Hepatic aneurysms are a rare sequela of vascular abnormalities in the liver, including trauma, infection, necrotizing vasculitis such as polyarteritis nodosa (PAN), and iatrogenic and arterial mediolysis. Presentation with intra-abdominal hemorrhage is associated with a high mortality rate. We describe life-saving transcatheter coil embolization of multiple isolated ruptured hepatic pseudoaneurysms in a patient with no history or clinical findings of PAN. We present angiographic findings and intra-arterial transcatheter embolization techniques in the treatment of ruptured large hepatic artery aneurysms. Endovascular specialists should recognize that PAN could present with classic angiographic findings and, in some cases, as life-threatening ruptured isolated hepatic artery aneurysms as its first presentation. (J Vasc Surg 2004;39:1122-4.)

CASE PRESENTATION

A 64-year-old woman with a past medical history of uncontrolled hypertension on four antihypertensive medications presented emergently to the hospital with 24-hour history of epigastric and midabdominal pain, with a blood pressure of 180/100. The patient underwent a non-contrast computed tomography scan of the abdomen and pelvis, which demonstrated high-density fluid surrounding the liver and spleen. The patient was brought directly to the operating room, with high suspicion for a perforated viscus. Surgical exploration was performed and demonstrated hemoperitoneum with hematoma in the hepatoduodenal ligament. There was no perforated viscus and the visceral organs were grossly normal. An intraoperative ultrasound demonstrated grossly normal hepatic, splenic, and gastroduodenal arteries. No source of active bleeding was identified during surgical exploration.

An emergent mesenteric arteriogram was obtained immediately following the exploratory laparotomy to define the source of the hemorrhage. An abdominal aortogram was performed with a 5F Pigtail catheter (Angiodynamics, Queensbury, NY). Selective cannulation of the superior mesenteric artery (SMA) and common hepatic arteries was performed with a 5F Cobra glide catheter (Terumo/Boston Scientific, Natick, Mass), which demonstrated two large hepatic artery aneurysms, measuring 2 × 1.5 cm and 1.5 × 1.5 cm, arising from the left hepatic artery (Fig 1). There was active extravasation of contrast from the most proximal left hepatic artery aneurysm consistent with acute hemorrhage. A second equal-sized aneurysm located distal to the first was also identified. A replaced right hepatic artery arising from the SMA demonstrated multiple smaller aneurysms of variable size with associated multifocal stenoses and ectasia within the distribution of the vessel (Fig 2). No aneurysms arising from the replaced right hepatic artery were actively extravasating at this time. The remaining visceral vessels demonstrated no aneurysms or vascular irregularities.

A 5F Cobra glide catheter was advanced into the left hepatic artery. A subselective microcatheter (Target Therapeutics/Boston Scientific, Fremont, Calif) was advanced into the neck of the proximal hepatic artery aneurysm. Brisk extravasation of contrast was identified from this aneurysm (Fig 3). Multiple helical coils (Target Therapeutics/Boston Scientific) were then packed within the neck of the proximal hepatic artery aneurysm. Immediate occlusion of the ruptured aneurysm was documented, with no further active extravasation or visualization of the aneurysm. The adjacent hepatic artery aneurysm was then evaluated, and although it was not actively extravasating at this moment, its appearance was similar to the previously embolized aneurysm. Therefore, the neck of this aneurysm was also embolized with helical coils (Target Therapeutics/Boston Scientific), with no further visualization of this aneurysm (Fig 4). A total of six 2 × 10-mm microcoils (Target Therapeutics/Boston Scientific) were deployed during this procedure. Following embolization, there was no further evidence of
bleeding. A rheumatology consult was called in light of the multiple hepatic artery aneurysms and irregularity of the hepatic artery vascularity. An elevated C4 complement 74 (16–47mg/dL), C reactive protein 13.5 (0.0 – 0.9), and erythrocyte sedimentation rate 30 (0-20) were documented on laboratory values, which helped reinforce our diagnosis of PAN. The patient was started on high-dose prednisone and cytoxan.

The patient was discharged from the hospital on postoperative day number 6 in good condition. She has done well clinically, with routine visits with her rheumatologist as an outpatient for over two years and no further evidence of bleeding or other clinical sequelae of PAN. During her acute presentation and over the last two years, no liver or muscle biopsies have been performed to evaluate for PAN activity. Also, there has been no further radiologic imaging.
for evaluation of aneurysm regression because her clinical condition has been excellent. Currently, her blood pressure is well controlled and is on a maintenance low-dose steroid.

DISCUSSION

There is a wide spectrum of clinical presentations of PAN. Our case provides another example of the various clinical and radiographic challenges in the management of PAN. The variety in symptomatology can wax and wane from acute life-threatening abdominal hemorrhage secondary to ruptured isolated hepatic aneurysms to an indolent subclinical vasculitis. Furthermore, these isolated hepatic aneurysms are extremely rare in the setting of PAN. Approximately seven cases have been described in the past 40 years. PAN manifests as small aneurysms, vascular ectasia, and vascular occlusion of small and medium sized vessels. Recently, Ryan et al described segmental arterial mediolysis of the hepatic artery that is radiographically similar to PAN without evidence of a vasculitic process. In their case, multiple aneurysms were identified without clinical and rheumatologic findings of PAN. Our case demonstrates similar radiographic findings; however, rheumatologic and radiographic findings helped confirm the diagnosis of PAN.

Although a tissue biopsy of the involved symptomatic nerve or muscle is ideal (65% sensitive), angiography is diagnostic in some classic cases. The acute activity of the disease is associated with the formation of aneurysms. Hypertension can be found in 75% of patients with renal involvement. The risk of aneurysm rupture increases once hypertension develops. As in our case, the risk of aneurysm rupture increased significantly with the patient’s uncontrolled hypertension; clinically, these patients do worse in light of the acute nature of ruptured aneurysms. Furthermore, we embolized the second nonruptured large hepatic aneurysm because its appearance angiographically was similar to the acutely ruptured aneurysm, and this aneurysm left untreated could be a potential source of subsequent rupture in the future. Stanson et al state that prophylactic treatment of large aneurysms should be considered in anticipation of the risk of rupture. The other smaller aneurysms arising from the replaced right hepatic artery did not have the appearance of impending rupture at the time of the procedure. Aneurysms of this caliber may resolve over time as clinical symptoms resolve.

Overall, transcatheter embolization of spontaneous perirenal hemorrhage secondary to PAN has been described with low incidence of procedure-related mortality (3.6%) and has the ability to spare adjacent non-involved organs. We used microcoil embolization during this procedure for immediate and permanent thrombosis of the aneurysms. Excellent results have been achieved consistently over the years with catheter-directed embolic agents such as microcoils, Gelfoam (Pharmacia and Upjohn, Kalamazoo, Mich), and particulate materials in the setting of acute gastrointestinal hemorrhage. Our patient has done well following her procedure and has been symptom-free over the last two years. She is normotensive on a regime of steroid and immunosuppressive therapies, with no further clinical sequelae of PAN. These aneurysms have been shown to regress with aggressive medical management and tight control of blood pressure. Disappearing aneurysms that reappear in different location have also been described on follow-up angiograms. Darras-Joly et al states that follow-up angiography is not indicated if clinical improvement is evident. However, recent data by Tarhan et al demonstrate that multidetector computed tomography is an excellent noninvasive modality for evaluation of aneurysmal involvement as small as 3 mm and for follow-up surveillance after treatment.

In summary, we describe both the diagnostic and therapeutic advantages of transcatheter angiography and lifesaving coil embolization in the setting of isolated acutely ruptured hepatic artery aneurysms in a previously undiagnosed patient with PAN. Endovascular specialists should be aware that PAN could present with acutely life-threatening ruptured hepatic artery aneurysms.

REFERENCES


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