

**SPONTANEOUS UTERINE RUPTURE IN A BICORNUATE BICERVICAL UTERUS
DISCOVERED INCIDENTALLY DURING DELIVERY HEMORRHAGING: A CASE
REPORT**

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ABSTRACT

The bicornuate bicervical permeable uterus is a relatively rare uterine malformation and is often diagnosed incidentally during an examination done for another purpose. It is asymptomatic and during pregnancy, this malformation may go unnoticed if no prenatal monitoring has been done. We report a case of a permeable bicornuate bicervical uterus discovered incidentally during a laparotomy for post-partum hemorrhaging on suspicion of uterine rupture. The interest of this case is to show the obstetrical prognosis in fertile women with this uterine malformation.

KEYWORDS: bicornuate bicervical uterus, delivery hemorrhage, uterine malformation, uterine rupture.**INTRODUCTION**

Most congenital uterine malformations are detectable at birth, others are diagnosed in the antepartum period and still others in puberty or adulthood. The latter often go unnoticed for a long time and are only discovered incidentally. Among them, we find uterine malformations, in particular bicervical uterus which can be discovered during an imaging examination or during an abdominopelvic surgery performed for another reason, or, as in our case, during a delivery bleeding. We report here a case of a permeable bicervical uterus discovered incidentally during a laparotomy for delivery hemorrhaging on suspicion of uterine rupture in a 38 years old multiparous woman. The interest of this case is to show the obstetrical prognosis in fertile women with this uterine malformation.

Patient and observation

Mrs. A.T, 38 years old, G4P4, with a history of 4 vaginal deliveries of pregnancies carried to term, the last delivery from an unmonitored pregnancy carried to term, dated back to 4 hours from the time of her admission to our structure. The patient was referred to our University Hospital from a birthing center for delivery bleeding. On admission, the patient was conscious, eupneic, pale, BP: 100/60 mm hg, HR : 115 beats/min, T : 37°, discolored conjunctiva. The obstetrical examination showed continuous reddish bleeding of endo-uterine origin, hence the realization of a uterine revision which objectified the presence of an anterior uterine rupture.

The patient was quickly taken to the operating room for an emergency laparotomy with concomitant reanimation. The exploration showed the presence of a large hemoperitoneum with two distinct hemi-uterus totally separated, with two bodies between them the posterior bladder wall is inserted (bladder V sign) each with annexes (ovary, fallopian tube, round and broad ligaments) on one side, as well as two isthmuses and two cervixes. The first one, on the left, is gravid with a well-formed lower segment on which the uterine rupture is located on the right third of the anterior face. The second one, on the right, is non pregnant, enlarged and globular, comparable to a pregnancy of 8 to 10 weeks of amenorrhea with adnexa on the right side (Figure 1). Uterine reconstruction was done to ensure hemostasis. Since the patient no longer wished to have children, and given her age, we opted to perform a tubal section ligation. The postoperative course was good and the patient had left our structure six days later accompanied by her baby. A renal-vesical ultrasound performed after 6 weeks did not show any abnormality.



Figure 1: Intraoperative observation after repairing the uterine wall showing the two hemi-uterus and their annexes.

DISCUSSION

The incidence of congenital uterine anomalies in a fertile population is 3.2%, of which 90% are uterine septa and 5% are either bicornuate uterus or didelphic uterus.^[1] The bicornuate uterus complicates pregnancy, but does not prevent it. It is often the pregnancy itself that reveals the malformation, as it can cause repeat abortions. While congenital uterine malformations are present in 3-4% of the fertile and/or infertile female population, their frequency rises to 5-10% in women consulting for recurrent miscarriage, and to 25% in women with late miscarriage or preterm delivery.^[2,3] The problem in these patients is not that of conceiving, but of carrying the pregnancy to term. Several factors explain this: uterine malformations are associated with a reduced uterine cavity size, less efficient musculature, inability to distend, myometrial and cervical dysfunction, inadequate vascularization and poorly developed endometrium. These abnormalities are responsible for recurrent miscarriages, preterm deliveries, dystocic presentations, intrauterine growth retardation (IUGR), and the realization of more cesarean sections, with an increased risk of uterine rupture.^[4]

As regards to embryology, bicornuate bicervical uterus are due to a defect in total fusion of the Müllerian ducts between the 10th and 13th week of pregnancy resulting in the formation of two non-communicating uterine cavities.^[5] A vaginal septum is absent in 25% of cases as in the case of our patient. A classification based on the degree of developmental defect of the reproductive system was proposed in 1979 by Buttram and Gibbons^[6], and modified in 1988 by the American Fertility Society.^[7] According to the American Fertility Society classification, bicervical bicornuate uterus correspond to class III. They are accompanied by unilateral urinary malformations such as agenesis in 10 to 50% of cases.^[7,8] In our case, no abnormality was found.

Clinically, bicornuate bicervical uterus remain asymptomatic, especially if they are permeable. The diagnosis is only made incidentally during an examination performed for another purpose. Thus, the diagnosis of a bicornuate bicervical uterus can be made at the time of a first pregnancy check-up or to discover the presence of a vaginal septum or two previously unrecognized cervixes during a vaginal delivery.^[8] Given its rarity, the age and the multiparity of the patient, it was surprising that this anomaly was never detected during prenatal monitoring of previous pregnancies.

As regard to obstetrics, uterine malformations as a whole account for a large number of dystocic presentations, and among these, a breech presentation is found in 23-61% of uterine malformation cases.^[9] As for the mode of delivery, the frequency of caesarean sections is significantly higher in cases of uterine malformation, with rates ranging from 27.5 to 83% according to authors^[9,10,11]. This is because uterine malformations are often associated with dystocic presentations, but they are also associated with a higher frequency of labor abnormalities in about 50% of cases, such as cervical and dynamic dystocia.^[9] In the case of our patient, no obstetrical complications had been observed in the three previous deliveries. It was only during the fourth pregnancy that she was able to present a delivery bleeding requiring a laparotomy for suspected uterine rupture during a uterine revision.

Some types of uterine malformations affect reproductive life and require surgery to restore continuity. But in the case of a permeable bicornuate bicervical uterus, the reunification surgery of two hemi-uterus described by Strassmann in 1952, has not shown any real benefit.^[12] It should be reserved only for patients whose obstetrical prognosis are extremely unfavorable and whose anamnesis reveals several late miscarriages. When the diagnosis of uterine malformation is made in early pregnancy, treatment will only be preventive (rest, lung maturation, ultrasound monitoring of fetal growth and cervical competence).^[13] Cervical cerclage should only be proposed in cases of proven cervical incompetence, which is observed in 25-30% of cases of uterine malformations.^[14]

CONCLUSION

Congenital uterine malformations are relatively common and often asymptomatic, but the reproductive consequences vary depending on the type of malformation. It should be remembered that when uterine malformation is diagnosed, urinary tract imaging should be performed because of the frequent associated abnormalities. A bicornuate uterus does not always lead to complications but it can lead to full term pregnancies. Given the obstetrical history of our patient, we can say that the obstetrical prognosis in women with a permeable bicornuate bicervical uterus, often seems to be very good. This type of malformation is very rare, but it is important to diagnose it by ultrasound in order to manage

the situation preventively, and to allow the extraction of the fetuses in good conditions before any complication.

Conflicts of interest

The authors declare no conflicts of interest.

Authors' contributions

All authors have read and approved the final version of the manuscript.

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