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Undiagnosed Cornual Pregnancy Complicated by Large Uterine Fibroid: A Rare Case Report

Julian Johozua Roberth Maker[1], Julita D.L Nainggolan[1], Alexsandro Ignatius Saverio Lao[1]

[1]Department of Obstetrics and Gynecology, Faculty of Medicine, Pelita Harapan University/Siloam Hospitals Lippo Village, alexsandroignatiussaveriolao@gmail.com

ABSTRACT
A cornual pregnancy is a rare form of ectopic pregnancy with the incidence rate of 2 – 4% of all ectopic pregnancies. It has a higher mortality rate, which is 6 – 7 times higher than other ectopics. Diagnostic approach can be done by transvaginal sonography and serum β-hcg test, however reaching the diagnosis itself is quite challenging. Due to the greater distensibility of myometrium covering interstitial segment of the fallopian tube, undiagnosed cornual pregnancy usually ends with rupture in 8 to 16 weeks of pregnancy, which lead to significant maternal hemorrhage then hypovolemic shock. Reported case is a 35 years old woman who presented to the emergency room with chief complain of severe abdominal pain in a state of hypovolemic shock. She was admitted for emergency exploratory laparotomy. On exploration, we found a point of rupture at the left cornual area from the 20-22 weeks gestational age sized uterus. The 12 weeks old fetus inside the intact gestational sac was found in the abdominal cavity. The patient then underwent cornual wedge resection, myomectomy and bilateral salpingectomy, since the uterus was not viable for anotherpregnancy. All procedures were done successfully, and the patient was discharged in a good condition.

Keywords: *Cornual Pregnancy*, *Ectopic Pregnancy*, *Rupture Ectopic Pregnancy*
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Introduction

Cornual ectopic pregnancy (CEP) by definition refers to the implantation and development of a gestation in a rudimentary horn (rudimentary horn pregnancy, RHP) or in one horn of a septate or bicornuate uterus. However, this term has also been used forpregnancies in the interstitial portion of the fallopian tube (interstitial pregnancy, IP) and those in the lateral angle of the uterine cavity (angular pregnancy, AP). Interstitial (cornual) pregnancy is a rare type of ectopic pregnancy (EP). It is accounting for 2-4% of all tubal pregnancies, with an estimated occurrence of one in every 2500-5000 deliveries. [1] But it is a life-threatening condition, being responsible for nearly 20% of all deaths caused by EP.[2.]

Gestation in the cornual part of the uterus is a rare form of pregnancy that poses a diagnostic and therapeutic challenge. There are some risk factors for EP include a history of previous EP or pelvic inflammatory disease, the use of assisted reproductive technology andtubal surgery. But etiology of cornual pregnancy remains unclear.[2.] Mortality in CEP is reported to be seven times higher than those with other tubal pregnancies and one of the main causes of higher mortality rate is the vascularity of the cornual part of uterine. The cornual part ofuterine itself is supplied by Sampson's artery which is connected to ovarian and uterine arteries. Hence, in case of rupture, there will be massive hemorrhage which may lead to hypovolemic shock. Rupture of cornual pregnancy usually occurs at 8th – 16th week of gestation. This occurs because the cornual part of uterine has thicker myometrium than distal

part of fallopian tube.[3,4.]

The clinical finding of cornual pregnancy depends upon whether or not it is ruptured. In unruptured cases, patients may present with abdominal pain or vaginal bleeding or both and history of repeated abdominal pain at few days interval is also noticed. When the fertilized ovum is implanted well within the cornual/interstitial portion, rupture usually do not occur until 14 to 16 weeks. Ruptured cases usually present with severe abdominal pain with hemodynamic instability caused by significant hemoperitoneum related to the advanced gestational age.[5] Ultrasonographic and quantitative β-HCG lab test are suggested in order to diagnose cornual pregnancy in its early stage. The ultrasonographic criteria for the CEP include: an empty uterine cavity, gestational sac located outside the uterine cavity and myometrial lining less than 5mm around the gestational sac, typically an echogenic line from the endometrial cavity to the corner which is next to the gestational mass is seen.[6.]

Cornual Ectopic Pregnancy can be treated clinically (with methotrexate) or surgically. The criteria for performing treatment with methotrexate are hemodynamic stability, absence of fetal cardiac activity at ultrasound examination and pre-treatment beta -HCG below 5,000 IU/ml. Surgical intervention is necessary in women with hemodynamic instability, suspected or confirmed tubal rupture, heterotopic pregnancy and contraindication or failure to treat with methotrexate.[7.]

Case Summary

Mrs. NH, 35 years old, gravida 2 para 1, presented to the emergency room with a complaint of severe abdominal pain in a state of hypovolemic shock. She was at 12 weeks pregnancy based on her last period (June 25th, 2019). She denied vaginal bleeding. Her last childbirth was 11 years ago, delivered normally assisted by midwife.

She had a history of uterine fibroid since 7 years ago. She did not do a routine checkup for the fibroid until 2 weeks prior to admission. She came to our outpatient department due toa delayed period and found out that she was pregnant at 10 weeks gestational age. The ultrasound examination showed an intrauterine gestational sac in the left lateral of uterus with a single live fetus. CRL was 3,56 cm which was appropriate to 10-11 weeks gestational age (Figure 1). A hypoechoic mass was also noticed in the right lateral of uterus, sized 13.3 x 11.3 x 8.2 cm.

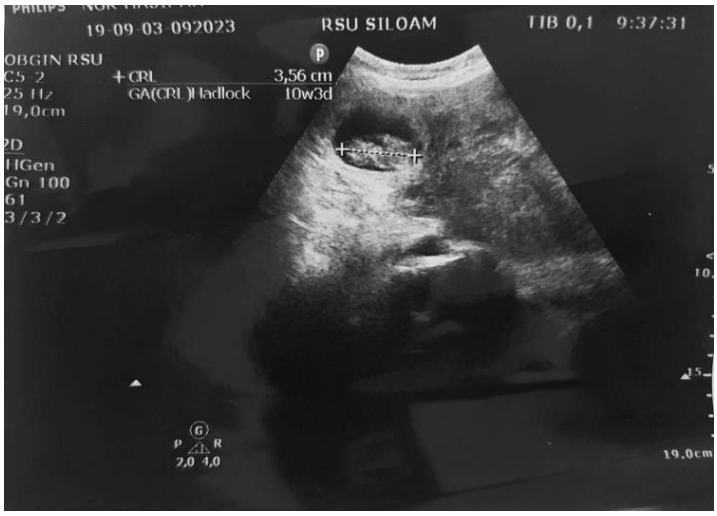

Figure 1

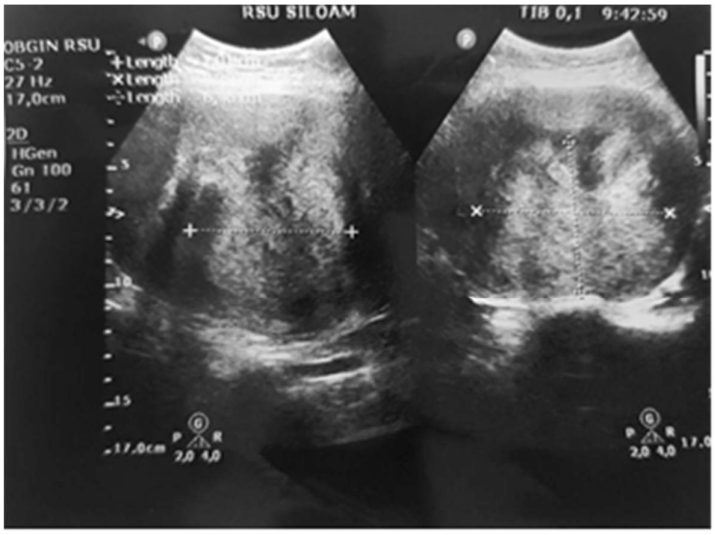

Figure 2

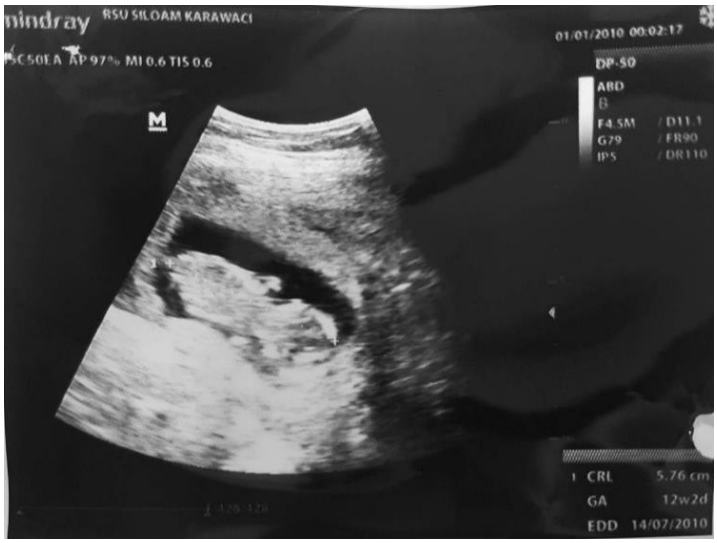

Figure 3

2 weeks after first outpatient visit, she was admitted to the emergency room due to severe abdominal pain. Her vital signs were within normal range. The significant physical examination showed a slightly anemic appearance with a mild to moderate tenderness during palpation on uterus. The uterus was larger than the gestational age (20-22 weeks sized) due tothe large uterine fibroid. At that time, we were considering whether the abdominal pain was dueto the uterine fibroid or any other surgical problems. Hence, she was referred to the surgical department. Ultrasound by obstetrician was done, revealed with a single live intrauterine fetus, CRL was measured appropriate to 12-13 gestational age. We noticed an unusual position of the gestational sac, which was pushed laterally to the left side of the uterus due to the large myoma. Laboratory examination indicated anemia (hemoglobin level 9.7g/dL, erythrocyte 28,30%) and possible infection (leucocyte 11780/mm3, ESR 65 mm/hours).Random Blood Glucose was 156. ECG was within normal limit.

A few hours after clearance from obstetrician, whole abdominal ultrasound examinationin radiology department was done, requested by the surgeon to rule out any other surgical problems. It showed hyperechoic heterogen mass with indistinct boundaries from intrauterine, sized +/- 14,12 x 13,86 cm (Figure 2) suggestive uterine fibroid. In addition, there was another lesion nearby, sized +/- 3,48 x 2,99 cm. Free fluid was also detected at upper right quadrant, perispleenic and around the uterus. The gestational sac was seen in left side of the uterus with fetal pole measured 3.56 cm corresponding to 12 – 13 weeks of gestational age (Figure 3).

Soon after the ultrasound examination from radiology department had been done, shecomplained of increased abdominal pain intensity, difficulty of breathing as her vital signs became worsen as if she developed a hypovolemic state. She was rushed to the operating theatre to undergo exploratory laparotomy.

On exploration, we found hemoperitoneum and blood clots estimated to be approximately 4.5 L. We found approximately 6x4 cm point of rupture at the left cornual areafrom the large uterus, sized about 20-22 weeks gestational age, with a protrude intact gestational sac in the abdominal cavity and a 12 weeks sized fetus within (Figure 4,5). The operator then performed left cornual wedge resection. The bleeding was completely controlled with two layers stitching, followed by myomectomy sized 15x13x9 cm (Figure 6). Uterine fibroid was sent to the laboratory for pathological examination. Gestational sac was given to the family. The patient underwent a left salpingectomy since the left fallopian tube was detached from the uterus. Since the uterus is no longer viable for another pregnancy, we decided to have another informed consent from the husband to remove the other fallopian tubeas well. The decision was also made with consideration of social-economic issues.

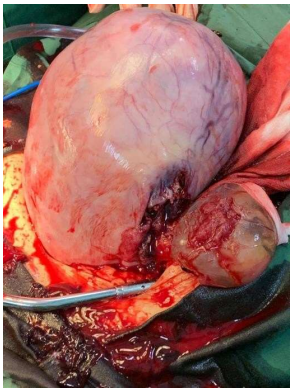

Figure 4

Figure 5

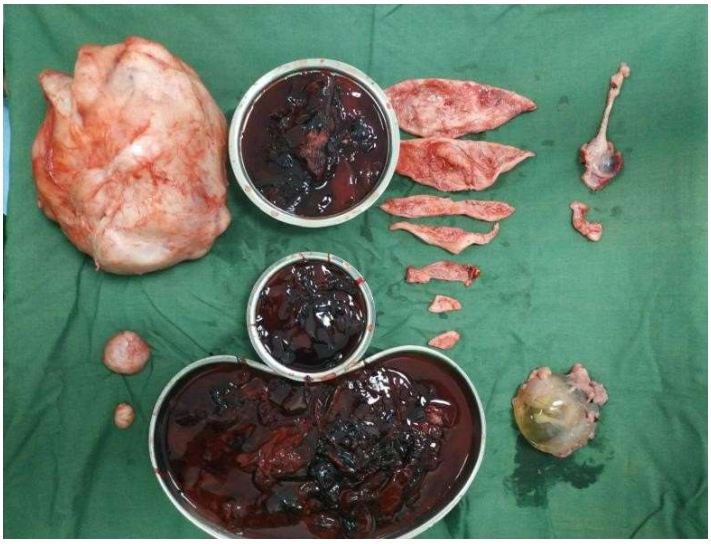

Figure 6

She was admitted to the Intensive Care Unit (ICU). Correction of anemia by packed red cell transfusion was done. The hospitalization was uneventful, and the patient was discharged five days after surgery in good condition.

Discussion

We present a case of an undiagnosed cornual pregnancy. The diagnosis itself is quite challenging because cornual pregnancy is an ectopic pregnancy inside the uterus. However, it is located in the part of the fallopian tube that penetrates the muscular layer of the uterus which makes physicians often misdiagnose it with normal uterine pregnancy. Moreover, this patient had a large myoma in the last 7 years prior to pregnancy. It makes us more difficult to assertain cornual pregnancy.

Ectopic pregnancy is the implantation of a fertilized ovum outside the uterine cavity, and it's thought to affect 1-2% of pregnancies. 93-97% of ectopic pregnancies are tubal. Interstitial (cornual) pregnancy is a rare type of ectopic pregnancy (EP), accounting for 2-4% of all tubal pregnancies.[8] The surrounding myometrial tissue allows progression of the pregnancy into second trimester but rupture at such an advanced gestation may result in catastrophic hemorrhage with mortality rate of up to 2%, as the location is close to the uterine blood supply, that leaves hysterectomy to be the only option.[9.]

Uterine fibroids or myomas are the most frequently recorded benign smooth muscle tumor of the uterus, affecting 20-60% of women in their reproductive age and may negativelyaffect fertility and outcome of pregnancy. Myomas have been related to a lot of ante-, intra-and postpartum complications such as spontaneous abortion, antepartum hemorrhage, placental abruption, malposition of the fetus, fetopelvic disproportion, premature rupture of membranes, retention of placenta, postpartum hemorrhage, preterm delivery, low birth weight infants, dysfunctional labor and increased need to cesarean delivery. Patient history ofuterine fibroid plays a role in this cornual pregnancy. The size of the fibroid is quite large +/- 13.3x11.3x8.2 cm which makes the embryonic implantation took place in the area intrauterine which was free from fibroid, in this case the left cornu.[10]

Clinical finding of cornual pregnancy depends upon whether or not it is ruptured. Inruptured cases, patients may present with severe abdominal pain with features of hemodynamicinstability. Aside from using abdominal

sonography, another diagnostic tool of cornualpregnancy should be transvaginal sonography. Findings from transvaginal ultrasound such asthe eccentric position of the gestational sac with an empty uterine cavity and the presence ofa thin (less than 5mm) or even absent myometrium surrounding the sac are highly suggestiveof cornual ectopic pregnancy. Some literature even recommended the use of MagneticResonance Imaging as a diagnostic tool for this case, which is not available in our settings.[2,11]Patient came to emergency unit because of pain in the lower region of abdomen that comes and goes for about 3 days with increasing intensity. She was known pregnant for about 12 weeks and denying any vaginal bleeding. Antenatal care was done once accidentally whenshe went to obstetrician to check her period and uterine fibroid. Transvaginal was not considered in this case because it is not a routine procedure for a 10 weeks pregnancy. Duringabdominal ultrasonography examination, gestational sac was found on the left lateral segmentof uterus (eccentric position), in which cornual pregnancy should be considered at that time. Unfortunately, the patient came back 2 weeks after the last examination presented with abdominal pain. At the time the patient came to emergency room, there was no sign of acute abdomen and the ultrasound by the obstetrician still showed the single live intrauterine pregnancy along with the large uterine fibroid. Although the position of the pregnancy was unusual, we still considered it was caused by the large uterine fibroid. Moreover, we were thinking about any other surgical problems, hence referred to surgical department. Whole abdominal ultrasound from the radiology department was done, showed free fluid on the right upper quadrant of theabdomen, perispleenic and surrounding the uterus.

Soon after the ultrasound examination, the patient was presented with acute abdominal pain and hypovolemic state, in which internal bleeding was suspected, hence she was rushed to the operation theatre to undergo exploratory laparotomy.

In this case, we did a left cornual wedge resection instead of hysterectomy with consideration of age (patient is still young) and preservation of the womb as requested by the patient. In this opportunity, we decided to remove the myoma, as well as the left fallopian tube since it was detached from the uterus. Considering the viability of another pregnancy is not suggested in this case, we did another informed consent to remove the other fallopian tube (right fallopian tube).

Conclusion

Interstitial (cornual) pregnancy is a rare type of ectopic pregnancy (EP), accountingfor 2-4% of all tubal pregnancies. The etiology of cornual pregnancy remains unclear.Uterine myoma in pregnancy can cause several complications. In this case, we highly considered that the cause of cornual pregnancy is because of the presence of the large uterine myoma.

Diagnosis and therapy of cornual pregnancy remains as a great challenge. In any cases of pregnancy with myoma, we should be very careful when an eccentric position of the gestational sac is noticed, in which we need to rule out the possibility of a cornualpregnancy. It is very important to make a proper diagnosis since this type of ectopic pregnancy has 6-7 times higher mortality rate than other ectopic pregnancies of the fallopian tube. With a proper diagnosis, we can do an appropriate management to reduce high morbidity and mortality.

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