Case Report

Diagnostic Challenges and Surgical Management of Non-Healing Uterocutaneous Fistula: A Case Report

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ABSTRACT

Uterocutaneous fistula (UCF) is a rare condition, with only a few case reports mentioned in the literature. This report describes the management of a 30-year-old female who presented with bloodstained purulent discharge from a previous Pfannenstiel incision after cesarean section. The patient had developed a fistulous communication between the uterus and the anterior abdominal wall, and the discharge from the wound site increased during menstruation. This condition was refractory to medical management. Radiological investigations, including a fistulogram, showed a blind-ended tract extending from the skin into the subcutaneous tissue. A laparotomy was performed in which a uterocutaneous fistula was identified and managed surgically.

Keywords: Uterocutaneous fistula; Cesarean section; Diagnostic imaging; Surgery

INTRODUCTION

Uterocutaneous fistula (UCF) is an uncommon, underreported, and often underdiagnosed condition that occurs due to abnormal communication between the uterus and skin.[1] UCF generally arises as a complication of cesarean section, often occurring due to faulty suturing technique, post-operative wound infection, or the use of drains.[2] This condition can present with symptoms such as persistent non-healing wounds, purulent discharge, or cyclical bleeding from a previous surgical incision during menstruation.[3, 4]

Previous reports have mentioned various treatment options for UCF, ranging from conservative management with antibiotic therapy and hormonal therapy to surgical repair of the fistulous tract, which is usually reserved for cases that fail to resolve with conservative measures.[5,6] There are currently no established guidelines for its diagnosis and management. In this report, a case of a 30-year-old woman is presented who developed UCF following multiple cesarean sections. This report highlights the diagnostic challenges and management strategies of this rare complication of multiple cesarean sections.

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CASE REPORT

In July 2024, a 30-year-old female, para 4+0, presented to the surgical outpatient department with infected discharge and menstruating blood from a previous Pfannenstiel incision. She had a history of previous three cesarean sections, with her last lower segment cesarean section performed six months ago at a tertiary care hospital. Following the cesarean section, the patient was discharged on her second post-operative day. After one week of surgery, her wound gaped from the right side along with a tender swelling, with clinical findings of a hematoma present on the left side of her stitch line. Stitches were removed, and she was advised oral antibiotics and anti-inflammatory drugs. She started noticing blood-stained discharge from the left side of the wound after the tenth post-operative day. The patient went to a private healthcare facility in her vicinity, where incision and drainage of the wound were performed. The wound did not heal, and she had a persistent blood-stained, purulent discharge from the left side of the wound. She was advised oral norethisterone and tranexamic acid for cessation of bleeding by a gynecologist. The bleeding remained controlled for a period of six months, following which the patient showed poor compliance with the medication, which resulted in re-bleeding from the wound.

In July 2024, debridement of her wound was performed under local anesthesia at a tertiary care hospital, and the patient was discharged on oral

antibiotics. After two weeks, the patient presented to our outpatient department with persistent discharge from the wound site. On examination, a 3 x 5 cm wound with purulent discharge and marked hyperemia was present, which corresponded to Southampton grade V (Figure 1). The patient was admitted to the surgical ward, and relevant investigations were ordered. Her ultrasound and computerized tomography (CT) scan of the abdomen and pelvis with contrast did not show any internal communication of the wound. The CT scan showed an anteverted uterus measuring 10.7×4.9×7cm with central endometrium and bilateral clear adnexa. A fistulogram with water-soluble contrast was performed, which highlighted a blind ending curvilinear sinus tract measuring approximately 1.7 cm with distal pooling of contrast noted in the subcutaneous plane in the anterior abdominal wall in the vicinity of the cesarean section scar. No branching, ramification of tract, or fistulous communication with rectus muscles or gut wall was noted. The case was discussed with the radiology department in the intradepartmental clinical meeting. On clinical suspicion of a possible fistulous communication, exploration of the tract was planned. As the patient had a history of menstrual bleeding through the tract, advice from the gynaecology department was sought, and consent for hysterectomy was taken from the patient in case communication with the uterine cavity was encountered.

The patient was operated on under general anesthesia. Peroperatively, Gentian violet was injected to delineate the tract. A skin incision was made along the previous scar line. A fistulous tract of about 4 cm extending from the cutaneous plane to the anterior lower uterine wall was identified (Figure 2). The tract was excised, and a hysterectomy was performed by ligating the bilateral tubo-ovarian ligaments, the round ligament, the uterine vessels, and the transverse cervical and uterosacral ligaments. Uterus and cervix were removed, and the vaginal vault was closed with an absorbable Polyglactin 2-0 suture. An intra-abdominal drain was placed, and primary closure of the abdomen was done. The histopathology report showed dense fibrosis, chronic inflammatory changes, and a collection of foreign-body giant cells. The tract was lined by skin. The patient was discharged the following day and was followed closely for three months. At her third-month follow-up, her wound had completely healed (Figure 3).

Figure 1: Pre-operative condition of the wound showing marked hyperemia and purulent discharge

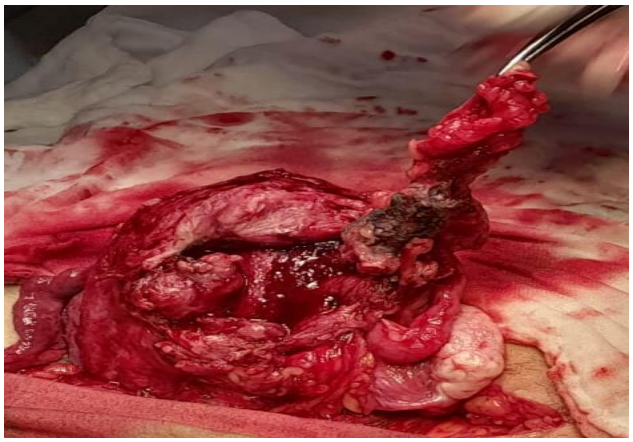

Figure 2: Per-operative image showing exploration of the fistulous tract delineated by methylene blue dye. The pointer shows the external opening of the tract.

Figure 3: Post-operative wound after 3 months of excision of the Uterocutaneous fistula

DISCUSSION

UCF is a rare condition, and the etiological causes and mechanisms underlying its pathogenesis are still not completely known. This report highlights the etiology, symptoms, diagnosis, and treatment options of this rare condition. The development of UCF has been linked to various causes. Most of the case reports have mentioned repeated cesarean section as a cause.[4,7,8] Wang et al. have reported a case of UCF after abdominal hysterectomy.[9]

Deborah et al. reported diagnostic hysteroscopy as a cause of UCF.[10] Other causes include use of non-absorbable sutures during cesarean section, necrosis of the uterine wall due to excessive suturing, wound infection, scar endometriosis, retained placental connective tissue, use of drains, hysterotomy, and incomplete hysterorrhaphy.[5, 11, 12]

The UCF in the present case was not identified on ultrasound and CT-scan of the abdomen and pelvis with contrast. A fistulogram done with water-soluble contrast showed a blind ending tract within the subcutaneous tissue in the vicinity of the wound, but no communication with the uterus was identified. The final diagnosis of UCF was only made intra-operatively after carefully following the tract. This highlights the diagnostic challenges that can make the diagnosis of UCF extremely difficult. Therefore, early identification of the characteristic symptoms of UCF becomes important. These symptoms include a non-healing wound after cesarean section, cyclical bleeding during menstruation, and discharge of pus or serosanguinous fluid.[7] Although in the present case, the imaging modalities offered little help, they nevertheless have a significant role in the diagnosis of UCF. The imaging modalities helpful in this regard are CT scan or MRI of the pelvis, hysterosalpingography, and hysteroscopy, while ultrasound generally has a very limited role in the diagnosis of USF.[2,13] The injection of methylene blue through the cervix is very helpful when the diagnosis is not clear, as it can show the spillage of dye from the wound in case of a patent fistulous tract.[14,15]

In the absence of a standard management plan, the treatment of UCF depends on multiple factors such as underlying cause, family planning concerns, and the personal preferences of individual patients.[10] In this case report, surgical management included hysterectomy, taking into account a history of long-standing refractory disease and the patient's decision of having achieved family completion. Literature shows that UCF can be managed conservatively, particularly in young patients where the diagnosis is made earlier.[9,16] Conservative management includes medical treatment with antibiotics and hormonal therapy or conservative surgery with excision of the fistulous tract without performing a hysterectomy.[17] UCFs can be treated with a gonadotropin-releasing hormone agonist that suppresses menstruation and causes healing of the tract. These hormones can be used in isolation or as an adjunct to surgical treatment.[18] In this case report, failure of conservative management was attributed to a deep fistula communicating with the uterine cavity and poor patient compliance with antibiotic treatment and hormonal therapy.

Surgical resection and repair of the fistula appears to be the treatment of choice in larger fistulae or those not responding to medical management.[19] Surgical treatment ranges from wide local excision to hysterectomy, depending on the extent, duration, and refractive nature of the condition.[20] In previous studies by Jadib et al. and Wang et al., patients of UCF have been successfully managed without hysterectomy, thus highlighting the effectiveness of fertility-sparing surgery.[9, 17] However, the fistulous tract excision in this case report was followed by hysterectomy as the disease process was longstanding and refractory to medication and local management of the wound. This approach is supported by the literature evidence. Anderson et al performed a hysterectomy with bilateral salpingo-oophorectomy for a UCF that developed after a uterine leiomyoma.[21] Thakur et al. performed a hysterectomy in UCF with necrosis of the uterine wall to prevent bleeding and sepsis.[22] Similarly, in the present scenario, late diagnosis, failure of medical management, and extent of disease were considered as ample evidence to support the adoption of a more aggressive approach. Had an earlier diagnosis been made, a more suitable approach would have been adopted in the initial stages of management in this case. Thus, the authors recommend that, in the absence of standard guidelines for management of UCF, the surgical team should determine the ideal treatment of UCF based on the extent and duration of disease, underlying cause, and the patient's preferences. The preexisting literature is more in favor of conservative management of UCF. Nevertheless, this case report points out that surgical intervention is valid and effective in cases of failure of conservative management. Also, this case highlights that UCF may not necessarily be noticeable during pre-operative radiography, including CT scan or fistulogram, and may only be detected peroperatively. This stresses the need to have a high index of suspicion and the readiness to have a definitive surgical response even after negative imaging outcomes.

CONCLUSION

UCF remains a rare but significant complication following cesarean section. Early suspicion and correct imaging can prompt fertility-sparing treatment. Surgical management remains the definitive treatment with a focus on excising the fistulous tract and addressing the underlying etiology. This case also highlights the importance of meticulous surgical techniques during cesarean section to avoid complications such as UCF.

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reviewed and approved the final manuscript and agree to be accountable for all aspects of the work.

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