

Different etiologies of an unusual disease: Colouterine fistula - Report of two cases

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Abstract

Colouterine fistula is an extremely rare condition, as the uterus is a thick, muscular organ. Here, we present two different etiologies for this rare condition—diverticulitis and malignancy. A 77-year-old female with colouterine fistula due to diverticulitis presented with complaints of lower abdominal pain localized particularly in the left iliac fossa and fever. Another case was of 73-year-old female with colouterine fistula due to malignancy who presented with abdominal pain, blood in stools, and whitish discharge from vagina. Both cases were evaluated with contrast-enhanced computed tomography (CECT). The presence of air and fluid within the uterus on ultrasound or CT scan, prompts the possibility of colouterine fistula with CECT providing accurate preoperative assessment.

Key words: Colon; diverticulitis; fistula; malignancy; uterus

Introduction

Colouterine fistula is very rare condition with only around 25 reported cases of post-diverticulitis colouterine fistula so far.^[1-6] Other causes include sigmoid malignancy, radiotherapy, and iatrogenic conditions such as insertion of intrauterine devices, endometrial curettage with uterine, and bowel perforation, or obstetrical injury.

The most common type of fistula associated with colon is colovesical fistula followed by a colovaginal fistula arising from the sigmoid colon. Because uterus is a thick muscular organ, it provides a protective barrier against invasion of benign or malignant disease. Therefore, a colouterine fistula is an extremely rare. Here, we present two cases of a colouterine fistula caused by diverticulitis and malignancy of sigmoid colon.

Case Reports

Case 1

A 77-year-old female came to the hospital with complaints of lower abdominal pain localized particularly in the left iliac fossa. On initial examination, she was febrile (100.4°F). The abdomen was distended with ill-defined palpable mass in the left lower abdomen. The pelvic examination showed a spontaneous malodorous discharge in the vagina and cervical os. She had leukocytosis (14,500/mm³) and raised erythrocyte sedimentation rate (ESR) (40 mm/h).

She underwent contrast-enhanced computed tomography (CECT) scan which revealed a large collection adjacent to the sigmoid colon. The collection was seen abutting the uterus with air-fluid level within the uterine cavity [Figure 1].

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Cite this article as: Aggarwal R, Indiran V, Maduraimuthu P. Different etiologies of an unusual disease: Colouterine fistula – Report of two cases. Indian J Radiol Imaging 2018;28:37-40.

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<p>Quick Response Code:</p> 	<p>Website: www.ijri.org</p> <p>DOI: 10.4103/ijri.IJRI_172_17</p>

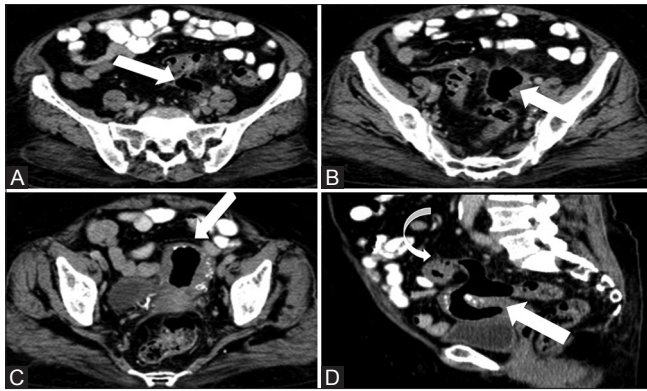


Figure 1 (A-D): Contrast-enhanced abdomen and pelvis CT scan (A and B) Axial images show air filled collection adjacent to sigmoid colon. (C) Axial image shows air within the endometrial cavity. (D) Sagittal reformat shows air filled collection extending between the air filled endometrial cavity (straight white arrow) and the thickened sigmoid colon (curved white arrow)

Based on CT findings, possibility of sigmoid diverticular abscess and colouterine fistula was considered. The patient underwent abscess drainage, hysterectomy, and colostomy. About 2 cm defect was noted in the posterior uterine wall communicating with the abscess and sigmoid colon. The patient improved clinically following surgery.

Case 2

A 73-year-old female patient came to the outpatient department with complaints of lower abdominal pain, blood in stools, and whitish discharge through vagina for 10 days. On examination patient's vitals were stable. The patient underwent a CECT scan abdomen which revealed irregular wall thickening involving the rectosigmoid region for a length of ~ 7 cm, with wall thickness of ~ 18 mm. There was loss of fat plane between the uterus and the thickened rectosigmoid colon with air pockets within the endometrial cavity [Figure 2]. Mesocolic fat and perirectal fat stranding was also seen with multiple adjacent subcentimetric lymph nodes. Based on CT findings, possibility of rectosigmoid malignancy and colouterine fistula was considered. The sigmoidoscopy and biopsy of the lesion revealed infiltrating moderately differentiated adenocarcinoma of colon. The *en bloc* resection of the uterus and sigmoid colon with colostomy was performed on the patient and adjuvant chemotherapy started.

Discussion

Colouterine fistula was first reported by Lejemtel in 1909.^[7] Three main etiologies were described at that time were uterine trauma, abscess rupture into the bowel, and the uterus and uterine or sigmoid carcinoma. Later radiotherapy was also identified as one of the etiological factors. Although, fistulas are usually caused by injury or surgery, they may also form after an infection which led to severe inflammation and decompresses by perforating into

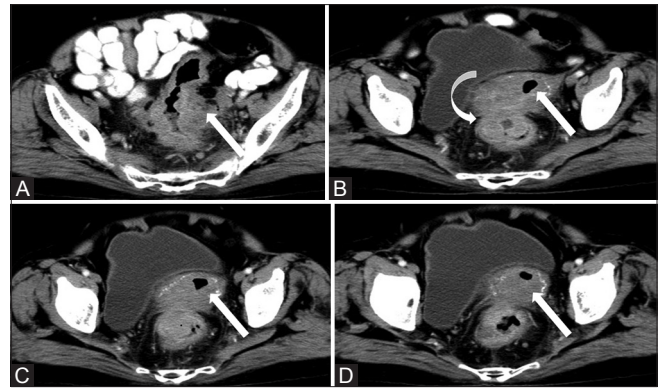


Figure 2 (A-D): Axial images of contrast-enhanced abdomen and pelvis CT scan (A) shows heterogeneously enhancing circumferential irregular wall thickening involving the recto-sigmoid region. (B-D): Straight white arrows show air pocket in the endometrial cavity. Curved arrow in B shows thickened rectum abutting the posterior wall of lower uterus with no fat plane in between

an adjacent viscus, or through the skin. The fistulation occurs between the colon and the urinary bladder (colovesical fistula) in 65% of the cases and between the colon and the vagina (colovaginal) in 25% of the cases. Colouterine fistula is a rare complication of diverticulitis of the colon.^[7]

Diverticular disease of the colon is common in developed nations and thought to result from structural abnormalities of the colonic wall, disordered intestinal motility, or deficiencies of dietary fibers. The signs and symptoms of diverticulitis include fever, abdominal pain, and leukocytosis. As per the data recorded among all the nations, there is high prevalence rate of left-sided diverticulosis.

Besides the usual signs and symptoms, in the course of the diverticulitis, several unusual complications like pylephlebitis, perforation, intestinal perforation, abscess, and fistula formation may be encountered.^[8] While colovesical fistula is the most common type of fistula associated with diverticulitis of the colon occurring in 2–22% of patients with known diverticulitis, colouterine fistulas are a relatively uncommon entity arising in the setting of the disease.^[9] The rarity of the condition is probably explained by the fact that the uterus is a thick muscular organ, which poses obstacles for invasion for both benign and malignant disease.

Colon cancer, the most common type of gastrointestinal cancer, is a multifactorial disease process, with etiology encompassing genetic factors, environmental exposures (including diet), and inflammatory conditions of the digestive tract. Among the cancers of the colon, the most common type is adenocarcinoma. As colon cancer grows and spreads beyond the colon mucosa, it is called invasive or infiltrating adenocarcinoma. Metastases to the female genital tract from extragenital malignancies are very uncommon, and the most common extragenital primary sites are breast and gastrointestinal system.^[7,8] The common

metastatic sites of colorectal cancer include the liver, lung, lymph nodes, and peritoneum, with uterus being a rare site. Mazur *et al.*^[10] reported that the endometrium was the metastatic site for colon and rectum carcinoma in only 3.6% of the 56 cases in their study.

Patients with the colouterine fistula usually present with malodorous fecal or purulent vaginal discharge as the colonic lumen and the uterus are linked by a fistula tract.^[2] The most common presenting symptom of uterine metastasis is vaginal bleeding whereas in cases of diverticulitis causing fistula, patients may present with fever and recurrent abdominal pain.

Many imaging modalities have been used for diagnosing colouterine fistula, but computed tomography (CT) is being increasingly used nowadays for evaluation of acute abdomen and diagnosing intraperitoneal and retroperitoneal lesions. In a colouterine fistula, air bubbles in the uterine cavity and colonic wall joined together may be identified on CT, but limitations are there as it may fail to demonstrate the fistulous tract. However, CT and magnetic resonance imaging (MRI) may play an important role in preoperative surgical planning.

Ultrasound is the preferred initial diagnostic modality for gynecologic disease and captures the real-time images of organs and blood flow without radiation hazards. Additionally, ultrasound is almost always the first modality used to analyze the endometrium and to guide further diagnostic work-up in these patients, such as endometrial biopsy. Takada *et al.*^[11] reported diagnosis of colouterine fistula by sonohysterography with contrast medium using it to track and diagnose the fistula tract.

Kassab *et al.*^[12] reported that MRI could be a versatile, non-invasive, detailed, and accurate diagnostic modality comparable to CT scan. In their report, MRI was used to identify fistulae, and T1-weighted images portrayed the extension of the fistula relative to the adjacent organ and showed inflammatory changes in fat planes. T2-weighted images and diffusion weighted images can depict the collections and abscesses better.

Beattie *et al.* suggested that the use of multidetector CT (MDCT) allows excellent multiplanar reconstructions and improves visualization of pathology with shorter acquisition time.^[13] If a positron emission tomography/computed tomography (PET/CT) scan is performed, an increased activity in the endometrium is often expected in cases of carcinoma, but sometimes infection or inflammatory change associated with diverticulitis may also raise standardized uptake values (SUV).^[14]

Additionally, the 'Charcoal Challenge Test' has also been described in diagnostic aid. This test helps in diagnosing

colouterine fistula through orally administered activated charcoal which passes across the bowel lumen into the cervical os, but it does not tell us the exact site of the fistula.^[15] Diagnostic hysteroscopy is also considered a very good choice for investigating patients with malodorous vaginal discharge, enabling direct visualization, and biopsy of the pathological area.^[1]

Most cases of colouterine fistula need surgical management. Various surgical approaches have been reported. An *en bloc* resection,^[2] according to the patient's condition, is mandatory in cases of colouterine fistula caused by malignancy. However, in benign conditions, the need for a hysterectomy has not been established. Resection of the colon alone and drainage of the purulent uterine lesion may be sufficient for definite treatment. In cases of colouterine fistula due to diverticulitis, a two-stage procedure involving resection and end colostomy, followed by reanastomosis later is most effective. Therefore, selection of an appropriate surgical approach according to the patient's condition is necessary.

Conclusion

Colouterine fistula is a rare complication of diverticulitis of colon and malignancy of colon. It should be suspected when patient presents with malodorous discharge from vagina. It can be diagnosed by air and fluid within the uterus on ultrasound or CT scan. However, CT scan is essential for an accurate preoperative assessment. The surgical treatment is indicated in almost all patients except in very high risk cases.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

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