



Recurrent perineal scar endometriosis: A case report

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ABSTRACT

Introduction: Endometriosis is a chronic disease with the presence of endometrium-like tissue containing endometrial glands and stroma outside the uterus. The incidence of episiotomy scar endometriosis after vaginal delivery is 0.06–0.7%.

Case presentation: A 28-year-old parous woman with two previous vaginal deliveries with episiotomy presented to the outpatient department with severe pain and swelling in the perineal region over the past year, leading to difficulty in sitting, coitus and routine essential activities like defecation. She had undergone local perineal nodule excision surgery twice, which confirmed her perineal scar endometriosis diagnosis one year earlier at another hospital with no improvement in symptoms. Physical examination revealed a firm, tender, deeply embedded palpable nodule measuring approximately 3 - 4 cm in the left posterolateral aspect of the distal vagina. Wide local excision of the nodule with a clear margin of 1 cm was performed. The nodule extended up to the left ischial tuberosity and apex up to the pudendal vessels complex.

Discussion: The classic diagnostic triad of perineal endometriosis were present in this patient. Surgical intervention with wide local excision with a clear margin of approximately 1 cm of healthy tissue reduces the chance of recurrence.

Conclusion: Awareness of this condition among medical practitioners will lead to early diagnosis and excision. Timely intervention in the form of excision with free margins is the definitive treatment and provides complete pain relief and good quality of life for endometriosis patients.

1. Introduction

Endometriosis is a chronic disease with the presence of functional endometrial-like tissue containing glands and stroma outside the uterus. Endometriosis is commonly observed in the pelvis, ovaries, peritoneum, pouch of Douglas and uterosacral ligaments [1]. Extra-pelvic sites of endometriosis are uncommon and pose a therapeutic challenge when found in umbilicus, bladder, lymph nodes, gastrointestinal tract, under the surface of the diaphragm, abdominal surgery scar areas following hysterectomy and caesarean section, and in the perineum following vaginal delivery with episiotomy. It is the second most prevalent benign gynaecologic disease in women of reproductive age, with a prevalence of 10–25%. The incidence of abdominal wall endometriosis after caesarean section is 0.03–1.7% and that of episiotomy region endometriosis after vaginal delivery is 0.06–0.7% (2). Nevertheless, the very low percentages reported in the literature can be attributed to a failure to consider

perineal endometriosis as a differential diagnosis for a perineal mass. The classic triad highly diagnostic of perineal endometriosis is progressive cyclical perineal pain, tender nodule or perineal mass on examination in a patient with a past history of vaginal delivery with an episiotomy [3,4]. Recurring cases of endometriosis are due to an in situ lesion, microscopic metastasis and insufficient surgical removal leading to residual disease [5].

The present case highlights the importance of correct diagnosis, mapping of lesions and excision of perineal scar endometriosis to reduce recurrence and multiple surgeries. The case is reported in accordance with CARE guidelines.

2. Case Presentation

A 28-year-old woman, P2L2, presented to the outpatient department with cyclical pain and swelling in the perineal region starting one year

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before presentation but with a significant worsening of pain and her quality of life in the last few months. The patient described her pain as severe, leading to difficulty in sitting, coitus and performing routine essential activities like defecation. She had had two vaginal deliveries necessitating an episiotomy 7 and 4 years earlier. Three years after her last delivery she started experiencing pain the perineal region and also noticed a swelling at her perineal episiotomy scar site, for which she underwent local excision with a confirmatory histopathology report of perineal scar endometriosis and a diagnosis of perineal endometriosis was made.

After local excision, her symptoms persisted and she went to several gynaecologists and underwent scar endo excision again in an outside setting, following which her symptoms worsened and her quality of life deteriorated to a point where she was having continuous pain. On a 10-point visual analogue scale she scored 10 for dyschezia and 10 for dyspareunia. After two incomplete excisions at other institutions she presented with a highly tender deformed distal vagina, with puckering and scarring. Examination led to a management conundrum as it was highly tender and ultrasound was inconclusive due to previous recurrent surgeries and could only pick up features of a puckered scar. Hence magnetic resonance imaging (MRI) was undertaken to confirm a diagnosis.

These findings were confirmed with high-resolution trans-perineal ultrasound and pelvic MRI. The pelvic MRI showed the spiculated mass in the left ischial fossa abutting the ischial tuberosity with deformity of the left side of vagina secondary to scarring. The mass was hypointense in T2 signal with hyperintensity foci in T2 and T1FATSAT was consistent with the blood signal. Irregular blooming was evident on an SW1 image with restricted DW1 and low ADC value of 0.7. These findings were consistent with episiotomy scar endometriosis extending up to the left ischial tuberosity (Fig. 1). Under general anaesthesia, a firm deeply embedded nodule was palpable in the left posterolateral aspect of the

distal vagina, measuring approximately 3–4 cm. The distal vagina was deformed and puckered with scarring (Fig. 2A). Digital rectal examination elicited normal anal sphincter tone and the perineal mass was abutting the rectal wall. Wide local excision of the nodule was performed with a clear 1 cm margin. The nodule was extending up to the left ischial tuberosity and apex up to the pudendal vessel complex (Fig. 2B). Wide local excision was done and it led to a dead space of 5 × 6x7cm which was allowed to heal by secondary intention. Although the endometriotic nodule was very deep, the mass was excised and anal sphincters were not involved in the procedure although it was abutting and in close contact with the rectal wall (Fig. 2C). The dead space was dressed daily. Cut specimen tissue revealed small cystic spaces containing chocolate-coloured fluid (Fig. 3). Microscopic examination showed intensive endometrial glands with typical stroma, blood, and hemosiderin-laden macrophages. The histopathology showed endometrial glands with columnar lining which was consistent with scar endometriosis and no evidence of malignancy (Fig. 4).

The patient was discharged on the third postoperative day. She had dressings changed daily for 2 weeks. At 4-week follow-up, she was asymptomatic and able to sit comfortably and perform regular activities and reported no pain during defecation or intercourse. This case has been reported to highlight the importance of surgical expertise in wide excision of endometriotic nodules with clear margins to prevent recurrence of lesions.

3. Discussion

Schickele reported perineal endometriosis for the first time in 1923; since then, several studies have reported the condition. [6]

The exact aetiology of endometriosis is unknown but it is thought that scar endometriosis occurs after direct inoculation of the endometrial cells during surgery into the subcutaneous tissue of the abdominal

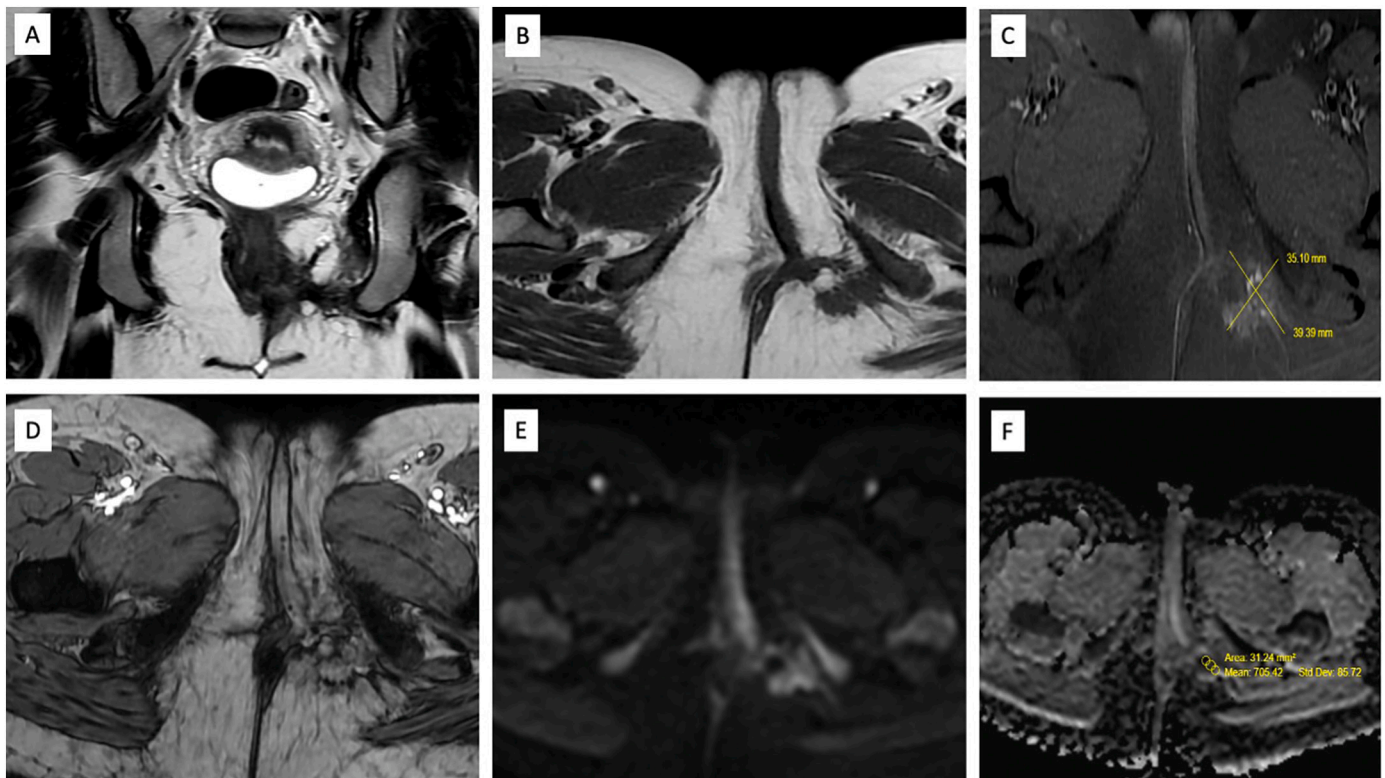


Fig. 1. : (A-F) Irregular spiculated mass in left ischial fossa abutting ischial tuberosity with deformity of left side of vagina secondary to scarring. Mass shows T2 hypointense signal with T2 hyperintense cystic foci on T2W images(A) and isointense signal on T1W (B). Hyperintense foci in T1FATSAT (C) consistent with blood signal. Irregular blooming on SW1 image (D) with restricted DW1 and low ADC value of 0.7 (E,F). These findings were consistent with scar endometriosis diagnosis extending up to the ischial tuberosity.

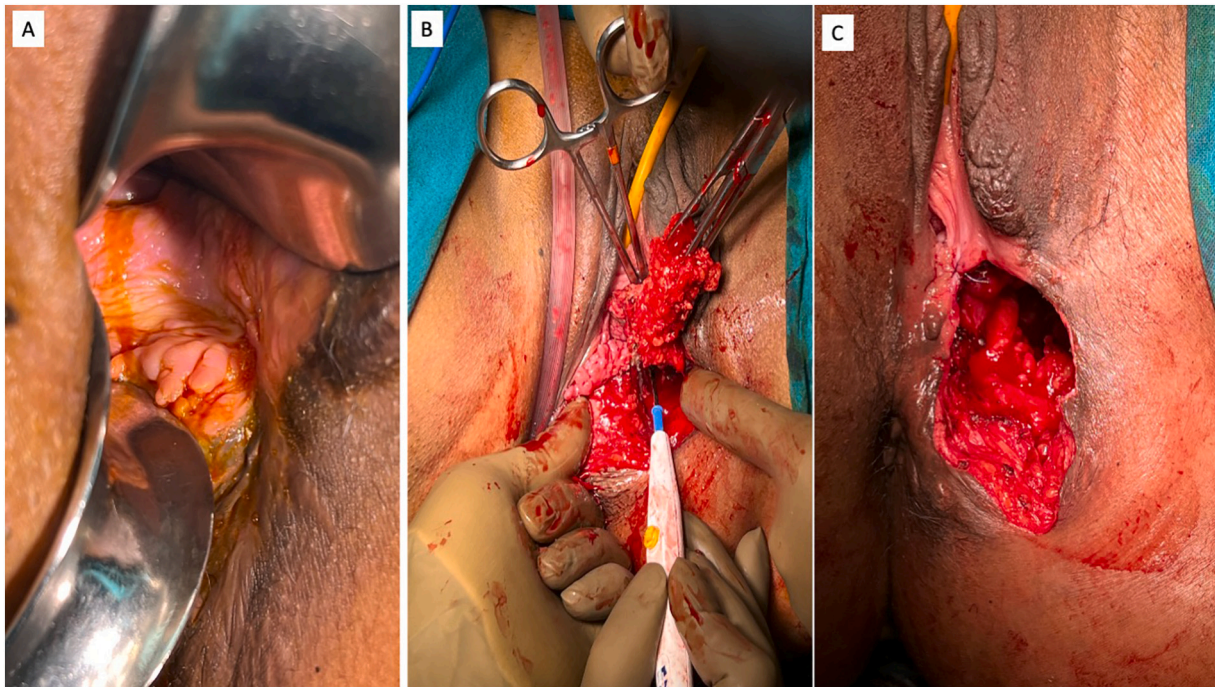


Fig. 2. (A) Preoperative image showing puckered scar with deformed distal lumen of vagina. (B) Wide local excision abutting rectal wall. (C) Post-excision picture demonstrating the intact sphincter ani complex and dead space 5x6cm, which was allowed to heal by secondary intention.

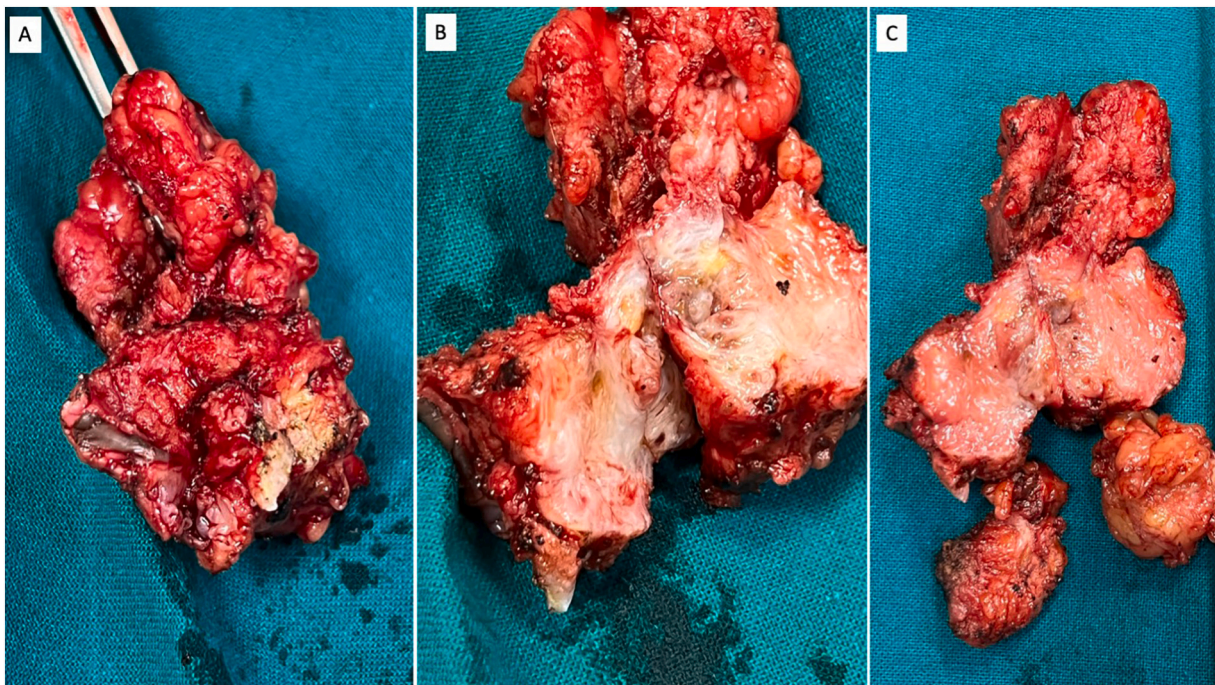


Fig. 3. (A) Excised specimen. B & C) Cut section showing full-thickness nodule.

wall and sometimes into the rectus muscle planes. Theory of implantation is considered in women with episiotomy scar endometriosis with no signs of pelvic endometriosis. [7]

The etiopathogenesis of endometriosis is unclear but several theories have been proposed. Minh theory states that endometriosis is caused by transformation of pluripotent peritoneal mesothelium. Other theories proposed are lymphatic dissemination causing migration of cells, direct implantation, hematogenous spread, familial, immunological factors, and metaplasia of mullerian remnants causing endometriotic nodule

formation. Predisposition is polygenic and multifactorial [8]. The risk of endometriosis rises 7-fold in women with a history of endometriosis in a first-degree relative. On the other hand, the theory of transplantation can also be considered for the cause of perineal endometriosis, with implantation of the shedding decidual tissue at the episiotomy wound site, as. Recently a neurologic hypothesis has been suggested, where the lesions are seen infiltrating the large bowel along the nerves, away from the site of primary lesions.

The classic triad highly diagnostic of probable perineal

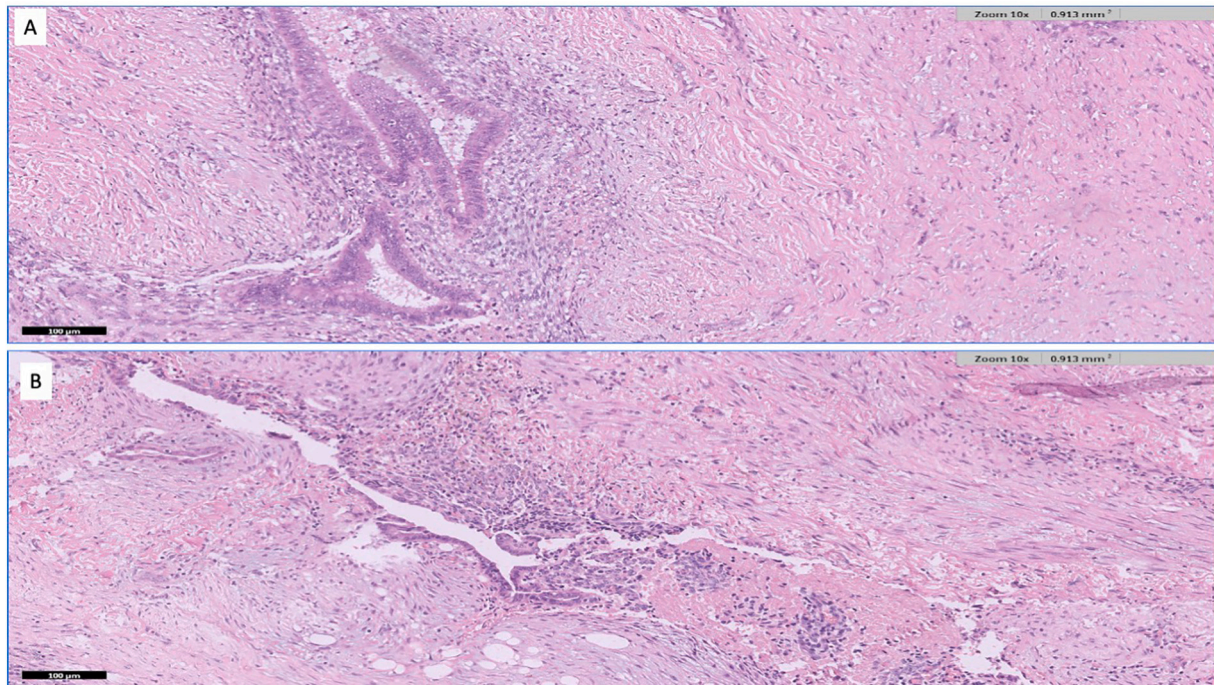


Fig. 4. (A) Endometrial glands showing columnar lining, 10 \times . (B) Irregular endometrial gland with surrounding compact stroma, 10 \times .

endometriosis, present in this patient, are [1] an episiotomy or past perineal tear during vaginal delivery; [2] a tender mass or nodule at the perineal lesion; and [3] cyclic and progressive perineal pain. Differential diagnoses of a suture granuloma, desmoid tumor, sarcoma, and metastatic malignancy should always be considered. The incidence of malignant transformation to a clear cell carcinoma is less than 1%, but should always be ruled out in a recurrent case of scar endometriosis. [9]

The clinical manifestations range from an asymptomatic mass to a cyclic and progressive painful perineal mass. Ultrasonography is the most commonly used imaging method and aids in confirming the diagnosis. On USG imaging, it appears as a hypoechoic or heterogeneous nodule (depending on the solid and/or liquid component) or sometimes as a hyperechoic nodule with blurred and irregular margins that infiltrate the surrounding tissue; the nodules vary in size, shape, amount of fibrosis, blood and the menstrual cycle. In cases with suspicion of deep invasion, MRI can aid the diagnosis [10].

Surgical intervention with wide local excision with clear margins of approximately 1 cm of healthy tissue helps to reduce the incidence of recurrence. Recurrence of scar endometriosis is due to incomplete surgical removal of the endometriotic nodule. Perineal scar endometriosis can be prevented by ensuring that the episiotomy scar is not contaminated with debris and blood. Gloves should be changed prior to performing an episiotomy wound repair. When the anal sphincter is involved, sphincteroplasty may be required to reduce the chances of faecal incontinence [11–13].

4. Conclusion

Heightened awareness of this condition among medical practitioners is important in order not to delay diagnosis and intervention. Diagnosis requires proper understanding of a patient's medical history and pelvic examination. Timely intervention and proper excision with free margins is the definitive treatment and provides complete pain relief and good quality of life for endometriosis patients. It is also important that the patient reaches the right specialist on time so as to avoid multiple surgeries as was noted in this case. The predisposing factors may not only be iatrogenic, as genetic predisposition is also responsible. Perineal endometriosis is rare but should be considered in women with cyclical pain at

the episiotomy site, history of vaginal delivery and a palpable mass in the perineal area. This case was unique as the patient underwent incomplete excision twice before this presentation and proper excision finally gave her symptomatic relief and ended her suffering. The disappearance of pain and improved quality of life lay emphasis on the timely referral to a specialist centre and the expertise required for complete excision without injuring other vital structures, such as the rectum, anal sphincter and pudendal nerves when dealing with such cases.

Contributors

Vimee Bindra contributed to patient management and the literature review, and drafted the manuscript.

Nikitha Reddy contributed to the literature review, and the drafting and editing of the manuscript.

C Archana Reddy contributed to patient management and the editing of the manuscript.

P Swetha contributed to the editing of the manuscript.

Kishore V Alapati contributed to patient management and the editing of the manuscript.

Madhavi Nori contributed to pre-operative diagnosis and the editing of the manuscript.

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Patient consent

Written informed consent was obtained from the patient for the publication of this case report and accompanying images.

Provenance and peer review

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Conflict of interest statement

The authors declare that they have no conflict of interest regarding the publication of this case report.

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