Visceral Artery Aneurysms

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ABSTRACT

Objective. Visceral artery aneurysms are rare vascular malformations and the literature lacks satisfactory general information about the pathology. The aim of this study was to review our experiences in the diagnosis and treatment of visceral artery aneurysms.

Materials and Methods. We retrospectively reviewed data on 10 patients who were diagnosed with visceral artery aneurysms at our institution between June 2002 and September 2005. All available clinical, pathologic, and postoperative data were reviewed and analyzed for postoperative outcome.

Results. Four splenic artery aneurysms, 2 hepatic artery aneurysms, 5 renal artery aneurysms, 1 superior mesenteric artery aneurysm, and 1 inferior mesenteric artery aneurysm (13 total visceral artery aneurysms) were diagnosed in 10 patients. All the patients were treated except 1 patient with bilateral renal artery aneurysms. One patient required emergent surgical treatment due to splenic artery aneurysm rupture. Only 1 patient underwent endovascular treatment (ie, coil embolization for a superior mesenteric artery aneurysm); otherwise all the patients were treated surgically on an elective basis. Surgical treatment modalities included ligation with exclusion in 4 patients (2 splenic artery aneurysms, 1 renal artery aneurysm, 1 hepatic artery aneurysm) and resection with revascularization in 4 patients (1 splenic artery aneurysm, 2 renal artery aneurysms, 1 hepatic artery aneurysm, 1 inferior mesenteric artery aneurysm). Histopathologic examination of the vascular materials revealed major atherosclerotic changes except one that showed inflammatory vasculitic changes. One patient required bleeding revision, and mortality did not occur in any of the patients.

Conclusions. Visceral artery aneurysms are rare and potentially life-threatening vascular disorders. The number of

cases diagnosed every year increases because of advanced radiologic diagnostic methods and screening programs. Careful consideration and early management of these malformations can be life saving.

INTRODUCTION

Visceral artery aneurysms are rare vascular pathologies with an incidence detected in autopsy series ranging between 0.1% to 0.2% among all vascular aneurysms [Drescher 2006]. The natural history and clinical course of the pathology are uncertain, and the indications for surgical management remain controversial. Patients with visceral artery aneurysms are mostly asymptomatic; however, the aneurysms can lead to life-threatening complications if rupture ensues. In recent decades, more cases have been diagnosed because of the refinements in radiological diagnostic methods as well as the increasing number of patients undergoing abdominal imaging, such as ultrasonography, computed tomography, or even angiography.

The aim of this study was to assess our 3 years of experience with visceral artery aneurysms and review our data in respect to the signs and symptoms, diagnostic work up, histopathological findings, treatment methods, outcome and follow-up of patients with different visceral artery aneurysms at our institution.

MATERIALS AND METHODS

The study was approved by the ethical committee of the institution and informed consent was obtained from each patient. We retrospectively reviewed the data on 10 patients with visceral artery aneurysms who were treated at Bezm-I Alem Vakif Gureba Hospital, Cardiovascular Surgery Service, over a 3-year period from June 2002 to September 2005. The study was approved by the ethical committee of the institution according to the regulations of the hospital on manuscript preparation.

Thirteen visceral artery aneurysms were diagnosed in 10 patients. There were 4 aneurysms of the splenic artery, 5 aneurysms of the renal arteries, 2 aneurysms of the hepatic artery, 1 aneurysm of the inferior mesenteric, and 1 superior mesenteric artery. Three of the patients were female and 7 were male. Mean age was 41.5 years (range, 31-55 years). All the patients except the one who presented with signs of intra-

Demographic Data of the Patients

Patient No.	Age, y	Sex	Type of Artery Aneurysm	Aneurysm Size, cm	Risk Factor	Treatment
1	55	M	Bilateral renal	2.1 and 2.2	Trauma (renal angioplasty)	Follow-up
2	42	F	Unilateral renal	3.1	Atherosclerosis	Ligation + nephrectomy
3	48	M	Bilateral renal	2.8 and 2.9	Atherosclerosis	Resection and revascularization
4*	31	M	Splenic	Ruptured	Behcet's Syndrome	Ligation and splenectomy
5	35	M	Splenic	2.5	Atherosclerosis	Ligation and splenectomy
6	34	F	Splenic	3.6	Atherosclerosis	Ligation and splenectomy
7	37	F	Splenic	2.6	Atherosclerosis	Ligation and revascularization
8	46	M	Hepatic	3.6	Atherosclerosis	Ligation and revascularization
9	42	M	Hepatic	2.4	Atherosclerosis + trauma (cholecystectomy)	Ligation and lobectomy
4*	32	M	Superior mesenteric	5.9	Behcet's Syndrome	Coil embolization
10	45	M	Inferior mesenteric	3.6	Atherosclerosis	Resection and revascularization

*Patient number 4 presented twice with visceral artery aneurysms; one was a ruptured splenic artery aneurysm and the other was a superior mesenteric artery aneurysm.

abdominal bleeding were diagnosed incidentally during investigation of various intra-abdominal vascular or nonvascular pathologies either by computed tomography, Doppler ultrasonography, or angiography. In all elective patients, clinical assessment, electrocardiogram, and biochemical analysis were performed as an initial diagnostic work-up except for the patient who presented with intra-abdominal bleeding. The definitive diagnosis was established by Doppler ultrasonography, contrasted or uncontrasted computed tomography, magnetic resonance imaging, and conventional angiography.

RESULTS

All demographic data, patient characteristics, preoperative findings, comorbidity factors, and treatment modalities of the patients are summarized in the Table.

Renal Artery Aneurysms

Renal artery aneurysms were diagnosed in 3 patients; 2 of the patients were male and 1 was female. Aneurysms included both of the renal arteries in 2 patients; the female patient had a unilateral aneurysm. Hypertension was the main presenting symptom (mean systolic, 155 ± 15 mmHg; range, 145-180 mmHg; mean diastolic, 105 ± 10 mmHg; range, 95-115 mmHg), and patients were diagnosed during the investigation of the etiology of hypertension. Blood urine nitrogen (mean, 31.2 ± 4.3 mg/dL; range, 28-34 mg/dL; normal, 8-22 mg/dL), blood creatinine (mean, 1.6 ± 0.7 mg/dL; range, 1.5-1.8 mg/dL; normal, 0.7-1.4 mg/dL), and 24-hour urine creatinine clearance values were slightly elevated in all the patients.

One of the male patients previously had bilateral renal artery

stenoses, which were treated by angioplasty. In the follow-up of that patient, aneurysm formation was observed on the renal arteries very close to the renal paranchyma. Sizes of the aneurysms on the left and right renal arteries were 2.1 cm and 2.2 cm, respectively (Figure 1). Because both of the aneurysms were in the vicinity of the renal tissues, follow-up of the patient and medical control of hypertension was chosen for treatment.

Renal ultrasonography of the female patient revealed atrophy of the left kidney together with aneurysm formation on

Figure 1. Bilateral renal artery aneurysms. Sizes of the aneurysms on the left and right renal arteries were 2.1 cm and 2.2 cm, respectively.
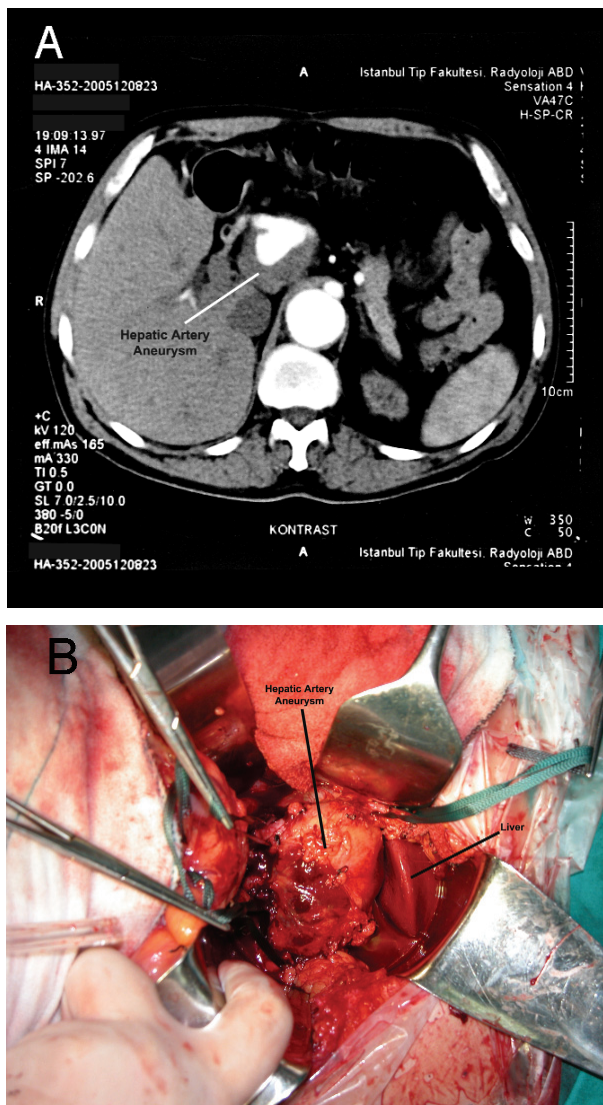
Figure 2. A, Contrasted computed tomography view of the hepatic artery aneurysm. B, Perioperative photography of the hepatic artery aneurysm with a size of 3.6 cm.

the respective renal artery. Computed tomography angiography confirmed the diagnosis and showed the aneurysm at the distal one third of the left renal artery, with a diameter of 3.1 cm and renal atrophy. Renal scintigraphy showed significant decrease of the functions (5% of the normal values) of the left kidney and relatively preserved functions of the right one. Ligation of the left renal artery and nephrectomy was the choice of treatment and were performed through a standard median laparotomy approach.

The diameters of the renal artery aneurysms of the third patient were 2.9 cm on the left and 2.8 cm on the right side. Location of the aneurysms was the proximal one third to middle portion of the renal arteries. Saphenous vein graft interposition was the choice of treatment for the aneurysms on both sides.

The operations and postoperative courses were uneventful and success of the operations were confirmed by renal

Doppler ultrasonography performed before the discharge of the patients. Hypertension was cured in 1 and improved in the other 2 treated patients and persisted in 1 patient.

Splenic Artery Aneurysms

Splenic artery aneurysms were observed in 4 patients; 2 of the patients were male and 2 were female. All were diagnosed incidentally except 1 patient who presented with signs and symptoms of intra-abdominal bleeding and was operated on immediately. Exploratory laparotomy revealed a ruptured splenic artery aneurysm. Ligation of the splenic artery and splenectomy were performed. Physical examination and questioning the patient about his medical history suggested Behcet's disease. Histopathologic examination of the vascular material showed inflammatory changes including thickening of both media and intima together with monocyte and plasmocyte infiltration in the intima and adventitia levels of the artery and only minor atherosclerotic changes. The patient demonstrated the classic triad of Behcet's disease (oral aphtous ulcers, genital lesions, and uveitis), had a positive pathergy test, and his condition was diagnosed as Behcet's disease. Immunosuppresives and high doses of prednisolone were started immediately. Postoperative magnetic resonance angiography was conducted to detect any other vascular abnormalities; however, the results were negative for aneurysms.

The remaining patients presented with symptoms of gastrointestinal discomfort. None of the female patients were multiparous. Mean diameter of the aneurysms was 2.9 cm (range, 2.5-3.6 cm); 2 were diagnosed by contrasted computed tomography and 1 by magnetic resonance. As splenectomy could be the treatment option during the surgery, all the patients were vaccinated 1 week prior to the surgery. Choice of surgical treatment was ligation and splenectomy in 1 male and 1 female patient, and saphenous vein graft interposition in 1 female patient. Histopathologic examination of the excised material revealed atherosclerotic changes in these patients. Postoperative courses were uneventful and Doppler ultrasonography confirmed the patency of the saphenous graft.

Hepatic Artery Aneurysms

There were 2 patients with hepatic artery aneurysms. Both were male and presented with symptoms of abdominal pain. Patients were diagnosed incidentally during investigation of any possible abdominal aortic pathologies; one by angiography and the other by computed tomography angiography. During clinical investigation of the patients, rheumatologic or vasculitic pathology test results were negative. Diameters of the aneurysms were 3.6 cm (Figure 2) and 2.4 cm. One of the patients had previously undergone an open cholecystectomy operation. The aneurysm in this patient was saccular and confined to the intrahepatic structures. It was treated with ligation of the aneurysmatic artery and left lobectomy. The postoperative course of the patient was complicated by bleeding, and re-exploration was required for homeostasis even though no active bleeding site could be found; otherwise no extra complications occurred. The aneurysm in the second patient was fusiform and treated with resection and saphenous vein graft interposition (Figure 3). Histopathologic
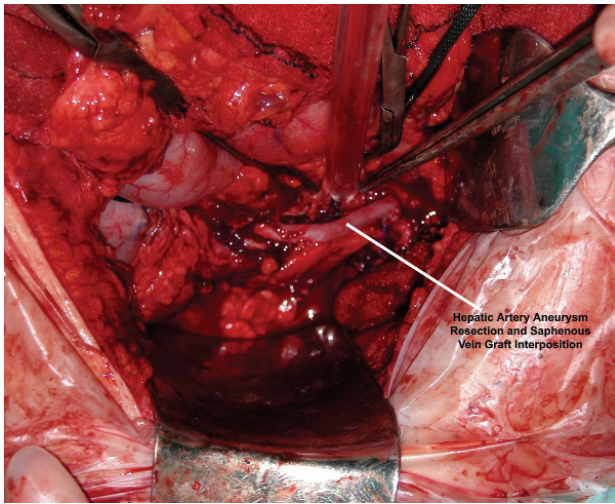
Figure 3. The hepatic artery aneurysm was treated with resection and saphenous vein graft interposition.

examination of the vascular specimens derived from the patients showed atherosclerotic changes; however, prior open cholocystectomy operation in one of the patients also indicated a mixed etiology of both trauma and atherosclerosis, but there was no infectious basis. Control Doppler ultrasonography before discharge indicated a patent graft.

Superior and Inferior Mesenteric Artery Aneurysms

The patient with a superior mesenteric artery aneurysm had been previously treated for a ruptured splenic artery aneurysm and diagnosed with Behcet's disease. Following discharge, the patient's compliance to an immunosuppressive regimen for vasculitis had been poor and he presented with complaints of abdominal pain. Thoracoabdominal magnetic resonance angiography was performed to rule out any life-threatening vascular malformations, and a saccular aneurysm with a diameter of 5.9 cm at the superior mesenteric artery was diagnosed. Immunosuppressive agents were started immediately, and as soon as remission was obtained coil embolization was performed in a private center for the treatment of saccular aneurysm. The procedure was uneventful and the patient was discharged from the hospital with remission-dose immunosuppressive agents.

The presenting symptoms of the patient with inferior mesenteric artery aneurysm were gastrointestinal disturbances, changes in defecation habits, and abdominal pain. Doppler ultrasonography was not remarkable except for aneurysmatic changes on the abdominal aorta at the origin of the inferior mesenteric artery. The pathology was further analyzed with computed tomography angiography and an aneurysm of the inferior mesenteric artery with a diameter of 3.6 cm was diagnosed at the proximal one third portion of the artery. The abdominal aorta showed tortuosities and multiple atherosclerotic plaque formations, but no aneurysmatic dilatations. Resection and saphenous vein graft interposition was performed. The postoperative course was uneventful and the patient was discharged from the hospital on the sixth postoperative day.

All the patients in our group tolerated treatment safely. One patient required re-exploration for bleeding control. On a routine basis, postoperative abdominal Doppler ultrasonography was carried out in all of the patients to examine vascular, neovascular, and nonvascular intra-abdominal structures. All the grafts used for vascular reconstruction were found to be patent in the early postoperative period prior to discharge with no further intra-abdominal pathologies related to surgery. All the patients were followed-up with a range of 1 to 4 years at 6-month intervals and control Doppler ultrasonography results were normal in all patients with patent grafts and no other significant abnormalities.

DISCUSSION

Visceral artery aneurysms are very rare vascular malformations with an incidence of 0.01% to 0.2% detected during a routine autopsy series [Larson 2002]. Since they are rare, the natural history and clinical course of the pathology are not certain and indications for management are controversial. On the other hand, the number of patients diagnosed is increasing because of increasing use of available intra-abdominal imaging modalities. These aneurysms can be saccular or fusiform as well as true or false in nature. They are generally asymptomatic but can lead to life-threatening complications, such as rupture. In general, nearly 20% of these cases present with a clinical emergency and nearly 10% of these emergent cases result in death [Zelenock 2000]. In contrast, there is not a single accepted consensus in the literature about the treatment of these pathologies, and the literature also lacks guidelines for treatment and follow-up of these patients. These aneurysms can occur, in decreasing order of frequency, on the splenic artery, hepatic artery, renal arteries, superior and inferior mesenteric arteries, celiac artery, gastric and gastroepiploic arteries, jejunal and ileal arteries, and gastroduodenal, pancreaticoduodenal, and pancreatic arteries. Among the visceral artery aneurysms, those on the inferior mesenteric artery, celiac artery, gastric and gastroepiploic artery, jejunal and ileal artery, and gastroduodenal, pancreaticoduodenal, and pancreatic artery are seen with an incidence <5%.

Splenic artery aneurysms are the most common of the visceral artery aneurysms, accounting for 60% of cases. The reported incidence is not certain and ranges between 0.16% to 10.4% in different series. It is usually a disease of elderly and young parous women. Women are 4 times more prone to the disease than men. The main etiologic factors that account for the pathology are multiparity because of the changes in the arterial wall due to the alterations in the hormonal local hemodynamic status during pregnancy, portal hypertension with splenomegaly, inflammatory vasculitidies, fibrodysplasia, hypertension, and atherosclerosis [Pescarus 2005]. Also, these aneurysms can be seen in association with other arterial aneurysms; when splenic artery aneurysms are present, the possibility of other aneurysms must be investigated. Viseral artery aneurysms are usually asymptomatic; however, they can lead to nonspecific gastrointestinal symptoms such as epigastric pain, flatulence, or pain radiating to the left subscapular region. The calculated mean diameter for splenic artery aneurysms is 2.1 to

2.2 cm in the larger series; cases larger than 3 cm in diameter are rare and accepted as large in the literature [Pescarus 2005]. The aneurysms can be fusiform or saccular, usually single. They can be located at any portion of the artery, but are commonly at the distal zones close to the splenic paranchyme. The main complication of these aneurysms is rupture, with a 2% to 10% risk that has a 20% to 30% mortality rate [Pescarus 2005; Hernandez-Lahoz 2006]. Even though treatment indications are unclear, surgery is indicated for symptomatic patients, patients with enlarging aneurysms, pregnant patients, patients anticipating pregnancy, patients with portal hypertension presenting for liver transplantation, and patients with aneurysms larger than 2 cm in diameter [Pescarus 2005]. Surgical treatment can be performed by conventional open or laparoscopic means and can include endovascular measures, such as endovascular grafting or coil embolization. Treatment of the aneurysms at the proximal segments of the artery are the least complicated and easiest to perform; however, aneurysms located in the middle third and distal segments are rather difficult due to the proximity to the pancreatic and splenic tissues. Although the ideal treatment is resection of the aneurysm and reconstruction of the artery, in complicated cases proximal ligation of the splenic artery with or without splenectomy should not be discounted. The important issue about splenectomy is appropriate preoperative immunization for all elective patients with a splenic artery aneurysm. Open surgery is the most frequently applied treatment method; however, with the refinements in radiologic and minimally invasive methods, these other options are commonly applied today [Moyer 2005]. In our series, 4 patients, 2 male and 2 female, were treated for splenic artery aneurysms with a mean aneurysm size of 2.9 cm; however, one was a large aneurysm with a size of 3.6 cm. One patient presented with a ruptured aneurysm and the aneurysm size could not be measured in this patient. At the same time, this was the only patient in the series with a different etiology, inflammatory vasculitic, rather than atherosclerosis. Resection and revascularization could be performed in only 1 of the patients, while splenectomy was the chosen procedure for the remaining patients.

Hepatic artery aneurysms are the second most common of the splanchnic tree, with an incidence of 20%. Men are 2 times more commonly affected than women. From 1960 to 1970, the major etiology was infection from a sequela of bacterial endocarditis; however, infections now account for 10% of hepatic artery aneurysms etiology, particularly in intravenous drug abusers. Other conditions associated with a hepatic artery aneurysm are mainly atherosclerosis, fibrodysplasia, vasculitidies, inflammatory changes around the artery, trauma, and long-term oral amphetamine use [Zelenock 2000; Abbas 2003; Stambo 2004], hence its presentation in a wide range of patient age groups. Hepatic artery aneurysms are usually solitary and can be confined to the intra- or extrahepatic structures. They can be saccular or fusiform in nature; smaller ones are of fusiform character more frequently. Hepatic artery aneurysms, in contrast to the other visceral artery aneurysms, have a higher risk of rupture, with a 14% to 80% rate and a 21% to 43% risk of mortality [Abbas 2003]. Rupture more commonly occurs in inflammatory than in atherosclerotic aneurysms. Most

aneurysms remain asymptomatic; gastrointestinal discomfort and epigastric and right upper quadrant pain are frequent symptoms. A characteristic triad of jaundice, hematobilia, and pain is seen in cases of aneurysmal rupture in the bile ducts. Treatment of a hepatic artery aneurysm is necessary because of the high risk of rupture. Treatment options include conventional open surgical resection with or without resection of the hepatic tissues or minimally invasive and endovascular measures. Intrahepatic multiple aneurysms usually require ligation of the branch hepatic arteries and resection of some parts of the hepatic structures; however, extrahepatic aneurysms can be treated with reconstruction of the artery as well as other, less invasive methods. Two patients in our series were treated for hepatic artery aneurysms, with one of the aneurysms being saccular and the other fusiform. Both were male patients in their 40s and the aneurysms were mainly due to atherosclerosis; no infectious origin could be identified. One of the aneurysms was relatively large, with a diameter of 3.6 cm. One patient was treated with resection of hepatic tissue; however, the other patient was treated with saphenous vein graft interposition.

Renal artery aneurysms are one of the most frequent aneurysms of the abdominal cavity, with an estimated incidence of 0.01% to 0.09% in an autopsy series and 0.6% to 1% found by arteriograms [Calligaro 2000; Pfeiffer 2003]. They are usually solitary,but occur bilaterally in about 10% of cases. The frequency in men and women is accepted to be equal. They can be saccular or fusiform in nature. Saccular aneurysms are usually located at the renal artery bifurcations and fusiform ones occur secondary to stenosis as a poststenotic dilatation on the renal artery itself. Mostly they are located at the extraparancymal regions of the artery. The main etiologic factors accounting for these aneurysms are atherosclerosis, fibromuscular dysplasia, and, with the increasing number of radiologic investigations and endovascular treatment methods, catheter-induced trauma [English 2004]. These aneurysms are generally asymptomatic; however, most of the patients are hypertensive. Flank pain or hematuria can be other symptoms. Compared hepatic artery aneurysms, the rupture risk of renal artery aneurysms is relatively low, with 2.8% to 5.6% rates, and there is no correlation between the size of the aneurysm and the risk of rupture for renal artery aneurysms [Pfeiffer 2003]. However, the pathology can be complicated, with hypertension, distal embolization, and loss of functional renal tissue and hematuria, which can all be cured or improved by the treatment of the aneurysm itself. Treatment, although unclear, is recommended in cases with a risk of rupture, hypertension, embolization leading to renal infarction, hematuria, or aneurysms larger than 1.5 cm [Reiher 2000; English 2004]. However, the risk and benefit to the patient must be weighed carefully because the surgery itself may lead to the loss of the kidney completely. The best surgical treatment option is resection of the aneurysm and reconstruction of the artery, but endovascular measures are also employed with high success rates for the treatment of renal artery aneurysms. There were 3 patients and 5 aneurysms of the renal arteries in our series with a mean diameter of 2.62 cm, all being fusiform in shape. One patient had a trauma-induced aneurysm, and the other 2 patients had artherosclerotic aneurysms leading to hypertension and derangements of

renal functions. Procedures were performed on 2 patients and 1 patient was followed medically. In the treated patients, hypertension and renal functions improved.

Superior mesenteric artery aneurysms with an incidence of 5.5% to 8% among splanchnic aneurysms and 0.8% in autopsy series are the third most common of the visceral artery aneurysms. Infection is the most frequent cause and, in decreasing order, atherosclerosis, arterial dysplasia, collagen vascular disorders, arteritis, or trauma also play an important role in the etiology. Usually it is older patients who are affected, and the aneurysms occur with similar frequency in men and women [Zelenock 2000; Dorigo 2004]. They can be saccular or fusiform in nature. They are often asymptomatic, but patients may present with abdominal pain and signs and symptoms of ruptured aneurysm because superior mesenteric artery aneurysms have a higher risk of rupture, as do hepatic artery aneurysms, compared with other visceral artery aneurysms. The rupture risk of superior mesenteric artery aneurysms ranges between 38% and 50% with a rupture mortality rate of 40% to 60%. Since these aneurysms have such high rupture and mortality rates, elective surgery remains the mainstay of treatment, with a mortality rate of <15% [Zelenock 2000; Liao 2006]. The aim of treatment is the preservation of organ perfusion, and therapy can be performed either by classic surgical methods, removal of the aneurysm and arterial reconstruction, or by means of endovascular measures such as coil embolization or grafting. Depending on the etiology, patients should receive medical therapy and close clinical follow-up. Our patient with a superior mesenteric artery aneurysm was a young man and had a large saccular aneurysm. The etiology in our case was Behcet's disease, and he was treated with coil embolization successfully before rupture ensued. Before and after the treatment, high doses of immunosuppressives were administered, and the patient was discharged with remission-dose immunosuppressive agents.

Aneurysms of the inferior mesenteric artery are rare, with an incidence of less than 1% among the visceral artery aneurysms and there are only sporadic cases reported in the literature [Saliou 1999; Zelenock 2000]. The etiology is usually atherosclerosis, but inflammatory changes and vasculitic disorders have also been documented [Saliou 1997; Zelenock 2000; Morimoto 2003]. They are mostly fusiform rather than saccular in character and are confined to the proximal segments of the vessel. Patients are usually asymptomatic and the aneurysm is diagnosed during radiologic work-up for other possible vascular abnormalities. Treatment of these aneurysms is usually resection and reimplantation, but also, rarely, vein graft interposition. There was a patient with inferior mesenteric artery aneurysm in our series with an aneurysm size of 3.6 cm who was treated with resection of the aneurysm with saphenous vein graft interposition.

Aneurysms of the celiac, gastric and gastroepiploic, jejunal and ileal, gastroduodenal, pancreoticoduodenal and pancreatic arteries belong to the group of splanchnic aneurysms. They are rare, with an incidence <5% [Zelenock 2000] and share common features with the other splanchnic aneurysms.

In conclusion, aneurysms of the visceral arteries are diagnosed more frequently today because increasing numbers of patients undergo abdominal imaging and because of refinements in radiological diagnostic methods. These aneurysms carry a considerable risk of rupture and consequent mortality. The etiology of these aneurysms is usually of an atherosclerotic and vasculitic nature, rather than mycotic, as they were in the past, due to the use of antibiotics and a decrease in the incidence of endocarditis. Open conventional surgical methods account for the major treatment modality, but endovascular treatment procedures are used frequently and with high success rates.

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