

# Management of visceral artery aneurysms

## *Conduta terapêutica em aneurismas de artérias viscerais*

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### Abstract

**Objective:** Visceral artery aneurysms, despite being uncommon, are important vascular diseases, since they frequently are life-threatening, and often fatal emergencies. The purpose of this study is to review our experience with treatments of visceral artery aneurysms.

**Method:** Between 1988 and June, 2004, 37 visceral artery aneurysms were treated in 35 patients (17 male and 18 female) with average age of  $56 \pm 14$  years. The most common locations were the splenic artery (18), the hepatic artery (10) and the superior mesenteric artery (four). 22 patients were asymptomatic, 13 patients were symptomatic. Emergency surgery was performed on three patients, elective open surgery on 29 patients, and endovascular treatment on seven patients.

**Results:** Perioperative mortality rate was 3.1% in the surgical group. The perioperative morbidity rate was 5.7%: one case of respiratory distress and one case of biliary fistula were manifested in the immediate postoperative period. None of the patients died in the endovascular group; perioperative morbidity rate was 14.3% (one case of hepatic artery thrombosis after failure of gastroduodenal artery aneurysm embolization). Failure of the procedure was 42.9% (three cases of aneurysm recanalization).

**Conclusion:** We believe that an aggressive surgical approach is justified even in case of asymptomatic visceral artery aneurysms, due to the low morbidity/mortality rates. Endovascular treatment should be reserved to selected cases.

**Key words:** splanchnic circulation, splenic artery, hepatic artery, superior mesenteric artery, celiac artery, aneurysm, surgery, endovascular.

Visceral artery aneurysms (VAA) are uncommon but important since they frequently present as life-threatening, often fatal, emergencies.<sup>1</sup> More than 3,000

### Resumo

**Objetivo:** Os aneurismas da artéria visceral, apesar de incomuns, são doenças vasculares importantes, já que muitas vezes oferecem risco de vida e são frequentemente fatais em emergências. O objetivo deste estudo é rever nossa experiência em tratamentos dos aneurismas da artéria visceral.

**Método:** Entre 1988 e junho de 2004, 37 aneurismas da artéria visceral foram tratados em 35 pacientes (17 homens e 18 mulheres), com idade média de  $56 \pm 14$  anos. As localizações mais comuns foram a artéria esplênica (18), a artéria hepática (10) e a artéria mesentérica superior (quatro). Vinte e dois pacientes eram assintomáticos, e 13 sintomáticos. Realizou-se cirurgia de emergência em três pacientes, cirurgia eletiva aberta em 29 pacientes e tratamento endovascular em sete pacientes.

**Resultados:** A taxa de mortalidade perioperatória foi de 3,1% no grupo cirúrgico. A taxa de morbidade perioperatória foi de 5,7%: um caso de dificuldade de respiração e um de fistula biliar ocorreram no período imediato ao pós-operatório. Nenhum dos pacientes morreu no grupo endovascular; a taxa de morbidade perioperatória foi de 14,3% (um caso de trombose da artéria hepática após falha na embolização do aneurisma da artéria gastroduodenal). A falha no procedimento foi de 42,9% (três casos de recanalização do aneurisma).

**Conclusão:** Acreditamos que uma abordagem cirúrgica agressiva justifica-se no caso de aneurismas assintomáticos da artéria visceral, devido às baixas taxas de morbidade/mortalidade. O tratamento endovascular deve ser reservado para casos selecionados.

**Palavras-chave:** circulação esplâncnica, artéria esplênica, artéria hepática, artéria mesentérica superior, artéria celiaca, aneurisma, cirurgia, endovascular.

cases have been reported in the literature. Most cases are small series or case reports;<sup>2</sup> few series reported more than 30 cases.<sup>3</sup> Common affected vessels include splenic, hepatic, superior mesenteric and celiac artery.<sup>3</sup> Gastric, gastroepiploic, intestinal (jejunal, ileal, colic), pancreaticoduodenal, pancreatic, gastroduodenal and inferior mesenteric arteries are less often involved.<sup>4-7</sup> The real incidence is unknown since most of them are asymptomatic. In the past, several VAA were diagnosed in rupture. Nowadays, there has been an increasing

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diagnosis of VAA due to the availability of advanced diagnostic imaging techniques. The broader use of sophisticated ultrasound, computed tomography (CT), magnetic resonance imaging (MRI), and contrast angiography allow the diagnosis of several asymptomatic lesions. However, visceral aneurysm rupture is often discovered only at autopsy. This emphasizes the need for a more aggressive treatment approach. Shanley et al. report the high mortality related to rupture in a review of the English literature from 1985 to 1995. The mortality rate ranged from 21% for hepatic artery aneurysms (HAA) to 100% for celiac artery aneurysms (CA).<sup>3</sup>

Therapeutic procedures can be performed either surgically or by endovascular treatment. The high incidence of aneurysm reperfusion at the follow-up of the endovascular procedure reported by some authors,<sup>8</sup> which we confirmed in a previous paper,<sup>9</sup> prompts us to frequently consider the surgical alternative.

The purpose of this study is to review our experience with surgical and endovascular VAA treatment.

## Material and methods

Between 1988 and June, 2002, 37 VAA were treated in 35 patients with an average age of  $56 \pm 14$  years. 17 patients were males and 18 were females. There were no pregnant women.

We treated 18 splenic artery aneurysms (SAA), 10 HAA, four superior mesenteric artery aneurysms (SMA), two pancreaticoduodenal artery aneurysms (PDA), one CA aneurysm, and one gastroduodenal artery aneurysm (GDA). Two patients had multiple VAA: one SAA

associated to SMA aneurysm, and one HAA associated to CA aneurysm. Other associated aneurysms involved the thoracic aorta (one case), the abdominal aorta (three cases) and the lower extremities (one case).

20 cases were atherosclerotic. Three cases were associated to fibromuscular dysplasia, two cases to blunt trauma, one case to pancreatitis, one case to duodenal ulcer, one case to infection, two cases to iatrogenic causes (abdominal surgery). In other cases, no obvious etiology was found for the development of the VAA. 16 patients were asymptomatic. The indication of treatment was given in case of aneurysm with diameter  $\geq 2$  cm. 13 patients reported abdominal or flank pain. These symptoms were associated to abdominal hemorrhage (two cases), gastrointestinal hemorrhage (two cases), and hemobilia (one case). In four patients an abdominal mass was present (Table 1). Diagnostic techniques that allowed to discover VAA are reported in Table 2. The techniques employed were always complemented with CT or MR and angiography (Figure 1). The mean maximum diameter of the VAA observed in our series was  $3.6 \text{ cm} \pm 2.3$ .

## Results

29 patients underwent elective surgery: 15 for SAA, eight for HAA, three for SMA aneurysms, two for PDA aneurysms, and one for CA aneurysm. The treatments are summarized in Table 3.

Three patients underwent emergency surgery. The first patient suffered a car accident. He had a blunt liver trauma with hemobilia and hemoperitoneum. Urgent laparotomy with pseudoaneurysm excision and hepatic

**Table 1** - Clinical presentation of the visceral artery aneurysms

Location	Cases	Mean age $\pm$ SD	M:F	Pain	Mass	Abdominal hemorrhage	Gastrointestinal hemorrhage	Hemobilia
Splenic	18	$56.6 \pm 8.2$	5:9	7	–	–	–	–
Hepatic	10	$59.2 \pm 20.9$	5:2	2	2	1	–	1
Superior mesenteric	4	$50 \pm 24.8$	4:0	1	1	–	1	–
Pancreaticoduodenal	2	$57 \pm 2.8$	0:2	2	1	1	–	–
Celiac	1	61.5	0:1	–	–	–	–	–
Gastroduodenal	1	40	1:0	1	–	1	1	–

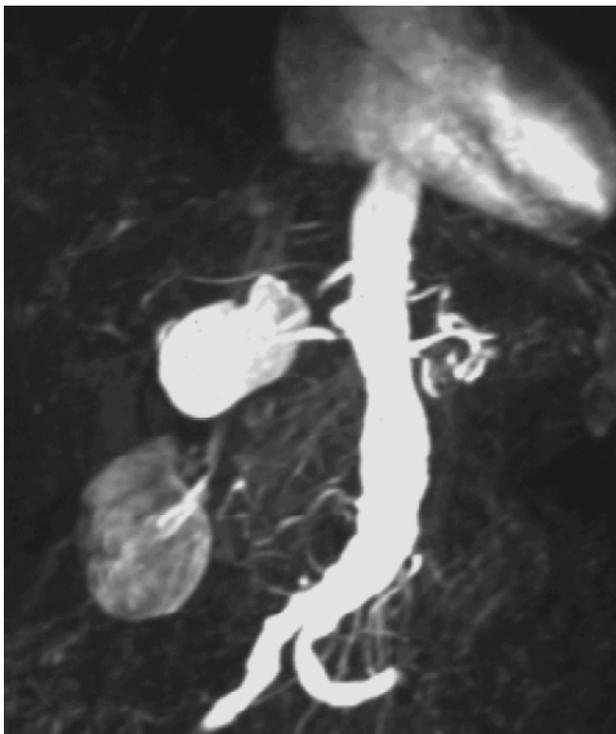
SD = standard deviation; M:F = male:female.

artery endoaneurysmorrhaphy were performed; no ischemic alteration was noticed intraoperatively during the clamping period. The intraparenchymal hepatic

**Table 2** - Diagnostic techniques used to discover visceral artery aneurysms and diameters of the aneurysms

Type of aneurysm	Diagnostic technique (n)	Diameter (cm) ± SD
Splenic	CT (9)	2.9 ± 0.8
	Echography (5)	
	X-ray (2)	
	Angiography (1) MRI (1)	
Hepatic	Echography (6)	6.3 ± 3.6
	CT (3)	
	Angiography (1)	
Superior mesenteric	Echography (4)	3.4 ± 1.7
Celiac	Echography (1)	2 ± 0
Pancreaticoduodenal	Echography (1)	3.3 ± 1.8
	CT (1)	
Gastroduodenal	Echography (1)	6

SD = standard deviation; CT = computed tomography; MRI = magnetic resonance imaging.



**Figure 1** - NMR angiography shows hepatic artery aneurysm

cavity with biliary tract damage was treated by mean jejunal loop anastomosis. The second patient was affected by chronic pancreatitis. He had developed a SMA pseudoaneurysm (previously treated with endovascular embolization) ruptured into a pancreatic cyst. He reported abdominal pain and hematemesis; surgical treatment was the pseudoaneurysm excision and the endoaneurysmorrhaphy. The last patient developed a GDA pseudoaneurysm that caused an intraabdominal hemorrhage after surgery for a retroperitoneal leiomyosarcoma. It was treated with transcatheter embolization but the procedure was not successful and it was complicated by hepatic artery thrombosis. An emergency laparotomy with thrombectomy and hepatic vein graft interposition, associated to gastroduodenal aneurysmectomy with artery ligation was performed (Figure 2). In the postoperative period the patient developed a bilious fistula treated with an endobiliary prosthesis during endoscopic retrograde cholangiopancreatography (Table 4).



**Figure 2** - Aneurysmectomy of hepatic artery aneurysm and expanded polytetrafluoroethylene prosthetic graft interposition

The perioperative mortality rate was 3.1% in the surgical group: only one patient died on the fourth postoperative day due to massive intestinal infarct after associated surgery for abdominal aortic aneurysms (AAA) and SMA aneurysm, despite SMA reconstruction. In one case, respiratory distress was manifested in the immediate postoperatively period, requiring prolonged

mechanical respiratory assistance, and one case of bilious fistula. No further complications were observed after surgery (morbidity rate: 5.7%). In the five cases of heterotopic splenic autotransplantation, technetium-labeled autologous red cells scintigraphy was performed after 6 months, showing that all autotransplantation was viable. The follow-up ( $72 \pm 51$  months) was obtained with duplex ultrasounds and/or with CT (Figure 3) in the surgical group. Six patients were lost at follow-up because they were referred from distant regions. In 25 patients no complications related to surgery and no recurrence of aneurysm were observed.

Transcatheter embolization was performed in seven patients with coils via femoral artery in all cases. The procedure was successful in three patients with saccular SAA (2, 2.2, 4 cm of diameter) and in a patient with saccular HAA (2 cm of diameter). The other three cases were treated surgically after failure of endovascular embolization. The first patient with saccular HAA (6 cm of diameter) repeated the endovascular treatment, with coils and angiographic guidewire after one year due to aneurysm enlargement (8 cm of diameter). At this time the patient was offered surgical treatment, which he refused; after three years of follow-up the

**Table 3 - Elective surgical treatment**

Involved artery	Patients	Surgery
Splenic (15 patients)	5	Splenectomy with splenic autotransplantation to omentum
	3	Aneurysmectomy with direct arterial reconstruction
	4	Splenectomy
	3	Aneurysmectomy with arterial ligation
Hepatic (eight patients)	3	Aneurysmectomy, interposition prosthetic graft (ePTFE)
	4	Aneurysmectomy, direct arterial reconstruction
	1	Ligation with excision of aneurysm (common HAA)
Superior mesenteric (three patients)	1	Aneurysmectomy, interposition vein graft
	1	Aneurysmectomy, interposition prosthetic graft (ePTFE)
	1	AAA resection and grafting
Pancreaticoduodenal (two patients)	1	Aneurysmectomy, interposition prosthetic graft (Dacron)
	1	AAA resection and grafting
	1	Aneurysmectomy, PDA - GDA anastomosis
Celiac	1	Pseudoaneurysm excision, arterial ligation
	1	Aneurysmectomy, prosthetic graft (ePTFE) interposition
<b>Total</b>	<b>29</b>	

ePTFE = expanded polytetrafluoroethylene; HAA = hepatic artery aneurysm; AAA = abdominal artery aneurysm; PDA = pancreaticoduodenal artery; GDA = gastroduodenal artery.

aneurysm enlarged to more than 11 cm and the patient underwent surgical excision of the aneurysm. The second patient with GDA iatrogenic pseudoaneurysms (6 cm of diameter) developed thrombosis of the hepatic artery during the embolization and consequently required emergent surgical treatment (Figure 4). The last patient had an unsuccessful SMA pseudoaneurysm embolization (5.5 cm of diameter). Urgent surgical treatment was performed due to gastrointestinal hemorrhage (Table 3).



**Figure 3** - Computed tomography scan shows reperfusion of embolized hepatic artery aneurysm



**Figure 4** - Aneurysmectomy: incomplete thrombus formation induced by percutaneous embolization of coils and angiographic guidewires

None of the patients treated with coil embolization died. The perioperative morbidity rate was 14.3% (one case of hepatic artery thrombosis after failure of GDA aneurysm embolization). Failure of the procedure was 42.9% (three cases of aneurysm recanalization). In four patients the endovascular treatment was successful; no aneurysm reperfusion or enlargement were observed by computerized tomography during the annual follow-up ( $81 \pm 18$  months).

**Discussion**

VAA are rare but insidious, due to their high incidence of rupture and life-threatening hemorrhage. The most common abdominal visceral vessel affected by aneurysmal disease was believed to be the splenic artery.<sup>5</sup> However, the hepatic artery has recently become almost the most frequently affected vessel. Shanley et al.<sup>3</sup> reported an increase of incidence of hepatic pseudoaneurysms caused by the higher number of endoscopic procedures worldwide. As reported in the literature, SAA are associated to many conditions. The most common are pregnancy and portal hypertension.<sup>6,7</sup> However, in our experience we did not observe this correlation. SAA are frequently asymptomatic: aneurysmal rupture is the most dramatic clinical presentation with a reported mortality rate of 36%.<sup>10</sup> Pregnancy is associated to 20-50% of all ruptures.<sup>6,7</sup> Rupture frequently develops into severe intraperitoneal hemorrhage.<sup>11</sup> Gastrointestinal bleeding is occasionally reported.<sup>12</sup> Open surgery seems to be justified for all

**Table 4** - Urgent surgical treatment

Involved artery	Patients	Surgery
Hepatic	1	Pseudoaneurysm excision, endoaneurysmorrhaphy, hepato-jejunostomy
Superior mesenteric	1	Pseudoaneurysm excision, endoaneurysmorrhaphy
Gastroduodenal	1	Aneurysmectomy, gastroduodenal artery ligation, hepatic artery thrombectomy and interposition vein graft
<b>Total</b>	<b>3</b>	

symptomatic SAA and in pregnant patients or in women of childbearing age due to the high maternal and fetal mortality related to aneurysmal rupture during pregnancy.<sup>6,13</sup> Elective surgery is more controversial: it is generally recommended for low risk patients with SAA greater than 2 cm in diameter. This criterion was also adopted in our service. Aneurysms of the proximal vessel may be treated by aneurysmectomy and end-to-end anastomosis or simple ligation/exclusion without arterial reconstruction; splenectomy must be performed when the aneurysms are localized at the splenic hilus. When possible, we prefer arterial reconstruction to simple ligation: if splenectomy is necessary, we generally use heterotopic splenic autotransplantation into the omentum in order to prevent the risk of overwhelming postsplenectomy infection which is related to high mortality.<sup>14</sup> Splenic autotransplantation was performed in five cases and in all of them the splenic function was confirmed by scintigraphy at 6 months. The patients were carefully followed clinically and no serious infection occurred during the follow-up period even without any prolonged antibiotic therapy.

HAA are traditionally considered the second most common VAA: they amount to about 20% of all VAA.<sup>15</sup> The reported incidence has been increasing over the past two decades with false aneurysms accounting for up to 50% of HAA.<sup>3</sup> This is the result of an increasing number of invasive biliary tract procedures as well as an increasing use of diagnostic CT imaging after blunt trauma. Most HAA remain asymptomatic. Rupture of HAA may occur into the intestinal or hepatobiliary tract and into the peritoneal cavity. Hemobilia<sup>16</sup> often occurs in case of traumatic intrahepatic pseudoaneurysms, as we observed in one case. Intraoperative bleeding is frequently caused by extrahepatic aneurysmal rupture.<sup>17</sup> Mortality rate is high, not less than 35%.<sup>18</sup> Only one case of post-traumatic HAA rupture was observed. The other patients were asymptomatic at presentation. Common HAA may often be treated with aneurysmectomy or aneurysmal exclusion without arterial reconstruction: nevertheless, restoration of arterial hepatic blood flow is important in the aneurysms involving the common hepatic artery and its extrahepatic branches. In our experience, we performed two bypasses: one to treat an aneurysm involving the right and left hepatic artery, another to treat a proper HAA.

Aneurysms of the SMA account for 5.5% of VAA.<sup>5</sup> These lesions are frequently infectious (mycotic) in

etiology.<sup>19</sup> In our series (four cases), one was mycotic. SMA aneurysms are frequently symptomatic at presentation, with intermittent upper abdominal pain.<sup>4</sup> Symptoms may be caused by thrombosis or embolization of the SMA. In our series, two patients complained of abdominal pain and one had gastrointestinal hemorrhage; the others were asymptomatic. Surgical treatment with aneurysmorrhaphy and simple ligation are the most used techniques.<sup>20,21</sup> An efficient collateral flow allows this approach to be usually successful. Arterial reconstruction with graft interposition or aortomesenteric bypass with aneurysm exclusion or excision are rarely reported.<sup>22,23</sup> We used the arterial reconstruction technique to treat two cases of SMA aneurysms associated with AAA. Unfortunately, one of the patients died of massive intestinal infarct due to thrombosis of the graft.

Aneurysms of the CA are unusual lesions that account for 4% of all visceral aneurysms.<sup>5</sup> CA aneurysms are frequently asymptomatic,<sup>24</sup> rupture is often associated to intraperitoneal hemorrhage, although communication with the gastrointestinal tract can occur. Shanley et al. found that mortality rates for rupture of aneurysms of the CA is up to 100%.<sup>3</sup> Aneurysmectomy and primary reanastomosis of the CA trunk are sometimes possible. If not possible, an aortoceliac synthetic prosthesis bypass (as we performed in the only case observed in our series) or autogenous vein bypass can be performed. CA ligation rarely results in hepatic necrosis, but it should not be performed in case of intraoperative hepatic ischemia.<sup>25</sup> Surgical treatment of aneurysms originating from the celiac axis, the superior mesenteric artery, or their branches may be carried out more safely after the control of the aorta: this may be performed by rotating the viscera medially after incising the left lateral peritoneum through the phrenicocolic and lienorenal ligaments, as clearly described by Murray et al.<sup>26</sup>

GA, PDA and pancreatic artery (PA) aneurysms are rare. Pancreatitis<sup>27</sup> and increasing blood flow through these vessels, such as in case of CA occlusion, are common etiological factors. Most patients are symptomatic at the time of diagnosis, presenting epigastric pain, frequently with radiation to the back. All our cases were symptomatic and one PDA was associated to CA occlusion; we did not have cases of pancreatitis as etiological cause. Aneurysmal rupture is manifested as gastrointestinal hemorrhage through stomach, duodenum, and less often through biliary or

pancreatic ductal system in about 75% of these cases.<sup>28</sup> When aneurysms are associated to pancreatic pseudocysts, cyst decompression should be undertaken; major resections of pancreatic tissue, including pancreaticoduodenectomy, may be necessary.<sup>29</sup>

Considering the natural history of the VAA and the risk of rupture, there is general agreement in the literature to also treat these lesions when they are asymptomatic. An aggressive surgical approach is justified when it is associated to low morbidity/mortality rates.

Percutaneous transcatheter coil embolization (PTCE) techniques have been used with increasing frequency in the treatment of VAA. Technical success of PTCE has been acceptable in most series: Carr *et al.*<sup>8</sup> report a primary success rate of 66.7%, Salam *et al.*<sup>30</sup> of 81%, Kasirajan *et al.*<sup>31</sup> of 75%, Gabelmann *et al.*<sup>32</sup> of 92%. However, disappointment has been expressed in preventing the pressure transmission to the aneurysmal wall,<sup>33,34</sup> which may be present even in successfully embolized aneurysm. Onohara *et al.*<sup>35</sup> reported a case of rupture into the stomach of embolized CA pseudoaneurysms. Salam *et al.*<sup>30</sup> observed two patients with evidence of aneurysms reperfusion at follow-up, Carr *et al.*<sup>8</sup> reported two patients with rupture at follow-up. Intraprocedural aneurysm rupture has also been reported.<sup>30</sup> Abscess and sepsis are potential complications in case of mycotic aneurysm even if any cases have been reported. Another complication of PTCE is arterial thrombosis/embolization that may result in organ infarction and subsequent infection.<sup>8</sup> Kasirajan *et al.*<sup>31</sup> reported favorable aneurysm morphology for PTCE treatment: saccular aneurysms with a narrow neck, aneurysms with adequate collateral flow and aneurysms of vessel that are not the only source of blood to the organ end. Other percutaneous techniques, such as permanent liquid embolic material<sup>5,36</sup> and covered stent,<sup>30,37</sup> have been recently proposed, but the results must be verified.

In our experience only few cases of VAA have been treated with PTCE, due to the high incidence of aneurysmatic sac reperfusion and relatively high morbidity correlated to the procedure. Three of the seven cases described needed surgical treatment after unsuccessful percutaneous embolization even if we consider that one case was treated with empirical techniques. In one case, an important complication related to embolization was manifested: hepatic artery thrombosis that required emergent surgical revascularization.

In the surgery group, aneurysmal recurrence was not observed at follow-up. One death due to associated treatment of AAA and SMA and two minor complications were observed in the perioperative period, with a mortality rate of 3.1% and morbidity rate of 5.7%. The low aneurysmal recurrence, mortality and morbidity rates related to surgery of VAA were confirmed in the literature by many authors.<sup>8,38-40</sup>

The worst results we obtained with endovascular treatment prompt us to reserve this technique only to selected cases: favorable aneurysm morphology and difficult aneurysm surgical access. We have observed a major incidence of aneurysm recanalization in large aneurysms, even if the small sample does not permit a statistical analysis.

## Conclusion

The recent increased use of sophisticated diagnostic techniques has allowed the discovery of an increasing amount of asymptomatic VAA. We believe that an aggressive surgical approach is justified even in case of asymptomatic VAA, due to the low morbidity/mortality rates related to this treatment. Endovascular techniques should be reserved to selected cases.

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