A Case of Endometriosis in Episiotomy Scar with Anal Sphincter Involvement and Extension into Ischiorectal Fossa

ABSTRACT

Episiotomy scar endometriosis is characterized by presence of endometrial tissue (glands and stroma) and is a rare condition. It may also extend and involve the anal sphincter. The ideal treatment is wide excision to prevent recurrence but it may cause fecal incontinence, if the anal sphincter is involved. We describe here a case with much deeper extension into ischiorectal fossa. She was treated with wide local excision and primary sphincteroplasty. She has remained free of recurrence up to 12 months follow-up.

Keywords: Perineal endometriosis, Episiotomy scar endometriosis, Anal sphincter repair.

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INTRODUCTION

Endometriosis is defined as the presence of functional endometrial tissue outside the uterine cavity. Since, Allen first described it in 1896, there have been a hundred published cases of extragenital endometriosis.1

Perineal site endometriosis is extremely rare, accounting for only 0.2% of ectopic endometriosis.1 The first reported case of endometriosis with perineal involvement was in 1923 by Schickele.2 Minvielle and de la Cruz (1968) first described endometriosis in the anal canal.3

Endometriosis is commonest in the pelvis. It is very occasionally found in bizarre sites, such as the pleura, umbilicus, cesarean section scars, perineum or vagina, diaphragm, arm, leg or kidney, but these cases are rare. Extra peritoneal locations include cervix (0.5%), vagina and rectovaginal septum, round ligament and inguinal hernia sac (0.3-0.6%), navel (1%), abdominal scars after gynecological surgery (1.5%) and cesarean section (0.5%). Endometriosis rarely affects extra-abdominal organs, such as the lungs, urinary system, skin and central nervous system.4

Here we give a very rare case report of scar endometriosis in episiotomy scar with anal sphincter involvement, with extension to ischiorectal fossa.

CASE REPORT

A 25 years female, Para 2, both full term spontaneous vaginal deliveries, presented in our outpatient with recurrent swelling and pain in perineum during her menstrual period for the last 6 years and used to subside with analgesics. She had delivered her first child 10 years back with episiotomy and the second one 7 years back without episiotomy. Pain in the perineum was localized without any radiation and used to become most intense just before and immediately after the onset of menstrual bleeding. Patient was symptom free during rest of the month. She was fully continent with normal bowel and bladder function.

Examination under anesthesia showed presence of a hard indurated mass about 2.5 × 2.5 cm in right lateral position associated with healed episiotomy scar and involvement of anal sphincter muscle.

Transperineal ultrasound revealed a heterogeneous echogenic mass of 17 × 13 × 12 mm with an inflammatory zone. Her biochemical parameters were all in normal limit. Proctosigmoidoscopy findings were normal.

MANAGEMENT

Endometriotic scar excision was done along with pelvic floor repair and perineal tear repair. Examination under anesthesia showed presence of a hard indurated mass about 2.5 × 2.5 cm in right lateral position associated with healed episiotomy scar and involvement of anal sphincter muscle.
PROCEDURE

Patient was put in lithotomy position. A radial incision was given and whole of the endometriotic mass extending into ischiorectal fossa was dissected out (Fig. 2). The excision extended medially and part of anal sphincter was also removed. Dissection was done between rectum and posterior vaginal wall. Bilateral sphincter ani muscles were strengthened using opposition technique. Bilateral levator ani were also strengthened. A closed suction drain was placed in right ischiorectal fossa. Episiotomy site wound was sutured (Fig. 3). The excised specimen showed characteristic dark-brown chocolate colored small cysts.

Histopathological examination: Excised endometriotic tissue was sent for histopathological examination which confirmed it to be endometriosis with invasion into anal sphincter muscles (Fig. 4).

The postoperative recovery remained uneventful. She had good resting and squeezing tone even 6 months after surgery.

DISCUSSION

Endometriosis was first described by Rokitansky in 1860 and was defined as the presence and proliferation of the endometrium outside the uterine cavity, commonest site being the pelvis. The etiology and pathogenesis of endometriosis are complex and still incompletely understood. So many theories have been developed:

- Implantation theory
- Coelomic metaplasia
- Transplantation of exfoliated endometrium
- Altered immunology

Transplantation theory is actually derived from the first theory (implantation theory). During vaginal delivery; viable endometrial cells become implanted in the perineum, including the site of episiotomy and result in endometriotic lesions. However, perineal endometriosis without history of delivery cannot be explained by transplantation theory. Lymphatic dissemination theory has been hypothesized for such cases. As there are rich lymphatic communications between uterus, cervix, vaginal and perineum, endometrial tissues can be transported by lymphatic routes and result in perineal endometriosis.

Contact of endometrial tissue with perineal injury often occurs during vaginal delivery, but the incidence of perineal endometriosis is rare. The reasons for rare incidence may include the following:

- Bacteria existing in the perineal wound which can cause infection or even necrosis of the local tissues. The infection and necrosis is not appropriate for transplanted endometrial cells to live.
- After delivery, the level of estrogen decreases, which also makes the growth of transplanted endometrial cells difficult.

Most patients have perineal cyclic pain corresponding to menstrual periods.

Mostly, the color of mass is normal. Sometimes the color of perineum is blue. Some patients have cyclic bleeding in perineal mass.

A high index of suspicion is recommended, when a woman presents with a postoperative scar site lump. A proper history, as well as a thorough examination with appropriate imaging techniques are necessary for arriving at the correct diagnosis. Anal endosonography can be performed with a B and K mechanical probe rotating through 360° with a frequency of 7 and 10 MHz which shows the endometriotic lesion as a hypoechoic area.\(^5\)

Perineal endometriosis (PEM) is the presence of endometrial tissues in the perineal sites. It has been published in obstetric and gynecologic literature since 1949. In nearly half of women with PEM, the lesions erode into anal sphincter.\(^6\) Thus, it is imperative that further examination

![Fig. 1: A tender hard bluish swelling 1.2 × 1.2 cm in size over episiotomy scar](image1)

![Fig. 2: Endometriotic tissue extending into ischiorectal fossa being removed](image2)
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Fig. 3: Episiotomy site repaired

Fig. 4: Histopathology showing proliferative endometrial glands and stroma with intervening muscle fibers and adipose tissue (H&E ×100)

Management of Scar Endometriosis

The treatment of choice is always total wide excision of the lesion, which is diagnostic and therapeutic at the same time. Complete surgical excision of perineal endometriomas should be curative. Medical treatment with the use of progestogens, oral contraceptive pills, and danazol is not effective and gives only partial relief in symptoms and does not ablate the lesion. Moreover, due to side effects, such as amenorrhea, weight gain, hirsutism, and acne, compliance is unlikely. Recently, there have been reports of the use of the gonadotropin agonist (Leuprolide acetate), but it has been found to provide only prompt improvement in symptoms with no change in the lesion size. GnRh analogs: Decapeptil can be prescribed in a dose of 3.75 mg/month for 3 months.

Follow-up and Prevention

Follow-up of endometriosis patients is important because of the chances of recurrence, which may require re-excision. In cases of continual recurrence, possibility of malignancy should be ruled out. Hence, good technique and proper care may help in preventing recurrent endometriosis. Recurrence, supposedly due to incomplete removal, usually appears within 1 year.

REFERENCES