

EC CLINICAL AND MEDICAL CASE REPORTS

Case Report

Successful Pregnancies in a Woman with Uterine Didelphys: A Case Report

Eweputanna LI^{1*}, Otuka OAI², Ezirim EO³, Okoye HC⁴ and Eke-Otuka UN⁵

- ¹Department of Radiology, Abia State University Teaching Hospital, Aba, Nigeria
- ²Department of Surgery, Abia State University Teaching Hospital, Aba, Nigeria
- ³Department of Obstetrics and Gynecology, Abia State University Teaching Hospital, Aba, Nigeria
- ⁴Department of Hematology and Immunology, University of Nigeria Teaching Hospital, Enugu, Nigeria
- ⁵Department of Obstetrics and Gynecology, Federal Medical Centre, Asaba, Delta State, Nigeria

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Abstract

Mullerian duct anomalies (MDAs) are congenital defects involving the paramesonephric ducts, occurring early in fetal life. Uterine didelphys or double uterus is one such rare embryological malformation of the Mullerian duct. This case report highlights the successful pregnancies of a 35-year-old woman diagnosed with uterine didelphys, a condition often associated with adverse reproductive outcomes. The diagnosis was made incidentally via ultrasound during an unrelated investigation. Subsequent abdominopelvic magnetic resonance imaging confirmed the presence of the double uterus and ruled out other associated genitourinary congenital anomalies. This case underscores the importance of comprehensive anatomical evaluation in women presenting with MDAs and provides hope for successful pregnancies in the presence of this uncommon condition. It further emphasizes the need for clinicians to possess a detailed understanding of the diverse presentations and implications of MDAs to provide appropriate counseling and management strategies.

Keywords: Uterine Didelphys; Successful Pregnancy; Ultrasound; Magnetic Resonance Imaging

Introduction

Mullerian duct anomalies (MDAs) are a diverse group of congenital abnormalities that result from aberrant embryological development of the paramesonephric ducts (also known as Mullerian ducts), which typically fuse together by the 18th day of fetal life [1]. These anomalies can manifest in various ways, with a spectrum of clinical presentations, implications, and potential complications. One of the most distinctive MDAs is uterine didelphys, or a double uterus, which is a result of the complete failure of the Mullerian ducts to fuse [2].

MDA may result from complete agenesis, defective vertical or lateral fusion, or resorption failure [3]. The incidence of MDA is 0.1 - 3% of all women [4,5]. Uterine anomalies are found in 3 - 15% of women with infertility/repeated spontaneous abortion. 25% of women with uterine abnormalities have fertility problems [4]. Uterine anomalies are frequently associated with urinary tract anomies in 20 - 50% and possibly increased familial occurrences of limb reduction [3,4]. Patients with Mullerian duct anomalies are at the risk of spontaneous abortion, prematurity, intrauterine growth retardation, abnormal fetal lie and shoulder dystocia.

^{*}Corresponding Author: Eweputanna LI, Department of Radiology, Abia State University Teaching Hospital, Aba, Nigeria.

According to American fertility society classification [6] there are nine categories of MDA. Uterine didelphys, previously type III MDA, results from failure of fusion of paramesonephric duct at 9 weeks of gestation resulting in complete duplication with two vagina and 2 cervices as well as 2 uterine horns.

It is a congenital uterine anomaly characterized by complete duplication of the uterine horns and cervix with no communication between them. It accounts for 11% of uterine anomalies. It is associated with 2 widely spaced uterine corpora with normal zonal anatomy or volume each with a single fallopian tube. Separate widely divergent uterine horns, wide fundal cleft, cervical duplication, and longitudinal vaginal septum in about 75%, unilateral hemivaginal septum, obstructing transverse vaginal septum between upper one-third and low two-third of vagina connecting to longitudinal septum commonly with ipsilateral renal agenesis, opacification of single deviated horn on HSG. Common complications are unilateral hydo/hematocolpus (if transverse vaginal septum is present) with reflux endometriosis.

Uterine didelphys is considered to be one of the rarest forms of MDAs, with a reported prevalence ranging from 0.1% to 3.8% in the general population [7]. Although this condition has been associated with various reproductive challenges, such as infertility, recurrent miscarriage, and preterm birth, successful pregnancies are indeed possible [8].

In this report, we described the unique case of a 35-year-old woman with uterine didelphys, who achieved successful pregnancies. The anomaly was incidentally discovered during an ultrasound examination for an unrelated issue, and was later confirmed by abdominopelvic magnetic resonance imaging (MRI) which revealed the presence of a double uterus without any other associated genitourinary congenital anomalies.

Case Presentation

Mrs CM, a 35-year-old multiparous woman (P2+0 A2) trader with last menstrual period 10 days prior to presentation who presented on referral from a private hospital with one month complain of lower abdominal pain. The pain was sudden in onset and located on the left lower quadrant of the abdomen. It was sharp, non-radiating, temporarily relieved by over-the-counter drugs. There was no correlation of her menses, there was prior fever but no abnormal vaginal discharge.

She has had no prior history of complaints, no prior abdominal surgery, trauma, no history of vaginal instrumentation. There was no history of lower urinary tracts symptoms, no change in bowel habits, no history of multiple sexual partners. She was placed on oral anti-biotics by referring clinic with resolution of fever but pain has persisted though now moderate to mild.

She attained menarche at the age of 13 years with a menstrual flow of 4 days in a regular 28 day cycle. There is no history of dysmenorrhoea, heavy menstrual flow or dyspareunia. She is aware of contraception but practices withdrawal method. There is no prior history of treatment for sexually transmitted diseases. Age at coitarche was 31 years.

Her first pregnancy was conceived 6 months after marriage which was spontaneously achieved, supervised in a maternity, was carried to term and delivered via spontaneous vaginal birth to a live male neonate that cried at birth with a birth weight of 3.2 kg. Baby was predominantly breast fed and immunized for age. The puerperium was uneventful. There was no scan done in this pregnancy.

Her second pregnancy was conceived 16 months after the first. It was spontaneously achieved and supervised in a maternity. She was treated for malaria at 17 weeks and 26 weeks gestational age (GA) with artemisinin-based combination therapy (ACT)s and had a scan done at 32 weeks GA following a complaint for reduced fetal movement which was said to be normal. Pregnancy was carried to term and she had a spontaneous vaginal delivery of a live female neonate that cried at birth who weighed 3.3 kg. Baby was predominantly breast fed and immunized for age.

Other aspects of history were essentially normal.

On examination, vaginal examination showed positive cervical excitation motion tenderness for which working diagnosis of pelvic inflammatory disease was made for which she was asked to do an abdominopelvic scan amongst other investigations: high vaginal swab for microscopy, culture and sensitivity (m/c/s), endocervical swab m/c/s, Urine m/c/s.

Her abdominopelvic scan showed two uterine horns with adnexal structures shown to be within normal limits and no fluid in the pouch of Douglas. An impression of? uterine didelphys was made and an MRI was advised.

The patient received treatment along antibiotic sensitivity from the culture result and was sent for MRI following adequate counselling on the benefit of MRI over other modalities as it will help identify other abnormalities if present.

Uterine didelphys was seen on ultrasonography. She was sent for pelvic MRI and the result is as follows.

Uterus: There was duplication of the uterus due to non-fusion of the uterine horns. The uterine horns were equal in size and fusiform in shape. They measured 88 mm x 45 mm and 86 mm x 46 mm on the right and left sides respectively (Figure 1 and 2). Each has a single fallopian tube arising from the apices of the horns. The endometrial cavities were normal (Figure 3). The junctional zones were preserved. The myometrial layers were also preserved. No myoma was seen. Between the uterine horns was a soft tissue as shown in figure 4. It was triangular in shape measuring 374 mm x 509 mm in the largest transverse and an anteroposterior planes. There were also two distinct cervical canals. There was a single vagina which was normal in size and outline.

Adnexa: Both ovaries were located in their respective pelvic side walls. They were normal in volume, outline, and intensity. A leading follicle was identified on the right.

There was no free fluid seen in the pouch of Douglas.

The urinary bladder and rectum were normal in outline and intensity.

Both kidneys were normal in position, size, outline, and orientation. They were adequate cortical thickness. Corticomedullary differentiation was preserved. The results of the investigations were relayed to the patient and the requesting doctor. The patient was counseled about plans for further pregnancy and had her fears allayed.



Figure 1: Axial view of the pelvis. Arrows point to uterine horns.

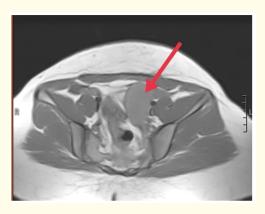


Figure 2: Red arrow points at the left uterine horn.

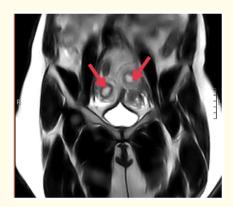


Figure 3: Coronal section of the pelvis. The red arrows point at the endometrial cavities.

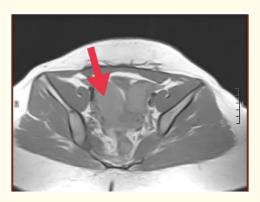


Figure 4: MRI axial section of the pelvis showing the right uterine horn (red arrow) and separating triangular shaped soft tissue (blue arrow).

Discussion

This case presents a fascinating, albeit unusual, example of successful pregnancies in a woman with uterine didelphys, a congenital Mullerian duct anomaly (MDA). It is crucial to stress the rarity of this condition, with incidence rates of MDAs varying from 0.1 to 3% in the general population [4,5] and uterine didelphys accounting for approximately 8% of these cases. Nevertheless, the majority of women with MDAs are asymptomatic and often undiagnosed until an incidental finding or the presence of fertility or obstetric complications triggers a further examination. These anomalies occur due to a failure in the fusion of the paramesonephric ducts during fetal development, normally completed by the 18^{th} day of fetal life [1,6].

In the present case, the double uterus was diagnosed via ultrasound conducted for left lower abdominal pain underscoring the silent nature of this condition. Following up with abdominopelvic magnetic resonance imaging not only confirmed the initial diagnosis but also ruled out other genitourinary congenital anomalies, highlighting the diagnostic power of this tool in assessing the full spectrum of MDAs. While ultrasound is commonly used for initial evaluation, MRI is more precise in diagnosing MDAs, as it provides a more detailed visualization of uterine and vaginal structures [11]. This reinforces the role of comprehensive imaging studies like MRI in confirming the diagnosis and ruling out other associated genitourinary anomalies.

Uterine didelphys can lead to complications, such as an increased risk of miscarriage, premature birth, and malpresentation, among others, which can potentially affect the course of pregnancy and delivery [12,13].

The aspect of successful pregnancies in this woman with uterine didelphys is particularly noteworthy. Literature indicates that women with uterine didelphys can have successful pregnancies, though often with increased monitoring and possible interventions due to the aforementioned complications. Thus, this case reinforces the current body of knowledge indicating that while uterine anomalies like didelphys can pose challenges, they do not necessarily preclude successful pregnancies. It further reinforces the fact that each case can have a varying clinical presentation and prognosis.

Diagnosis of uterine didelphys typically involves imaging modalities like ultrasound and MRI. In our case, ultrasonography initially detected the anomaly, while abdominopelvic MRI confirmed the diagnosis and ruled out associated genitourinary anomalies, which are commonly seen in 25% - 50% of cases with Müllerian duct anomalies [14]. This emphasizes the importance of thorough investigation in these patients to avoid overlooking related abnormalities, thus enabling comprehensive management.

Management of uterine didelphys largely depends on symptomatology and the patient's desire for fertility. In asymptomatic cases like our patient, expectant management is the norm. However, counseling on potential pregnancy-related complications should be undertaken, as in this case [15,16]. In cases where fertility is desired, assisted reproductive techniques may prove helpful.

This present case highlights the complementary role of different imaging modalities in evaluating patients with known or suspected MDAs. As we see in this case, a combination of ultrasound and abdominopelvic magnetic resonance imaging was crucial in the diagnosis of patient's condition. It also highlights the necessity for personalized care, considering the unique anatomy and potential complications associated with MDAs.

While several case reports discuss pregnancy outcomes in women with MDAs, the factors influencing these outcomes remain poorly understood. Our case adds to this growing body of evidence and highlights the need for further investigation into the molecular and physiological mechanisms underlying MDAs and the associated reproductive outcomes.

Conclusion

A high index of suspicion, along with effective use of diagnostic modalities like ultrasound and MRI, are essential for accurate diagnosis of uterine didelphys. Each case is unique and requires a tailored approach to management and counseling. The successful conception and

23

delivery in this case highlight the potential for normal reproductive function in women with uterine didelphys. It also emphasizes the importance of accurate diagnosis and comprehensive understanding of such congenital anomalies, as this can provide necessary information for counseling and management of reproductive issues in women affected by similar malformations. Further research is needed to fully elucidate the implications of these anomalies on fertility and pregnancy outcomes. This report should serve as a reassuring reference for women with similar conditions who wish to conceive, and for the clinicians involved in their care.

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Conflict of Interest

There is no conflicting interest to declare.

Bibliography

- 1. Acién P., et al. "Müllerian anomalies "without a classification": from the didelphys-unicollis uterus to the bicervical uterus with or without septate vagina". Fertility and Sterility 91.6 (2019): 2369-2375.
- 2. Rock JA., *et al.* "Pregnancy success following surgical correction of imperforate hymen and complete transverse vaginal septum". *Obstetrics and Gynecology* 59.4 (2022): 448-451.
- 3. Behr S., et al. "Imaging of Mullerian Duct Anomalies". Radiographics 32.6 (2012): E233-E250.
- 4. Dahnert W. "Radiology Review Manual". 7th Edition. Walters Kluwer/Lippincott Williams and Wilkins. Chapter 2 Central Nervous System (2017): 269-272.
- 5. Troiano R and McCarthy S. "Mullerian Duct Anomalies: Imaging and Clinical Issues". Radiology 233.1 (2004): 19-34.
- 6. The American Fertility Society classifications of adnexal adhesions, distal tubal occlusion, tubal occlusion secondary to tubal ligation, tubal pregnancies, müllerian anomalies and intrauterine adhesions". *Fertility and Sterility* 49.6 (2018): 944-955.
- 7. Raga F., et al. "Reproductive impact of congenital Müllerian anomalies". Human Reproduction 12.10 (2017): 2277-2281.
- 8. Heinonen PK. "Clinical implications of the didelphic uterus: long-term follow-up of 49 cases". European Journal of Obstetrics, Gyneacology and Reproductive Biology 91.2 (2020): 183-190.
- 9. Chan YY., et al. "Reproductive outcomes in women with congenital uterine anomalies: a systematic review". *Ultrasound in Obstetrics and Gynecology* 38.4 (2011): 371-382.
- 10. Miller DS and Diaz-Arrastia DP. "Müllerian anomalies and their effect on pregnancy outcome". *American Journal of Obstetrics and Gynecology* 207.4 (2012): 293-300.
- 11. Troiano KL and McCarthy RN. "Müllerian Duct Anomalies: Imaging and Clinical Issues". Radiology 233.1 (2004): 19-34.
- 12. Saravelos SH., et al. "Prevalence and diagnosis of congenital uterine anomalies in women with reproductive failure: a critical appraisal". *Human Reproduction Update* 14.5 (2008): 415-429.
- 13. Robboy SJ., *et al.* "The Developing Human Female Reproductive Tract: A 21st-century perspective". *Endocrine Reviews* 39.4 (2018): 383-428.

- 14. Raga F, et al. "Reproductive impact of congenital Müllerian anomalies". Human Reproduction 12.10 (2017): 2277-2281.
- 15. Grimbizis GF., et al. "Clinical implications of uterine malformations and hysteroscopic treatment results". Human Reproduction Update 7.2 (2021): 161-174.
- 16. Pfeifer SM., et al. "ASRM Mullerian anomalies classification". Fertility and Sterility 116.5 (2021): 1238-1252.

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