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Postpartum Rupture of Right Internal Iliac Artery Aneurysm Case Report from a Tertiary Hospital in Kenya

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Abstract

Iliac artery aneurysm is an extremely rare vascular disorder occurring in about 0.03% of the population. Sudden hemodynamic changes along with the radiological findings of free abdominal fluid warrants for further surgical intervention, as the heavily gravid uterus during pregnancy that compresses on the aneurysm undergoes a sudden reduction of size postpartum and the associated changes in pressure gradient on the artery post-delivery may cause sudden rupture of the iliac artery aneurysm. Rupture of the internal iliac artery is a rare complication in pregnancy and in postpartum but should be considered as a differential diagnosis of abdominal pain post-partum. We are presenting a 29-year-old para 3+0 who delivered via SVD with normal antenatal, intrapartum and immediate postpartum vitals. On day 1 Post-Partum the patient went into cardiovascular arrest secondary to hypovolemic shock, the postmortem revealed ruptured right internal iliac artery aneurysm.

Keywords: Ruptured internal right iliac artery; Aneurysm; Post-mortem

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Introduction

Iliac artery aneurysm is an extremely rare vascular disorder occurring in about 0.03% of the population [1]. Approximately 30% to 50% are bilateral [2,3]. The iliac arteries are normally situated deep in the pelvis and, it divides into the internal and external iliac arteries, hence the diagnosis of aneurysms of these vessels are difficult. They are detected when they are enlarged or ruptured or as an incidental finding during imaging studies for other abdominal pelvic disorders.

Spontaneous rupture of the internal iliac artery in pregnancy or postpartum is rare and has a lethal outcome, it was first described in 1778 [4]. Till current date, the exact etiology of it is unclear and it is believed to be multifactorial.

Some hypothesize that disorders such as endometriosis, congenital diseases of connective tissue (such as Ehlers-Danlos syndrome type IV), and congenital malformations of blood vessels such as aneurysm, including aneurysm of the internal iliac artery, may result in arterial rupture in pregnancy, postpartum or in non-pregnant patients [5-10].

Incidences of aneurysm maybe found in 0.1% of general population, with aneurysm of the iliac artery accounting for 1% of these cases. Common iliac artery is involved in 85%, internal iliac artery in 10% and external iliac artery in 1% of cases [11-13].

High blood pressure in the uterine or ovarian arteries during muscle activity and strain during pregnancy or delivery is a risk factor for rupture of the artery [14,15]. Differential diagnosis that may contribute to rupture of aneurysm maybe premature placenta abruption, spontaneous uterus rupture, and spleen rupture i.e., aneurysm of splenic arteries.

Nevertheless, nonspecific symptoms of artery rupture may result in unrecognizable severity and urgency of the problem, which leads to life threatening circumstances to the mother and the child [16]. The mortality of mothers with spontaneous rupture of uterus artery has dramatically decreased from 49% to 3.6% owing

to improvement of intensive intraoperative and postoperative treatment [17]. Early diagnosis, radiological studies and intensive surgical-anesthetic treatment helps in prevention of mortality.

We present a case of incidental postmortem finding of a ruptured right internal iliac aneurysm in a postpartum woman.

Case Report

A 29-year-old Para 2 gravida 3 was admitted into a Tertiary Hospital with complaints of lower abdominal pains for 1 day at 43 weeks gestation by her LNMP, She perceived normal fetal movements. She had no history of any chronic medical conditions or prior history of surgeries. She attended antenatal clinics throughout her pregnancy, her antenatal profile was unremarkable. She had history of postpartum Hemorrhage in her previous two pregnancies. On admission, the patient was in Fair General Condition with vitals within normal ranges (BP - 133/82mmHg, Pulse - 106 beats per minute, Respiratory rate - 20 per minute, Temperature - 35.7, SpO_2 - 100%). There was also no pallor on general examination. Antenatally her HB was 12.8 g/dl. Hemodynamically, the patient was stable. On abdominal examination- there was no tenderness on palpation, the fundal height was term, fetus in cephalic presentation with a longitudinal lie, fetal heart rate was 140 beats per minute. On vaginal examination- The cervix was 5cm dilated, thick and centrally positioned. The other systems were essentially normal. She was admitted with a Diagnosis of Active phase of labor. The Patient progressed well according to strict partograph monitoring and successfully delivered a Live Male Infant, 3.6 kg with a good Apgar score. Post-partum vitals were within normal ranges. Two hours post-delivery, patient was taken to the normal postpartum ward for 12 hours monitoring. On the following day, the patient was fit for discharge, as she was leaving the ward she started complaining of dizziness and she collapsed, on examination the patient was very pale, sweaty with cold extremities. Vitals were 86/ 60, pulse- 146 beats per minute, random blood sugar - 7.0 mmol/L. Abdominal examinations revealed right iliac fossa tenderness, the uterine fundus was not well appreciated. Vaginal examination was normal. An impression of Hypovolemic Shock secondary to Primary Post-Partum Hemorrhage was made. She was resuscitated with intravenous fluids, one unit of whole blood, and a Pelvic ultrasound was done immediately which revealed a right iliac fossa hematoma (measuring 210 cm^3) **(Figure 1),** with minimal ascites noted (hepato-renal space) **(Figure 2)** Full hemogram showed Hemoglobin of 12.8 g/dL, Platelets of 273 × 10^3/ uL. CT scan of the abdomen and pelvic was suggested for further evaluation but patient was unstable to be taken for the scan. Decision to perform exploration laparotomy was made, however patient started gasping upon arrival to the operation theater. Cardiopulmonary resuscitation as per National ACLS protocol was done, however resuscitation was unsuccessful. Clinical postmortem was performed, more than 2L of hemoperitoneum found in the abdominal cavity. Large amount of blood clot seen in the retroperitoneum space spreading all the way to the diaphragm **(Figure 3).** There was a soft tissue hematoma on the board ligament with normal postpartum uterus **(Figures 4 and 5).** The aorta and left illiac artery was normal but found a ruptured Right internal iliac artery aneurysm. All other internal organs were normal.

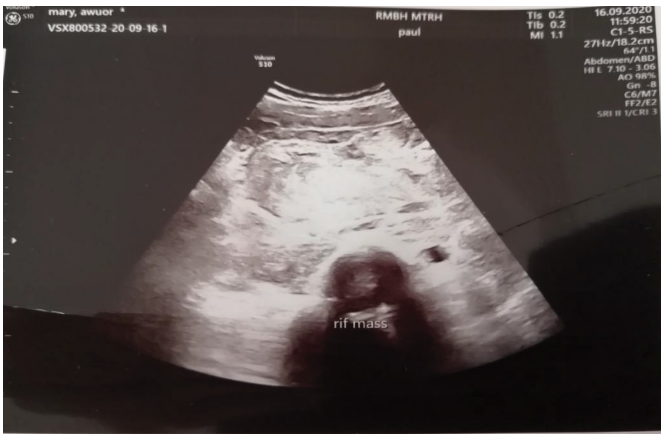

Figure 1 Ultrasound finding of right iliac mass.

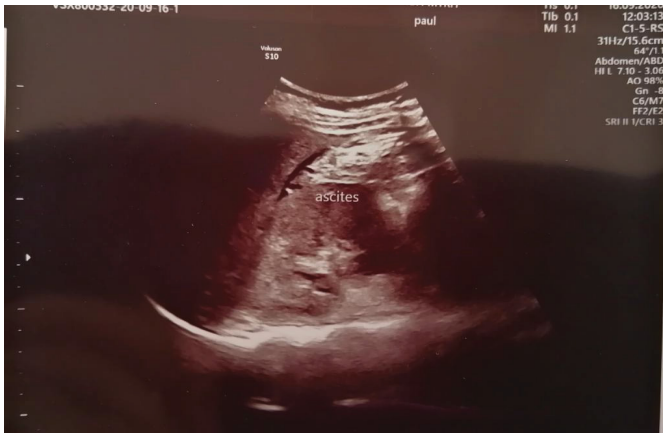

Figure 2 Minimal ascites noted in the hepato-renal space.

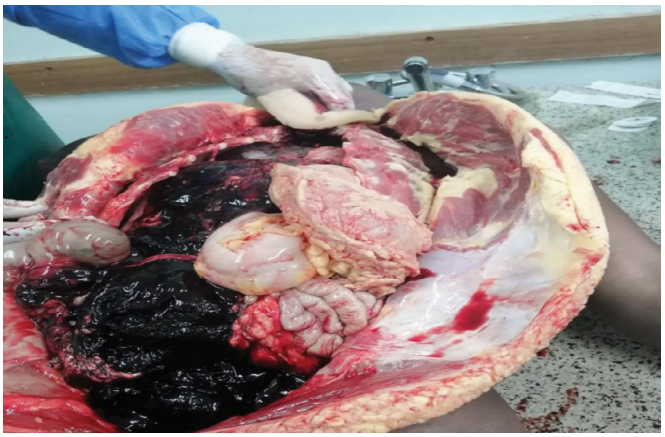

Figure 3 Large amount of blood clots in the retroperitoneal space.

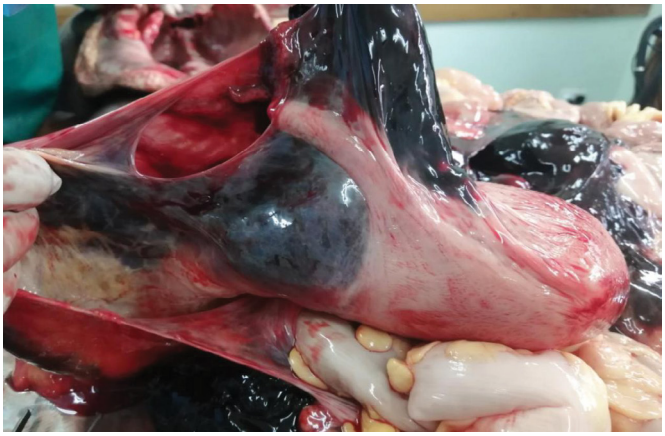

Figure 4 Soft tissue hematoma on the right board ligament.

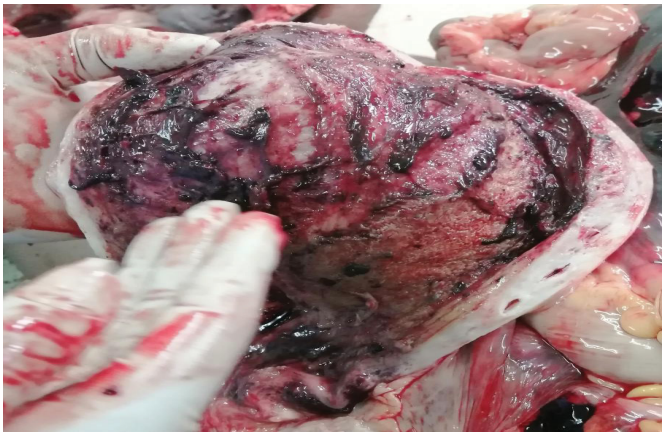

Figure 5 Normal postpartum uterus.

Discussion

In this patient, the exact etiology of the spontaneous arterial rupture was not established. We believe that there might be an existing arterial pathology, but we were not able to show evidences of any arterial pathology existence with either histological or radiological examination. The pathogenesis of iliac artery aneurysms is multifactorial and similar to that of Abdominal Aorta Aneurysm (AAA). Etiologies that have been reported to contribute to iliac aneurysmal dilatation include atherosclerotic changes, inflammation, wall stress and wall tension, proteolytic degradation of arterial wall tissue, and molecular genetics. Histologically, an iliac aneurysm, similar to an AAA, is characterized by macrophage degradation of the medial elastin lamellar architecture by metalloproteinases. The less common etiologies that may cause Internal iliac Artery Aneurysm (IIAA) include trauma, fibromuscular dysplasia, infection, cystic medial necrosis, collagen vascular diseases and pregnancy [18].

There is an ongoing debate as to whether pregnancy may contribute to the pathogenesis of vascular disease. Cardiovascular changes during pregnancy may enhance the stress on the arterial wall. High levels of female hormones during pregnancy alter the histological structure of the arterial wall, thereby resulting in a predisposition to aneurysmal dilatation. Pregnancy may be an initiator of arterial degeneration, which is additive with multiple pregnancies and changes may be permanent [19].

Aneurysm rupture during pregnancy is often associated with nonspecific symptoms and is life-threatening. The maternal mortality rate for vascular dissection and rupture is 0.74–0.76 per 100,000 live births. It is important to have differential diagnoses in mind as this contributes to the complexity of diagnosing this condition. Differential diagnoses include: uterine rupture, placental abruption, massive pulmonary embolism, cholecystitis, perforated ulcer, and urolithiasis [20].

Diagnosis can be made by computed tomography angiography, MRA and abdominal/transvaginal ultrasound, which is quick, easy and free of ionizing radiation. US is used extensively within obstetrics and gynecology and many clinical situations are diagnosed via a combination of clinical experience and ultrasound skill. A detailed evaluation of the kidneys (size, location, tumors, stasis and concrements), a specific description of fluid collections (free fluid, hematoma, serous, mucinous etc.) and use of color flow mapping provided valuable information for exact diagnosis of an extremely rare condition in this case.

Iliac artery aneurysm has a potential to cause spontaneous rupture and commonly presents with aneurysm of the abdominal aorta, with the incidence of 10%. Isolated finding of internal iliac artery aneurysm is rare, with the incidence of 2% [21-23]. While single iliac artery aneurysm is uncommon, accounting for around 0.03% of cases. It is expressed as enlargement of the artery to double the size without any signs of any other aneurysm in other locations [24-27].

Limited amount of literature concerning internal iliac artery aneurysm or rupture in pregnant or postpartum women was found. However, pre-rupture finding of aneurysm can be managed with elective surgery, thus reducing the mortality rate to less than 10%. Endovascular techniques have become the mainstay in IIAA's treatment. Endovascular treatment of IIAA can be based on a pure endovascular approach or a hybrid one as part of an endovascular aneurysm repair (EVAR) procedure to treat an aortoiliac aneurysm [compromising internal iliac artery (IIA) patency bilaterally], in which a bypass is performed, usually from the distal external iliac artery or the common femoral artery, to revascularize one of the internal iliac arteries [28].

Pure endovascular techniques may be allocated in two main philosophies: with or without IIA flow preservation. While the second option is inarguably easier to perform, it can be associated with ischemic complications, especially in case of bilateral IIA occlusion. The worst of these complications include gluteal/perineal skin necrosis [29,30], colon ischemia [31], and spinal cord ischemia [32]. Despite representing a more benign complication, the incidence of buttock claudication can reach up to 28% in patients with unilateral occlusion of IIA and 42% in patients with bilateral IIA embolization [33].

In order to achieve internal iliac arteries flow preservation, several endovascular techniques were developed, including external iliac to internal iliac artery stent grafts coupled with aorto-uni-

iliac repairs, "sandwich" stent grafts, and iliac branch stent grafts (IBSGs). An intra-peritoneal rupture are commonly associated with sudden death and has extremely poor prognosis compared to Retroperitoneal ruptures.

Acute abdominal pains can be a sign of arterial rupture causing acute bleeding along with hemodynamic changes. Symptoms become more prominent after delivery as the extensively gravid uterus undergoes sudden decrease in size and weight causing sudden arterial rupture due to the pressure changes in the vessels. Any sign of abdominal pain in pregnant or postpartum woman should be taken seriously along with radiological finding of free liquid in the abdominal cavity and immediate surgical intervention is warranted. To increase awareness of this rare condition, we report this case which is an important differential diagnosis to consider in pregnant women with abdominal pain.

Conclusion

Rupture of the internal iliac artery is a rare complication in pregnancy and postpartum. Abdominal pain of unknown origin will be important hallmark in diagnosis of arterial rupture. Pre-rupture Iliac artery aneurysm diagnosed early in pregnancy could reduce mortality and morbidity risk to less 10%. Bleeding becomes more severe intra-operatively when the compressive factor of the gravid uterus is removed. Since rupture of the internal iliac artery is an extremely rare complication in pregnancy or during postpartum, never the less it should be considered as a differential diagnosis of abdominal pain or sudden hemodynamic instability.

Conflict of Interest

The authors declare that there is no conflict of interests.

Acknowledgement

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Informed Consent

Written informed consent was obtained from the patient's relative for publication of this case report and any accompanying images. Permission was sought and granted from Moi Teaching & Referral Hospital (MTRH) prior to publication of this case report.

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