

CLINICAL ARTICLE

Obstetrics

Splenic artery aneurysm in obstetrical patients: A series of four cases with different clinical presentation and outcome

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Abstract

Objective: To describe four consecutive cases of splenic artery aneurysm (SAA) with different clinical patterns of presentation among obstetrical patients.

Methods: A series of four cases of SAA diagnosed in pregnant or postpartum women at our University center between January 1998 and December 2020. Clinical and radiologic data were retrospectively obtained by reviewing paper and electronic medical records after acquiring patient's consent.

Results: One case was completely asymptomatic and incidentally identified at the beginning of pregnancy, thus allowing for multidisciplinary treatment. The other three cases were unknown: two manifested with maternal collapse due to aneurysm rupture in the third trimester of gestation, whereas one presented with acute abdominal pain during the postpartum period and was successfully managed before rupture occurred.

Conclusion: Although extremely rare, SAA rupture in obstetrical patients can be associated with dramatic consequences. Early suspicion and prompt intervention are essential to avoid fatal outcomes, so promotion of knowledge of all the potential clinical patterns of presentation of SAA rupture among obstetrical patients is mandatory.

KEYWORDS

aneurysm, postpartum, pregnancy, shock, splenic artery

1 | INTRODUCTION

Splenic artery aneurysms (SAA) are defined as more than 1-cm pathologic dilation of the splenic artery. They are the most common type of visceral arterial aneurysms, accounting for approximately 60% of the diagnoses.¹

In contrast to other types of arterial aneurysms, SAA are found more frequently in women than in men, with a 4:1 female to male

ratio. Of note, 58% of SSA are diagnosed in women of childbearing age.¹

Splenic artery aneurysms are usually asymptomatic or cause non-specific, mild abdominal symptoms until rupture occurs.^{2,3} Rupture is an extremely rare although life-threatening complication of SAA. Interestingly, over half of SAA rupture events occur during pregnancy, especially during the third trimester, or the postpartum period, possibly due to hormonal influences and changes in portal flow.⁴⁻¹⁰

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Alongside pregnancy, major risk factors for SAA rupture include portal hypertension, pancreatitis, liver transplantation, rapid growth, and aneurysm size greater than 2 cm.¹¹

Rupture of SAA in pregnancy is associated with an exceedingly high maternal and fetal mortality rate, reaching 75% and 95%, respectively.^{1,9} Good perinatal outcomes can only be achieved by early diagnosis and prompt multidisciplinary treatment. Unfortunately, SAA rupture during pregnancy is often misdiagnosed because symptoms can be vague or similar to other more common obstetrical emergencies, such as ruptured extrauterine pregnancy, placental abruption, and uterine rupture. Therefore, high levels of awareness of and suspicion for this condition among obstetricians and other frontline staff is mandatory to improve preparedness and, subsequently, perinatal outcomes.

Here we describe four consecutive cases of SAA in obstetrical patients with different clinical presentation and outcomes.

2 | MATERIALS AND METHODS

This is a case-series including four consecutive cases of SAA diagnosed in pregnant or postpartum women at our University center between January 1998 and December 2020. Relevant clinical and radiologic data were obtained and herein described by reviewing paper and electronic medical records after obtaining patient's consent.

The study was approved by the Institutional Review Board of the University of Milan-Bicocca, Italy (protocol No. 236).

3 | RESULTS

3.1 | Case 1 (year 1998)

A 29-year-old primigravida was admitted to our obstetrical Emergency Department (ED) at 29 weeks of pregnancy complaining of sudden onset of nausea associated with severe epigastric pain radiating to the back.

At the first clinical examination, vital signs were normal (blood pressure 125/85 mm Hg, heart rate 88 beats per minute [bpm]), abdominal palpation was regular and there were no uterine contractions. An ultrasound assessment showed normal fetal movements with a fetal heart rate of 140 bpm, and no free fluid in the abdomen. Thirty minutes after this first assessment, the patient became pale and distressed, started sweating profusely, and reported major pain in her left-sided abdomen. Vital sign assessment revealed blood pressure of 90/60 mm Hg and heart rate of 100 bpm. An ultrasound evaluation was immediately performed, which showed intrauterine fetal demise and blood clots overlying the fundus of the uterus. Supposing placental abruption, the patient was transferred to the operating room for emergency cesarean delivery. After xiphopubic incision of the abdomen, a massive intraperitoneal hemorrhage was identified. Also, a large retroperitoneal hematoma within the lesser

sac and extending to the hilum of the spleen was found. There were no signs of placental abruption. With the intervention of a consultant vascular surgeon, the lesser sac was opened and a ruptured 18-mm aneurysm of the splenic artery located at the splenic hilum was identified. Splenectomy was then performed. Estimated blood loss was 4300 mL; the patient was transfused with 10 units of red blood cells and 5 units of fresh frozen plasma. The postoperative course was uncomplicated and the patient was discharged on postpartum day 10.

3.2 | Case 2 (year 2018)

A 42-year-old woman presented at our outpatient Obstetrical Unit for her first scheduled visit at 14 weeks of pregnancy.

The obstetrical history included a previous first-trimester miscarriage following heterologous assisted reproductive technology in 2017. The current pregnancy was also conceived by heterologous assisted reproductive technology. In addition, the patient reported a hospital admission for pyelonephritis in 2016. By examining the discharge medical records, the obstetrician in charge found an abdominal computed tomography scan report describing the presence of an aneurysm of the splenic hilum of 16 mm, for which the patient did not receive any treatment at the time of diagnosis or follow-up assessment.

The woman was completely asymptomatic. An abdominal magnetic resonance imaging scan was immediately requested and confirmed a 16-mm SAA at the splenic hilum (Figure 1a,b). A multidisciplinary team including obstetricians, interventional radiologists, general surgeons, and anesthesiologists was assembled to appropriately assess potential management options. Considering the gestational age at diagnosis and the radiation exposure risk for the fetus in case of embolization,^{12,13} the team opted for laparotomic resection of the aneurysm with concomitant splenectomy.¹⁴ Laparotomy was performed at 19⁺⁴ weeks of pregnancy, via an incision parallel to the left costal arch. The postoperative course was regular and she was discharged 12 days after surgery.

Pregnancy was complicated by gestational diabetes, which required diet and oral hypoglycemic agents to adequately control blood glucose levels. Fetal growth was regular. At 39 weeks of gestation, the patient went into spontaneous labor and delivered a healthy female baby of 2950 g. The postpartum course was uncomplicated and both the mother and the newborn were discharged on day 3 postpartum. Counseling on the need for vaccination against polysaccharide-encapsulated bacteria like pneumococci was performed before discharge.

3.3 | Case 3 (year 2018)

The third case is a 39-year-old woman at her first pregnancy who presented at our general ED at 29 weeks of pregnancy complaining of mild epigastric pain, fatigue, and general malaise. Pregnancy

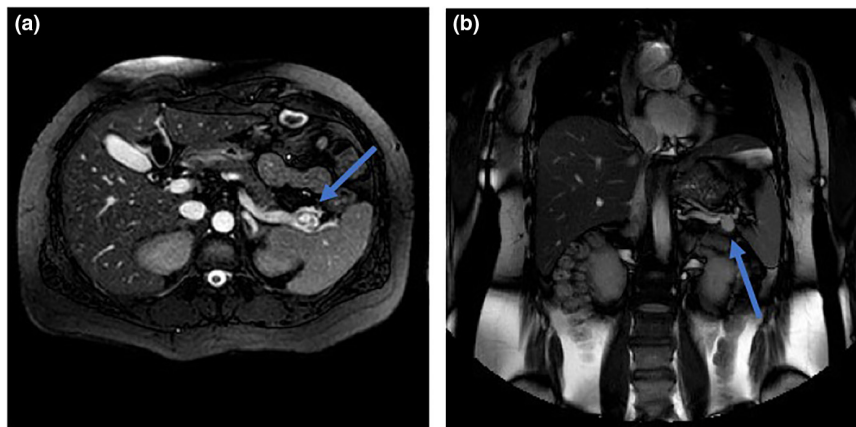


FIGURE 1 Magnetic resonance imaging of abdomen with contrast shows the splenic artery aneurysm at splenic hilum (arrow). Axial scan (a) and coronal scan (b)

was complicated by gestational diabetes, which was adequately controlled by diet.

Upon ED arrival, the woman had hypotension (blood pressure 80/55 mm Hg) and mild tachycardia (heart rate 102 bpm). Her blood examinations showed anemia with a hemoglobin of 9.4 g/dl; electrocardiogram was normal. Abdominal palpation was negative for specific areas of tenderness. Three hours after the initial assessment, symptoms were partly improved with rest and intravenous hydration. She was then transferred to the obstetrical ED for evaluation of fetal well-being before discharge. During the transfer, maternal syncope occurred. A focused assessment with sonography for trauma (i.e. FAST) examination showed massive hemoperitoneum and fetal bradycardia. An emergency cesarean section was performed. During surgery, the woman experienced cardiac arrest, and notwithstanding cardiopulmonary resuscitation maneuvers, she expired in the surgery room. A male neonate of 1165 g was delivered. Apgar score was 1 at the first minute and 4 at the fifth minute; umbilical artery gas analysis at birth showed a pH of 6.75 with a Base Excess of -24 . The newborn was admitted to the neonatal intensive care unit and was managed with mechanical ventilation and surfactant administration. He was discharged after 55 days in good condition.

Autopsy examination of the woman confirmed that the cause of death was the rupture of a 17-mm SAA, located 4 cm from the splenic hilum.

3.4 | Case 4 (year 2020)

A 33-year-old woman in her second pregnancy experienced an intrauterine fetal demise at 35 weeks of gestation associated with a true umbilical cord knot. Pregnancy course had been otherwise uncomplicated.

Eight days after vaginal delivery, she referred to our obstetrical ED for upper abdominal pain started 2 days before and not responsive to painkillers. Her vital signs and blood tests were regular. The pain was localized in the epigastrium and left hypochondrium; the abdomen was soft and non-tender. Analgesics and proton-pump

inhibitors were administered unsuccessfully. A focused assessment with sonography for trauma (i.e. FAST) examination was performed: no renal pelvic dilatation and no free fluid in the abdomen were observed, but a focal dilatation of the splenic artery close to the splenic hilum was suspected. The diagnosis was confirmed by computed tomography scan with contrast: a voluminous and bilobate 3-cm aneurysm in the proximal third of the splenic artery was observed. Thin parietal thrombosis was present (Figure 2a). A multidisciplinary team was assembled and it was decided to perform arteriography with super-selective embolization of the SAA; this was performed 2 days after the diagnosis (Figure 2b–d). Subsequent serial abdominal ultrasound scans documented the complete exclusion of the SAA, with a focal 3-cm area of ischemia containing a 1.5-cm abscess in the middle third of the spleen. The postoperative course was regular. An ultrasound scan performed at 10 months after the procedure showed a small area of fibrosis substituting the focal ischemia in the middle third of the spleen, with regular vascularization of the upper and lower thirds of the organ.

4 | DISCUSSION

Here we describe four consecutive cases of SAA with different clinical presentation and outcome diagnosed in obstetrical patients at our Institution over a 23-year period. Specifically, one case was completely asymptomatic and incidentally identified at the beginning of pregnancy, thus allowing for a planned surgical approach and a successful outcome. In turn, three SAA cases were unknown: two manifested with maternal collapse due to spontaneous rupture of the aneurysm during pregnancy, leading to maternal death in one case and stillbirth in the other, whereas one case presented with acute abdominal pain during the postpartum period and was successfully treated by angiographic embolization before rupture occurred.

Although the true prevalence of SAA and ruptured SAA in pregnant or postpartum women is still unknown, these conditions are considered very rare.^{1,7,15} Over a two-decade period and 63826 deliveries, we identified four cases of SAA in obstetrical patients, two

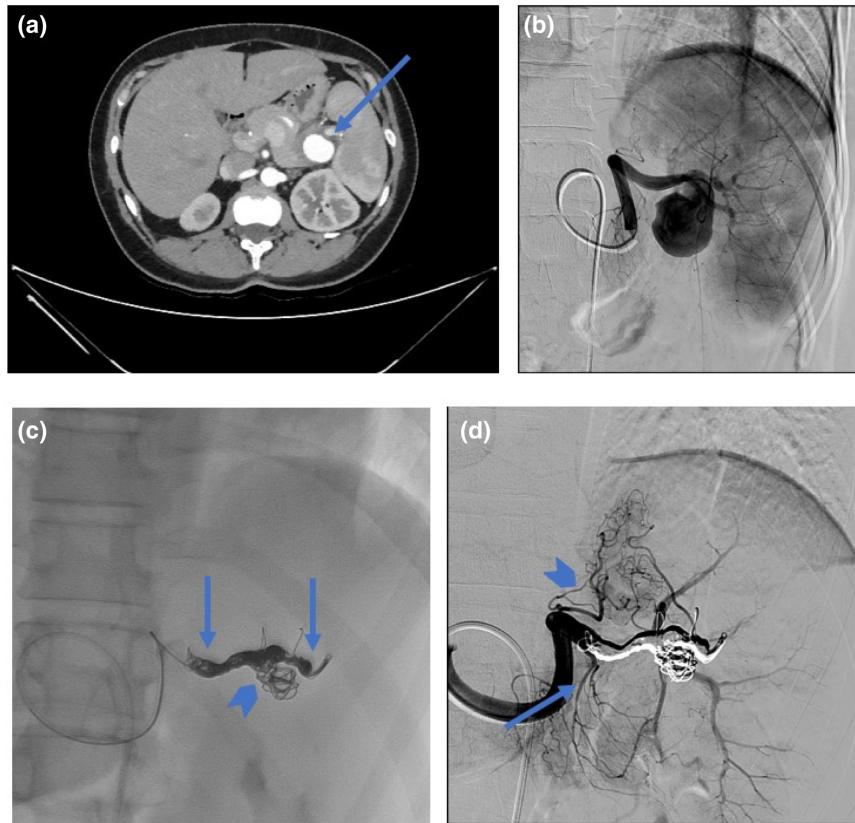


FIGURE 2 Computed tomography of abdomen with contrast shows a voluminous 3-cm aneurysm in the proximal third of the splenic artery (arrow) (a). The splenic artery aneurysm as seen in the splenic arteriography before embolization (b). Aneurysm was embolized by release of multiple detachable coils with the “sandwich technique” (arrows) and neck occlusion (arrowhead) (c). Postembolization splenic arteriography demonstrated complete exclusion of aneurysm sac and patency of distal splenic artery branches recanalized with collateral pathways through short gastric arteries (arrowhead) and pancreatic magna artery (arrow) (d)

of which were complicated by rupture, for a prevalence of 0.006% and 0.003% for SAA and ruptured SAA, respectively. These data are consistent with the findings of McMahon et al.,¹⁵ who identified a 0.004% prevalence of ruptured SAA among 27587 deliveries at a military center. Also, our data are in line with the results reported by Nanez et al.,⁷ identifying no cases of SAA rupture among more than 67000 deliveries at a university center in the USA.

Due to its rarity, diagnosis of SAA rupture is challenging. In addition, diagnosis is made even more difficult by the clinical presentation of SAA rupture.

It can be a rapid process, associating with acute-onset of severe mid and left upper abdominal pain, hemodynamic instability, and massive hemoperitoneum.^{2,16} However, SAA rupture can also occur in two stages. If so, the initial clinical presentation is deceitful because the hemorrhage is contained in the lesser sac by clots blocking the foramen of Winslow and presents with vague symptoms, such as nausea, general malaise, and mild to moderate upper abdominal pain, with no evidence of free intraperitoneal fluid at sonography. This phase can last from minutes to up to a few days, thus providing additional time for diagnosis. When the bleeding reaches the greater sac, the abdominal pain becomes more intense and maternal shock occurs. Misdiagnosis can also happen at this

stage because of similarities in clinical presentation with other more common obstetrical emergencies, including ruptured extra-uterine pregnancy for women in the first trimester and placental abruption and uterine rupture for women in the second and third trimesters.^{1,17}

In both our cases complicated by a two-stage SAA rupture in the third trimester (cases 1 and 3), this condition was not suspected before performing emergent surgery for maternal hemodynamic instability and intraperitoneal bleeding. In particular, placental abruption was hypothesized in case 1.

Before rupturing, SAA can be asymptomatic and incidentally diagnosed or manifest with non-specific upper abdominal symptoms, as occurred in our cases 2 and 4, respectively. Importantly, diagnosis of SAA before rupture allowed for multidisciplinary management and planned treatment, which have been reported as pivotal to markedly reduce maternal fatality rate.^{2,3,18} Mortality rate rises from less than 3% for elective treatment to 75% for emergency intervention after rupture occurs, with a fetal mortality rate reaching 95%.^{19,20} These high percentages are mostly the result of delayed diagnosis.^{1,9,21} In our series of four SAA cases, there was one maternal death and one fetal demise, and SAA was not suspected in either.

Clinical presentation of SAA can vary widely, so this should be taken into account in the diagnostic imaging process for pregnant and postpartum women.¹

In hemodynamically unstable patients with hemoperitoneum, ultrasonography should be the first imaging technique because it is rapidly available without removing the woman from the clinical arena; it is safe, non-invasive, and allows accurate assessment of the degree of intraperitoneal free fluid, thus identifying cases who may require laparotomy.²² Importantly, this imaging can be performed by obstetricians as well as by adequately trained frontline health-care professionals, without requiring a radiology consultation.^{23,24} In addition, sonography performed by an obstetrician can provide information regarding the fetal status, the placenta, the uterus, and the adnexa, which can aid in the process of differential diagnosis.^{1,17}

Ultrasonography with color Doppler also represents the first imaging choice among hemodynamically stable women with vague symptoms. In this case, it should be performed by an expert radiologist and if inconclusive, due to bowel gas, advanced gestation, or small-sized vascular lesions, it should be followed by computed tomography or magnetic resonance imaging.¹ Computed tomography with contrast is the reference standard imaging to identify SAA because it allows precise definition of the size and location of the aneurysm and so allows adequate plan management.^{14,25} However, potential fetal risks related to ionizing radiation and iodinated contrast material exposure contraindicates its use in favor of magnetic resonance imaging, if available, before 25 weeks of pregnancy.^{12,13}

Advanced imaging can also help in differentiating SAA from vein aneurysms. Although much rarer than SAA, splenic vein aneurysms (SVA) share similar etiology, presenting symptoms, complications, and management with SAA.²⁶ Also, high parity and advanced gestation represent substantial risk factors for rupture. As such, SVA and SVA rupture should always be included in the differential diagnosis of non-uterine abdominal pain and hemoperitoneum in pregnant and postpartum women.²⁷

Recently, updated guidelines for management of visceral artery aneurysms have been published.¹⁴ Considering the substantial risk of rupture of SAA in the third trimester of pregnancy and the related exceedingly high fatality rates, these guidelines suggest that SAA diagnosed in asymptomatic or mildly symptomatic pregnant women should always be treated regardless of their size. Hence, the 2-cm cut-off previously used to decide for intervention is no longer valid. Of note, both our cases of SAA rupture had a size smaller than 2 cm. The guidelines also suggest treatment of SAA of any size in women of childbearing age because of the risk of rupture.¹⁴

Management options include endovascular treatment with angiographic embolization and surgery with ligation or resection of the aneurysm and potential concomitant splenectomy.^{3,28,29} Gestational age at diagnosis and SAA localization are key elements in guiding the choice of the therapeutic approach.^{1,30,31} The updated guidelines on management of visceral artery aneurysms suggest a preference for an endovascular approach in electively managed cases, except for distal SAA adjacent to the hilum of the spleen, which should preferably be approached with open surgical techniques.¹⁴ Arterial

embolization can be considered after 25 weeks of pregnancy to avoid potential problems related to fetal radiation exposure^{12,13} and only for SAA located in the proximal or middle third of the splenic artery to prevent splenic abscesses due to ischemia.¹⁴ Splenectomy concomitant to ligation or resection of SAA is required in the case of distally located aneurysms. The choice of open versus laparoscopic surgery depends on the team's experience and the duration of the pregnancy.²⁹⁻³¹

Additional recommendations included in the updated guidelines on management of visceral artery aneurysms include periodic surveillance after endovascular intervention to assess potential endoleaks or continued aneurysm perfusion, and screening of patients with SAA for other intra-abdominal, intrathoracic, intracranial, and peripheral artery aneurysms.¹⁴

Although including all the potential clinical presentations of SAA and SAA rupture in obstetrical patients, this series lacks cases of SVA or SVA rupture. Also, there are no cases of SAA rupture during the first trimester of pregnancy.

In conclusion, SAA and SAA rupture are extremely rare. However, SAA rupture in obstetrical patients can be associated with dramatic consequences. Early recognition and prompt multidisciplinary management are pivotal to avoid fatal outcomes. Knowledge of all the potential clinical patterns of presentation of SAA is mandatory. Our case series represents all such patterns, including two-stage rupture as well as diagnosis before rupture, so provides an extensive update and promotes awareness of this condition as differential diagnosis among ED healthcare professionals managing pregnant or postpartum women with acute onset of upper abdominal pain or hypovolemic shock.

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CONFLICT OF INTEREST

The authors have no conflicts of interest.

DATA AVAILABILITY STATEMENT

Data sharing is not applicable to this article as no new data were created or analyzed in this study.

AUTHOR CONTRIBUTIONS

IC contributed to conception of the work, acquisition and interpretation of data, and drafting of the first version of the manuscript. SO contributed to interpretation of data, and drafting of the first and revised versions of the manuscript. SDN contributed to acquisition and interpretation of data, and drafting of the first version of the manuscript. VG and LLM contributed to acquisition and interpretation of data and critical revision of the manuscript. RC and PV contributed to interpretation of data and critical revision of the manuscript. NR contributed to conception of the work, interpretation of data, and critical revision of the manuscript.

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