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Case Report

An episiotomy scar site endometriosis: a rare case report and review of literature

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ABSTRACT

Endometriosis can be defined as the presence and growth of functioning endometrial tissue (glands and or stroma) outside the endometrium. Occasionally, surgical scars are the sites of endometriotic implantation particularly perineal scars (episiotomy). Perineal scar endometriosis is a very rare condition, its incidence rate was about 0.03-0.15%. Perineal scar endometriosis mainly affects young females at the episiotomy site following a previous vaginal delivery. A 29 years old patient presented with swelling at the site of episiotomy which was associated with pain during menstruation. On examination a palpable mass was revealed without induration at close proximity to episiotomy scar site during menstruation. MRI pelvis is the most commonly used imaging technique for the diagnosis of perineal endometriosis as well as to assess the deeper extent of the lesion. Mass excised and sent for histopathological examination. Wide local excision is the definitive treatment for surgical site endometriosis. Follow up is recommended for recurrence. The rare possibility of malignant transformation, can be excluded by histopathological examination of excised tissue.

Keywords: Endometriosis, Surgical scar site endometriosis, Perineal scar endometriosis, Episiotomy scar endometriosis

INTRODUCTION

Endometriosis can be defined as the presence of functioning endometrial tissue (glands and stroma) outside the uterine cavity. Endometriosis more commonly affects reproductive age group women (0.03-1.7%) whereas, 0.7%-44% prevalence rate was observed in general population.[1] Pelvis is the commonest site for endometriosis, especially ovaries, round ligaments and uterosacral ligaments. Extra pelvic sites such as bladder, gastrointestinal tract, lungs and skin also involved in some cases, especially after obstetric and gynaecological interventions. Surgical scar endometriosis is a rare condition and the main cause for it is obstetric and gynaecological surgeries such as hysterectomy and caesarean section in abdominal surgeries whereas in perineum following vaginal delivery.[2] To describe the occurrence of scar endometriosis, several theories have been proposed but most accepted theory is the direct implantation of endometrial tissue in the scars during surgical procedures. Reported incidence rate of Perineal endometriosis is about 0.3-1%.[3]

At the time of delivery, episiotomy is frequently performed. So, one should know about this rare entity and certain measures can be taken to prevent its occurrence. We report a case of episiotomy site endometriosis from a tertiary care institution at Puducherry.

CASE REPORT

A 29 years old female patient of G3 P3L3, came to Gynaecology OPD with complaints of cyclical pain and swelling in the perineum during her menstrual periods for past 4 years, that used to subside with analgesics. Pain in the perineum was localized, non-radiating and more intense just before and immediately after the onset of menstrual bleeding. Patient was asymptomatic during the rest of the month. Her menstrual cycles were regular with average flow which is not associated with dysmenorrhea. Patient also had complaints of dyspareunia. She had 3 vaginal deliveries, her first child was delivered 10 years back with episiotomy, second child delivered 7 years back without episiotomy and her third child was delivered 5 years back without episiotomy. There was no intrauterine manipulation at the time of deliveries. She underwent tubal ligation 4 years back. Her bladder and bowel habits were normal. There was no significant personal and family history of endometriosis.

Her general examination and systemic examination findings were normal, with stable vitals. On local examination, external genitalia were normal. A tender nodular mass of size 3cm×3cm, with well-defined margins, which was firm to hard in consistency was felt at the site of previous left mediolateral episiotomy. Per speculum and per vaginal examination findings were within normal limits. On Per Rectal examination anal sphincter and rectal mucosal were found to be uninvolved.

Based on the clinical trial of cyclical pain, tender modular mass at the episiotomy scar site with history of vaginal delivery, the diagnosis of episiotomy scar site endometriosis was considered provisionally. There was no signs and symptoms suggestive of associated pelvic endometriosis.

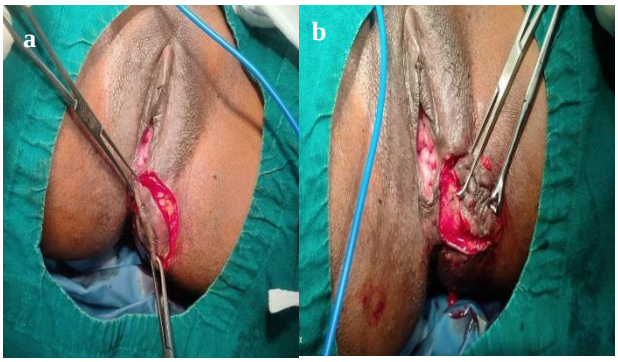

Figure 1 (a and b): Endometriotic nodule on surgical exploration.

MRI pelvis done to rule out anal sphincter complex involvement and also to assess deeper extension. MRI pelvis suggested a well-defined heterogeneous signal intensity lesion of size 2.0cm×2.0cm noted in the left para midline perineal region, about 16mm deeper to the subcutaneous plane and 4mm left towards midline. On T1-weighted and T2-weighted images of MRI pelvis, it was relatively hyper intense to muscle. As per the report of MRI pelvis, endometriotic lesions were confined to the perineum and lower half of vagina, without any involvement of anal sphincter complex. In addition to the above findings, it also revealed Sub septate Uterus and bilateral polycystic ovarian changes with multiple dilated pelvic veins. Pelvic organs such as rectum and urinary bladder were normal. The diagnosis of episiotomy scar site perineal endometriosis was confirmed by classical clinical trial, with hyper intense imaging findings of MRI pelvis.

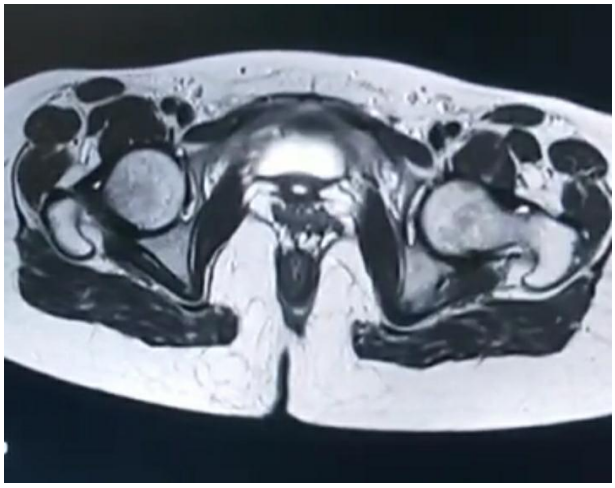

Figure 2: MRI Pelvis - T2 hyperintense images.

Preoperative evaluation was done. Wide local excision was advised and proceeded under spinal anaesthesia. A well circumscribed mass with 2cm margin of surrounding normal tissue was removed to prevent recurrence and sent for histopathological examination. Perineal reconstruction was done in layers. A closed suction drain was kept. Post operatively, tablet Dienogest were given to prevent its recurrence. Postoperative recovery period remained uneventful.

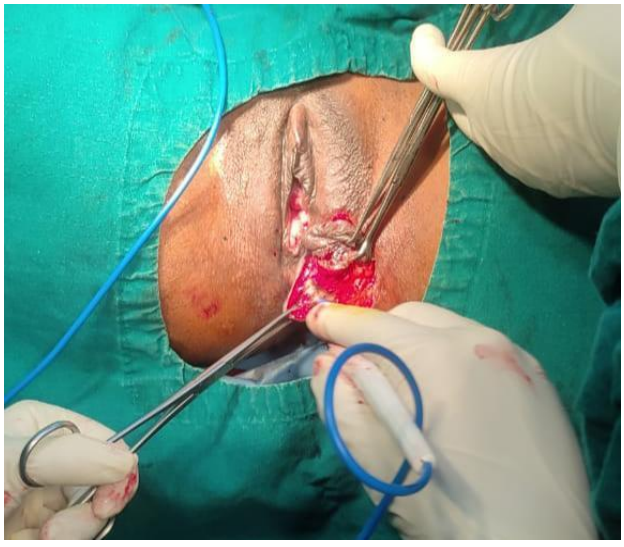

Figure 3: Wide local excision of mass.

Figure 4: Episiotomy site repaired.

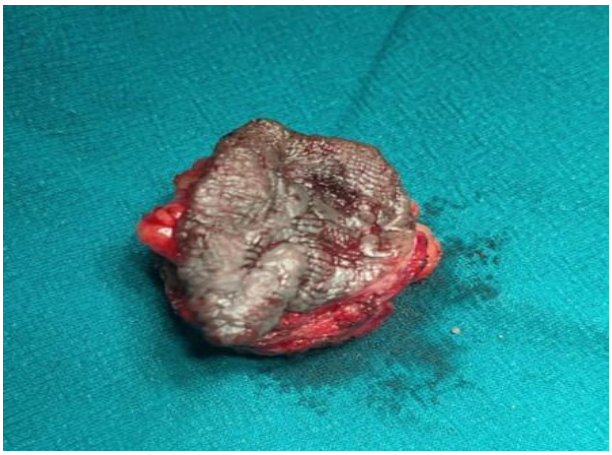

Figure 5: Excised endometriotic mass.

Figure 6: Macroscopic appearance.

Histopathological examination of excised tissue, revealed the presence of endometrial glands and stroma along with areas of hemosiderin and hemosiderin laden macrophages which confirmed the diagnosis of Episiotomy scar endometriosis. Endometrial stroma shows focal evidence of decidualisation. The lesion was found to be extending into focal subcutaneous plane which includes only endometrial stroma.

Our patient had a follow up after 1 month and 3 months interval and she was found to be normal, without any clinical symptoms and signs of recurrence. Informed consent was obtained from the patient before the case report was written.

Figure 7: Follow up after wide local excision. (a) after 1 month, (b) after 2 months and (c) after 3 months.

DISCUSSION

In 1860, Rokitansky first describe endometriosis which was defined as the presence and proliferation of endometrium outside the uterine cavity, commonest site being the pelvis.[4] Allen in 1896, described Extra genital endometriosis whereas Scheckele in1923, reported the first case of Perineal endometriosis.[5] Most of the reported cases of perineal endometriosis in Obstetrics, have occurred following episiotomy. Perineal scar endometriosis also called as Episiotomy scar endometriosis, which is extremely rare. Severe morbidity and prolonged illness were observed in patients due to damage of the adjacent structures like anal sphincter or rectum at later stages.[6]

Proposed etiopathogenetic theories of endometriosis were direct implantation theory, colemic metaplastic theory, haematogenous spread, lymphatic dissemination, macrophagic theory and iatrogenic implantation theory.

After vaginal delivery, in some cases there may be autologous transplantation of viable endometrial cells on episiotomy wounds which attributes to perineal endometriosis. This was supported by Iatrogenic implantation theory as well as Ridley and Edward experiment which was done in 1958.[7] Procedures like

manual uterine exploration and postpartum curettage after vaginal delivery increases the risk of perineal endometriosis.

Classical triad of cyclical pain, perineal mass and previous history of episiotomy scar or perineal tear is sufficient for the diagnosis of perineal scar endometriosis, which was seen in only 50% of cases. Hence, MRI pelvis plays a useful role in the diagnosis. Asymptomatic window period for perineal endometriosis can be more than 8 years following surgery. Differential diagnosis for this entity includes suture granules, abscesses, haematoma, myeloid, lions, sebaceous cysts, desmoids and malignant tumour.[8]

FNAC rules out differential diagnosis by providing a rapid, inexpensive and accurate diagnosis. But some authors stated that it was controversial, because it might result in new endometriotic implants. Imaging modalities like Ultrasound perineum using high frequency probe 6.5MHz and MRI perineum were indicated in case of larger lesions, to rule out anal involvement, to assess the various approaches, surgically and also to elucidate the potential complications of sphincteroplasty to the patient. MRI perineum also rules out coexisting pelvic endometriosis, which can be seen in 25% cases. Due to lack of resolution and radiation hazards, CT scan is rarely advised. Saloum et al, stated that using MRI contrast in suspected cases of malignancy, has more advantage than using non-contrast MRI.[9] Any two of the following three features must be present for the cytological diagnosis of endometriosis- endometrial glands, stromal cells and hemosiderin laden macrophages.

The definitive treatment of choice for surgical site endometriosis is Wide local excision.[10] Surgical site endometriosis usually does not respond to medical management. Hormonal suppression of the lesion using oral contraceptives, pregestational agents and androgenic agents have been tried.[11] But it is partially effective but it helps to prevent recurrence after excision.

Wide local excision of the lesion with 2cm marginal clearance on all sides along with sphincteroplasty can be performed if there is anal sphincter involvement.[12] Incomplete excision of the lesion leads to recurrence. Advantage of surgical excision is to provide a sample for histopathological confirmation of the diagnosis and also to rule out malignancy. Although malignant transformation is very rare, few cases were reported so far. Reported cases of malignant transformation in extra general pelvic was 21.3% and 4% in scars after laparotomy.[13] Early diagnosis and treatment is very imperative, for prevention of progressive involvement of anal sphincter. Hence, the level of suspicion should be high, while diagnosing this rare entity. At the time of vaginal delivery, episiotomy is frequently performed but the incidence rate of perineal endometriosis is rare.[14] The reason for this includes the following; infection and necrosis of the local tissue was caused by the presence of existing bacteria in perineal wound. These circumstances are not appropriate for transplanted endometrial cells to live. Estrogen levels after delivery decreases, which makes difficult circumstances for the growth of transplanted endometrial cells.

Further, the incidence rate of episiotomy scar site endometriosis can be reduced by adopting the following measures such as washing the episiotomy wound with normal saline before suturing, by avoiding manual uterine exploration and postpartum curettage and gloves should be changed before suturing the episiotomy in cases where the uterine cavity was checked and products of conception were removed manually.[15] Suturing materials should not be contaminated with blood and debris from uterine cavity.

Any woman presenting with pain at episiotomy incisional site especially after pelvic surgery, a thorough history and physical examination should always be performed. Also, every surgeon should consider this entity in their differential diagnosis.

Effects of pregnancy on scar site/perineal endometriosis is an area for future research, as progesterone can cause decidual reaction in endometriotic deposits, resulting in remission and resolution of symptoms.

CONCLUSION

Episiotomy scar site endometriosis is extremely a rare clinical diagnosis. When a reproductive age group women presents with painful mass in an episiotomy scar site, high level of suspicion should be kept in mind for diagnosing this rare entity. Best modality of treatment for perineal endometriosis is wide local excision. Before proceeding surgical treatment, anal sphincter involvement should be ruled out because its involvement needs careful evaluation. Morbidity due to progressive involvement of anal sphincter can be prevented by timely diagnosis and treatment. Follow up is essential as recurrence is not uncommon.

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