




## Case report

# Spontaneous rupture of splenic artery aneurysm in pregnancy: A case report and review of the literature

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## ABSTRACT

**Introduction:** Splenic artery aneurysms (SAAs) are the most common (60%) of visceral artery aneurysms with the possibility of rupture. SAA rupture is associated with 20% mortality, which increases disproportionately to 75% in pregnant women, with fetal the mortality rate of 95%.

**Aim:** We present a discussion based on the case report with a review of the literature on ruptured SAA in pregnancy as well as possible treatment options, complications and prognosis.

**Case study:** Patient, 40-year-old pregnant woman, gravida 2, para 1, 33 week of pregnancy, with ruptured SAA, underwent cesarean section, splenectomy, and hypovolemic shock treatment. The outcomes of the mother and neonates were good. Postoperative recovery was complicated by right-sided pneumothorax.

**Results and discussion:** 87% of patients with SAA are females. In our analysis of cases, with ruptured SAA in pregnancy, the parity may be the factor that influences the maternal and fetal outcomes. In third trimester of pregnancy (as in our case study) the most often mistaken indication for laparotomy was a suspected placental abruption.

**Conclusions:** Ruptured SAA during pregnancy should be considered as one of the non-obstetric causes of intraabdominal bleeding. Careful observation of the patient from the onset of symptoms is a key approach that increases maternal and fetal survival.

## 1. INTRODUCTION

Splenic artery aneurysms (SAAs) have been recognized since 1770.<sup>1</sup>

The incidence of SAAs is estimated to be less than 1%.<sup>2</sup> SAAs are diagnosed incidentally, after they rupture, in symptomatic cases.<sup>3,4</sup> However, about 80% of patients are asymptomatic.<sup>1,3-4</sup>

SAA rupture is associated with 20% mortality, which increases disproportionately to 75% in pregnant women, with foetal mortality of 95%.<sup>2</sup> The foetal distress and mortality usually results from hypovolemia, shock and their consequences.<sup>5</sup>

## 2. AIM

We present the case of spontaneous SAA rupture during the third trimester of pregnancy manifested as ‘double rupture.’ After emergency surgical operation and shock treatment, maternal and foetal outcomes were good. Diagnostic evaluation and management are discussed along with a review of relevant literature.

## 3. CASE STUDY

Patient, 40-year-old pregnant woman, gravida 2 para 1, 33 week of pregnancy, was consulted at Emergency Department because of epigastric pain lasting for about 2 h, probably related to indigestion. The previous clinical and obstetrical history was unremarkable. Examination revealed weak tenderness in epigastric region, sweaty hands and normal tension in the uterus. No signs and symptoms of hypovolemic shock were noted.

Cardiotocography (CTG) record showed moderate pattern of fetal heart rate (FHR) oscillation, short term variation over 4 ms, FHR 140–155 beats/min, no contractions. Fetal ultrasound scan revealed no signs of placental abruption, vertex position of the foetus, amniotic fluid index in normal range and estimated fetal weight – 2062 g. In digital examination cervix was formed, cervical canal was closed and no evidence of vaginal bleeding was found. The patient was admitted to the maternity ward for observation and she demonstrated stabbing abdominal pain and nausea 7 h after admission. In examination, positive peritoneal irritation symptoms and the uterine muscle with increased tension were found. Respiratory rate was 22 breaths/min, 98% oxygen saturation level, heart rate 89 beats/min, blood pressure 90/65 mmHg. CTG record demonstrated marked oscillation and regular contractions every 2–3 minutes. Emergency Pfannenstiel incision was performed to open the abdomen, due to suspected placental abruption, fetal distress symptoms and positive peritoneal symptoms. A viable male infant was delivered via a low transverse hysterectomy, with an Apgar score 4 and 10 points at 1 and 5 minutes after birth, respectively. Carbetocin was administered intravenously after placental removal.

During caesarean section, hemoperitoneum was found (approximately 500 mL), and longitudinal cutting into the xiphoid and packing of five large compresses were performed to control bleeding. Suspected localization of bleeding was situated in left epigastric region, abdominal layers were closed to perform CT scan. During transferring to radiology department, the patient developed symptoms of hypovolemic shock. Immediate relaparotomy with splenectomy, packing of two large compresses and drainage of abdominal cavity, as the best choice of surgical treatment in suspicion of ruptured SAA, were done.

Due to intraoperative blood loss in a total volume of 2500 mL, unstable patient’s general condition with hypovolemic shock parameters, 6 units of packed cells and 6 units of fresh frozen plasma, followed by 1200 IU of human prothrombin complex (Prothromlex), 4 mg of recombinated 7a factor (NovoSeven) and 1.0 g of tranexamic acid (Exacyl) were administered in the operating theatre. As a result, the patient was transferred to Intensive Care Unit with an intravenous infusion of noradrenaline (Levonore) 0.1 mL/h. Two days after splenectomy, relaparotomy with unpacking of two large compresses was performed. Postoperative recovery was complicated by right-sided pneumothorax.

Histopathological report, obtained after surgery, confirmed ruptured SAA.

The mother and her son were discharged in good physical condition three weeks after the operation. The woman was informed about the consequences of her present asplenic situation, the need of vaccination and immediate start of antibiotic treatment as soon as she develops fever.

## 4. RESULTS AND DISCUSSION

Visceral artery aneurysm is an uncommon pathology, with the possibility of rupture. SAA are the most common (60%), followed by hepatic (20%), superior mesenteric (6%) and celiac (4%) artery aneurysms.<sup>5</sup> The incidence of SAA is generally estimated to be less than 1%.<sup>2</sup> A majority of patients with SAA are 50–70 years of age, however 20% are 20–50 years of age. Among these younger patients, the female-to-male ratio is 20 : 1.<sup>1</sup>

We have analysed cases from PubMed database since 1944, search rules: ‘Splenic artery aneurysm’ and ‘Pregnancy.’ As the result, 28 cases of ruptured SAA in pregnancy with maternal and foetal survival were described in literature.<sup>6-32</sup>

In analysed cases of ruptured SAA in pregnancy with good maternal and foetal outcome the median patient age is 33 years.<sup>6-32</sup> The patient presented in our case study is one of the oldest. The range of the age in analysed articles is 23–41 years.<sup>6-32</sup>

Among patients with SAA 87% are females, and 80% are multiparous.<sup>1</sup> In our analysis of cases with ruptured SAA in pregnancy with good maternal and foetal outcome, the percentage of multiparous patients is estimated at 61% comparing to 80% in overall pregnant patients with SAA. It may suggest a presence of some factors that decrease a

risk of rupture in multiparous patients with SAA but the amount of cases up to date is still very small. This observation needs further precise analysis. In the group of patients with ruptured SAA 95% are pregnant females.<sup>1</sup> It has led to a common speculation that changes in arterial connective tissue as well as increased blood volume, portal congestion, and splenic arteriovenous shunting related to pregnancy contribute to splenic artery medial degeneration and aneurysm formation. This relationship ‘hypertension-vascular fragility’ may worsen as a result of a congenital abnormality of connective tissue and an increase in vascular flow in the spleen during pregnancy.<sup>1</sup> Hormonal changes have also been suspected, especially high levels of estrogens.<sup>2</sup>

The clinical phenomenon of ‘double rupture’ gives an opportunity for diagnosis and surgical intervention before the onset of hemorrhagic shock.<sup>5</sup> The patient described in our case study has typical two-step abdominal pain divided by few hours of good general condition with no specific signs or symptoms. As mentioned in the literature, the first sign is due to bleeding to the lesser sac with typical epigastric pain.<sup>1</sup> This is the moment when most patients need medical support. Ruptured SAA in pregnancy are diagnosed mostly incorrectly, because they present with symptoms like: nausea, vomiting, hypotension that are similar to those of other obstetric emergencies. Gynaecological/obstetrical examination and gynaecological/obstetrical ultrasound scan reveals no evident pathology. Free fluid in abdominal cavity, which is supposed to be essential to suspect ruptured SAA, may not be found. Sometimes a small deviation in physical examination could be crucial for the patients’ prognosis. In our case study, sweaty hands were a symptom that was significant during the decision to keep the patient under observation. The symptoms of aneurysmal rupture in a pregnant woman may be mistaken for the common obstetrical emergencies such as uterine rupture, placental abruption or amniotic fluid embolism, as 20% of these patients will develop SAA rupture during labour.<sup>5</sup> At most cases, third trimester of pregnancy was taken under analysis (as well as in our case study) and the most often repeated indication to laparotomy was suspected placental abruption. Ultrasound scan should be considered as the best additional test to distinguish placental abruption from ruptured SAA.

Patients with SAA should undergo surgery if they are or may become pregnant,<sup>1</sup> if the aneurysm is greater than 1.5–2.5 cm or if they have symptoms referable to the abdomen.<sup>1</sup> Some patients may benefit from non operative percutaneous catheter embolisation.<sup>1</sup> Due to low SAA incidence in population, we do not recommend any screening towards SAA.

## CONCLUSIONS

Ruptured SAA in pregnancy must be considered as one of the reasons of non obstetrical intra-abdominal bleeding. The crucial management which increases maternal and fetal survival rate is a careful observation of a patient from the

beginning of symptoms. The clinical phenomenon of ‘double rupture’ gives an opportunity for diagnosis and surgical intervention before the onset of hemorrhagic shock.<sup>5</sup>

## Conflict of interests

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