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Case Report of Rupture of Splenic Artery Aneurysm during Pregnancy with Poor Outcomes

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Authors' contributions

This work was carried out in collaboration between all authors. Authors RA, AS, AS and KHN wrote the manuscript. Author JSL provided critical feedback of manuscript and help prepare submission. Author RAD performed the editing and clinical input on manuscript. All authors read and approved the final manuscript.

Case Study

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ABSTRACT

Aims: To highlight the importance of considering non-obstetrical etiologies for acute abdominal pain in gravid patients with risk factors for vasculopathies including diabetes and hypertension. Specifically, we report a tragic case of splenic artery aneurysm (SAA) rupture during the third trimester in a diabetic patient resulting in maternal-fetal mortality. Traumatic vascular events during pregnancy may be associated with a high rate of maternal and fetal morbidity or mortality. Therefore early and rapid intervention is critical to the obstetrical outcome.

Presentation of Case: A 35 year old multiparous hypertensive diabetic patient presented with acute abdominal pain at 33 weeks of gestation. The presumptive diagnosis was concealed placental abruption with diabetic ketoacidosis. Although non-obstetrical diagnoses were not initially considered, postmortem analysis revealed a non-obstetrical etiology of SAA rupture with catastrophic consequences for the patient and fetus.

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Discussion: The likelihood of aneurysm rupture of SAA is heightened due to vasculopathic medical comorbidities such as hypertension and diabetes. The vascular congestion of pregnancy increases flow through arteries, leading to increased likelihood of aneurysm rupture without warning or preceding symptoms. Preconception screening and imaging modalities to confirm splenic artery aneurysms and elective repair are also discussed. Early consideration and accurate identification of SAA rupture is critical to saving the lives of both mother and fetus.

Conclusion: SAA rupture, in the differential diagnosis of acute abdominal pain in pregnancy, should be considered more likely in multiparous patients and in the presence of comorbidities such as diabetes and hypertension.

Keywords: Splenic artery aneurysm rupture; diabetes; hypertension; pregnancy.

ABBREVIATION

SAA: splenic artery aneurysm.

1. INTRODUCTION

The physiologic changes associated with pregnancy, including increased vascular load and changes in arterial composition, often unmask previously undiagnosed pathology and heighten the risk of vascular anomalies such as aneurysms. Several case reports describe patients with previously undiagnosed splenic artery aneurysms (SAA) manifesting during pregnancy with subsequent rupture that results in death [1-5]. Current estimates of maternal and fetal mortality from rupture during pregnancy are 75% [6] and 95%, respectively, which is markedly increased compared to the 25% mortality among non-pregnant patients [7]. Differential diagnoses of acute abdominal pain in the gravid patient is broad including placental abruption, uterine rupture, perforated ulcer, or rupture of other arterial aneurysms [8].This report underscores the importance of early and accurate diagnosis of SAA given the extremely high mortality associated with rupture [5]. Indeed, the mortality rate of SAA rupture approximates that for amniotic fluid embolism, one of the leading causes of maternal death worldwide [9,10].

2. PRESENTATION OF CASE

A 35 year-old woman, gravida 5, para 3-0-1-3, at 33 weeks' gestation presented to an outside facility complaining of decreased fetal movement and the acute onset of abdominal pain. Her medical history was significant for chronic hypertension and class B diabetes mellitus. While the patient was noted to be alert, oriented, and appropriately responsive, she was also observed to be diaphoretic. Her gravid abdomen was remarkably distended with fundal tenderness upon examination. There were no overt signs of vaginal bleeding and vital signs demonstrated a persistently elevated systolic and diastolic blood pressure of 160/100mmHg. The patient's random finger stick blood sugar was 273mg/dL. Doppler evaluation for fetal cardiac activity was notable for absent fetal heart tones. Given the patient's history of diabetes, the preliminary diagnosis was diabetic ketoacidosis with intrauterine fetal demise. The patient was admitted to the outside facility by the attending obstetrician and insulin with supportive therapy was initiated.

Per hospital transfer report, following an interval of four hours the patient continued to have abdominal discomfort and started to become hypotensive. Her recorded blood pressure fell to 70/40mmHg, following which she went into cardiac arrest. Standard cardio resuscitative measures were initiated, and cardio version with defibrillation achieved normal sinus rhythm. At this point, in order to protect her airway and provide respiratory support, the patient was intubated and placed on a ventilator. She then received the inotropes dopamine, and phenylephrine for vascular support per protocol. The patient's baseline presenting hemoglobin of 11.0g/dL rapidly decreased to 6.8g/dL, requiring resuscitation with multiple units of packed red blood cells (pRBC) and fresh frozen plasma (FFP). At this juncture, the outside facility care team assessed that the patient was adequately stabilized for transfer to our tertiary care center. The patient was transferred by helicopter to our center with a presumptive diagnosis of placental abruption with fetal demise. During transport, the accompanying healthcare team noted worsening hemodynamic instability and experienced difficulty maintaining the patient's vascular pressure. Throughout transport, fluid resuscitation was continued with pressor support.

Upon arrival at our facility, the patient was met at the emergency entrance by a critical care team comprised of obstetrician/gynecologists and trauma surgeons. Given the critical condition of the patient, immediate management was initiated in the emergency bay. Assessment upon arrival demonstrated that the patient was unresponsive to verbal or painful stimuli. She remained hemodynamically unstable with poor vascular parameters of hypotension and bradycardia with cool extremities. Neurologic evaluation revealed pupils to be fixed and dilated. The gravid abdomen remained markedly distended without any vaginal bleeding upon pelvic examination. The cervix was noted to be dilated at 6cm. The working diagnosis remained acute placental abruption with intrauterine fetal demise. Efforts to save the patient's life continued with aggressive fluid resuscitation and cadiopulmonary resuscitation. In addition to intravenous crystalloids, at presentation the patient received an additional 6 units of pRBCs, 2 units of FFP, and 1 unit of cryoprecipitate with pressor support. A total of 10mg epinephrine, 1 mg lidocaine, 2 ampules calcium chloride, and 5 ampules bicarbonate were administered. Intrauterine fetal demise was confirmed by Doppler and ultrasonography. An emergent laparotomy was considered but deferred due to the patient's hemodynamic instability. Given the presumptive diagnosis of concealed placental abruption, continued attempts were made to deliver the fetus vaginally to help stabilize the patient. Oxytocin was administered and vacuum extraction attempted but was unsuccessful. Serial examination of the abdomen demonstrated progressively increasing 'fundal height', which reaffirmed the suspicion of placental abruption.

The patient again went into cardiac arrest and aggressive cardiopulmonary resuscitative efforts resumed with pressor support using norepinephrine, epinephrine, dopamine, and phenylephrine. Unfortunately, she never regained sinus rhythm. Following two hours of resuscitative attempts, the patient was pronounced dead. The presumed cause of death was cardiopulmonary arrest secondary to hemorrhagic shock as a consequence of acute placental abruption, superimposed on diabetic ketoacidosis. Postmortem examination instead revealed rupture of a splenic artery aneurysm, retroperitoneal hemorrhage, and hemoperitoneum of 2900mL clotted blood. The spleen was congested with focal disruption of the splenic artery near the neck of the pancreas. A 33-week, normally formed, intrauterine pregnancy was found with fetal demise due to maternal exsanguination and hypotension. The placenta was found attached to the endometrium and easily stripped from the uterine wall.

3. DISCUSSION

Splenic artery aneurysm (SAA) is the most common type of visceral artery aneurysm and represents over 50% of all reported aneurysms. The incidence of SAA in gravid patients is less than 1% [11], however, of the 400 reported cases of ruptured SAA, half occurred during pregnancy [6,12]. A majority of the ruptures occur during the late third trimester [5,11,13,16] likely due to physiologic and hormonal effects of pregnancy that exacerbate congenitally-present vascular defects, including a high flow rate through the splenic artery secondary to increased plasma volume, distal compression of the aorta and iliac arteries by the gravid uterus, a state of relative portal congestion, and a progressive weakening of the arterial media [2,8,17,18]. Other conditions associated with increased risk of SAA include essential hypertension, portal hypertension, diabetes, splenomegaly, medial fibrodysplasia, intracranial aneurysm, polyarteritis nodosa, liver transplantation, autoimmune diseases such as dermatomyositis, and autosomal dominant polycystic kidney disease. Additionally, atherosclerotic disease is strongly associated with both splenic and extra-splenic artery aneurysms [6]. In gravid cases, fetal mortality rate is almost certain at over 90%, and maternal mortality closer to 70-75% [3,7,12,14]. This is a stark contrast to the 25% mortality rate of SAA in non-pregnant patients [7]. Pregnancy-related factors become more problematic with higher parity [19], with the mean parity at 4.5 in women with SAA rupture [20].

Early and accurate clinical diagnosis remains the most important strategy for management of patients with SAA. As in the case presented above, a minority of patients present with sudden or intermittent acute abdominal pain, sometimes located in the left upper quadrant, which rapidly progresses to hypovolemic shock [14,21]. There may be a period of temporary stabilization subsequent to initial rupture as blood fills the lesser sac and is contained by omentum or the omental foramen. This is followed by massive hemorrhage, resulting in maternal shock, fetal demise, and ultimately maternal demise. This type of “double rupture,” occurring in two stages, has been reported [22-24] and should be taken into account in differential diagnosis. The second stage of hemorrhaging may have resumed during transfer of the presented patient to our facility. Taking this case into consideration, a high index of suspicion is warranted in patients who present with any abdominal symptoms and a concurrent history of chronic hypertension, autoimmune disease, diabetes, multiparity, or other factors suggesting subclinical vasculopathy.

Unfortunately, SAA is generally asymptomatic and is usually discovered post-rupture in the emergency department. However, the diagnosis is sometimes made during imaging studies for unrelated abdominal conditions [6]. Due to the overall rarity of SAA in gravid subjects and the cost of widespread screening, patients with preexisting conditions predisposing them to SAA rupture would benefit from SAA screening [25]. Hypertensive, diabetic patients in particular may benefit from preconception screening for vasculopathies, as angiographic screening prior to portal hypertensive treatment has successfully identified SAA risk [26]. The preferred diagnostic imaging technique for SAA detections is angiography. Of the investigative imaging modalities that have been employed in the setting of a hemodynamically stable patient, ultrasonography has also demonstrated utility in identifying massive hemoperitoneum and clotting [1]. The high spatial resolution of computed tomography (CT) scans and ability to construct reformations can also improve evaluation of putative SAA [27,28]. Furthermore, in emergent cases such as that of our patient, a CT scan may help rapidly and accurately identify the pathology and result in improved outcomes.

Detection and treatment of SAA prior to rupture has great life-saving potential for both

mother and fetus. A splenic artery aneurysm larger than 2cm in size detected in a pregnant woman or woman of childbearing age or in a pregnant patient should be treated electively because of the increased risk of rupture associated with pregnancy, in contrast to low mortality rate of 0.5%-1.3% associated with the elective repair [6,11,29]. Surgical options include conservative measures such as laparoscopic splenic artery aneurysmectomy and embolization. With laparotomy, treatment may involve splenic artery ligation with or without aneurysmectomy and splenectomy [30]. Alternatively, when arterial anatomy permits, stent grafts preserve end-organ blood supply and avoid perioperative risks for patients with portal hypertension [31,32].

4. CONCLUSION

In summary, we stress that misdiagnosis and delay in treating SAA rupture in the gravid patient with acute abdominal pain will likely contribute to maternal and fetal mortality. SAA in pregnancy is uncommon enough that healthcare providers rarely identify a case prior to rupture. Early and immediate surgical intervention is crucial to preventing negative outcomes for both mother and fetus [11,13,14]. Laparotomy and Cesarean delivery of the viable infant followed by aneurysm repair is the optimal management. If the infant is pre-viable, immediate laparotomy, repair of the aneurysm or splenectomy without delivery of the infant may be an option depending on the outcome of the surgical intervention and maternal condition [21].

Unfortunately, the differential diagnosis in this case included complications associated with both pregnancy and diabetes that seemed reasonable to explain the patient's presentation. In patients with predisposing factors such as those listed above, heightened suspicion of SAA rupture is warranted in the differential diagnosis of abdominal pain hemodynamic instability and sudden cardiovascular collapse.

Although placental abruption, uterine rupture and amniotic fluid embolization are included in the differential diagnosis, it is important to consider visceral artery aneurysm, such as SAA, rupture in pregnant patients with high-risk factors for cardiovascular and vasculopathic diseases. Furthermore, patients at risk for vasculopathic disease should receive extensive counseling on exacerbation of pregnancy associated risks as it pertains to their specific comorbidities.

CONSENT

Not applicable for anonymized case report

ETHICAL APPROVAL

Not applicable.

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COMPETING INTERESTS

Authors have declared that no competing interests exist.

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