Rupture Risk and Etiology of Visceral Artery Aneurysms and Pseudoaneurysms: A Single-Center Experience

Susanne Regus, MD and Werner Lang, MD

Abstract

Background: The aim of this study was to analyze differences in rupture risk and etiology of visceral artery aneurysms (VAAs) and visceral artery pseudoaneurysms (VAPAs) in a single-center experience. Materials and Methods: We retrospectively reviewed all patients with a VAA or VAPA after treatment by open surgical or endovascular repair (ER) in our institution. Patient history, treatment details, and outcome were recorded and analyzed. Results: From January 1996 to April 2014, 29 (12 women) patients with 33 aneurysms (26 VAAs and 7 VAPAs) were treated in elective and urgent settings by open repair or ER. Etiology was quite different, most common was atherosclerosis (61.5%) in VAA and chronic pancreatitis (85.7%) in VAPA. Rupture rate was 19.2% in VAA and 42.9% in VAPA, whereas mean size of ruptured VAA was 4.4 cm and of ruptured VAPA was 2 cm. Open repair (suture, ligation, and aneurysmectomy with or without arterial reconstruction) and ER (coil embolization in the packing technique) were performed in half of all cases. After follow-up (72-month VAA and 82-month VAPA), aneurysm-free survival was reported to be 95% in VAA and 100% in VAPA. Conclusion: Chronic pancreatitis seems to be a prominent risk factor for the development of VAPA in this single-center experience. Modern endovascular techniques with promising short- and long-term results could broaden indications to treat asymptomatic VAA and VAPA.

Keywords

visceral artery aneurysms, visceral artery pseudoaneurysm, rupture risk, chronic pancreatitis

Background

True visceral artery aneurysm (VAA) and visceral artery pseudoaneurysm (VAPA) are a rare and uncommon condition of vascular disease with an estimated incidence in autopsies and angiographic series between 0.1% and 2%.1 The increased use of duplex ultrasound scan (DUS) and cross-sectional body imaging for intra-abdominal pathologies has raised the prevalence and incidental identification of VAA and VAPA in the last 20 years.

Affected arteries are the splenic (60%), hepatic (20%), superior mesenteric (5%), celiac (4%), gastric and gastroepiploic (3%), intestinal (2%), pancreaticoduodenal (1%), and other (5%).2 Pathogenesis of true and false aneurysms are fundamentally different. The most commonly reported etiology of VAA is atherosclerosis, rarely fibromuscular dysplasia, or hereditary diseases as Marfan- or Ehlers-Danlos syndrome.3-5 In contrast, most VAPAs are caused by abdominal traumas or infection of adjacent organs, in particular chronic pancreatitis. Formation of VAPAs results from weakening of the arterial wall caused by partial enzymatic digestion.6 Furthermore, increased use of minimal invasive laparoscopic and percutaneous procedures has raised the incidence of iatrogenic arterial lesions.7 Regardless of etiology, rupture is a hazardous and life-threatening complication with a reported mortality rate between 21% and 100%.8

Visceral artery aneurysms have been historically managed with either expectantly with ultrasound scan monitoring at regular intervals or with open surgery. In the last 20 years, endovascular approaches offer an alternative to conventional open surgery with the benefit of low periprocedural morbidity and mortality.

The aim of this study was to analyze the etiology and rupture risk of 26 VAAs and 7 VAPAs after open (OR) or endovascular repair (ER) at a single center.

Materials and Methods

From January 1996 to April 2014, 29 patients with the diagnosis of 33 splanchic artery aneurysms (26 VAAs und 7 VAPAs) underwent surgical repair or ER at a single center.

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Demographic and clinical data of the 29 patients as well as special features of aneurysms were obtained retrospectively through review of physician and hospital records and collected in a computerized database. The analysis was performed separately for VAA and VAPA. The statistical evaluation was done using IBM SPSS Statistics 21 software.

Diagnostic imaging consisted of DUS, computed tomographic angiography (CTA), magnetic resonance angiography (MRA), or digital subtraction angiography.

Indications to treat were aneurysm size of >2 cm in asymptomatic patients, in fertile female, and symptomatic aneurysms without reference to size. Any degree of bleeding was considered as an indication for intervention. Due to high rupture rate without reference to size, each VAPA was treated by OR or ER.

Treatment modalities included both traditional open surgery and percutaneous endovascular techniques using coil embolization. Specifically, coils were packed into the efferent and subsequently the afferent artery, thus excluding the aneurysm. In cases of persistent blood flow, coils were packed into the aneurysm sac as well. There were no other endovascular (ie, stent placement or embolization using liquid agents) or laparoscopic techniques employed.

Primary technical success was defined as thrombosis of the aneurysm sac or successful occlusion at the completion of the procedure. Major complications were determined as bleeding with subsequent surgical revision, splenic infarction, pancreatic infarction, and mesenteric ischemia. Postoperative deaths during the in-hospital stay were statistically evaluated as perioperative mortality.

After discharge from hospital, follow-up of VAA included clinical examination and ultrasound scan after 6 months and once a year in the following period. Any abnormal findings lead to further imaging with CTA or MRA. All patients undergoing ER were imaged with MRA or CTA scan at least once 6 months after treatment.

Each death in the follow-up period was subdivided into aneurysm-related and unrelated event, from which the calculation of aneurysm-free and overall survival was made.

**Results**

**Visceral Artery Aneurysm**

Twenty-two patients with 26 VAAs were included in this study. Eleven (50%) patients were female (2 pregnant). More detailed information about patients’ demographic data, aneurysms etiology, and symptoms are listed in Table 1.

Three patients had multiple VAAs and 4 had multiple synchronous aortic, peripheral, or cerebral aneurysms. Mean diameter was 3 cm (range 1-7.5 cm). Ruptured VAA (rVAA) had a diameter of 4.4 cm, while those without rupture measured about 2.5 cm. The rupture rate depending on location of the aneurysms is listed in Table 2.

Of 26 VAAs, 25 were treated by open or endovascular surgery. The remaining 1 untreated VAA was a patient with multiple VAA. It was a small (1 cm) splenic artery (SA) and underwent surveillance ultrasound imaging twice yearly. Open repair was done in 13 (50%) cases without reconstruction by aneurysmectomy and ligation in 6 (24%) cases. Treatment data are listed in Table 3. End-organ resection was necessary in 5 (20%) cases, particularly in 4 cases as splenectomy and in 1 case as partial resection of small bowel.

Endovascular repair was performed in 12 (46.2%) aneurysms by coil embolization, most commonly via embolization of the parent artery as well as sac packing technique with detachable coils (Figure 1A and B). No subsequent end-organ resection was necessary in the ER cohort.

The preferred treatment of the 5 rVAAs was OR in 4 (80%) cases and ER in 1 (20%) case.
After OR, the primary success rate was 100%, whereas second treatment was necessary in 3 (25%) cases of ER. In the first case, embolization was successful only after the second attempt. In the second case, a ruptured aneurysm of SA was treated in emergency with coils to stop bleeding, after stabilization aneurysmectomy was performed by OR. In the third case, embolization of a SA was not possible due to tortuous course of the feeding artery, but afterward OR was done successfully by aneurysmectomy and end-to-end anastomosis.

After OR, 1 patient with a ruptured hepatic artery aneurysm died, while there was no death in the endovascular group. Thus the overall perioperative mortality rate was 3.8% (1 of 26), particularly 7.7% (1 of 13) for OR and 0% (0 of 13) for ER and conservative treatment. Perioperative mortality was 20% (1 of 5) for rVAA and 0% for VAA without rupture. Mean duration of follow-up was 71.7 months (range 5-186). During follow-up, 2 (9%) patients were lost and 1 (4.5%) aneurysm-related death occurred. Furthermore, 2 nonaneurysm-related deaths were recorded. One patient died because of liver cancer and the other sustained a lethal cerebral bleeding. In 60% (12 of 20) of the patients, post-operative surveillance imaging with MRA or CTA was chosen, while 40% (8 of 20) of the patients were investigated using DUS or clinical examination. Aneurysm-free survival was 95% (19 of 20) and total survival was 85% (17 of 20).

### Table 3. Intraoperative Data, Primary Success, Morbidity, and Mortality of VAA and VAPA.

<table>
<thead>
<tr>
<th>Location (n)</th>
<th>Treatment (n)</th>
<th>Primary Success (n)</th>
<th>Complications (n)</th>
<th>Mortality (n)</th>
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<tbody>
<tr>
<td>VAA (26/33)</td>
<td>Aneurysmectomy, splenectomy 18.8% (3/16)</td>
<td>86.7% (13/15)</td>
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<td>SA (16)</td>
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<td>Conservative 6.3% (1/16)</td>
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<td>HA (5)</td>
<td>Suture, tamponade 20% (1/5)</td>
<td>100% (5/5)</td>
<td>Death 20% (1/5)</td>
<td>20% (1/5)</td>
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<tr>
<td>PDA (3)</td>
<td>Vein interponat 80% (4/5)</td>
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<td>SMA (2)</td>
<td>Aneurysmectomy 100% (2/2)</td>
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<td>VAPA (7/33)</td>
<td>Aneurysmectomy 33.3% (1/3)</td>
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Abbreviations: VAA, visceral artery aneurysm; VAPA, visceral artery pseudoaneurysm; OR, open repair; ER, endovascular repair; SA, splenic artery; HA, hepatic artery; PDA, pancreaticoduodenal artery; SMA, superior mesenteric artery.

Visceral Artery Pseudoaneurysm

Six of the seven patients with a VAPA were male (85.7%). Mean age was 55.7 (range 40-67). All patients with ruptured aneurysms were male. Mean diameter was 3 cm (ranging from 2 to 7.2). Mean diameter of ruptured VAPA (rVAPA) was 2 cm, while those without rupture measured 3.5 cm.

Etiology of VAPAs was quite different from those of true aneurysms. Six (85.7%) had a history of chronic pancreatitis and 1 (14.3%) developed a VAPA after laparoscopic cholecystectomy 3 years ago. The etiology of chronic pancreatitis included alcohol in all cases, none of them were suffering from gallstones. Treatment details and outcome are listed in Table 3.

Rupture rate in the presence of chronic pancreatitis was 50%. The location was hepatic artery (HA) in 2 cases (male, 2 cm diameter and male 7.2 cm) and pancreaticoduodenal artery (PDA) in 1 case (male, 2 cm).

Follow-up consisted of clinical and ultrasound examinations. Mean duration of follow-up was 82.6 months (range 12-156); no patient was lost. One nonaneurysm-related death occurred 2 years after surgery due to malignant hematologic disease. Aneurysm-free survival was 100% (7 of 7), and total survival was 85.7% (6 of 7).

Discussion

The frequency of lethal outcome after aneurysm rupture of a splanchnic artery and the abdominal aortic aneurysm are comparable, but the world literature numbers only approximately 30 series involving more than 25 VAAs and VAPAs at a single institution.

Currently, the more widespread use of CTA and MRA has permitted an increasing number of early diagnosis, thus allowing for the treatment of a large number of asymptomatic lesions and reducing urgent treatment of ruptured aneurysms. Nevertheless, the increased use of laparoscopic, endoscopic, and
endovascular therapeutic and diagnostic procedures results in an augmented number of VAPA, especially of the hepatic and mesenteric artery.

In this study, statistical analysis was performed separately for VAA and VAPA due to major differences in pathogenesis and natural history.

The most common etiology of VAA reported in the literature is atherosclerosis in elderly and congenital syndromes such as Marfan or Klippel-Trenaunay in young patients. Visceral artery pseudoaneurysms are often caused by abdominal trauma or infection often related to chronic pancreatitis and iatrogenic lesions resulting from laparoscopic or percutaneous treatment. In the study presented, the previously reported fundamental differences in origin of true and false aneurysms can be confirmed: VAAs are mostly caused by atherosclerosis (61.5%) or high flow (15.4%) situation in the presence of a stenosis or occlusion of the celiac trunk or superior mesenteric artery. In contrast, VAPA was most commonly a result of chronic pancreatitis and not atherosclerosis.

Visceral artery aneurysm and VAPA present emergently in 22% with a mortality rate ranging from 20% to 100%. The rupture risk depends on several properties including location, size, etiology, and gender. While the prevalence of SA among VAA is approximately 50% to 70%, the risk of rupture is relatively low (2%-10%). In contrast, while the prevalence of HA (20%) and PDA (5%) is very low, the corresponding risk of rupture is reported to be as high as 20% to 80%.

Gender seems to be an important risk factor for rupture in our experience. Despite an equal distribution in incidence between the genders, 95.7% patients with rVAA and 100% with rVAPA were male in our study. However, this observation is limited by the small number of cases in our study and does not allow for any conclusion suggesting a low rupture risk in women.

Clear indications to treat VAAs include the presence of symptoms and/or evidence of rupture. Furthermore, asymptomatic patients with aneurysms greater than 2 cm or a rapid growth of more than 0.5 cm/year, as well as visceral aneurysms irrespective of size in a pregnant woman, those of childbearing age, or in patients undergoing an orthotopic liver transplantation. In addition, the well-known different rupture and mortality rate of various visceral aneurysms depending on location may guide treatment decisions. The most common VAA is SA (60%), followed by HA (20%), SMA (5%), and PDA (1%). Nevertheless, HA and PDA present the highest rupture rate with about 70% to 80% and should therefore be treated more aggressively than SA. Visceral artery pseudoaneurysms carry a high risk for rupture independent of size and should therefore be treated when identified.

Concomitantly, the number of minimally invasive techniques available to treat VAAs has expanded. Now that less invasive methods are available, the indications for intervention may have broadened. Historical size criteria for intervention are still referenced, but the overall trend has been toward earlier intervention instead of serial surveillance.

Traditional open surgery is considered to be the treatment of choice for VAA and VAPA. However, endovascular management has become a valid therapeutic option in recent years, especially in patients who are not suitable for OR.

Open surgery offers the advantage of durability, decreased necessity for follow-up examinations, and low mortality rates of <0.5% in elective repairs. The simplest surgical treatment technique is ligation of the feeding artery without arterial reconstruction. This approach can be used for elective and emergent treatment of aneurysms involving the splenic and superior mesenteric artery, the celiac trunk, and common hepatic artery with a low risk of hepatic ischemia, provided that ligation is performed proximal to a functional gastroduodenal artery. A risk of small intestine ischemia has been reported in up to 40% of patients.

In our study, end-organ resection was performed after aneurysm ligation in 71.4% (5 cases), in particular splenectomy in 4 cases, and segmental small intestine resection in 1 case.

Figure 1. A, Saccular asymptomatic aneurysm of the splenic artery in a 65-year-old male patient before endovascular repair. Arrow: saccular aneurysm sac; arrowhead: efferent splenic artery. B, After successful embolization of a splenic artery aneurysm, the aneurysm sac was thrombosed with no persistent flow. Arrow: aneurysm sac; arrowhead: coils in the feeding artery.
While splenectomy has historically been the most common surgical treatment in patients with SA aneurysms, several studies have highlighted the importance of splenic preservation to prevent subsequent infectious complications.\(^1,18,19\) In this study, no splenectomy was necessary after coil embolization, even though postprocedural asymmetric splenic infarction was diagnosed in 25% of cases. These results suggest that the need for routine splenectomy should be critically evaluated, particularly after arterial ligation without SA reconstruction.

Endovascular repair can also be done with or without preservation of the parent’s artery. Isolation techniques whereby the parent artery is sacrificed using coils, vascular plugs, or liquid embolic agents are relatively easy to perform, while preservation of the main artery by stent implantation or balloon-assisted thrombin injection of liquid agents represents more challenging procedures.\(^20\)

A common technique is to pack coils into the aneurysms sac, preserving the afferent and efferent arteries.\(^21\) This is best achieved in a saccular aneurysm with a narrow neck and so as to avoid occlusion of the parent artery.\(^22\) Alternatively, the parent artery can be embolized as well so long as there exists a robust collateral network. In this study, coil embolization was the exclusively chosen endovascular technique for 12 VAAs and 4 VAPAs (11 SAs, 4 PDAs, and 1 HAs). In all cases, the parent artery was sacrificed; subsequent end-organ infarction was documented in 25% \((n = 4)\) of these cases. After the endovascular procedure, no patient had abdominal pain or other symptoms reduced to splenic infarction, thus there was no need for splenectomy in any case. In 2 cases, necessity for OR was caused by ineffective occlusion and therefore reperfusion of the aneurysm, while in both cases we assume fusiform aneurysms with broad necks are the reason for technical failures. We hypothesize that these 2 technical failures are due to the short landing zone for coils in the presence of large and fusiform aneurysms. Our experience in endovascular treatment is limited to embolization with coils being packed into the efferent and afferent arteries, sometimes additionally into the sac, whereas we cannot demonstrate results after placement of different stents with different designs. In this study, 6 of the 19 SAs were located near the hilum, which was the reason for choosing OR, especially ligation of the feeding artery and aneurysm resection with adjunctive splenectomy. In those cases, we were unable to perform ER or open reconstruction. The remaining 13 SAs were located in the main trunk, while in 1 case OR and splenectomy were necessary due to rupture and hemorrhagic shock.

Despite our favoring coil embolization of both sac and parent artery, flow-diverting stents are a promising alternative in treatment of VAA, especially in fusiform aneurysms with reduced collateral flow. These stents are specially designed to reduce flow velocity in the aneurysm sac and therefore to promote thrombosis, while maintaining flow in the parent artery.\(^23\) Mid-term results after endovascular treatment of 5 VAAs by the Cardiatis multilayer stent (Cardiatis, Isnes, and Belgium) are not promising.\(^24\) After a 2-year follow-up period, success rate defined as stent patency and sac thrombosis was achieved in only 60% patients. Two stents became occluded, but there was no reperfusion of the aneurysm sac and patients were asymptomatic. Therefore, the final result was comparable with coil embolization and initial sacrifice of the parent artery, but stent placement is more challenging and much more cost expensive. Indeed, the validity of this study is limited due to the small number of cases.

In conclusion, we recommend OR in all patients with VAA or VAPA presenting with hemorrhagic shock as well as in those patients whose anatomy precludes an adequate landing zone for stent placement. Otherwise, we prefer ER utilizing coil embolization of parent artery and selective packing of aneurysm sac for every VAA and VAPA in the presence of sufficient collateral flow.

Importantly, ER carries the risk for reperfusion of the aneurysm sac during follow-up, which means that regular imaging is necessary.\(^25,26\) CT angiography represents a potential disadvantage for patients having renal failure or contrast medium allergy, furthermore, and it is contraindicated in women of childbearing age.

Contrast-enhanced magnetic resonance angiography (CEMRA) may offer an alternative follow-up imaging technique after treatment with coil embolization as it is also a noninvasive, and safe technique that can evaluate the state of aneurysms.\(^27\)

**Conclusion**

In contrast to VAA, there seems to be no correlation between aneurysm size and associated rupture rate in VAPA. Thus, all VAPAs should undergo prompt repair via either ER or OR. In particular, chronic pancreatitis carries a serious risk for rupture in cases of VAPA. Those patients are often not suitable for open surgery due to chronic alcohol abuse and poor general condition. The promising results of endovascular treatment in elective settings could lead to broader indications, thus reducing life-threatening rupture.

**Declaration of Conflicting Interests**

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