Case Report　　　　　　　　　　　　　　　　　　　　　　　　　　　　**Open Access**

Management of Spontaneously Ruptured Splenic Artery Aneurysm in Pregnancy with Endovascular Stent-Graft

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Abstract

Rupture of splenic artery aneurysm (SAA) is a rare condition that mostly occurs during pregnancy and carries a potentially high maternal and fetal mortality. A 24 year-old female in her 20th week of gestation presented with epigastric abdominal pain and hemorrhagic shock related to spontaneous rupture of a large SAA in the mid portion of the splenic artery. The aneurysm was successfully excluded from circulation with endovascular placement of a covered stent-graft. This technique is less invasive than open surgical approach and maintains the patency of splenic artery, which is preferred over endovascular embolization.

Introduction

Splenic artery aneurysm (SAA) is the third most common intra-abdominal aneurysm following abdominal aorta and iliac arteries [1]. It is also the most common location for splanchnic vascular aneurysms constituting 60% of cases followed by hepatic artery 25% and superior mesenteric artery 5% [2]. Yet, it is an overall rare entity with an incidence of 0.02% to 0.1% [3]. Autopsy studies have reported the prevalence of SAA to be between 0.2% to 2% in all age groups and 10.4% in individuals above age 60 [4]. This condition is more predominant among women with a ratio of 4:1 [5]. SAA usually forms in the distal portion of the splenic artery and has a saccular appearance. In about 20% of cases, there are multiple SAAs [6]. The etiologic factors have been proposed to be congenital defect, arteriovenous malformation, portal hypertension, atherosclerosis and pregnancy [6-10].

While the exact pathogenesis of SAA during pregnancy is not clearly understood, a number of anatomic and physiologic changes are thought to contribute to the development of this condition [11,12]. Partial compression of aorta and iliac arteries by the gravid uterus [12,13] and increased blood volume during pregnancy can lead to portal congestion resulting in increased blood flow through the splenic artery and higher stress level on the arterial wall [14]. The arterial wall is also weakened by the hormonal effect of estrogen and progesterone [10] in addition to another hormone named relaxine, which augments vascular elasticity [15]. All these factors can predispose the splenic artery to aneurysmal dilatation.

The most serious complication of SAA is rupture. It occurs in approximately 3% to 10% of cases and carries a mortality rate of 10% to 25% due to circulatory shock [6,12,13,16,17]. The mortality rate has been reported to be as high as 70% in pregnant females [2]. The risk of fetal loss can exceed 90% [18,19]. In pregnancy, rupture tends to occur more frequently during the third trimester perhaps secondary to a more hypertensive picture [20]. In about 20% to 25% of cases, the aneurysm first ruptures into the lesser sac where it is tamponaded by a blood clot at the foramen of Winslow. It is then followed by a free rupture into the greater sac as the pressure within the lesser sac is increased [12,15,21]. This "double rupture" phenomenon provides a very short window of opportunity for diagnosis and treatment.

Patients with ruptured SAA usually present with acute onset epigastric or left upper quadrant abdominal pain, nausea and sometimes emesis. These patients may or may not have peritonitis. Hemodynamic instability and evidence of circulatory shock is sometimes initially absent in patients whose SAA has ruptured into the lesser sac. Anemia is evident a few hours after the onset of symptoms [12]. Plain abdominal x-ray may show a calcified ring with central lucency [22]. CT scan or ultrasonography often confirms the diagnosis by demonstrating both SAA and hemoperitoneum. Angiography is the most accurate method of establishing the diagnosis [23].

Emergent surgical or endovascular intervention is the treatment of choice for patients with ruptured SAA. Laparotomy and aneurosectomy with or without splenectomy has been the mainstay approach to management [12,20,24]. Recently, laparoscopic and endovascular modalities have also been introduced [5,20]. While successful endovascular embolization has been reported for ruptured SAA, endovascular stent-graft placement has been mostly studied in the elective setting [1,25-27]. In this case report, we present a successful endovascular exclusion of a ruptured SAA with expanded polytetrafluoroethylene (ePTFE) stent-graft. Informed consent for this publication has been obtained from the patient.

Case Presentation

A 24-year-old, otherwise healthy, G1P0 female in her 20th week of gestation presented with a 3-hour history of sudden onset epigastric and left upper quadrant abdominal pain, nausea and one episode of emesis. She was initially hemodynamically stable with no peritonitis and appropriate fetal ultrasonographic study. She became progressively tachycardic (heart rate in the 140s bpm), tachypneic (respiratory rate 26/min) and hypotensive (systolic blood high 80's mmHg) over the next 2 hours, however, and required transfusion of blood products for maintenance of hemodynamic status. Base deficit was 6.8 with

HCO3 of 17 and Hb had decreased to 6.5 from 8.2 on admission. CT of abdomen and pelvis revealed hemoperitoneum and a large saccular SAA in the mid portion of the splenic artery (Figure 1).

Endovascular stent-graft placement

The patient was emergently taken to the angiography suite. The celiac tree was accessed via right common femoral artery. Injection of contrast confirmed the presence of a large mid SAA with active leak. The aneurysm was noted to have a wide neck with the guidewire preferentially selecting the aneurysm itself. To facilitate the selection of the efferent limb of the splenic artery out of the aneurysm, a 6-French flexor guiding sheath (COOK MEDICAL® FLEXOR® RAABE) was introduced and placed at the origin of the splenic artery. Selection of the distal aspect of the splenic artery was accomplished with the help of a "hairpin wire" after about 11 minutes.

The aneurysm was first excluded from the circulation by a 6×25 mm expanded polytetrafluoroethylene (ePTFE) stent (GORE® VIABAHN®) but then required deployment of a 6×50 mm ePTFE stent due to questionable leak at the efferent end of the initial stent (Figure 2).

The total duration of the procedure was 1 hour and 27 minutes. The hemorrhage was fully controlled in about 36 minutes from the start of the procedure. Total fluoroscopy time was 49.5 minutes. Total radiation dose was 1960 mGy. Fetal monitoring was performed during the procedure and care was taken to minimize radiation delivered to the fetus by protecting the lower abdominal and pelvic regions with a lead gown.

Outcome

The patient regained hemodynamic stability almost immediately following the procedure. She required laparoscopic evacuation of approximately 2,800 mL of old hemoperitoneum two days after the procedure. During the diagnostic laparoscopy, no evidence of new hemorrhage or splenic infarct noted. She was discharged home after 10 days of hospitalization. The fetus, unfortunately, did not survive the maternal circulatory shock and was delivered one day prior to discharge. At 18-month follow up, the patient remains asymptomatic. She is in her third trimester of an uncomplicated second pregnancy.

Discussion

The endovascular approach to management of SAA has gained increasing popularity due to its high technical success rate and improved morbidity and mortality in comparison to open surgery [5]. Two main principle techniques have been described: exclusion
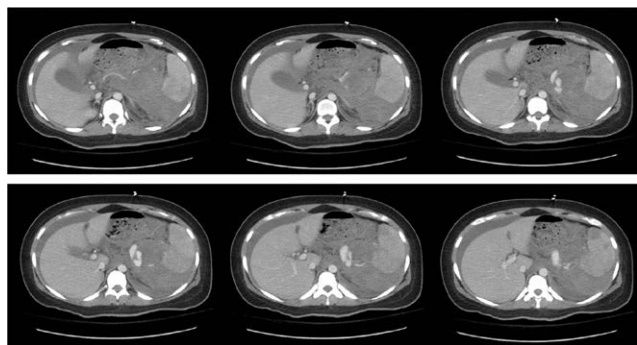

Figure 1: Axial views from contrast enhanced CT demonstrating hemoperitoneum and a ruptured SAA.
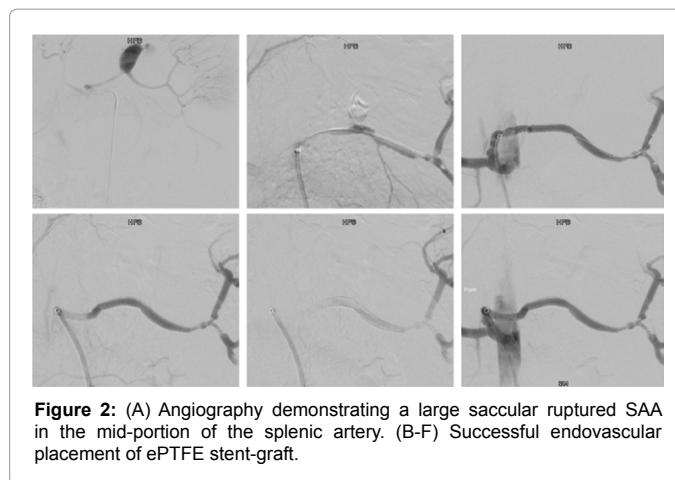

Figure 2: (A) Angiography demonstrating a large saccular ruptured SAA in the mid-portion of the splenic artery. (B-F) Successful endovascular placement of ePTFE stent-graft.

of SAA with occlusion of splenic artery and SAA exclusion with maintaining splenic artery patency. Examples of the first technique include angioembolization with coils, n-butyl cyanoacrylate (n-BCA), or application of expandable nitinol mesh vascular plug (ST. JUDE MEDICAL® AMPLATZER VASCULAR PLUG®) [5,25]. The second technique was traditionally achieved either by coil embolization and dense packing of the aneurysm itself ideal for saccular aneurysms with a narrow neck [5,28] or aneurysm coiling with application of a bare stent [5,29]. With the advent of covered stents, their successful utilization in the management of SAA has been reported in multiple case reports [1,5,27,30-33].

While the placement of covered stent-grafts in the management of SAA offers advantages including the ability to exclude an aneurysm with a wide neck and maintaining the native blood flow through the splenic artery [27], most studies have been focused on the application of this technique in the elective setting [1,5,27,30-33]. Some general indications for elective repair of SAA include a diameter greater than 2 cm, pregnancy, women of childbearing age, cirrhosis, portal hypertension, patients undergoing liver transplantation, and any pseudoaneurysm (often caused by vascular damage following pancreatitis or trauma) [5,25]. In any of these cases, the vascular surgeon has adequate time to select both afferent and efferent limbs of the aneurysm and safely place a covered stent-graft. Splenic artery tortuosity is often the main limiting factor preventing rapid advancement of a guiding sheath or guiding catheter to a suitable level of deployment [5].

In ruptured SAA, the priority is prompt control of hemorrhage. The most rapid means of achieving that goal is, therefore, considered the management of choice. The variable degree of experience with open or endovascular techniques among different surgeons plays a vital role in selecting the approach to management. Many vascular surgeons continue to consider laparotomy, open vascular control, aneurosectomy with or without splenectomy as the first line intervention [12,20,24,34]. Yet, satisfactory outcome has been reported with endovascular embolization [25,26,35]. In this case report, timely vascular control was achieved by placement of an ePTFE stent-graft was performed in a 24 year-old pregnant female with ruptured SAA and evidence of circulatory shock.

Two main factors contributed to our successful exclusion of SAA with the use of covered stent-graft. First, the use of an endoprosthesis with reduced delivery profile allowed prompt progression of the catheter through a relatively tortuous splenic artery. Second, the mid-portion location of the aneurysm with respect to the splenic artery

provided enough proximal and distal limbs to deploy a 5 cm stent-graft to fully exclude a wide-necked SAA from the circulation. Advancing the stent-graft, despite its low delivery profile, to a more distal aspect of the inherently tortuous splenic artery can potentially lead to a longer duration of hemorrhage before obtaining vascular control. Angioembolization may be a more suitable for distal SAAs [1].

We reported how long it took to control the hemorrhage (36 minutes) due to the importance of timely vascular control in a hemodynamically unstable patient with ruptured SAA. Unfortunately, we could not find similar data in the literature for comparison. A retrospective study of 40 patients who underwent proximal splenic artery embolization for reasons other than SAA by Zhu et al. in 2010 reported mean occlusion time of 31.5 minutes and 24.4 minutes with the use of coil and vascular plug respectively. The difference between the two modalities was found to be not significant [35]. The primary reason that emergent endovascular stent-graft placement in managements of patients with ruptured SAA has not been widely accepted is the presumed longer duration of obtaining hemostatis. Additional studies and experiences will be needed to elucidate whether this presumption holds true in most clinical settings. In the meantime, the experience and comfort level of the vascular surgeon should be the main determinant of the technique.

Conclusions

Endovascular placement of a stent-graft is a less invasive alternative to open surgical repair in hemodynamically unstable patients with ruptured SAA. While the period to achieve hemostasis can be comparable to endovascular embolization, this technique also offers the maintenance of native blood flow through the splenic artery.

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