

Successful pregnancy after excision of cesarean scar endometriosis with uterovesicocutaneous fistula: A rare case report

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ABSTRACT

Scar endometriosis is an infrequent type of extrapelvic endometriosis. The most common extrapelvic form of endometriosis is cutaneous endometriosis, involving scar tissues occurring after obstetric or gynecologic procedures such as episiotomy, hysterotomy, cesarean section, and even laparoscopic surgery. The clinical presentation of scar endometriosis, i.e., tender swellings, mimics other dermatological and/or surgical conditions and delays the diagnosis. Scar endometriosis very rarely can get complicated with uterocutaneous fistula with a reported incidence of very few cases in world literature. We report a case of a 36-year-old woman presenting with scar endometriosis with complicated uterocutaneous fistula 11 years after her last lower segment cesarean section, managed successfully with laparotomy-fistulectomy and sleeve resection of the bladder with repair followed by successful subsequent spontaneous conception and pregnancy terminated by lower segment cesarean section.

Key words: Endometriosis, lower segment cesarean section, uterovesicocutaneous fistula

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INTRODUCTION

Endometriosis is defined as the presence of endometrium-like stroma and glands outside the uterine cavity. It generally occurs in the pelvic sites such as the ovaries, cul-de-sac, uterine ligaments, pelvic peritoneum, bowel, and rectovaginal septum. Extrapelvic endometriosis can be found in unusual places such as in the nervous system, thorax, urinary tract, gastrointestinal tract, and cutaneous tissues; among them, the most frequent location is the abdominal wall. There are various theories concerning the scar endometriosis. One

of them is the direct implantation of the endometrial tissue in scars during the operation. Under the optimal hormonal stimulus, these cells may proliferate (cellular transport theory) or the neighborhood tissue may undergo metaplasia, which leads to scar endometriosis (coelomic metaplasia theory). By lymphatic or vascular pathways, the endometrial tissue may reach the surgical scar and then cause scar endometriosis.

CASE REPORT

A 36-year-old female P2L1 presented in October 2013 with the complaint of pain and swelling on the cesarean scar on and off for 11 years. In addition, she described cyclic bleeding along with pus from this cesarean scar for 11 years. She previously had one spontaneous vaginal

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delivery 14 years ago and cesarean section 12 years back in view of nonprogression of labor. She described pain over the cesarean scar at the lower end that increased during the menstruation and then noticed a swelling over cesarean scar at the lower end. She noticed mild bleeding from scar within the 1st days of her menstruation followed by pus. Examination revealed a well-healed cesarean scar, moderately pigmented area, tenderness at the lower end of the vertical infraumbilical scar on palpation. Ultrasound [Figure 1] showed that ill-defined hypoechoic lobulated lesion 3.3 cm × 2.7 cm is seen in relation to the superior surface of the uterus till skin surface at the site of scar. A linear fluid-filled tract was also seen within it. Based on characteristic history and examination findings, the most probable diagnosis of scar endometriosis with uterocutaneous fistula was kept. The patient underwent exploratory laparotomy. Intraoperatively, an inverted Y-shaped fistula was identified with the help of a thin infant feeding tube connecting abdominal skin with the uterus and to the dome of the bladder. Fistulous tract excised out and the specimen was sent to the pathology department. Left salpingectomy was done in view of hydrosalpinx, and as bladder dome had some swelling attached to the fistulous tract, sleeve resection of the bladder with repair was done in single continuous layer. Uterus was also closed in two layers. Histopathology of the excised tract confirmed endometriosis. The patient's postoperative course was uneventful and her pain subsided. There was no bleeding and discharge from scar afterward. She conceived spontaneously in May 2014, she was a booked pregnancy in our hospital, and her antenatal period was uneventful. She underwent emergency lower segment cesarean section in view of fetal distress and preterm premature rupture of membranes at 34 weeks 2 days of gestation. She delivered a healthy male baby of birth weight 2.42 kg. The patient's postoperative period was uneventful.



Figure 1: Ultrasound picture showing fistulous tract

DISCUSSION

Endometriosis is the presence of endometrial tissue outside the uterine cavity and can be seen in intra- and extra-abdominal locations. Extrapelvic endometriosis is rare, but there are reports of endometriosis in almost all locations, including kidneys, lungs, and central nervous system.^[1] The previous reports of endometriosis in cesarean section scars have reported the incidence as between 0.03% and 0.4%.^[2] A study by Akbulut *et al.*^[3] in 2010 reported the incidence as 0.1% of women who undergo a cesarean section.

Direct mechanical implantation seems to be the most plausible theory for explaining scar endometriosis.

During cesarean section, endometrial tissue might be seeded into the wound, and under the optimal hormonal influences, these cells proliferate. The endometrial tissue may have certain abilities that make implantation and transplantation possible during pregnancy. The interval between prior surgical treatment and onset of symptoms ranged from 1 to 20 years.^[4] The incidence of concomitant pelvic endometriosis with scar endometriosis has been reported to be ranging from 14.2% to 26%.^[5] Ideally, all patients must be examined for concomitant pelvic endometriosis, and at this point, postoperative follow-up should be done for a couple of years and the patient should be under the observation of the gynecologist. Diagnosis of scar endometriosis should involve detailed history taking and pelvic examination. Sonography and fine-needle aspiration cytology can be used, but it is usually diagnosed by surgical excision^[6] and histopathological examination.

Although a rare event, malignant transformation of abdominal wall endometriosis is a possibility.^[1] A case study by Stevens *et al.* suggested that endometriosis in a cesarean scar can transform into metastatic adenocarcinoma.^[7] Complete surgical excision including the adjacent fascia or skin is the recommended treatment for scar endometriosis.^[8] Therefore, wide excision with at least 1 cm margin is considered the treatment of choice, even for recurrent lesions. Several prophylactic procedures have been proposed to prevent residual contamination of the wound. As it is accepted, inoculation of endometrium into the surgical area is the most common cause of abdominal wall endometriosis, it is strongly recommended that the used sponge should be discarded immediately after cleaning the uterine cavity, the suture material used for uterus should not be reused while closing abdominal wall, and finally the surgical area should be cleaned thoroughly and irrigated with saline solution before closure.^[9]

CONCLUSION

Endometriosis should be considered a diagnosis in a patient presenting with a pain in an abdominal scar, particularly with a history of a cesarean section, even many years after the original surgery. Careful closure and avoidance of contamination (changing gloves, needles before closure) following cesarean section may prevent scar endometriosis.

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Conflicts of interest

There are no conflicts of interest.

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