Case Report

Scar endometriosis as a complication of surgically treated utero-cutaneous fistula

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ABSTRACT

Extra-pelvic endometriosis is a rare form of endometriosis, however cesarean section scar is a common site for the rare condition. Scar endometriosis can be associated with obstetric or gynecologic surgeries. Utero-cutaneous fistula is an abnormal communication between the endometrium and the skin, this is another rare clinical condition that may complicate cesarean section. We report a case of a 23-year-old woman presenting with features suggestive of scar endometriosis about 20 months after surgical treatment of utero-cutaneous fistula. The utero-cutaneous fistula developed 2 months after cesarean section. This case is presented to highlight scar endometriosis as a complication of surgical management of utero-cutaneous fistula and emphasize the importance of complete surgical excision in the management of both rare conditions.

Key words: Cesarean section; scar endometriosis; surgical treatment; utero-cutaneous fistula.

Introduction

Both scar endometriosis and utero-cutaneous fistula are rare clinical conditions. Endometriosis is the presence of functional endometrial tissue outside of the uterine cavity. Scar endometriosis is a rare form of endometriosis with estimated prevalence of 0.03% to 0.45% of all cases of endometriosis.[1-3] Though the most commonly accepted theory for the pathogenesis of scar endometriosis is the iatrogenic transplantation into the wound during surgery, scar endometriosis has however been reported after pelvic surgeries even when the endometrium was not breached.[1,4,5] Cesarean section is the most common surgery associated with scar endometriosis.[2]

Utero-cutaneous fistula on the other hand is a communication between the endometrial cavity and the skin. It is a rare complication of cesarean section. It is usually associated with postoperative complications and classical cesarean section. Other possible associated risk factors include pelvic abscess, intrauterine device, use of abdominal drain, trauma, pelvic malignancies, and chronic granulomatous pelvic infection such as tuberculosis.[6-8] Thakur *et al*. reported a case following B-lynch suture for the management of post-partum hemorrhage.[9] The relationship between endometriosis and utero-cutaneous fistula is complex; endometriosis is a rare cause of cutaneous fistula and there are reported cases of endometriosis developing within the tract of utero-cutaneous fistula.[5,10]

It may be clinically difficult to distinguish between scar endometriosis and utero- cutaneous fistula as both will usually present with distressing symptoms around the menstrual period. Typically, utero-cutaneous fistula is

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associated with cyclical bleeding, whereas endometriosis presents with cyclical pain and typical dark skin discoloration with or without cyclical bleeding.[2,3,10]

Case Report

Mrs A.A., a 23-year-old $P1^{+1}$ with no living child. She presented with cyclical caesarean section scar pain of 18 months duration with associated hypertrophy and hyper pigmentation of lower 3rd of the scar. She had an emergency caesarean section in a local hospital in her village 2 years prior to presenting to our institution with above complaints. We could not access her medical records hence the details of the operation were scanty. We however established that the indication for the cesarean section was failure to progress in labor and that a midline infra-umbilical incision was used on the anterior abdominal wall but could not determine the type of incision on the uterus. She was delivered of a live male neonate who died shortly after delivery.

She had wound infection and breakdown post operatively, which was initially managed with medication and dressing for 2 weeks. She then had a secondary wound closure in the same facility. The suture was removed 10 days post secondary closure and the wound appeared to have healed. She however subsequently noticed the wound was discharging blood during her menstruation. She presented to a general hospital with the complaint and surgery was done. Though the wound stopped discharging blood during her menses and appeared to have healed, but she thereafter started having cyclical scar pain during her menses with associated dark pigmented nodules. She then presented to our facility with the complaints of scar pain and unsightly appearance of her scar.

Examination revealed a healed mid-line infra-umbilical scar with hyperpigmented nodules in the lower 3 cm of the scar. The diagnosis of scar endometriosis was suspected and skin melanoma was entertained as a possible differential diagnosis. She had a wide excision of the scar. At the operation, her uterus was found to be attached to the anterior abdominal wall through a sinus tract communicating between the endometrium and the diseased portion of the scar. The tract was surrounded by fibrous tissue of about 2 cm in cross-sectional diameter. The scar was excised along with the sinus tract and the surrounding fibrous tissue.

A circumferential incision was made on the normal skin around the scar. The dissection of the scar was commenced in a stepwise manner, starting with normal scar portion above and below the diseased portion. The scar tissue along with the underlying subcutaneous tissue was dissected away from the rectus sheath. The dissection was done circumferentially around the fibrous tissue connecting the diseased part of the scar with the uterus. Subsequently, a longitudinal incision was made on the exposed rectus sheath, starting from the cephalad aspect of the fibrous band, about 8 cm incision was extended towards the umbilicus. The incision was then extended circumferentially around the fibrous tissue thereby freeing it from the rectus sheath attachment. The uterus was then exposed with the dissected scar tissue attached to it through the sinus tract with surrounding fibrous tissue. A circumferential incision was made on the uterus to dissect the tract and fibrous tissue away, thereby leaving about 3 cm defect on the uterus. The defect was closed in 2 layers and anterior abdominal wall was subsequently closed in layers. There was no evidence of endometriotic deposit in any other part of the body.

She was discharged on 5th post operative day and was seen at the clinic 4 weeks later. The wound has healed well and she had menstruated without having scar pain. The histology report confirmed the presence of both scar endometriosis and sinus tract.

Discussion

Mrs A.A presented with scar endometriosis about 18 months following the surgical repair of utero-cutaneous fistula. The utero-cutaneous fistula developed 2 months after a cesarean delivery. The cesarean delivery was complicated by surgical wound infection and wound breakdown necessitating secondary wound closure.

Post cesarean section wound infection is a predisposing factor for utero-cutaneous fistula and is the most likely cause in this index case. The secondary wound closure provides additional possibility of inadvertently including the underlying sub-involuted uterus in the stitch during the closure. The presentation of Mrs A.A at the general hospital where she had surgical treatment for utero-cutaneous fistula was bleeding from the scar during menstruation. This is a typical presentation of utero-cutaneous fistula.[3,4] The surgical treatment of utero-cutaneous fistula can be challenging, therefore it is not unusual to have multiple surgeries as documented by Maddah *et al*.[11] It however appears that the surgery at the general hospital effectively treated the utero-cutaneous fistula because the cyclical bleeding from the scar was resolved following the surgery. The possibility therefore exits that either iatrogenic transposition of endometrium during the surgical treatment of her utero-cutaneous fistula or migration from the transected fistulous tract was responsible for the scar endometriosis.

The diagnosis of endometriosis can be suspected clinically but requires the histological evaluation of excised tissue for confirmation.[1,2] Investigations, such as hystersalpingogram and fistulograpghy to confirm utero-cutaneous fistula were not done because the history was not suggestive. On the other hand, computer tomography (CT) scan and magnetic resonance imaging (MRI) that could have helped in determining the depth of tissue involvement in endometriosis and also utero-cutaneous fistula were not available in our facility.

Both scar endometriosis and utero-cutaneous fistula can be managed with medication, but surgical excision is the preferred management modality.[12] Surgery requires complete excision to prevent recurrence. We had factored in the possibility of migration from the transected fistulous tract as the cause of the scar endometriosis, hence planned to completely excise both the scar endometriosis and the suspected remnant of the fistulous tract. Offiong *et al*.[13] cannulated the fistula during surgery to guide dissection, this was not possible in this case because there was no fistulous opening on the skin, hence we had to rely on tissue planes and tissue texture to guide our dissection. Hysterectomy was not considered for this patient because she is desirous of pregnancy. She had complete resolution of her symptoms confirming we achieved complete excision of both the scar endometriosis and the remnant fistulous tract.

Conclusion

Incomplete excision of a fistulous tract should always be entertained in a woman who presents with the symptoms of scar endometriosis following the surgical treatment of utero-cutaneous fistula. Meticulous attention to tissue plane and tissue texture is of significant benefit towards complete excision.

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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