Interventional Management of Arc of Buhler Aneurysm

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ABSTRACT

The Arc of Buhler (AOB) represents a persistence of the ventral anastomosis between the superior mesenteric artery (SMA) and the celiac arterial systems. The exact incidence of the AOB is not known, but it is believed to be $\leq 4\%$. Aneurysms of this rare anomaly are even more uncommon. We report a case of an aneurysm of the AOB with an intact pancreaticoduodenal artery arcade (PDAA) and near occlusive celiac origin stenosis. Stenoses or occlusions of the celiac origin have been reported in association with AOB aneurysms, as well as in patients with PDAA aneurysms. Transcatheter embolization (TCE) was successfully performed, thereby excluding the AOB aneurysm while preserving flow through the PDAA. To our knowledge, this is the first report of successful percutaneous treatment of an AOB aneurysm. The pathophysiology and management AOB and PDAA aneurysms are reviewed. Review of the literature suggests that TCE, when feasible, is at least as effective as conventional surgery in patients with PDAA aneurysms, but with lower morbidity and mortality. Based on this data and our experience, we believe that TCE should be the initial treatment of choice in patients with PDAA or AOB aneurysms.

KEYWORDS: Visceral aneurysm, pancreaticoduodenal arcade, Arc of Buhler, pancreaticoduodenal artery

Objectives: Upon completion of this article, the reader should be familiar with the Arc of Buhler and understand the pathophysiology, natural history, and interventional and surgical treatment options for aneurysms of the Arc of Buhler and the pancreaticoduodenal artery arcade.

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The following case report details an unusual case of an aneurysm of the Arc of Buhler (AOB). The AOB is a ventral communication between the superior mesenteric artery (SMA) and the celiac arterial system.¹ The AOB is uncommon and has been described in both asymptomatic patients undergoing imaging for other reasons as well as in symptomatic patients with celiac axis (CAx) stenosis

related to atheroma or median arcuate ligament syndrome (MALS). Aneurysms of this anatomical variant would appear to be even rarer, with only a handful of cases described in the literature. Closely related anatomically to the AOB is the pancreaticoduodenal artery arcade (PDAA). The PDAA, consisting of both the superior (SPDA, anterior and posterior) and inferior

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(IPDA, anterior and posterior) pancreaticoduodenal arteries, provides the normal collateral flow pathway between the SMA and celiac arterial system in most patients. The interventional management of an AOB aneurysm is described and serves as a paradigm for discussion of the evaluation and management of these relatively uncommon but dangerous aneurysms.

CASE REPORT

A 54-year-old man was referred by his primary care physician for consultation regarding a visceral aneurysm.

Routine blood work performed 3 months earlier had revealed elevation of liver function tests (aspartate transaminase [AST] and alanine transaminase [ALT]). Serological testing eventually led to a diagnosis of hepatitis C. The patient subsequently had a screening ultrasound that revealed no hepatic lesions but suggested a mass near the pancreatic head. Follow-up computed tomography (CT) (Fig. 1A) and magnetic resonance (MR) angiography (Fig. 1B) of the abdomen documented a peripherally calcified $2.4 \times 2.5 \times 3.0$ cm aneurysm that appeared to arise from a branch of the IPDA (Fig. 1B). High-grade stenosis of the celiac origin was also noted (Fig. 1C). The

Figure 1 (A) Contrast-enhanced computed tomography shows aneurysm with peripheral calcification (arrow). (B) Magnetic resonance (MR) angiogram delineates aneurysm (arrow) in relation to adjacent pancreaticoduodenal artery arcade and shows communication with celiac arterial system near origin of the common hepatic artery.

Figure 1 (Continued) (C) Lateral MR angiogram image documents long segment high-grade stenosis (arrow) of origin of celiac artery with configuration suggestive of median arcuate ligament syndrome.

patient's past medical history was significant for non-insulin-dependent diabetes mellitus, recently controlled primarily by diet. His past surgical history was negative with the exception of a percutaneous liver biopsy (showing mild inflammation without fibrosis) performed after MR imaging to stage the level of cirrhosis. He reported no abdominal pain or weight loss and denied prior episodes of pancreatitis, intra-abdominal infection, or significant trauma. He also denied use of alcohol or drugs. After consultation with an interventional radiologist and a vascular surgeon, the patient was scheduled for arteriography and potential intervention.

Initial aortogram confirmed a 2.5-cm aneurysm arising from the SMA branch supplying the IPDA. The SMA was selectively catheterized with a 5F Simmons 2 catheter (Cook, Bloomington, IN). Selective arteriography (Fig. 2A) confirmed an intact PDAA (anterior and posterior IPDA, SPDA, and gastroduodenal artery [GDA]), as well as a separate branch from which the aneurysm arose and that distally communicated with the

CAx near the origin of the common hepatic artery (CHA). This branch was confirmed by lateral arteriography to be consistent with an AOB. Multiple oblique views confirmed that the aneurysm arose at the origin of the AOB and had a wide neck, making preservation of the AOB virtually impossible. It was believed that even a stent graft would not allow sufficient wall contact to allow preservation of the AOB collateral pathway. The long segment CAx origin stenosis suggestive of MALS was confirmed and deemed not favorable for percutaneous treatment. The decision was made to treat the aneurysm by transcatheter embolization (TCE) with hope of achieving distal control of the outflow and then packing the aneurysm with coils to exclude the aneurysm itself as well as its origin while maintaining patency of the PDAA to preserve hepatic and splenic blood flow.

The IPDA was selected with the Simmons 2 catheter. Over a guidewire, a 6F Pinnacle Guiding sheath (Terumo Medical, Somerset, NJ) was placed to the origin of the aneurysm. A 0.018 safety wire was left in the IPDA. Using a 120-cm vertebral catheter (Cook, Bloomington, IN), a guidewire was placed into the AOB just beyond the aneurysm. A 6F Envoy guiding catheter (Cordis, Miami Lakes, FL) was then placed into the aneurysm to improve stability for TCE. An Excelsior microcatheter and 0.014 Transcend wire (Target/Boston Scientific, Natick, MA) combination was placed through the 6F guiding catheter and used to select the outflow vessel of the aneurysm (Fig. 2B). Distal control of the aneurysm was first achieved using Trufill DCS detachable coils (Cordis, Miami Lakes, FL), ranging in diameter from 5 to 7 mm. A 10 mm × 30 mm DCS coil was then deployed as the microcatheter was retracted from the outflow vessel into the aneurysm itself. The aneurysm was then packed using DCS coils as well as 0.052 MWCE and Nester coils (Cook, Bloomington, IN). Once the aneurysm was densely packed with coils (Fig. 2C), a postembolization angiogram was performed, which demonstrated no residual flow within the aneurysm (Fig. 2D) and preserved flow through the PDAA branches and subsequently to the liver and spleen. The patient was monitored overnight and placed on clopidogrel and aspirin with the hope of maintaining patency of the PDAA. Follow-up CT performed 3 months later confirmed patency of the PDAA as well as exclusion of the aneurysm (Fig. 3).

DISCUSSION

The AOB was described by Buhler in 1904.¹ Tandler² reported that the SMA and CAx arise in prenatal life from the 10th and 13th segmental arteries, respectively, arising from the aorta. These segmental arteries are connected by a ventral anastomosis, which usually regresses. Failure of this regression leads to persistence of the ventral communication between the SMA and the

Figure 2 (A) Superior mesenteric artery (SMA) injection shows the aneurysm and intact pancreaticoduodenal artery arcade (PDAA), both arising from same SMA branch vessel. (B) Pre-embolization catheterization showing 0.018-inch guidewire in inferior pancreaticoduodenal artery, 6F sheath in SMA, 6F guiding catheter in aneurysm, and microcatheter in outflow segment of the aneurysm. (C) Postcoiling image documents dense packing of aneurysm and outflow channel. (D) Postcoiling arteriogram demonstrates exclusion of aneurysm with patent PDAA and filling of hepatic and splenic circulation.

CAX or one of its branches (common hepatic artery). The AOB, because of its relative scarcity, has received little attention in the literature since 1904.^{1,3-7} Its exact incidence remains unclear. Although it has been described in association with CAX stenosis, it would appear to be seen just as frequently in patients without CAX stenosis. Three series exist in the English literature describing the incidence of AOB as 1%⁷ and 4%³ in patients undergoing arteriography for mesenteric arterial stenoses (symptomatic) and 3.3%⁶ in individuals undergoing mapping arteriography for potential living related liver transplant donation (asymptomatic). The anomaly may be asymptomatic and without clinical impact. Knowledge of it becomes critically important when

contemplating pancreaticoduodenal surgery, hepatic surgery, hepatic embolization, or ablation in this region.⁸

Aneurysms of the AOB are indeed rare in the literature, with two cases described to date. Kugai⁴ described the treatment of a 3-cm saccular AOB in a patient with a celiac artery occlusion. Aneurysmectomy was performed without revascularization of the CAX after two attempts at TCE had failed. Myers⁵ described the surgical resection of an AOB aneurysm with SMA to splenic artery bypass using saphenous vein in a patient with neurofibromatosis type 1. The patient had near occlusive stenosis of the celiac origin, but reconstruction of the CAX was not performed. Both patients recovered from surgery uneventfully; however, little follow-up is provided in either of these case reports.

Figure 3 Computed tomography arteriogram performed 3 months post-TCE documents preserved patency of PDAA and hepatic and splenic blood supply with no filling of aneurysm. Note some metallic artifact from embolization coils.

A larger body of experience exists for PDAA aneurysms. Although uncommon ($\sim 2\%$ of all visceral artery aneurysms⁹), >100 cases are described in the literature.¹⁰ PDAA aneurysms seen in the setting of pancreatitis, intra-abdominal infection, or trauma are most often pseudoaneurysms, whereas true aneurysms are more typically associated with atherosclerosis and/or CAx stenosis. It has been postulated that the high flow state through the PDAA induced by CAx obstruction (stenosis, occlusion, or MALS) may predispose to aneurysm formation,¹¹ and spontaneous thrombosis of PDAA aneurysms after decompression of the CAx has been reported.¹² PDAA aneurysms have also been reported in association with vasculopathy such as fibromuscular dysplasia and Takayasu's arteritis.

PDAA may be discovered incidentally in asymptomatic patients undergoing imaging for unrelated reasons but appear in the literature to present more often with retroperitoneal, intraperitoneal, or gastrointestinal bleeding. Common presentations also include abdominal pain, vomiting, pulsatile mass, and gastric outlet obstruction. The propensity of PDAA aneurysms to rupture is significant, with $>60\%$ of cases in the literature presenting as rupture. Pseudoaneurysms seem to present more often with bleeding than true aneurysms. In contradistinction to aneurysms in other locations, there does not appear to be direct correlation between the size of the aneurysm and risk of rupture,¹³ making even small aneurysms a threat.

Early reports of treatment were primarily surgical. In patients presenting with rupture, surgical mortality

rates were unacceptably high ($\geq 40\%$ ^{10,14}). Over the past 15 years, TCE has been performed in an increasing number of patients with both ruptured and unruptured aneurysms. Although reports of failed embolizations as well as ruptures during attempts at TCE exist,¹⁵ these are the exceptions with successful exclusion of aneurysms achieved in 80 to 90% or more cases.¹⁰ Embolization has been performed using Gelfoam, glue, and coils. The complexity of catheterization and need to preserve collateral pathways has led to an increased use of microcoils and microcatheters, which can improve success and safety in this region. Three reviews on the management of PDAA aneurysms recommended TCE as the initial treatment of choice due to its high success rate and excellent safety.^{9,10,16} In patients with occlusive disease, especially where collateral flow cannot be preserved, consideration should be given to revascularization of CAx stenosis or occlusion (due to CAx atherosclerotic disease) using stents or surgery (for MALS) where appropriate.

CONCLUSION

In summary, PDAA and AOB aneurysms present formidable risks to patients and diagnostic and therapeutic challenges to physicians. Their propensity to rupture makes even small aneurysms dangerous, and treatment is generally warranted for all aneurysms in this location. Literature review suggests that TCE with coils, using microcatheters where necessary, is the optimal initial therapy for both ruptured and unruptured aneurysms. In cases where CAx obstruction exists, consideration of treatment by surgical or interventional means is reasonable but not mandatory for success.

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