Case Report

Uterine Didelphys: A Rare Anomaly Unveiled at Repeat Caesarean Section in A Multiparous Woman at Term

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Abstract

Background: Uterine didelphys (UD) is an unusual congenital Mullerian duct anomaly with significant obstetric and gynaecological implications.

Case Presentation: We present the case of a 32-year-old gravida 4 para 2 plus 1 (1 alive) multiparous woman, who underwent repeat elective caesarean section for breech presentation at 37 weeks of gestation. Despite a prior ultrasound suggesting uterine didelphys during her first confinement (the diagnosis was missed intraoperatively), and no findings on subsequent scans during the index pregnancy, the anomaly was definitively diagnosed during the current caesarean section. The fetus was successfully delivered via breech extraction from one hemi-uterus, and a separate second hemi-uterus with its cervix was identified intraoperatively. The patient's immediate postoperative course was uneventful.

Conclusion: This case highlights the diagnostic challenges of uterine didelphys, underscoring the importance of a high index of suspicion and thorough intraoperative exploration to accurately identify such rare anomalies, even in multiparous women with a history of uterine surgery.

Keywords: Uterine didelphys, congenital Müllerian anomaly, hemiuterus

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INTRODUCTION

Genital tract embryological anomalies can occur at different stages of mesonephric and paramesonephric development.[1] This depends on the period during which these vital processes are affected.

Uterine didelphys (UD) is one of the congenital genital anomalies which results from the failure of medial fusion of Müllerian ducts, specifically at the distal portions from the fundus to the cervix during 7 to 9 weeks of fetal life.[2, 3] The reason for the lack of this fusion is not well understood. It is characterised by double uterine bodies and cavities, with each possessing its own cervix.[4] Other variants include: the bicornuate uterus, septate uterus, and the unicornuate uterus.[5]

The prevalence of uterine didelphys is about 0.3% in the general population.[5] It is found in about 0.2-0.3% of infertile women and 0.6-2.1% of women with recurrent pregnancy losses.[6, 7] It may remain unnoticed during childhood and puberty, as most affected individuals with (UD) are usually asymptomatic until during pregnancy, with incidental diagnosis on ultrasound or at operative delivery.[8] However, it may become apparent during reproductive years when the affected individuals may present with gynaecological concerns of infertility, and perhaps symptoms of dysmenorrhea.[8, 9]

The diagnosis of Uterine didelphys is usually made by ultrasonography, hysterosalpingography, and MRI.[3] Transvaginal ultrasound scan is the first-line imaging technique for evaluating the female reproductive tract and is most often the initial diagnostic modality of congenital genital malformations, including uterine didelphys.[10]However, the initial identification of these anomalies oftentimes necessitates further, and more precise evaluation.[10] Magnetic Resonance Imaging (MRI), with its superior soft tissue contrast and multiplanar capabilities, emerges as an important investigative modality that effectively complements the ultrasound findings, providing a more definitive diagnosis of this condition.[10]

Aside from the possible gynaecological challenges and symptoms associated with uterine didelphys, pregnancies in women with uterine didelphys are considered to be high risk due to potential complications of preterm delivery, intrauterine growth restriction, fetal abnormalities, abnormal lie, and perhaps fetal death.[3, 5]

Interestingly, not fewer than 40% of pregnancies in women with uterine didelphys end in spontaneous miscarriage.[11]Hence, early recognition of the anomaly and prompt implementation of prenatal care and support might enhance the preservation of the pregnancy and quality of neonatal life at birth.[12,13]

This case report unveils the discovery of uterine didelphys during a repeat caesarean section in a multiparous woman. Notably, this anomaly had remained undiagnosed despite a previous caesarean delivery and a history of adverse obstetric outcomes, including a first-trimester spontaneous abortion and a preterm birth resulting in early neonatal death.

CASE REPORT

A 32-year-old gravida 4 para 2 plus 1, 1 alive, booked woman at estimated gestational age of 37 weeks. Who had a repeat elective caesarean section at our facility on account of breech presentation with 1 previous caesarean section scar. She booked for antenatal care at about 16 weeks ' gestational age. The index pregnancy was desired and spontaneously conceived, diagnosed by serum pregnancy test and confirmed by pelvic ultrasound scan. Antenatal care follow-ups were well-attended, with no adverse events identified.

She has had 2 confinements in the past. Her first pregnancy was in 2019, and she had an emergency caesarean section on account of severe oligohydramnios. She was then delivered of a live female neonate with good APGAR Scores and a birth weight of 3.8kg. The child is alive and well. She had a spontaneous miscarriage in 2022 at an estimated gestational age of 8weeks. She had another pregnancy in 2022, had spontaneous vaginal delivery at 35weeks gestational age, the baby suffered early neonatal death.

An ultrasound scan done in her first pregnancy showed evidence of uterine didelphys, which was however, not recognised in her first Caesarean section. Several other pelvic ultrasound scans done in the past did not show uterine didelphys, and in the index pregnancy. At last ANC visit, abdominal examination revealed symphysio-fundal height of 38weeks size, longitudinal lie and breech presentation, with no palpable contractions, fetal heart rate of 136 beats per minute. It was also confirmed by an obstetric ultrasound scan. She was counselled for elective caesarean section.

Pre-operative packed cell volume was 38%. She had a unit of blood grouped and cross-matched for the surgery. Informed consent was obtained, and pre-operative anaesthetic review was done.

Intraoperative findings showed a live female neonate, delivered via breech extraction, with good APGAR scores. Postero-fundal placenta was delivered via cord

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traction. Following delivery of the baby and exteriorization of the uterus, a single fallopian tube and a left-located normal ovary were identified. This prompted further inspection, and a second uterus was found deep in the pelvis, to the right of the initial hemiuterus, slightly bulky, with a single fallopian tube on the right and a normal ovary. It has no connection with the second uterus (complete separation between the two uterine cavities). She also has two cervices, each uterus with its own cervical canal. The uterus where the fetus was delivered had two fibroid nodules, measuring 2cm by 2cm in the largest diameter. Speculum examination excluded vaginal septum. Vital signs were stable, and she was discharged on the fourth post-operative day.

DISCUSSION

Partial or complete failure of embryological fusion of the Müllerian ducts often leads to a spectrum of genital tract malformations.[1] These anomalies frequently remain undetected until they become symptomatic during puberty or the reproductive years, or are incidentally identified during radiological imaging for unrelated concerns.[14] One of the resultant anomalies of this non-fusion is the uterine didelphys, which is associated with potential reproductive challenges and significant obstetric complications.[12]

In the presented case, the definitive diagnosis of uterine didelphys was made incidentally during a repeat caesarean section. Despite the patient's history of a prior caesarean delivery and the utilization of sonographic imaging in both previous and current pregnancies, the anomaly remained undetected. This underscores a well-documented challenge in the management of uterine didelphys—its frequent underdiagnosis or misdiagnosis, especially in settings with limited imaging resources or insufficient diagnostic expertise.[12]

The diagnostic sensitivity of transvaginal ultrasound (TVS), although commonly employed as the first-line imaging modality, may not always be sufficient to accurately differentiate UD from other Mullerian anomalies like bicornuate or septate uteri.[10] Furthermore, uterine anomalies can be entirely overlooked, particularly when imaging is performed in the mid to late stages of pregnancy, as the enlarging gravid uterus may obscure vital anatomical landmarks.[15] In this patient, the repeated failure of antenatal ultrasounds to identify the anomaly underscores the diagnostic limitations of conventional 2D ultrasonography, especially later in gestation. More advanced imaging techniques, such as three-dimensional (3D) ultrasound and magnetic resonance imaging (MRI), have demonstrated improved diagnostic accuracy, with MRI providing superior soft tissue contrast and the ability to delineate uterine anatomy in multiple planes.[10,15,16]

Notably, the obstetric history of the patient presented aligned with several documented adverse outcomes associated with UD. Her history includes a first-trimester miscarriage and a preterm delivery resulting in early neonatal death. These complications are consistent with existing literature, which reports elevated rates of recurrent miscarriage, fetal malpresentation, intrauterine growth restriction (IUGR), and preterm birth in women with UD.[3,5,15] The breech presentation, which was the indication for the repeat caesarean section in this case, is particularly a common finding in pregnancies complicated by UD due to the reduced intrauterine space and altered uterine contour.[15] Additionally, the presence of fibroid nodules within one of the hemi-uteri could potentially impact uterine compliance and placental function.

The missed diagnosis during the patient's first caesarean section suggests a lack of systematic intraoperative assessment. Such oversights can occur when the primary focus during surgery remains solely on foetal delivery, without a comprehensive exploration of uterine morphology. This case therefore emphasizes the critical need for heightened intraoperative vigilance, particularly in women with a history of adverse obstetric outcomes or previous imaging findings suggestive of uterine anomalies. Routine exteriorization of the uterus during caesarean delivery, combined with a thorough inspection of adnexal structures and uterine contours, could facilitate the earlier recognition of such anomalies.

While UD inherently raises concerns about fertility and pregnancy outcomes, it is important to recognise that successful term pregnancies, as achieved in this patient, are indeed possible and better than other variants of mullerian duct anomalies.[3] Nevertheless, optimal obstetric management necessitates early and accurate diagnosis, individualised prenatal care, and meticulous intrapartum planning to mitigate potential risks. The absence of a longitudinal vaginal septum in this patient is noteworthy, as this feature often coexists with UD and can introduce additional complexities during both delivery and gynecological management.[9]

For future care, comprehensive counseling for this patient regarding the nature of her anomaly and its

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potential implications for subsequent pregnancies is essential. Preconception evaluation, potentially including advanced imaging techniques, should be considered prior to any future pregnancies to allow for informed risk stratification and tailored obstetric care planning.

Implications of the finding

This case highlights the need for surgeons to imbibe the culture of thorough exploration of the pelvic visceral for any identifiable anomaly during Caesarean section and perhaps other pelvic surgeries irrespective of prior multiple pregnancies or C-section, as a rare condition like uterine didelphys (UD) can go undiagnosed. This underscores the need for high index of suspicion, especiallywhen there are positive history of unexplained pregnancy issues like miscarriage or preterm birth. The fact that the diagnosis was missed during the first caesarean section, even after a prenatal ultrasound suggested it, points to a major gap in practice.

The policy of pelvic visceral exploration during caesarean section will be far reaching in timely diagnosis of (UD) if integrated in the standardised checklists for C-sections. These lists would prompt surgeons to perform a comprehensive uterine and adnexal inspection, ensuring that nothing is missed. This simple change could prevent similar oversights in the future.

This report is a wake-up call for all obstetricians to refresh their knowledge on rare congenital anomalies and a reminder that a patient's medical history, no matter how seemingly straightforward, can hide a complex story.

CONCLUSION

This case reinforces the importance of maintaining a high index of suspicion for Müllerian anomalies, even in multiparous women with previous caesarean deliveries. It underscores the need for thorough intraoperative evaluation and, when possible, preoperative imaging with high-resolution techniques in women with unexplained adverse pregnancy outcomes or suggestive antenatal findings. Timely diagnosis of UD can significantly influence counselling, prenatal management, and delivery planning, ultimately improving maternal and neonatal outcomes.

Declarations

Ethical Consideration: Patient consent was sought to report the case for the scientific knowledge and value to practice

Authors' Contribution: Ogunlaja IP-conceptualized the study and contributed to the initial write-up of the case report. drafted the discussion and reviewed the report. Abdullahi MO- Sought patient consent for pictorial evidence and drafted the summary of the case. Olasinde A- prepared the detailed description of the study, reviewed the relevant literature, and reviewed the report for publication adapted for editor consideration. Oladipo A contributed to the writing of the case report. Zainab Abdulkadri contributed significantly to the critical review and revision of the manuscript.

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