

# Atypical Obstruction Praevia: A Case Report and Review of the Literature

Mohamed Mahmoud Emed ,Ahmedna Elbechir , Karam Mohammed Saoud, Nisrine Mamouni, Sanae Errarhay, Chahrazed Bouchikhi, Abdelaziz Banani

Department of Gynecology and Obstetrics I, Hassan II University Hospital, Fes, Morocco

**Abstract:** A didelphic uterus is a congenital anomaly due to the lack of fusion of the mullerian ducts, resulting in the development of two uterine bodies, each with its own cervix, fallopian tubes and often a septal vagina; To draw the attention of clinicians, we report the case of a 21-year-old parturient, without any notable pathological history, primigravida primiparous woman referred from a birthing center for pelvic pain on a full-term pregnancy. On admission, the patient was algic with a uterus that was contracted on palpation. On vaginal touch, the cervix is lateralized to the left and dilated to one finger, the presentation is mobile cephalic. The water sac is intact with a sensation of a bulging mass at the level of the Douglas lateralized to the right. The emergency ultrasound noted a pregnancy of 37-38 weeks with a retro-cervical mass lateralized to the right of 10 cm × 8cm, enclosed in the cul-de-sac of Douglas whose echo structure favors a normal myometrium. An emergency caesarean section allowed the extraction of a male newborn, Apgar 10/10 and weight 3100 grams, on the basis of the information provided by the ultrasound, the surgeon decided to further explore the pelvic cavity after total exteriorization of the uterine mass. This procedure revealed a second non-gravid uterus that was enclosed in the cul-de-sac of Douglas which corresponded to the Prævia mass seen on ultrasound. Clinicians must have a high index of suspicion for uterine abnormality to make an early diagnosis of Didelphic uterus. A pregnancy in a Didelphic uterus deserves early diagnosis of the anomaly, and meticulous care during pregnancy and delivery to avoid the associated negative consequences.

## Introduction:

A didelphic uterus is a congenital anomaly due to the lack of fusion of the mullerian ducts, resulting in the development of two uterine bodies, each with its own cervix, fallopian tubes and often a septal vagina [1].

This congenital anomaly of the female reproductive system is associated with various reproductive difficulties: reduced possibility of natural or assisted conception, rate of miscarriage in the first and second trimester, premature birth, placental abruption, low birth weight and fetal growth retardation, poor presentation at delivery and perinatal mortality [2].

Among the mullerian duct anomalies (MDAs), septal uterus is the most common (35%), followed by bicornuate uterus (25%), arcuate uterus (20%), then unicornuate (9.6%), and complete agenesis (3%). The didelphic uterus is the second least common with an incidence of approximately 8.3% of all mullerian canal anomalies.

The prevalence of didelphic uterus is estimated to be 1 in 1,000 to 1 in 30,000 women [3]. Among the causes of prævia obstruction, the most frequent are masses (uterine fibroids, tuboovarian masses) and especially scars: surgical in the West or ritual in Africa. Scars account for 80-90% of cases [4].

More rarely, the cause may be another organ such as an ectopic kidney [5], an endometrial sarcoma or a uterine malformation [6-7]. In this last case, we report an observation of this exceptional etiology of prævia obstruction, underlining the interest of a careful ultrasound and MRI examination during the prenatal work-up, in order to draw the practitioner's attention to this possibility of prævia obstruction.

## Presentation of Case:

The 21 year old parturient, with no notable pathological history, primigravida primiparous referred from a birthing center for pelvic pain on a full term pregnancy. On admission, the patient was algic with a uterus that was contracted on palpation. On vaginal touch, the cervix is lateralized to the left and dilated to one finger, the presentation is mobile cephalic. The water sac was intact with a sensation of a bulging mass at the level of the douglas lateralized to the right. The tococardiography showed a basic fetal heart rate of 140 beats per minute with preserved variability and fetal reactivity. Emergency ultrasound noted a 37-38 week pregnancy with a right lateralized retro-cervical mass of 10 cm × 8 cm, embedded in the cul-de-sac of Douglas with echo structure in favor of a prævia obstruction by a probable posterior cervico-isthmic pedicle myoma or uterine hemibody. An emergency caesarean section allowed the extraction of a male newborn, Apgar 10/10 and weight of 3100 grams, on the basis of the information provided by the ultrasound, the surgeon decided to further explore the pelvic cavity after total exteriorization of the uterine mass. This procedure revealed a second non-gravid uterus that was enclosed in the cul-de-sac of Douglas which corresponded to the Prævia mass seen on ultrasound (Figure 1).

Pelvic exploration established the diagnosis of a didelphic uterus with two hemibodies (Figure 2), two open necks in a vaginal cavity. The postoperative course was simple. Currently, there are different classification systems for the categorization of congenital malformations of the reproductive system. The oldest and most commonly used classification is the 1988 American Society for

Reproductive Medicine (ASRM, formerly the American Fertility Society). The ASRM classification divides mullerian duct anomalies into seven main types based on the anatomical changes of the uterus and the embryonic processes responsible for them. However, it does not account for complex urogenital malformations [15]. In 2013, the European Society of Human Reproduction and Embryology (ESHRE) and the European Society of Endoscopy(ESGE) published a classification of female genital anomalies. It is designed and developed primarily on the basis of anatomical findings. Abnormalities are classified into major classes and subclasses, separately reflecting anatomical anomalies and variations; uterine, cervical, and vaginal abnormalities are independently classified into subclasses. This congenital anomaly is identified by the ESHRE/ESGE classification system as U3b/C2 (complete bicorporeal uterus/"normal" double uterine neck)[8].

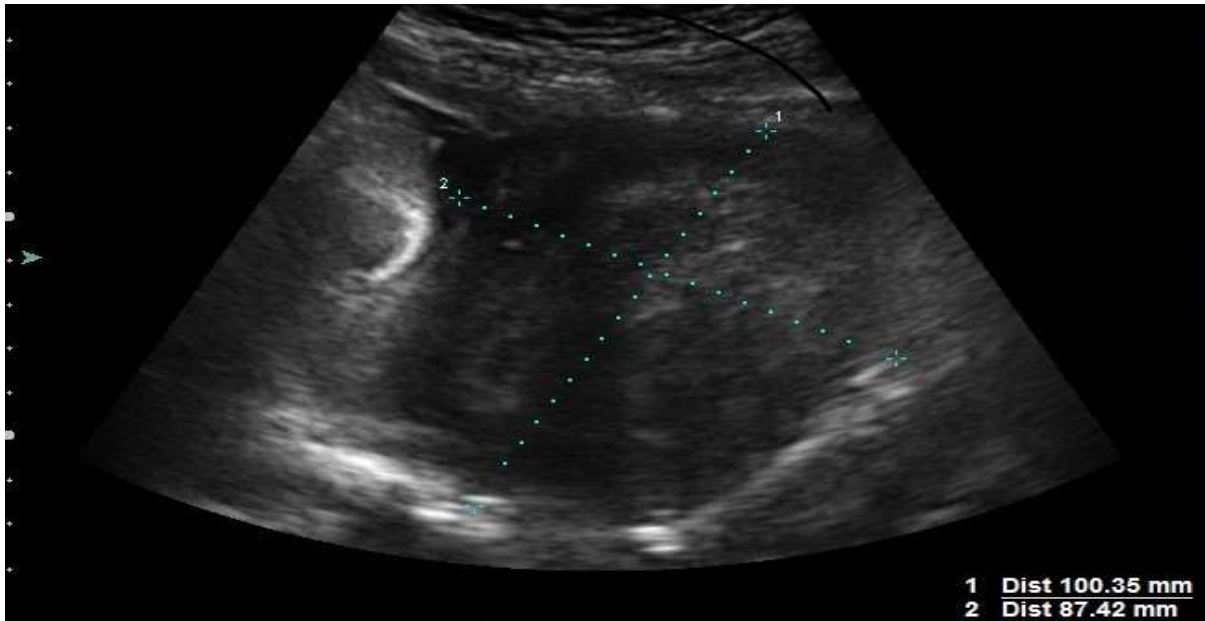
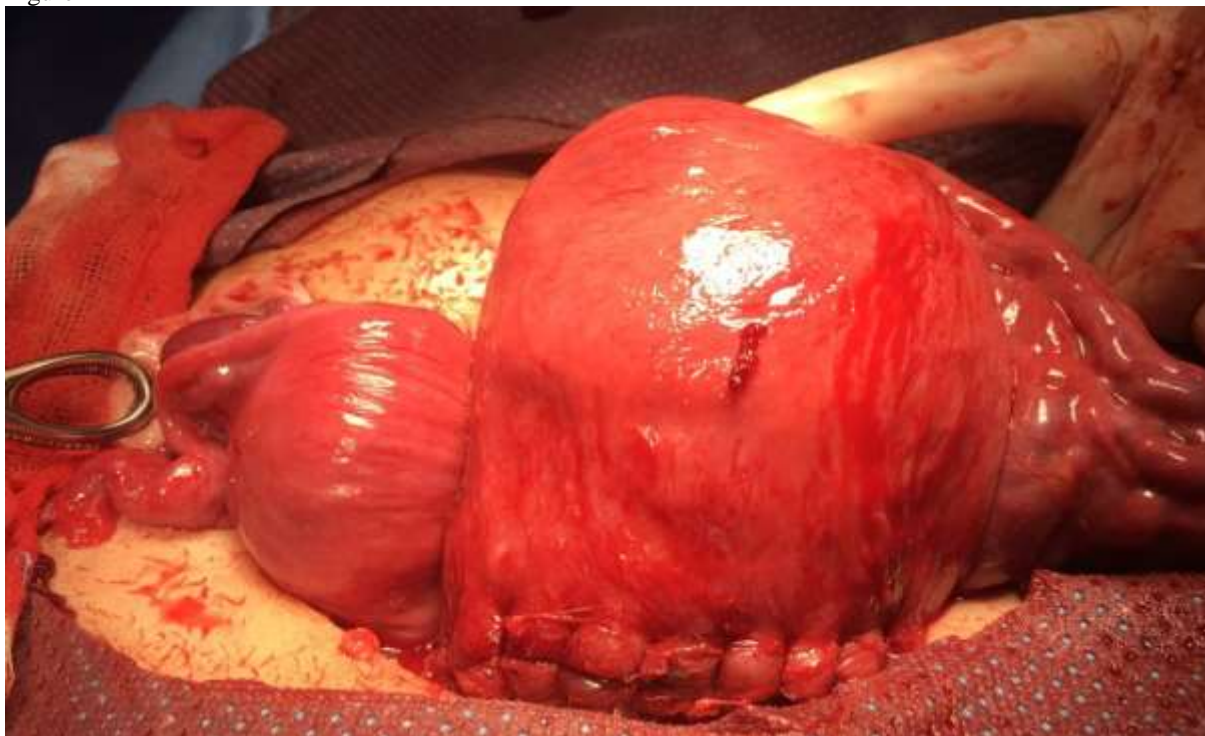


Figure 1



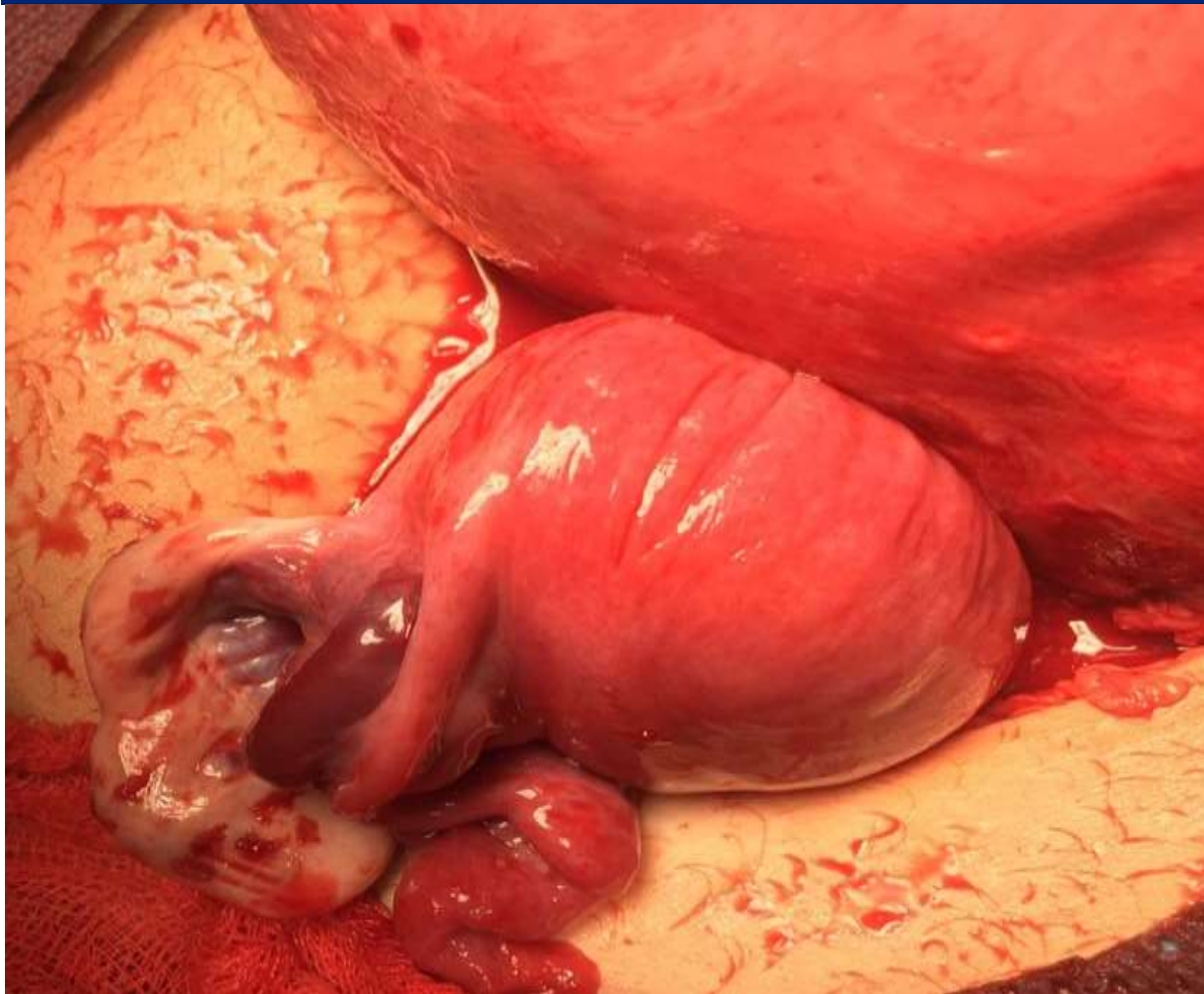


Figure 2

### Discussions:

The didelphic uterus is a rare anomaly and represents 8% of congenital anomalies of the female reproductive system [8]. It occurs in 0.3% of the total population. In the population of women with a history of abortion and infertility, its occurrence rate is more frequent, 2.1% [2].

Most often, this uterine malformation is asymptomatic and detected accidentally, which probably explains the inaccurate assessment of its frequency. Its clinical manifestations may present as pelvic discomfort, dyspareunia, dysmenorrhea, hematocolpos and hematometra [9]; and may be compatible with a pregnancy that will normally progress to term in about 50% of cases, and end with a normal delivery [10].

Heinonen PK (2000) [12], evaluated the long-term clinical consequences, and reproductive performance of 49 women with a Didelphic uterus who were followed for 6.3 years.

He found hemi-vaginal obstruction in 9 patients (18%) with 8 (16.3%) having ipsilateral renal agenesis. Five (13%) had primary infertility.

Thirty-four of 36 women (94%) who wanted to conceive became pregnant, 21% had a miscarriage and 2% had an ectopic pregnancy. The fetal survival rate was 75%, prematurity 24%, fetal growth retardation 11%, perinatal mortality 5.3%, and the cesarean delivery rate 84%. The pregnancy was located in the right uterus in 76% of cases.

Many patients with a Didelphic uterus have a normal sex life, pregnancy and childbirth[13]. Dystocia due to a prævia obstacle is mainly due to cervico-isthmic myomas; 1-5% of myomas [5]. However, ampliation of the lower segment at the end of pregnancy or at the beginning of labor, associated with spontaneous migration out of the pelvis of the segmental myoma, which then ceases to be prævia. This situation can also be observed with the non-gravid hemi-uterus in the case of didelphic uterus [10].

Pankaja et al. reported three cases of didelphic uterus and their pregnancy outcomes. Cesarean section was performed in two cases due to lack of labor progression and previous uterine surgery. The authors concluded that the didelphic uterus is associated with a high incidence of cesarean section due to labor dystocia [14].

Although the initial diagnosis of didelphic uterus is made by ultrasound or hysterosalpingography, MRI offers the most accurate diagnosis.

MRI is the best option for classification of different anomalies because of its better anatomical assessment compared to other diagnostic modalities [12]. An accurate diagnosis is essential to determine the most effective treatment during delivery.

The obstetrical complications of this malformation are numerous. The chances of reaching term for pregnancies with didelphic uterus are reported to be 20-30%. Premature rupture of the membrane; intrauterine growth retardation or fetal hypotrophy at birth; labor dystocia. Pregnancy is often complicated, and follow-up must be planned. Cesarean section is not systematic.

## Conclusion

Müller congenital anomalies are challenging case scenarios for obstetrician-gynecologists in terms of diagnosis and resolution of reproductive problems.

Prenatal obstruction by the non-gravid hemi-uterus on didelphic uterus is a rare cause of praevia obstruction; it is easily detected at ultrasound by a careful prenatal examination that takes into account not only the fetus but also the uterus and its appendages, the maternal urinary system. Clinicians must have a high index of suspicion for uterine abnormality to make an early diagnosis of Didelphic uterus. A pregnancy in a Didelphic uterus deserves early diagnosis of the anomaly, and meticulous care during pregnancy and delivery to avoid the associated negative consequences. Women with uterine anomalies are more likely to experience adverse pregnancy outcomes, which requires accurate knowledge, early diagnosis, and appropriate treatment.

## References

1. Al Yaqoubi HN, Fatema N. Successful vaginal delivery of naturally conceived Dicavitary twin in didelphys uterus: a rare reported case. *Case Rep Obstet Gynecol* 2017;2017:1-4.
2. Venetis, C.; Papadopoulos, S.P.; Campo, R.; Gordts, S.; Tarlatzis, B.C.; Grimbizis, G.F. Clinical implications of congenital uterine anomalies: A meta-analysis of comparative studies. *Reprod. Biomed. Online* 2014, 29, 665-683. [CrossRef] [PubMed]
3. O. Ozyuncu, M. Turgal, A. Yazicioglu, and A. Ozek, "Spontaneous twin gestation in each horn of uterus didelphys complicated with unilateral preterm labor," *Case Reports in Perinatal Medicine*, vol. 3, no. 1, 2014.
4. Koné M, Ba P, Diarra S. Soft tissue dystocia. *Encycl Méd Chir (Elsevier, Paris), Obstetrics*, 5-060-A-10, 1996, 7 p.
5. Samuel N, Feldberg D, Dicker, Goldman J. Pelvic kidney presenting as tumor praevia: report of a case diagnosed by ultrasound. *Europ J Obstet Gynec reprod Biol* 1985;20:325- 8.
6. Cooney MJ, Benson CB, Doubilet PM. Outcome of pregnancies in women with uterine duplication anomalies. *J Clin Ultrasound* 1998;26:3-6.
7. Cooney MJ, Benson CB, Doubilet PM. Outcome of pregnancies in women with uterine duplication anomalies. *J Clin Ultrasound* 1998;26:3-6.
8. Grimbizis, G.F.; Camus, M.; Tarlatzis, B.C.; Bontis, J.N.; Devroey, P. Clinical implications of uterine malformations and hysteroscopic treatment results. *Hum. Reprod. Update* 2001, 7, 161-174. [CrossRef] [PubMed]
9. Rezai, S.; Bislam, P.; Alcantara, I.L.; Upadhyay, R.; Lara, C.; Elmadjian, M. Didelphys Uterus: A Case Report and Review of the Literature. *Case Rep. Obstet. Gynecol.* 2015, 2015, 865821. [CrossRef] [PubMed]
10. Muller P, Barrat J. Pregnancy and delivery in cases of uterine malformations. In, Roger Vokaer, traité d'obstétrique. Ed, Masson Paris, 1985; volume II: 458-71.
11. Fukunaga, T.; Fujii, S.; Inoue, C.; Mukuda, N.; Murakami, A.; Tanabe, Y.; Harada, T.; Ogawa, T. The spectrum of imaging appearances of müllerian duct anomalies: Focus on MR imaging. *Jpn. J. Radiol.* 2017, 35, 697-706. [CrossRef] [PubMed]
12. Heinonen PK (2000), "Clinical implications of the didelphic uterus: long-term follow-up of 49 cases," *European Journal of Obstetrics & Gynecology and Reproductive Biology*, vol. 91, no. 2, pp. 183-190.
13. Mohd Subhail, Hina Khan, Sofia Suhail (2010). Uterus Didelphys having Single Pregnancy in her right horn: A Case Report. *Journal of Chinese Clinical Medicine* Volume 5 (1): 46-49.
14. Pankaja, S.; Ip, P.; O'Mahony, F. Successful Pregnancy with Uterus Didelphys. *J. Androl. Gynaecol.* 2016, 4, 3.
15. The American Fertility Society. The American Fertility Society classifications of adnexal adhesions, distal tubal occlusion, tubal occlusion secondary to tubal ligation, tubal pregnancies, Mullerian anomalies and intrauterine adhesions. *Fertil. Steril.* 1988, 49, 944- 955. [CrossRef]