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(CASE REPORT)

Double pregnancy in Didelphys Uterus

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Abstract

Uterus didelphys is a congenital malformation of the Müllerian duct that occurs due to complete failure of the ipsilateral ducts. Müllerian duct anomalies have a prevalence of 2-3%, with 11% and 43% having uterus didelphys and renal anomalies, respectively. We want to report the case of a patient with uterus didelphys and pregnancy in both uteri. Case presentation: A 26-year-old patient had a previous cesarean section in 2016 due to breech presentation. She was referred with a diagnosis of twin pregnancy at 26 weeks and 2 days. The ultrasound and the report indicated uterus didelphys with one fetus in each uterus. On examination, the speculum showed two cervices, but no septum was observed. The patient was treated as a normal twin pregnancy until 34 weeks. After lung maturation, the patient was scheduled for elective cesarean section. A 2600g female baby girl with an Apgar score of 8/8 and placental weight of 415 g was delivered. An 1840 g female baby girl with an Apgar score of 9/9 and placental weight of 300 g was delivered. The patient and both babies performed well. Conclusions: Uterus didelphys is a rare anomaly and accounts for 8% of congenital anomalies of the female reproductive tract, often undiagnosed. It occurs in 0.3% of the total population and 0.1% to 0.5% of the healthy fertile population. In most cases, this anomaly is asymptomatic and is detected accidentally, which probably explains the inaccurate assessment of its frequency.

Keywords: Didelphys; Multiple Pregnancy; Previous Cesarean Section; Müllerian Duct

1. Introduction

Uterus didelphys is a congenital malformation of the Müllerian duct that results from the complete failure of the ipsilateral ducts. In the general population, Müllerian duct anomalies have a prevalence of 2–3%, with 11% and 43% having a uterus didelphys and renal anomalies, respectively.

Congenital uterine anomalies result from the abnormal formation, fusion, or resorption of the Müllerian ducts during embryological development. Their prevalence is estimated at 5.5% in the general population, 13.3% in women with a history of miscarriage, and up to 24.5% in patients with a history of infertility and miscarriage. In fertile women, the incidence of Müllerian anomalies is estimated at 1 case in every 200 to 600. [1]

Currently, there is no universally accepted system for Müllerian anomalies; however, different systems have been proposed throughout history. [2]

The most up-to-date is the new proposal from the American Society of Reproductive Medicine in 2021. This is the most recent classification system, which retains the simplicity of the previous system proposed in 1988 and seeks to

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strengthen its weaknesses. It is, therefore, a system of nine descriptive categories (Müllerian agenesis, cervical agenesis, unicornuate uterus, uterus didelphys, bicornuate uterus, septate uterus, longitudinal vaginal septum, transverse vaginal septum, and complex anomalies), which opens the possibility of some anomalies appearing in more than one category to allow classification of anomalies with combined elements. The guide to this system offers a virtual example, with educational elements to help medical personnel approach patients. [3]

3D transvaginal ultrasound has the highest sensitivity, ranging from 92% to 100%, and overall diagnostic accuracy. This technique provides coronal visualization of the uterus, which is ideal for diagnosing Müllerian anomalies by this route. The combination of hysteroscopy and laparoscopy remains the gold standard for diagnosis because it allows evaluation of the endometrial cavity and uterine contour, although the procedure is invasive. [4,5]

A bicornuate uterus is a risk factor for gestational hypertension and preeclampsia. The increased risk of hypertensive disorder in women with uterine anomalies is related to underlying renal malformations [6]. Our case did not present renal impairment, although she did suffer from chronic hypertensive disease.

For this reason, we decided to report the case of a patient with a didelphys uterus and pregnancy in both uterine bodies.

2. Case report

A 26-year-old patient, gravida 2 para 1 (previous caesarean section in 2016 due to breech presentation). Class 2 obesity, with a diagnosis of chronic hypertension treated with methyldopa 500 mg every 8 hours, but well controlled. No other comorbidities.

The patient was referred to our institution with a possible diagnosis of multiple pregnancies, at 26 weeks 2 days by the date of her last menstrual period. Physical examination revealed a fundal height of 39 cm and a uterus that appeared gravid without palpable contractions. A vaginal examination was not performed, and a speculum was placed, where two cervices were observed without a vaginal septum. An obstetric and abdominal ultrasound was performed, confirming the diagnosis of a didelphys uterus, with no renal abnormalities, and the presence of two fetuses in each uterine cavity. The gestational age was approximately 29 weeks 2 days for the first fetus and 26 weeks 4 days for the second fetus.

The patient was immediately admitted to the antenatal care unit for follow-up and monitoring. Betamethasone steroids were administered 12 mg intramuscularly every 12 hours, two doses, and the patient was treated as a di-chorionic diamniotic twin pregnancy until 34 weeks, when lung maturity was reached. It was decided to prepare the patient for elective caesarean section.

2.1. Surgical procedure

Under general anesthesia, a midline infraumbilical incision is made and two gravid uteri are observed (Fig 1)

Figure 1 Visualisation of two uteruses

A lower segmental incision was made in the right uterus. Clear fluid was observed, and a female fetus weighing 2600 g was extracted, with an Apgar score of 8/8 and a placental weight of 415g. The same procedure was subsequently

performed on the left side. A clear fluid was observed, and a female fetus weighing 1,840 g was extracted, with an Apgar score of 9/9 and a placental weight of 300g.

Figures 2 and 3 Visualization of both uterine cavities with placentas in situ

Both histerorrhaphies were performed in two layers (Figure 4) using 1.0 Vycril suture, and the abdominal cavity was closed in layers down to the skin (Figure 5). The procedure lasted approximately 36 minutes, and the estimated blood loss was approximately 800 ml.

Figure 4 Histerorrhaphy

Figure 5 Closure of the abdominal cavity

Figure 6 Both babies after the procedure

3. Results and discussion

Early diagnosis of the condition is a very important factor, allowing, in cases of gestating uterus didelphys, adequate prenatal care, carefully considering the potential complications to which the patient with these anomalies is exposed and being prepared to successfully resolve them. Another benefit of early diagnosis, no less important, is avoiding an erroneous diagnosis, which is sometimes only rectified in the operating room, with the performance of a cesarean section. [7]

Patients may be asymptomatic, but the anomaly may be associated with dysmenorrhea, dyspareunia, infertility, spontaneous abortion, premature delivery, fetal mal-presentation, intrauterine growth restriction (IUGR), Premature rupture of membranes, renal agenesis, decreased live births, and cesarean delivery [8-13]. The incidence of cesarean delivery in uterine didelphys can be as high as 82% [9]

Multiple pregnancies are very rare. The usual presentation is twins in each horn separately. Most multiple pregnancies with a didelphys uterus result in premature delivery. The incidence of triplets with a didelphys uterus is 1 in 25 million [14]

The case presented in this study was operated on for breech presentation in 2016, but outside the province, and previous records were never found. Another aspect to consider is that our institution is at the secondary level, and most patients are referred when there is a complication. In this case, the patient was referred for a possible diagnosis of twin pregnancy, which is usually treated at this level.

Another interesting finding is the difference in the weight of both fetuses. This could be due to independent intercourse, i.e., on different days, IUGR in the second fetus, or even from different fathers. This aspect was ruled out subjectively, given that the patient confirmed that she only had intercourse with the same partner.

Uterus didelphys is associated with poor reproductive outcomes, with only a 20–30% chance of carrying a pregnancy to term. However, we present a case of a successful, full-term pregnancy with one fetus in each uterine body. [15-18]. Most data on the clinical significance and outcomes of this uterine anomaly are based on small retrospective, observational, or case-reported studies. The results of these studies are of mixed significance due to the low incidence of uterus didelphys in the population and the small number of cases. Therefore, further studies are warranted. [19]

4. Conclusion

Uterus didelphys is a rare anomaly, representing a small percentage of cases in the general population and often undiagnosed. It occurs in 0.3% of the total population and in 0.1% to 0.5% of the healthy fertile population. In most cases, this anomaly is asymptomatic. Although our case presented menstruation twice a month, the absence of symptoms probably explains the incorrect assessment of its frequency.

Compliance with ethical standards

Acknowledgments

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Disclosure of conflict of interest

All the authors confirm that there are no conflicts of interest.

Statement of informed consent

For the presentation of this case, the authors requested the patient's consent, which was obtained verbally and in writing.

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