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Utero-Cutaneous Fistula as a Rare Complication After Cesarean Delivery: Case Series

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Abstract

Introduction: Utero-cutaneous fistula is a rare condition following uterine surgeries especially cesarean section. This kind of fistula has various etiologies including drain use, iatrogenic trauma, endometriosis, multiple abdominal surgeries, incomplete closure of uterine wound during cesarean delivery, inflammatory processes related to intra-abdominal sepsis or infectious, and dislocation of intrauterine devices.

Case Presentation: This report deals with two unusual cases of utero-cutaneous fistula. The patients referred with discharge from abdominal wall. The first one had vesico-cutaneous fistula simultaneously. Both of them had a second cesarean section. After four months of cesarean section, in fistulography report of the first case, it was found irregular fistula tract associated with vagina following cannulation and contrast injection. In the second case, ultrasonography revealed the attachment of uterus to abdominal wall as well as accumulation and communication of the small amounts of fluid from uterine cavity to abdomen wall. After confirming the diagnosis, the repairing surgery was successfully planned.

Conclusions: Cesarean has some rare morbidity such as uterocutaneous fistula that needs awareness of physician and patient. The early diagnosis and repairing of this abnormality is essential.

Keywords: Utero-Cutaneous Fistula, Cesarean Section, Perinatal Morbidity, Side Effects

1. Introduction

Utero-cutaneous fistula is a rare condition following uterine surgeries especially cesarean section (1). This kind of fistula has various etiologies including drains use, iatrogenic trauma, endometriosis, multiple abdominal surgeries, and incomplete closure of uterine wound during cesarean delivery, inflammatory processes related to intra-abdominal sepsis or infectious, and dislocation of intrauterine devices (2, 3). Due to the serious complications of this fistula such as repeated abortions, postpartum and postoperative complications (4), the early diagnosis and repairing of this abnormality is essential. This is the report of two unusual cases of utero-cutaneous fistula that one case had simultaneously vesico-cutaneous fistula.

2. Case Presentation

Case 1: this case was a G2P2L2 woman with the history of two cesarean sections in Yas hospital, Tehran, Iran, affiliated to Tehran University of Medical Sciences. One month after the recent cesarean delivery, the patient noticed an unusual discharge from the incision site, consequently admitting to the hospital. The suture was washed and sutured and then, she was discharged with appropriate antibiotic therapy. On discharge, the patient did not report any symptoms including abdominal pain, fever, nausea, vomiting or other gastrointestinal manifestations. About four months after the surgery, the patient found a leakage of menstrual blood during menstruation from a hole at the site of incision beginning a few months ago and thus referred to the hospital again. In ultrasonography assessment, the uterine showed normal shape, size, and echogenicity with the dimension of $70 \times 31 \times 39$ mm^3. Endometrial lining thickness was normal in the range of 5 to 6 mm. We found the accumulation of small amounts of fluid in endocervical channel. Endometrial cavity also spread to the surgical incision site in the wall of the uterus body and it seemed to spread even to the outside surface of the uterus. It also revealed a mild accumulation of fluid with inflammatory changes around the uterus especially in the margins of the recent surgical incision. The ovaries were reported to be normal with the diameter of 27×15 mm^2 in the right and 27×14 mm^2 in the left. It was also found a normal adnexal condition without evidence of solid or cystic masses. Contrast computed tomography (CT) revealed that dye leaked into the uterus via narrow fistula. In fistulography report, it was found dye collection in-

dicating probable fistula connected to the uterus. Based on the available evidence, the diagnosis of utero-cutaneous fistula plus vesicocutaneous fistula was suspected.

Case 2: The patient referred with discharge of abdominal wall between umbilical site and pubis. She had a history of cesarean section four months ago due to repeated cesarean section in Vali-e-Asr hospital, Tehran, Iran, affiliated to Tehran University of Medical Sciences.

In physical examination, we found a hole about 2 to 3 mm with serosanguinous discharge from this site. Ultrasonography revealed the attachment of uterus to abdominal wall, accumulation, and communication of the small amounts of fluid from uterine cavity to abdomen wall. The uterus and ovaries were in the normal size, and there was not free fluid in pelvic cavity. Laboratory tests were normal in the two patients. In addition, tumor markers and abdominal X-ray graphy were normal. After confirming the diagnosis, the repairing surgery was planned. For the first case, diagnostic laparotomy was done to find the fistula; the fistula was separated from the skin and resected. Then, enterolysis was performed and omental flap was implanted intra abdominally and the uterine and skin were repaired in the site of fistula. Intra operatively, extra peritoneal adhesion to the bladder appeared that was sharply separated from the bladder. Finally, cysto-ureterolysis was performed and border of the uterus to the bladder was determined and separated. The patient was discharged 72 hours after surgery. She has not shown any problem until now, one year after surgery. For the second case, laparotomy was done, and the extensive and severe adhesion band between uterus and abdominal wall was separated. The fistula was removed and the bleeding from the separated adhesion points was controlled by continuous and separate suture (in some places). Physicians, who conducted previous surgery for this patient (cesarean section), said that at the end of the procedure, the patent had pain and was not calm. As a result, the repair of facial layer was not perfect that may lead to traumatized uterus or attached uterus to the facial layer.

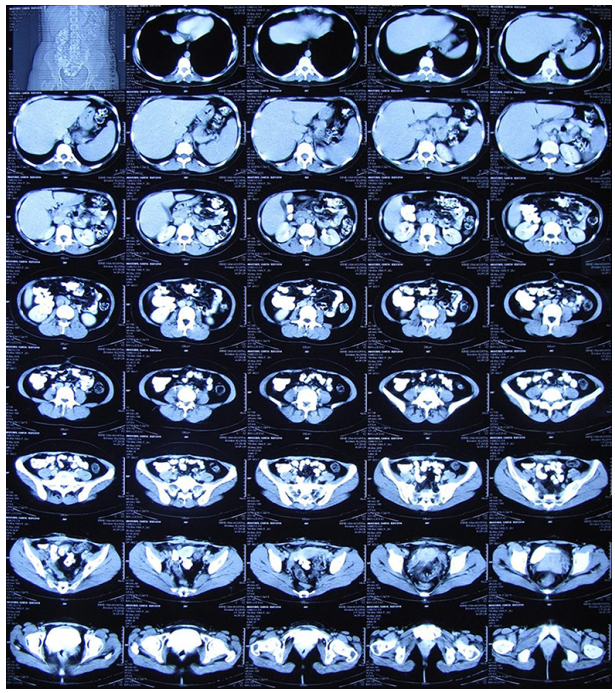

Figure 1. Contrast Computed Tomography (CT) Revealed That Dye Leaked Into the Uterus Via Narrow Fistula

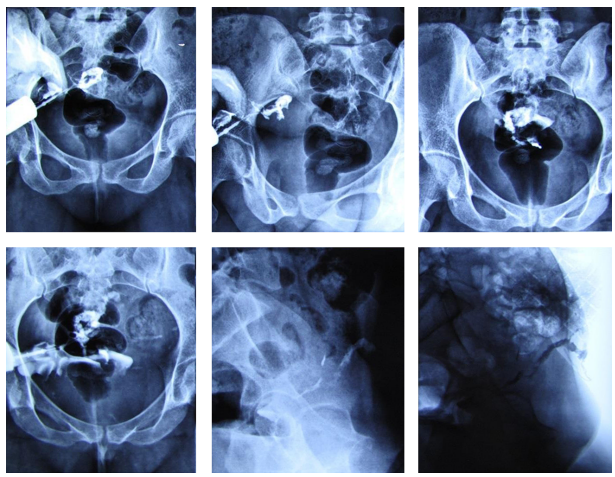

Figure 2. In fistulography Report, it Was Found Dye Collection Indicating Probable Fistula Connected to the Uterus

3. Discussion

Utero-cutaneous fistula is a rare condition with difficulty in diagnosis and management. The most common cause of this condition has been shown to be incomplete closing wound after cesarean section and other uterine interventional procedures. Diagnosis of this abnormality frequently requires radiological assessments using fluoroscopic or cross-sectional modalities (5). Fistulography is the choice direct mean of visualizing the fistula that can be considered when feasible such as in cutaneous fistula (6). Other diagnostic mainstays of investigations include intravenous urography and pyelography or ureterography or even magnetic resonance imaging (7). Reviewing the literature found a few case reports on uterocutaneous fistula and its medical and surgical management (5-7). The surgical repairing is the treatment of choice; however, preoperative medical treatment with gonadotropin-releasing

hormone agonists has been accompanied with better outcome; however, some cases may end up with hysterectomy. The present cases that occurred following cesarean section were successfully diagnosed by fistulography and ultrasound and finally managed properly by the repairing surgery. Although intraoperative extra peritoneal adhesion to the bladder occurred, this complication was also well managed and the patient was discharged without any complication. Our patients had one risk factor; the first one was at risk of infection in the location of surgery and second repair; the risk for the second one may be due to the training hospital system and uncooperative patient and anesthesiologist to calm the patient, leading to ineffective repair.

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