



Menstruating Skin: Utero-Cutaneous Fistula Coexisting with Scar Endometriosis: A Case Report

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Abstract

Rationale: Both scar endometriosis and utero-cutaneous fistula complicating caesarean section is a rare occurrence, only a few cases have been reported in the literature.

Patient Concern: She developed cyclical bleeding from the previous operation site associated with abdominal pain. The bleeding occurred whenever she was having her menstrual period.

Diagnosis: An initial diagnosis of scar endometriosis was made.

Intervention: She had surgical excision of the fibrotic subcutaneous tissue and rectus sheath with the repair of the fistulous uterus. The histopathological examination was scar endometriosis and utero-cutaneous fistula Outcome: She did well post-surgery and was discharged home and the follow-up clinic visits were not eventful.

Lessons: Caesarean section wound infection is a risk factor for utero-cutaneous fistula formation and scar endometriosis. Meticulous excision of the endometriotic skin, the fistulous tract, and repair of the fistulous uterus is an effective modality of management.

Keywords: Caesarean section; Scar endometriosis; Utero-cutaneous fistula

Introduction

Scar endometriosis and utero-cutaneous fistula are uncommon and rare clinical presentations encountered in the gynecology clinic. Endometriosis is often seen as functional endometrial tissue outside the uterine cavity while utero-cutaneous fistula on the other hand is a communication between the endometrial cavity and the skin [1,2]. It is rare to have both presenting at the same time in a patient. Such rare presentation is usually associated with postoperative abdominopelvic surgeries especially caesarean section in women. Other possible risk factors include pelvic abscess, intrauterine contraceptive device, use of abdominal drain, trauma, pelvic malignancies, and pelvic infections like tuberculosis [2,3].

The relationship between endometriosis and utero-cutaneous fistula is complex; endometriosis can complicate utero-cutaneous fistula and there are reported cases of endometriosis developing within the tract of utero-cutaneous fistula [4,5].

Here we report a case of scar endometriosis coexisting with utero-cutaneous fistula in a 31-year-old woman post-caesarean section.

Case Presentation

Mrs. KA, is a 31 years old para 4+1, 3 alive women, whose last menstrual period was 31/01/2023. She presented at the gynecological clinic on 23/02/2023 on account of 18 month's history of cyclical bleeding from the previous operation site associated with abdominal pain. She had a history of emergency caesarean section two and half years ago in an outside facility on account of intrauterine fetal death in a transverse lie at term. Even-though, we could not assess her medical record and the details of the surgery done at the referral center, she gave a detailed account of the events of the postoperative period. There was complication of the wound site by infection and subsequent breakdown of the wound. She was placed on antibiotics and the wound was dressed for 12 days. After which secondary wound closure was performed; the removal of the suture was done 10 days post-secondary closure and the wound appeared to have healed.

Six months after the delivery she began to feel pain at the left edge of the operation site. It

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Figure 1: Showing the uterus with fistulous tract separated from the subcutaneous tissue.

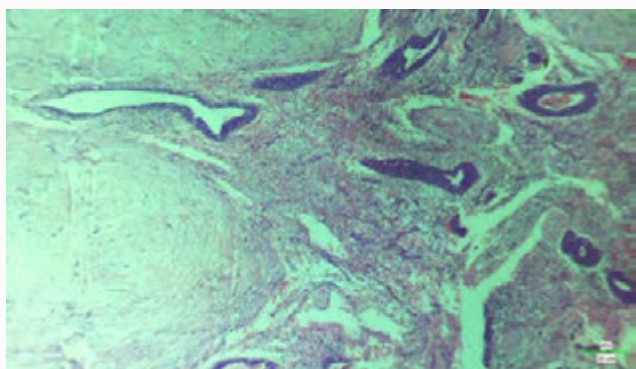


Figure 2: Showing endometrial tissue in the scar tissue of the skin.

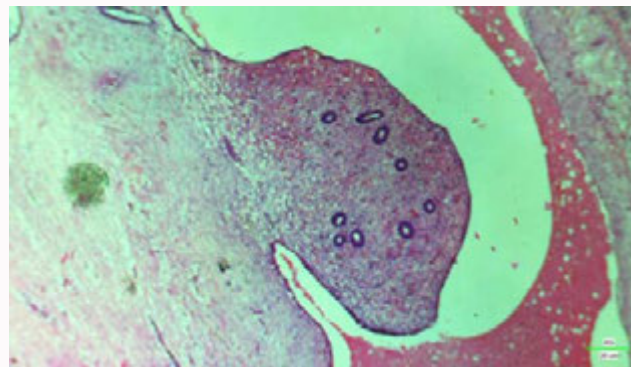


Figure 3: Showing endometrial tissue within the utero-cutaneous fistula.



Figure 4: Dissected gross specimen of scar tissue.

was persistent and associated with darkening and swelling of the skin around the area. Two months after the onset of the pain she observed bleeding from the same area, the bleeding occurred during her menstruation with an increase in the severity of the pain at the site. After several complaints and no resolution of the problem, she was referred to our facility.

At presentation, she complained of cyclical bleeding from the operation scar; examination revealed an old poorly healed Pfannenstiell scar with hyperpigmentation of the skin surrounding the scar. There was a hyperpigmented nodule at the left edge with a sinus at the center of 1 cm diameter. There was a firm, tender, and undefined mass palpable below the nodule, which appeared to be within the anterior abdominal wall. A provisional diagnosis of scar endometriosis was made given the patient's history of bleeding from the incision site and pain with menstrual periods. She was planned for excision of the scar. The pelvic ultrasound scan and fine needle aspiration cytology results were unremarkable. The blood profile tests were essentially normal.

At the operation, an elliptical incision was made around the old scar ensuring that the normal skin margins were created, and stepwise dissection of the scar tissue was carried out starting with the normal scar portion from the right edge. She had a wide excision of

the endometriotic mass, the findings included fibrotic subcutaneous tissue and rectus sheath with discharge of a minimal amount of brownish exudate. A fistulous tract connecting the diseased portion of the scar on the anterior abdominal wall and the endometrial cavity in the uterus was seen and severed.

The uterus was thereafter exposed with the fistulous tract (Figure 1); the fistula was repaired and the anterior abdominal wall closed. No gross evidence of endometriotic seedlings in any other part of the body is seen.

She was discharged home on the 5th day post-surgery on antibiotics and combined oral contraceptives for three cycles. Her first three visits on follow-up at the gynecology clinic at an interval of two weeks showed that the wound had healed well and she had no complaint of abdominal pain; the subsequent visits were also uneventful.

At the histopathology lab we received formalin-fixed five pieces of fibrous tissue, one of which is skin bearing. It aggregates to 8 cm × 6 cm × 3 cm in dimension. The skin-bearing tissue is elliptical in shape and has a length of 9 cm and a dimple of 1 cm in diameter on the skin (Figure 4). The cut surface of the skin tissue is grey and solid.

Histopathologic finding of the sample was that of skin tissue beneath the epidermis with infiltration in places by endometrial glands and stromal, the glands are cystically dilated and contain pale secretion (Figure 2). Also seen is a dilated fistula located within the subcutaneous layer with endometrial tissue infiltrating a polypoid growth on the wall of the tract. Also seen within the tract is fresh hemorrhage (Figure 3).

Discussion

Scar endometriosis is a rare form of endometriosis with an estimated prevalence of 0.03% to 0.45% of all cases of endometriosis

[1]. 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 [6,7]. Caesarean section is the most common surgery associated with scar endometriosis, and the diagnosis is made several months or up to several years following the procedure [8]. The clinical appearance varies with the depth and localization of the lesion. The symptomatology is not always present, and the diagnosis is difficult. However, the most frequent complaint is cyclical pain and a positive history of surgery may be a clue for the correct diagnosis, as seen in this case [8,9].

On the other hand, Utero-Cutaneous Fistula (UCF) which is also a rare entity, is clinically difficult to differentiate from scar endometriosis. Both will usually present with distressing symptoms around the menstrual period. Typically, utero-cutaneous fistula is associated with cyclical bleeding, whereas scar endometriosis presents with cyclical pain and typical dark skin discoloration with or without cyclical bleeding [3,10-12]. It was when we saw the fistulous tract on the uterus (Figure 1) at surgery and after the result of the histopathological examination, that we could establish the coexistence of both utero-cutaneous fistula and scar endometriosis in this patient.

Scar endometriosis usually affects women in their reproductive age (mean age 30-40 years). It is also hormone-dependent and its clinical presentation starts with a pigmented or skin-colored papule or nodule with an average diameter of 2 cm and their persistence ranges from 2 months to 2 years. In addition, the average period to the onset of symptoms is between 3 months to 18 years [1,3,13]. Mrs. KA, at 31 years of age, was within the reproductive age; the site of the lesion was at the left edge of the caesarean section scar and the nodule was 1 cm in diameter; furthermore, the lesion appeared 6 months after surgery as a subcutaneous nodule. She also experienced bleeding or monthly swelling of the nodule thus, raising our index of suspicion for scar endometriosis.

It has been reported that Utero-Cutaneous Fistula (UCF) does not appear in the immediate postoperative period but requires some days in order to create a connection between the skin and the uterine cavity [8]. Post-surgical infections are often responsible for it. Other possible causes include placenta remnant, repeated previous abdominal operations, long-term stay of drains, incomplete closure of uterine incision, and wound dehiscence. Additional causes of UCF include Intrauterine Contraceptive Device (IUCD) migration or infection by actinomycetes [14-16]. The index case could have been due to the infection and dehiscence of the wound, thus creating direct contact between the uterus and the abdominal wall. In addition, secondary wound closure would have provided the possibility of inadvertently including the underlying sub-involuting uterus in the stitch during the process [17].

We know that surgical treatment of utero-cutaneous fistula can be challenging, therefore it is not unusual to have multiple surgeries as documented by Maddah et al. [14]. However, the surgery she had in our facility effectively treated the utero-cutaneous fistula, as her symptoms resolved and there was no further complaint.

Seyhan et al. had previously described a non-surgical method involving the use of Gonadotropin-Releasing Hormone agonist (GnRHa) [18]. It works by suppressing menstruation and rendering the endometrial-like lining of the fistulous tract atrophic, with

resultant spontaneous closure of the fistula. However, medical treatment is not readily available, because we are in an underserved area.

Even-though, both scar endometriosis and utero-cutaneous fistula when they occur together in the same patient can be managed with medication, surgical excision is the preferred management [19]. Investigations, such as hysterosalpingogram, Fistulography, Computerized Tomography (CT) scan, and Magnetic Resonance Imaging (MRI) to confirm utero-cutaneous fistula were not done due to lack of availability locally and serious financial constraints.

Surgery requires complete excision to prevent recurrence. We had considered the possibility of migration from the fistulous tract as the cause of the scar endometriosis (Figure 3), hence the decision to completely excise both the scar endometriosis and the suspected fistulous tract. Offiong et al. [20] cannulated the fistula during surgery to guide dissection; thus, showing another surgical procedure. From our experience, we carefully dissected, relying on tissue planes and texture in the process.

There is no evidence-based treatment modality currently available, the mainstay is still surgery. Total and subtotal abdominal hysterectomy has been performed in patients who have completed their families [5,15]. Hysterectomy was not considered for this patient because she is desirous of pregnancy.

Conclusion

This case report highlights the very rare possibility of scar endometriosis and utero-cutaneous fistula occurring together in a woman a few months after cesarean section. While there is no evidence-based treatment schedule, we advocate surgical excision of the scar and repair of the fistulous tract. A hysterectomy may be useful if the patient has completed her family. Further research is necessary to explain the mechanisms of this condition.

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