

CASE REPORT

Perineal scar endometriosis

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Abstract

Endometriosis is defined as the presence of ectopic endometrial tissue outside the uterine cavity. It is a benign condition commonly observed in women of the reproductive age group. It can occur in both pelvic and extra-pelvic sites. Moreover, pelvic endometriosis is relatively common, as compared to extra-pelvic endometriosis. The most frequent site for pelvic endometriosis is ovary. It can also affect rectum, uterosacral ligaments, rectovaginal septum, urinary bladder. Extra-pelvic endometriosis is rare and when it occurs, does so more frequently in surgical scar sites, especially in caesarean section scar. Endometriosis in an episiotomy scar is extremely rare but can lead to significant morbidity in patients due to local infiltration. This condition can be diagnosed by the presence of the classical clinical triad of history of episiotomy, tender nodule at the scar site and cyclical pain. Magnetic resonance imaging (MRI) is a very useful imaging modality to diagnose and assess the deeper extension of the lesion. Herein, we report one such case of episiotomy scar endometriosis in the perineum.

Key words: Episiotomy scar endometriosis; perineal scar endometriosis; scar endometriosis

Introduction

Perineal scar endometriosis, at the episiotomy scar site, is a very rare entity occurring in only about 0.03–0.15%.^[1,2] Although the classical clinical triad of this condition which includes, history of episiotomy, tender nodule at the scar site and cyclical pain, is described in the literature, it is seen in only half of the cases.^[1,3,4] Therefore, imaging plays a crucial role in the diagnosis of cases with not only atypical presentation but also for the vital preoperative evaluation.^[3] Magnetic resonance imaging (MRI) is the preferred imaging modality, due to its high contrast resolution. Besides diagnosis, it can also accurately delineate the extent of the disease and its relation to the nearby structures.^[1,3,5] Early diagnosis is vital, as delayed diagnosis can lead to increased morbidity resulting from anal sphincter or rectal involvement.^[3,5] Recurrence and malignant transformation are the other complications of this condition.^[1,5]

Clinical History

A 33-year-old female patient presented with a history of swelling and pain in the perineal region for the past 6 months. She was para 1 and live 1, with previous vaginal delivery 2 years back. Her menstrual cycle was regular for the last 1 year and she mentioned that her pain got worsened around the time of menstruation. There was no history of fever or weight loss. Clinical examination revealed a palpable mass with tender induration at close proximity to the episiotomy scar site. The overlying skin was healthy, without any discharge. Based on the clinical triad of vaginal delivery with visible episiotomy scar, palpable mass at the scar site and cyclical pain, the clinical diagnosis of scar endometriosis was considered.

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Ultrasound (US) of the perineal region with high frequency, linear probe showed complex multi-loculated cystic lesion with low-level internal debris at the scar site [Figure 1]. No intra-lesional vascularity was demonstrated in color doppler study. MRI was performed to assess the deeper extension of the lesion and to exclude involvement of anal sphincter complex.

MRI showed a well-defined multi-loculated lesion of size (2.8 × 2.0 × 3.1 cm) (AP × ML × CC) in the perineal region, on the left side, corresponding to the scar site. It was relatively hyper-intense to muscle on T1-weighted and T2-weighted images [Figures 2 and 3]. The lesion also appeared bright on the T1-fat saturation (FAT-SAT) sequence [Figure 4]. Irregular margin with radiating strands was noted on the post-contrast T1-FAT-SAT image [Figure 5]. However, no definitive enhancement was noted, within the lesion, on subtraction image [Figure 6]. The lesion was confined to the perineum and lower vagina, without disturbing the anal sphincter complex [Figure 7].

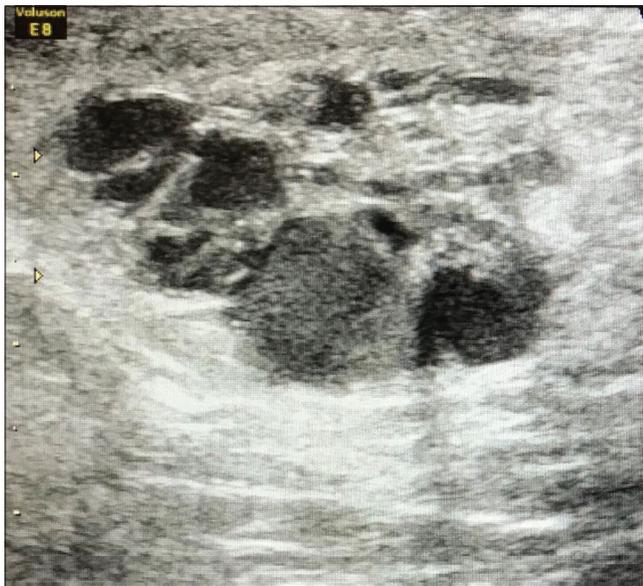


Figure 1: Transperineal ultrasound at scar site shows well-defined multi-loculated complex cystic lesion with internal debris

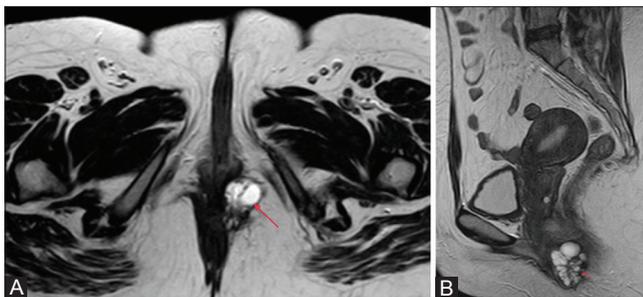


Figure 3 (A and B): (A) Axial T2-weighted image shows a multi-loculated T2 hyperintense lesion with thin dark internal septations (red arrow) in the left paramedian aspect of the perineum. (B) Sagittal T2-weighted image of the same lesion (red arrow)

Pelvic organs such as uterus, bilateral ovaries, rectum, and urinary bladder were normal.

Based on the above imaging features of T1 and T2-hyperintensity, absence of signal loss in fat suppression sequence along with the classical clinical triad of symptoms, the diagnosis of perineal scar endometriosis was made. Surgical excision was advised. A well-circumscribed mass, with a wide 1 cm margin of surrounding normal tissue was removed and sent for histopathology [Figure 8]. Histopathology confirmed the diagnosis of scar endometriosis, characterised by the presence of endometrial glands with stroma and hemosiderin-laden macrophages

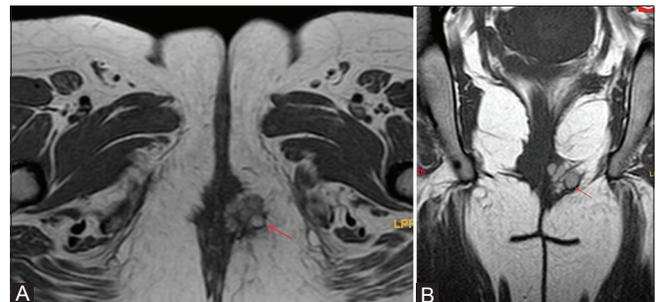


Figure 2 (A and B): (A) Axial T1-weighted image shows a multi-loculated T1 hyperintense lesion (red arrow) in the left paramedian aspect of the perineum. (B) Coronal T1-weighted image shows a multi-loculated T1 hyperintense lesion (red arrow) in the left paramedian aspect of perineum

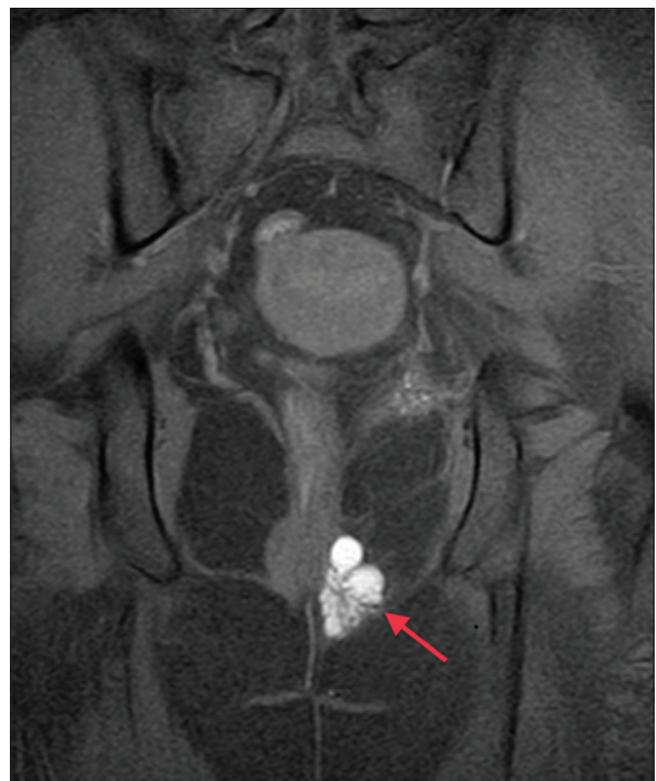


Figure 4: Coronal T1 fat saturation image shows the lesion continues to appear bright without signal suppression (red arrow)



Figure 5: Coronal post-contrast T1 fat saturation image shows the irregular margin and radiating strands from the lesion (red arrow)

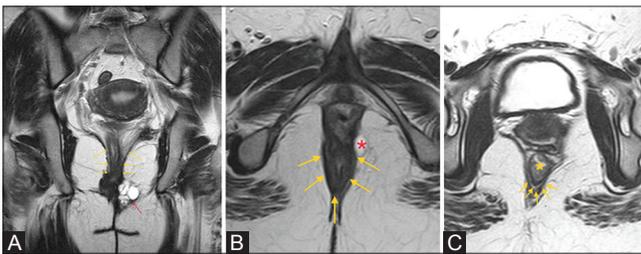


Figure 7 (A-C): (A) Coronal T2-weighted image shows the relation of the lesion (red arrow) to the levator ani and puborectal muscles (yellow arrows). (B) Axial T2-weighted image shows the pubo-rectalis muscle at the level of anorectal junction (yellow arrows) and the superior extent of the lesion (red *). (C) Axial T2-weighted image (at a higher level) shows normal appearance of rectum (yellow *) and the sphincter complex (yellow arrows)

[Figure 9]. Our patient had a follow-up after 6 months and she was normal, without any clinical suspicion of recurrence.

Discussion

Rokitansky described endometriosis, for the first time, almost 150 years back.^[6] There are various theories about its development that include, retrograde menstruation, metaplasia, vascular theory, mechanical transposition and the more recent macrophagic theory.^[6,7] Endometriosis, the presence of ectopic endometrial tissue, can occur in both pelvic and extra-pelvic sites.^[1] The latter is relatively rare and constitutes only about 12% of all cases of endometriosis.^[1]

Extra-pelvic endometriosis at the surgical scar is known as scar endometriosis. It is commonly seen in the caesarean

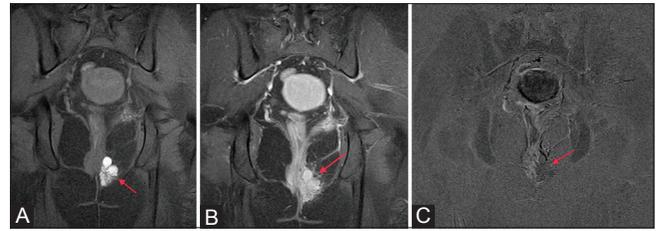


Figure 6 (A-C): (A) Coronal T1 fat saturation image shows the lesion appears bright after fat suppression (red arrow). (B) Coronal post-contrast T1 fat saturation image shows the perilesional enhancement with radiating strands (red arrow). (C) Subtraction image (T1 FAT-SAT post-contrast and T1 FAT-SAT plain) shows no significant enhancement within the lesion

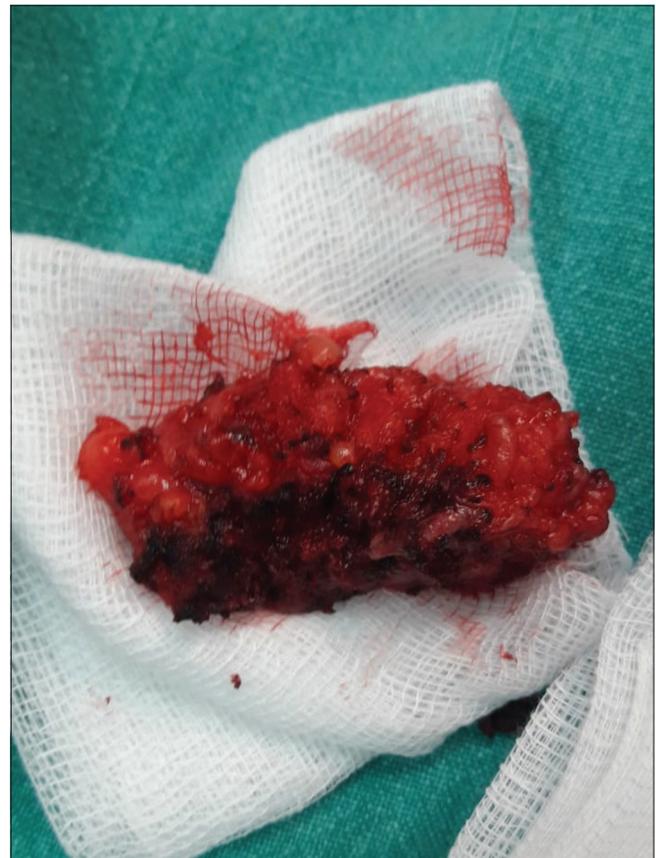


Figure 8: Postoperative picture of the excised lesion

section scar.^[3,8] The possible cause for the development of scar endometriosis is through mechanical transposition that occurs during surgery, as per the iatrogenic implantation theory.^[1,8] This theory is also supported by the experiment done by Ridley and Edward in 1958.^[8]

Perineal scar endometriosis also called episiotomy scar endometriosis, is extremely rare (0.03–0.15%).^[1,2] This condition, however, can result in severe morbidity and prolonged agony in patients, due to damage of the adjacent structures like anal sphincter or rectum at later stages.^[5] Hence, early diagnosis is crucial and MRI plays an important role in diagnosis and preoperative evaluation of the disease.^[1,3]

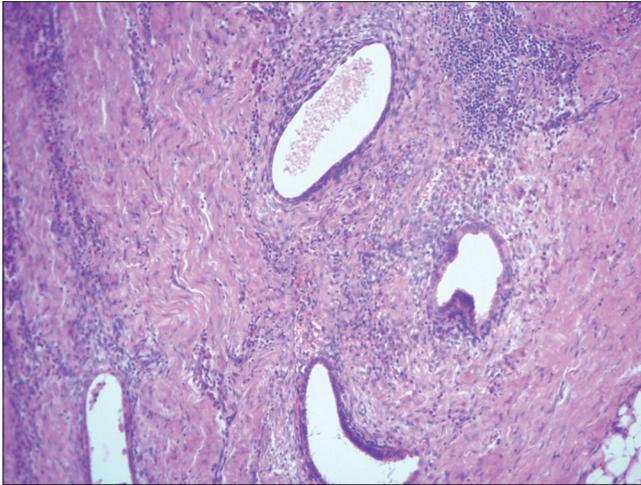


Figure 9: Histopathology image of the lesion showing an endometrial glandular pattern with stroma, confirming the diagnosis of endometriosis. No evidence of malignancy reported in this case

Perineal scar endometriosis can be diagnosed clinically by the presence of the classical triad of symptoms as described in the literature.^[1] However, this classical triad is seen in only 50% of cases and hence, imaging plays a useful role in the diagnosis.^[3] The asymptomatic window period, following surgery, can be more than 8 years for perineal endometriosis.^[8] Associated pelvic endometriosis is seen in about 25% of cases.^[8]

US is often the initial imaging modality for perineal scar endometriosis. A high frequency (7.5 MHz) linear probe is used for the trans-perineal US imaging. The US findings include hypo-echoic or anechoic lesion with fine internal echoes, at the scar site, and peripheral vascularity may be seen.^[5] Trans-rectal US with high-frequency probe (6.5 MHz) can be used to diagnose rectal, recto-vaginal or recto-sigmoid endometriosis but its penetration is poor.^[1] Transvaginal ultrasound and computed tomography (CT) are less useful in the evaluation of perineal scar endometriosis.^[1]

MRI, being a non-invasive imaging modality, is very useful in the diagnosis and preoperative assessment of the local extent of disease.^[1,8] The standard MRI protocol includes T2-weighted fast spin-echo (FSE) sequence (in sagittal, coronal and axial planes), T1-weighted FSE sequence (axial plane) and T1-weighted FSE fat saturation (FAT-SAT) sequence (axial and sagittal planes). The presence of T1 and T2 hyper-intensity and the absence of fat saturation favour the diagnosis of endometriosis.^[1] T1-FAT-SAT sequence is very useful for small lesions (<1 cm) and it can also differentiate hemorrhagic from lipid components.^[1] Saloum *et al.* stated that the addition of MRI contrast has no specific advantage over the non-contrast MRI, except in cases where malignancy is suspected.^[1]

The involvement of anal sphincter is a rare complication of perineal scar endometriosis.^[3] Kamble *et al.* reported

a case of perineal scar endometriosis with anal sphincter involvement and highlighted the role of MRI in the preoperative evaluation of this condition. MRI, due to its high contrast resolution, can identify the extent of the lesion and its relation to the anal sphincter complex.^[6] This helps the surgeon to decide upon the sphincter reconstruction if there is evidence of anal sphincter involvement by the lesion.^[6,7] Accurate delineation of the extent of lesion is very much essential for the complete excision of the lesion and thereby, preventing recurrence.^[1,3,8] Another advantage of MRI is that it can also rule out coexisting pelvic endometriosis, which can be seen in 25% of cases.^[8]

The treatment of choice for perineal scar endometriosis is primarily surgical excision with clear wide margins.^[1,8,9] Recurrence can occur. Hence, complete excision of the lesion is of primary importance.^[1,3,8] Jeyaseelan *et al.*, also stated that the complete excision of the lesion with wide margin is the only way to prevent recurrence. The use of leuprolide, a gonadotropin-releasing analogue, after surgery can also prevent recurrence.^[5,10] Malignant transformation is rare but possible. Therefore, cytology should be performed in all cases.^[1,5,8]

Conclusion

Perineal scar endometriosis is a rare benign condition. A high index of suspicion is needed to diagnose this condition in patients, who present without classical clinical symptoms. Early diagnosis and treatment significantly reduce morbidity. Preoperative MRI is very useful in the diagnosis and to assess its local extent. It can also rule out coexisting endometriosis at other pelvic sites. Postoperative follow-up is essential to rule out recurrence.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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