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A Rare Case Report of Uterine Didelphys, in Which One Uterus Carried a Pregnancy While the Other Carried Twice, with a Successful Pregnancy Outcome Resulting in an Alive-Term Delivery

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Abstract

Uterine didelphys is a rare congenital anomaly of the female reproductive organs, designated by the presence of the uterus as a pair of organ, the chance of having a pregnancy in one of the uterus while the other one had successfully pregnant twice is very low. In this case, report we present a 31-year-old gravida four para three deliveries (two Cesarean section, alive and healthy, one spontaneous abortion) woman, who received C-section to terminate pregnancy due to intrauterine hypoxia and oligohydramnios, with the outcome of a 3000 g male alive neonate. Intraoperatively, there was uterine didelphys, one uterus holding the pregnancy while the other with caesarean section scar.

Subject Areas

Gynecology & Obstetrics

Keywords

Didelphic Uterus, Pregnancy, Müllerian Duct Anomalies

1. Introduction

Müllerian duct anomalies (MDAs) are a group of congenital defects of the female reproductive tract which occur due to the abnormal formation, fusion, or resorption of Müllerian ducts in utero. It is classified as an abnormality of formation (agenesis), lateral fusion defects (arcuate, bicornuate, didelphys, septate,

and unicornuate), and vertical fusion defects (transversevaginal septum) [1] [2]. The didelphicuterus results from a lateral fusion defect and is one of the rare types of MDAs occurring in 1/3000 of all women and 11% of women with Müllerian anomalies. It arises from incomplete fusion of the upper portion of the Müllerian ducts that results in two entirely separate hemiuteri, two cervices, and usually two vaginas or a longitudinal vaginal septum. The cause of the failure of fusion is not known. But there are several risk factors identified as contributing factors. Patients with a didelphic uterus may have associated defects in the renal system, vagina, and, rarely, the skeleton [3] [4] [5]. The diagnosis of MDAs, including didelphys, is challenging and usually made during the reproductive period. This is believed to be because they are associated with symptoms like amenorrhea, dysmenorrhea, dyspareunia, pelvic pain, and obstetric complications such as recurrent pregnancy loss, premature delivery, malpresentation, intrauterine growth restriction (IUGR), placental abruption, and cervical insufficiency [5] [6] [7] [8]. It is very rare for such patients to successfully conceive to term, and there are no delivery guidelines for this kind of uterine malformation pregnancy, so obstetrician need to individually assess the condition and formulate delivery plans.

2. Case

A 31-year-old woman (gravida 4, para) came to our hospital with chief complain of lower abdominal pain and minor vaginal bleeding for 1 hour at the gestational age of 39w2d.

In regards of her medical history, she was diagnosed with Muller's duct abnormalities (MDAs) in her first pregnancy (in the right uterus) in 2017, there were two vaginas and two cervixes. Cesarean section was performed at 41 weeks of pregnancy, and a boy weighing 3000 g was delivered successfully with 300 ml intraoperative bleeding, she had a quick recovery after surgery. The woman got pregnant in the right uterus again two years later, regular prenatal examinations were unremarkable, she had another C-section with surgical indications of scarred uterus at 38 weeks' gestation age after active phase of labor a live baby girl weighing 3000 g was delivered successfully with 200 ml intraoperative bleeding, she recovered well after that. The third pregnancy still occurred in the right uterus 2 years ago, unfortunately, she had the missed abortion at 8 weeks of pregnancy.

Ultrasound revealed the pregnancy in the left uterus this time (6 weeks of gestation) (**Figure 1**).

Her prenatal examinations were unremarkable during this pregnancy, she had paroxysmal abdominal pain accompanied by a small amount of vaginal bleeding at 38w2d, obstetrical examinations were as below: Fundal height was 32 cm, abdominal circumference 98 cm, cephalic presentation, LOA, fetal heart beat 154 times/min with irregular contractions, we estimated fetal weight 3400 g. Vaginal examination revealed two vaginas, left cervix was soft, anterior, 0% of efface-

ment, uterine orifice was closed, Station-3, the amniotic membrane was intact, a bishop score of 3 was concluded. The right cervical was soft and deviated toward backwards. The amniotic fluid volume was low as shown in the ultrasound (AFI: 40 mm), Pregnant women are willing to request vaginal delivery, the examination of color ultrasound indicated: amniotic fluid volume is too low (AFI40 mm), the fetal heat monitor showed no acceleration, the condition didn't improve after management and intrauterine hypoxia was considered, given to the patient's cervical condition, we informed the woman and her family members with the situation and had a discussion of chances and risks, together, we decided to take C-section to terminate the pregnancy. The right uterus was slightly enlarged with the scar in the lower segment was observed, and the right ovary and fallopian tubes were in normal shape (**Figure 2**).

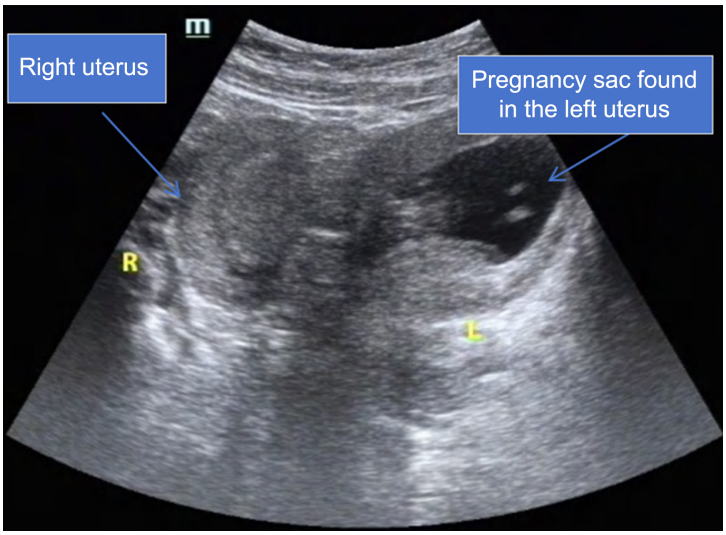

Figure 1. Two-dimensional ultrasound showing two uteruses and Pregnancy sac found in the left uterus.

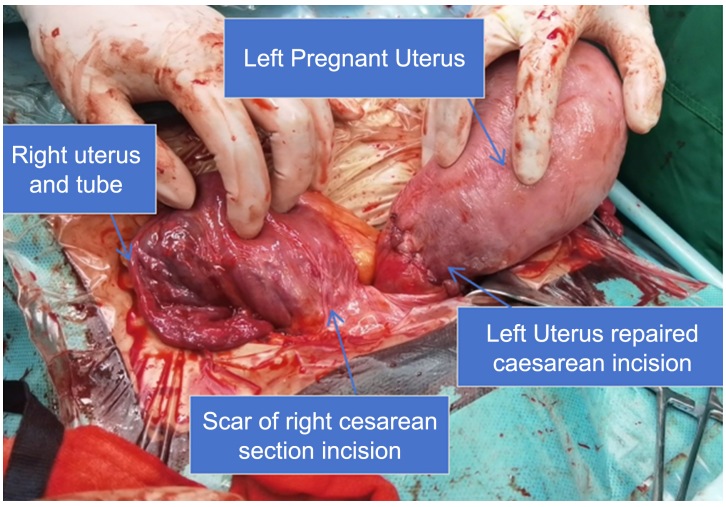

Figure 2. An anterior view of the double uterus with their tube and ovary, and both the left right uterus with a repaired cesarean incision.

Her operation was successful and a live male neonate weighing 2800 g was delivered, she recovered very well and the mother and baby were discharged 4 days after the surgery.

3. Discussion

Congenital uterine dysplasia is the most common type of female reproductive system dysplasia, and its incidence rate is about 0.1% - 0.2% [9]. At present, it is believed that the occurrence of congenital uterine developmental abnormalities is related to the abnormal alignment, merging, and fusion of the Mullerian tubes during the embryonic period [10]. Mullerian tube developmental abnormalities are also closely related to unilateral kidney deficiency in patients, with approximately 29.8% of patients with Mullerian tube developmental abnormalities accompanied by unilateral kidney deficiency, which is common in patients with both uteri, with an incidence rate of 81.3%, The reason for its occurrence may be related to the fact that the genitourinary tract belongs to the extracellular mesoderm during embryonic development [11] [12]. Congenital uterine developmental abnormalities can be classified into the following categories according to the revised Buttram classification standards of the American Fertility Association: mediastinal uterus, bicornuate uterus, saddle shaped uterus, unicorn uterus, and residual horn uterus. The bicornuate uterus includes double uterus, double vagina, double uterus, single vagina, and the mediastinal uterus includes complete and incomplete mediastinum [13]. Due to axial abnormalities in the uterine cavity and changes in the shape of the uterine cavity, the volume of the uterine cavity is relatively reduced, fetal activity is limited, and the incidence of abnormal fetal position is significantly increased. At the same time, abnormal uterine development is often accompanied by abnormal blood flow and uterine dysfunction, leading to an increase in both natural abortion and premature birth rates. Congenital uterine development abnormalities are often accompanied with the abnormal distribution of uterine muscle fibers, resulting in uncoordinated contraction of uterine muscle fibers, Prone to Postpartum haemorrhage [14]. Research has shown that the miscarriage rate of pregnant women with uterine malformations is as high as 14.3%, the premature birth rate is 18.4%, and the full term birth rate is 67.3%. The miscarriage rate and premature birth rate are significantly higher than the normal control group, and the full-term birth rate is significantly lower than the control group [15]. As for pregnant patients with MDAs, the mode of delivery must be decided individually. The literature reports are almost equal for those who prefer CS and those who prefer vaginal delivery. It is quite common to get abnormal presentations and positions in patients with uterus didelphys, but there are reports of successful labor in this group of women. 10CS was the mode of delivery for 82% of patients having MDAs, as reported by Heinonen *et al.* [16].

In this case, the signs of labor begin at 39 weeks of gestation, she had two C-sections in the right uterus pregnancies before and this pregnancy was in the

left uterus, the delivery mode was the focus of discussion. Some believed that the vaginal delivery was possible due to the pregnancy was in the left uterus this time and the probability of uterine rupture is small, and this was the patient's first choice at the beginning, while the opposite opinion stands with the risks of fetal distress, prolonged labor, and postpartum hemorrhage in rare condition as uterine didelphys. Unfortunately, the amniotic fluid volume is low and the fetal monitoring was undesirable, we decided together with the patient to terminate the pregnancy with C-section which was successful.

4. Conclusion

We recommended strengthening pre-pregnancy examination for women with reproductive malformations to get timely management. It is necessary to pay attention to the situation such as abortion, premature delivery, abnormal fetal position and postpartum hemorrhage, actively prevent the adverse events. There are no delivery guidelines for this kind of uterine malformation pregnancy, and hospitals at all levels need to individually assess the condition and formulate delivery plans. Once C-section comes as first choice, surgical risks should be fully assessed before delivery and detailed surgical plans should be formulated to ensure patient safety.

Acknowledgments

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Conflicts of Interest

The authors declare no conflicts of interest.

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