INTRODUCTION

Traditionally, therapeutic options for treating biliary cutaneous fistula (BCF) have included diversion of flow from the fistula with an external-internal biliary drainage and/or the deployment of biliary stents. The majority of reports describing obliteration with fibrin, acetic acid, ethanol or N-Butyl Cyanoacrylate (NBCA) injection involved isolated bile ducts without communication to the biliary tree (1-3).

Percutaneous injection of NBCA (4) was used to successfully treat a case of biliary leakage communicating with the main biliary tree. In the case reported, a persistent BCF detected after a biliary drainage catheter removal could not be resolved with diversionary techniques and Gelfoam administration in the fistulous tract. Using a cutaneous approach, an AMPLATZER Vascular Plug (AVP; AGA Medical Corp., Plymouth, MN, USA) was deployed in the fistulous tract with successful exclusion of the fistula.

AMPLATZER Vascular Plug is a device recently proposed for extra-cardiac procedures, such as arterial and venous embolization. In our experience and as reported in the early literature, the device is easy to use, versatile, and achieved a high technical success rate (5). AVP is composed of a self-expanding cylinder made of Nitinol, which is fixed by a microscopic screw, and an introduction wire made of stainless steel. The wire is to be turned anticlockwise in order to unscrew it from the cylinder, thereby allowing controlled release. Several studies on cardiologic applications reported a high success rate (97% successful deployment and 96% occlusion rate of atrial defect at 2 years) and a low complication rate (1.5%) (6). In literature, high versatility in the extra-cardiologic use of AVP systems is described, associated with high technical success (treatment of lung and renal artero-venous malformation, subclavian artery aneurysms, hypogastric, pulmonary, and cerebral aneurysms, occlusion of shunts in patients with portal hypertension during transjugular intrahepatic
portosystemic shunt, and occlusion of the hypogastric artery in patients with aortoiliac or iliac aneurysms) (5).

In literature, there is only a case report (7) in which an AVP was used to prevent a biliary leak; to the best of our knowledge, we report the first case of BCF embolized successfully with AVP.

AMPLATZER Vascular Plugs have been shown to be cost-effective alternatives to multiple coils (5). Gelfoam slurry and glue are comparatively cheap embolic agents, but they were thought to be unsuitable for use in this case, because it was a bile leak rather than bleeding (5, 7).

**CASE REPORT**

A 69-year-old female patient with painless obstructive jaundice was referred to the interventional radiology for decompression of biliary obstruction. The cause of obstruction was an intrahepatic tumor, a cholangiocarcinoma. A percutaneous puncture of a peripheral right hepatic duct branch was performed using a 22 G needle for cholangiography. Multisegmental bile duct obstruction was demonstrated, most likely due to the intrahepatic tumor.

Subsequently, a 18 G Ring Needle (DPLN-40-25-RING Drainage Access Catheter Needle Set, William Cook, Bjaeverskov, Denmark) was used for the puncture of the adequate right bile duct. With this access, the stricture was negotiated with a biliary manipulation catheter (William Cook) and an angled hydrophilic guidewire (Glidewire; Terumo, Tokyo, Japan); an internal-external drainage was deployed. After about one week, indices of biliary stasis were not improved in relation with the multisegmental bile duct obstruction; therefore another drainage was positioned. The cholangiography performed through the first drainage was used for the correct deployment of the second internal-external drainage. The drainage was maintained for 10 days during which the jaundice resolved and bilirubin levels normalized. The patient was submitted to a laparoscopic view that confirmed the impossibility of surgical removal of the tumor. Therefore, a biliary permanent stent was deployed [10 x 80 mm, (Gore-Viabil Endoprosthesis, USA)]. In that same procedure, a biliary cutaneous fistula (BCF) consequent to the first internal-external drainage removal was observed. Moreover, a biliary leak was observed. A conservative approach was used as first line treatment. An external drainage was maintained for the time necessary to ensure that the stent functioned and for the diversion of bile flow away from the BCF and biliary leak. Subsequent cholangiograms revealed the persistence of BCF and of the biliary leakage and the correct functioning of the stent. The biliary leakage was successfully embolized using coils (Fig. 1A).

Using the percutaneous approach, a fistulography was performed using a 7 Fr sheath (Fig. 1B). Through the same approach, a 7 mm AVP IV (AVP-AGA Medical Corp., Plymouth, MN, USA) was deployed. Immediate fistulography and cholangiogram revealed the complete exclusion of the fistula (Fig. 1C). External drainage was maintained for a few days to ensure the definitive embolization of the BCF.

Following the procedures, the patient remained well, with resolution of her jaundice and of the leakage and BCF.
DISCUSSION

Bile leakage and BCF are complications that can occur after a biliary injury (surgical, laparoscopic or percutaneous). Immediate surgical re-exploration in such patients, who are often critically ill, is difficult due to infection, edema, and scarring in the periportal area. Certainly, the diversion of flow away from the BCF or biliary leakage should be attempted before more invasive methods are attempted.

When the leakage or the fistula persists, the obliteration of the contributing bile duct may be considered. It can be performed using embolizing agents, such as fibrin, acetic acid, ethanol or NBCA injection (1), particularly when the injury involved isolated bile ducts without communication to the biliary tree.

Stent graft or coils can be used to embolize leakage or fistula communicating with the biliary tree.

Percutaneous injection of NBCA (4) was used by our group to successfully treat a case of biliary leakage communicating with the main biliary tree.

In particular, we would like to focalize attention to the treatment of BCF in communication with the biliary tree.

Vu et al. (2) described the successful treatment of a BCF with two sessions of NBCA instillation in two patients whose fistula had connections to the remainder of the biliary tree.

Eicher et al. (3) described a successful laser ablation of a bile duct in continuity with the remainder of the biliary tree, which contributed to a persistent BCF.

We report the successful use of the AVP within the liver to stop the cutaneous bile leak following percutaneous liver puncture preceding biliary drainage deployment.

The original AVP was designed as a permanent occluding flexible cylindrical device derived from the Amplatz Septal Occluder and the Amplatz Duct Occluder (5, 6). This specialized device is most useful for occluding medium and large vessels that would otherwise require multiple coils. All models of the AVP contain 2 basic components: a vascular plug and a delivery wire (5). All plugs are built with nitinol braids, which have a self-expanding feature. The plugs have radiopaque platinum marker bands at both ends that allow for high visibility under fluoroscopy. A stainless-steel screw on one of the platinum marker bands attaches to a delivery cable (5).

AMPLATZER Vascular Plug is commonly used for vascular embolization, in unusual cases too: congenital and not intrahepatic portosystemic venous shunt (7, 8), a ruptured pulmonary artery aneurysm in a patient with Behçet’s disease (9).

Recently, the use of AVP has been expanded to nonvascular disease with promising results. Endobronchial closure of a small postoperative bronchopulmonary fistula using the AVP was recently reported as a safe and effective treatment (10). AVP has also been used for the treatment of esophago-pleural fistulae (assisted with endoscopy) (11), ureteral occlusion for vesicovaginal fistula (12), and treatment of intraperitoneal bile leak (13).

To the best of our knowledge, the case presented is the first BCF treated with AVP. As this was a bile leak rather than bleeding, coils were thought to be inappropriate, as they rely on the promotion of thrombosis for their occlusive effect. Nondetachable coils were also thought to be difficult to deploy into the biliary system in a controlled fashion. We note a publication on the use of HydroCoils (MicroVention, Aliso Viejo, CA, USA) to successfully occlude a cystic duct stump leak (14). We opted to use an AVP in this case, for its mechanical occlusion effect. Further, as other colleagues prior postulated (13), the richly vascular liver would provide an adequate environment for clotting, and therefore occlusion, to occur. As the patient remained well and the follow-up CT scan demonstrated no evidence of leak, we were reassured that the device provided adequate occlusion.

Theoretically, using the AVP can lead to significant cost savings if a single device can achieve a complete occlusion (5). As Pellerin et al. (15) estimated, the cost of a single AVP (accounting for a mean cost of 485 euros) is cheaper than that of more microcoils (accounting for a mean cost of 250 euros for each).

We assume a novel successful use of AVP to occlude a BCF. More studies using a major number of clinical evidences are necessary to confirm this application of the AVP.

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