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A Case Report on a Rare Uterine Congenital Anomaly: Uterine Didelphys with Three Spontaneous Pregnancies in Opposite Horns of the Uterus and Delivered by Caesarean Sections.

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ABSTRACT

Background: Müllerian duct anomalies (MDAs) are a group of congenital female reproductive tract defects which occur due to the abnormal formation, fusion, or resorption of Müllerian ducts in utero. Uterus didelphys is a rare congenital anomaly, representing approximately 11% of female reproductive system anomalies. Most cases are asymptomatic, but they can present with dysmenorrhea, pelvic pain, dyspareunia, haematocolpos, and haematometra. It is usually diagnosed by routine ultrasound, hysterosalpingography, abdominal laparoscopy, and laparotomy, but an MRI remains the gold standard.
Case: We presented Mrs V. E. a 39-year-old G7P2+4 2A woman presented with two previous caesarean section scars. She registered for ante-natal care in our facility at 24 weeks gestational age with no complaints. She was a known patient of the facility with uterine didelphys earlier diagnosed following evaluation for recurrent miscarriages. Pregnancy was supervised and she presented in labour at 36 weeks gestational age and had an emergency repeat caesarean section for two previous caesarean section in labour with good feto-maternal outcome.
Conclusion: Uterus didelphys is a complex congenital anomaly that presents unique challenges and considerations in obstetric management. Current evidence suggests that, although the link between uterus didelphys and fertility is still debated, successful pregnancies can occur in either hemiuterus, highlighting the adaptability of the reproductive system.

Introduction:

Müllerian duct anomalies (MDAs) are a group of congenital female reproductive tract defects 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 (transverse vaginal septum)[1]. Its general prevalence is 5.5-8.0% and 24.5% among women with infertility and a history of miscarriages[2].

Uterine Didelphys (UD) belongs to the third class of MDAs, according to the American Society for Reproductive Medicine (ASRM-1988)[3]. The didelphic uterus 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 hemi-uteri, two cervices, and usually two vaginas or a longitudinal vaginal septum. The cause of the failure of fusion is not known. However, various risk factors have contributed to the occurrence of UD. Patients with a didelphic uterus may have associated defects in the renal system, vagina, and, rarely, the skeleton[4-6].

Having a uterine defect heightens the likelihood of obstetric complications, underscoring the importance of regular monitoring throughout pregnancy. Notably, this condition is associated with a greater risk of spontaneous miscarriage, preterm deliveries, breech presentations, and a lower rate of live births compared to those with a normal uterus[7]. Conception in patients with UD is not impaired and is considered better than in patients with other Müllerian duct anomalies. However, they are primarily associated with poor pregnancy outcomes [7,8]. Various reports have shown spontaneous vaginal delivery in about 50% of women with uterine didelphys, approximately 1 in 3 pregnancies lead to abortions, about half result in premature deliveries and about 1 in 5 reach term[9].

This case reports a known multiparous woman with uterine didelphys, with an initial diagnosis following four recurrent miscarriages in a private facility. In all confinements, she conceived spontaneously with individual pregnancies found in different hemiuterus after close monitoring. The first two were in the same hemi-uterus-right horn, and the last was in the left horn. The first two pregnancies were carried to term, and the last pregnancy was up to 36 weeks; all were delivered by caesarean sections with good pregnancy outcomes.

Case presentation:

A 39-year-old G7P2+4 2A woman presented with two previous caesarean section scars. She registered for ante-natal care in our facility at 24 weeks gestational age with no complaints. She was a known patient of the facility with uterine didelphys earlier diagnosed following evaluation for recurrent miscarriages. Pregnancy was planned and spontaneously conceived, confirmed via serum pregnancy test following a missed period and ultrasound at eight weeks gestation. Booking parameters were within normal ranges. An ultrasound scan done at booking (24 weeks gestation) showed a singleton fetus in one hemi-uteri; no fetal abnormality was detected. She had four uneventful ante-natal visits, during which she had two doses of tetanus toxoid (TT) and two doses of intermittent preventive therapy for malaria (IPT). The antenatal period in the index pregnancy was uneventful and was planned for elective repeat caesarean section at term.

Her first confinement was in 2017; it was spontaneously conceived, supervised in our facility, carried to term and delivered with an elective lower segment caesarean section at 38 weeks gestation on account of breech presentation. Intra-op findings showed a complete uterine didelphys consisting of two uteri, two cervixes and a single vagina, with a foetus in the right hemiuterus. She delivered a live female neonate that weighed 3.6kg with good Apgar scores. The baby was breastfed, immunised for age, and is alive and well. The postpartum period was uneventful.

Her second confinement occurred spontaneously, two years following the first, carried and delivered electively at term via caesarean section at 37 weeks on account of a previous caesarean section with congenital anomaly of the uterus and patient request. She delivered a live female neonate that weighed 2.5kg and had good Apgar scores. This pregnancy was also found in the right uterus.

Clinical presentation: She presented at 36 weeks gestational age with complaints of the passage of show and labour pain. There was no drainage of liquor and no vaginal bleeding. Clinical examination revealed a young woman, healthy-looking, afebrile, not pale, well hydrated, with nil pedal oedema. Her pulse rate was 85 beats per minute, full volume, regular, blood pressure was 124/70mmHg, and respiratory rate was 20 cycles per minute. The abdomen was enlarged with a bulging on the left and some depression on the right, symphysiofundal height was 36 weeks, and there were two moderate palpable uterine contractions in 10 minutes lasting for 35 seconds. There was a single fetus with longitudinal lie and cephalic presentation, and the descent was 4/5th palpable. The fetal heart rate at presentation was 136 beats per minute. Pelvic examination revealed a single vaginal opening with two cervical os, the left os was 3 cm dilated, and the right cervix admitted one finger.

Investigations: Her packed cell volume was 38%, blood group O was positive, HIV 1 and 2 were non-reactive, and Hepatitis B and C screening was negative. Urinalysis was essentially normal.

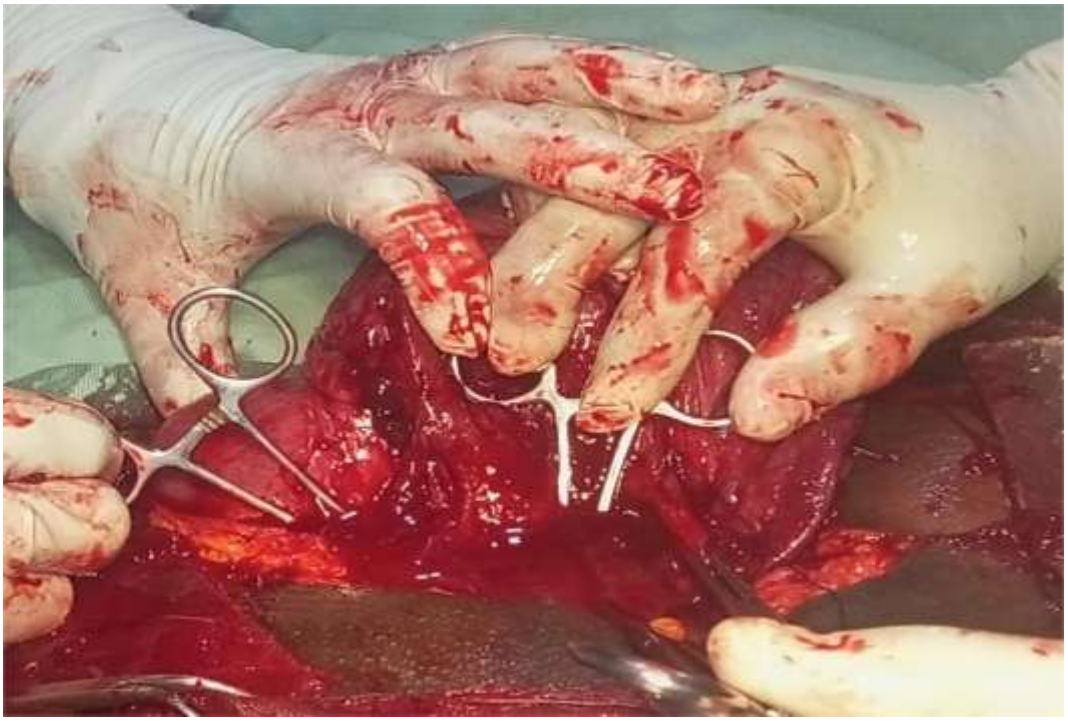
Diagnosis: Uterine Didelphys with preterm labour in a multigravida with two previous caesarean sections was made.

Therapeutic interventions: informed consent was obtained for an emergency repeat caesarean section, and an anaesthetist was invited. She had a subarachnoid block done, followed by a lower segment caesarean section. Intra-operative findings were a healed Pfannenstiel scar, moderate anterior abdominal wall and uterine adhesions, didelphylic uterus, each bearing a single uterine tube and ovary, and a gravid left hemi uterus. The right hemi uterus was slightly bulky (Figures 1, 2 & 3). A live male neonate was delivered cephalad, with a birth weight of 2.0kg and Apgar scores of eight in the first minute and nine in the fifth minute, respectively. The placenta was anterior and fundal, and the estimated blood loss was 450mls. She did well post-operatively and was discharged home on the fifth-day post-surgery after counselling for family planning.

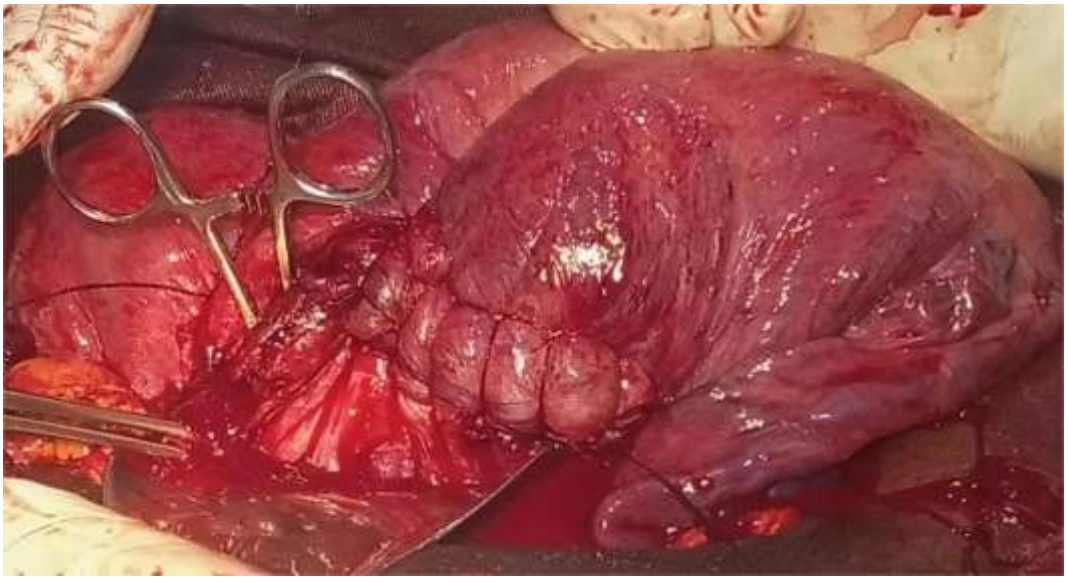
Follow-up: She had no complaints during the second and sixth week post-discharge review at the post-natal clinic. She was then counselled on family planning and opted for Jadelle.

Patient perspective: She was delighted with all aspects of the care she received from the antenatal clinic, including the promptness of intervention, the neonatal outcome, and the post-natal follow-up.

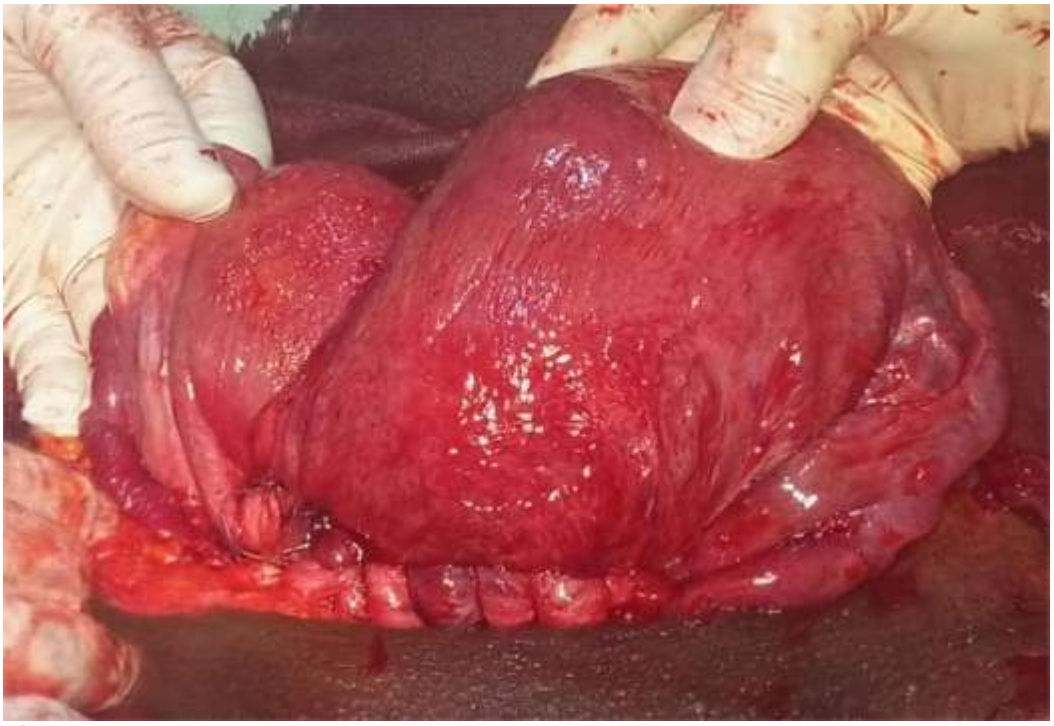
Consent: We obtained the patient's informed consent for her images and the information in this case report.



Figures 1



Figures 2



Figures 3

Discussion

Uterus didelphys is a rare congenital anomaly, representing approximately 11% of female reproductive system anomalies. Most cases are asymptomatic, but they can present with dysmenorrhea, pelvic pain, dyspareunia, haematocolpos, and haematometra[2]. It is usually diagnosed by routine ultrasound, hysterosalpingography, abdominal laparoscopy, and laparotomy, but an MRI remains the gold standard. An MRI is the most effective modality for classifying various anomalies due to its superior anatomical assessment compared to other imaging techniques[10].

The link between UD and fertility remains a subject of debate. Some studies have shown a relationship between uterine didelphys and higher rates of maternal complications, including infertility, intra-uterine growth restriction (IUGR), intra-uterine fetal death (IUFD), spontaneous abortion and postpartum haemorrhage[11]. However, a meta-analysis that compared 25 studies showed no association between UD and fertility but recognised a higher frequency of preterm delivery, abnormal fetal presentation, IUGR and IUFD[12]. Our patient had four previous miscarriages but no inability to conceive spontaneously.

Pregnancy can occur in any hemiuterus in UD. Our patient's two previous pregnancies were implanted in the right hemiuterus; however, the index pregnancy developed in the left horn. Previous reports have shown similar findings of spontaneous pregnancies occurring on opposite sides of the hemiuterus[13]. Another report showed spontaneous twin gestation occurring in each uterine horn with no complications and delivered via caesarean section[14]. These cases show the adaptability of uterus didelphys, suggesting that successful pregnancies can occur in both horns. Understanding these patterns is essential to inform clinical practice and improve patient outcomes.

Labour management in UD depends on the patient's clinical manifestation and the depth of the anomaly. A didelphys uterus is not an absolute contraindication for vaginal delivery; therefore, vaginal delivery should be considered as the first option. However, there is poor development of the muscular component of the uterus, and the unaffected hemiuterus may rotate posteriorly and become trapped in the pelvis, resulting in mechanical obstruction[15]. Previous studies have also reported successful vaginal deliveries; however, most cases undergoing caesarean delivery[16]. Our patient had complete uterine didelphys with two uteri, two cervixes and a single vagina, and had two previous caesarean sections; thus, she delivered surgically and had good fetal outcomes.

Conclusion

Uterus didelphys is a complex congenital anomaly that presents unique challenges and considerations in obstetric management. While most cases remain asymptomatic, the potential for complications and variations in pregnancy outcomes necessitates a thorough understanding of the condition. Current evidence suggests that, although the link between UD and fertility is still debated, successful pregnancies can occur in either hemiuterus, highlighting the adaptability of the reproductive system. Careful assessment and individualised labour management strategies are essential, as vaginal delivery may be feasible in select cases despite the anatomical challenges. Ultimately, continued research and clinical experience will enhance our understanding of uterus didelphys, improving outcomes for affected patients and their families.

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