

Research Letter

Intrapartum uterine rupture associated with a scarred cervix because of a previous rupture of cystic cervical endometriosis

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Accepted 15 May 2009

Endometriosis is defined as the growth of ectopic endometrial tissue beyond the uterine cavity. The incidence of endometriosis has been reported to range from 4% to 17% in all women of reproductive age. The most commonly reported presentations of endometriosis are dysmenorrhea, dyspareunia, pelvic pain, and infertility. Most endometrial deposits are confined to the pelvis but extrapelvic endometriosis has also been occasionally documented in the literature [1]. Cervical endometriosis is an uncommon and generally asymptomatic condition. We report an extremely rare case of intrapartum uterine rupture through the scar of the previously ruptured cystic cervical endometriosis.

A 33-year-old nulliparous woman visited our clinic in 2003 because of lower abdominal pain. She had a history of laparoscopic cystectomy because of bilateral ovarian endometriomas in 2000. Dysmenorrhea recurred 2 years after the operation. Transvaginal sonography at our clinic revealed a homogeneous cyst measuring approximately 3 cm × 4 cm over the posterior endocervix (Fig. 1). Abdominal sonography showed a fluid-filled cervical cyst. The patient presented at our emergency department a few hours after her initial visit to our clinic because of lower abdominal pain and persistent vaginal bleeding. She denied having undergone sexual intercourse or trauma before the vaginal bleeding began. Speculum examination revealed a ruptured cystic lesion approximately 2 cm × 2 cm with active bleeding over the posterior lip of the cervix (5-o'clock direction) near the vaginocervical junction without extending to the external orifice of the cervix. Chocolate-like fluid also flowed out from the wound, and cervical endometriosis was suspected. We removed the fragile cyst wall

for pathological study, and the ruptured wound was repaired using absorbable suture materials to achieve adequate hemostasis. Pathological examination revealed scattered endometrial tissue in the cervical stroma covered with squamous epithelium layers (Fig. 2). The patient was discharged after the operation and underwent laparoscopic surgery 4 months later because of infertility. Pelvic endometriosis with adhesion was found, and electrocauterization of endometriosis was performed. She achieved pregnancy through intrauterine insemination. The pregnancy course was smooth until the third trimester, at 35 weeks of gestation, when she was admitted to our ward for tocolysis for 2 days and then discharged under stable condition. Recurrent regular uterine contractions occurred at 37 weeks of gestation, and she was admitted to our hospital again. Cardiotocography showed regular uterine contractions about three times in 10 minutes, and the external orifice of the cervix was dilated by about 1 cm. Because there was no indication for cesarean section, she was admitted to our ward for a trial of vaginal delivery. Although oxytocin for augmentation was administered conservatively because of her previously scarred cervix, massive vaginal bleeding was found at 23 hours after admission, and the cervical orifice dilated suddenly from 1.5 cm to 5 cm in 1 hour. Fetal monitor also showed repeat variable and then late deceleration, and emergent operation was arranged. On opening the abdominal wall, we found internal bleeding of about 300 mL, and the uterus was split with an oblique laceration wound from the previous cervical wound to the left lateral side of the lower uterine segment. A viable female baby was delivered through cesarean section with Apgar score of 4 at 1 minute and 6 at 5 minutes. We repaired the ruptured uterus and cervix through the abdomen and vagina, respectively, with a total blood loss of approximately 1,800 mL. Five days after the operation, the patient complained of left flank soreness, and sonography revealed left hydronephrosis. Cystoscopy and ureteroscopy were arranged, and they showed segmental narrowing

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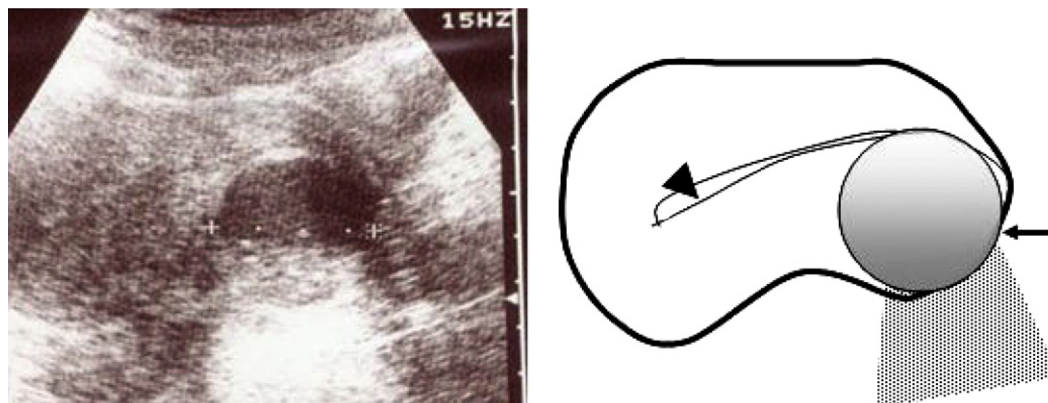


Fig. 1. Cystic lesion (arrow) with internal echo and acoustic shadow about 3.8 cm \times 3.5 cm in size over the posterior endocervix. Arrowhead in the right diagram denotes the endometrium.

of the lower third of the left ureter. Ureteral stricture because of adjacent tissue fibrosis was suspected. Ureter stenting was performed and then removed 2 months after insertion without long-term complications.

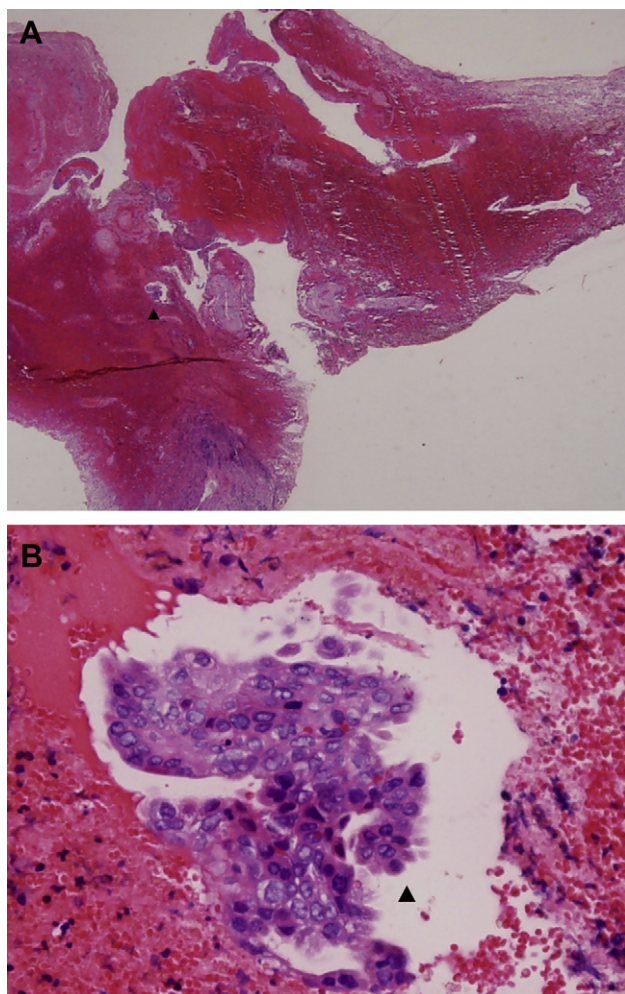


Fig. 2. (A) Cervical specimen showed normal cervical squamous epithelium with acute hemorrhage and endometrial gland (arrowhead) in the stroma (Hematoxylin and eosin, $\times 40$). (B) Cervical specimen showed endometrial glandular tissue (arrowhead) with extravasated red blood cells in stroma (Hematoxylin and eosin, $\times 200$).

Cervical endometriosis is uncommon, and the incidence was about 0.11–2.4% in one colposcopic examination series [2]. Cervical smears in cases of cervical endometriosis can be misinterpreted as high-grade squamous intraepithelial lesion, atypical glandular cells, or adenocarcinoma *in situ* [3], and it is usually only diagnosed retrospectively on histology. Most patients were asymptomatic, and sporadic cases presented with persistent postcoital bleeding [4] or intractable vaginal bleeding [5,6]. On colposcopy or speculum examination, elevated, well-defined, hemorrhagic lesions may be seen. Only a few cases presenting with uterus-like masses arising from the cervix [7] or polypoid masses in the cervix have been reported [8,9].

Several theories have been postulated to explain cervical endometriosis. Previous cervical trauma, such as biopsy, conization, or laser vaporization, was widely accepted as a possible pathogenic pathway for endometriosis implantation. A previous report reviewing the specimens of post-conization hysterectomy, surprisingly, found the incidence of cervical endometriosis to be as high as 43% [10]. Another report demonstrated that 15% of the cervical endometriosis cases may relate to a history of extensive cervical cautery [11]. Another hypothesis to explain cervical endometriosis in the nontraumatic cervix is the development of residual Mullerian tissue in cervical stroma.

Expectant management is appropriate in asymptomatic patients because any destructive therapy, such as excision or cauterization, may carry a risk of reintroducing the endometriosis tissue. In patients with bothersome vaginal bleeding, loop-wide excision of the transformation zone with gonadotropin-releasing-hormone treatment for superficial endometriosis was suggested [3]. In the circumstances of deep cervical endometriosis with rectovaginal septum implantation or coexistence of pelvic endometriosis with symptoms of dysmenorrhea and dyspareunia, total hysterectomy can be considered as a rational therapy.

Cervical endometriosis, which forms a cystic mass like an ovarian endometrioma, is extremely rare. Recently, Iwase et al [12] reported successful management of a case of massive hemorrhage because of the rupture of a cystic cervical endometriosis using a loop electrosurgical excision procedure to avoid hysterectomy. In our patient, heavy hemorrhage was also

noted because of the rupture of a cystic cervical endometriosis, and we performed a simple closure of the wound to achieve hemostasis.

On the other hand, uterine rupture is a major obstetric hazard and more commonly involves a scarred uterus [13]. Although most cases have been associated with a previous cesarean delivery, our patient developed intrapartum uterine rupture that was only associated with a scarred cervix because of a previous rupture of the cystic cervical endometriosis. We described this rare case here to remind the reader of this journal of this unusual complication. Vaginal delivery of a viable baby in cervical endometriosis has been documented in the literature [9]. Despite a scarred cervix not being an indication for cesarean section, a previous rupture of the cystic cervical endometriosis in our case caused her cervix to become vulnerable. This case illustrates that cesarean section may be considered as an alternative mode of delivery to prevent the complication of intrapartum uterine rupture in cases with a history of rupture of cystic cervical endometriosis.

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