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CASE REPORT

Term Pregnancy with a Live Fetus in Non-communicating Rudimentary Horn with Placenta Percreta

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Introduction

Abnormalities of embryogenesis of mullerian duct system resulting in congenital anomalies of female genital tract are relatively common [1]. The prevalence of congenital uterine anomalies in the general population is 6.7 % [2]. In women with a history of repeated pregnancy loss, the rate of mullerian anomalies increases to 3–25 % [2, 3]. Unicornuate uterus accounts for 5 % of all mullerian anomalies. Unicornuate uterus is thought to occur in general population at a rate of 1:4,020 [1]. Unicornuate uterus is a type II mullerian anomaly according to the American Fertility Society classification system [1, 4] that occurs due to a complete or partial failure of development of one mullerian duct and incomplete fusion with contralateral side [1]. The failed mullerian duct fusion leads to the formation of an isolated hemiuterus without a contralateral structure (in complete failure) to various degrees of a rudimentary horn (in partial failure) [1]. This rudimentary horn is subclassified into communicating or non-communicating with uterine cavity and a horn with no cavity [1]. In about 84 % of these cases, a contralateral rudimentary horn exists, almost always of a non-communicating type [1, 5]. The attachment of the rudimentary horn may vary from a fibromuscular band (separated variety) to an extensive fusion between the two horns where there is no external separation between them [4, 6]. This was a case with an extensive fusion between the two horns.

Pregnancy in such a rudimentary horn is extremely rare, tenfold less common than an abdominal pregnancy [1, 2, 4]. The natural course of a rudimentary horn pregnancy is

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rupture during the first or mid-second trimester with potentially life-threatening bleeding [1, 5]. In the majority of cases, horn rupture occurs before 20 weeks of gestation [7]; reports of rupture varying from 5 to 37 weeks are described [1, 7]. The uterine wall being thicker and more vascular, bleeding is more severe in rudimentary horn pregnancy rupture, therefore it commonly manifests with acute abdominal pain and intraperitoneal hemorrage [1, 7].

Case Report

Our patient was a 26-year-old G3P2L2 with 37 weeks pregnancy. She had previous two full-term vaginal deliveries. Her present pregnancy was supervised by a local private doctor. Up to fifth month, her pregnancy was uneventful and then she developed abdominal pain which was non-specific, generalized, and continuous, not associated with nausea, vomiting, dizziness, and syncopal attack or pervaginal bleeding. She consulted the doctor; ultrasound and some analgesics were advised. Pain was relieved by taking medicine, but after some time pain reappeared. She had loss of fetal movement since the sixth month of pregnancy. USG was done, and bicornuate unicollis uterus, within 26 week ± 5 days of pregnancy, in right horn was diagnosed. USG of 32nd and 35th week also suggested the same findings. At the 37th week of pregnancy, she was admitted to the district hospital in Ujjain for continuous abdominal pain where pain was relieved and she was referred to M.Y. Hospital because of the USG finding of pregnancy with bicornuate uterus, transverse lie with placenta praevia, and oligohydramnios.

On admission, the patient was found to be stable. Her pulse was 88/m and BP was 110/70 mm Hg. On obstetric examination per abdomen, she was 32 weeks pregnant, oblique lie, and her lower pole was empty, and the uterus was relaxed and nontender. FHS was 132/m regular. As per speculum examination, the cervix was posterior, and her cervical os was closed. Her Hb% was 6.4 g/dl. Repeat ultrasound in our center was also suggestive of the same findings of "36 weeks pregnancy with bicornuate uterus, transverse lie, placenta praevia and oligohydramnios (AFI = 1.5)." Cesarean section was done for transverse lie and placenta praevia. After opening the abdomen, baby was delivered by a vertical incision on the upper part of saccular structure lying in the supra-pubic space. Placenta was densely adhered and encroached up to the serosa of the sac. The upper part of the sac adhered to the omentum. After exteriorization of the sac, we found a normal-sized uterus with left tube and ovary was attached to the left side of the sac corresponding to the left horn of the uterus. The sac was attached to the right tube and ovary, corresponding to a rudimentary horn. Rudimentary horn along with the placenta, right tube, and ovary was excised en bloc. There was no intraperitoneal hemorrhage. After the removal of the rudimentary horn, brisk hemorrhage occurred from the margins of the right horn of uterus, and to control it, hysterectomy was done, leaving the left tube and ovary in place. There was no communication between the right and the left horn. Patient delivered a live-term female baby, weighing 2.5 kg with good apgar score of 5 and 7 at 1 and 5 min, respectively, and no apparent gross deformity. Four units of packed red cells were transfused to the patients along with 2 units FFP. Post-op recovery was uneventful. Patient was discharged on the 10th postoperative day with good condition (Figs. 1, 2).

Discussion

Pregnancy in rudimentary horn is a rare condition. It was first described in 1669 by Mauriceau and Vassal [2, 4]. The incidence of rudimentary horn pregnancy is quoted as 1:76,000–1:140,000 [4]. Worldwide, it has been described up to now in about 700 cases [1]. The term pregnancy in rudimentary horn with live fetus is an extraordinary unusual combination, only 10 % reach term, and the newborn survival rate is about 2 % [1]. Because of reduced expansibility, 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].

In addition to the morbidity associated with uterine rupture, abnormal placentation like accreta or percreta may also be encountered in these pregnancies and add further complication [2, 8]. The endometrium of the rudimentary horn has been described as thinner and sometimes dysfunctional leading to pathologic placentation, with placenta accreta being described with this condition [8]. Nine cases of placenta accreta in a pregnant rudimentary uterine horn have been reported, and this state of affairs was followed by hemorrhagic rupture of the uterus in 8 cases, while the ninth case of RHP with placenta accreta presented by Henriet et al. was diagnosed before rupture [2, 9]. Oral et al. anticipated on the basis of literature review that the prevalence of placenta accreta in rudimentary uterine horn pregnancies may be greater than 10 % [2, 9].

Early diagnosis of the rudimentary horn pregnancy is essential, to prevent life-threatening complication of rupture. An early bimanual palpation showing a deviated uterus with a palpable adnexal mass, a mass extending outside the uterine angle (Baart de la faille's sign) or displacement of fundus to contralateral side with rotation of uterus and elevation of affected horn known as Ruge Simnn Syndrome, should lead to a suspicion of a mullerian anomaly [1]. The availability of technological advances like ultrasonography (USG) and magnetic resonance

imaging (MRI) has made the diagnosis of rudimentary horn pregnancy possible at an early gestational age. But in advance pregnancy diagnostic accuracy decreases. The sensitivity of USG as a late gestation diagnostic tool is 26 % [8, 9]. MRI appeared to be the "gold standard" for diagnosing and grouping utzerine anomalies because of its 98–100 % accuracy [2, 6].

Our case is a rare case with unruptured term rudimentary horn pregnancy with placenta percreta. The patient had previous two vaginal deliveries at term. Her pregnancy was misdiagnosed as a pregnancy with bicornuate uterus in routine ultrasound scan. She was referred to our hospital at 37th week of gestational age for transverse lie, with placenta praevia for which cesarean section was done. During operation, she was diagnosed as a case of term rudimentary horn pregnancy with placenta percreta. Tsafrir et al. suggested ultrasound criteria for early diagnosis of this condition that includes (1) a pseudo-pattern of asymmetrical bicornuate uterus, (2) absent visual continuity between the cervical canal and the lumen of the pregnant horn, and (3) the presence of myometrial tissue surrounding the gestational sac [7, 8, 10]. These criteria can help in differentiating the pregnancy from cornual pregnancy, intraabdominal pregnancy, and bicornuate uterus [8, 10].

Once a diagnosis of the rudimentary horn pregnancy is made, the treatment is laparotomy with excision of rudimentary horn and ipsilateral salpingectomy to prevent spontaneous rupture and possible catastrophic consequences [1, 4, 8]. Hysterectomy may be necessary in case of massive hemorrhage [1]. Medical management with methotrexate or fetocide (in a later pregnancy), and posterior pregnancy rudimentary horn excision by laparoscopy is proposed by Cutner et al. with the aim to shrink the horn and allow a less invasive surgery [1]. Dicker et al. reported the case of women benefiting from laparoscopic surgery of rudimentary horn [8, 10].

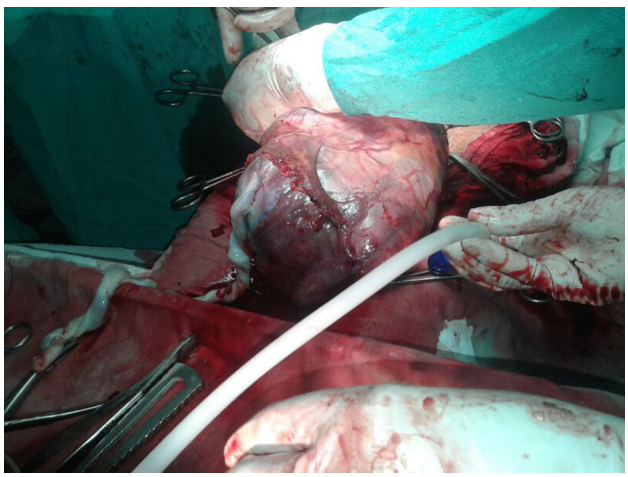

Fig. 1 Rudimentary horn sac with placenta percreta before excision

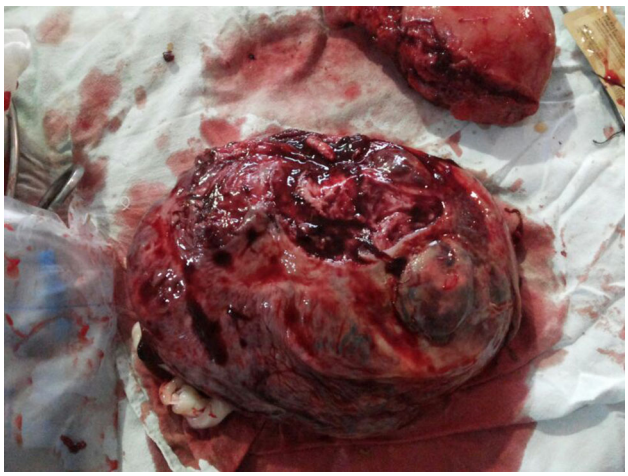

Fig. 2 Rudimentary horn with placenta percreta after excision

Conclusion

Pregnancy in rudimentary horn is a rare but a life-threatening condition. Diagnosis is difficult and challenging. Careful clinical examination and imaging techniques such as USG and MRI can help in the diagnosis of this condition.

Compliance with ethical requirements and Conflict of interest Informed written consent was obtained from the patient for publication of her case. This study was approved by the ethical committee of our institute and the authors declare that they have no conflict of interest.

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