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Uterocutaneous fistula: A rare complication of cesarean section

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Abstract:

A uterocutaneous fistula is an abnormal tract between the endometrium of the uterus and the skin. It is a rare but significant postcesarean section complication. This is a case of a 39-year-old female who presented with cyclical bleeding from her cesarean section scar during menstruation over the past 4 years. The assessment revealed a discharging sinus in the scar and an acutely anteverted, fixed uterus. Symptoms persisted despite medical treatment for the provisional diagnosis of endometriosis. The diagnosis was updated as uterocutaneous fistula based on a computed tomography scan of the pelvis and abdomen and a hysterosalpingogram. The fistula was surgically excised, and a hysterectomy was performed. This report elaborates on the rare presentation, diagnosis, and definitive surgical management. The surgical management resulted in a successful resolution, highlighting the importance of choosing the appropriate treatment modality while adhering to a patient-centered approach.

Keywords:

Case report, cesarean section, uterocutaneous fistula

Introduction

A fistula is a tract between two epithelium-lined surfaces.[1,2] Uterocutaneous fistula is a tract communicating the endometrium of the uterus and the skin. It is most likely lined by endometrium. It occurs as postsurgical complications, where there is incomplete closure of incision, poor techniques, use of drains, and postsurgical infections.[2] The prevalence of uterocutaneous fistulas is extremely low due to their rarity as a medical condition. There are no large-scale studies providing exact prevalence rates because the condition is typically reported as isolated case reports or in small series in the medical literature. Managing this rare condition is challenging due to the scarcity of existing literature and the lack of standardized protocols, necessitating an individualized approach for each case.[1,3]

Case Report

A 39-year-old woman presented with abnormal, painless bleeding from her lower segment cesarean section scar during menstruation, which began 6 weeks after her last cesarean section. This bleeding had persisted intermittently over the past 4 years. Initially, she also experienced a watery discharge from the scar between menstrual cycles, which had resolved by the time of presentation.

The patient is a mother of four children, all delivered through cesarean sections over an 11-year period. Her most recent cesarean section, performed 4 years ago, was free of intraoperative or immediate postoperative complications, and her initial postpartum recovery was uneventful without any infections. She exclusively breastfed her baby for the first 4 months.

Abdominal examination revealed skin discoloration and subcutaneous thickening

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over the cesarean section scar with a discharging sinus toward the left side. Vaginal examination revealed an acutely anteverted, less mobile, almost fixed uterus adhered to the anterior abdominal wall.

Routine basic investigations, including full blood count and urinalysis, were normal. The ultrasound scan of the abdomen and pelvis revealed a bulky uterus adhered to the anterior abdominal wall with no focal lesion or adnexal masses. A provisional diagnosis of cesarean scar endometriosis was made based on the cyclical nature of the bleeding and the poor response to initial antibiotic treatment. She was subsequently treated with hormonal therapy, including GnRH agonists and progestogens, which provided partial relief. However, she continued to experience significant bleeding from the scar site during menstruation.

This prompted a computed tomography (CT) scan of the abdomen and pelvis for further evaluation, which revealed findings suggestive of a uterocutaneous fistula extending from the skin to the anterior wall of the uterus. A hysterosalpingography (HSG) was subsequently performed to better characterize the fistulous tract, which showed contrast extravasating into the skin through a fistulous tract extending from endometrial cavity [Figure 1].

Fistulous tract dissection with total abdominal hysterectomy was performed, as the patient had completed her family. The fistula was meticulously traced using an aneurysm needle [Figure 2], followed by careful dissection that included the removal of the thickened rectus sheath, which was firmly adhered to the uterus. Dissection of the urinary bladder was particularly challenging, as it was closely fused with the fistulous tract and the anterior uterine surface. An *en bloc* resection was carried out, removing the entire fistulous tract along with the uterus [Figure 3]. The postoperative recovery was uneventful.

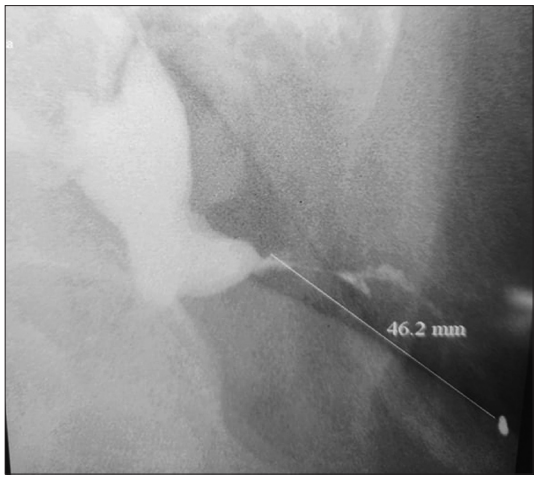

Figure 1: Hysterosalpingogram; the fistulous tract, begins from the midline deviating 4 cm away to the left side of the abdomen

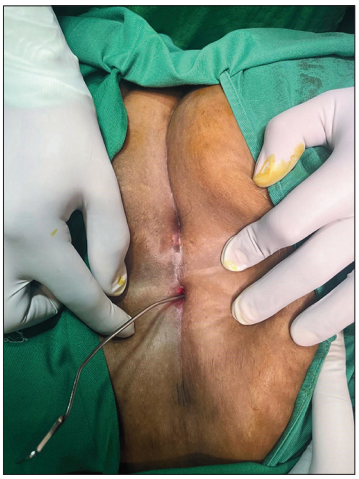

Figure 2: Probing of the fistula with an aneurysm needle

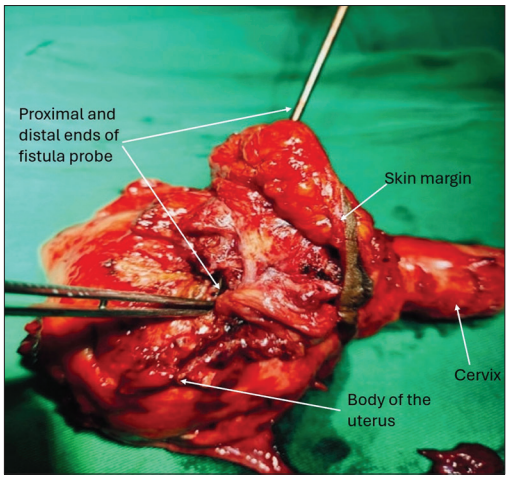

Figure 3: *En bloc* resection of the fistulous tract from the skin to the endometrium. The metal probe indicates the fistulous tract

Discussion

Fistulas, though uncommon, are recognized in gynecology and are most often associated with postsurgical complications or mismanaged obstetric care. Uterocutaneous fistulas typically present with blood or blood-stained discharge from the skin, especially during menstruation.[2]

Etiologies include multiple abdominal surgeries; incomplete closure of the uterine wound during myomectomy or cesarean section; use of drains; and postoperative complications, such as infection and inflammation. Septic abortions, migration of intrauterine devices, endometriosis, and retained placenta after abdominal pregnancy are more extreme etiologies.[1]

Differential diagnoses, such as extrapelvic endometriosis or postsurgical wound infections, can pose significant diagnostic challenges. Advanced imaging modalities,

including ultrasonography, CT, magnetic resonance imaging, hysteroscopy, and hysterosalpingography (HSG), as well as simpler techniques like the methylene blue test, play a pivotal role in establishing the diagnosis.[2] Furthermore, fistulography offers direct visualization of the fistula and uterine cavity through radiographic imaging following the injection of contrast medium into the fistulous opening.[2,3] These diagnostic tools are complementary, and their selection should be tailored based on resource availability and the expertise of the clinical team.

Uterocutaneous fistulae are exceedingly rare, with only two documented cases in the literature achieving successful medical management using GnRH analogues.[4] Medical management is successful when the lining of the fistulous tract is endometrial in origin. In the presence of GnRH analogs, the endometrium will undergo atrophy leading to the resolution of symptoms. However, this will not differentiate uterocutaneous fistula from subcutaneous endometrioma with discharging sinus. Treatment with GnRH analogue is associated with notable limitations, including variable success rates influenced by the size and chronicity of the fistula, prolonged treatment duration, and potential drug-related adverse effects. Consequently, surgical intervention is widely regarded as the treatment of choice in most reported cases. As demonstrated in our case, surgical management not only offers a more definitive resolution but also achieves faster and more reliable outcomes.[5] A laparoscopic approach, when feasible, may be a preferable option following meticulous patient selection, especially in fertility-preserving cases. However, challenges such as pelvic adhesions, obesity, fibrosis around the fistulous tract, and concurrent comorbidities can complicate surgical procedures, necessitating advanced skills and careful planning.[1]

In conclusion, uterocutaneous fistula is a rare yet significant complication that affects both physical and mental health of the patient. Its occurrence can be minimized through meticulous surgical techniques, precise tissue approximation, and rigorous infection prevention measures. Treatment strategies, whether medical or surgical, should be tailored to the severity of the condition, the expertise of the healthcare team, the patient's overall health, and their fertility preferences.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given her consent for her images and other clinical information to be reported in the journal. The patient understands that name and initials will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

Authorship contributions

Dr Sampath Gnanarathne - Involved in the conceptualization, patient management, manuscript drafting and editing.

Dr Ashani Ratnayake- Involved in the conceptualization, patient management, manuscript drafting and editing.

Dr Ayodhya Kariyawasam - Involved in manuscript drafting and editing.

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Conflicts of interest

There are no conflicts of interest.

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