use a gastroscope to reach papillary region. Therefore cannulation of papilla was achieved through the gastroscope thanks to the high expertise of the endoscopist (Fig. 2A). Cholangiography showed common bile duct without any defect, but it showed a cystic duct with a biliary stone inside, compressing common bile duct with an hepatic duct and intrahepatic duct dilated (Fig. 2B–D). Diagnosis of Mirizzi syndrome was made, and it was classified as Type I according to Csendes et al. Classification [20]. A biliary drainage with a biliary “pigtail” probe was performed and surgical treatment was planned. Endoscopic procedure was well tolerated and patient did not develop any complication post-ERCP.

Laparoscopic cholecystectomy was performed, observing a cystic duct adhering to common bile duct, confirmed during an intra-surgery cholangiography once again (Mirizzi Syndrome – Fig. 3). Cystic bile duct was identified and moved away from common bile duct, and successively isolated and sectioned. Cholecystectomy was concluded and histological analysis of gallbladder showed a chronic cholecystitis.

![Fig. 1. Two different scans (A e B) of MRCP showing infundibulum of gallbladder next to common bile duct.](image-url)
Fig. 2. Cholangiography during ERCP: A, cannulation achieved with a gastroscope; B,C, Opacification of biliary tree with infundibulum of gallbladder compressing common bile duct (Mirizzi Syndrome Type I); D: Complete opacification of biliary tree, including gallbladder.

Antibiotic prophylaxis with piperacillin/tazobactam was administered immediately before operation and it was kept for 48 h after surgery. Post-surgery laboratory results showed white blood cells $11.05 \times 10^3$/ul, 80% of which were segmented neutrophils, hemoglobin 11.4 g/dL, platelets $210 \times 10^3$/ul, INR 0.95, albumin 3.9 g/dL, total bilirubin 0.9 mg/dL, direct bilirubin 0.5 mg/dL, $\gamma$GT 350 U/L, alkaline phosphatase 136 U/L, AST 225 U/L (3–50 U/L), ALT 305 U/L, normal lipase and amilase.

A second look of the CT scan previously performed showed infundibulum adhering common bile duct (Fig. 4). Two days after surgery cholangiography through biliary “pigtail” drainage was performed, confirming the lack of a biliary leak, a normal CBD and a correct biliary flow into duodenum (Fig. 5).

Patient was discharged asymptomatic with normal laboratory tests, except for a residual cholestasis ($\gamma$GT 230 U/L), and follow-up at 1 and 3 months was scheduled. At subsequent follow-up appointment with his primary care physician, the patient reported no biliary symptoms and normal laboratory tests.

3. Conclusions

The present case report highlights the importance of including Mirizzi syndrome in the differential diagnosis of biliary obstruction in patients with upper GI altered anatomy, as those with partial gastrectomy with Billroth II anastomosis. In fact in this subset of patients, as showed in our case, even when second level imaging indicates common bile duct stone, a wrong initial radiological diagnosis may be made. In fact, although a non-invasive 2nd level imaging as CT and MRCP narrows the differential diagnosis, Mirizzi syndrome might be discovered or confirmed only at ERCP, especially in those patients with altered anatomy. Moreover, as in our case, if cholangiography during ERCP shows Mirizzi syndrome, the management includes to schedule cholecystectomy as soon as possible. Physicians practicing in a community hospital with expertise in ERCP should always consider altered anatomy as a factor which may confuse radiologist in diagnosis of biliary stone disease.
Fig. 3. Cholangiography during laparoscopic cholecystectomy: cystic duct and infundibulum (arrow) brought away from common bile duct before section of duct itself. CBD does not result compressed anymore.

Fig. 4. CT scan of abdomen: a mediocre view of common bile duct (red arrow) adhered and partially compressed from infundibulum (white arrow).

Fig. 5. Cholangiography through biliary “pigtail” drainage two days after cholecystectomy: normal biliary tree with regular contrast flow into duodenum (arrow).

Declaration of Competing interest

The authors report no declarations of interest.

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Ethical approval

This report is exempt from ethical approval in our institution.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contributions

Dr. G.E.M.R.: Conceptualization, Methodology, Resources, Literature search, manuscript writing, design and image providing.
Dr. G.D.C.: Software.
Dr. G.R.: Writing, Methodology and image providing.
Dr. G.F.: Literature search and image providing.
Dr. G.C.: Software.
Prof. C.S.: Supervision, Comments and data analysis.

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