Rupture of internal pudendal artery aneurysm following spontaneous vaginal delivery: An uncommon cause of post-partum bleeding

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Abstract

Pregnancy-related rupture of an arterial aneurysm is an unusual occurrence associated with increased risk of morbidity and mortality. Various pregnancy-related physiologic changes appear to make pregnancy a high-risk situation for rupture of either preexisting arterial aneurysms or those that develop throughout the course of pregnancy. Splenic artery aneurysms are the most common (60%), followed by hepatic (20%), superior mesenteric (5.9%), celiac (4%), ovarian, uterine, and renal (<2%) artery. Even rarer are aneurysms involving the internal iliac artery and its branches, to which there is only one published case report. In this report, we present a case of a 34-year-old pregnant gravida₁ para₀ who, following a normal vaginal delivery, had a severe rupture of the right internal pudendal artery and subsequently developed a massive hematoma which ultimately required embolization treatment.

Keywords

postpartum hemorrhage, hematoma, internal pudendal artery, aneurysm rupture, pregnancy

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Introduction

Pregnancy-related rupture of an arterial aneurysm is an unusual occurrence associated with increased risk of morbidity and mortality.

Various pregnancy-related physiologic changes appear to make pregnancy a high-risk situation for rupture of either preexisting arterial aneurysms or those that develop throughout the course of pregnancy.¹ Splenic artery aneurysms are the most common (60%), followed by hepatic (20%), superior mesenteric (5.9%), celiac (4%), ovarian, uterine, and renal (<2%) artery aneurysms.^{2–5} Even rarer are aneurysms involving the internal iliac artery and its branches, to which there is only one published case report.⁶ The internal pudendal artery branches off the anterior trunk of the internal iliac artery and is the main supplier of blood to the external genitalia and perineum via its perineal, labial, and hemorrhoidal branches. In this report, we present a case of a 34-year-old pregnant gravida₁ para₀ who, following a normal vaginal delivery, had a severe rupture of the right internal pudendal artery and subsequently developed a massive hematoma which ultimately required embolization treatment.

Case

A 34-year-old gravida₁ para₀ at 39 weeks gestational age presented to the labor and delivery emergency department with complaints of loss of fluid. She denied any past medical, surgical, or family history, vaginal bleeding, decreased fetal movement, headache, abdominal pain, nausea, vomiting, or visual disturbances. The patient had routine prenatal care and her prenatal course had been uncomplicated thus far. Following admission, she progressed through labor appropriately without the aid of induction agents or artificial rupture of membranes and spontaneously delivered one live, viable female infant with

Figure 1. Pelvic sonogram. Pelvic sonogram revealed an enlarged heterogeneous uterus and (blue *) compressed and displaced bladder in the midline pelvis and (blue *). The compressing hematoma is identified by the red *.

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Figure 2. CT of abdomen and pelvis with contrast (a–d). CT scan revealed a large soft tissue density within the right side of the pelvis extending inferiorly, displacing the uterus and urinary bladder to the left (arrows), consistent with large pelvic hematoma (*) with dimensions of 13 cm $\times$ 8.5 cm transversely. Additionally, poor definition of the soft tissue planes was seen in the pelvis with a hypodensity extending from the anterior mid pelvis into the pelvic cul-de-sac reflecting hemoperitoneum. Hyperdense fluid was also seen in the perihepatic space, along the right paracolic gutter and less prominently along the left paracolic gutter also consistent with hemoperitoneum.

APGAR scores of 9 and 9. Estimated blood loss was 400 mL and the total duration of labor was 5 h. The newborn weighed 3.7 kg. Three hours postpartum, she complained of dizziness, lower abdominal pain, and near-syncope. A physical exam was performed and her vital signs revealed hypotension with blood pressures of 62/38 mmHg, tachycardia of 139 beats/min, and was found to be afebrile with a temperature of 36.8°C with associated skin pallor and lethargy. A vaginal exam was performed and revealed no significant bleeding apart from that expected postpartum. Her hemoglobin and hematocrit were noted to fall from 11.9 g/dL and 34.4% to 6.6 g/dL and 19.4%, respectively. She was given two units of packed red blood cells, and sent for pelvic sonogram and a CT of the abdomen and pelvis with contrast.

Pelvic sonogram revealed an enlarged heterogeneous uterus and cystic structure in the midline pelvis representing a compressed and displaced bladder, but was limited due to the patient's postpartum status, abdominal pain, and tenderness (Figure 1).

CT scan (Figure 2) revealed a large soft tissue density within the right side of the pelvis extending inferiorly, displacing the uterus and urinary bladder to the left, consistent with large pelvic hematoma

with dimensions of 13 cm $\times$ 8.5 cm transversely. Additionally, poor definition of the soft tissue planes was seen in the pelvis with a hypodensity extending from the anterior mid pelvis into the pelvic cul-de-sac reflecting hemoperitoneum. Hyperdense fluid was also seen in the perihepatic space, along the right paracolic gutter and less prominently along the left paracolic gutter also consistent with hemoperitoneum.

Following these imaging results, interventional radiology was consulted and the decision was made to perform a pelvic angiogram and possible embolization. A contained extravasation was identified to the right of the pubic symphysis on the initial nonselective pelvic arteriogram. The subsequent right internal iliac angiogram demonstrated bleeding arising from the distal aspect of an anterior division of the internal iliac artery, the right internal pudendal artery. There was a deviation of the artery due to the formed hematoma. The vessel was successfully catheterized and the subsequent angiogram confirmed that the bleed did arise from this branch, likely due to the rupture of an aneurysm therein (Figure 3). This area was treated with coil embolization and there was no angiographic evidence of continued bleeding. The patient was stabilized postembolization, had an otherwise normal

Figure 3. Pelvic angiogram. A contained extravasation was identified to the right of the pubic symphysis on the initial non-selective pelvic arteriogram. The subsequent right internal iliac angiogram demonstrated bleeding arising from the distal aspect of an anterior division of the internal iliac artery, the right internal pudendal artery(*). There was a deviation of the artery due to the formed hematoma.

hospital course, and was discharged in stable condition on postpartum day 3.

Discussion

Pregnancy-related rupture of an arterial aneurysm, of any location, is an unusual occurrence associated with increased risk of morbidity and mortality. Various pregnancy-related physiologic changes appear to make pregnancy a high-risk situation for rupture of either preexisting arterial aneurysms or those that develop throughout the course of pregnancy. As demonstrated by this case, these changes in normal pregnancy are sufficient to complicate an otherwise normally healthy pregnant patient. The patient in this case had no comorbidities, had a normal body mass index of 22, and was able to proceed through labor without any medical assistance apart from the actual delivery itself.

Most often, aneurysms and their subsequent rupture involve an underlying pathology that is further exacerbated by the physiological alterations of normal pregnancy. These include, but are not limited to, congenital abnormalities, inherited vascular disease, arterial degeneration, inflammatory process, and trauma. Conversely, it is possible that these conditions could have existed in a very mild state in this patient but did not become apparent until exacerbated by the changes of pregnancy. It appears that the changes during normal pregnancy in both blood flow dynamics and hormonal concentrations are important in causing the arterial changes predisposing to aneurysm formation which include hyperplasia of the arterial intima and distortion of the composition and organization of arterial media.⁶ In this specific patient, it is likely that these arterial changes began in the first trimester and worsened throughout her pregnancy leading to the development of

an aneurysm in the internal pudendal artery. Given that she had no past medical history of congenital diseases or aneurysms in other locations throughout the body, the possibility of the patient having a pre-existing pudendal artery aneurysm before her pregnancy is less likely, although cannot be completely excluded.

Further increases in blood pressure and resultant stress on the already compromised blood vessel during the labor process are likely to have caused the previously formed aneurysm therein to rupture. The rupture of the internal pudendal artery aneurysm was not likely to have been caused from the direct pressure effect of the fetal head because of its deeper location in the pelvis as evidenced by the pelvic angiogram (Figure 3): the aneurysm would need to have been sufficiently superficial in the pelvis for enough pressure to be exerted on it from the descending fetal head. If this were to occur, superficial bleeding would be observed and would be more likely to manifest as a vulvar hematoma or frank bleeding from the vagina or perineum rather than the internal bleeding observed in this patient.

A literature search was performed using MEDLINE, PubMed, and Google Scholar for the period from 1930 to 2015 to assess the current literature regarding aneurysms in pregnancy. The incidence of internal pudendal artery aneurysms or related conditions of this artery location such as dissection is not well described, and there have been no case reports of this specific situation to the best of our knowledge, thus to comment about a given population's risk of developing aneurysms and ruptures of such is limited. Aneurysms, in other locations, however, are well described and are themselves rarely seen with an incidence of 0.02 to 0.1% for splenic artery aneurysms and far less for hepatic, superior mesenteric celiac, for uterine, for ovarian, and for renal artery aneurysms respectively.⁷

The prognosis of ruptured aneurysms largely depends on the site of aneurysms and how quickly they are identified and treated. In situations during pregnancy, this prognosis is also dependent on when these aneurysms rupture: there will be significantly more risk for an intrapartum patient versus a postpartum patient given the need to provide adequate circulation and homeostasis to two organisms versus one. Because there is limited literature concerning right pudendal artery aneurysms and rupture in pregnancy, categorical conclusions of this situation's prognosis cannot be made, but as evidenced by the management in this case report, it can be concluded that this condition, although serious, can be managed effectively with prompt identification, evaluation, and treatment: this patient's condition was promptly identified with the proper clinical reasoning and physical examination, the appropriate radiological tests were ordered to elucidate the problem, and the definitive treatment of arterial embolization was conducted, thus this patient was able to be discharged home in stable condition with no sequelae other than acute blood loss, which was managed medically with the appropriate amount of packed red blood cells and outpatient oral ferrous sulfate.

Conclusion

The formation and rupture of an internal pudendal artery aneurysm is an exceedingly rare and thus far undocumented situation to our knowledge. Management approaches and risk stratification is based upon the existing literature that concerns other visceral artery aneurysms and was successfully employed in this case to manage this patient's cause of postpartum bleeding. This case highlights the necessity to promptly identify the correct cause of hemorrhage in women postpartum to avoid a potentially catastrophic outcome. With prompt identification and treatment, cases of internal pudendal artery aneurysm rupture can be quickly and effectively managed resulting in a patient with no serious sequelae.

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Guarantor

DM

Contributorship

The manuscript's design and conception was performed by Drs. FX Martingano and Ruggiero-DeCarlo; the manuscript was written by Dr. Daniel Martingano; the images were prepared by Dr. Daniel Martingano; the manuscript was proofread and edited by Drs. Daniel Martingano, FX Martingano, and Ruggiero-DeCarlo.

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