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A uterus didelphys with a spontaneous labor at term of pregnancy: a rare case and a review of the literature

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Abstract:

Uterus didelphys accounts for 13% of uterine anomalies and has been correlated with preterm delivery and fetal malpresentation at delivery. A 37-year-old pregnant woman reported a spontaneous pregnancy in the right horn of a uterus didelphys. The course of the pregnancy was complicated by gestational diabetes, but no miscarriage threat or preterm delivery threat was reported during this pregnancy. She arrived at our division, in labor, at 39.2 gestational weeks'. She delivered by cesarean section due to failure to progress at 5 cm. Her post-operative course was uneventful. Vaginal delivery could be a safe option and the induction of labor or the use of oxytocin could be helpful in such cases, but recommended doses and labor time should be evaluated, so cesarean section is to date the most frequent delivery route in uterus didelphys at term.

Keywords: Müllerian malformation, term pregnancy, uterus didelphys

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Introduction

Müllerian anomalies are congenital defects of the female genital system due to abnormal embryological development of the Müllerian ducts (failure of its development, fusion, canalization or reabsorption). The incidence of uterine malformation is difficult to report considering that the data is usually referred to an infertility cohort, but poor information has been reported in the general population.

Uterus didelphys is characterized by the presence of two separate uterine horns and two different uterine cervix due to an absence of fusion between the two Müllerian ducts. In a study by Hua et al., uterus didelphys accounted for 13% of the uterine anomalies and was correlated with preterm delivery [1]. In a systemic review published in 2011 this malformation presented a significant increase of preterm labor and fetal malpresentation at delivery, however, no higher incidence of first and second trimester miscarriage was found in comparison with the general population [2]. A rate of 45% of pregnancies with uterus didelphys delivered at term [3]. A vaginal delivery could be an option even if a cesarean section is usually the most frequent means of delivery, considering high possibility of the alteration of cervix dilatation or abnormal fetal presentation.

Case report

A 37-year-old pregnant woman reported a spontaneous pregnancy in a malformed uterus. Her gynecological and obstetrical history reported a uterus didelphys and a longitudinal vaginal septum, which divided the right and left uterine cervix, and a first trimester miscarriage. The patient was a normal weight (pre-gestational weight was 55 kg, with 11 kg of weight gain during pregnancy), she is tall, 175 cm; she had been a smoker, who stopped smoking at the beginning of the pregnancy; she had no medical disease and she reported no surgical intervention. The pregnancy was spontaneous and implanted into the right horn. The course of the pregnancy was complicated by gestational diabetes, well controlled by diet. She never had bleeding, a miscarriage threat

or a preterm delivery threat during this pregnancy, so she did not receive any medical therapy. The ultrasound examination during pregnancy reported a normal growth fetus with a normal placenta implantation on the fundus of the right uterine horn. She arrived to our division, in labor, at 39.2 gestational weeks'; on arrival, the right cervix was smoothed and the dilatation was 2 cm; the left cervix presented a 3 cm length and was closed. She required and was given an epidural anesthesia. After 4 h, the dilatation was 5 cm. Despite other 5 h of valid contractions, the dilation remained unchanged. We preferred not to use oxytocin for labor augmentation, so she was delivered by cesarean section. At the surgical intervention, we saw a small a left uterine horn and a right one containing the fetus. We performed a transversal incision on the low right uterine segment, with the birth of a healthy male neonate, weighing 2900 g, whose Apgar scores at 1 and 5 min were, respectively, 9 and 10 and the pH of the umbilical cord was 7.31. Blood loss at cesarean delivery was 200 mL. Her post-operative course was uneventful. At discharge, 3 days after surgery, the right horn was contracted and the left horn was not appreciable on abdominal palpation. The ultrasound evaluation demonstrated a regular right horn with a thin intrauterine cavity and a small and regular left horn.

Discussion

Here, we describe a rare case of spontaneous labor at term in a pregnant woman with a uterus didelphys. Even if a higher risk of preterm delivery in cases of this uterine malformation has been reported, our patient's gestation was regular. During the pregnancy, she underwent frequent obstetrical observations every 2–3 weeks, to evaluate the likely development of complications. She never required progesterone or tocolytic therapy. She desired a vaginal delivery, but unfortunately, she required a cesarean delivery due to the failure of cervical dilation to progress beyond 5 cm.

We describe our case because of the rarity of uterus didelphys and the peculiar gestational age at which our patient came to our division in labor, considering that she never presented a risk of preterm labor in pregnancy, without any preventive therapy [1]. Considering that vaginal delivery has described in the literature as a safe option in such cases, we assisted the labor of our pregnant woman. We did not administer oxytocin as a labor augmentation, even if in the literature, it is occasionally used. We preferred to wait through the spontaneous course of her labor due to the possibility of a uterine rupture in the malformed uterus and because a specific protocol for these cases, with doses and time of augmentation, was not available in our institution.

In the literature, there were only nine cases delivered at term (Table 1). In two cases, the pregnant women had successful vaginal deliveries. Another case described the first twin "had a successful vaginal delivery" while a cesarean section was performed for the second twin. Cesarean section was frequently done, it was performed in 72% of cases, due to abnormal fetal presentation, maternal desire or during labor due to the alteration of the cervix dilatation or a not reassuring cardiotocography. In the cohort reported in Table 1, a cesarean section was performed in six cases (66.7%).

Table 1: Cases of term delivery in patients with uterus didelphys reported in the literature.

Case no.	Author/year	Pregnancy course	Gestational age at delivery	Delivery mode and indication
1	Narayansingh 1998 [4]	Vaginal bleeding during pregnancy	38 weeks	Cesarean section after premature rupture of the membrane
2	Fedele 1987 [5]		Term	
3	Giannopoulos T 2004 [6]	Uncomplicated pregnancy	38 weeks	Cesarean section (patient on her third pregnancy, one termination in the first trimester; second pregnancy delivered by cesarean section at 38 weeks)
4	Chen 2006 [7]	No preterm labor during pregnancy	38 weeks	Cesarean delivery for breech presentation
5	Rana 2008 [8]	Pyocolpos at 10 weeks treated with antibiotics and surgical incision	37 weeks	Vaginal delivery
6	Isango 2013 [9]	Uncomplicated pregnancy	39.5 weeks	Diagnosis of uterus didelphys at cesarean delivery performed for cervix dystocia

7	Park 2013 [10]	Uterus didelphys with a blind hemivagina complicated by pyocolpos due to <i>Pediococcus</i> infection, drained and treated with antibiotics	39 weeks	Cesarean section for patient refuse to vaginal delivery
8	Maki 2014 [11]	Uncomplicated pregnancy	37 weeks	Twin pregnancy. Spontaneous labor after premature rupture of the membrane. First twin delivered vaginally after oxytocin augmentation; second twin delivered by cesarean section due to a not reassuring cardiotocography
9	Rezai 2015 [12]	Uncomplicated prenatal care; no bleeding or threatened preterm labor	38.4 weeks	Induction of labor with misoprostol for membrane rupture; Pitocin augmentation of labor; vaginal delivery

Close monitoring during pregnancy is recommended in patients with uterus didelphys. No routine use of therapy for preventing preterm contractions is required. Future studies, which will define adequate doses and time of augmentation of labor, are required. These data could increase the rate of vaginal delivery in the population with a uterus didelphys.

Conclusion

Vaginal delivery could be a safe option and the induction or augmentation of labor could be helpful in bringing about successful vaginal deliveries, the recommended doses and specific management should be evaluated in future studies, cesarean section is to date the most frequent mode of delivery in uterus didelphys at term.

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References

- [1] Hua M, Odibo AO, Longman RE, Macones GA, Roehl KA, Cahill AG. Congenital uterine anomalies and adverse pregnancy outcomes. *Am J Obstet Gynecol.* 2011;205:558.e1–5.
- [2] Chan YY, Jayaprakasan K, Tan A, Thorneton JG, Coomarasay A, Raine-Fenning NJ. Reproductive outcomes in women with congenital uterine anomalies: a systematic review. *Ultrasound Obstet Gynecol.* 2011;38:371–82.
- [3] Rezai S, Bisram P, Alcantara IL, Upadhyay R, Lara C, Elmadjian M. Case report didelphys uterus: a case report and review of the literature. *Case Rep Obstet Gynecol.* 2015, Article ID 865821, 5 pages.
- [4] Narayansingh CV, Serrao C, Ramsewak SS. Innocuous recurrent vaginal bleeding during pregnancy in a patient with uterus didelphys. *J Obstet Gynaecol.* 1998;18:592.
- [5] Fedele L, Zamberletti D, D'Alberton A, Vercellini P, Candiani GB. Gestational aspects of uterus didelphys. *J Reprod Med.* 1988;33:353–5.
- [6] Giannopoulos T, Croucher C. Successful consecutive pregnancies in separate horns of a uterus didelphys. *J Obstet Gynaecol.* 2004;24:314.
- [7] Chen FP, Ng KK. Term pregnancy at the site of atresia following vaginal canalization in a case of uterus didelphys with hemivaginal atresia and ipsilateral renal agenesis. *Taiwan J Obstet Gynecol.* 2006;45:366–8.
- [8] Rana R, Pasrija S, Puri M. Herlyn-Werner-Wunderlich syndrome with pregnancy: a rare presentation. *Congenit Anom (Kyoto).* 2008;48:142–3. DOI: 10.1111/j.1741-4520.2008.00195.x.
- [9] Isango YI, Mukuku O, Ilunga PM, Kakisingi CN, Nsambi J, Kabamba C, et al. Utérus biconne bicervical perméable: découverte fortuite lors d'une césarienne d'urgence chez une multipare lusophone. *Pan Afr Med J.* 2013;15:75. DOI: 10.11604/pamj.2013.15.75.2745.
- [10] Park TC, Lee HJ. Pregnancy coexisting with uterus didelphys with a blind hemivagina complicated by pyocolpos due to *Pediococcus* infection: a case report and review of the published reports. *J Obstet Gynaecol Res.* 2013;39:1276–9. DOI: 10.1111/jog.12049.
- [11] Maki Y, Furukawa S, Sameshima H, Ikenoue T. Independent uterine contractions in simultaneous twin pregnancy in each horn of the uterus didelphys. *J Obstet Gynaecol Res.* 2014;40:836–9. DOI: 10.1111/jog.12219s.

[12] Rezai S, Bisram P, Lora Alcantara I, Upadhyay R, Lara C, Elmadjian M. Didelphys Uterus: A Case Report and Review of the Literature. *Case Rep Obstet Gynecol*. 2015;2015:865821. DOI: 10.1155/2015/865821.