Case Report

Long-term results from endoscopic choledochoduodenostomy closure using a cardiac septal occluder device in a patient with refractory sump syndrome

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Abstract

Sump syndrome can be managed effectively by biliary sphincterotomy and duct clearance. However, options for the management of sump syndrome in patients who have not responded to sphincterotomy are limited. We report a case of refractory sump syndrome managed with endoscopic closure of the choledochoduodenostomy using a cardiac septal occluder device.

Keywords: Cardiac septal occluder, Choledochoduodenostomy, Endoscopic retrograde cholangiopancreatography, Sump syndrome

Introduction

Choledochoduodenostomy (CDD) is occasionally performed in conjunction with bile duct exploration for severe or refractory choledocholithiasis.1,2 In some patients, food debris collects in the portion of the common bile duct that is distal to the CDD. This may cause pain and cholangitis and is termed “sump syndrome”. Usually sump syndrome can be managed effectively by biliary sphincterotomy and duct clearance.3 However, options for management of sump syndrome in patients who have not responded to sphincterotomy are limited. We report a case of refractory sump syndrome managed with endoscopic closure of the CDD using a cardiac septal occluder device.

Case report

An 82-year-old Caucasian female underwent open common bile duct exploration and CDD due to multiple large primary bile duct stones that manifested many years after cholecystectomy. Unfortunately, approximately 1 year after this surgery the patient began to complain of right upper quadrant pain and fever and was found to have abnormal liver biochemical tests. She then underwent a total of six endoscopic retrograde cholangiopancreatography (ERCPs) over the next 18 months with interventions including biliary sphincterotomy, subsequent sphincterotomy extension, large diameter papillary balloon dilation, and placement of multiple biliary stents. Despite these interventions, she continued to present repeatedly with clinical bacterial cholangitis, and cholangiography invariably demonstrated abundant organic solid material in the bile duct despite both the biliary orifice and CDD being widely patent, compatible with refractory sump syndrome. The patient was offered surgical correction (CDD closure and conversion to hepaticojejunostomy) but she declined.

Attempted endoscopic closure of the CDD using a cardiac septal occluder device was elected after thorough discussion with the patient and informed consent. At ERCP, the CDD was again observed (Fig. 1A), with soft yellow stone material emanating from it. The duodenoscope was passed to the second part of duodenum, where a previously placed biliary stent was removed and the bile duct was cleared of debris using a 15–18 mm extraction balloon (Fig. 1B). The duodenoscope was withdrawn to the duodenal bulb and a 9–12 mm biliary occlusion balloon was used to cannulate the CDD for the purpose of sizing the orifice. At 12 mm, the balloon met significant resistance to withdrawal across the CDD (Fig. 1C), but at 9 mm the balloon met only mild resistance. Based on this sizing, an Amplatzer septal occluder device with a 10 mm waist diameter was selected. A 450 cm × 0.89 mm super-stiff guidewire was passed via the CDD into the bile duct (Fig. 1D), and eventually across the papilla into the third portion of the duodenum. As the delivery sheath for the septal occlude device was too short to pass through the working channel of the duodenoscope, the scope was withdrawn over the guidewire entirely out of the patient. The 8F, 90 cm long delivery sheath was then passed over the wire using fluoroscopic guidance until it just traversed the CDD into the bile duct (Fig. 1E). Next, the duodenoscope was passed alongside the delivery sheath to the duodenal bulb. The septal occluder device was then placed.
advanced inside the delivery sheath. With the tip of the device positioned in the bile duct, the guide wire was removed. The internal 20 mm disk was then deployed in the bile duct. Traction was applied to the delivery system, tenting the bile duct toward the duodenum. With the biliary disk well seated, the 18 mm duodenal disk was deployed, with a 10 mm waist occupying the CDD orifice. The device appeared well positioned on endoscopy (Fig. 1F) and fluoroscopy (Fig. 1G).

The patient did well immediately postprocedurally. Four months after placement, she presented with signs and symptoms compatible with bacterial cholangitis. At follow-up ERCP, the septal occluder device was found to be properly positioned endoscopically (Fig. 1H) and fluoroscopically (Fig. 1I). A 15 mm occlusion balloon could be trolled past the biliary disk with minimal resistance in the patient’s diffusely dilated bile duct. Minimal sludge and no stones were swept from the biliary tree. The bile duct was explored using an ultraslim gastroscope. The biliary disk was properly seated and contained only a thin film of yellow stone material. The patient was treated with oral antibiotics and quickly recovered. Ursodeoxycholic acid 500 mg twice daily was prescribed, as was maintenance ciprofloxacin at a dose of 250 mg twice daily during the first week of each month, both with the intent of potential prophylaxis against further instances of cholangitis.

Over the ensuing 2.5 years, the patient largely did well from a clinical standpoint. She developed symptoms and signs suggestive of mild bacterial cholangitis approximately twice a year during this time. This was typically managed with a short course of intravenous antibiotics, with success. No further ERCPs were undertaken. The patient died from causes unrelated to her biliary issues (complications of cardiac disease) 3 years after the septal occluder device was used to close the CDD.

Discussion

Sump syndrome is occasionally not resolved by biliary sphincterotomy, leading to a management dilemma. In patients who refuse surgery or in whom surgery is contraindicated, endoscopic management may represent the best therapeutic option. Refractory sump syndrome must be distinguished from stenosis of the CDD, which may present similarly. To date, very few endoscopic treatment approaches have been reported for refractory sump syndrome. A single German case report described the successful use of the Amplatzer septal occluder to close a CDD. The Amplatzer septal occluder (AGA Medical Corporation, Golden Valley, MN, USA) is a proprietary device constructed of Nitinol wires shaped into two flat disks and a central waist (Fig. 2). The device has been used in humans since 1997 to repair cardiac atrial septal defects. It is available in a variety of disk and waist diameters, and is capable of closing defects up to 40 mm in diameter. Aside from its initial
purpose, the device has also been used for closure of tracheoesophageal and bronchopulmonary fistulae.\textsuperscript{5,6}

In our patient, the device appeared to successfully close the CDD. On follow-up ERCP there was only minimal sludge in her bile duct, whereas on all prior examinations the bile duct was noted to contain copious amounts of solid organic material. This technical success appears to have translated into partial clinical success. Unfortunately our patient developed mild clinical cholangitis approximately twice a year in the 3 years following CDD occlusion. However, this represented a marked decrease in the frequency and severity of episodes of cholangitis.

In conclusion, in cases of refractory sump syndrome, a cardiac septal occluder may be used to occlude the CDD, preventing further food entry into the bile duct. However, the device wires may be lithogenic, and the problem of infected bile remains. The role for ursodeoxycholic acid and prophylactic antibiotics to address these issues empirically remains uncertain and, as such, this procedure is best considered experimental at this time.

Conflict of interest

All contributing authors declare no conflicts of interest.

References