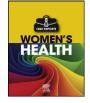
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Successful management of the recurrent uterine rupture after the uterine septum resection^{*}



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ABSTRACT

Uterine septum is associated with an increased rate of recurrent miscarriages which can be reduced significantly by performing endoscopic resection. Perinatal outcome improvement due to resection is favorable but still remains controversial. Uterine rupture is a late complication of hysteroscopic surgery that may be a life-threatening condition in terms of perinatal and maternal morbidity-mortality. This article presents the first case of the related literature dealing with the successful management of the third recurrent uterine rupture in the 31st week of pregnancy subsequent to the hysteroscopic resection of uterine septum. The patient observed in this article had experienced uterine rupture twice; firstly, in the 27th week of pregnancy resulted with post-partum exitus and secondly, in the 29th week of pregnancy that ended with stillbirth while her last pregnancy eventuated in a live healthy birth. Surgeons who accomplish these hysteroscopic procedures must weigh out pros and cons; and must also acquaint their patients with the probable risk of recurrent and multiple uterine ruptures during the subsequent pregnancy.

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1. Introduction

The prevalence of uterine anomalies is estimated to be 1/200 and uterine anomalies are relatively infrequent among general population; however, they have been observed among 15% of women with recurrent pregnancy losses defined as three sequential pregnancy losses before the 20th gestational week [1]. Women with recurrent pregnancy losses have a 3.2–6.9% likelihood of having an uterine septum anomaly [2]. Uterine septum is an important cause of decrease in the live birth rate that can significantly be reversed by performing endoscopic resection, in other words "metroplasty" [1]. Hysteroscopy which is the gold standard intervention to evaluate the uterine cavity is used not only to evaluate the type of uterine anomalies like uterine septum but also to treat this malformation concurrently [3]. Since endoscopic procedure is risky, it should only be performed if the patient has recurrent miscarriages as a result of uterine structural defect caused by uterine septum [1].

Uterine rupture during pregnancy following the operative hysteroscopy seems to be a rare and late complication of hysteroscopic surgery that may be a life-threatening condition in terms of perinatal and maternal morbidity-mortality. Although uterine rupture seems to be rare in the literature, it is being reported more and more in the pregnancies subsequent to endoscopic septum resection. It should be suspected if there appears sudden fetal heart rate deceleration during labor or unexpected intrapartum hemorrhage particularly in the patients with a history of uterine septum resection or uterine ruptures [4]. Here, we present a case of the third recurrent uterine rupture in the 31st week of pregnancy; the pregnant had previously experienced recurrent pregnancy losses and later, had undergone the hysteroscopic resection of uterine septum.

2. Case Presentation

A 35 year-old woman – gravida 5, para 2, abortion 2 – applied to our high-risk pregnancy clinic in the 24th week of pregnancy regarding her last menstrual period. When her obstetrical history was searched, it was seen that she had two first-trimester miscarriages in the 6th and 10th weeks with no obvious etiological reason, except for an uterine septum found during her complete examination that includes a sonogram demonstrating normal fundal uterine configuration without any indentation. Following the two early pregnancy losses, uterine septum on the fundal part of uterus was suspected after the hysterosalpingography, and diagnostic office hysteroscopy confirmed the evident HSG findings which were compatible with uterus septus (Fig. 1). Upon an explanation of the risks and possibilities of uterine rupture in the future, patient had agreed to undergo hysteroscopic metroplasty. After the diagnosis, she had a diagnostic laparoscopy (L/S) and then, operative

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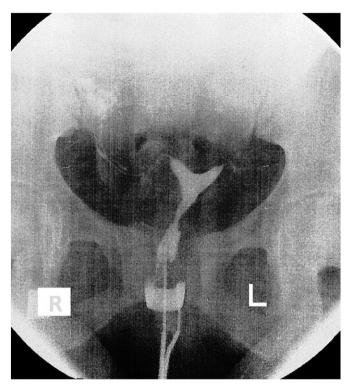


Fig. 1. Hysterosalpingography (HSG) image of the patient still showing a pattern of arcuate uterus after resection of uterine septum by cutting electrosurgery.

hysteroscopy (O/H) was performed. During the L/S, minimally arcuate shaped uterus with bilateral normal tubes and ovaries were noted; thus, bicornuate configuration had been ruled out. In the O/H, 3 cm thickness of longitudinal uterine septum occupying more than half of the uterine cavity in transverse section and lying 1 cm away from the fundus had been diagnosed and resected, using cutting electrosurgery with bipolar needle electrodes. The resection level elongated to the level of the tubal ostiums by means of the modulated waveform and the 70 watt-electrical current. The resection was performed all along the septum until myometrial muscle fibers became visible at the uterine fundus. At this point, no further incision into the myometrium was performed in order to prevent uterine perforation. To monitor the operation and to prevent the occurrence of myometrial invasion, sonography was employed during the septal resection. Uterine bleeding was minimal and postoperative recovery was without any problems. No uterine perforation had occurred and no rigid scissors or other non-electrosurgical modalities like laser were used during the resection of uterine septum.

After the endoscopic resection operation, she had two pregnancies without any live infants as she had experienced two uterine ruptures in the 27th and the 29th week of pregnancy resulted with postpartum exitus and stillbirth respectively. There was a seven year gap between the first rupture in the 27th week and the third rupture in the 31st week, the patient's latter pregnancy resulted with a live birth.

In the first rupture she consulted to the hospital, complaining of severe groin pain and vaginal bleeding that ended up with an emergent laparotomy. It was required because of severe fetal bradycardia and uterine rupture causing maternal hemodynamic hypovolemic shock. Her baby was born 950 g with an Apgar score of 8-10-10 but unfortunately died on the 2nd day of postpartum. Five days after the laparotomy, she recovered without any problems and was discharged. The second uterine rupture occurred in the 29th week of her 4th pregnancy -4 years before the successful live birth. She applied to the emergency service with severe abdominal and groin pain that once again ended up

with an emergent laparotomy. The laparotomy was performed because of uterine rupture and a stillbirth of a 1510 g baby. There was a 10 cm rupture of uterus extending from one cornu to the other cornual part, and all abdomen was filled with fibrinated blood.

In the 24th week of the fifth pregnancy, she was hospitalized in case of a possible uterine rupture. She had no contraction on palpation and there was no evidence of cervical dilatation during the bimanual pelvic examination. All routine antenatal tests revealed no abnormality and all vital signs were normal. Her antenatal care was completely normal until hospitalization. The combined first-trimester screening test and the detailed ultrasonography scan for fetal anomaly screening were normal. Her vital signs were monitored every 4 h during her stay in the hospital. In the 27th week, antenatal corticosteroid (betamethasone) was intramuscularly injected for the maturation of the fetal lung in case of a preterm delivery stemming from uterine rupture. Biophysic profile was performed three times a week routinely and additionally, whenever the patient suspects of the fetal health. Non-stress test (NST) the day before the delivery was reassuring and showed no signs of contractions. All ultrasonographic fetal growth parameters were concordant with the pregnancy week. The placenta was located posteriorly in uterus with no evidence of placenta previa and no signs of thinning, or no disruption of uterine scar was observed during the examination by ultrasonography. There were also no signs of infection regarding her routine tests and clinical situation leading to preterm delivery.

In the 31st week of her fifth pregnancy subsequent to the resection, she had complained of sudden and severe abdominal pain. Ultrasound revealed severe fetal bradycardia. After it was confirmed by ultrasonography, within 7 min, an emergent delivery was performed via laparotomy because of an indication of suspected uterine rupture and of severe fetal distress. The delivery was accomplished from the rupture, since there was no need for low uterine segment incision as the fetus was unbound in the abdominal cavity beside all the placental products. An entubated 1300 g male baby with a 0 Apgar score was born and the venous umbilical fetal blood pH was 6.95. Intraoperatively there was a nearly 12 cm rupture of myometrial section of the uterus extending from one cornu to the other cornual part and the whole abdomen was filled with approximately 500 cm³ of fibrinated blood. The uterus had ruptured from fundal middle part. As shown in the intraoperative overhead image, it looks like a blooming of a "daisy flower" (Fig. 2). As can be observed, the complete uterine rupture occurred at the site of the previous uterine septum which was the middle fundal part of the uterus. The deformed and ruptured part was carefully repaired as three layers through the interrupted 1.0 vicryl sutures; as a result, the normal



Fig. 2. Intraoperative overhead image of the middle fundal part of the uterus photographed instantly after the complete uterine rupture at the site of the previous uterine septum.

anatomy of the uterus nearly disappeared. To remove necrotic and traumatized tissue, repair of the uterine defects was accomplished by trimming the edges. The trimmings and the placenta were sent to histopathology and the possibility of placental implantation disorder was ruled out. The postoperative recovery period was fine and she was discharged 6 days after the operation. Before discharge, she was informed carefully about the possible risks in case of a subsequent pregnancy and labor, and was recommended to use effective contraception. Because of the cumulative effect of exponentially increased risk of recurrence of uterine rupture, tubal ligation was suggested but she declined. Her baby is now 4-years-old and healthy, normally grown and completely concordant with his peers with no neurodevelopmental sequelae.

3. Discussion

Data obtained from the literature confirms that patients with septate uterus are associated with higher rates of fetal loss, spontaneous abortion, preterm labor, intrauterine growth restriction, breech presentation, cesarean delivery and complications that increase perinatal mortality and morbidity [1]. Our patient had spontaneous abortion twice, also one fetal loss, one neonatal loss, three preterm labors and three uterine ruptures. Hysteroscopic metroplasty significantly improves pregnancy outcomes among women with uterine anomalies [5]; as in the case of our patient, no abortions had occurred after the operation, and delivery accomplished in spite of the multiple uterine ruptures. It is also important to note that there is no randomized trial regarding the outcome of metroplasty based on pregnancy losses. When the related literature is searched, it is seen that the 65-75% of women with 2 prior losses have successful subsequent pregnancies without any intervention. The reason for better perinatal outcomes and higher success may be related to the gestational age at which the rupture occurs. The aforesaid patient was hospitalized when the rupture occurred; therefore, the immediate extraction of the fetus was possible. The hysteroscopy had been performed under the laparoscopic guidance; however, the laparoscopy did not prevent the undesirable deep myometrial damage causing perforation, in this particular case.

The resectoscope and the microscissors are equally valid instruments to correct a septate uterus, with an optimal feasibility rate. In our case, cutting electrosurgery method like resectoscope was performed and it resulted with the successful correction of intrauterine cavity. Among the patients selected for metroplasty because of their recurrent pregnancy losses, the rate of 7% successful pregnancy has risen to 75% as a result of metroplasty [6].

The late complications of operative hysteroscopy caused by myometrial damage during the surgery can trigger catastrophic consequences during a subsequent pregnancy. These long-term problems should lead the operating physician to select metroplasty patients carefully, in order to minimize the likelihood of unnecessary myometrial damage. Sentilhes et al. included 12 patients with a history of hysteroscopic metroplasty from the 14 cases in which uterine rupture occurred between the 19th and 41st weeks, the 66.5% of cases were reported without any sign of labor while 4 fetal deaths and 1 maternal death followed the uterine rupture [7]. There is no agreement on the secure interval period between hysteroscopic septum resection and a subsequent pregnancy. In our case gestational weeks were the 27th, the 29th and the 31st respectively and the interval times between hysteroscopy and the subsequent pregnancies complicated with uterine ruptures were 1, 4 and 8 years respectively. This situation may be explained through the interval time; the longer the time is, the longer the pregnancy continues.

Congenital weakness of uterine wall during the uterine augmentation resulting in rupture is repaired by sutures, and it recovers with fibrosis; therefore a more resistant myometrial tissue against rupture emerges; but more data is needed to confirm this hypothesis.

In our case, serial ultrasound scans had been performed during the pregnancy to detect impeding rupture. Sentilhes et al. also reported that hysterosalpingography or ultrasonography scan, long interval between the operative hysteroscopy and the subsequent pregnancy, and the elective cesarean sections are not effective to prevent and to detect the impeding ruptures in the subsequent pregnancies; only favorable use of scissors for hysteroscopic metroplasty has been found as an accurate method to prevent it [7]. In our case, electrosurgical energy had been used instead of rigid scissors and this confirms the results of this study. If scissors had been performed in our patient, no uterine perforation might have been occurred then. Uterine rupture occurs mostly between the 28th and the 32nd weeks of gestation; however, in literature, a patient in the 41st week who underwent hysteroscopic resection of an uterine septum for recurrent miscarriage and experienced uterine rupture has been reported. Multiple hysteroscopic procedures may significantly increase the risk of uterine rupture as in the case of our patient who has a history of hysteroscopic uterine septum resection. In literature there are many reliable and secure methods for hysteroscopic resection. Intraoperative ultrasonography guidance for intrauterine endoscopic procedures seems to offer a noninvasive means of assessing the precision and the adequacy of resectoscopic procedures and seems to provide the exact location of the instruments within the uterine cavity and uterine wall. In our opinion, intraoperative imaging may be employed for other intrauterine endoscopic procedures and may preclude the need for simultaneous laparoscopy which constitutes an additional surgical risk for the patient and prolongs the procedure.

Suggesting routine cesarean section for patients who have undergone metroplasties is somewhat excessive if there are no details of the operations and/or ultrasound imagings of the uterine fundal wall. Attention must be paid to identify the specific uterine anomaly. The subsequent rupture cases in the literature present so many distinct features with respect to the size of septum, the method of dissection, the complications of the operation (including perioperative uterine perforation), the age, the parity, the time interval between the surgery and the pregnancy, the gestational week during the rupture, the singleton or the multiple pregnancy and other pregnancy complications. Certainly, attentive surveillance in the peripartum period is necessary and it is very important to know the details of the operation, particularly how deep into the myometrial wall the resection was performed [4].

In conclusion, we want to point out that physicians dealing with the patients with previous hysteroscopic metroplasty should be aware of the potential risks of uterine rupture during the following pregnancy until a sensitive and specific predictive method is discovered. Although hysteroscopic resection of an intrauterine septum reduces the frequency of pregnancy loss among women having recurrent abortions; the possibility of uterine rupture after operative hysteroscopy might have negative maternal and perinatal outcomes. Currently, there is no agreement on the standard follow-up protocol that decreases perinatal adverse outcome among these patients. For this purpose, patients must be well-informed about the symptoms of uterine rupture during their pregnancy and must be hospitalized, and clinicians must regard the possibility. Surgeons who accomplish these hysteroscopic procedures must weigh out the pros and cons; and must warn their patients against the probable risk of recurrent uterine ruptures during the pregnancy following the contingent uterine rupture.

Declaration of Statement

The authors report no declarations of interest.

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