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DOI: 10.17235/reed.2018.5815/2018
Link: PubMed (Epub ahead of print)

Please cite this article as:

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Endoscopic sump syndrome secondary to EUS-guided choledocho-duodenostomy with a lumen-apposing metal stent

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Received: 15/07/2018
Accepted: 30/09/2018

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SUMMARY
Sump syndrome (SS) is associated with choledocho-duodenostomy (CDD) dysfunction, which occurs due to accumulation of detritus, biliary mud and food remains in the suprapapillary distal common bile duct. The prevalence is low after CDD. Currently, biliary drainage endoscopic ultrasound (EUS)-guided with a lumen-apposing metal stent (LAMS) is a new minimally invasive alternative for biliary stenosis for patients in whom endoscopy retrograde cholangial-pancreatography (ERCP) is not feasible. CDD via EUS-guided LAMS is increasing. Thus, SS has become a potential associated complication that was previously unreported in the literature.

Conflicts of interest: Dr. Manuel Pérez-Miranda is a consultant for Boston Scientific and M.I. Tech and is a speaker for Boston Scientific and Olympus.
The other authors have no conflicts of interest. The work had no sponsorship or funding.

INTRODUCTION
The sump syndrome is a complication that is rarely associated with biliary-enteric anastomosis (1). Although infrequent, it has traditionally been associated with CDD dysfunction. Food debris, detritus and biliary mud are deposited in the distal section of the suprapapillary common bile duct, which results in repeated cholangitis due to over infection of the detritus and the formation of abundant biliary mud and calculi (2). The possible complications associated with this syndrome include cholangitis, pancreatitis, liver abscesses and secondary biliary cirrhosis, which are often the cause of death in these patients. Different studies show a low but variable prevalence, between 0 and 9.6% (3-6). The prevalence is lower nowadays as less lateral anastomosis are performed in patients with benign biliary obstruction. CDD is also performed less frequently as a palliative procedure for unresectable distal choledochal cancer, as it allows bile drainage whilst avoiding the diseased and obstructed biliary segment. Recently, biliary drainage EUS-guided LAMS has been used as a minimally invasive alternative for the management of biliary strictures in patients in whom ERCP is not feasible or in non-surgical candidates.

CASE REPORT
A 61-year old male with multiple comorbidities and ASA III (American Society Anesthesiology) was admitted due to obstructive jaundice, pruritus and weight loss. He had an emaciated physical appearance, with marked jaundice, no abdominal pain and no ascites. The analytical tests showed total bilirubin levels of 34.2 mg/dl, alkaline phosphatase of 1,100 mg/dl and INR of 1.34. A failed ERCP was performed at his reference hospital. A biliopancreatic EUS identified a neoplastic duodenal stenosis with infiltration of the duodenal papillary, and a 32 mm nodular lesion was also observed in the head of the
pancreas (Fig. 1A-C). Thus, an EUS-guided puncture with a 22 G needle (three passes) and mucosal duodenal biopsies were performed. Multiple peripancreatic adenopathies, ascites and space-occupying lesions in the liver were identified. Thus, the bile duct was drained by a transduodenal insertion of an 8 x 8 mm Hot Axios® LAMS and a coaxial double pigtail catheter (Fig. 1D). Furthermore, the duodenal stenosis was resolved using a short metal prosthesis to minimize the risk of interference with the CDD. A clinical improvement was registered and the control BT was 2.01 mg/dl after two weeks of follow-up. The biopsies and pancreatic FNA identified stage IV pancreatic adenocarcinoma. Thus, it was considered as an advanced pancreatic cancer and a non-surgical candidate.

Three months later, the patient presented to the Emergency Room due to severe jaundice (total bilirubin 19.9 mg/dl), fever and vomiting. The control bilirubin level was 19.9 mg/dl, gamma glutamyl transpherase was 996 mg/dl and alkaline phosphatase was 1,739 mg/dl. An abdominal echography identified a moderate dilatation of the intrahepatic biliary duct and poor visualization of the extra-hepatic duct. An abdominal computed tomography (CT) scan identified a severe dilatation of the intra and extrahepatic duct, with a suspicion of a liver and peri-choledochal abscess (Fig. 2A and B).

The duodenal stenosis was resolved during the previous procedure, thus, ERCP was initially performed at this time. In this procedure, sphincterotomy was performed and abundant biliary mud and purulent content were obtained. A metallic stent of 6 cm x 10 mm and double pigtail were also placed in order to minimize the risk of migration. Liver and peri-choledochal abscesses were ruled out by echoendoscopy. Furthermore, a coaxial covered metal stent of 6 cm x 8 mm was placed through the CDD (Fig. 2C). Purulent content, food debris and biliary drainage were observed (Fig. 2D).

**DISCUSSION**

The sump syndrome is a rare complication that is classically described in surgical CDD. Until now, no cases of SS have been described in relation to endoscopic CDD, probably due to the fact that this technique is performed in international reference centers with expert echoendoscopists. On the other hand, the complications most frequently described
in larger series have occurred early in relation to the procedure and not during the long-term follow-up, which is often limited by the underlying malignant pathology.

The experience with EUS-guided biliary drainage was first published in 2001 by Giovannini et al. (7), followed by a small case in 2004 where EUS was used to perform rendez-vous in pancreatic and biliary obstructive lesions (8). Two large multicenter studies were recently published in reference centers with expert endoscopists, with failed cases of endoscopic drainage via ERCP (9,10). The complications described in relation to the procedure were pneumoperitoneum in 5% of cases, bleeding in 11%, biliary leak/peritonitis in 10%, cholangitis in 5% and abdominal pain (10).

It is possible that the prevalence of this entity will increase due to the increased use of endoscopic choledocho-duodenostomy to palliate symptoms of double stenosis (duodenal and biliary) or ERCP failure in patients with periampullary cancer. The reported case could be resolved by means of a combined technique with transpapillary drainage of the common bile duct and the placement of a metallic prostheses that is covered coaxially through the common duct with PMAL. This can therefore be considered as a valid rescue technique in these patients. On the other hand, it may be advisable to approach the extrahepatic bile duct in order to minimize the risk of sump syndrome, with the most distal/juxtapapillary access possible during EUS-guided choledocho-duodenostomy.

REFERENCES


Fig. 1. A. EUS with evidence of a neoplastic lesion in the pancreas head. B. Duodenal infiltration stenosis. C. Papilla duodenal stenosis. D. Endoscopic CDD with LAMS and a double pigtail and metal stent coaxial.
Fig. 2. A. Abdominal CT scan, with evidence of severe dilation of the intrahepatic bile duct and suspected hepatic and pericoledocyan abscess. B. Tomographic view of the CDD (red arrow shows LAMS). C. Fluoroscope with transpapillary biliary drainage by means of a metallic covered stent and coaxial double pigtail. D. Endoscopic view of metal stent via the endoscopic CDD.