



Case report

Duodenal perforation secondary to stent migration after ERCP for hepatobiliary tuberculosis: Case report of a lethal complication in a young patient

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ABSTRACT

Introduction: Interventional internal drainage of the biliary tract has become an established procedure for the temporary and definitive treatment of biliary obstruction due to malignant or benign disease. The complication rate is reported to be so low that when feasible, this technique is preferred over a surgical drainage procedure.

Presentation of a case: A 26-year old woman was referred to the hepatopancreaticobiliary surgery service due to severe abdominal pain for 3 days after undergoing endoscopic retrograde cholangiopancreatography (ERCP). She underwent biliary dilatation and stent insertion for obstructive jaundice secondary to biliary stricture from hepatobiliary tuberculosis. The patient underwent exploratory laparotomy, peritoneal lavage, duodenorrhaphy and tube jejunostomy for bilious peritonitis and duodenal perforation from biliary stent migration. The patient died one day post-operation due to septic shock from secondary bacterial peritonitis.

Discussion: ERCP and other interventional endoscopic biliary interventions are increasingly being used for biliary obstruction. Despite the various complications which arise from these diagnostic and therapeutic modalities, complications are relatively uncommon. Duodenal perforation from biliary stent migration is a rare complication after undergoing ERCP and stenting. However, in patients presenting with severe pain and physical signs of acute abdomen after the procedure, it should always be a consideration.

Conclusions: Despite the relative safety of interventional techniques for biliary obstruction, complications like pancreatitis, hemorrhage and perforation may occur. Early recognition and high index of suspicion allows for early intervention with good outcomes. Duodenal perforation from stent migration can occur and when intervention is delayed may lead to morbidity and mortality.

1. Introduction

Endoscopic intervention has become an integral part of the management of complex biliary diseases. The utility of endoscopy has gone beyond the diagnostic capabilities of endoscopic retrograde cholangiopancreatography (ERCP), enabling placements of stents and prosthesis to bypass biliary obstruction both as therapeutic and palliative procedures [1]. Despite the minimally invasive nature of the procedure, these interventions may lead to complications. ERCP by itself may lead to pancreatitis, hemorrhage, duodenal injury etc. Likewise, migration of biliary stents applied after ERCP has been reported in the literature. Dislocation and migration is a late complication after

endoscopic stenting and it has been reported to occur in about 7% (0–40%) of cases [2]. Proximal migration, the further advancing of the stent into the biliary duct, may lead to biliary obstruction. Its correction is technically challenging, but it can usually be achieved endoscopically by forceps, snare, or balloon. In the early stages of distal migration, the stent can be replaced endoscopically. Endoscopic treatment is limited to early and endoscopically accessible cases. In most other cases, the stent passes naturally due to the contractions and distensibility of the intestinal wall. Complications due to distal migration other than the recurrence of biliary obstruction have only been reported anecdotally. With distal migration, bowel obstruction or perforation may occur, requiring surgical intervention. This occurs in about 10% of patients, with a higher

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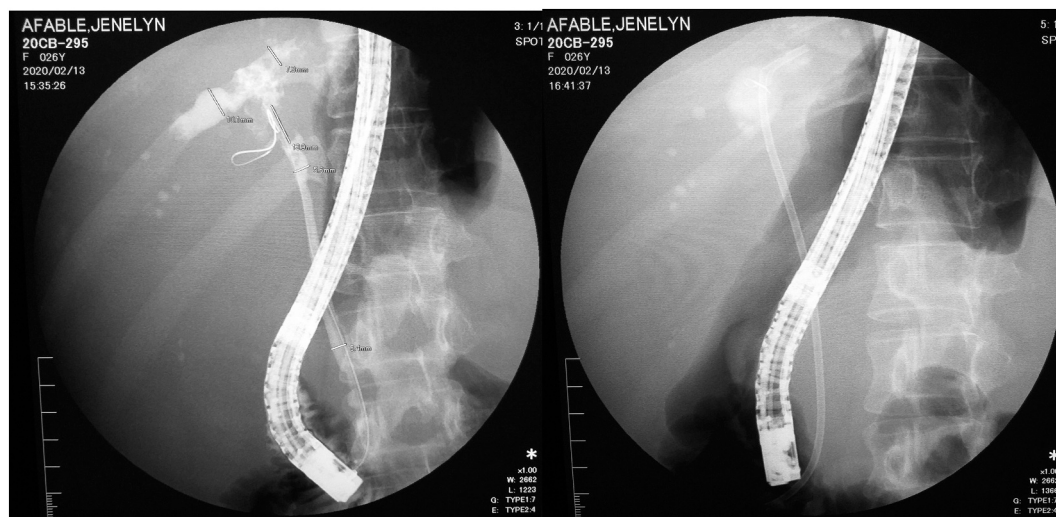


Fig. 1. Endoscopic retrograde cholangiopancreatography which showed nondilated common bile duct, tapering of the common hepatic duct and dilated intrahepatic ducts (left) and insertion of Fr 8.5 17 cm biliary plastic stent.

risk for patients with benign biliary strictures. Less than 1% of migrated stents cause intestinal perforation with a mortality rate below 1% [3].

Presented herein is a case of a rare complication of stent migration after biliary stenting for hepatobiliary tuberculosis. This unfortunate complication led to the demise of a young patient with benign biliary disease. Despite the low incidence of this complication, intestinal perforation secondary to biliary stent migration should be a differential diagnosis in all patients presenting with fever and abdominal pain after biliary stent insertion. This rare and unfortunate complication is being reported in line with the SCARE guidelines [4].

2. Case presentation

The patient is a 26-year-old female from Manila presented with a 5-month history of jaundice associated with intermittent epigastric pain, weight loss, pruritus and tea-colored urine. She was initially seen at the outpatient department and diagnosed as a case of obstructive jaundice probably secondary to choledocholithiasis based on ultrasound done in an outside institution. The patient had no other comorbidities or previous surgeries. On physical examination, the patient was normotensive with a blood pressure of 110/70 mmHg, heart rate of 80 bpm and afebrile. She had icteric sclerae, soft, nondistended, nontender abdomen

with generalized jaundice.

Transabdominal ultrasound revealed a non-dilated common bile duct with note of intrahepatic ductal dilatation with areas of segmental dilatation. There was note of bile duct sludges. The diagnosis was revised to choledocholithiasis with possible biliary stricture secondary to hepatobiliary tuberculosis. Magnetic resonance cholangiopancreatography was deferred due to financial constraints. The plan was to do possible ERCP extraction of stones and possible stenting of the biliary stricture suspected on ultrasound.

The scope was inserted up to the second part of the duodenum, and the ampulla was visualized to be patulous with minimal bile egress. Cannulation was done using the sphincterotome-guidewire technique with slight difficulty. Initial cholangiogram showed a non-dilated common bile duct with tapering at the common hepatic duct measuring 13.9 mm long. Intrahepatic ducts were dilated, with the right IHD measuring 10.1 mm and the left IHD measuring 7.3 mm. No filling defects were noted. Gradual dilation of the CHD stricture was done using French 7–10 dilators, with subsequent passage of the sphincterotome beyond the site of the stricture. Partial sphincterotomy was done and a 17 cm Fr 8.5 plastic biliary stent was inserted with good bile egress (Fig. 1). TB polymerase chain reaction (TB-PCR) testing of bile samples revealed tuberculosis.

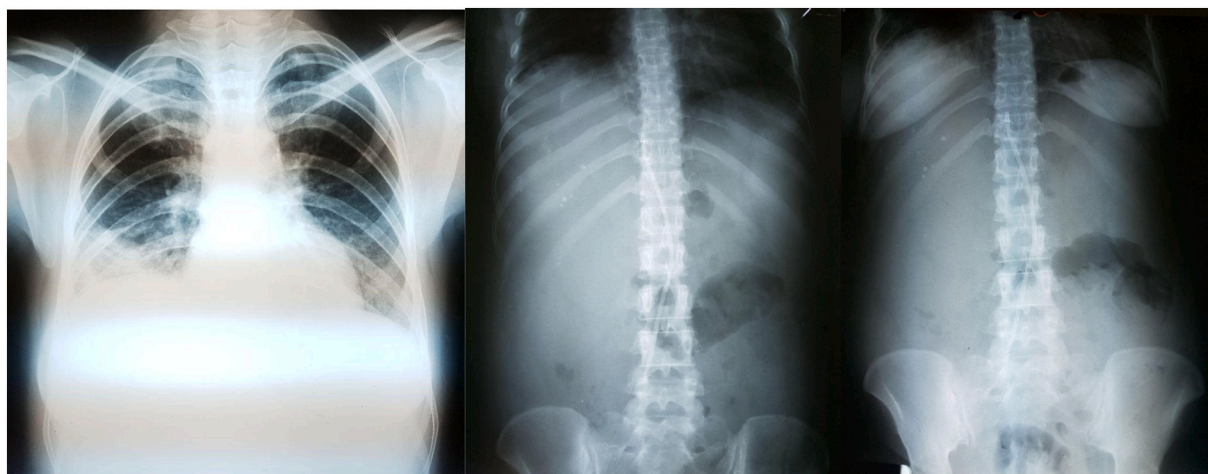


Fig. 2. Chest X-ray (left), Abdominal X-rays supine (middle) and upright (right) with no signs of pneumoperitoneum and perforation. There is note of right basal density probably due to effusion or subdiaphragmatic fluid.

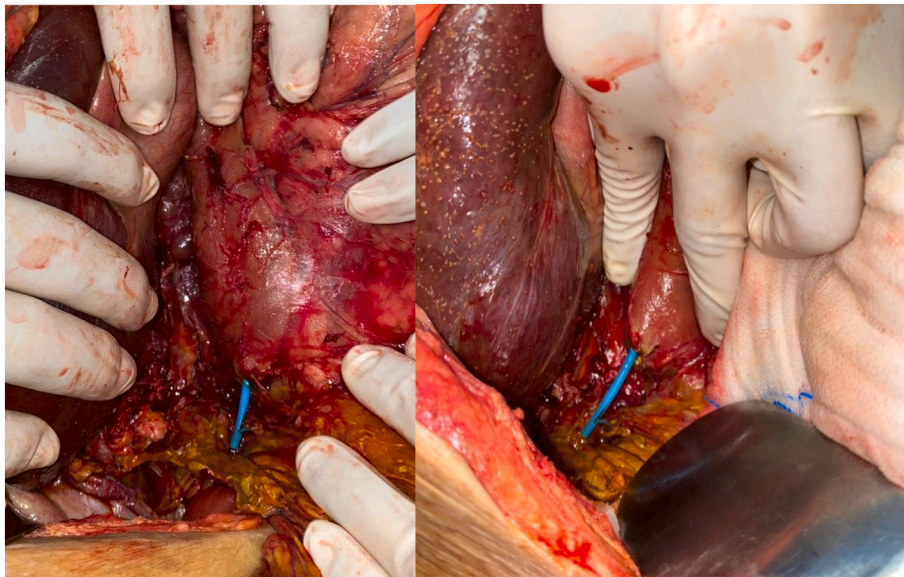


Fig. 3. Intraoperative image showing the plastic stent perforating the duodenum and protruding into the peritoneal cavity.

After undergoing endoscopic retrograde cholangiopancreatography, the patient was admitted for monitoring. She had episodes of occasional abdominal pain with visual analogue score of 8/10 (using a pain scale of 0 pain with 10 as maximal pain) during the first post-endoscopy day which was slightly relieved with analgesics. Discharge was deferred the following day when she developed fever, tachycardia and epigastric tenderness. The impression at that time was post-ERCP pancreatitis. Blood tests done revealed a slightly elevated amylase level of 125 U/L and a normal lipase level of 234 U/L. Creatinine (43 $\mu\text{mol/L}$), albumin (35 g/L), hemoglobin (101 g/L), hematocrit (31%), WBC count ($5.30 \times 10^9/\text{L}$), platelet count ($185 \times 10^9/\text{L}$), prothrombin time (13.9 s), PT% (86%), and PT-INR (1.11) were all normal.

On her third post-endoscopy day, the patient still had febrile episodes but now with generalized abdominal pain and hypotension as low as 70/40 mmHg, which only slightly improved with hydration. The abdomen was slightly distended, with direct and rebound tenderness on all quadrants, which prompted referral to the hepatopancreaticobiliary surgery service.

When the patient presented with generalized abdominal pain on the 3rd day post-procedure, an acute abdomen was considered possibly from iatrogenic bile duct injury or intestinal perforation. Chest and abdominal radiographs done were negative for pneumoperitoneum and showed no signs of perforation (Fig. 2). An abdominal ultrasound revealed nondilated intrahepatic ducts and common bile duct with the stent in place. At this time, elevations in the WBC count ($12.9 \times 10^9/\text{L}$), prothrombin time (22.7 s), PT-INR (1.83) and activated partial thromboplastin time (43.9 s) were seen. Broad-spectrum antibiotics were started, and Vitamin K infusion and FFP transfusion were given due to deranged bleeding parameters. Norepinephrine was started due to persistent hypotension despite hydration. The patient underwent urgent exploratory laparotomy, performed by a senior surgical resident supervised by an attending where 300–400 cc of bile was initially drained from the peritoneal cavity. There was note of generalized peritonitis with preponderance of purulent material at the right paracolic gutter. There was note of a perforation between the 2nd and 3rd portion of the duodenum through which the stent was seen to be protruding (Fig. 3). Duodenorrhaphy with primary repair, peritoneal lavage and tube jejunostomy were performed.

After surgery, the patient remained intubated and was admitted to the surgical intensive care unit. She had increasing pressor requirements, no response to painful stimuli with slowly reactive pupils. The patient died on the first day post-surgery after developing persistent

hypotension despite pressor support and supraventricular tachycardia.

3. Discussion

Endoscopic retrograde cholangiopancreatography with biliary stenting is an established treatment for obstructive jaundice secondary to benign or malignant strictures. It is generally regarded to cause less morbidity than surgery and is also a valuable treatment option for postoperative biliary leaks, obviating the need for reoperation. In the Philippines, the prevalence of hepatobiliary tuberculosis is relatively high compared to developed countries. Cases of Hepatobiliary tuberculosis in the country requiring intervention usually undergo transhepatic drainage, endoscopic stenting and occasionally liver resection with or without biliary bypass. These procedures are performed complementary to medical management of extrapulmonary TB. However, the use of and deployment of biliary stents may be associated with several complications such as pancreatitis, bleeding, infection, obstruction and repeated procedures. Stent migration can occur in up to 10% of patients undergoing biliary stenting, resulting in mortality in less than 1% of cases [3,4]. Migration is more common in patients with benign biliary pathologies [5,6], possibly due to strictures loosening once ductal inflammation and mucosal edema subside. Tumor growth in malignant strictures may also help anchor the stent and prevent migration. Malignant strictures, larger diameter stents, and short stents are known to be associated with proximal biliary stent migration. Stent-related factors such as the type, length, and caliber of the stent offer potential avenues to minimize the risk of migration. Distal migration and related duodenal injury are more likely to present during the early period after biliary stenting [5]. Diameter and length of stents may also have a relation with the rate and direction of migration, as shorter stents tend to migrate proximally and longer stents tend to migrate distally in benign biliary strictures [6].

Most migratory stents can be retrieved using upper gastrointestinal endoscopy and ERCP. If the stent migrates to the intestines, 43% may pass spontaneously [3]. Occasionally, biliary stents may impact and perforate the fixed intestinal segments such as the duodenum or right side of the colon, or in other areas which may become fixed by adhesions from a previous operation. Intestinal perforation may cause biloma formation or biliary peritonitis. When this is suspected, surgery is the treatment of choice.

Similar cases were reported in a retrospective observational study by Gromski et al., where in six cases of duodenal perforation involving

Table 1

Clinical and procedural information of patients with intestinal perforation from stent migration.

Patient, age (years), gender	Indication for ERCP	Biliary stent used	Time to perforation identification (days post-ERCP)	Treatment of perforation
1. 51, male ^[3]	Choledocholithiasis	10 Fr by 10 cm plastic stent in the distal common bile duct	14	Antibiotics, laparotomy
2. 79, female ^[4]	Benign biliary stricture	10 Fr by 9 cm stent	30	Exploration of left groin, drainage of abscess
3. 63, female ^[5]	Pancreatic head mass	10 Fr by 10 cm plastic stent	1	Laparoscopy
4. 52, female ^[6]	Extrahepatic cholangiocarcinoma	8.5 Fr by 10 cm plastic stent	21	Laparotomy
5. 79, female ^[7]	Biliary stricture	7 Fr by 12 cm plastic stent into the right hepatic duct, 10 Fr by 15 cm plastic stent into the left hepatic duct	1	Endoscopy
6. 65, female ^[8]	Pancreatic cancer, cholangitis	10 Fr by 10 cm plastic stent	62	Laparotomy

biliary stents were reported after endoscopic retrograde cholangiopancreatography for both benign and malignant cases [7]. The mean age was 65.2 years, 4 of the procedures were performed for malignant biliary obstruction, and plastic biliary stents were used in 6 cases. Stent perforation was discovered at a mean of 7.5 days after stent placement. All perforating plastic stents were noted to have a length greater than 9 cm.

Compared to the other case reports reviewed as summarized in Table 1 [8–11], the patient's age in this case is very much younger. The French size of the stent used in this case (Fr 10) is also comparable with most of the other cases but is noted to be much longer (17 cm). More benign cases were also noted which supports the literature earlier cited. Laparotomy was also used in majority of the cases but with more favorable outcomes than in this case. Time to perforation identification varied greatly among these reports.

The unfortunate outcome in this case was due to the delay in recognition of the problem, attributing the deterioration to progressing pancreatitis. In terms of stent characteristics, the length of the stent used warrants further investigation. The decision to use the 17 cm stent is probably due to the proximal location of the stricture. The fulminant course of the sepsis may have been aggravated by the immunocompromised status of the patient, having been afflicted with hepatobiliary tuberculosis.

In this case, the surgical team elected to do primary repair and tube jejunostomy. In more favorable situations, surgery for duodenal perforation may entail pyloric exclusion or other techniques of covering the defect like patching. The decision to do a less than ideal procedure was brought about by the poor overall condition of the patient and the need to limit the operative time.

4. Conclusion

While duodenal perforation from biliary stent migration is a rare complication, it should always be included in the differential diagnosis in patients presenting with abdominal pain after ERCP. Early detection of such complications is essential since a delay in diagnosis is a major risk factor for morbidity and mortality. Immediate surgical intervention is warranted in cases of patient deterioration due to perforation and peritonitis. A high index of suspicion for intestinal perforation and peritonitis is required when patients present with signs of intra-abdominal sepsis after endoscopic biliary interventions. Appropriate imaging should be done to confirm the diagnosis and difficult cases are best treated in tertiary hospitals, managed operatively by experienced surgeons.

Provenance and peer review

Not commissioned, externally peer-reviewed.

Consent

Written informed consent was obtained from the patients next of kin 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.

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Guarantor

Anthony R. Perez, MD, MHA.

Ethical approval

Ethics approval obtained from University of the Philippines Ethics Review Board.

Registration of research studies

Not applicable.

CRediT authorship contribution statement

Anthony R. Perez, MD: Study concept, writing the paper, final draft.
Hans Jesper F. Del Mundo, MD - Data collection, review of literature, writing the paper.

Brent Andrew G. Viray, MD: Data collection, study design, manuscript editing.

Juan Carlos Abon, MD: Data collection, review of literature, final draft.

Derek C. Resurreccion, MD- writing the paper, final editing.

Declaration of competing interest

There were no conflicts of interest.

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