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# Splenic artery aneurysms during pregnancy: An obstetric nightmare

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ABSTRACT

*Objective:* In this study we report our experience in the management of Splenic Artery Aneurysm (SAA), diagnosed during pregnancy.
*Study Design:* The current manuscript describes three different events, treated in out our department, involving SAAs diagnosed during pregnancy. Each case presents an unusual course and a unique clinical challenge.
*Results:* The first case is of a 25 week's gestation twin pregnancy with ruptured SAA ending in maternal and fetal death. Another case of SAA rupture presented at 27 week's gestation with consequent emergency cesarean section and splenectomy. In the last case, two SAAs were incidentally diagnosed at 25 weeks' singleton gestation. The patient was managed conservatively and delivered by an elective cesarean section at 34 weeks followed by postpartum angiographic embolization of the aneurysms.
*Conclusions:* Health care providers and especially obstetricians should be aware of the diagnosis of ruptured SAA in a pregnant woman with abdominal discomfort and hemodynamic deterioration. In addition, once an asymptomatic pregnant patient is diagnosed with a SAA, conservative surveillance may be allowed.



## Introduction

Splenic artery aneurysms are the third most common form of abdominal visceral artery aneurysms following aortic and iliac artery aneurysms. Those aneurysms are relatively rare with an incidence of 0.16–0.78% [1]. SAA is diagnosed more frequently in young women, with up to 95% of reported cases detected during child-bearing years, prominently during pregnancy [1,2].

The indications for intervention in diagnosed SAAs are related to the natural history of this condition with emphasis on factors that increase the risk of spontaneous rupture. All symptomatic SAAs should be treated as a matter of urgency. Trastek et al. [3] further propose that all incidental, asymptomatic aneurysms detected during or before a planned pregnancy should be managed immediately as well. In addition, all asymptomatic aneurysms greater than 2 cm in diameter and SAAs that increase in size during follow up should be treated without delay [2,4]. Lang et al. recommend that even an aneurysm of $\leq$ 2 cm in diameter should be treated during pregnancy since diameter of the aneurysm does not reflect the probability of its rupture [5].

Pregnancy itself can increase the risk of SAA rupture as a result of hormonal changes (estrogen, progesterone, relaxin) and mechanical changes (increased plasma volume, cardiac output, and portal hypertension) with reported maternal and fetal mortality as high as 75% and 95%, respectively [6]. Major contributing factors to mortality are the minimal prodromal symptoms and acute deterioration after aneurysmal rupture. Furthermore, SAA rupture is often confused with other common obstetric emergencies [1,2,7]. The diagnostic evaluation of unstable patients may include the use of a CT scan, MRI, or angiography, all of limited role because of their time consuming nature. Ultrasound can be used to diagnose common obstetric emergencies such as placental abruption, placenta previa, and uterine rupture yet several case reports address the use of bedside ultrasound to detect intra-abdominal free fluid and assist in the

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diagnosis of less common causes of non-traumatic hypotension and hemodynamic collapse [8].

In the current manuscript we present three different events of SAAs during pregnancy, treated in our department, their management and their outcome.

*Case 1*

A 27 year-old G2P0 healthy woman in her first twin pregnancy at 25 + 0/7 weeks gestation with an unremarkable past medical history was admitted to the Obstetric Emergency Department (OED)complaining of sudden onset of left lower abdominal pain and vomiting. She was previously treated with Methotrexate for an ectopic first pregnancy and her current pregnancy was achieved by IVF.

Upon admission she looked pale, and seemed weak and painful. Her blood pressure was 106/67 and pulse 100 with no respiratory distress. She was alert and oriented and was able to complete full sentences. She had no fever and no urinary complaints, denied having uterine contractions or vaginal discharge, and felt fetal movements well. On physical examination her abdomen was soft with minor tenderness on the left flank at percussion and the uterus was not tender. Speculum examination demonstrated a normal looking cervix with a small amount of bloody show and no exposed membranes. Gynecological examination found the cervix to be was long and closed measuring 11–16 mm length on transvaginal ultrasound. Abdominal ultrasound revealed two vital fetuses with a posterior, normal looking placenta with no apparent signs of abruption and adequate amniotic fluid in both sacs. No uterine activity was recorded on CTG (fetal cardiotocography).

Laboratory workup included WBC = 13,000, CRP (C-reactive protein) = 7, Hemoglobin was 10.9 g/dl, blood clotting indices and chemistry were normal as well. Urine dip stick showed insignificant WBC, RBC and mild ketone levels and ECG showed sinus tachycardia.

Betamethasone for fetal lung maturity was given and IV Atosiban (Oxytocin receptor antagonist) started due to abdominal pain resembling contractions. She also received IV Ceftriaxone, suspecting pyelonephritis.

Shortly after, she began complaining of weakness and acute left upper quadrant pain. She was dyspneic, respiratory rate 36, SpO2 86% at room air and 96% using an oxygen mask.

With rapid deterioration in the patient's condition and impaired blood gasses the diagnosis of amniotic fluid embolism was suggested. CPR was initiated and the patient was urgently transferred to the operating room for an emergency cesarean section (CS).

Entering the peritoneal cavity with the uterus still intact, about 1.5 L of fresh blood and blood clots were seen. A low segment uterine incision was performed and two non viable infants weighing 745/725 g were delivered with no apparent evidence of placental abruption. The patient remained hypotensive with systolic blood pressure of 60 mmHg. General surgeons were called in and the skin incision was extended to the xyphoid. She received colloid, crystalloid fluids and blood products and aortic supraceliac pressure was applied followed by an increase in systolic blood pressure to 120 mmHg. An active bleeding site was not identified, yet diffuse abdominal oozing was prominent. The patient was hypothermic (32 °C), and acidotic with arterial pH = 6.9) with clinical and laboratory signs of DIC. After hemodynamic stabilization the abdomen was closed with Bogota suture.

Within a few hours, re-laparotomy was performed due to hemodynamic instability. Exploration of the abdomen detected bleeding from the splenic artery and an emergency splenectomy and uterine B-Lynch sutures were performed. The patient was transferred to the ICU unit and succumbed a few days later due to multiple organ failure and sepsis. Histopathology of the spleen noted thinning of the splenic artery, characteristic of splenic artery aneurysm.

*Case 2*

A 30 year-old female G1P0 at 27 + 0/7 week's gestation presented to the OED (Obstetric Emergency Department) complaining of sudden diffused abdominal pain, with no other gastrointestinal or genitourinary complains. She denied having uterine contractions, vaginal bleeding or discharge and reported feeling good fetal movements. Her past medical history was significant for resection of Wilm's tumor with left nephrectomy at the age of one. Ten years later she underwent explorative laparotomy for bowel obstruction, adhesiolysis and an appendectomy. This was her first, spontaneous, so far uneventful pregnancy. Upon admission she was afebrile with normal vital signs but looked painful. On physical exam the abdomen was tender and the uterus was soft. Gynecological exam found the cervix long and closed. Abdominal ultrasound revealed a viable fetus, posterior placenta with no signs of abruption. Fetal monitoring was reassuring with uterine contractions that ceased after hydration and NSAIDS (100 mg suppository Indomethacin) and she received IM Betamethasone to enhance fetal lung maturity. Examination by a general surgeon was unremarkable. Soon after and unexpectedly, she became pale and weak, and complained of dyspnea. A prolonged fetal deceleration was noted on the CTG and the woman was transferred to the operating room. A low segment abdominal pfannenstiel incision was performed revealing hemoperitoneum. After performing a corporeal uterine incision, a live 1000 g infant was delivered (Apgars 3 and 10 at 1'and 5' respectively, umbilical cord pH = 6.9) with no signs of placental abruption. Bleeding from the splenic hilum was noted and splenectomy was accomplished. Mother and infant, were both discharged, in good health. Histopathology of the spleen revealed ruptured splenic arteries, consistent with splenic artery aneurysm.

*Case 3*

A 26 year-old G2P0 healthy female with an unremarkable past medical history was referred to the OED at 25 + 1/7 weeks' gestation because of progressive dyspnea and shortness of breath.

Her physical exam was unremarkable with BP = 110/70, pulse 120, respirations 22 with SpO2 100% in room air. Blood gases, ECG and chest x-ray were normal. Venous ultrasonography Doppler study (VUS) was performed revealing a thrombus in the right popliteal vein. She was admitted to the High Risk Pregnancy (HRP) department and treatment with LMWH 60 mg bid was initiated. Twelve hours after admission she reported difficulty breathing and was tachycardic and dyspneic Saturation was 100%, bed side cardiac echo was normal. CT pulmonary angiography showed low probability for pulmonary embolism, but two SAAs measuring 13 and 11 mm were noted as an "incidental finding" (Fig. 1).

Reevaluation of the VUS Doppler exam ruled out DVT and the LMWH was discontinued. A multidisciplinary consult with the participation of general and endovascular surgeons, invasive radiologists and perinatologists was held At this time and due to the implausible clinical signs, suggesting rupture of the SAA, their size and the early gestational age, the forum proposed prolonged hospitalization and close observation before any intervention. The patient was monitored daily for vital signs, saturation and blood count. Four packed blood units were kept available upon immediate demand and an ultrasound evaluation of the aneurysms was performed twice a week by the obstetric

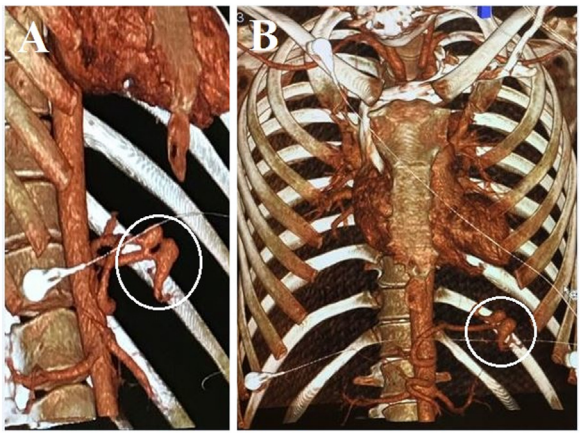


**Fig. 1.** Three dimensional reconstruction using arterial-phase computed tomography imaging. (A) Demonstration of tortuous splenic artery with the incidentally discovered "golf-ball" appearance of two mid-splenic artery aneurysms (circle). (B) Magnification of SAAs measuring 11 and 13 mm (circle).

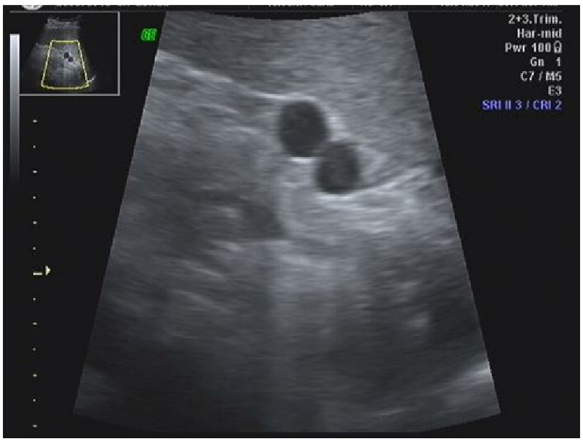


**Fig. 2.** Sagittal view ultrasound of the maternal spleen at 28 weeks gestation. Two rounded, cystic appearing aneurysms of the splenic artery are seen in the para-hilar region of the spleen with no sign of free fluid.

ultrasound senior staff for aneuryms' size and evidence of hemorrhage (Fig. 2).

At 34 + 1weeks gestation an elective, uneventful cesarean section was performed and a healthy boy weighing 2115 g was delivered with good APGAR scores. The following week, the patient underwent angiographic embolization of both SAAs with splenic preservation (Fig. 3). Two weeks later she reported having LUQ pain. A splenic abscess was detected and she was treated with IV antibiotics with clinical improvement.

## Discussion

The etiology leading to the development of SAAs is not fully understood. SAAs can be atherosclerotic and related to arterial degenerative syndromes, may be associated with arterial degeneration, attributed to medial fibrodysplasia, autoimmune disorders, portal hypertension or collagen vascular diseases. Despite its rare occurrence in the general population with low rupture rate, symptomatic SAAs have a 2–10% risk of rupture and asymptomatic SAAs not associated with pregnancy carry a mortality rate of 10%–20% [9]. More than 400 cases of ruptured SAAs have been reported in the international literature, approximately 30% of these described during pregnancy [10]. Still the true prevalence of SAAs in pregnancy or in women of childbearing age is estimated to be less than 0.1% [11,12]. While some study populations claim that ruptured SAAs are exceedingly rare in young women with no identified ruptures during pregnancy [13], others report rupture of twelve percent during the first and second trimesters [14], 13% during labor, 6% postpartum and 69% of SAA ruptures which will occur in the third trimester [1]. In an attempt to ascertain a possible benefit of screening for SAA in pregnant women, McMahon et al. studied a total of 27,527 pregnancies with reported SAA prevalence of 0.1% as previously recounted and 0.004% rupture rate, or 3.6 occurrences of rupture per 100,000 pregnancies [13].

While 80% of splenic artery aneurysms are asymptomatic they are increasingly detected on abdominal imaging studies. Pre-rupture symptoms may be variable and not specific and include vague or sharp left upper quadrant, epigastric, or flank pain, sometimes radiating to the left shoulder (the Kerr sign). Other related symptoms include nausea, vomiting, dyspepsia, and anorexia [15].

Although the precise mechanism of arterial rupture is unknown, when ruptured, 40% of cases may present with sudden cardiovascular collapse due to intraperitoneal hemorrhage and hypovolemic shock with high mortality rate [1]. Aneurysms may initially rupture into the lesser sac with a limited, transient tamponade and minimal clinical symptoms. Within 6–96 h, the blood overflows into the peritoneal cavity through the foramen of Winslow, ("double-rupture phenomenon") resulting in hemorrhagic shock. This delayed rupture may manifest itself in 25% of these cases [16,17].

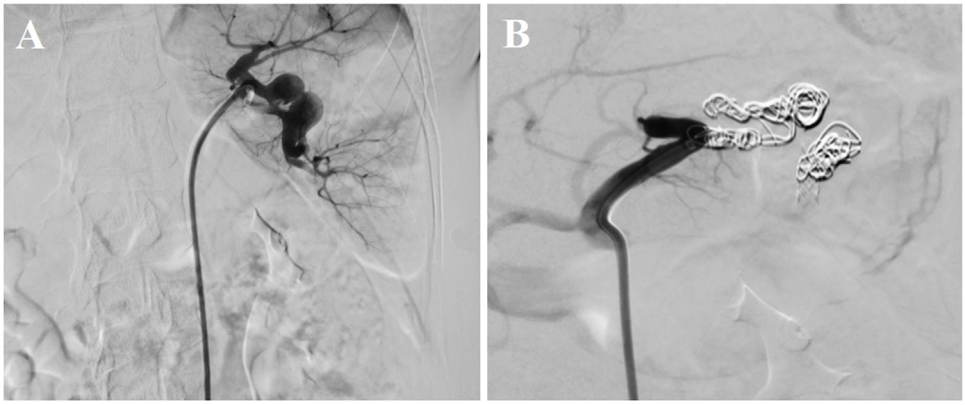


**Fig. 3.** (A) Selective splenic artery angiography via right common femoral artery access revealed two aneurysms in the upper and lower branches of the splenic artery. (B) Wire was introduced to the artery at the lower pole, distal to the aneurysm. To preserve the artery 4 mm covered stent (Bently) was deployed at the area of the aneurysm. Arteriogram showed endoleak around the stent. A microcatheter was introduced to the aneurysm and detachable coils deployed until embolization to the sac was achieved. Embolization was performed form distal to proximal pole due to the sharp angle of the artery.

Special consideration is prudent in cases of SAAs diagnosed in pregnant women where high risk of maternal and fetal mortality has been reported. Hormonal changes during pregnancy affect vascular arterial wall, causing internal elastic lamina disruption and medial fibrodysplasia. The resultant mural degeneration combined with increased blood flow can lead to weakening of the vessel wall and aneurysm formation. The delay in the diagnosis of ruptured SAA during pregnancy is attributed to the pursue of the more common obstetric and non obstetric emergencies such as placental abruption, uterine rupture, pyelonephritis or amniotic fluid embolism [1,2,7] and imaging may expose mother and fetus to high doses of radiation. Ultrasound can be a helpful tool for rapid assessment of acute abdominal catastrophe [8] and FAST (Focused Assessment with Sonography for Trauma) scan has been shown to detect as little as 100 mL of intra-abdominal fluid with 88% sensitivity [18].

The first successful treatment of ruptured SAA and maternal survival was described in 1940 by MacLeod, and the first fetal-maternal survival reported only in 1967 [1,19,20]. Ha et al. [1] reviewed the records of 32 patients, diagnosed with SAA during pregnancy with mean SAA size of 2.25 cm (range 0.5–4 cm) and reported rupture of half of the SAAs that were less than 2 cm in diameter. Due to the high risk of rupture and mortality during pregnancy, elective SAA surgery is now recommended for asymptomatic women of child bearing age, or even during pregnancy [1,19]. If diagnosed during pregnancy, the optimal timing of surgery is suggested to be after the first trimester [5]. In their case report, Parrish et al. [12] describe a non-pregnant woman who was diagnosed with an incidental 9 X 13 mm hilar SAA during an ultrasound performed for abdominal pain and was offered treatment but presented nine weeks later with an eight weeks gestation. She underwent embolization of the aneurysm at 31 weeks, developed splenic abscess three weeks later and delivered vaginally at 36 weeks [21]. Expectant management for asymptomatic, less than 2 cm SAAs, was previously discussed with the exception of aneurysms detected in populations at high- risk for rupture such as pregnant women or those anticipating pregnancy [22].

In the third case described in our series, two SAAs were incidentally discovered and managed conservatively. This unique approach was made possible by the joint multidisciplinary team in our hospital and the availability of trained staff. Accordingly, a prolonged hospitalization with close follow up including frequent maternal and fetal assessment. These essential conditions set the basis for permitting prolongation of the pregnancy without risking maternal or fetal complications due to early intervention, surgical or radiological exposure while sparing the spleen and avoiding post embolization complications during pregnancy.

The strength of our manuscript lays in the diverse nature of SAA and its possible complications, presented in our three patients. However, small number of cases and lack of obstetric experience in this vascular complication during pregnancy is the main weakness of the current document.

Based on our experience and literature search we propose a flow-chart scheme for the management of asymptomatic/incidental finding of SAA during pregnancy (Fig. 4).

In conclusion, health practitioners and especially obstetricians should be aware of the possibility of ruptured SAA in pregnant women with unspecific abdominal symptoms and complaints. Once an asymptomatic pregnant patient with a SAA less than 2 cm in size is diagnosed, conservative surveillance may be offered. Planned cesarean delivery at 34–36 weeks' gestation followed by elective SAA embolization will therefore evade extreme premature delivery as well as splenectomy and radiation hazard during pregnancy. Additional information of SAA's, diagnosed in pregnant women is still needed in order to determine the right time for intervention balancing the risks and benefits of embolization and radiation exposure versus surgery or expectant management.

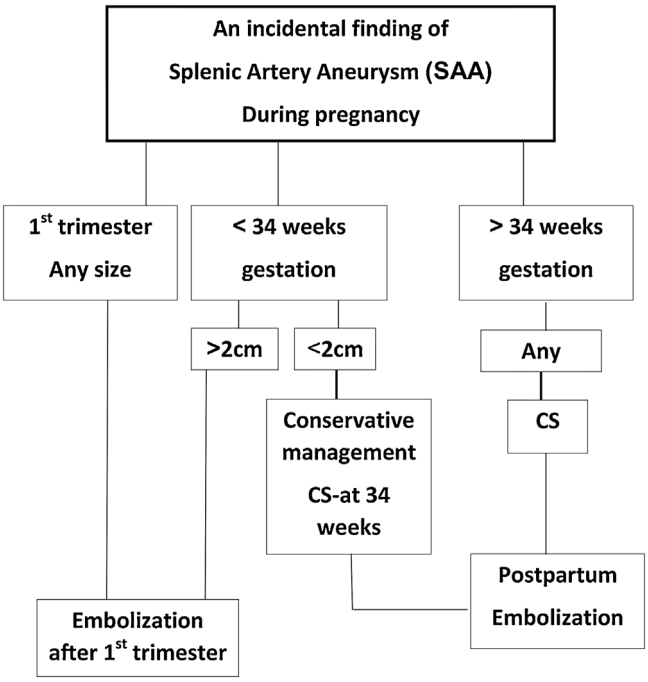


**Fig. 4.** Flow chart scheme – management of splenic artery aneurysm (SAA) detected during pregnancy.

## Comment

Ruptured SAA is a medical emergency and should also be considered in the differential diagnosis of hypovolemic collapse during pregnancy.

## Conflicts of interest

None.

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None.

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