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CASE REPORT PEER REVIEWED | OPEN ACCESS

A case of uterine rupture in second term delivery after septum resection

Alexandra Huttler, Maureen Kelly

ABSTRACT

Introduction: The decision to resect a uterine septum remains individualized. Hysteroscopic approach is the gold standard for uterine septum resection. Uterine rupture in subsequent pregnancy is a known but rare complication without identified methods of prevention.

Case Report: A 32-year-old woman underwent uncomplicated hysteroscopic uterine septum resection followed by uncomplicated term vaginal delivery, with uterine rupture and intrauterine fetal demise at 38 weeks gestational age in the subsequent pregnancy.

Conclusion: Term uterine rupture is possible when the only history of uterine surgery is hysteroscopic septum resection, even with an intervening delivery. Providers should have a low threshold to initiate diagnostic evaluation with fetal or maternal distress in pregnancy after any uterine surgery. Modifiable risk factors for uterine rupture after septum resection may include residual septa, short interval between surgery and pregnancy, and labor induction methodology. Providers should consider these factors when counseling regarding the risks and benefits of resection.

Keywords: Operative hysteroscopy, Uterine rupture, Uterine septum resection

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INTRODUCTION

A uterine septum represents the most common uterine anomaly, with an incidence of 0.2–3% among reproductive-aged women [1, 2]. Uterine septa confer an increased risk for miscarriage, preterm birth, malpresentation, fetal growth restriction, placental abruption, and perinatal mortality [1, 3].

The role of uterine septum resection remains controversial, as the existing evidence does not uniformly support a fertility or obstetric benefit [4]. As per the American Society of Reproductive Medicine, resection is reasonable in selected clinical circumstances [1]. Hysteroscopic resection is the first-line approach and can be accomplished with use of mechanical scissors, electrosurgery, vaporizing or bipolar electrodes, lasers, and mechanical morcellators [2]. Possible complications of a hysteroscopic septum resection include bleeding, uterine perforation, intrauterine adhesions, and uterine rupture in subsequent pregnancies [2].

A 2012 meta-analysis described 18 confirmed reports of uterine rupture during a pregnancy subsequent to hysteroscopic septum resection [2]. Since then, additional case reports have been published, including two cases of recurrent uterine rupture in three subsequent pregnancies [5, 6] and uterine rupture as early as ten weeks gestational age [7]. There has only been one case reported of uterine rupture after uncomplicated hysteroscopic resection with an intervening uncomplicated term delivery [8]. This

patient underwent septum resection via potassium titanyl phosphate laser, had an uncomplicated term vaginal delivery, and then presented with preterm uterine rupture at 33 weeks gestational age in her next pregnancy [8]. There exist no reports of pregnancies complicated by uterine rupture at term that were preceded by an uncomplicated delivery in the period after hysteroscopic resection.

CASE REPORT

A patient was initially evaluated as a gravida zero at age 32 for unexplained infertility. She was found to have two separate endometrial cavities on hysterosalpingogram. Magnetic resonance imaging demonstrated a 2.8 × 3.7 cm muscular uterine septum extending to the level of the internal os with a 4 mm fundal indentation, a pseudoseptum of the cervix, and a single vagina, consistent with a septate uterus. Bilateral endometriomas were also noted. She underwent an uncomplicated hysteroscopic septum resection with use of cold scissors within the resectoscope under ultrasound guidance, using glycine as the distension medium. A 10 mL Foley balloon was maintained in the cavity postoperatively. She was prescribed a 28-day course of postoperative oral estradiol. Ultrasound performed five months later demonstrated residual indentation of the fundal aspect of the endometrial cavity measuring approximately 1.5 cm. She conceived her first pregnancy after letrozole ovulation induction with timed intercourse five months postoperatively. She underwent an induction of labor at 39 weeks 1 day gestational age for gestational diabetes. Her induction was completed with 25 μg misoprostol vaginally, placement of a cervical ripening balloon, and 24 hours of oxytocin with maximum dose of 28 mU/min. She delivered a 3.12 kg infant via spontaneous vaginal delivery complicated by uterine atony that was controlled with oxytocin and methylergonovine. She had an uncomplicated postpartum course and was discharged on postpartum day two.

One month after delivery, a screening pap smear suggested a placental site nodule. Ultrasound demonstrated mild heterogeneity of the endometrial complex in the region of septoplasty without abnormal vascularity.

Thirteen months postpartum, she spontaneously conceived a dichorionic-diamniotic twin pregnancy with demise of one twin around six weeks gestational age. The pregnancy was otherwise uncomplicated. She presented to the hospital with complaint of contractions at 38 weeks 3 days gestational age at 22:53. Her cervical exam did not change over 2 hours, she had a reactive fetal non-stress test, and she was subsequently discharged. She returned to the triage unit at 06:39 the following day at 38 weeks 4 days gestational age with complaint of worsening contractions. At that time, she was found to have an intrauterine fetal demise. Ultrasound evaluation did not comment on free fluid or myometrial integrity. Her vital signs were normal and hemoglobin was 12.7 g/dL.

She underwent an induction of labor initiated with 50 μg of vaginal misoprostol around 09:30. Shortly thereafter, an epidural analgesia catheter was placed. Three hours later, she experienced a hypotensive episode with 15 seconds of syncope of undifferentiated etiology. She was treated with intravenous fluids and vasopressors. Membranes were artificially ruptured without fluid return and she was started on oxytocin for labor induction at 13:37 at 4 mU/min, which was increased to a maximum of 8 mU/min by 14:00 and stopped by 15:16. She remained persistently hypotensive and became tachycardic. At 17:30, transthoracic echocardiogram was performed with findings of left ventricular ejection fraction of 75% and hyperdynamic left ventricular systolic function. Hemoglobin was found to have dropped from 10.2 to 8.1 g/dL. Decision was made to perform exploratory laparotomy. Upon entrance to the peritoneal cavity, there was expulsion of copious dark red blood and clot. Fetal parts were noted in the abdomen, with the head entrapped inside a well-contracted, firm uterus. Upon exteriorization of the uterus, a full thickness rupture of the uterine fundus was appreciated, approximately 10 cm in length from the posterior to anterior uterine body. Full thickness repair was performed after delivery of the fetus. The patient was discharged on postoperative day three. She had an uncomplicated postoperative recovery. Standard evaluation of intrauterine fetal demise including laboratory work-up for placental abruption, maternal infection, antiphospholipid antibody syndrome, endocrine abnormalities, and illicit drug use in addition to gross placental pathologic examination did not identify an etiology. Six months postpartum, ultrasound demonstrated a fluid collection in the uterus. Magnetic resonance imaging showed a 17 mm thickened endometrial complex and postsurgical changes related to septum resection and rupture repair. Follow-up ultrasound was recommended but not completed.

DISCUSSION

Hysteroscopic septum resection is a relatively safe procedure, but there are still inherent risks [2]. Uterine rupture is a rare complication, but the consequences can be catastrophic and involve maternal and/or fetal death, as demonstrated in this case of intrauterine fetal demise. Risk of uterine rupture has been correlated with excessive septal excision, penetration of the myometrium, uterine wall perforation, and excessive use of cautery or laser energy during removal [1]. None of these risk factors were present in this case.

As aforementioned, there are multiple ways to perform hysteroscopic septum resection, and American Society of Reproductive Medicine guidelines state that there exists insufficient evidence to recommend a specific method for hysteroscopic septum incision [1]. Electrosurgery exposes the myometrium to thermal vascular damage and

weakening of the tissue, and use of monopolar electric current has been substantiated as a risk factor for uterine rupture after laparoscopic myomectomy or operative hysteroscopy [9]. In the only other report of a case of uterine rupture after hysteroscopic septum resection preceded by a term delivery without rupture, the authors theorized that the myometrium was damaged by the laser used during resection and then further weakened by the stress of the initial pregnancy, ultimately leading to preterm rupture during the second pregnancy [8]. In the case presented here, the myometrium had no exposure to monopolar or laser energy, but may have experienced initial myometrial insult via mechanical disruption with scissors that was similarly exacerbated by the stress of the primary pregnancy. However, uterine integrity was sufficiently maintained for 38 weeks of the second pregnancy.

There is insufficient evidence to designate a safe interval between hysteroscopic septum resection and conception. Studies assessing endometrial repair after septum resection suggest that complete healing may occur as quickly as eight weeks [10]. However, it is known that inter-delivery intervals of less than 18 months are associated with increased risk of uterine rupture in women undergoing trials of labor after previous cesarean section [11]. While hysterotomy is a more invasive procedure than septum resection, it seems plausible that a longer period of healing could be associated with a lower risk of rupture. However, longer interval between generalized operative hysteroscopy and subsequent pregnancy has not been found to be effective in preventing rupture [9]. In this case, the interval between hysteroscopy and conception of the first pregnancy was five months, with thirteen months between delivery and conception of the second pregnancy. Perhaps the short intervals between each of these events did not permit adequate myometrial recovery in this patient.

There are no restrictions regarding method of labor induction after operative hysteroscopy. The American College of Obstetrics and Gynecology recommends avoiding misoprostol for cervical ripening or labor induction in patients at term who have had a prior cesarean delivery or major uterine surgery due to the increased risk for uterine rupture [12]. While operative hysteroscopy is not considered major uterine surgery, no data currently exist validating the safety of misoprostol after hysteroscopic septum resection. It is plausible that its use in this case increased the risk of rupture.

While the impact of septum size on fertility and pregnancy outcomes has been evaluated, there are no data discussing the role of septum size in the risk for uterine rupture after septum removal [13]. In the case presented, the septum was both long and broad. It is biologically plausible that septum size may contribute to risk of uterine rupture after septum removal, as longer and more extensive surgery is likely to be associated with removal of a larger structure. Additionally, as in this case, there have been other reports of uterine rupture after hysteroscopic septum resection in which a small residual septum remained. It has been hypothesized that residual septa may result in a weaker uterine wall that, when stretched during the uterine enlargement of pregnancy, becomes a risk factor for rupture [14, 15]. Histological analyses have suggested that septa have similar amount of but more irregular muscle fibers and looser connective tissue compared to normal myometrium [16]. It is also possible that an anomalous uterus may be congenitally weakened, so that even after removal of the septum, the myometrium remains abnormal. Although not substantiated with histologic evidence, the residual septum and large septum size in this case may have been risk factors for rupture.

It is not known how uterine rupture can be predicted or prevented. Neither hysterosalpingogram or ultrasound follow-up of myometrial thickness nor extended intervals between surgery and pregnancy have been effective in preventing or detecting impending ruptures [9]. Further, data remain insufficient to support prophylactic elective cesarean section before the onset of labor to prevent rupture [15]. The clinical presentation of uterine rupture during pregnancy is extremely variable and can include abdominal pain, fetal bradycardia, and maternal shock with concomitant fetal distress or demise due to hemorrhage. It can be challenging to distinguish abdominal pain suggestive of uterine rupture from the pain associated with normal uterine contractions of labor. Specifically in the case presented, the presence of an intrauterine fetal demise may have impacted the medical team's ability to assess for signs of uterine rupture, as fetal distress could not be evaluated in the absence of fetal heart rate tracing. Diagnosis prior to hemodynamic instability can reduce maternal blood loss and facilitate timely performance of necessary surgical repair. While it is unknown at what point in this patient's clinical course the uterine rupture occurred, additional maternal imaging or withholding of epidural anesthesia to allow more accurate assessment of pain may have permitted earlier recognition of this complication. Maintaining a higher degree of suspicion for uterine rupture could have resulted in a lower threshold to initiate diagnostic evaluation in the setting of maternal distress.

CONCLUSION

This case argues that the presence of fetal or maternal distress and/or severe abdominal pain during pregnancy after a previous uterine surgery, even after uncomplicated hysteroscopic septum resection and intervening normal pregnancy, should prompt suspicion of uterine rupture and subsequent diagnostic evaluation to minimize adverse outcomes. The impact of residual septa, short interval between surgery and pregnancy, and prostaglandin use on risk of uterine rupture could be considered in these clinical contexts.

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Author Contributions

Alexandra Huttler – Conception of the work, Design of the work, Acquisition of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Maureen Kelly – Conception of the work, Design of the work, Acquisition of data, Interpretation of data, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

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Authors declare no conflict of interest.

Data Availability

All relevant data are within the paper and its Supporting Information files.