

## CASE REPORT

# Sudden Rupture of Splenic Artery Aneurysm in a Pregnant Woman Resulting in the Death of the Fetus: A Case Report and Review of Literature

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**Abstract: Background:** Visceral artery aneurysm is an uncommon pathology with a potential risk of rupture and disastrous complications, especially during pregnancy. Sudden rupture is associated with higher fetal mortality. The exact etiology of splenic artery aneurysm remains unclear. However, intimomedial mucoid degeneration is a rare vascular disorder that can lead to aneurysms in young individuals.

**Case Presentation:** In this case, we present a case of unexpected and sudden intra-abdominal hemorrhage in a 26-year-old woman in her first pregnancy (27<sup>th</sup> week) with fetal death but maternal survival. At the onset, blood pressure was 90/70 mmHg, and blood tests showed anemia. The trans-abdominal ultrasound showed fluid in the sub-hepatic area and a significantly slowed fetal heart-beat. Cesarean section was performed, and upon opening the abdomen, there was a massive hemoperitoneum, and the incision of the uterus led to the extraction of a dead fetus. Surgical pathology revealed an aneurysm of the splenic artery, and histological examination supported a diagnosis of intimomedial mucoid degeneration of the vessel.

**Conclusion:** Abdominal pain in pregnancy is a frequent presentation in obstetric departments with a wide possibility of diagnosis. Clinicians face significant diagnostic challenges due to the wide range of potential causes of abdominal pain. The rarity of this pathology and the objective diagnostic difficulties make this condition obscure and unpredictable. Routine screening of the splenic artery with ultrasound and Doppler can be considered in pregnant women with predisposing factors. Timely surgical intervention is necessary to spare the life of the mother and the fetus.

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## 1. INTRODUCTION

The exact etiology of splenic artery aneurysm (SAA) currently remains unclear. Pregnancy represents a recognized risk factor for SAA, with approximately 78% of cases occurring during the third trimester [1]. SAA affects approximately 0.01% to 0.2% of the general population [2].

To date, the literature has established an association between the development of splenic artery aneurysms and several risk factors, namely fibromuscular dysplasia, collagen vascular diseases, female gender, history of multiple pregnancies, and portal hypertension [3].

Increased blood pressure and weakened arterial walls are two factors that can contribute to SAA rupture during pregnancy [4].

Physiological changes during pregnancy promote stress on the arterial wall (*i.e.*, increased blood volume, increased cardiac output, hypertension, and relative portal congestion) [4, 5].

SAA in pregnancy recognizes two favorable factors: the increase in blood pressure and the weakening of the arterial wall [4, 6]. During gestation, hormonal influence could cause a histological alteration with degeneration of the vascular tunica media and consequent aneurysmal dilatation. In these cases, medial fibrodysplasia, fragmentation of the internal elastic lamina, endothelial thickening, and accumulation of acid glycosaminoglycans in the subintimal and medial layers have been documented [6-9].

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The elevated blood volume and cardiac output characteristic of pregnancy can increase blood flow through the splenic artery, resulting in elevated portal venous pressure [2].

The effects of these hormones can be amplified by the release of relaxin, which acts on the elasticity of the artery [4].

Aldosterone and renin can promote thinning of the arterial wall, and alpha-1-antitrypsin deficiency can further promote aneurysm formation [4, 10].

During pregnancy, rupture rates vary from 2-3% to 10%. The rupture can happen suddenly or in two stages. The latter occurs in approximately 25% of cases and is characterized by the preliminary containment within the omentum or blood clots blocking the foramen of Winslow. After approximately 6-96 hours, the rupture releases the peritoneum due to increased tension within the sac minor [11, 12].

Regarding treatment, aneurysms larger than 2 cm should be treated [4]. Minimally invasive techniques, such as transcatheter embolization, percutaneous angiographic embolization, or laparoscopic ligation/resection, can be used. In cases of rupture, caesarian section laparotomy and resuscitation are necessary [13].

In case of rupture of the aneurysm, the symptoms are abdominal pain in the epigastrium and left hypochondrium, also associated with pain in the left shoulder [14].

The diagnosis is not simple as this pathology can be confused with other pathological conditions, such as placental abruption, amniotic fluid embolism, appendicitis, peptic ulcer perforation, cholecystitis, or uterine rupture [1, 15]. For this reason, many cases can be under-diagnosed.

In this case report, we present a case of unexpected and sudden rupture of a splenic artery aneurysm in a 26-year-old woman in her first pregnancy (27<sup>th</sup> week). Histological examination supported a diagnosis of intimomedial mucoid degeneration of the vessel. The dissection of the vessel resulted in massive intra-abdominal hemorrhage (hemoperitoneum) with the death of the fetus.

## 2. CASE REPORT

A 26-year-old woman (native of Italy), in the 27<sup>th</sup> week of gestation (early 3<sup>rd</sup> trimester), went to the emergency room of a city hospital due to the sudden onset of stabbing abdominal pain and a syncopal episode. The patient was in her first pregnancy. Standard prenatal check-ups were performed, and the pregnancy was classified as low risk based on clinical evaluations and the absence of known comorbidities or risk factors.

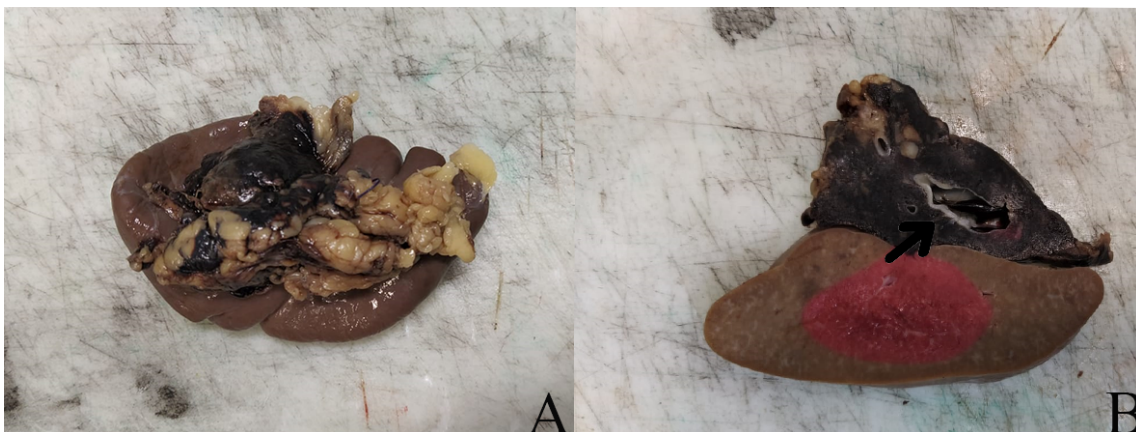
At the onset, blood pressure was 90/70 mmHg. Blood tests were performed in the emergency room which showed anemia (Hb 6 g/dl). Substance abuse was ruled out.

The trans-abdominal ultrasound showed fluid in the sub-hepatic area and a significantly slowed fetal heartbeat. Therefore, a cesarean section was performed. Upon opening the abdomen, there was a massive hemoperitoneum, and the incision of the uterus led to the extraction of a dead fetus. The laparotomy surgical access was enlarged, and peri-splenic bleeding was recognized. Following a section of the gastrocolic ligament, a large clot was identified with an active bleed at the splenic hilum. Splenectomy with ligation of the splenic artery was therefore performed.

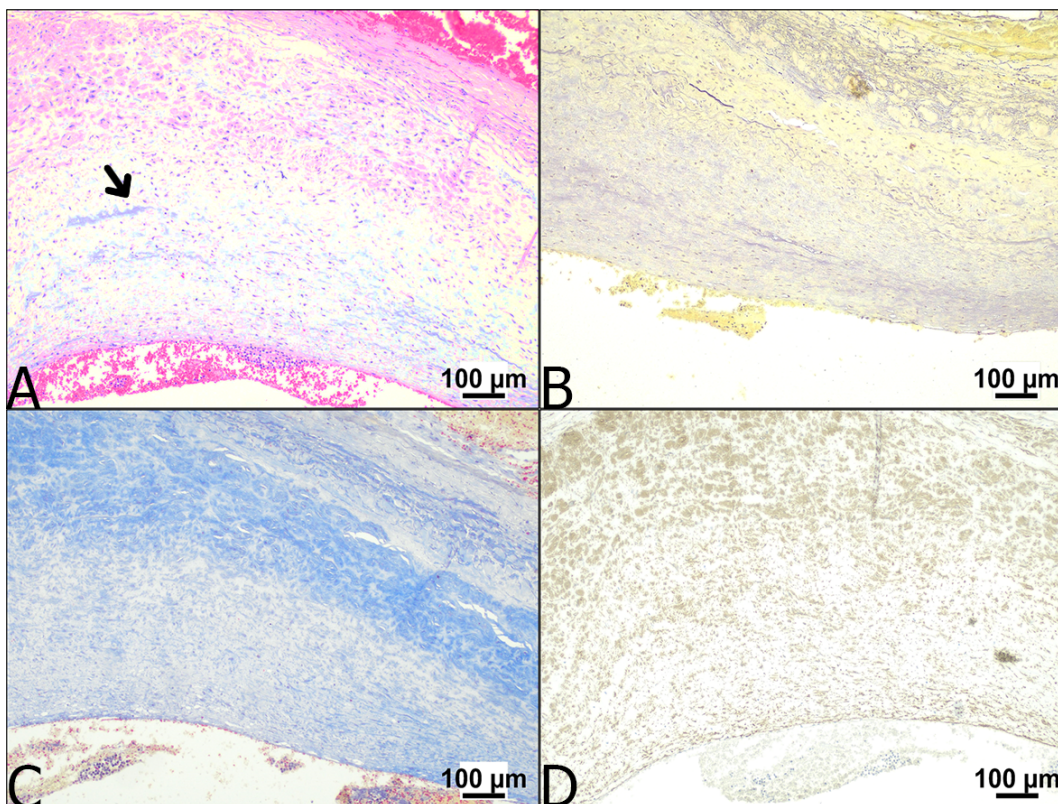
The postoperative course was unremarkable, with rapid and progressive improvement in the woman's condition.

After fixation in formalin, the spleen was analyzed. A significant hematoma was evident in the splenic hilum with a maximum diameter of 4.5 cm. The splenic artery aneurysm showed a wall rupture (Fig. 1).

Histological analysis of the splenic artery showed the progressive reduction of the wall thickness associated with thinning of the muscular layer. The artery exhibited significant wall thinning and lumen dilation with mucoid degeneration of the intima and media layers (Fig. 2A). The internal elastic lamina was fragmented and largely absent (Fig. 2B). Masson trichrome staining showed increased deposition of fibrous tissue within the mucoid degeneration area (Fig. 2C). Immunohistochemistry staining (smooth muscle actin) showed a loss of smooth muscle cells in the region of mucoid degeneration, contributing to the overall weakening of the arterial wall (Fig. 2D). Adjacent to the rupture site (in a different area of the splenic artery), the vessel wall was reduced to a thin fibrous band.



**Fig. (1). Macroscopic findings.** A) Hematoma at the level of the splenic hilum. B) Dilatation of the splenic artery with interruption of the wall (arrow). (A higher resolution / colour version of this figure is available in the electronic copy of the article).



**Fig. (2). Histology.** **A)** Hematoxylin and Eosin (H&E) stain; Original magnification 100X: Demonstrates the overall tissue architecture, highlighting areas of mucoid degeneration with a distinct pale, slightly basophilic appearance (arrow). **B)** Elastic fiber stain; Original magnification 100X: Reveals disruption and fragmentation of elastic fibers within the area of mucoid degeneration, indicating weakening of the arterial wall. **C)** Masson trichrome stain; Original magnification 100X: Differentiates collagen fibers, showing increased deposition within the mucoid degeneration area, suggestive of a reparative process. **D)** Immunohistochemistry for smooth muscle actin; Original magnification 100X: Demonstrates a loss of smooth muscle cells in the region of mucoid degeneration, contributing to the overall weakening of the arterial wall. (A higher resolution / colour version of this figure is available in the electronic copy of the article).

Therefore, mucoid deposition of the intima and the media was detected, while no aspects of atherosclerosis or cystic necrosis of the media were observed. These findings suggested an intimomedial mucoid degeneration of the splenic artery.

The histological examination of the placenta showed tissues within normal morphological limits for the gestational age.

The fetal autopsy showed a male fetus weighing 740 grams with no malformations. The thoracic, abdominal, and pelvic visceral organs were congested. No particular macroscopic or histological findings were noted. Based on the analyses performed, the fetus experienced intrauterine asphyxia secondary to maternal hemorrhage.

### 3. DISCUSSION

Intimomedial mucoid degeneration (IMMD) is a rare vascular disorder that is mostly diagnosed in young people [16]. This pathological condition is characterized by the deposition of mucoid material in the two internal layers of the vessels with the degradation of elastic fibers and a reduction in smooth muscle cells [17, 18]. Moreover, smooth

muscle cells show the deposition of mucin vesicles, causing muscle cell necrosis [19].

A striking characteristic of IMMD is the lack of a luminal thrombus in the aneurysm sac during vascular procedures [19].

Hypertension aggravates the risk of rupture of the degenerated vascular wall. In the study by Abdool-Carrim *et al.* [20], approximately 60% of patients suffering from aortic mucoid degeneration had a history of arterial hypertension. All conditions that cause increased blood flow in an anatomical region (such as pregnancy) can lead to the rupture of vessels, even extra-aortic ones, in patients with vessel degenerative pathology, such as IMMD [21].

Sudden rupture is commonly associated with a very high maternal mortality rate (approximately 75%) with a higher fetal mortality (95%) [1, 22].

The most frequently affected vessels are the aorta and its main branches, especially large and medium-sized vessels, such as the renal arteries, the subclavian, the common carotid, and the common iliac arteries. Although the disease mainly involves the aorta, extra-aortic disease can be found even in the absence of aortic involvement [23].

Momoh *et al.* [24] reported a case of a ruptured splenic aneurysm in a 36-year-old woman in the twenty-sixth week of gestation. The woman had undergone splenectomy and survived, while the fetus showed no signs of life after caesarean section.

Mathelin *et al.* [25] reported a case of a 30-year-old woman at term who experienced sudden hypotension and fetal distress. Emergency caesarean section revealed a ruptured splenic artery aneurysm, leading to fetal death and maternal hemoperitoneum. The patient underwent a distal pancreatectomy and splenectomy.

Cases of fetal survival are also described in the literature. For example, Vaughan *et al.* [26] described a case of a 35-week pregnant woman who presented with sudden abdominal pain and shortness of breath. Imaging revealed a ruptured splenic artery aneurysm. Emergency surgery, including coiling of the aneurysm, caesarean section, and splenectomy, was performed. The patient had a complex postoperative course, but she was discharged home with her healthy baby.

Moreover, Fuji *et al.* [22] demonstrated a case of a 35-week pregnant woman who presented with acute abdominal pain and fetal distress. Emergency caesarean section was followed by exploratory laparotomy, which revealed a ruptured splenic artery aneurysm. Due to severe bleeding, the patient required splenectomy, distal pancreatectomy, and temporary abdominal packing. Despite a complex postoperative course, the patient and her baby recovered fully.

In the present case, the pathology remained asymptomatic and began suddenly with massive hemoperitoneum, severe anemia, and hemorrhagic shock. The rapid diagnosis and surgical intervention allowed the woman's life to be saved but did not allow the fetus to survive. The histological analysis allowed us the diagnosis of intima-medial mucoid degeneration of the splenic artery. Therefore, in this case, the rupture of the aneurysm was essentially determined by important risk factors: the vascular effects of pregnancy with intimomedial mucoid degeneration of the splenic artery.

According to our knowledge, this case is particularly significant for the uncommonness of rupture of the splenic artery with intima-medial mucoid degeneration in pregnancy.

From a clinical point of view, we must keep in mind that abdominal pain in pregnancy is a frequent presentation in obstetric departments with a wide possibility of diagnosis. Clinicians meet significant diagnostic challenges due to the wide range of potential causes of abdominal pain [26]. The rarity of this pathology and the diagnostic difficulties make this condition obscure and unpredictable. The correct balance of a radiation risk against the hypothesis of active hemorrhage is an extremely difficult choice, and there are no clear guidelines [26]. Exposure of the fetus to radiation should be minimized due to the teratogenic risk. Regarding MRI, obvious fetal damage is not documented, but the National Radiological Protection Board advises against the use of MRI during the first trimester [4].

Diagnostic CT scans in pregnancy typically utilize low radiation doses, minimizing the risk of teratogenic effects. However, intravascular contrast agents should be avoided [4].

Ultrasonography and Doppler are preferable due to their non-invasive nature and the absence of maternal-fetal risks (Diagram 1 summarizes the diagnostic and treatment pathway). However, the clinical utility of this technique can be hindered by several factors, including operator-dependent image acquisition, limitations in obese patients, interference from bowel gas, and the presence of arteriosclerosis [4].

The diagnostic difficulty often causes a delay in bleeding control with the real risk of hemorrhage. In this case, the diagnosis of intra-abdominal hemorrhage was intraoperative, and the rupture of the aneurysm was identified only during the post-operative histological examination.

The catastrophic consequences of splenic aneurysm rupture require a call for action; further research and clearer guidelines for the diagnosis and management of similar cases are urgently needed. Given the very high fetal and maternal mortality, ultrasound and Doppler could be performed in pregnant women with abdominal pain, nausea, and vomiting. Furthermore, routine screening of the splenic artery with ultrasound and Doppler can be considered in pregnant women with predisposing factors, such as hypertension, and liver and pancreatic diseases [11]. Timely surgical intervention is necessary to spare the life of the mother and the fetus.

## CONCLUSION

In these cases, clinicians should follow a stepwise approach to evaluate abdominal pain during pregnancy: 1) assess the severity and intensity of the symptoms: a thorough evaluation of the patient's pain, including its location, duration, and character, is crucial, 2) identify potential risk factors, 3) perform diagnostic imaging: Ultrasound and Doppler ultrasound are essential for visualizing the spleen and assessing blood flow, and 4) consider management options; if an aneurysm is detected, Minimally invasive interventions, such as endovascular embolization, may be considered. In cases of hemoperitoneum, immediate surgical intervention, including laparotomy and potentially cesarean section, may be necessary to control bleeding and ensure maternal and fetal well-being.

From a forensic point of view, in cases of fetal and/or maternal death from intra-abdominal hemorrhage, an in-depth macroscopic and histological analysis of the spleen and splenic artery is necessary to correctly identify the cause of the bleeding.

## AUTHORS' CONTRIBUTION

The authors confirm their contribution to the paper as follows: validation: FV; analysis and interpretation of results: VGV; draft manuscript: RB and MD. All authors reviewed the results and approved the final version of the manuscript.

## LIST OF ABBREVIATIONS

IMMD = Intimomedial Mucoïd Degeneration  
 SAA = Splenic Artery Aneurysm

## ETHICS APPROVAL AND CONSENT TO PARTICIPATE

Not applicable.

## HUMAN AND ANIMAL RIGHTS

Not applicable.

## CONSENT FOR PUBLICATION

Informed consent was obtained from the participant.

## STANDARDS OF REPORTING

CARE guidelines were followed.

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## CONFLICT OF INTEREST

The authors declare no conflict of interest, financial or otherwise.

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