

Case Report

Endometriomas: Two Case Reports and Review of Literature

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Received: 20/12/2013

Revised: 17/01/2014

Accepted: 30/01/2014

ABSTRACT

Objective: Scar endometriosis may develop after pelvic operations, such as cesarean section, tubal ligation, hysterectomy, hysterotomy or secondary to obstetric or surgical trauma. Perineal endometriosis in episiotomy scar and endometriosis in post hysterectomy abdominal scar endometriosis are quite rare. This prompted us to demonstrate the proper diagnosis; management and prophylactic procedure of scar endometriosis.

Study Design: In these case reports we present females of age 27 and 38 years with complaints of growth at episiotomy site and post hysterectomy abdominal scar since 2 and 1 years respectively. In first case there is previous history of normal delivery 4 years back with episiotomy. The mass was increasing in size with pain and itching over the area during menses. A diagnosis of ?foreign body granuloma, ?endometriosis was done. In second case there was history of abdominal hysterectomy 2 years back. The patient presented with subcutaneous lesion on the anterior abdominal wall. Diagnostic tools used included USG Abdomen, transvaginal and endorectal ultrasonography (USG), magnetic resonance imaging (MRI) and biopsy. The mass was wide-excised together with scars in both cases. The recovery was uneventful with excellent functional and esthetic results. The excised skin covered specimens measured 3.5x3cm and 5x4.5cm. Cut sections showed small cystic spaces. The microscopy revealed endometrial glands and stroma embedded in the fibrous scar tissue.

Results: Six months after operation, women were asymptomatic and there was no evidence of recurrence.

Conclusion: Diagnosis of scar endometriosis should involve detailed history taking and pelvic examination. The role of needle aspiration cytology still remains controversial. Several prophylactic procedures have been presented to prevent decidual contamination of the wound. Complete surgical excision including the adjacent fascia or skin is the proper treatment of scar endometriosis. Increasing awareness of this condition among doctors can help in early diagnosis and treatment with gratifying results

Key words: Endometriomas, episiotomy scar, post hysterectomy scar.

INTRODUCTION

Endometriosis was first described by Rokitansky in 1860 and was defined as the presence of functioning endometrial glands and stroma in an anatomical location other than uterine cavity, which most commonly affects peritoneal surfaces, ovaries and

uterine ligaments. Even it is quite rare, endometriosis may affect the vulva, vagina, rectosigmoid septum or perineal region, generally secondary to obstetric or surgical trauma. The episiotomy scar is a fairly rare site for endometriosis. The actual incidence of abdominal wall endometriosis is

unknown but one series reported that only 6% of these were unrelated to scars. In another series the prevalence of surgically proven endometriosis in scars was 1.6%.^[1] The most common site is cesarean section scar.^[2] But there are case reports of involvement of the rectus abdominus muscle in a virgin abdomen.^[3,4] Endometriosis involving only the rectus muscle and sheath is very rare.^[5] The simultaneous occurrence of pelvic endometriosis with scar endometriosis has been found to be infrequent.^[6] Scar endometriosis is rare and difficult to diagnose, often confused with other surgical conditions. We report two cases of scar endometriosis following episiotomy and hysterectomy which were misdiagnosed as stitch granuloma initially and review relevant literature.

CASE REPORT

In these case reports we present females of age 27 years and 38 years with complaints of growth at episiotomy and abdominal scar sites since 3 years and 1 year respectively. In first case there is previous history of normal delivery 4 years back with episiotomy. The mass was increasing in size

with pain and itching over the area during menses. A diagnosis of? *foreign body granuloma, ?endometrioma* was done. In second case there was history of abdominal hysterectomy 2 years back. The patient presented with subcutaneous lesion on the anterior abdominal wall. Diagnostic tools used included USG abdomen, transvaginal and endorectal ultrasonography (USG), magnetic resonance imaging (MRI) and biopsy. The masses were wide-excised together with the scar in both the cases. The recovery was uneventful with excellent functional and esthetic results. The excised skin specimen in first case was discolored, measured 3.5x3cm (fig1). Cut section grayish-white to yellow and showed small cystic spaces. In second case the specimen excised was skin covered tissue bit measuring 5x4.5cm (fig 3) and cut section showed small cystic spaces. The microscopy of both the cases revealed endometrial glands and stroma embedded in the fibrous scar tissue along with epidermis and dermis (fig 2 & 4). Six months after operation, both the patients were asymptomatic. They were given Danazol 100mg twice daily for initial six months.



Fig 1: Specimen of perineal endometrioma (Case 1).

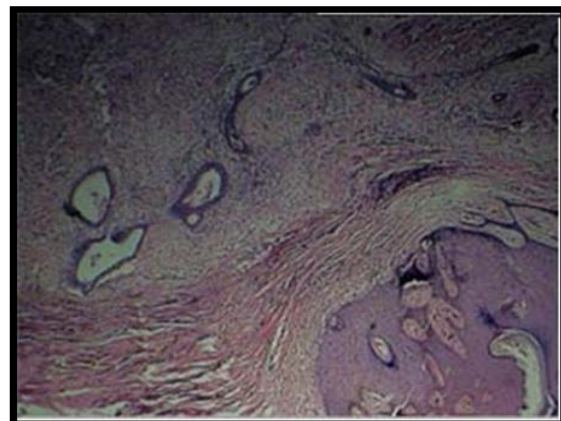


Fig 2: H&E x100. Microscopy: Presence of endometrial glands and stroma in between the fibrous tissue along with epidermis and dermis. (Case 1)



Fig3: Specimen of post Scar endometrioma. (Case 2)

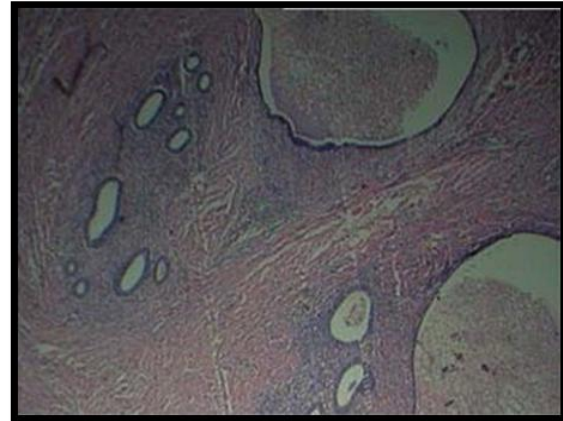


Fig 4: H&E x100. Microscopy: Presence of endometrial glands and stroma in between the fibrous tissue (Case 2).

DISCUSSION

Endometriosis is presence of functioning endometrial tissue outside the uterine cavity, whereas endometrioma is its well-circumscribed mass. The various sites for extra pelvic endometriosis are bladder, kidney, bowel, omentum, lymph nodes, lungs, pleura, extremities, umbilicus, hernia sacs and abdominal wall.^[7] Endometriosis involving the abdominal wall is an unusual phenomenon that should be considered in the differential diagnosis of abdominal wall masses in women.^[5] The usual clinical presentation is a painful nodule in a parous woman with a history of gynecological or obstetrical surgery. The intensity of pain and size of nodule vary with menstrual cycle.

Pathophysiology: The proposed theories^[8] of endometrioma formation are:

- Retrograde spread of collections of endometrial cells during menstruation.
- Blood, lymphatic or iatrogenic spread.
- Metaplasia of the pelvic peritoneal cells.
- Immune system dysfunction and autoantibody formation.

The development of intrapelvic endometriosis may involve retrograde menstruation, maturation of extrauterine

primordial cell remnants of embryogenesis, or hematological or lymphatic spread of endometrial cells. Extrapelvic endometriosis in the lung, skin, and extremities are not associated with surgical violation of the uterus is believed to be the result of hematogenous or lymphatic spread of endometrial tissue.^[9,10] Scar endometriosis are believed to be the result of direct inoculation of the abdominal fascia or subcutaneous tissue with endometrial cells during surgical intervention and subsequently stimulated by estrogen to produce endometriomas. This theory is convincingly demonstrated by experiments in which normal menstrual effluent transplanted to the abdominal wall resulted in subcutaneous endometriosis.^[11] In clinical practice, its occurrence has been well documented in incisions of any type where there has been possible contact with endometrial tissue, including episiotomy, hysterectomy, ectopic pregnancy, laparoscopy, tubal ligation, and cesarean section.^[12-14]

The presence of endometrial tissue can induce metaplasia of the surrounding fascial tissue to form an endometrioma. Alternatively, endometrial cells may reach a cesarean scar via lymphatic or hematogenous routes and subsequently grow into an endometrioma by one of the

mechanisms described above. This may cause rare occurrence of abdominal wall endometrioma without any surgical intervention.^[15] Scar endometriosis most commonly occurs after operation the uterus and tubes. The incidence following hysterectomy is 1.08-2% whereas after cesarean section the incidence is 0.03-0.4%.^[5] The reason for higher incidence after hysterectomy has been given as the early decidua has more pluripotential capabilities and can result in cellular replication producing endometrioma. Time interval between operation and presentation has varied from 3 months to 10 years in different series.^[5] In the present cases it was 3 years and 1 year.

Diagnosis: Scar endometriosis is rare and difficult to diagnose, often misdiagnosed as stitch granuloma, inguinal hernia, lipoma, abscess, cyst,^[16] incisional hernia,^[17] desmoid tumor, sarcoma, lymphoma, or primary and metastatic cancer.^[2] A high index of suspicion is recommended when a woman presents with post-operative abdominal lump.^[18] A good surgical and gynecological history, as well as a thorough examination with appropriate imaging techniques (ultrasound, CT or MRI) will usually lead to a correct diagnosis. The presence of cyclic pain in an incisional mass associated with a cesarean section scar is almost pathognomonic for the condition. Some authors believe that when the diagnosis is made on clinical grounds, no further studies are necessary before wide surgical excision.^[16]

On the contrary, review of the surgical literature indicates that postoperative diagnosis is often incorrect.^[19,20] Hence, whenever the diagnosis is uncertain, efforts should be made to make a preoperative diagnosis with the help of imaging techniques and FNAC. FNAC has been reported to be accurate in diagnosis.^[21] But in a recent report by Dwivedi et al,^[22]

this was not diagnostic in any of the four patients who underwent this procedure. Nonetheless it may be helpful in eliminating malignancy from the diagnosis. Caution should be used if suspicion of incisional hernia is present.^[23] Sonographic and color doppler when combined with clinical data may substantially contribute to the preoperative diagnosis.^[24] These lesions appear as hyperechoic, vascular, and solid, with some cystic changes. CT usually shows a solid, well-circumscribed mass. MRI can be more helpful when the lesion is small because of its high spatial resolution, furthermore it is better than CT scan in detecting the planes between muscles and abdominal subcutaneous tissue.^[25]

Management: Treatment of choice is always total wide excision of the lesion and may sometimes require mesh placement.^[2,4,6,8] Medical treatment with the use of progesterone, 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 has been report of use of gonadotrophin agonist (Leuprolide acetate) but has been found to provide only prompt improvement in symptoms with no change in the lesion size.^[26]

Malignant risk: Malignant change of endometriosis in a cesarean scar (CS) is rare.^[27] Long-standing recurrent scar endometriosis could undergo malignant changes and clinician should be aware. Only 21.3% of cases of malignant transformation of endometriosis occur at extra-gonadal pelvic sites, 4% of cases in scars after laparotomy.^[28] Clear-cell carcinoma is the most common histological sub-type, followed by endometrioid carcinoma.^[29] In the literature, survival rate reaches only 57% only after a short follow-up of 20 months. Treatment is radical surgical resection

followed by prosthetic abdominal wall repair. Compared with endometriosis-associated ovarian carcinoma, the prognosis of abdominal scar complication is poor.^[29]

Follow-up and prevention: These patients need to be followed up because of the chances of recurrence, which require re-excision.^[10] In cases of continual recurrence possibility of malignancy needs to be ruled out. Good technique and proper care during hysterectomy, cesarean section and normal delivery may help in preventing endometriosis. It has been suggested that at the end of surgery especially on uterus and tubes, the abdominal wall wound should be cleaned thoroughly and irrigated vigorously with high jet solution before closure.^[30]

CONCLUSION

General surgeons are infrequently involved in the management of cesarean section scar lesions. The lack of awareness makes the preoperative diagnosis unnoticed. The presence of cyclic pain in an incisional mass associated with a hysterectomy scar, cesarean section scar and episiotomy scar is almost pathognomonic for the condition. When the diagnosis is made on clinical grounds, no further studies are necessary before wide surgical excision. However, imaging techniques, laparoscopy along with FNAC are indicated towards better diagnostic approach. In presence of frequent recurrences, malignancy should be suspected, which carries a poor prognosis.

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How to cite this article: Bembde AS, Manzoor I, Mulay SS. Endometriomas: two case reports and review of literature. *Int J Health Sci Res*. 2014;4(2):207-212.
