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Utero-Cutaneous Fistula after Caesarean Section Delivery: Diagnosis and Management of a Rare Complication

Aalaa A. Wahab Marzooq*#, Maryam Mohamed Mahmood, Hiba Moayad Saeed, Rabab Mustafa Mohamed Khalafalla

Salmaniya Medical Complex, Manama, Bahrain
Email: #alaa_a.wahab@hotmail.com

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Abstract

Utero-cutaneous fistula following cesarean section is a rare occurrence. We present the case of a 34-year-old woman who presented to our department four years after her second cesarean section with a history of pain and blood discharge from a previous Pfannenstiel incision, during menstruation, with an absence of vaginal menstrual flow. Despite a prior surgical repair operation, her symptoms persisted. A pelvic MRI was done to confirm the diagnosis of utero-cutaneous fistula, and surgical management was pursued. This case report aims to contribute to the existing literature on utero-cutaneous fistula and provide insights into the diagnostic considerations and management strategies for this rare complication.

Keywords

Cesarean Section, Uterocutaneous Fistula, Complication, Uterus

1. Introduction

A fistula is defined as an abnormal connection between two epithelial-lined surfaces and often occurs as a complication following surgery, infection, or chronic inflammatory conditions [1]. Utero-cutaneous fistula, an exceedingly rare condition, involves abnormal communication between the anterior wall of the uterus and the abdominal wall, with only limited reports documented in the existing literature. Potential causes are varied and include iatrogenic injuries during surgery, endometriosis, incomplete hysterorrhaphy, uterine cavity revisions, reten-

*First author.
#Corresponding author.

tion of placental material after delivery, prolonged abdominal drain usage, radiation injuries, trauma, post-operative infections or injuries, and the use of intrauterine devices [2].

Typically, these fistulas present as cyclical bleeding from an abnormal opening in the scar of a previous cesarean section [3]. Diagnosis underscores the necessity for a comprehensive diagnostic toolkit, including CT scans with intravenous injection of contrast medium and MRI, which are essential for gynecologists facing possible utero-cutaneous fistulas [2].

Given the rarity of utero-cutaneous fistulas and the limited clinical cases documented, diagnosing and determining the appropriate therapeutic approach remains a challenge for gynecologists. The management of utero-cutaneous fistulas depends on the underlying cause and is usually tailored to individual patients, as there is no one-size-fits-all management plan.

The objective of this paper is to detail the clinical presentation, diagnostic challenges, surgical interventions, and management of a 34-year-old female with a rare utero-cutaneous fistula following cesarean sections.

2. Case Report

We present a unique case involving a 34-year-old female with a history of two cesarean sections. The patient presented with persistent bloody discharge from a previous Pfannenstiel incision, occurring monthly in tandem with her menstrual cycle, yet with no vaginal flow. She had undergone a lower segment cesarean section at a different hospital four years ago. She was diagnosed with a utero-cutaneous sinus, which was repaired three years ago in the same hospital. A histopathology examination confirmed a sinus tract with endometrial tissue (endometriosis), with no subsequent improvement.

The general examination of the patient was within normal limits. The previous Pfannenstiel incision had a solitary fistulous opening approximately 5mm in diameter, with sprouting granulation tissue noted at the opening. A vaginal examination revealed fibrotic stenosis of the cervical os, through which a uterine sound could not be passed.

The diagnostic process included advanced imaging techniques at our hospital, such as magnetic resonance imaging (MRI). The MRI was chosen for its excellent soft tissue visualization capabilities, providing detailed information about the fistula tract, the uterine wall, and surrounding pelvic anatomy and proving particularly useful in complex cases. The MRI revealed thinning in the anterior uterine wall related to the previous cesarean section. It showed a well-defined ovoid lesion measuring 2.9 × 4 × 4.2 cm, located intraperitoneally at the pelvic midline between the rectus muscle and the inferior wall, inseparable from the uterine fundus. The anterior wall was indenting a subcutaneous scar from previous surgery. The lesion showed an isointense signal on T1-weighted imaging and a heterogeneous iso-to-high signal on T2-weighted imaging, with small cystic areas within. Additionally, it demonstrated heterogeneous enhancement

on the post-contrast series. The lesion was also seen as closely related and inseparable from the adjacent small bowel loops, a feature of endometriosis from a cesarean section forming a utero-cutaneous fistula (Figure 1).

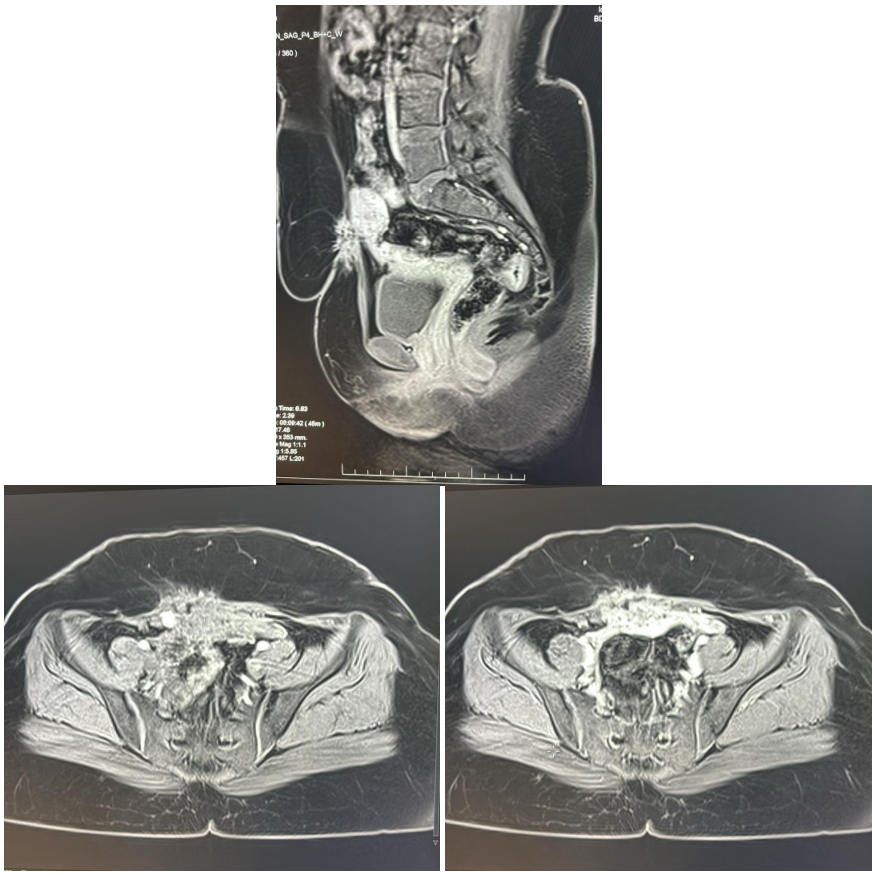

Figure 1. These images collectively depict the presence of a fistula, providing detailed anatomical context and highlighting the extent of the abnormality in the pelvic region.

After counseling, the patient was scheduled for a laparotomy with excision of the fistula tract and a possible hysterectomy.

Exploratory laparotomy revealed an irregular anterior abdominal wall scar with an invaginated area in the middle. There were significant adhesions of the anterior abdominal wall with omentum and bowel. The uterus was irregular and directly adherent under the skin, distorting the anatomy. Both fallopian tubes and ovaries appeared normal. Multiple non-absorbable sutures from the previous surgery were visible in the anterior abdominal wall and on the uterus. No communication was observed between the uterine cavity and the cervical canal. Intraoperatively, attempts to perform hysteroscopy were unsuccessful due to occlusion of the internal os.

The excision of the anterior abdominal wall endometriosis, the fistula tract, and a total abdominal hysterectomy with bilateral salpingectomy were performed, with both ovaries preserved, followed by abdominoplasty by a plastic surgeon. The patient's immediate post-operative condition was deemed satis-

factory.

The specimens were sent to the pathology laboratory for definitive histological examination. Microscopic examination of sections from the uterus showed mild endocervicitis, secretory phase endometrial glands, and a few foci of adenomyosis in the myometrium. Examined sections of fibroadipose tissue displayed multiple foci of endometrial glands and stroma (endometriosis, highlighted with CD10 and vimentin). Adjacent abdominal wall skeletal muscle bundles with focal atrophy were also noted. The pathologist confirmed the surgical suspicion of a utero-cutaneous fistula.

The immediate postoperative course was uneventful and satisfactory, without any immediate complications. During the 6-week follow-up appointment, the patient reported no postoperative complications, with no history of bleeding or discharge. The surgical wound was healed entirely on physical examination, with no signs of skin infection, wound dehiscence, or discharge. Additionally, there were no indications of recurrence of the uterocutaneous fistula. The patient's smooth postoperative recovery and the absence of any complications or recurrence of the fistula during the follow-up suggest a successful surgical intervention and effective management of the condition.

3. Discussion

Fistulous communications of the uterus are relatively rare. We report a case of a uterocutaneous fistula that developed following a cesarean section, which was further complicated by surgical interventions and the presence of non-absorbable sutures that may have contributed to the fistula's persistence. Despite previous surgical attempts, the persistence of such a complication highlights the complexity of management and the need for a meticulous surgical approach.

Our patient experienced a fistula following a cesarean section, similar to the case reported by Maddah *et al.* and Thubert *et al.* [4] [5].

In contrast, other reports have indicated a similar presentation after septic abortion. To our knowledge, this is the first case of a uterocutaneous fistula developing following a septic abortion induced by the introduction of a Laminaria tent into the uterus [6].

Another report described an unusual case of a uterocutaneous fistula that developed in a multiparous woman after the surgical evacuation of an incomplete first-trimester septic abortion [7].

In a case of uterovaginal malformation, a young woman who underwent surgical intervention for cryptomenorrhea three years ago developed menstrual discharge from the abdominal scar [8].

We also observed a reported case of pelvic actinomycosis due to intrauterine devices presenting as a cutaneous fistula [9].

Additionally we observed cases of utero-cutaneous fistula following abdominal myomectomies. These cases highlight the potential for this rare complication to occur even after multiple abdominal myomectomy procedures, un-

derscoring the importance of careful post-operative monitoring and management [10] [11].

The diagnosis of uterine fistula can often be straightforward, with pathognomonic clinical signs such as bloody discharge through an abdominal scar during menstruation. Other investigative approaches include fistulography, where the injection of contrast material through the skin opening shows a connection to the uterus, and hysterosalpingography and MRI with contrast as other diagnostic modalities [5].

The treatment of utero-cutaneous fistulas requires a customized approach, considering the unique circumstances of each case. Although no standardized, evidence-based treatment modality is currently available, various options have been reported, including medical treatment and surgical resection. The selection of treatment method depends on the underlying cause and the size of the fistula. Successful management typically involves a surgical intervention to excise the fistulous tract, followed by meticulous repair of the uterine and abdominal wall defects [2] [3]. While surgical intervention, including hysterectomy and excision of the fistulous tract, remains the mainstay of treatment as evidenced in published cases [6], there are reports of successful non-surgical management using gonadotropin-releasing hormone agonist (GnRHa) administration [12]. GnRHa works by suppressing menstruation and rendering the endometrial-like lining of the fistulous tract atrophic, resulting in the spontaneous closure of the fistula.

Additionally, another case reported a combined conservative surgical and medical treatment of a utero-cutaneous fistula, where GnRHa was administered for six months, followed by surgical repair via laparoscopy and laparotomy [5]. This suggests a potential for conservative treatment options in select cases, although surgery is often required for definitive management.

In our case, medical treatment was not an option as the patient chose surgical intervention. Despite these findings, more studies are required to inform evidence-based treatment, including understanding the mechanisms of fistula formation following cesarean sections.

4. Conclusion

Uterocutaneous fistula is a rare condition that can occur following cesarean section or other pelvic surgeries. In the case reported, the use of non-absorbable suture material in uterine suturing has been identified as a contributory factor. Additional factors implicated in its development include infection, necrosis, the presence of foreign bodies, and malignancy. Effective management requires precise delineation of the fistula tract and control of any associated infection. Complete excision of the fistula tract followed by suturing the uterus with absorbable suture material is essential for successful treatment. This case highlights the importance of maintaining a high index of suspicion for uterocutaneous fistula in postoperative patients presenting with unusual abdominal wall drainage. Prompt diagnosis and surgical management are crucial to resolving this uncommon but

challenging complication.

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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