CASE REPORT Uterocolon Fistula Formation in 50 Year Old Patient with History of 16 Years Intrauterine Device Use: A Case Report

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Introduction: Uterocolon fistula is one of the complications of intrauterine device (IUD) insertion. Not only may IUD materials cause perforation, but some other risk factors may contribute to its development including uterine abnormalities, thus IUD is contraindicated in patients with anatomical anomaly.

Case: P3A1 woman, 50 years old with a history of IUD use for 16 years presented with complaints of fecal discharge from the vagina 8 months ago which worsened after IUD extraction. Physical examination revealed no abdominal tenderness. Speculum examination found feces in the cervical canal. CT scan examination showed multiple uterocolon fistulas and uterine didelphys. Diagnostic laparoscopy and hysteroscopy were carried out and found a recto-uterine fistula, then the patient was scheduled for colostomy and reanastomosis with the stapler method.

Conclusion: Diagnosis was very difficult to establish despite proper imaging modalities. The use of direct visual diagnostics (hysteroscopy and laparoscopy) can be a good alternative for the diagnosis of uterocolon fistula. To the best of our knowledge, this is the first case report on recto-uterine fistula in a patient with long-term use of IUD and uterine didelphys. Keywords: uterocolon fistula, IUD, uterine didelphys, diagnostics

Introduction

Intrauterine device (IUD) is the most widely used contraception worldwide. IUD complication occurs in 18% of its user. Uterine perforation is rare with the estimated incidence between 0.2 and 3.6 cases per 1000 insertions and the incidence of uterocolon fistula is even rarer.¹

Patients with uterocolon or recto-uterine fistula may remain asymptomatic or they may present with abdominal pain, bleeding, or fecal incontinence. The exact mechanism between IUD and recto-uterine formation is not fully understood. Some authors suggest that not only may IUD materials cause perforation but some other risk factors may contribute to its development including lactation, history of recent child birth, adenomyosis, insertion of IUD by an inexperienced clinician, and uterine abnormalities.^{2,3} In this report, a spontaneous recto-uterine fistula in a 50 year old patient with 16 years of IUD use will be discussed. This case will review and evaluate the treatment and highlights the role of imaging modalities to accurately diagnose uterocolon fistula.

Case

A 50-year-old, P3A1 woman was referred to our urogynecology clinic due to malodorous fecal passage through the vagina for 6 months. The condition worsened since she had an extraction of copper T 380 mm IUD. She had IUD insertion for contraception 16 years ago. She had no history of any surgery. The patient denied any abdominal pain or fever. Upon admission, her laboratory examinations were within the normal range: Hemoglobin of 14.2g/dl, white blood count of 6600/ µL, ureum 23.9 mg/dL, serum creatinine 0.73 mg/d, CRP 0.62, and fecal analysis within normal limit. Pelvic examination

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revealed vaginal atrophy and malodorous yellowish vaginal discharge. Vulva was within normal limits. A vaginal speculum examination showed the passage of feces from the cervical canal increased with coughing and straining.

Abdominal ultrasonography found normal size uterus measuring $4.00 \times 4.51 \times 7.55$ cm with homogeneous parenchyma. No abnormality was found on the adnexa. Fecal passage through the vagina is highly suggestive of the presence of a fistula (colovaginal or uterocolon), thus after discussion with the digestive department, the patient was referred for several imaging examinations. Fistulography showed contrast via anal verge and filled rectum until distal sigmoid colon (Figure 1).

Abdominal and pelvic CT with contrast confirmed multiple rectovaginal fistulas with a diameter of 0.87 cm and a 0.34 cm communication that connected the 1/3 medial vagina to the distal rectum. Another fistula was 0.54 cm in diameter and 0.31 cm in length that connected the distal 1/3 of the vagina with the distal rectum. Rectum was normal without luminal narrowing or visible lesions. The uterine fundus was divided into 2 separate structures, suggesting uterine didelphys. MSCT scan of the abdomen and pelvis showed multiple rectovaginal fistulas, suspected uterine didelphys, and enlargement of the right paraaortic lymph node.

Colonoscopy did not show the fistula. Diagnostic laparoscopy and diagnostic hysteroscopy were performed and revealed severe adhesion between the sigmoid colon and the right posterior fundus of the uterus, the fistula was found during hysteroscopy and it connected the posterior right fundus to the sigmoid colon (Figure 2). The umbilicus was



Figure I Fistulography with contrast.



Figure 2 Diagnostic laparoscopy and diagnostic hysteroscopy.



Figure 3 Reanastomosis using intraluminal stapler with good result.

incised by 2 cm and veress needle was inserted. A hanging drop test was positive, then insufflation was conducted with CO2 gas, 14 mmHg. In this incision a 10 cm trocar was inserted. Another 5 mm trocar was inserted in the right hypogastric region. Laparoscopy was started and after peritoneum was penetrated, exploration was carried out and it appeared that the sigmoid colon had adhesions with the posterior uterus, bilateral adnexa within normal limits. After making sure there was no bleeding, the trocar from the laparoscope was removed with direct observation. Hysteroscope was then inserted through the cervical canal. On exploration, we found a uterine defect in the right posterior section with a size of $\pm 2x1x1$ cm, connecting with the colon (visible haustra in the lumen).

History taking, physical examination, and imaging findings suggested recto-uterine fistula. The patient was scheduled for elective colostomy with adhesiolysis, reanastomosis using intraluminal stapler, and hysterorrhaphy (Figure 3). This was a joint surgery between urogynecology and digestive surgeon.

Discussion

Uterine abnormality is included as a contraindication for IUD insertion. Product information for the CuT380A and the levonorgestrel intrauterine system stated that contraindication to IUD included any conditions that cause uterine cavity distortion. The report on uterine perforation in patients with uterine anomaly is limited since it is contraindicated. Interestingly some studies report successful IUD insertion in patients with uterine didelphys.^{4,5} However, the success was due to comprehensive imaging identification and careful preparation of IUD insertion, while in this case, uterine didelphys was found incidentally.

Clinical presentation of recto-uterine fistula varies between studies but mostly involved fecal passage through the vagina. Weerasekera et al reported a uterocolon fistula in a pregnant woman with retained IUD. She was asymptomatic, and no fecal passage or symptom of uterine perforation was present. Fistula was identified when she underwent abdominopelvic CT to locate the retained IUD.⁶ Another study by Robayo-Amortegui et al, reported pelvic pain and intense dysmenorrhea.⁷ Another study by Zeino et al, on a case of uterocolon fistula due to IUD perforation also reported worsening back pain as the presenting symptom.² In this case, the presenting symptom was fecal passage through the vagina. Lower abdominal or back pain was denied by the patient. This could be due to the removal of IUD in this patient. Before IUD removal, the patient also reported chronic lower abdominal pain, thus she went to a local midwife for IUD removal. Previous studies that reported pain as the presenting symptom found retained IUD intra or extra-uterine.

Colonoscopy or barium enema may be helpful in identifying colonic lesion such as diverticulosis, however it inaccurately detects recto-uterine or uterocolon fistula due to the small size of the fistula. Thus we performed pelvic CT scan and fistula.^{8,9} Kassab et al reported a uterocolon fistula identified by MRI and the extension of the fistula relative to the adjacent organ was delineated on T1-weighted images. Another imaging modality using charcoal challenge test may show the orally-administered charcoal flowing through the cervical os and could be used to diagnose uterocolon fistula; however, this method does not identify the fistula tract or the exact fistula site.⁹ Another study by Choi et al demonstrated that sonohysterography with contrast medium was able to visualize the fistula tract, the uterine wall, and the sigmoid colon in a patient with uterocolon fistula. Furthermore, the use of ultrasound prevented patient from X-ray exposure.¹⁰ In our case, CT findings suggested the diagnosis of a uterocolon fistula, while fistulography and other imaging studies are still not clear. To confirm the diagnosis the patient underwent a hysteroscopy and laparoscopy procedure and the result showed a uterocolon fistula.

The management of uterocolon or recto-uterine fistula requires surgery. The data supporting hysterectomy in benign conditions are still limited. Colon resection and drainage is sufficient in most cases. Additional reanastomosis after resection and end colostomy have been the most effective procedures in such patients.¹ In our case the patient underwent colostomy followed by reanastomosis with stapler method.

The recommended duration for copper T 380 mm IUD usage is maximum of 5–10 years. Systematic review by Ti et al, evaluated the safety of extended IUD duration and found expulsion rate of 1.3 per 100 participants in years 11–12 in one study but did not finf perforations or cases of pelvic inflammatory disease in years 11–12. Any gynecological symptom in patients with IUD should be evaluated for IUD-related complication and IUD removal should be done if necessary. However, in this case the patient did not seek medical treatment until she had her symptom for 6 months. The painless nature of uterocolon fistula may contribute to patient negligence.¹¹

A secondary reanalysis of the EURAS data in 2021 found that the different copper content, design, and size were associated with different rates in bleeding, pain, expulsion and continuation rates between devices. Larger size IUD (including 380 mm IUD) was more likely to cause adverse events than the smaller size IUD. Timing of IUD insertion was also associated with perforation rate. The risk of perforation increases if IUD adverse events include anatomic abnormalities, such as cervical stenosis and a retroverted uterus. However, a study by Tepper et al, evaluated 19 case reports or case series of IUD in women with anatomic anomaly and could not make a conclusion on specific patient characteristics or uterine abnormalities for which an IUD might be safe. Despite limited evidence, the UKMEC guideline 2016 recommended that in such patients with known distortion of the uterine cavity the risk associated with increased risk of perforation, expulsion or malposition. In this case, uterine didelphys was found incidentally upon CT scan examination. In Indonesia, IUD insertion can be done without prior imaging of the uterus, thus patients with anatomical anomaly remain unidentified.¹²

The limitation of this report was that we did not follow-up the patient after surgery and detailed discussion on the surgical method. However, this case report highlights the entity and diagnostic modality for such a rare case in urogynecology.

Conclusion

Uterocolon fistula in patients with IUD is very rare. Despite good imaging modality, the diagnosis remains challenging. Operative diagnostic procedures (hysteroscopy and laparoscopy) can be a good alternative for diagnosis. Risk factors that may have been involved in the pathogenesis in this case were IUD use duration and uterine didelphys. To the best of our knowledge, this is the first case report on rectouterine fistula in a patient with long-term use of IUD and uterine didelphys.

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Consent for Publication

The patient voluntarily participated in this study. Written informed consent has been provided by the patient to have the case details and the accompanying images published. The patient agreed that she had been given the opportunity to ask questions and had them answered satisfactorily. Subject had been notified that her imaging findings would be published for scientific purpose. Patient has received a copy of consent form signed by the researcher. Institutional approval was not required by our institution regarding the case publication.

Disclosure

The authors report no conflicts of interest in this work.

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