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Case Report

# Atipical presentations of endometriosis: a case series

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## Abstract

Endometriosis has a broad spectrum of presentation The purpose of this article is to familiarize the gynecologist with the wide spectrum of pelvic and extra pelvic endometriosis and to review the distinctive imaging findings. Its clinical diagnosis could be challenging since it is often confused with infection, abscess, hematoma, and tumors. We hereby report a series of multiple endometriosis presentations appearing during pregnancy, at episiotomy and perineum, as well as after cesarean delivery and hysterectomy. The primary modality of treatment is surgical removal of the lesions, though hormonal therapy is also applied.

**Keywords:** Endometriosis; Episiotomy; Hysterectomy

## Introduction

Endometriosis is the growth of endometrium outside the uterus [1]. Endometriosis lesions are mainly located in the pelvis though it could occur at other sites [2]. It is estrogen-dependent and occurs in around 10% of women in the reproductive age group [3]. Endometriosis has a broad spectrum of presentation [4]. It could be internal located at the walls of the uterus or external located anywhere in the pelvis such as the ovaries, pouch of Douglas, or rectovaginal [5]. It could also develop in scars after surgical procedures affecting the endometrium such as cesarean section, hysterectomy, and episiotomy after normal delivery [6-8]. Symptoms vary among women though some could be asymptomatic. Yet, it is often associated with chronic pelvic pain especially during menses [1, 2]. Other symptoms include dysmenorrhea, dyspareunia, and infertility [9]. The diagnosis of endometriosis could be challenging due to its variable presentations [6]. Ultrasonography, computerized tomography, and magnetic resonance imaging can assist in preoperative diagnosis; however, they are associated with some degree of uncertainty [10]. The mainstay of diagnosis is the visualization of lesions surgically whether by laparoscopy or laparotomy [3]. Nevertheless, histologic examination is needed to confirm the presence of endometriosis [1]. We hereby report a series of multiple endometriosis presentations appearing during pregnancy, at episiotomy and perineum, as well as after cesarean delivery and hysterectomy.

## Cases Series

### Case 1: Endometriosis during pregnancy and Endometriosis during pregnancy

A 23-year-old woman gravida 0 para 0 presented with pain and amenorrhea 2. Pelvis ultrasound revealed 3 cm pelvic mass

at the right ovary with heterogeneous endometrioma-like aspect (Figure1) and luteinique cyst on the left ovary 3cm. After 3 months, the patient became pregnant. During pregnancy, the patient had intermittent severe pain treated with analgesics (paracetamol and non-steroidal anti-inflammatory drugs). There was no evidence for cyst during pregnancy. At 37th week the patient had cesarean section (CS) for fetal distress. During CS, there were multiple lesions of endometriosis in the pelvis (utero sacral ligament, pouch of Douglas, and vesico uterine space) as well as a ruptured ovarian mass that was diagnosed before pregnancy with diffuse opaque dark fluid in the pelvis (Figure 2). Cauterization and hemostasis were done for some lesions. The newborn had an Apgar score of 8 but was admitted to the Neonatal Intensive Care Unit (NICU) for respiratory distress.

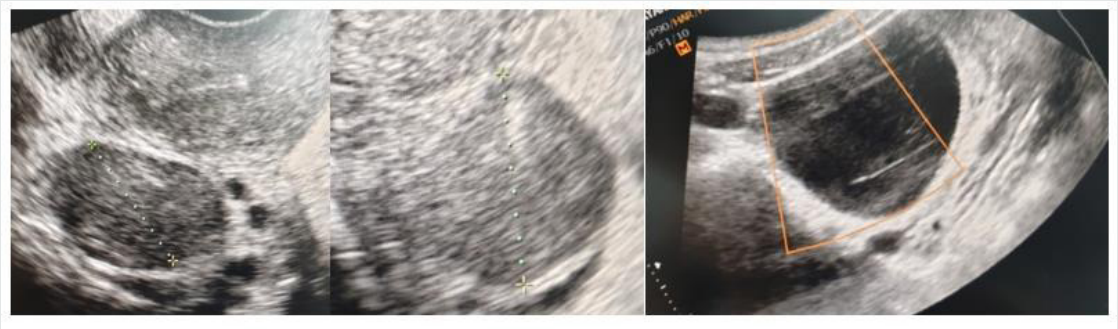

**Figure 1:** Ultrasound image showing endometrioma on the on the right ovary before pregnancy. endometrioma (arrow) with homogeneous low-level echoes no color flow will be seen and luteinique cyst on the left ovary.

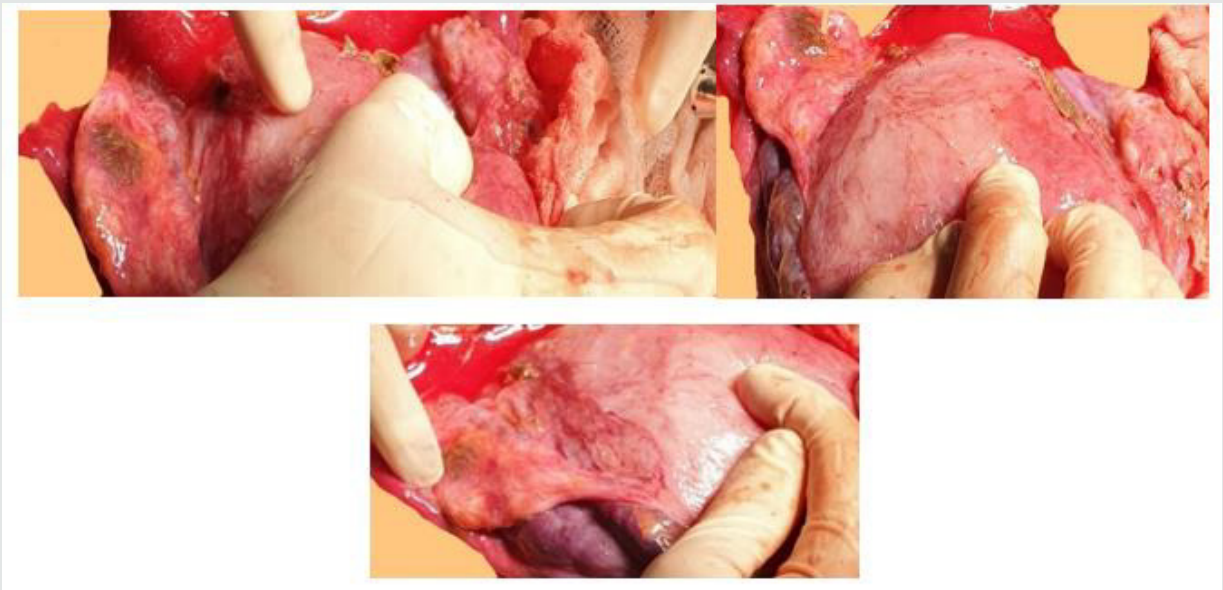

**Figure 2:** During CS, there were multiple lesions of endometriosis in the pelvis (utero sacral ligament, pouch of Douglas, and vesico uterine space) as well as a ruptured ovarian mass that was diagnosed before pregnancy Dark red or bluish cysts or nodules on the surface of peritoneal and pelvic organs.

## Case 2: Recurrent endometriosis on the cesarean section scar

A 27-year-old female gravida 2 para2 who had two previous cesarean deliveries due to fetopelvic disproportion without any complication. She had a history of severe bleeding during menses without experiencing severe pain. She takes estrogen and progesterone contraceptives. The patient presented with pain on the cesarean wound after 2 years from first cesarean. She stopped contraceptive pills 5 months ago. Upon examination, a nodule on the wound was found an approximately 3 cm wide, tender, strict,

and immobile right subcutaneous mass beneath the low segment cesarean scar. Pelvic and abdominal ultrasound showed a 3 cm mass with heterogeneous echo structure (Figure 3a). Surgery was done to remove the mass (Figure 3b). Histology confirmed the diagnosis of endometriosis. After two years, the patient presented with pain. Ultrasound examination revealed a 3 cm nodule on other side of the wound. (Figure 4a). She had surgery for recurrent endometriosis (Figure 4b).

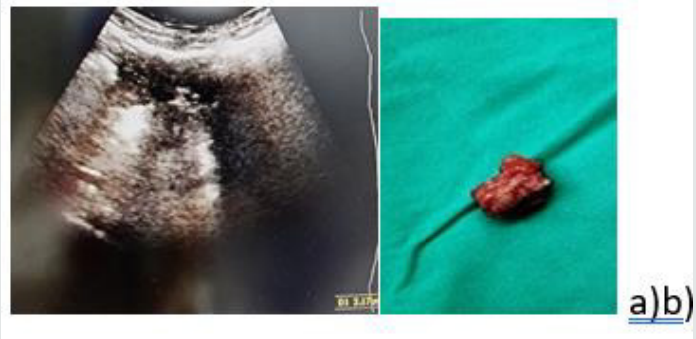


**Figure 3a:** Pelvic and abdominal ultrasound showed a 3 cm × 3 cm × cm, oval-shaped heterogeneous mass within the right rectus abdominus muscle, with no abnormalities of the uterus and ovaries Partly cystic and partly solid intramuscular lesion in the abdominal wall.
**Figure 3b:** Excised mass having brown aspect inside.

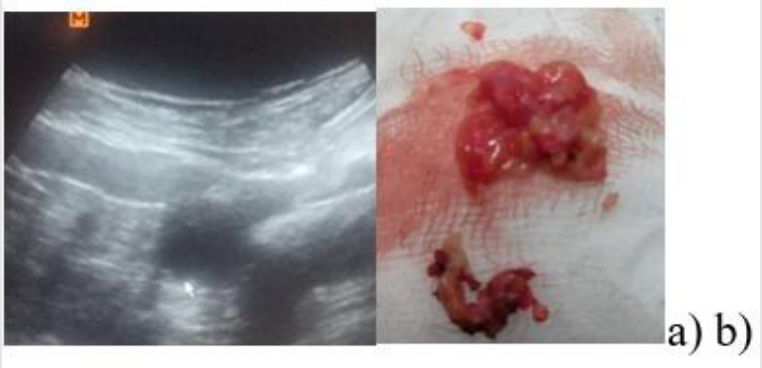


**Figure 4a:** pelvic and abdominal Ultrasound image showing heterogenous aspect mass under the skin of cesarean scar at the left side: solid intramuscular lesion in the abdominal wall.
**Figure 4b:** Excised mass having brown aspect inside.

## Case 3: Endometriosis in the groin near cesarean section scar

A 27-year-old woman gravida 3 para2 1 abortion (1 NVD and 1 CS) patient presented with a painful swelling in her groin. She had been symptomatic for 8 months. Inguinal hernia was initially suspected. She reported that her groin pain increased during her periods. She had a cesarean delivery 1year ago. Abdominal examination revealed a palpable 3 cm mass in the groin area at the right side and lateral to her CS scar. MRI was done and showed a 3 cm mass with a mixed signal intensity near her CS scar. Endometriosis was suspected. The patient had a laparotomy at her groin during which lesions were found and completely removed. Histology confirmed the diagnosis of endometriosis.

## Case 4: Endometriosis on the episiotomy

A 26-year-old woman gravida 2 para1 1 abortion (1 NVD) patient presented with a painful nodule over the episiotomy site for two years. She had forceps delivery seven years ago. Upon examination, a tender, irregular, dark-colored nodule measuring 4 cm was found in the right perineal region over the previous episiotomy scar. She underwent surgery during which the scar and the nodule were excised. Histopathology showed endometriosis with chronic inflammation at the episiotomy scar. The patient was relieved of her presenting complaints after the surgery. She took progestin medication Dienogest (Visanne) for endometriosis treatment for 6 months and then she became pregnant without any complication.

### Case 5: Deep perineal endometriosis behind episiotomy

A previously healthy 28-year-old woman gravida 2 para1 1 abortion had a 1-year history of a painful palpable lesion within the deep left perineum. The pain was correlating with her menstrual period. Gynecological ultrasound examination was normal. Physical examination revealed a deep firm mass in the perineum inferior to the left labia majora. A punch biopsy was performed. Histopathology showed multiple foci of endometrial glands and dense stroma surrounded by tissue. The patient was diagnosed as having endometriosis and was treated with Dienogest for 6 months. She was asymptomatic but when she stopped the medication the severe pain returned. She was advised to undergo surgery during which multiple lesions of endometriosis were excised. The patient now is asymptomatic and is being treated with contraceptives and Dienogest.

### Case 6: Endometriosis of the vaginal vault

A 41-year-old woman gravida 0 para0 presented with prolonged lower pelvic discomfort, constipation, and severe vaginal bleeding. The patient had a history of abdominal hysterectomy 3 months ago due to symptomatic adenomyosis and multiple leiomyomas. After 2 months of surgery, the patient started to have severe vaginal bleeding. She consulted a physician who detected polypoid lesions protruding from the suture of previous hysterectomy at the vaginal vault (Figures 5a & 5b). Her vital signs were stable without fever. Pelvic and vaginal exam induced severe pain during manipulation of the vault site and polypoid lesions were suspected as an old hematoma of the hysterectomy site. Pelvic and abdominal ultrasound examinations showed no signs of active bleeding at the surgery site. For further investigation, under general anesthesia, polypoid lesions were removed with sponge forceps and Di thermocoagulation. Bleeding was controlled with sutures and tampon gauzes that were inserted intra-vaginally and were removed after one day. Histology confirmed endometriosis on the scar. The patient underwent treatment with Dienogest.

**Figure 5(a b):** Polypoid lesions protruding from the suture of previous hysterectomy at the vaginal vault.

## Discussion

Endometriosis is a gynecologic pathology that most commonly develop in the pelvis, such as ovaries, pouch of Douglas, uterosacral ligaments, and anterior abdominal wall [4,5]. The endometrial lesions are composed of glands and stroma that functionally respond to exogenous and endogenous hormonal stimuli. The presentation and evolution are variable, ranging from few lesions on pelvic organs to massive extensive adhesions involving the intestinal and urinary systems [3-5]. Endometriosis occurs in women of reproductive age. It may also develop during pregnancy [11-14]. Bean et al. reported that the prevalence of ovarian endometriomas and deep endometriosis in women in their early pregnancy was 4.9% [15]. Ovarian endometriomas during pregnancy are usually diagnosed by ultrasonography due to its high accuracy and safety [11]. Complications of endometriosis during pregnancy are rare [11]. These include increased risk of placenta previa, placental abruption, hypertension, spontaneous abortion, preterm birth, and cesarean delivery [14]. However, there is no evidence that endometriosis has a detrimental effect on pregnancy outcome [11,12]. In our case, the patient did not have any complication. She only had diffuse pain that was medically treated. On the other hand, endometriosis can develop in the scar after obstetric or gynecologic surgeries [6,16,17].

The incidence of scar endometriosis after cesarean delivery was reported to range between 0.08% and 0.95% [10,18]. The symptoms of scar endometriosis are nonspecific, usually involving pain and swelling at the incision site [6,17]. It can be clinically misdiagnosed as granuloma, hernia, abscess, lipoma, or hematoma [6,10]. Preliminary diagnosis is made through medical history and physical examination in addition to imaging. Ultrasonography is commonly used. Sonographic features are generally not specific showing irregular borders, heterogeneous echotexture, and increased vascularity [6,16]. Accurate diagnosis can be done by histopathological examination of the tissue [6,10,17]. Treatment involves the surgical removal of the lesion [6,10,17]. Kaplanoglu et al. suggested that the complete excision of the lesion together with

around 1 cm of healthy tissue could help to prevent local recurrence [6] since recurrence rate ranged from 1.5% to 9.1% [19, 20]. In our second case, the patient had recurrent scar endometriosis but on a different side since the first lesion was on the right of the scar while the second lesion was central. Scar endometriosis could also rarely occur in scars resulting from episiotomy [8,21,22]. The incidence of endometriosis at episiotomy site after normal vaginal delivery was reported 0.01%-0.04% [6,21]. It may be misdiagnosed as granuloma, cyst or abscess [23]. The presence of a perineal nodule or tender mass, along with cyclic perineal pain with a history of an episiotomy, could be highly indicative of endometriosis [8]. Physical examination usually reveals a dark blue perineal mass [5,8]. The first choice of treatment is complete excision of the perineal endometrial tissue. Other treatment modalities include hormonal suppression [8]. Early treatment is essential since extensive perineal endometriosis can extend to the anal sphincter necessitating primary sphincteroplasty in addition to surgical excision [24]. Another rare type of endometriosis is vaginal vault endometriosis that could develop after hysterectomy [7,25,26].

Choi et al. and Chen et al. reported two cases of vaginal vault endometriosis post vaginal hysterectomy who presented with sudden vaginal bleeding without having previous evidence of endometriosis. A possible pathophysiology could be endometrial implantation during hysterectomy [25,26]. Similar to other forms of endometriosis, the primary treatment is total surgical excision [25,26]. Although surgical removal of the lesions is recommended; the use of medications such as Dienogest has shown to be effective [27]. Dienogest is a progestin that binds to progesterone receptors and inhibits gonadotropin secretion. It also has anti-inflammatory and antiproliferative effects on endometriotic lesions; thereby, providing several advantages over combined hormonal contraceptives [27].

## Conclusion

Endometriosis is associated with considerable morbidity. It is still an underdiagnosed disease. Further studies are needed to determine preventative measures. Clinical trials are also necessary to determine effective medical therapies, along with surgical treatment, to prevent disease progression, minimize pain, and improve fertility.

## Conflict of Interest

No conflict of interest was reported.

## Funding Information

None.

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