Case Report

Vesicouterine Cutaneous Fistula Post-cesarean Section — A Case Report

Seham M. Abufraijeh[1], Seif Jankhout[2], Basil Al-Tah[2]*, Abdallah Daradkeh[2], Suhaib A. Allauzy[3]

[1]Department of Obstetrics and Gynecology, Faculty of Medicine, Mutah University, [2]Faculty of Medicine, Mutah University, Al-Karak, [3]Department of Obstetrics and Gynecology, Al-Basheer Hospital, Jordanian Ministry of Health, Amman, Jordan

Abstract

This case report presents the diagnosis and management of a 41-year-old woman who developed a vesicouterine cutaneous fistula after a cesarean section. She initially presented with pus discharge from the cesarean wound site, and later exhibited cyclical hematuria and fluid leakage from the skin incision, prompting further investigation. Diagnostic modalities, including computed tomography with contrast, confirmed the presence of a complex fistula involving the bladder, uterus, and skin. Conservative management with gonadotropin-releasing hormone therapy was attempted but discontinued due to worsening symptoms. Surgical intervention was performed successfully, leading to the resolution of symptoms. This case underscores the importance of prompt recognition and appropriate management of rare postcesarean complications, emphasizing adherence to anatomical principles.

Keywords: Case report, cesarean section, complex fistula, cyclical hematuria, diagnostic imaging, gonadotropin-releasing hormone, surgical management, vesicouterine cutaneous fistula, wound infection

Introduction

The prevalence of cesarean sections (CSs) has significantly increased in various countries, emerging as the primary surgical intervention for childbirth.[1,2] Over the past three decades, there has been a consistent and notable increase in the rate of CS procedures, surpassing the recommended optimal proportion of 10%15% of births.[1,3] As the rate of CS rises, it is important to acknowledge the well-established high rate of complications associated with this procedure.[4] The emergence of obstetric fistulas stands out as a notable postoperative complication following CS. Obstetric fistula has devastating consequences for affected women, including physical challenges, social and marital relationship disruptions, economic incapability, and mental health issues.[5]

Vesicouterine cutaneous fistula is an exceedingly rare pathology. These unique conditions arise from factors such as trauma, neoplastic developments, inflammatory processes, or iatrogenic injuries.[6] In this report, the first documented case of a vesicouterine cutaneous fistula following a CS is presented.

Case Report

A 41-year-old gravida 14, para 5 + 8 (para 5, living 5 children, +8 abortions) woman with a history of five previous cesarean deliveries and one ectopic pregnancy. Her medical and surgical history is otherwise unremarkable. She underwent a difficult low transverse CS due to severe adhesions. Adhesiolysis and dissection of the bladder were performed as needed to safely expose the lower uterine segment. The Foley catheter was removed the following day, and she was discharged on the 4th day postoperation.

Twenty-seven days post-CS, the patient presented to the emergency room with a complaint of discharge from the

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Address for correspondence: Dr. Basil Al-Tah, Faculty of Medicine, Mutah University, Al-Karak 61710, Jordan.
E-mail: basilhussam2000@gmail.com

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wound site. Upon examination, it was found that the cesarean wound had a 3 cm centrally located defect in the skin and subcutaneous tissue, with purulent discharge, for which she was treated as a case of wound infection. Five months later, the patient came back with cyclical hematuria and fluid leakage from the wound. At that time, she denied having symptoms of urinary incontinence or abnormal fluid leakage per vagina.

This symptom raised suspicion regarding the potential presence of a fistula. A significant amount of fluid discharge with purulent material was coming out of the wound. The discharge was tested for creatinine levels, which were found to be similar to the levels typically seen in urine. This indicated that the bladder was involved in the fistula, as the discharge contained creatinine levels equivalent to those of normal urine.

A computed tomography (CT) scan with intravenous (IV) contrast was conducted as part of the investigative process. The report showed strong evidence of a vesico-cutaneous fistula, manifested as a tract between the bladder and the skin measuring about 5 cm in length, along with contrast spillage through the skin wound. However, findings regarding the vesicouterine part of the fistula were less evident [Figure 1]. A methylene blue test was then performed, followed by a cystoscopy. Methylene blue-dyed saline was injected into the bladder through a Foley catheter after placing a white gauze into the vagina to help detect any leak. The bladder was filled with the dyed saline until two small dots of the dye appeared on the vaginal gauze, confirming the presence of a vesicouterine fistula. A cystoscopy was then performed by the urology team. The findings included a defect at the dome of the bladder. The procedure also included an attempt to treat the defect by cauterization, which failed due to the small diameter of the fistula according to the cystoscopy report.

In the end, there were two continuous fistulas: one connected the dome of the bladder to the uterus, and the other connected it to the skin.

Treatment with gonadotropin-releasing hormone was initiated as triptorelin (Decapeptyl, 3.75 mg IM injection once monthly, Ferring pharmaceuticals, Switzerland), aiming to treat the symptoms of cyclical hematuria until definitive surgical intervention. However, this medication was later discontinued due to the patient's complaints of increased fluid discharge from the skin wound. Based on the urologists' opinion, the decision was made to try conservative management using an indwelling catheter for 4 weeks. During this period, the patient developed deep vein thrombosis. Therefore, the conservative management was extended for 3 months until she completed her treatment for DVT. Despite prolonged use of the Foley catheter, symptoms failed to improve. Thus, the patient was planned for surgical correction of the fistula conducted by a multidisciplinary team.

The surgical report highlights details of the procedure that was conducted. An initial infra-umbilical midline skin incision was performed, revealing the rectus sheath, which was tightly adherent to the uterus and bladder. The tightly adherent bladder was dissected retrogradely starting from the pouch of Retzius toward the adhesive area at the site of the fistula. An injury to the bladder wall was encountered during dissection, so the bladder was then approached by opening the broad ligament laterally to reach the vesicouterine space. A band between the dome of the bladder and the uterus (which contained the vesicouterine fistula) was cut after achieving proper dissection. After that, a hysterectomy was performed, and the vaginal vault was closed. The vesico-cutaneous tract of the fistula was then resected and the bladder defect was closed in two layers. Following the bladder repair, a methylene blue test was carried out and returned a negative result, confirming the successful resolution of the fistula and proper repair of the bladder defect.

Five days after the surgery, the patient was discharged home with antibiotics, and a Foley catheter was left in the bladder, which was removed 2 weeks later. The patient had a full recovery with an uneventful 1-year follow-up.

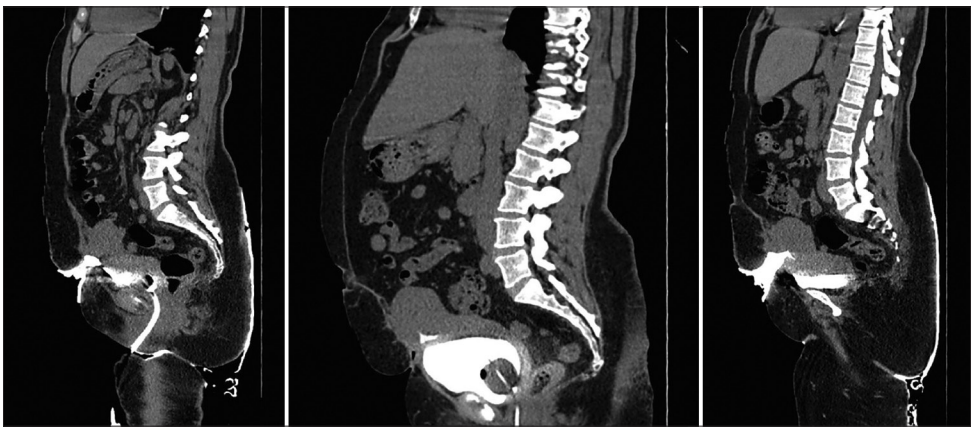

Figure 1: Lateral computed tomography scan with contrast, the fistula tract is clearly seen

Discussion

This case report presents a patient who developed a vesicouterine cutaneous fistula after a CS. What distinguishes this case is the uncommon occurrence of the three components coexisting together, resulting in a challenging diagnostic and management process.

The diagnosis of a vesicocutaneous fistula can be primarily achieved clinically.[7] However, in this instance, it was initially misdiagnosed as a wound infection due to its rare occurrence and unpredictable presentation. During the initial examination, the late presentation of wound infection should have raised suspicion of underlying pathology, but investigations toward the fistula only began when the patient presented after 5 months with cyclical hematuria and persistent fluid discharge from the wound site. The nature of the discharge was identified after an analysis of its creatinine levels, which consistently aligned with the patient's urinary creatinine level.[8]

The patient underwent a CT scan with contrast as part of the initial investigations, leading to the identification of a 5 cm × 1 cm vesicouterine cutaneous fistula. Imaging modalities such as IV urography, a contrast-enhanced CT scan, and magnetic resonance imaging can be valuable tools in the diagnosing and managing of patients with vesicocutaneous fistula.[7,9,10] However, it is noteworthy that the inability to identify a vesicocutaneous fistula through imaging studies does not conclusively exclude its existence.[9] Cystoscopy and methylene blue leakage test were performed because the patient complained of cyclical hematuria. Many authors consider cystoscopy and dye instillation to be helpful in diagnosing vesicouterine fistula and excluding other pathologies such as endometriosis.[10] [Table 1].

Consistent with prior cases, the onset of cyclical hematuria suggests that this case presents as Youssef syndrome.[13,14] Youssef syndrome is a rare medical condition characterized by vesicouterine fistula. In this syndrome, urinary incontinence is notably absent, but individuals experience amenorrhea and cyclic voiding of blood-stained urine during menstruation.[13] The methylene blue test conducted showed evidence of leakage of the dye-stained saline injected into the bladder into the vagina through the uterus. This finding might seem conflicting in the absence of complaints of urinary incontinence by the patient. The incompatible results may be explained by the unique circumstances of our patient. Having a cutaneous component to the fistula may have directed urinary leakage through the lower-pressure vesicocutaneous tract, manifesting as leakage through the skin defect and absence of continuous fluid leakage through the vagina. However, during the dye test, bladder filling with the dyed saline was much more rapid than in the physiological state. This led to bladder expansion and intra-vesical volume that is not usually reached outside the test environment, revealing the fistula through the passage of a small amount of the dyed saline through the uterine component. It is important to highlight that this case marks the first documented instance of Youssef syndrome coexisting with a cutaneous fistula. Due to its rarity, the exact mechanisms and causes of Youssef syndrome remain poorly understood.[13] However, previous literature reveals that repeated CSs are the most common risk factors.[15]

After diagnosing the vesicouterine cutaneous fistula, a trial of conservative management was attempted. This decision was influenced by the limited data available regarding cases where conservative management has proven effective in treating this type of fistula.[11] Unfortunately, in our case, conservative management was not effective in treating the fistula. According to the literature, it is advised to try conservative management for fistulas <1cm and to keep in mind if there is an inflamed tissue surrounding the fistula wall, or if there is an infection, to wait until the surrounding tissue heals.[16] The main challenges of the surgery were the severe adhesions between the anterior abdominal wall, uterus, and bladder.[17]

Table 1: A review of the relevant cases from the literature

Title	Authors and origin	Key point
Vesicocutaneous fistula[11]	Toufique and Merani Origin: Karachi, Pakistan	The case involves a 30-year-old woman who underwent a hysterectomy after multiple cesarean sections. She presented with urinary leakage from her abdominal scar and a urinary leak. Diagnostic tests confirmed a VCF between the bladder and abdominal scar. Conservative management proved successful due to early detection, small fistula size, and absence of additional urinary issues
Vesicocutaneous fistula after cesarean section – A curious complication: case report and review[6]	Tatar *et al*. (corresponding author) Orgin: Turkey	The case involves a 44-year-old woman with a history of two CSs, the most recent of which included a bladder injury in the last one. She presented with recurrent urinary tract infections and periodic urinary leakage through the abdomen. MRI revealed a VCF defect connecting the anterior bladder wall to the scar site, which was treated surgically. This case highlighted the superiority of MRI and CT scans for accurate diagnosis. Successful surgical treatment was attributed to various factors such as defect size, type, associated symptoms, and time since the injury
A rare case of combined vesicovaginal and vesicocutaneous fistulae treated by one-stage surgical repair[12]	Lengmang *et al*. Origin: Nigeria	The case involves a 24-year-old woman who developed combined vesicovaginal and VCF following a difficult cesarean delivery for prolonged obstructed labor, resulting in urinary incontinence that was repaired surgically. This rare case highlights the potential complications of fistula formation postlabor and stresses the importance of early diagnosis and management

VCF: Vesicocutaneous fistula, CT: Computed tomography, MRI: Magnetic resonance imaging, CSs: Cesarean sections

It is well-established that no single surgical technique can be applied to close all types of fistulas.[18] Hence, the surgery aimed to dissect the uterus from the anterior abdominal wall, identify the bladder, and isolate it from the rectus sheath anteriorly to the lower uterine segment posteriorly. A hysterectomy was performed when the surgical team encountered severe bleeding from the anterior abdominal wall due to extensive dissection, with prior consent from the patient who completed her family. The approach to treating complications from CS, including vesicouterine cutaneous fistula, has advanced to include minimally invasive surgical techniques. These methods reduce patient morbidity and promote quicker recovery.[19] Recent developments in minimally invasive surgical techniques have shown encouraging results for treating complex obstetric fistulas. For example, Yahata *et al*. reported a successful laparoscopic repair of a vesicoperitoneal fistula accompanied by a vesicouterine abscess, highlighting how laparoscopic methods can decrease recovery periods and lessen postoperative complications.[20] In conclusion, it is believed that poor surgical technique and failure to adhere to the important surgical principles of restoring the anatomy and identifying structures in the surgical field will increase the incidence of vesicocutaneous and uterine fistula. It is very likely that the fistula was caused by occult injury to the bladder during dissection or by a hemostatic stitch taken during hemostasis without recognizing the injury intraoperatively. In addition, to supplement the findings, a comprehensive table is included in the report presenting a literature review of the only three documented cases of vesicocutaneous fistula, all of which are closely related to the circumstances outlined in this case report.

Ethics statement

This study was conducted in accordance with the ethical principles outlined in the Declaration of Helsinki and its amendments. The authors certify that they have obtained all appropriate patient consent form. In the form, the patient has given her consent for her 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.

Author contributions

Seham M Abufraijeh: Writing the abstract, introduction and the literature review table. Other contribution: Supervision, Reviewing; Seif Jankhout: Collecting information from the patient (History) Writing the Case Presentation; Basil Altah: Writing the discussion and the case presentation. Other contribution: Collecting the information for the literature review; Abdallah Daradkeh: Writing the Discussion and the conclusion. Other contribution: Organizing the paper and the references; Suhaib A. Allauzy: Provided the Patient history and Physical examination findings and also the other investigation (CT scan) He helped writing the introduction and abstract. All authors have read and agreed to the final version of the manuscript.

Data availability statement

All data generated or analyzed during this study are included in this published article.

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Conflicts of interest

There are no conflicts of interest.

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