



# Vaginal Hemorrhage Associated with Decidualized Rectovaginal Deep Infiltrating Endometriosis during the Third Trimester of Pregnancy: A Case Report

임신 중 탈락막 변화를 동반한 직장질부위 심부자궁 내막증에서 발생한 대량 질출혈: 증례 보고

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Endometriosis-related symptoms are believed to be alleviated during pregnancy. However, pregnancy complications, such as pseudoaneurysm of the uterine artery, rupture of ovarian or uterine vessels, and intraabdominal bleeding from decidualized deep infiltrating endometriosis (DIE) lesion have been rarely reported. Owing to the potential risk of rupture and resultant life-threatening complications, proper diagnosis and close monitoring of decidualized endometriotic lesion are very important despite its low relative risk. Till date, massive vaginal bleeding from decidualized rectovaginal DIE during pregnancy has not been in English literatures. Here, we present the first case of spontaneous massive vaginal bleeding due to decidualized rectovaginal DIE that occurred in the late third trimester of pregnancy.

**Index terms** Endometriosis; Pregnancy; Vaginal Bleeding; Uterine Artery Embolization

## INTRODUCTION

Deep infiltrating endometriosis (DIE) is a type of endometriosis that shows subperitoneal

invasion by ectopic endometrial tissue exceeding 5 mm in depth. It can present in the rectovaginal septum, uterosacral ligament, cul-de-sac, rectosigmoid colon, and urinary tract (1). Main clinical manifestations of DIE are dysmenorrhea and cyclic pain including defecation pain, dysuria, and dyspareunia (1). Although endometriosis is known as a common gynecologic condition among reproductive age, pregnancy is considered a protective factor for endometriosis in the past (2). The consistently high level of estrogens and progestogens during pregnancy can prevent the progress of endometriosis. However, serious pregnancy complications such as intraabdominal bleeding from the rupture of uterine or ovarian vessels during late pregnancy related to DIE have been reported, although rare (3, 4). Life-threatening complications associated with decidualized endometriotic implants and modified vascularization can occur (4). Due to the potential risk of rupture, close monitoring of decidualized endometriotic implant is important despite its low relative risk. In addition, a decidualized endometriosis may mimic malignancy (5). Its symptoms include abdominal pain and vaginal bleeding that are non-specific during late pregnancy, making it difficult to diagnose and treat during pregnancy. Understanding of this complication may help clinicians make proper diagnosis and prevent adverse outcomes. Here, we present the first case of spontaneous massive vaginal bleeding due to a decidualized rectovaginal DIE that occurred in the late third trimester of pregnancy.

## CASE REPORT

A 33-year-old primigravid female visited the emergency unit at 36 weeks gestation with unusually spontaneous vaginal bleeding and lower abdominal pain. Visual inspection of the uterine cervix could not be performed due to active bleeding oriented from posterior vaginal fornices, which were compressed with gauze packing. Her past medical history included dysmenorrhea and painful defecation during menstruation with suspected rectovaginal DIE. She had never been treated with any medication or surgery prior to the current pregnancy. The pregnancy occurred spontaneously, with a single fetus. US screening of the fetus revealed no abnormalities. She had complained irregular vaginal spotting during the pregnancy. On vaginal examination at 34 weeks of gestation, an irregular shaped tender protruding vaginal mass of white-yellow in color with touch bleeding was found in the posterior vaginal fornix. Transvaginal ultrasonography showed a 6.1 cm × 4.1 cm sized ill-defined heterogeneously hypoechoic lesion in the rectovaginal pouch with abundant vascularity in the Doppler exam (Fig. 1A).

Her hemoglobin (Hb) level changed from 13.6 to 11.1 g/dL two hours later. Her vital sign got unstable with hypotension (80/40 mm Hg). Moreover, a non-stress test revealed a non-reassuring fetal status with bradycardia and recurrent late decelerations with regular uterine contraction. We conducted an emergency cesarean section and revealed placental abruption in view of a dark blood clot at the placental margins. However, the cervix, uterus, cul-de-sac, and adnexal regions had unremarkable findings. Uterine contraction was normal. The newborn weighed 2460 gram, with Apgar scores of 2 and 7 points at 1 and 5 minutes, respectively. The pH of umbilical cord was 7.08. However, severe active vaginal bleeding continued after surgery. We could not exclude the diagnosis of vaginal malignancy. Thus, we conducted a

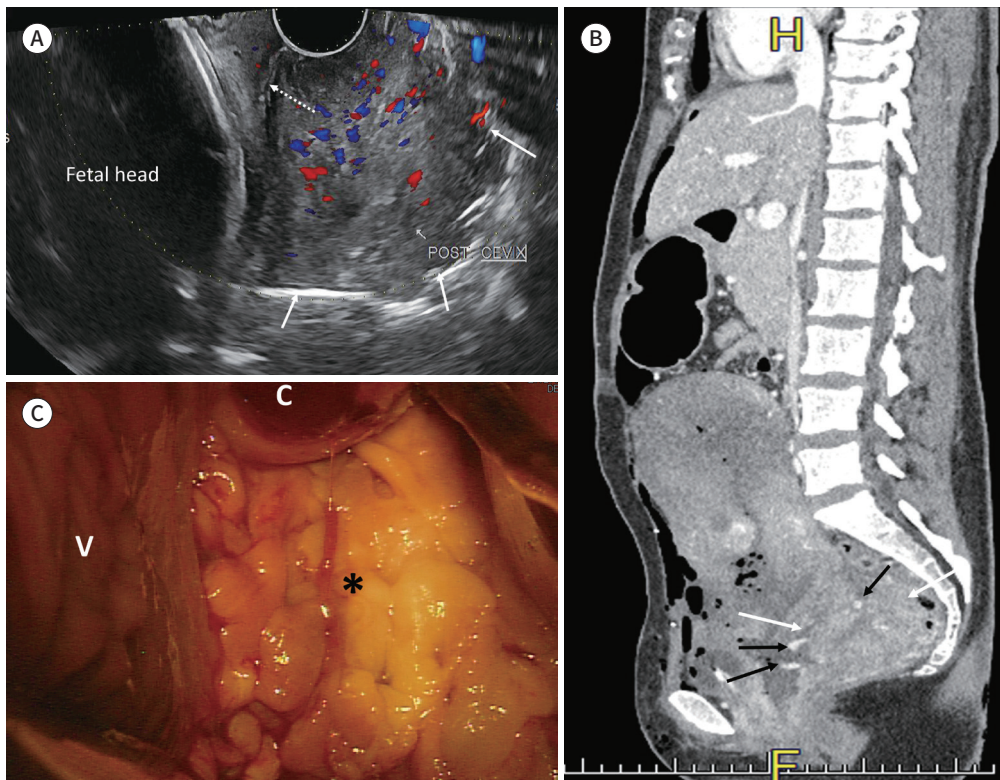
**Fig. 1.** Vaginal hemorrhage from decidualized endometriosis in pregnancy in a 33-year-old female at 36 weeks gestation, complaining vaginal bleeding and low abdominal pain.

**A.** Transvaginal ultrasonography image 2 weeks before the delivery shows an ill-defined heterogeneously hypoechoic lesion measuring 6 cm, in the rectovaginal pouch (arrows) beneath the cervix (dotted arrow) with an abundant vascularity in Doppler image.

**B.** Sagittal reconstruction image of contrast enhanced abdomen and pelvic CT scan shows ill-defined heterogeneously enhancing soft tissue mass in the rectovaginal pouch involving the posterior vaginal wall and closely abutting on the anterior wall of rectum (white arrows). Note that pseudoaneurysms within the lesion and into the vaginal cavity (black arrows) suggest active bleeding.

**C.** Vaginal examination performed through colposcopy shows a vaginal mass (asterisk) that is grossly irregular and protruding in the surface of the posterior wall of the vagina and cervix. The lesion combined with white-yellowish multiple necrotic foci and bled on touch.

C = external cervix os, V = normal vaginal wall



histologic study of the vaginal lesion and placenta. Attempted hemostasis using sutures was ineffective due to vulnerable tissue condition at the posterior vaginal wall and compression applied. Blood tests showed a Hb concentration of 7.7 g/dL after receiving blood transfusion with 1 unit of red blood cells (RBCs). Contrast enhanced abdomen and pelvic CT scan was performed to localize active bleeding focus. Active bleeding was identified from a heterogeneously enhancing mass with indistinct margin in the rectovaginal pouch (Fig. 1B). Subsequent emergency uterine artery embolization was performed with successful hemostasis.

The next day, her Hb concentration was 6.3 g/dL without additional bleeding. We performed a vaginal examination through colposcopy and identified the vaginal mass that was grossly irregular and protruding in the surface of the posterior wall of the vagina and cervix. The lesion was combined with white-yellowish multiple necrotic foci and it bled on touch (Fig. 1C). A digital rectal examination demonstrated no colonic mucosal involvement. The final pathology report noted endometriosis with decidual reaction (decidualized stroma with



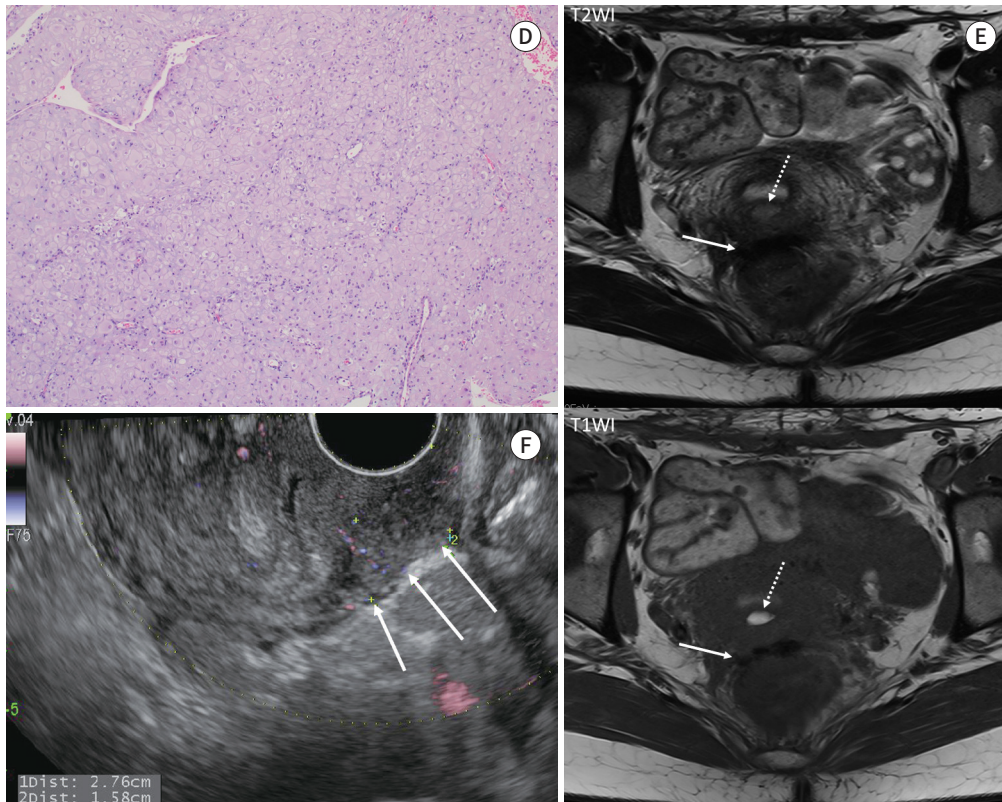
**Fig. 1.** Vaginal hemorrhage from decidualized endometriosis in pregnancy in a 33-year-old female at 36 weeks gestation, complaining vaginal bleeding and low abdominal pain.

**D.** Few glandular structures with decidualized stroma are observed in the subepithelial layer of the vagina (hematoxylin & eosin stain,  $\times 100$ ).

**E.** Axial pelvic MRI (T2W1 and T1W1) 3 months after the delivery shows ill-defined dark SI lesion in the rectovaginal pouch extension into posterior vaginal wall (arrows). Note that T1 and T2 high SI in the posterior vaginal wall suggests hemorrhage in dilated ectopic endometrial gland (dotted arrows).

**F.** Transvaginal Doppler ultrasonography 8 months after the delivery shows decreased size of the ill-defined heterogeneously hypoechoic lesion in the rectovaginal pouch (arrows) and internal blood flow.

SI = signal intensity, T1WI = T1-weighted image, T2WI = T2-weighted image



focal ischemic necrosis, decidualized endometriosis) at the site of vulnerable vaginal mass that caused bleeding. Placenta and umbilical cord histologic examination revealed focal infarction with calcification and intervillous fibrin deposition in the placenta, suggestive of placenta abruption. chorioamniotic membrane and umbilical cord showed no unremarkable finding. On the second postoperative day, her Hb concentration was 5.1 g/dL without bleeding from the vaginal lesion. She received blood transfusion of 3 units of packed RBCs and IV iron. The patient was discharged on the 6th postoperative day with a healthy newborn.

Pelvic MRI was conducted at 3 months after delivery. There was an irregular T1 and T2 dark signal intensity (SI) lesion in the rectovaginal pouch involving the posterior vaginal wall with foci of T1 and T2 high SI foci, suggestive of DIE (Fig. 1E).

Eight months later, inspection of vagina and cervix examination did not find any lesion. Transvaginal ultrasonography showed a mass of 2.7 cm  $\times$  1.5 cm in size and blood flow at the posterior cervix and vaginal wall (Fig. 1F). Tumor marker carbohydrate antigen 125 (CA-125)

was decreased from 50.7 U/mL (1 week after delivery) to 8.2 U/mL (normal range, 0–35 U/mL). Menstruation restarted six months after delivery with continuing breastfeeding. She did not complain of dysmenorrhea, defecation pain, or abnormal bleeding during that period.

This study was approved by the Institutional Review Board of our hospital and the requirement for informed consent was waived (IRB No. 2022-02-007).

## DISCUSSION

To the best of our knowledge, this is the only report of decidualized DIE complicated with massive vaginal bleeding in late pregnancy. In this case of spontaneous vaginal hemorrhage, a close intraoperative inspection of the vaginal lesion with nonremarkable finding of intraperitoneal and uterus indicated that decidualized endometriosis was the cause of the hemorrhage.

Ectopic decidualization defined as extrauterine decidual changes during pregnancy can occur on normal sub-coelomic mesenchymal cells and ectopic endometrium, i.e., endometriosis. The precise mechanisms of aberrant decidualization of endometriosis are incompletely understood. It might be attributed to increased progesterone signaling (5, 6). It usually remains asymptomatic. However, decidualized endometriotic implant during pregnancy can increase in size and change in morphologic feature on US examination (e.g., thick and irregular walls, hyperechogenic papillary excrescences, and abundant vascularity on Doppler exam). It may mimic a malignant tumor (5). In our patient, although she had a history of rectovaginal DIE, malignancy cannot be excluded in the same manner. Thus, we performed biopsy of the lesion.

Notably, when the decidualization of endometriotic implant is extensive, it can result in clinically significant hemorrhage and become life-threatening. Several cases of the rupture of uterine or ovarian vessels contained endometriosis characterized by prominent vascularization and decidualization of the lesion have been reported (3, 4). Besides decidualization, chronic inflammation, adhesion formation, tissue damage, and fibrotic thickening of endometriosis lesions have been suggested as mechanisms of these obstetric complications (2). Zwimpfer et al. (7) have reported that a decidualization of the rectovaginal DIE with uterine artery pseudoaneurysm is successfully treated with uterine artery embolization before delivery. The present case was interesting in that the massive vaginal hemorrhage caused by ectopic decidualization with pseudoaneurysm of vaginal artery occurred in a rectovaginal space of previously suspected endometriosis that accompanied by placental abruption. It was demonstrated that women with endometriosis have an increased risk for placental abruption (2). It can be speculated that placental hypoperfusion resulting from maternal hemorrhage and hypovolemia can cause decidual ischemia with subsequent infarction leading to placental abruption and non-reassuring fetal status (8, 9). Moreover, despite the acute and sudden clinical presentation, chronic inflammation can be implicated in placental abruption (9). Production of inflammatory mediators with decidualized endometriosis may play a part in this complication.

Possible explanations for the hemorrhage of this case are as follows: 1) decidualization of endometriosis can present an abundantly vascularized excrescence (5) and make pseudoaneurysm of adjacent vaginal arteries (7), 2) chronic inflammation and adhesion associated with

endometriosis might have concomitantly weakened the vaginal mucosa and vessels (2). Consequentially, vulnerable tissue might have been exposed in the vaginal lumen, and 3) Mechanical and inflammatory stimulation caused by cervical change with uterine contraction and a progesterone withdrawal trigger at the late third trimester might have facilitated involution of decidualized lesion and bleeding (3, 10).

In conclusion, we reported a case of a complicated decidualized rectovaginal DIE presented with spontaneous massive vaginal bleeding in late third trimester that was successfully treated with uterine artery embolization. Follow-up imaging showing typical DIE features was presented. Rectovaginal DIE is rarely linked to complications during late pregnancy. However, it should be considered in a pregnant patient with unusual vaginal mass or preexisting DIE lesion because resultant hemorrhage is a life-threatening obstetric complication, although it is rare.

### Author Contributions

Conceptualization, L.E.J.; data curation, O.J., J.Y.; investigation, O.J.; methodology, O.J.; resources, J.Y.; supervision, L.E.J.; writing—original draft, O.J.; and writing—review & editing, L.E.J.

### Conflicts of Interest

The authors have no potential conflicts of interest to disclose.

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## 임신 중 탈락막 변화를 동반한 직장질부위 심부자궁 내막증에서 발생한 대량 질출혈: 증례 보고

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임신 중 심부자궁내막증의 증상은 대부분 호전되는 것으로 알려져 있다. 그러나 심부자궁내막증과 관련하여 자궁동맥의 가성동맥류, 난소 또는 자궁동맥의 파열 및 탈락막화가 진행된 병변에 의한 복강내 출혈과 같은 심각한 산과적 합병증이 임신후반기에 드물게 보고되었다. 특히, 심부자궁내막증에 발생한 탈락막화가 진행될 경우 파열 및 출혈로 인한 심각한 모체/태아의 합병증이 발생할 수 있어 정확한 진단을 하고 임신 중 상태를 집중 감시하는 것이 필요하다. 그러나 이러한 경우는 매우 드물어 잘 알려져 있지 않으며, 저자들이 아는 한, 현재까지 보고된 심부자궁내막증에 의한 대량출혈은 모두 복강내출혈이 발생한 경우였다. 저자들은 임신 중 직장질부위 심부자궁내막증의 탈락막화가 진행되고 커지면서 질강내로 노출된 병변과 임신 후반기 병변에서 자연히 발생한 대량 질출혈의 증례를 보고하고자 한다.

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