Third cesarean section in an undiagnosed uterus didelphys

Salmeh Dadgar¹ · Asieh Maleki^{1,2} · Massoumeh Mirteimouri¹ · Ghazal Ghasemi¹

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About the Author

Salmeh Dadgar, MD is an Assistant Professor of Obstetrics and Gynecology, Department of Obstetrics and Gynecology, Emam Reza Hospital, Faculty of Medicine Mashhad University of Medical Sciences (mums), Mashhad, Iran. She is Co-editor of Journal of Obstetrics and Gynecology of Iran. She is 39 years old and married.

Salmeh Dadgar, MD, is an Assistant Professor of Obstetrics and Gynecology in Mashhad University of Medical Sciences; Asieh Maleki, MD, is an Assistant Professor of Obstetrics and Gynecology in Mashhad University of Medical Sciences; Massoumeh Mirteimouri, MD, is an Associate Professor of Obstetrics and Gynecology in Mashhad University of Medical Sciences; Ghazal Ghasemi, MD, is an Resident of Obstetrics and Gynecology in Mashhad University of Medical Sciences.

✉ Asieh Maleki
Malekiaa@mums.ac.ir

¹ Department of Obstetrics and Gynecology, Mashhad University of Medical Sciences, Mashhad, Iran

² Department of Obstetrics and Gynecology, Omollbanin Hospital, Behjat Street, Mashhad, Iran

Introduction

Uterus didelphys, also known as duplicated uterus, is an embryological abnormality resulting from failure of fusion of Mullerian ducts, causing full uterine development to erroneously occur bilaterally [1]. Women with didelphic uteri may be asymptomatic, but some present with dysmenorrhea, dyspareunia, or may be associated with infertility, spontaneous abortion, preterm labor, fetal malpresentation, intrauterine growth restriction, PROM, renal agenesis, decreased live births and cesarean delivery [1, 2]. This condition is less common than other uterine malformations and has been estimated to occur in 1/3000 women [3]. But the rate of twin pregnancy in this condition with two babies growing in two separate uteri is very rare [4].

Case Report

A 41-year-old woman, G4P2A1, with spontaneous twin pregnancy, was admitted to our hospital at 35 weeks of gestation due to premature rupture of membrane and uterine contractions. Although her previous cesarean section surgery's notes were not available, she did not mention any problem or unusual history in her two previous sections 12 and 15 years ago. Vaginal examination revealed a partial longitudinal vaginal septum and 3-cm dilation of cervix. Three sonograms were done at 12, 18 and 30 weeks of gestation, and in all of them bicornuate uterus was reported.

Cesarean section was performed, and after opening the abdominal wall, there was a severe rotation and displacement of uterus to lateral side; just after its correction, diagnosis of uterus didelphys was confirmed (Figs. 1, 2). We found double uterus, double cervix and septate vagina and two complete separate pregnancies in each uterus. Each uterus has its separate ovary and fallopian tube. Kerr incision was done on right uterus, and a healthy male baby of 2.3 kg with breech presentation was delivered. Then with another incision on left uterus, a female baby of 2.2 kg with cephalic presentation was delivered. Both had good Apgar score. Two separate placentas were delivered, respectively. At the end of surgery, speculum examination was done and two separate cervices were revealed.

Discussion

Uterus didelphys is a rare embryological abnormality resulting from failure of fusion of Mullerian ducts [2]. Because of its low occurrence (1/3000 women) and because it may be asymptomatic, sometimes it may go

Fig. 1 Uterus didelphys, right uterus with Kerr incision and undelivered placenta (with arrow), left uterus (yellow arrow)

Fig. 2 Double uterus; left uterus is repaired (yellow arrow)

undiagnosed [2, 3]. An increased rate of obstetric complications have been found in singleton pregnancies complicated by uterus didelphys (spontaneous miscarriage 30%; breech presentation 43%; preterm rupture of the membranes 53%; and preterm labor 95%). There is a paucity of information on twin pregnancy in uterus didelphys, because of its extremely low incidence of about one per million pregnancies [5]. Most of the data on clinical manifestations, outcomes and preferred route of termination of pregnancy in these patients are based on small observational or case studies. Most uterine malformations can be detected by sonography. In didelphys uterus, the endometrial cavities are widely separated and there is a profound indentation on the fundal contour [1]. But sometimes, there is misdiagnosis as bicornuate uterus. Rezai et al. [6] reported a case of term pregnancy in uterus didelphys that was similar to our case and was reported as a bicornuate uterus in pelvic sonogram. In most reported cases, the external uterine contour was not visualized and the diagnosis of a true uterus didelphys was not confirmed; thus, many cases of 'twin pregnancy in a double uterus' are not truly dicavitary, but represent twin pregnancies in a single horn in a bicornuate uterus [5]. We represented here a rare case of twin pregnancy in a didelphys uterus that was diagnosed in her third pregnancy. In all her pelvic sonograms, didelphys uterus was reported. Although her pregnancy was not affected with any prenatal complications, undiagnosed uterus didelphys in some cases is reported, that is associated with adverse prenatal outcomes. Okafor II et al. [2] reported an undiagnosed uterus didelphys in term pregnancy that was complicated with prolonged PROM, intrauterine fetal death, failed induction of labor and emergency cesarean delivery.

In one retrospective cohort study in 2014, in the USA, that includes 158 patients with singleton pregnancy and

uterine anomaly, adverse pregnancy outcomes and cesarean delivery were increased in patients with major fusion defects like uterus didelphys [7]. Due to such complications that can affect pregnancy, most women with related malformations need to be monitored closely throughout their pregnancy. In our case, due to spontaneous rupture of membranes at 35 weeks of gestation, appropriate growth of fetus in each horn of a didelphys uterus, and attention to uterine rotation and its displacement during the surgery, neither the patient nor her twins encountered any additional complications.

Conclusion

There must be a high index of suspicion of uterine didelphys, especially in sonographic reports suggestive of bicornuate uterus. Special attention should be given to initial pelvic examination to make early diagnosis and to prevent some associated adverse prenatal outcomes.

Compliance with Ethical Standards

Conflict of Interest The authors declare that they have no conflict of interest.

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