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Extremely Rare Case of Utero-cutaneous Fistula in Post Cesarean Section Setting with Successful Surgical Management

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Abstract **Introduction**: Fistula is defined as abnormal communication between two epithelium-lined surfaces. Among all types of uterine fistulae, the utero-cutaneous one is infrequently encountered, especially following a lower uterine segment cesarean section. The classic presentation of patients with utero-cutaneous fistula is cyclical hemorrhage from the incision site of previous CS. Surgical intervention is considered the most effective therapeutic option. **Clinical report:** A 34-year-old female patient came to our attention complaining of abdominal pain and bloody discharge from the incisional site 2 months after a lower uterine segment cesarean section (LUCS). Utero-cutaneous fistula was detected by using Pelvic MRI. Patient had successful surgical resection of the fistula. **Conclusion:** Although it's extremely rare, utero-cutaneous fistula should be included in the differential diagnosis of cyclical abdominopelvic pain and/or discharge after lower uterine segment cesarean section (LUCS).

Keywords: *utero-cutaneous fistula, cesarean section, fistulous tract, magnetic resonance imaging, surgical resection*

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1. Introduction

Fistula is created by the formation of an anomalous tract that connects two epithelialized surfaces together [1]. The vast majority of uterine fistulae result from abnormal communication between the uterus and urinary bladder (known as utero-vesical fistula) or intestine (known as utero-colic fistula) because of a variety of underlying causes including accidental injuries during surgical procedures, trauma, intra-uterine devices, long-lasting drains, locally invasive malignancies, chronic infectious diseases, radiation exposure, endometriosis, and imperfect closure of surgical sites especially that made in the uterus [2,3,4]. However, utero-cutaneous fistula is an extremely rare variant of uterine fistulae that results from pathological communication between the uterus and the skin [5]. Utero-cutaneous fistula was first identified in 1993 [1]. Given the paucity of utero-cutaneous fistula, the precise incidence of this condition is yet unclear, with very few numbers of cases have been reported in the literature during the previous 2 decades worldwide [6,7]. Utero-cutaneous fistula is considered one of the rarest consequences of cesarean section (CS) [8]. Patients with post-CS utero-cutaneous fistula are typically presenting with periodic episodes of bleeding from the opened surgical scar of preceding CS [6]. Although, utero-cutaneous fistula can be detected by several imaging studies such as computed tomography (CT) scan with intravenous contrast, hysterosalpingography and/or fistulography, the diagnostic modality of choice is magnetic resonance imaging (MRI) [2,8]. Despite of absence of standard therapeutic guidelines, treatment is dependent on the nature of the underlying cause and the size of the fistula. Although medical management can be used in some cases, surgical intervention remains the most effective and definitive option [2,6,9,10].

We herein introduce a 34-year-old woman who developed utero-cutaneous fistula ten days following CS as the first documented case of the condition in Palestine with early prompt recognition and effective therapeutic intervention.

2. Clinical Report

I.T., a previously healthy 34-year-old Palestinian woman, Gravida 5, para 3 presented on January 16, 2022 following 10 days of an uneventful lower uterine segment cesarean section (LUCS) to the Department of Obstetrics and Gynecology at Al-Istishari Arab Hospital, Ramallah, Palestine, complaining of lower abdominal pain associated with whitish discharge from CS surgical wound. At that time, appropriate cleaning and dressing of surgical wound were done and the patient was prescribed an analgesic medication and then discharged. On March 8, 2022, the patient returned to the hospital due to persistently increasing abdominal pain with a mixed, bloody and whitish discharge from the previous CS incision site. The patient's medical history is free and surgical history is significant for complicated appendicitis with perforation at the age of 13 years, diagnostic laparoscopy and hysteroscopy for infertility issues in 2016, and CS prior 2 years. Upon admission, physical assessment showed hemodynamically stable patient with slight pallor. Examination of the Pfannenstiel incision site revealed pink-to-red fluid leakage from a small pinpoint opening measuring approximately 1 cm at the periphery of the scar. No urine or fecal discharge from the opening. Speculum examination of the vagina was normal. The patient had no fever, genitourinary complaints, and/or changes in bowel habit. Laboratory evaluation was done and revealed anemia with hemoglobin of 10.4 g/dl, leukocytosis with White blood cells counts of 11400/L and elevated C-reactive protein of 51.3 mg/L. Urinalysis showed no abnormalities. Pelvic MRI was performed and disclosed a fistulous tract measuring about 69 mm between the uterine cavity and the skin (Figure 1). Patient diagnosed to have utero-cutaneous fistula and underwent surgical resection and repair of the fistula under general anesthesia. The patient was then discharged from the hospital with a complete satisfactory recovery.

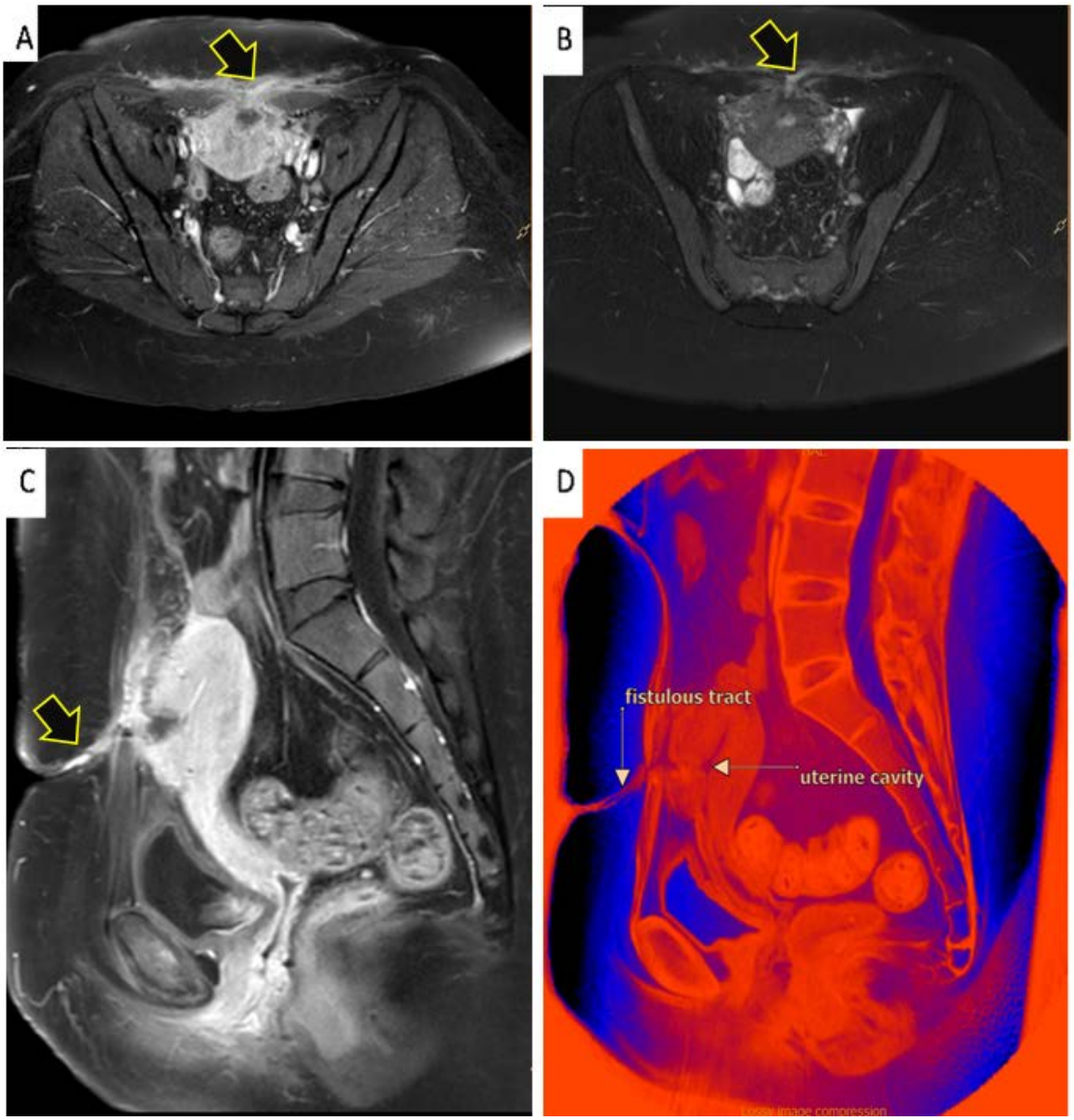

Figure 1. (A, B, C, D): figure 1 is a pelvic MRI with **A** in an axial T1W1 post contrast view, **B** in an axial T1W1, **C** in a sagittal T1W1 and **D** in a sagittal T2W1 with color remap technique, shows a fistulous tract (arrows) measuring about 69 mm arising from the anterior uterine wall at cesarean section site and directed anteriorly, downward, and ending in the left sided anterior abdominal wall, connecting the endometrium with the cutaneous area of anterior abdominal wall. It is associated with surrounding fat stranding and enhancement after IV contrast administration

3. Discussion

Utero-cutaneous fistula is an extremely rare but serious medical condition that results from abnormal communication between the uterus and the skin, and that was first described in 1993 [1]. In the past 30 years, very few numbers of cases have been documented, with most of those cases being post-CS [4]. The frequency of utero-cutaneous fistula is much more common with the classical type of CS compared to lower uterine segment one [11]. Besides cesarean section, septic abortion, uterovaginal malformations, multiple abdominal surgeries, pelvic actinomycosis due to intrauterine devices, prolonged use of drains, incomplete closure of incisions, pelvic abscesses, incomplete placenta removal, and curettage, are all have been implicated in utero-cutaneous fistula formation [2,3,4,11].

Patients with post-CS utero-cutaneous fistula are commonly presented with bloody discharge from a previous Pfannenstiel scar while menstruating classically within a period of 2 months to 6 years following CS. Although this is considered a pathognomonic clinical sign for utero-cutaneous fistula, the occurrence of endometriotic implants on the scars may also present with the same picture and should be excluded [6,11].

Radiological assessment is the mainstay for the diagnosis of utero-cutaneous fistula. Multiple radiological modalities are available including, fistulography, hysteroscopy, hysterosalpingography, CT, and MRI. Fistulography is a straightforward way to visualize the utero-cutaneous fistula by injecting contrast material into the cutaneous fistula. Although it's a simple test to start with, it doesn't give details about fistula communication. In cases of a small cutaneous opening where it's impossible to do fistulography, hysterosalpingography can be performed by injecting contrast material into the cervix [2,5,10]. Computed tomography (CT) scanning with IV contrast and MRI are useful methods for accurately identifying the fistulous tract. Indeed, MRI is preferred over CT due to its superior soft-tissue contrast resolution that aids in the proper characterization of the fistulous tract and its relations to adjacent intraabdominal organs. Therefore, it's considered the diagnostic modality of choice [2,8,12]. Regarding our case, the diagnosis was confirmed by pelvic MRI that showed the presence of a fistulous tract measuring about 69 mm connecting the endometrium with a cutaneous area of the anterior abdominal wall.

Different therapeutic methods have been described to manage utero-cutaneous fistula, including medical treatment and surgical resection. The selection of treatment choice depends on the underlying cause and the size of the fistula. Medical management represented by Gonadotropin-releasing hormone (GnRH) agonists is effective in the treatment of small-sized fistulae by causing epithelial alterations and decreasing fistula drainage with subsequent obliteration of abnormal tract. Meanwhile, large-sized fistulae and/or large skin openings usually require surgical treatment, which results in a highly satisfactory and significant fast healing. That's why, surgical intervention remains the most effective and definitive management of utero-cutaneous fistula [2,6,9,10].

4. Conclusion

Although the introduction of the lower uterine segment cesarean section (LUCS) has reduced the prevalence of utero-cutaneous fistulas, they can still occur following it. Therefore, the diagnosis of post-LUCS utero-cutaneous fistula requires a high clinical suspicious index. In this case report, we highly recommend considering the extremely rare possibility of utero-cutaneous fistula in women who are suffering from abdominal pain and/or discharge following LUCS. Imaging modalities play an essential role in accurate diagnosis, aiding clinicians in early prompt recognition and effective therapeutic intervention.

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