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## Uterine rupture before the onset of labor following extensive resection of deeply infiltrating endometriosis with myometrial invasion



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The risk of uterine rupture (UR) before the onset of labor has been investigated in women who have undergone cesarean deliveries with previous classical incision and in those with thin lower-uterine-segment defects [1,2]. Previous myomectomies are also a known risk factor for UR [1]. Additionally, evidence published in the past three decades suggests that extensive resection of endometriosis developing within the uterine wall could increase the chances of UR [3–6].

Deeply infiltrating endometriosis (DIE) is characterized by the presence of endometrial glands and stroma outside the uterus. Deep infiltration into the myometrium is a form of DIE that begins at the uterine serosa and advances toward the endometrium. The richly vascularized uterine smooth muscle offers a favorable pathway for DIE to develop [7]. Surgical radical resection is the best option to control DIE. Unfortunately, complete resection of DIE within the uterus can cause substantial thinning of the uterine wall, leaving these areas susceptible to UR during pregnancy. Additionally, the modified blood supply in the scar tissue is associated with local ischemia.

The aim of the present report is to describe two cases of UR following radical excision of DIE. In March 2010, a nulliparous woman aged 31 years (G0P0) who had been experiencing dysmenorrhea, dyspareunia, and infertility for 3 years presented to Huntington Centro de Medicina Reprodutiva, São Paulo, Brazil. Frozen pelvis was diagnosed by a laparoscopy. She subsequently underwent four unsuccessful rounds of in vitro fertilization (IVF).

Two years later, she was evaluated for intense pelvic pain by transvaginal ultrasonography with bowel preparation, which demonstrated

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severe DIE. After counseling, she underwent extensive laparoscopic treatment of grade IV endometriosis (American Society for Reconstructive Microsurgery staging criteria). Extensive uterine endometriosis was identified at the posterior uterine fundal wall, in addition to DIE lesions present in the retrocervical space, posterior vaginal fornix, and uterosacral ligaments. Far-reaching resection of uterine DIE resulted in a considerable thinning of the uterine wall. Bilateral salpingectomies due to hydrosalpinges and removal of small subserosal leiomyomas were also performed. Endocavitary single layer sutures were placed using polydioxanone suture (PDS 2–0, Ethicon, Johnson & Johnson, CA, USA). Rectosigmoid resection, appendectomy, and partial cecal resection were performed by a laparoscopy-assisted transvaginal segmental approach. The estimated blood loss was 250 mL.

Five months later, the patient underwent IVF with the transfer of three embryos. Pregnancy was achieved and was uneventful until the 32nd week, when the patient experienced acute abdominal pain. She was promptly admitted to hospital and stable vital signs, weak uterine contractions, and abdominal wall tenderness were recorded. Ultrasonography showed one fetus with a normal heart rate. One hour later, repeat ultrasonography demonstrated UR with prolapse of the amniotic membranes across the uterine wall near the posterior left uterine horn (Fig. 1). An emergency cesarean was performed under spinal anesthesia. A male newborn weighing 2175 g was delivered with Apgar scores of 1 and 10 at 1 and 5 minutes, respectively. UR was noted at the left cornual wall (Fig. 2), which was effectively closed with coated 0-Vicryl sutures (Ethicon, Johnson & Johnson, CA, USA). Mother and newborn were admitted to the intensive care unit (ICU). The mother underwent an uneventful recovery and the newborn was discharged from the ICU after 2 weeks, weighing 2105 g.

In June 2012, another patient (G1Ab1P0), who was aged 35 years, presented to Huntington Centro de Medicina Reprodutiva after 4 years of infertility for laparoscopic treatment of pelvic endometriosis. Bilateral tubovarian adhesions and superficial endometriosis were treated by adhesiolysis and fulguration. Thereafter, she underwent three unsuccessful IVF attempts. Transvaginal ultrasonography with bowel preparation was performed due to dyspareunia and revealed advanced DIE. Additionally, laparoscopy revealed a frozen pelvis, widespread DIE throughout the pelvis, and an endometrioma concealed within the left iliac vein, obturator nerve, and spreading to the ipsilateral ureter. Hemostasis of the DIE was accomplished using bipolar cauterization followed by intracorporeal suturing. Extensive resection of DIE lesions at the left fallopian tube was carried out with monopolar energy. The uterine wall was reapproximated via intracorporeal suturing with 2–0 absorbable polyglactin (Ethicon, Somerville, NJ, USA).

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**Fig. 1.** Transabdominal ultrasonography demonstrating uterine rupture associated with prolapse of the amniotic membranes across the uterine wall near the posterior left uterine horn (arrow).

Nine months later, three fresh embryos were transplanted as part of IVF. A single gestation was identified and an embryonic heartbeat was detected by ultrasonography at 6 weeks of pregnancy. There were no further complications until week 33, when the patient was hospitalized to inhibit preterm labor. Twelve hours after intravenous administration of the tocolytic agent, the patient became pale, her blood pressure dropped to 80/50 mm Hg, and abdominal ultrasonography demonstrated massive hemoperitoneum, UR, and fetal bradycardia. A cesarean was performed and a newborn weighing 2710 g was delivered in cardiorespiratory arrest. The newborn was actively resuscitated and did favorably in the neonatal ICU. UR was visualized at the posterior uterine wall next to the left uterine cornua (Fig. 3), and the placenta was 90% abrupted. Because of uterine atony, a hysterectomy was performed. The mother was discharged from the hospital 7 days later and the newborn after 32 days.

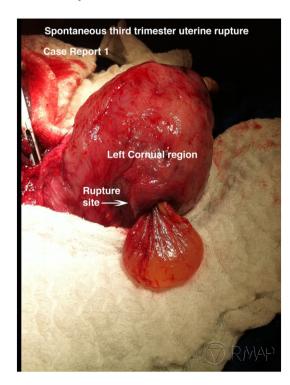
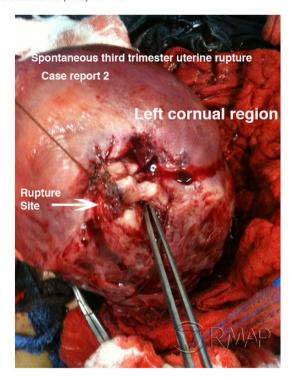


Fig. 2. Uterine exposure during cesarean revealing uterine rupture at the left cornual wall (arrow) with amniotic sac prolapsed.



**Fig. 3.** Uterine exposure during the cesarean demonstrating the site of uterine rupture at the posterior uterine wall next to the left cornua (arrow).

Laparoscopy is an effective treatment for either reduction or cure of endometriosis associated with pelvic pain, dysmenorrhea, and infertility. Endometriosis has also been associated with pregnancy complications (e.g. spontaneous hemoperitoneum, preterm birth, prepartum hemorrhage, placenta accreta, and pre-eclampsia [9]) after extensive resection of DIE. However, uterine dehiscence and UR have been described as late complications of DIE surgery [6,8,9]. Furthermore, these complications can lead to significant maternal and fetal morbidity and mortality [8,9].

The outcomes of pregnancies following surgery for DIE have not been entirely evaluated. Furthermore, the cases of UR reported here were through-and-through tears of the uterine muscle throughout the serosa at the site of extensive and deepest resections. There were a number of commonalities between the two cases. Both patients presented with extensive DIE characterized by deep infiltration into the myometrium at the posterior uterine wall (Fig. 4). To achieve intraoperative hemostasis, these patients required extensive electrocoagulation, leading to decreased vascularization in a thin uterine wall due to extensive myometrial resection, thus increasing the uterine wall vulnerability leading to UR.

A supplementary video is available online, showing the cesareans for both cases (Supplementary Material S1).



**Fig. 4.** Macroscopic longitudinal section of myometrial tissue resected from the posterior uterine wall during laparoscopy, showing deep infiltration of endometriosis.

Supplementary data to this article can be found online at http://dx.doi.org/10.1016/j.ijgo.2015.01.007.

## **Conflict of interest**

The authors have no conflicts of interest.

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# Unexpected uterine malignancy in women who have undergone myomectomy



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Myomas—the most frequent benign tumors of the uterus—affect approximately 25%–40% of women of reproductive age [1]. Traditional surgical options for symptomatic myomas are hysterectomy and myomectomy via various routes. Uterine malignant neoplasms are identified by histological analysis of specimens in 0.22%–0.49% of patients after hysterectomy or myomectomy for a presumed benign leiomyoma [2,3].

The US Food and Drug Administration has stated that use of power morcellators in women with presumed leiomyomas could cause dissemination of leiomyosarcomas [4]. Therefore, the incidence of unexpected uterine malignancy diagnosed after myomectomy (UUM-M) for leiomyoma rather than hysterectomy is important because of frequent morcellation during myomectomies. However, few data on this issue are available. The aim of the present study was to assess the incidence of UUM-M after myomectomy.

In a retrospective study, data from the South Korean Health Insurance Review Agency National Inpatients Sample (HIRA-NIS)

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for 2009–2011 were obtained using a sex- and age-stratified random sampling method (serial number: HIRA-NIS-2009-0066, 2010–0084, 2011–0063) [5]. The Korean National Health Insurance covers almost all citizens of South Korea (approximately 49 million people [5]), and the Health Insurance Review Agency reviews all medical fees. Inpatients with malignant neoplasms of the corpus uteri (diagnostic code C54 according to the International Classification of Diseases version 10) as a primary discharge diagnosis after a myomectomy were analyzed. Approval was obtained from the institutional review board of MizMedi hospital (Seoul, South Korea), but informed consent was not needed because all data were anonymized.

R version 3.0.3 (R, Vienna, Austria) was used for all statistical analyses. The  $\chi^2$  test for proportions was used to compare sample proportions. P < 0.05 was considered statistically significant.

During the study period, 1 843 451 women (approximately 610 000 per year) were enrolled in HIRA-NIS, of which 7020 women (approximately 2300 per year) underwent myomectomy via laparoscopy or laparotomy. The peak age of myomectomy is 36–45 years. UUM-M was recorded in 9 (0.1%) women, with the highest frequency at age 31–35 years (Table 1). No difference in the incidence of UUM-M was noted between women who underwent laparotomy or laparoscopy during 2010 and 2011 (P=0.718).

The incidence of UUM-M in the present study was similar to that reported in a previous study [2,3]. However, given that the mean age at diagnosis for uterine malignancy is 50–60 years, it is clinically notable that the peak age of UUM-M was 31–35 years in the present study.

Although the present study has several limitations—diagnosis miscoding, inability to verify pathology, and lack of follow-up—clinicians should counsel patients, even those at a young age, about UUM-M before myomectomy.

## **Conflict of interest**

The authors have no conflicts of interest.

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