

Near Term Rudimentary Horn Pregnancy with Term Intrauterine Pregnancy: A Case Presentation

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Abstract

Introduction: Unicornuate uterus accounts for 5 percent of all Mullerian anomalies. Rarely it could be a site for ectopic pregnancy, where natural course is rupture during second trimester, with a potentially life-threatening heavy bleeding. However, in about 10 percent of cases horn pregnancy will go to term or form a lithopedion. We describe an unexpected horn pregnancy reaching near term with live term intrauterine pregnancy, an unusual presentation.

Case presentation: 25 years old Gravida III, Para II woman presented to our hospital with complaint of right upper quadrant swelling of 5 months duration. Thirteen months back, she was diagnosed to have IUD with femoral length of 34 weeks and misoprostol used for induction of labor but failed. Patient went home and she came again with the above complaint. On examination, uterus was 26 weeks sized, positive fetal heartbeat. She has 20 cm × 20 cm firm, non-tender right upper quadrant mass. Ultrasound showed singleton intrauterine alive pregnancy of 26 weeks + 5 days and dead fetus of 34 weeks by femur length in the right abdominal cavity. Transverse lower uterine segment cesarean was done to effect 3800 gm male alive neonate. Resection of rudimentary horn and right salpingectomy was done. The horn was connected to the isthmus right wall of the uterus by a thin fibromuscular tissue and contained 2200 gm macerated fetus and placenta. No right ovary seen. The patient and neonate progressed well and were discharged.

Conclusion: An ectopic pregnancy in a rudimentary horn is rare and carries severe maternal and fetal consequences; antenatal diagnosis is challenging. Therefore, high index of suspicion is recommended to prevent morbidity and mortality after failed induction. The life-threatening complication is rupture of the horn.

Keywords: Rudimentary horn pregnancy; Ectopic pregnancy; Lithopedion; Gondar

Introduction

Unicornuate uterus accounts for 5 percent of all Mullerian anomalies, occurring in general population, approximately, 1 in 4020 women [1]; in about 84 percent of these cases a contralateral rudimentary horn exists, almost always of a noncommunicating type [2]. The presence of a rudimentary uterine horn with cavity leads to gynecologic and obstetric complications [1]. Most rudimentary horns are asymptomatic; however, some contain functional endometrium, although not necessarily normal [1]. Cyclic or chronic pelvic pain, hematometra, and endometriosis are often associated symptoms. Besides, the uterine horn could be a site for ectopic pregnancy, where natural course is rupture during second trimester, with a potentially life-threatening heavy bleeding [3]. However, in about 10 percent of cases horn pregnancy will go to term or form a lithopedion [4]. Pregnancy in such a rudimentary horn is extremely rare, 10-fold less common than an abdominal pregnancy. We describe an unexpected horn pregnancy reaching near term with live term intrauterine pregnancy, an unusual presentation.

Case Report

25 years old Gravida III, Para II Amhara woman presented to our hospital with complaint of right upper quadrant swelling of 5 months duration. Two weeks back she had one episode of minimal vaginal bleeding. Her antenatal care (ANC) follow up was at health center. Previous two deliveries were at home and uneventful. Thirteen months back, she presented with absent fetal movement of one month duration after 8 months of amenorrhea. At that time ultrasound was done. It showed intrauterine pregnancy, negative fetal heart beat; gestational age by femoral length was 34 weeks and breech presentation. For this labor induction with misoprostol 50 microgram every six hours for five doses tried, but failed. Ultrasound was not repeated. Patient went

home because of social reason. Currently, she presented with the above complaint. Up on physical examination, vital signs were in normal range, no pertinent finding on chest and cardiovascular system. On abdominal examination, uterus was 26 weeks sized, positive fetal heart beat. There was 20 cm × 20 cm firm, non tender right upper quadrant mass. Cervix was closed, uneffaced and posterior.

Ultrasound showed singleton intrauterine pregnancy with positive fetal heart beat and gestational age of 26 weeks + 5 days. There was also a dead fetus in the right abdominal cavity, adherent to the intrauterine pregnancy and gestational age by femoral length was 34 weeks. All laboratory investigations were in normal range. She was admitted with the impression of second trimester intrauterine pregnancy with lithopedion at third trimester. She was managed conservatively with strict follow up, steroid was given; fetus reached to term. It was breech presentation; placenta was low lying and patient started to have active bleeding. Emergency laparotomy was done. There was intact term sized gravid uterus with well-formed lower uterine segment and 20 weeks sized intact uterine horn on the right side. The horn was connected to the isthmus right wall of the uterus by a thin fibromuscular tissue. No right ovary seen. Left tube and ovary looks normal, both kidneys were in their normal site (Figure 1). Transverse lower uterine segment cesarean

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Figure 1: Rudimentary horn pregnancy.



Figure 2: Transverse section of rudimentary horn with macerated fetus inside.

was done to effect 3800 gm male alive neonate with Apgar score of 4, 6 and 8 in 1st, 5th and 10th minutes respectively. Placenta was low lying. Resection of rudimentary horn and right salpingectomy was done. The horn contained 2200 gm macerated fetus and placenta. Postoperative Hct was 23% and patient was discharged at 6th postoperative day (Figures 2 and 3).

Discussion

Unicornuate uterus accounts for 5 percent of all Mullerian anomalies, occurring in general population, approximately, 1 in 4020 women [1]; in about 84 percent of these cases a contralateral rudimentary horn exists, almost always of a noncommunicating type [2]. This case is being reported because no similar case has been reported so far, coexistence of term intrauterine and near term rudimentary horn pregnancies diagnosed at laparotomy. Pregnancy in a non-communicating rudimentary horn is uncommon, estimated to occur in 1 per 100,000 to 140,000 pregnancies [3]. Conception in the rudimentary horn arises either from a small communication with the uterine cavity or due to transperitoneal migration of the spermatozoon or fertilized ovum through contralateral tube [2-6]. Most rudimentary horn are asymptomatic but they can present with ectopic pregnancy as seen in our case [3]. In majority of cases, horn rupture occurs before 20 weeks [7]; reports of rupture vary from 5-37 weeks [5,7], depending on the horn musculature, variable thickness, under development with poor distensibility of the myometrium and the propensity for abnormal placentation contribute to the risk of uterine rupture [8]. But, in about 10 percent of cases horn pregnancy will go to term or form a lithopedion [4]. This is unique in our case

where induction failed and horn pregnancy stayed for 16 months without rupture. Because of reduced distensibility, relatively small volume and anomalous vasculature supplying the rudimentary horn a malformed fetus, fetal growth restriction, oligohydramnios and fetal malpresentation represent other forms of presentation of this condition [1,9,10]. Breech presentation was one of the finding in our case. Failed termination of pregnancy by medical method and uterine evacuation has been reported in obstructive Mullerian anomalies [2,11]. Failed abortion should be investigated with a high index of suspicion. Buntungu et al. reported a rudimentary horn pregnancy in a 6th gravida with all previous normal deliveries. Diagnosis of intrauterine fetal demise where induction with misoprostol failed would have raised the suspicion of ectopic pregnancy in our case [12] and ultrasound examination would have been repeated. Use of labor induction agents for termination of pregnancy in a rudimentary horn is unsuccessful and can lead to rupture of the horn. Samuels and Awonuga reported rupture after use of misoprostol due to misdiagnosis [13].

The possibility of a uterine anomaly should be considered by clinicians and sonographers in unexplained intermittent early pregnancy bleeding [14]. As it was observed in our patient who had history of vaginal bleeding two weeks back from admission in early pregnancy and cesarean delivery done for low lying placenta with active bleeding. The availability and advance in ultrasound and magnetic resonance imaging ameliorate the diagnosis of rudimentary horn pregnancy principally at an early gestational age. However, as the gestational age increases, the enlarged pregnant horn can obscure adjacent anatomic structures which makes the diagnosis more difficult [1]. Even it is difficult to diagnose rudimentary horn pregnancy in the presence of intrauterine pregnancy as seen in our case. It was taken as abdominal lithopedion. The sensitivity of ultrasound to diagnose a pregnant uterine horn could be as low as 30 percent [1]. Tsafirir et al. reported 2 cases of rudimentary horn pregnancy diagnosed in the first trimester by sonography and confirmed by MRI. They outlined a set of criteria for diagnosing pregnancy in the rudimentary horn. These are (1) a pseudo pattern of asymmetrical bicornuate uterus; (2) absent visual continuity of tissue surrounding the gestational sac and the uterine cervix; (3) presence of myometrial tissue surrounding the gestational sac. Nonetheless, most of the cases remain undiagnosed until it ruptures and present as emergency. Cases of late and false diagnosis leading to uterine rupture have been reported [1,6,7]. MRI has a major role for the diagnosis of Mullerian anomalies and should be considered when a pregnant rudimentary horn is suspected [1,6] which is not available in most resource limited areas.



Figure 3: Calcified fetus with placenta.

The classic management of a rudimentary horn pregnancy had been laparotomy with excision of the rudimentary horn and ipsilateral salpingectomy in order to prevent rupture, future ectopic pregnancies, and dysmenorrhea [1,2,6]. Hysterectomy may be necessary in massive hemorrhage [2]. Recently, early diagnosed cases have been treated by laparoscopic approach [2,6] which is not available in low resource centers. Renal abnormalities can coexist up to 40 percent of cases of unicornuate uterus, palpation during laparotomy to rule out urinary anomalies is fundamental in these patients [1]. Other associated anomalies such as an ectopic ovary tissue and, more rarely, absent ipsilateral gonad could occur [1]. In this case ipsilateral ovary was absent.

An ectopic pregnancy in a rudimentary horn is rare and carries severe maternal and fetal consequences; antenatal diagnosis is challenging, which is missed twice in this case. Therefore, high index of suspicion is recommended to prevent morbidity, especially in high risk groups like previous history of pelvic pain and infertility, recurrent miscarriages or late miscarriage, preterm labor, fetal malpresentation, fetal growth restriction, abnormal placentation, preeclampsia or failed induction for termination of pregnancy. This condition is clearly described in our case. Despite the removal of the rudimentary horn pregnancy, the patient should be advised of the increased risk of future ectopic pregnancy and associated risks with unicornuate uterus like malpresentation and abnormal placentation as seen in the current term intrauterine pregnancy.

Conclusion

An ectopic pregnancy in a rudimentary horn is rare and carries severe maternal and fetal consequences; antenatal diagnosis is challenging. Therefore, high index of suspicion is recommended to prevent morbidity and mortality after failed termination of pregnancy. The life-threatening complication is rupture of the horn.

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