



Rupture of splenic artery aneurysm in pregnancy with double-rupture phenomenon: A case report

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ABSTRACT

Background: Rupture of a splenic artery aneurysm (SAA) is a rare and often life-threatening complication of pregnancy. The reported incidence is 0.01% to 10.4%. Maternal and fetal mortality have been reported to be as high as 75% and 95% respectively.

Case Description: A 26-year-old woman, gravida 5 para 3, presented at 32 weeks of gestation with diffuse abdominal pain and several episodes of syncope. An anchor diagnosis of abruption was made secondary to findings on presentation of intrauterine fetal demise and extensive history of substance abuse.

Discussion: This case is an example of anchoring bias despite good outcomes due to misleading prodromal and warning symptoms with initial favorable response to resuscitation.

Conclusion: A ruptured SSA should be considered in the differential of severe and unexplained pain in the left upper quadrant in pregnancy. A high degree of suspicion is required to make this diagnosis.

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1. Background

Rupture of a splenic artery aneurysm (SAA) is a rare and often life-threatening complication of pregnancy. The reported incidence is 0.01% to 10.4% [1]. Maternal and fetal mortality have been reported to be as high as 75% and 95% respectively [1,2]. Presenting symptoms vary widely and include diffuse abdominal pain, especially in the left upper quadrant, syncope, shortness of breath, or nausea and vomiting. This is known to occur most frequently in the third trimester [3]. Typically, this is a catastrophic event. However, 20–25% of patients have an initial small contained rupture which responds to intravenous fluid resuscitation that precedes complete circulatory collapse [3]. This is called the double-rupture phenomenon. The sudden onset of abdominal pain and hypotension is explained by rupture of the splenic artery into the lesser omental sac, which is then tamponade by the omentum or blood clots. Typically, within 48 h of this latent period there will be acute circulatory failure due to building tension in the lesser sac causing eventual free rupture into the peritoneum [3]. The double-rupture phenomenon is believed to increase chances of survival, giving time for diagnosis and presentation to emergency department or labor and delivery unit [2].

True splenic artery aneurysms in general are commonly associated with conditions of increased blood flow such as pregnancy (particularly multiparity), arteriovenous malformations and portal hypertension. In pregnancy, predisposing factors include the hormonal changes of

increased levels of estrogen, progesterone and relaxin, combined with portal hypertension and increased cardiovascular output [1,3]. Other risk factors associated with SSA include congenital connective tissue disorders, advancing age, arterial hypertension, arteriosclerosis, infection or inflammation, trauma and substance abuse [2].

2. Case Description

A 26-year-old woman, gravida 5 para 3, presented originally at 32 weeks of gestation to hospital due to diffuse abdominal pain and several episodes of syncope. At that hospital she was reported to be hypotensive, with tachycardia, and anemic to 5.9 g/dl. A bedside ultrasound scan indicated intrauterine fetal demise (IUFD). She was transferred to a tertiary-care obstetrical floor for further stabilization and management of her IUFD.

Her medical history was significant for intravenous drug abuse, hepatitis C, tobacco usage and anxiety, with urine drug screen on admission positive for amphetamines. Her presenting blood pressure at the tertiary-care center was 136/60 mmHg and her heart rate was 131 bpm upon admission. The fetus was confirmed demised in cephalic presentation on bedside ultrasound scan. No active vaginal bleeding was noted on examination. On admission she complained of significant diffuse abdominal pain that was worst in the left upper quadrant. Intravenous fluid resuscitation was started and 2 units packed red blood cells were administered. She responded appropriately with her hemoglobin rising to 7.9 g/dl and her tachycardia improved. After the patient was stabilized the decision was made to proceed with induction of labor.

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During induction of labor the patient's tachycardia spiked to 180 bpm and she was noted to be in severe pain from her contractions. An epidural was placed and her tachycardia improved to 110–120 s bmp. Several hours later the patient became fully dilated with fetal head at +3 station. She was instructed to push, and she delivered the demised fetus followed by placenta without complication. There was no evidence of abruption with delivery or with examination of the placenta. Estimated blood loss from delivery was 100 ml. Her uterus was found to be firm and her abdomen was noted to be distended. The patient desired time to hold her baby and delivery personnel exited delivery room. Several minutes later, the emergency bell was pulled. The patient was pale, minimally responsive and hypotensive. Anesthesiology was called to the room and phenylephrine was administered but the patient remained hypotensive. Her stomach was noted to be increasingly distended. Fast scan was performed and was positive for free fluid around the liver. She was immediately brought to the operating room for emergency midline laparotomy.

Upon abdominal entry, a large amount of blood was evacuated, approximately 12 L in total at conclusion of case. Massive transfusion protocol was initiated during the course of her surgery and included 14 units pRBCs, 8 units fresh frozen plasma, 2 units platelets, 1 unit cryoglobulin and 1 g tranexamic acid. Upon entry, the pelvis was inspected and there was no active bleeding of gynecologic origin. The source was noted to be omental in origin and trauma surgery was called into the operating room. A tear in the splenic hilum as well as omental vasculature injury were identified. The patient ultimately underwent splenectomy, oversew of tail of pancreas, oversew of omental bleeding and drain placement. She was transferred to the intensive care unit. The rest of her course was uncomplicated, and she was discharged home in stable condition 4 days later.

3. Discussion

This case is an example of anchoring bias, as it is more common for an obstetrician to encounter placental abruption rather than splenic artery rupture, considering the patient's presentation of abdominal pain, hypotension, fetal demise and positive amphetamines on urine drug screen. The diagnosis of severe abdominal pain in pregnancy may be attributed to several sources, including placenta abruption, uterine rupture, ruptured ectopic, appendicitis, cholecystitis, or perforation of a peptic ulcer. Splenic artery rupture is often misdiagnosed as other much more common obstetrical emergencies, given its rarity. Therefore, a high degree of clinical suspicion should be practiced and diagnoses should be revisited even if patient responds to resuscitation well. Limited case studies in the literature have concluded the involvement of

general surgery or vascular surgery improves outcomes, with decreased mortality, and therefore should be included in cases of SAA whenever possible [2].

4. Conclusions

While routine screening of all pregnant women for splenic artery aneurysms is not cost-effective or practical due to very low incidence, screening high-risk patients could be beneficial. In this case study, the patient had several risk factors. A ruptured SSA should be considered in the differential diagnosis of severe and unexplained pain in the left upper quadrant in pregnancy. A high degree of suspicion is required to make this diagnosis.

Contributors

Jessie Huff is the sole author of this case report.

Conflict of Interest

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