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## CASE REPORT

# Uterocutaneous Fistula Complicating Caesarean Section: A Case Report

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## Summary

A uterocutaneous fistula (UCF) is a rare clinical presentation following Caesarean section (CS) and other pelvic surgeries. Presently, there are limited reports on the clinical management of UCF. This report describes a 35-year-old woman who presented with a four-year history of abdominal pain and cyclical bleeding from the skin after CS. Fistulography was carried out, and a diagnosis of UFC was made. She had a fistulectomy with the fistula tract excised, followed by debridement of the necrotic tissue and repair of the uterus. At the third-month postoperative follow-up, there was no recurrence of symptoms. A UCF is a rare condition that needs detailed investigation and timely medical and surgical management.

**Key words:** *Caesarean section, Cyclical bleeding, Fistulectomy, Fistulography, Uterocutaneous fistula.*

## Introduction

Uterocutaneous fistula (UCF) is an abnormal communication between the uterus and the skin. Commonly, uterine fistulae are known to occur between the uterus and the bowel or between the uterus and the urinary bladder due to postoperative injuries or infectious conditions. However, UFC is a rare entity [1,2]; hence, its exact incidence is unknown because of this pathology's rarity. Indeed, only 20 cases were reported in the literature over the last decade. [3, 4]

This unusual complication can follow surgeries such as caesarean sections (CS), hysterotomy and myomectomy. [5] Other causes that may result in this pathology include endometriosis, chronic infection, malignancies with local invasion to the adjacent organs, and the incomplete closure of wounds, particularly on

the uterine wall. [1,3] The clinical presentation is usually characterized by cyclical bleeding from the abnormal opening along the scar site on the abdomen. [3,6] A high index of suspicion is required to make a diagnosis, and this can be confirmed using imaging modalities such as hysterosalpingography (HSG), fistulogram, magnetic resonance imaging (MRI) of the pelvis, hysteroscopy, and computed tomography scan (CT scan) with intravenous contrast. Fistulous tract demonstration is necessary for definitive diagnosis; hence, ultrasonography is said to have a limited role in diagnosing UFC.[6]

The global rise in the CS rate has necessitated the need to create awareness about this rare complication of CS. Due to a lack of information on this complication of surgery, the most appropriate treatment remains unclear. This report is to discuss our experience in the management of this condition.

## Case description

A 35-year-old para 3<sup>+0</sup> woman with two living children presented to our Gynaecological clinic with a history of cyclical oozing of blood from her abdominal scar of four years duration. The cyclical bleeding coincided with her menstrual periods. The symptoms were noticed after undergoing emergency CS for preterm prelabour rupture of membranes (PPROM) at approximately 31 weeks of gestation four years ago. The baby died on the seventh day of life. She was transfused with two units of blood, and there was a history of prolonged wound care. She initially noticed pain along the Pfannenstiel scar, followed by swelling a few weeks after surgery. About two months after surgery, she noticed bleeding along the

operation site, which had continued until presentation. At presentation, bleeding from the operation site occurred during her monthly menstrual flow from three different points and stopped when the menstrual flow ceased. In between her menses, she did not experience bleeding from this site. She had had two previous vaginal deliveries (2007 and 2011) and was still desirous of pregnancy.

General physical examination was unremarkable, and vital signs were stable. There was a Pfannenstiel scar on abdominal examination, two swellings about 1 cm × 2 cm above the Pfannenstiel scar and a 1 cm × 1 cm swelling along the scar (Figure 1). Vaginal speculum examination and bimanual examination were not remarkable.



Figure 1: Two swellings about 1 cm × 2 cm above the Pfannenstiel scar and a 1 cm × 1 cm swelling along the scar

The full blood count and urinalysis parameters were normal. Abdominal ultrasound showed no abnormalities. Fistulogram revealed a

fistulous tract between the uterus and abdominal wall (Figure 2).



Figure 2: Fistulogram with a fistulous tract between the uterus and abdominal wall

A diagnosis of UFC was made based on the clinical findings and fistulogram report. Following counselling, she consented to exploratory laparotomy for excision of the fistulous tract and repair of the fistulous uterus. Intra-operatively, there was a thick fibrotic

fistulous tract extending from beneath the scar, involving the rectus sheath and peritoneum with subsequent attachment to the body of the uterus anteriorly. The tubes and ovaries were normal. The opening on the uterine body measured 2 cm x 2 cm (Figure 3).

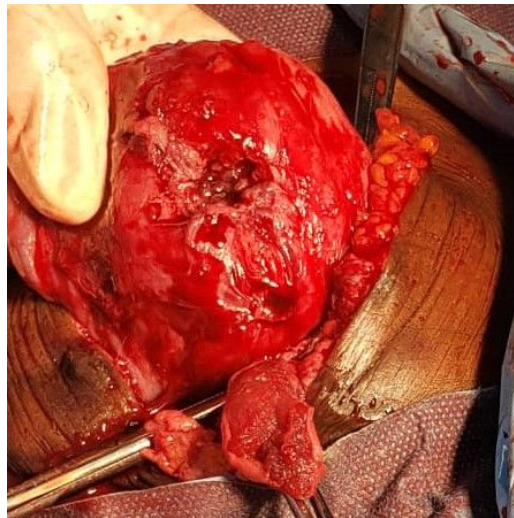


Figure 3: A 2 cm x 2 cm hole on the uterine wall

The fistulous tract was excised completely. The edges of the opening in the uterine wall were trimmed, and the uterine wall was repaired in two layers. Histopathologic report on the excised tissue showed a fistula tract lined with benign stratified squamous epithelium superficially and granulation tissue with no evidence of endometriosis. The postoperative course was uneventful. She was discharged on the seventh postoperative day. Upon review six and twelve weeks later, she was asymptomatic and menstruating normally without skin discharge. The abdominal scar was intact. She was advised to delay the subsequent pregnancy for another six months after surgery and register early for antenatal care in a specialized unit in all subsequent pregnancies. She was also counselled to have CS deliveries for subsequent pregnancies.

*Informed consent:* Consent was obtained from the patient to publish this case and accompanying images.

## Discussion

Uterocutaneous fistula is a rare gynaecological complication known to occur after CS, myomectomy, postoperative infection, postpartum sepsis, and uterine procedures. [3,7] Most UFC have been reported to originate from infective processes that disrupt the continuity of tissues. Poor surgical technique has also been suggested to contribute to the development of the UFC. [6,8] Both factors can be suspected in the index patient because of the history of PPROM and the need for blood transfusion postoperatively.

Almost always, UFC could be suspected in the presence of a history of abdominal pain and or bloody discharge from the skin during menstruation in a patient with a history of CS or myomectomy. [6,9] These were noticed in the index patient a few weeks after surgery. Uterocutaneous fistula may be confused with scar endometriosis; hence, a demonstration of the fistulous tract is necessary to make a

definitive diagnosis. [8] Unfortunately, at the moment, no gold standard has been adopted in the diagnosis of UFC. [5,9] However, imaging techniques such as fistulogram, HSG, CT or MRI could be explored. [9] Contrast CT scan and MRI could have demonstrated the tract, but they were not available in our centre and most centres in Nigeria. [8] These imaging techniques are also costly. Hence, they are out of reach of most patients. Therefore, the diagnosis of UFC in the index patient was made based on a high index of suspicion, and it was confirmed with a fistulogram which demonstrated the pathological pathway between the skin and the uterine cavity.

Due to the rarity of this condition, there is, at present, no standard treatment guideline. However, authors have suggested different treatment options, ranging from conservative treatment to surgeries or a combination of the two. [6, 7, 10] The types of surgeries varied from the excision of the fistula tract to hysterectomy, which presents a challenge in young patients still desirous of pregnancy. [7, 10] Conservative treatment with the use of gonadotropin-releasing hormone (GnRH) agonists have been reported to be successful in the treatment of UFC by inducing amenorrhea resulting in cessation of menstrual flow through the fistula tract hence facilitating the closure of the tract. [5, 9, 10] This is achieved by inducing atrophic changes in the endometrium-like epithelium of the fistulous tract. However, it was documented that this therapy was more appropriate for UFC caused by endometriosis. [5] Similarly, some authors have reported failed cases of UFC with medical treatment alone. [5,9] This patient was still desirous of future fertility; hence she had a fistulectomy and repair of the uterine wall rent. She recovered fully, and she has remained asymptomatic.

## Conclusion

With the rising CS rates, particularly in low-resource settings such as Nigeria, it has become imperative for medical practitioners who offer maternity care to have additional training in conventional CS techniques with an emphasis on infection prevention.

**Authors' Contributions:** AAK, PMA, and OAO participated in the clinical management of the case and conceptualized the case report. AAK and OAO contributed to the literature review. AAK, PMA and OAO drafted the manuscript. AAK and OAO revised the manuscript for sound intellectual content. All three authors read and approved the final manuscript.

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**Patient's Consent Statement:** The patient's consent was obtained to publish this case report.

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