




Case Study

Uterocutaneous fistula managed medically: A case report.

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Doi: 10.29052/IJEHSR.v11.i2.2023.104-109

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Received 30/01/2023

Accepted 17/03/2023

First Published 28/03/2023



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Abstract

Background: The uterocutaneous fistula is an abnormal connection or passageway between the uterus and the skin. The most common causes include infections, trauma, or surgery, a severe medical condition requiring prompt medical attention and treatment.

Case Presentation: Here, we describe the case of a 36-year-old female who developed a uterocutaneous fistula after cesarean delivery. Her post-cesarean recovery was complicated by wound infection, and after healing the wound, she presented with cyclical bloody discharge from a pinpoint opening in the healed wound scar.

Management: On ultrasound and MRI, she was diagnosed as having a uterocutaneous fistula. She opted for non-surgical management and underwent successful medical management with GnRH agonists. She remained symptom-free after the resumption of her menstrual cycle.

Conclusion: In younger populations hoping to avoid surgery, pharmacological care with GnRH agonist therapy can be a reasonable alternative to surgical resection of the fistulous tract. Healthcare professionals should have a high index of suspicion to diagnose this condition.

Keywords

Laparotomy, Uterocutaneous Fistula, Myomectomy, Septic Abortion, GnRH Agonists.



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Introduction

Uterocutaneous fistula (UCF) is one of the rarest complications of cesarean delivery. To date, only 120 cases are reported in the literature¹. In the era of classical cesarean deliveries, several instances of UCF were reported, but the frequency declined after the advent of lower-segment cesarean section. It has also been reported after septic abortion², myomectomy³, and laparotomy. As a rare entity, there can be considerable delay in the diagnosis, and there is no consensus on the treatment.

The standard gold treatment of any fistula is surgical excision. Since the demographics of this patient population are primarily young and require fertility preservation, successful conservative management of UCF is highly desirable, as done in our case study. The exact incidence of this clinical entity is unknown due to its occurrence rarity. Authors searched literature on PubMed/Medline using the mesh term uterocutaneous fistula, and it only reveals case reports.

Case Presentation

A 36-year-old patient who is Gravida 4 Para 3 Abortion 0 was being followed up in our obstetric clinic for pregnancy. Her history is significant for the previous three uncomplicated cesarean sections. She was diagnosed with placenta accrete overlying the previous uterine scar. She underwent elective cesarean sections at 37 weeks gestation. Her cesarean section was performed with an inverted T-shaped incision on the skin and abdominal wall. The uterus was incised via midline longitudinal incision, avoiding the placenta. The placenta was delivered incompletely, as a small portion was deeply adherent and was left in situ. Uterine conservative surgery was done. Her immediate postoperative recovery went smoothly.

She presented to the clinic one month post cesarean with an abdominal wall wound dehiscence, horizontally around 4 cm in size, and an intact rectus sheath. There was no pus drainage, and her serum inflammatory markers were normal. The wound swab confirmed staphylococcal infection and was treated well with oral antibiotics. The wound healed with secondary intention.

A month later, she presented with a bloody discharge from her cesarean scar. On examination, there was a pinpoint opening in her wound with fresh bloody discharge. She was also menstruating at that time.

Management & Results

Ultrasound of soft tissue showed an irregularly marginated hypoechoic area size 3 x 1.5 x 1.6 cm extending from the uterine cesarean scar to the midline of the abdominal wall up to the cutaneous plane. Increased vascularity was noted in this area, and minimal cystic fluid in the deeper part. Figure 1, a provisional diagnosis of UCF was made based on this imaging.

Magnetic resonance imaging of the pelvis with contrast was done for further evaluation. It showed that there is a linear track appearing hypointense on T1w, hyperintense on T2w and STIR with no obvious contrast enhancement measuring approximately 2.5 cm in length and 0.15 cm in diameter seen arising from the endometrial lining of the uterus at LSCS niche and extending superficially outside the uterus through the subcutaneous tissue of the anterior abdominal wall till the LSCS scar, likely a uterocutaneous fistula. The diffuse hyperintense signal is seen in surrounding subcutaneous tissue near the LSCS scar, likely subcutaneous edema (Figure 2).

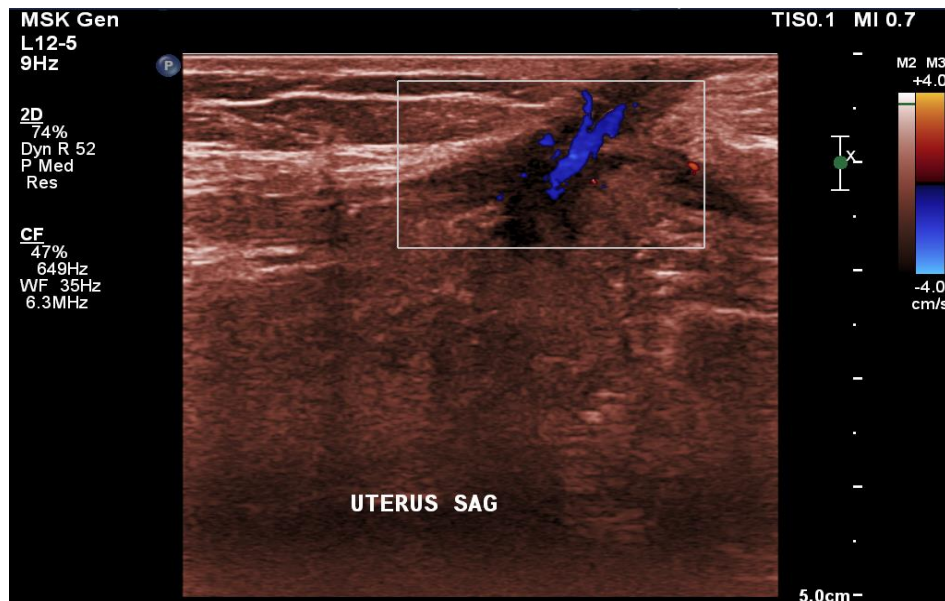


Figure 1: Ultrasound pelvis with color doppler imaging demonstrates linear hypoechoic uterocutaneous fistula with increased vascularity.

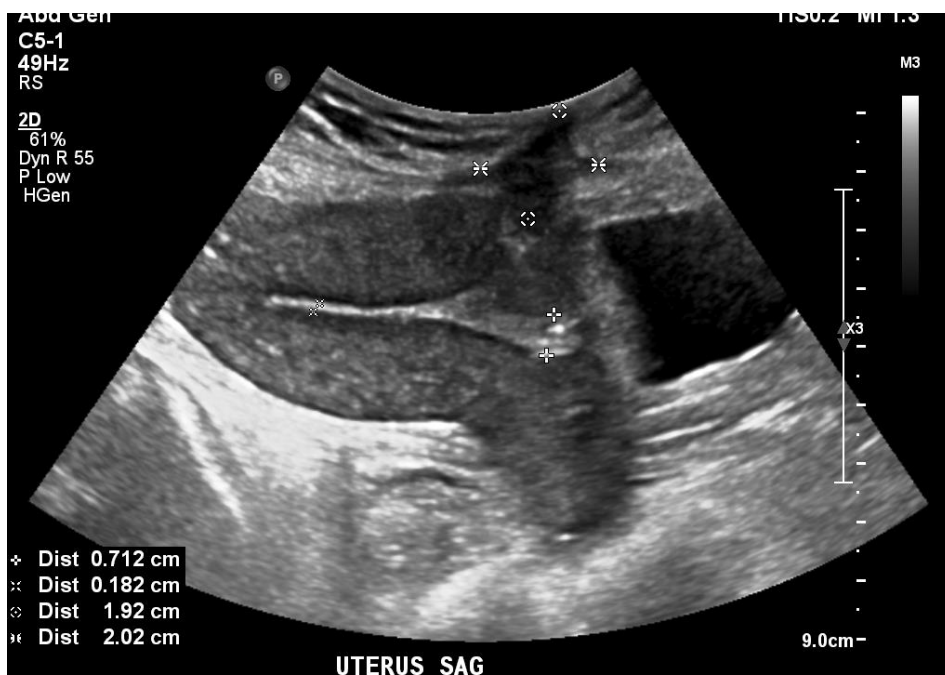


Figure 2: Ultrasound pelvis sagittal image demonstrating linear hypoechoic fistulous tract extending from the lower uterine wall up to skin.

Differentials of a uterocutaneous fistula include infections, intraperitoneal or abdominal wall sites, and tumors. Medical management by gonadotrophin-releasing hormone agonist and the surgical option of hysterectomy with fistulous tract removal were discussed with the patient. She decided to conserve her uterus and opted for medical management. She received a GnRH agonist injection of 12.5 mg for 3 doses, each 3 months apart.

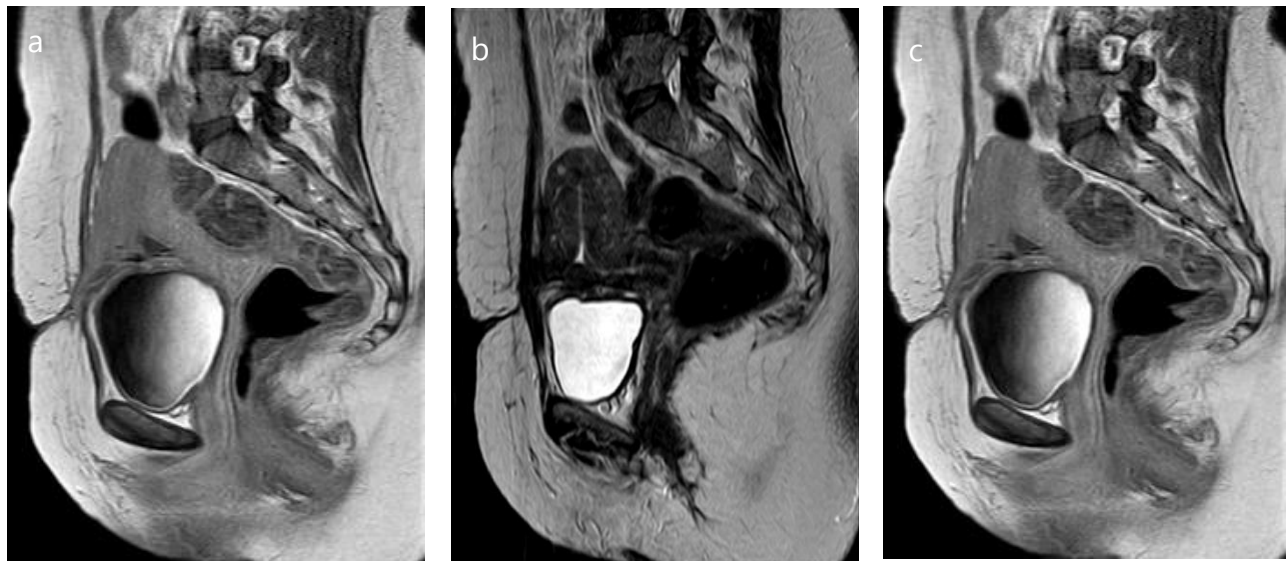


Figure 3 (a-c): MRI pelvis T1W, T2W, and T1W postcontrast sagittal images showing a focal defect in the lower uterine wall (scar site) with linear tract extending from the endometrial canal up to overlying skin without significant enhancement representing uterocutaneous fistula.

On a follow-up visit 3 months after the last injection, she was asymptomatic with no more menstrual bleeding from her incision site. She had regained her normal menstrual periods. Repeat Ultrasound imaging showed that the fistulous tract had closed. However, an incidental finding of an incisional hernia in the anterior abdominal wall exists. She was followed up for up to 22 months after her diagnosis of UCF, and there was no recurrence of her symptoms.

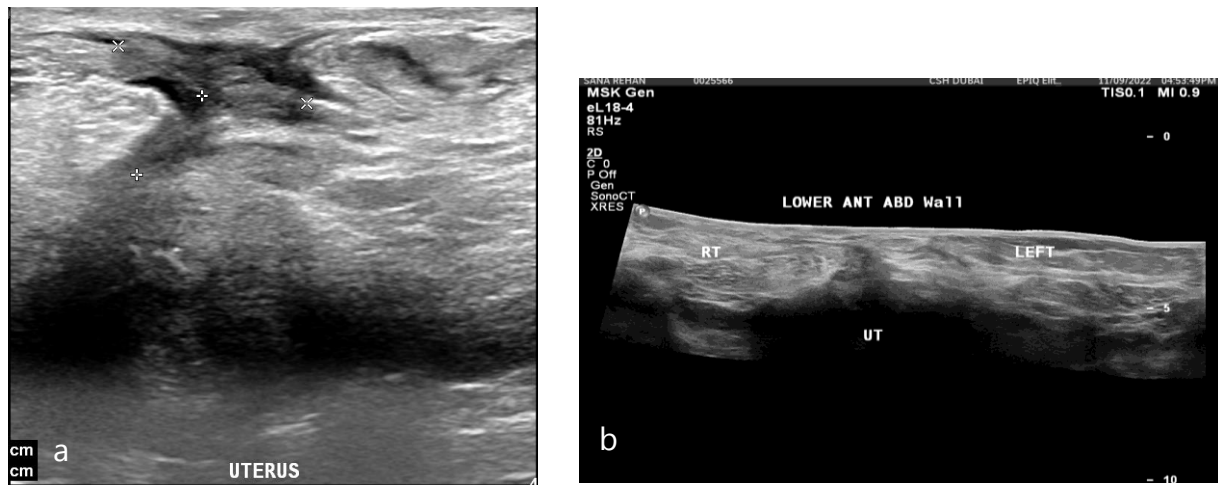


Figure 4 (a & b): Post-medical management ultrasound.

Figures 4a and 4b show an ultrasound done after 22 months of follow-up showing obliteration of the fistulous tract.

Discussion

Fistula is the name given to abnormal communication between two epithelial surfaces. As the name suggests, a uterocutaneous fistula communicates between the uterus and the skin. While it can happen due to pathologies like endometriosis, infection, or malignancy⁴, many of the causes are iatrogenic. These included cesarean section⁵, septic abortion⁶, intrauterine device migration⁷, radiation, or prolonged use of abdominal drains.

Due to vague presentation and mostly arising in a septic context, there is considerable delay in the diagnosis of uterocutaneous fistula, ranging from 15 days to 11 years⁸. Most case reports describe the initial presentation as bleeding from the scar site during menstruation, similar to our patient's presentation. Other presentations include abdominal pain or discharge.

The diagnostic approach includes clinical examination along with imaging. There is no standard gold test, but imaging modalities like Ultrasound and MRI can help. Hysterosalpingogram is another investigation where direct visualization of the fistulous tract can be done.

There is no consensus on the management strategy for this rare diagnosis. As with any other fistulous tract, surgical management with fistulectomy remains a valid management option. A literature review revealed two case reports showing successful medical management with GnRH agonist^{9,10} and one case report employing GnRH agonist after fistulectomy¹¹. The epithelialized tract between the uterus and skin may be lined with an endometrium that undergoes atrophy due to GnRH agonist, and the tract closes due to menstruation cessation and subsequent fibrosis. Both surgical and medical management options should be discussed with the patient, and informed consent should be obtained. If any patient wants to avoid complications associated with surgery, a conservative approach with GnRH agonist is a valid option. However, this option has its limitations, and the literature review reveals that

it is successful in 50% of the cases in which it is attempted¹². In this case report, the absence of estrogen and progesterone receptors signifies the diverse therapeutic effect of GnRH agonist independent of its hormonal effect, and it needs to be researched further. Nonetheless, medical management with GnRH agonists remains a reasonable therapeutic choice for women aiming to conserve their uterus and avoid surgery.

Conclusion

Uterocutaneous fistula is a rare clinical entity and can have multiple causes, most frequently arising in the context of cesarean delivery or infection. Though diagnosis is delayed due to vague presentation, diagnostic tests like Ultrasound, MRI, and hysteroscopy can aid in diagnosis. Surgical resection of the fistulous tract can be definitive management, but in younger populations aiming to avoid surgery, medical management with GnRH agonist therapy can be a reasonable alternative.

Conflicts of Interest

No relevant conflict of interest to declare.

Acknowledgement

The authors would like to acknowledge the assistance and support of doctors and paramedical staff of the Canadian Specialist Hospital in this case report.

Funding

The authors received no financial support for this article's research, authorship, or publication.

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