

---

## CASE REPORT

### Deep Perineal Endometriosis in an Episiotomy scar with IHC findings

Shweta Joshi-Warpe<sup>1</sup>, Bhushan M. Warpe<sup>1</sup>, Abhay Y. Desai<sup>2</sup>, Rahul Sakpal<sup>3</sup>

Associate Professors' (Pathology)<sup>1</sup>, Associate Professor (General Surgery)<sup>2</sup>, Assistant Professor (Pathology)<sup>3</sup> at B.K.L. Walawalkar Rural Medical College & Hospital, Shree-kshetra Dervan, District-Ratnagiri, Pincode- 415606, State-Maharashtra, India

---

Article: Received 17-12-2020 , Accepted 13-01-2021 , Published 18-01-2021

---

**Abstract:** Perineal endometriosis (PE) is a rare entity which can be explained by direct implantation of endometriotic cells over episiotomy wound and subsequent development of scar endometriosis. Present patient presented with current pain in her perineal swelling at episiotomy scar which was associated with menstruation. First-line simple procedure of FNAC detected endometrial cells and stromal fragments from scar tissue. Therapeutic wide local excision (WLE) was done and sent for histopathological examination (HPE). HPE and Immunohistochemistry (IHC) confirmed it as PE, staining the endometrial glands and endometrial stromal component in scar tissue. The post-surgical stay was uneventful. Patient was followed up after six months with no signs/ symptoms of recurrence.

**Keywords:** Perineum, endometriosis, episiotomy-scar, ER, PR

#### Introduction:

Rokitansky first described the presence of endometrial glands and stroma outside the uterine-cavity about 150 years back. In 1896, Allen described extra-genital endometriosis [1]. Perineal endometriosis (PE) was first reported in 1923 [2].

Clinically, it can be difficult to diagnose PE at times due to long latency period in years before symptoms develop, like in this case and due to under-reporting. Differential diagnosis of this entity includes suture granulomas, abscesses, hematomas, lipomas, sebaceous cysts, desmoids or malignant tumour [3]. Pelvic-MRI/Endo-anal ultrasonography (USG), FNAC, HPE report and associated novel

Immunohistochemistry (IHC) with ER/PR antibodies can help to diagnose this condition confidently.

#### Case Report:

A 36-year-old P<sub>1</sub>L<sub>1</sub> woman after eight years of her only per-vaginal delivery presented with perineal swelling since past two years which increased each time in size during her periods. She had cyclical pain in the perineum for past one year, which brought her to the hospital. This localized, bearable pain lasted throughout her period associated with itching. General practitioner advised her analgesics for pain relief during her periods since past one year. There was no history of dyspareunia or difficulty in

passing stool/urine. Patient did not have any major illness.

Her only normal vaginal delivery was done with episiotomy with post-partum curettage. Her menstrual cycles were regular; flow was normal and had mild pain in the abdomen as well as episiotomy scar. On examination vitals were stable and systemic examination was normal. On local examination of perineum: overlying skin was healthy but an old right medio-lateral episiotomy scar was seen. Examination during menstruation revealed reddish brown spots with blood oozing from the scar.

On palpation, a 2×2 cm indurated tender, firm reddish-blue swelling was felt over the episiotomy scar. Abdomen ultrasonography, per-speculum, per-vaginal examinations was within normal limits. As clinically there was no ano-rectal involvement on per-rectal examination and patient denied any history of anorectal complaint, endo-anal ultrasonography was not performed. FNAC done showed benign ectopic endometrial cells in sheets, clusters with benign endometrial stromal fragments suggestive of episiotomy scar endometriosis (Fig. 1).

### FIGURES:

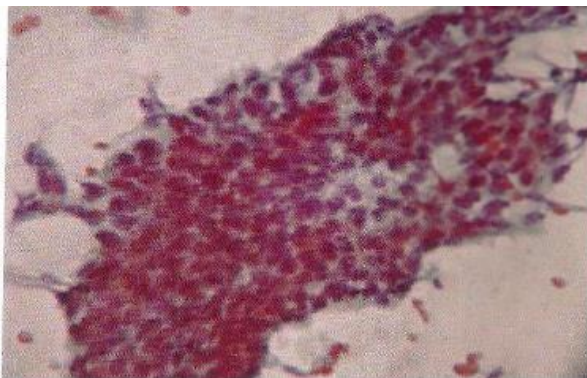


Fig.1A

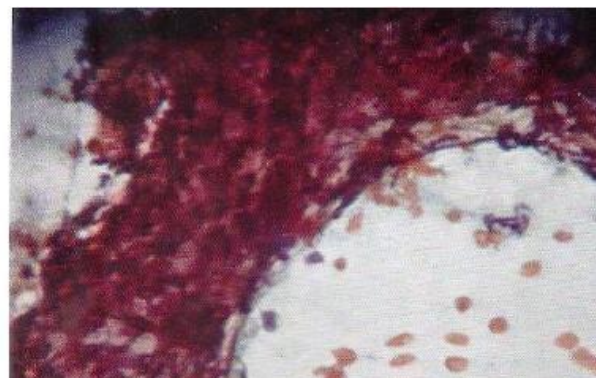


Fig. 1B

**Fig. 1: Photomicrograph of FNAC of perineal swelling- Benign ectopic endometrial glandular cells in sheets (IA); Fragment of benign spindled endometrial stromal component (IB); (PAP x400).**

Incision was given on the skin, overlying the swelling. 3×3cm lesion was separated from surrounding tissue by sharp dissection. WLE of firm mass with 1 cm margin was done to prevent recurrence under spinal anesthesia. Reconstruction of perineum was done in layers. Post-operatively injection Leuprolide 3.75 mg intramuscular was given to prevent recurrence.

The excised WLE swelling was sent for HPE. This swelling revealed unremarkable stratified squamous lining epithelium with deep sub-epithelial fibro-collagenous tissue showing few

ectopic endometrial glands in early proliferative phase surrounded by highly-compact endometrial stroma within the scar tissue (Fig. 2).

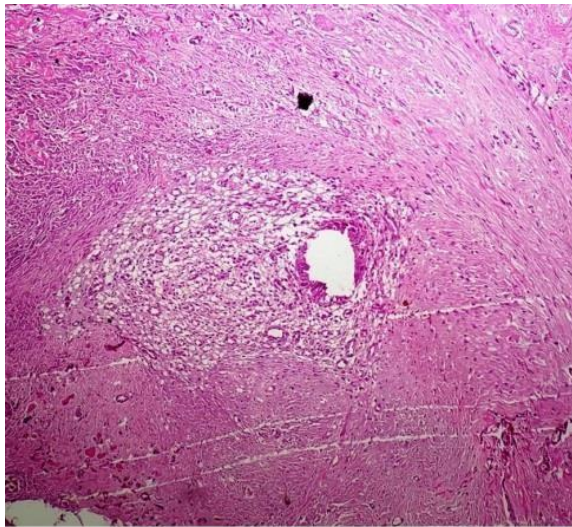


Fig. 2A

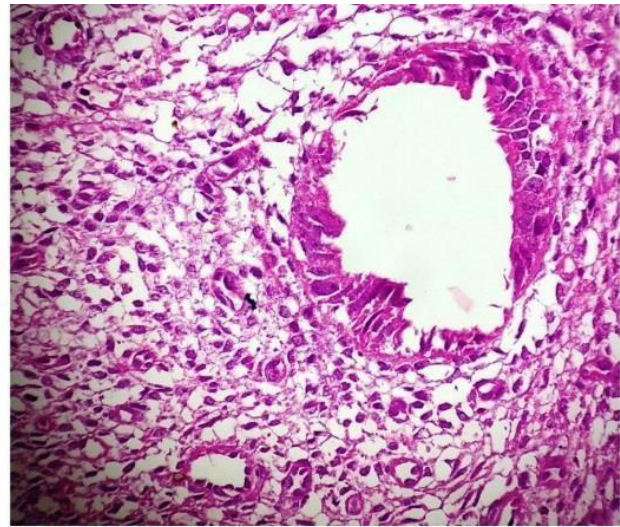


Fig. 2B

**Fig. 2: Photomicrograph of perineal biopsy - Ectopic endometrial glands and surrounding compact endometrial stroma within scarred fibro-collagenous tissue (2A: H&E, x40 ; 2B : H&E, x400).**



Fig. 3A: ER positive

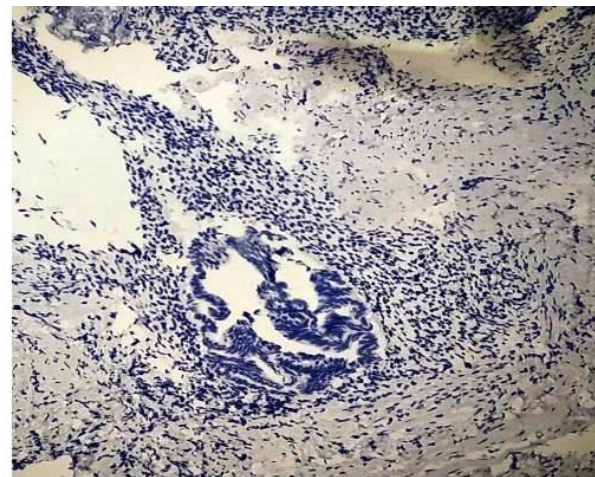


Fig. 3B: PR negative

**Fig. 3: Photomicrographs of IHC staining: Nuclei of ectopic endometrial glandular epithelium and surrounding endometrial stromal nuclei are strongly and diffusely stained by ER antibody (3A). PR antibody does not stain the same component (3B). Patient was in early proliferative stage of menstrual cycle, as per clinical details.**

IHC was done at our set-up to confirm HPE report with Estrogen receptor (ER) and Progesterone receptor (PR) antibodies (Fig. 3). ER revealed strong, diffuse nuclear positivity in endometrial glands and surrounding endometrial stroma in scar site. PR did not stain the nuclei of endometrial glands and stromal cells. This IHC report confirmed that patient was in early-proliferative stage as per her menstrual-cycle details.

Her final (HPE + IHC) report was Deep perineal endometriosis in an episiotomy scar. There was no recurrence after six months of follow-up.

#### **Discussion:**

Endometriosis refers to ectopic functional endometrial glands and stroma lying outside the uterine cavity. Incisional /scar endometriosis is endometriosis occurring in a surgical scar [4]. The incidence has been estimated to be only 0.03% to 0.15% of all cases of endometriosis [5]. 10-15% of ladies aged, 25 to 35 years of age have scar endometriosis [6].

Endometriosis is mostly an intra-pelvic disease which occurs in the ovaries with 30% ovarian incidence, also called as endometriomas. Other sites for endometriosis include cervix, uterine ligaments, fallopian tubes and pelvic peritoneum. Extra-pelvic endometriosis is a relatively rare condition, accounting for approximately 12% of all cases which includes abdominal wall and diaphragm [7].

Endometriosis predominantly locates on peritoneal surfaces, but can also affect the vagina, vulva, recto-vaginal septum and

perineum, usually secondary to surgical or obstetric trauma or scar [4]. Scar endometriosis has been reported after:

1.) After LSCS or abdominal surgeries like hysterectomy, tubal ligation, ectopic pregnancy surgery and 2.) After per-vaginal delivery with episiotomy scar or other vaginal surgeries [6,7].

Out of the many theories proposed for the etio-pathogenetic of endometriosis (direct implantation, lymphatic dissemination, coelomic metaplasia or hematogenous spread), PE can be attributed to autologous transplantation of viable endometrial cells on episiotomy wounds after vaginal delivery, the risk is increased when manual uterine exploration or postpartum curettage are done (as in our case) [8].

The cyclic nature of localized swelling/pain, which worsens at the time of menstruation, and a frequently reported history of surgery, are pathognomonic of PE [6-8]. Usually a triad of mass, cyclic pain and previous episiotomy is observed in PE.

Imaging modalities such as ultrasound and MRI are indicated in case of large lesions, anal involvement and surgical planning. MRI is also useful to differentiate it from another lesion like lipoma, abscess. The endo-anal ultrasonography is particularly reliable for visualizing anal sphincter involvement. CT-scan lacks resolution and so is rarely advised [8]. For cytological diagnosis of endometriosis there must be presence of any two of three features on cytological smears: endometrial glands, stromal cells and hemosiderin-laden macrophages [3].

The treatment of choice is WLE of the lesion with 1 cm margin on all sides. Surgical method has an advantage of providing biopsy sample to confirm diagnosis and rule out malignancy, which occurs few months to over 40 years after PE (9). Biopsy reveals ectopic endometrial glands and endometrial compact stroma in perineal scar. IHC with ER, PR antibodies show nuclear positivity based on the phase of her menstrual cycle. ER and PR are both strongly positive in late proliferative stage. [10].

### Conclusion

To prevent progressive involvement of the anal sphincter in PE, early diagnosis and treatment with WLE and Hormonal therapy is imperative. As episiotomy is very frequently performed at the time of vaginal delivery, incidence of PE can be reduced by washing the episiotomy wound with normal saline before suturing, avoiding manual uterine exploration and postpartum curettage. FNAC, HPE report and IHC for ER/PR status are useful for confirmation of diagnosis.

### Conflict of Interests:

The authors declare that there is no Conflict of Interests.

### References

1. Al-Jabri K. Endometriosis at caesarian section scar. *OMJ* 2009; 24: 294-5.
2. Laadioui M, Alaoui F, Jayi S, Bouguern H, Chaaara H, Melhouf MA. L'endométriose périméale profonde sur cicatrice d'épisiotomie: à propos d'un cas rare [Deep perineal endometriosis on

episiotomy scar: about a rare case]. *Pan Afr Med J.* 2013;16:112.

3. Andola US, Andola SK, Sanghvi KJ. FNAC diagnosis of Scar endometriosis: A report of 3 cases with review of literature. *J Basic Clin Reprod Sci* 2013; 1: 62-4.

4. Gauna B, Rodriguez D, Cabré S, Callejo C. A case of endometriosis in episiotomy scar with anal sphincter involvement. *International Journal of Clinical Medicine* 2011;2:624-6.

5. Danielpour PJ, Layke JC, Durie NC. Scar endometriosis, a rare cause for a painful scar: A case report and review of the literature. *J Plast Surg* 2010;18:19-20.

6. Tam T, Huang S. Perineal endometriosis in an episiotomy scar: case report and review of literature. *Journal of Gynaecology* 2012;4:93-6.

7. Uzuncakmak C, Guldaz A, Ozcam H. Scar endometriosis: A casereport of this uncommon entity and review of the literature. *J Clin Case Rep* 2013;3:1-3.

8. Demir M, Yildiz A, Ocal I, Yetimlar MH, Kilic D, et al. Endometriosis in episiotomy scar: A case report. *J Cases Obstet Gynecol* 2014; 1: 8- 10.

9. Saloum NM, Qureshi S, Ibrahim SA, Alrashid A. A Rare Case Report of Endometriosis in an Episiotomy Scar without Anal Sphincter Involvement. *EC Gynaecology* 2018; 7(12): 466-70.

10. Khan KN, Fujishita A, Kitajima M, Hiraki K, Nakashima M, Masuzaki H. Occult microscopic endometriosis: undetectable by laparoscopy in normal Peritoneum. *Human Reproduction* 2014;29(3):462–72.