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Colouterine fistula: A case report of a rare complication of diverticular disease managed during the pandemic

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ABSTRACT

INTRODUCTION AND IMPORTANCE: Colouterine fistulas related to diverticulitis are very rare due to the thickness of the uterine myometrium. Other causes related to colouterine fistula formation particularly malignancy, have to be considered. Diagnosis by imaging or endoscopy may be inconclusive.

CASE PRESENTATION: We are presenting a case of a 70-year-old female who presented with malodorous vaginal discharge and painful labial lesions. No previous history of surgery, gynecologic malignancy or other possible causes of the fistula was elicited. CT scan imaging suggested a colouterine fistula. The patient was admitted and underwent Exploratory laparotomy, Hartmann's procedure and total hysterectomy with bilateral salpingo oophorectomy. The patient was discharged without perioperative complications.

CLINICAL DISCUSSION: Colouterine fistulas are extremely rare complications of diverticular disease. Diagnosis entails clinical astuteness and judicious use of imaging and endoscopic modalities. Accurate diagnosis is essential to select the appropriate surgical approach, along with intraoperative findings patient status and prevailing conditions.

CONCLUSION: This case is being presented not only for the rarity of the case but also for the complexity of the management and decision making during the period of the pandemic.

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1. Introduction

Colonic diverticular disease is fairly common in Western countries. In recent years, the incidence of diverticulosis in Asian countries has been rising and this has been attributed to changes in dietary patterns mirroring that of the West. The complications that arise from inflamed diverticula may at times require surgery and include hemorrhage, abscess, perforation, or fistula formation. Fistulas resulting from diverticular disease are more commonly colovaginal or colovesical. Colouterine fistulas due to the diverticulitis are extremely rare that and very few cases have been reported in the literature worldwide. Colouterine fistulas from other causes are more common and include spontaneous rupture of a gravid uterus, obstetric trauma, pelvic malignancy and radiation.

Colouterine fistulas present similarly with colovaginal fistulas but the latter almost always occur after hysterectomy. Almost all patients present with foul-smelling, hemorrhagic, purulent, or feculent vaginal discharge. Some may present with vague or at times well delineated abdominal pain. Patient complaints at initial presentation vary widely from minimal symptoms to florid sepsis. It is at times difficult to come up with an accurate diagnosis given the unreliability of imaging and other diagnostic tests. The clinician must always be aware of the possibility of uncommon fistulas complicating the fairly common diverticular disease to come up with an appropriate treatment plan.

This case is being presented not only for its rarity but also for the decision-making challenges the management of such a complicated case during the pandemic entails. This report was written in line with the Statement: Updating Consensus Surgical Case Report Guidelines (SCARE) [1].

2. Presentation of a case

This is the case of a 70-year-old female who presented in our institution, a tertiary care, university-based hospital, complaining of a 4-month history of a foul-smelling vaginal discharge. This was accompanied by intermittent left lower quadrant pain

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associated with episodes of loose bowel movement. The patient self-medicated with analgesics affording temporary relief. She also complained of occasional constipation. A month prior to admission, she noted increased severity of left lower quadrant abdominal pain and loose bowel movement. She was subsequently admitted at a local hospital where she was given analgesics and antibiotics which afforded partial relief of symptoms. Colonoscopy was performed with no note of masses, polyps or bleeding lesions. A spastic segment of colon was noted 20–30 centimeters from the anal verge. She was discharged with relief of symptoms. 2 weeks later, patient developed fecaloid foul-smelling discharge per vagina requiring her to wear diapers. There was no note of abdominal pain, fever, pneumaturia or fecaluria. She was subsequently admitted by the Gynecology service and referred to General Surgery for further management.

The patient has a previous diagnosis of hypertension of 10 years duration and chronic renal disease for which she was taking maintenance oral anti-hypertensives. She has had no previous surgeries. The patient had normal vital signs on admission. Physical examination revealed a vague palpable left lower quadrant mass which was non tender on palpation, an erythematous labial area and fecaloid material on internal examination. Digital rectal exam revealed no masses or any palpable lesions.

Admitting impression was Rectovaginal fistula probably secondary to chronic diverticulitis vs malignancy.

Transvaginal ultrasound was requested and revealed the following findings:

Anteverted uterus with regular contour and echo pattern. There is a well-circumscribed hypoechoic mass (nodule) within the left anterofundal intramural subserous area $0.85 \times 0.89 \times 1.19$ cm. The endometrium is dilated to 0.92 cm containing jelly-like heterogeneous structures with echogenic debris, representing fluid (pus). Occupying the left adnexa is a complex mass probably enlarged left fallopian tube, measuring $4.03 \times 2.66 \times 1.92$ cm. It envelops the left ovary wherein the latter is visualized, as measured but cannot be separated from it. The interstitial segment is dilated to 0.91 cm containing heterogeneous, jelly-like structure with echogenic debris continuous into the endometrial cavity. No tenderness was elicited on probe manipulation. This was interpreted by the sonologist as possible pyometrium.

A lower abdomen Computed Tomography (CT) scan with intravenous and rectal contrast was likewise requested. It revealed circumferential mucosal wall thickening of the sigmoid with a greatest axial thickness of about 1.2 cm. Irregular contrast-filled sigmoid lumen with some areas of narrowing is seen. Adjacent mesenteric fat strandings are noted. A faintly contrast-opacified narrow tract from the sigmoid lumen into the endometrial cavity is noted with a length of 1.0 cm. The uterus exhibits heterogeneous enhancement with its walls partially indiscernible or inseparable from the sigmoid. Moreover, there is spillage of rectal contrast and air into the endometrial cavity (Figs. 1–3). The rest of the opacified bowels, urinary bladder, and visualized ureter segments are unremarkable. The working impression for this patient was thus revised to Colouterine Fistula probably secondary to diverticulitis.

Preoperative laboratories for cardiopulmonary risk assessment revealed no significant abnormalities. Though the patient was diagnosed previously as having chronic hypertensive renal disease, laboratory tests for renal function were within acceptable limits. Chest x-ray revealed right upper lobe pulmonary tuberculosis of undetermined activity and mild right lower lobe pneumonia. COVID 19 Reverse transcription Polymerase chain reaction testing (RT-PCR) yielded negative result. The patient subsequently consented for surgery.

She underwent an exploratory laparotomy via a midline incision under general anesthesia. There were findings of a consolidated mass at the left lower quadrant consisting of the sigmoid adherent

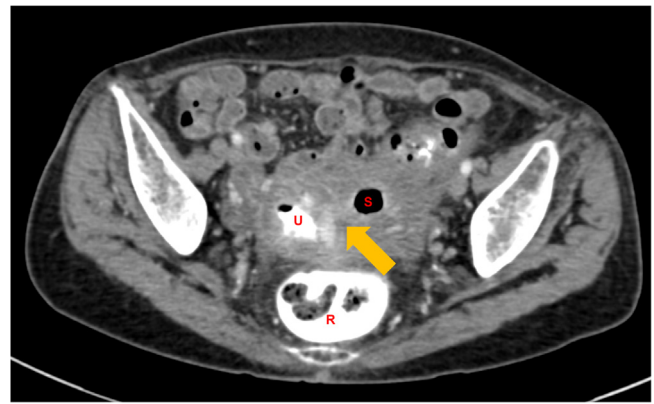


Fig. 1. CT scan demonstrates via transversal view the faintly opacified fistulous tract between the Uterus (U) and the sigmoid (S). Rectum is marked (R) for reference.

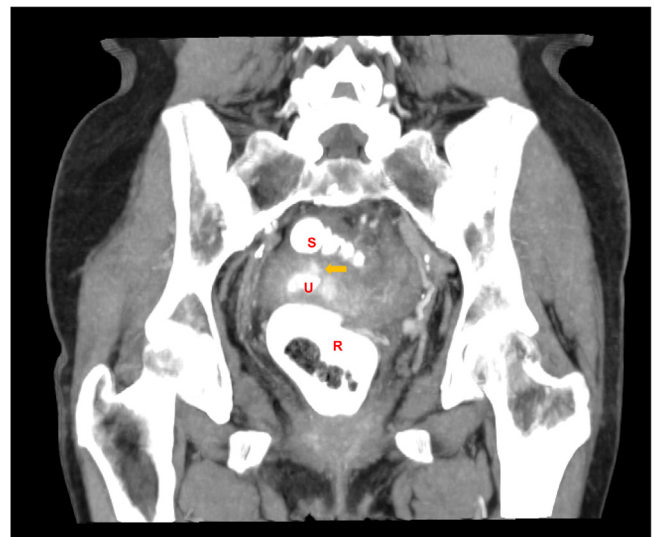


Fig. 2. CT scan, coronal view, demonstrating the fistulous tract between the uterus (U) and the sigmoid (S).

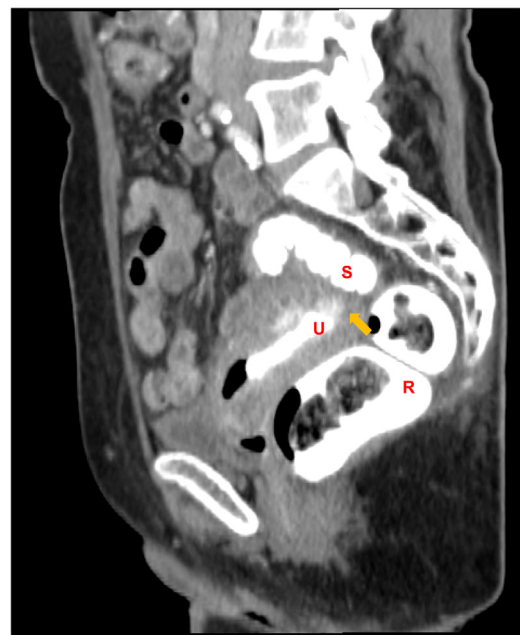


Fig. 3. CT scan, sagittal view, demonstrating the same fistulous tract evident on transversal and coronal views.

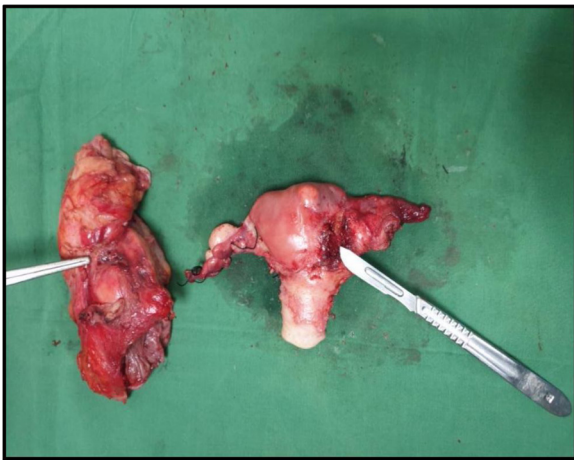


Fig. 4. Photograph of the specimens showing the excised sigmoid with an instrument being inserted into the defect and the resected uterus with the tip of the scalpel pointing at the perforation on the uterine side.

to the uterus enveloped by omentum from the transverse colon, resulting in a phlegmon. After adhesiolysis, the dissection freed the sigmoid colon and upper rectum, with note of minimal abscess draining from the phlegmon. The connection between the sigmoid and the uterus was disrupted, revealing the defects both at the sigmoid side and the corpus of the uterus. The surgical team proceeded with resecting the diseased sigmoid and performing a Hartmann's procedure. The operation was performed by a senior attending general surgeon assisted by 2 senior residents as dictated by hospital policy during the pandemic. The Ob-gyne service completed the procedure by performing a total hysterectomy and removal of the adnexae. A closed suction drain was used but was eventually removed prior to sending the patient home after 6 days. There were no perioperative complications and the patient was sent home with home care instructions, including stoma care.

Gross examination of the specimen demonstrated a fistula connecting the sigmoid to the uterus with no masses seen on the cut specimen (Figs. 4–7).

Final histopathologic report revealed: Diverticulosis with acute on chronic diverticulitis. Acute on chronic transmural inflammation with focal abscess. Viable surgical margins. One reactive lymph node and Leiomyoma uteri, subserous. Acute on top of chronic myometritis. Chronic cervicitis. Focal mild chronic salpingitis, left. Unremarkable bilateral ovaries and right fallopian tube. Final pathologic diagnosis was Colouterine fistula secondary to Diverticulitis.

3. Discussion

Colonic diverticular disease is fairly common, particularly in the older age groups. Diverticulosis is a very common colonic pathology occurring in more than 65% of the population by the age of 80. It involves the sigmoid colon in majority of cases. Fistula formation may complicate diverticulitis but is seen in only 17–27% of patients requiring surgery. The most common types of fistulas are colovesical fistulas (65 percent) and colovaginal fistulas (25 percent) [2–9]. Fistulas to unusual adjacent sites and organs have been documented and, in some reports, multiple pelvic organs have been involved [10–13]. Colouterine fistulas due to diverticulitis however are so rare that the literature only consists of case reports numbering less than 30 worldwide. Several institutions reporting large series of surgically managed diverticular disease spanning decades report only 1 or 2 cases at most of colouterine fistula as a complication [14–20]. This is the first published reported case in the Philippines of a colouterine fistula due to diverticular disease. The



Fig. 5. The uterus with an instrument inserted through the defect demonstrating the perforation through all layers of the uterus.



Fig. 6. Photograph of the cut specimen demonstrating the perforation and absence of any associated intraluminal mass lesions.

pathophysiology of fistulas includes destruction of two serosas of two epithelialized surfaces in close proximity. Several mechanisms have been theorized on the formation of fistulas between the colon and the uterus. It has been proposed that the inflammatory adhesion of the bowel wall to the uterus presumably results in necrosis and subsequent fistulization. Fistulas may also develop after localized perforations of diverticula and eventual erosion of the pericolic



Fig. 7. The sigmoid with an instrument passed through the perforation with surrounding remnants of adherent tissue forming part of a phlegmon.

abscesses into neighboring hollow viscera. The thickness of the uterine wall apparently affords protection and must be responsible for the low incidence of colouterine fistulas. This attribute of the uterus precludes invasion even from malignancies. Other causes of colouterine fistulas have been documented to be more common and these include obstetric and instrumentation trauma, malignancy, irradiation and rupture of a gravid uterus [20]. Thus, a thorough history taking and a complete physical exam is thus essential in arriving at an accurate diagnosis.

Colouterine fistulas present similarly with colovaginal fistulas. Despite the similarities, colovaginal fistulas almost always occur after a hysterectomy. All patients present with a history of foul-smelling, hemorrhagic, purulent, or feculent vaginal discharge. Some may also present with lower abdominal pain and other symptoms of acute diverticulitis. Presentation may range from asymptomatic to florid sepsis. Findings on physical examination may be unremarkable or may demonstrate a pelvic mass. In some patients, abdominal examination is suggestive of a diverticular disease, particularly when there is active diverticulitis, phlegmon or frank abscess. Vaginal speculum examination typically reveals the aforementioned vaginal discharge per os in contrast to a colovaginal fistula which typically drains from an opening at the apex of the vagina [18].

Our patient presented with a malodorous vaginal discharge which she has been complaining of for several months before admission. She has been wearing adult diapers to keep dry and to mitigate the excoriation on the skin. On physical exam, she has erythematous labia and there was a vague palpable mass at the left lower quadrant of the abdomen which was tender on palpation. In the absence of previous surgery and based on her age, malignancy is always a consideration, particularly in our patient who is post-menopausal.

Most patients such as ours are evaluated with endovaginal ultrasound and/or dilation and curettage with hysteroscopy to exclude malignancy. Having the benefit of a transvaginal ultrasound showing pyometrium and the demonstration of a fistula on CT scan, it was decided by the surgical team to forego with other gynecologic

procedures like dilatation, curettage and hysteroscopy. Cervical dilation is indicated to improve uterine drainage in the presence of pyometrium but considerations brought about by the pandemic warranted decreasing the number of procedures and limiting risk of exposure both to patient and health care workers. Other series suggested the usefulness of other imaging modalities including magnetic resonance imaging (MRI) for the diagnosis of a colouterine fistula. Although CT scanning is both sensitive and specific in making a diagnosis of diverticulitis, detection of a fistula tract or differentiation of complicated diverticulitis from colon cancer may be limited in CT. Fortunately, it was highly informative in our patient. CT scanning can also be useful in preoperative surgical planning by demonstrating the extent and degree of pericolic inflammation. Magnetic resonance imaging is versatile, noninvasive, and is a more sophisticated, detailed and accurate diagnostic tool compared to CT scanning. It can be used to identify fistulae and at times may provide delineation of the extension of the fistula relative to the adjacent organ, inflammatory changes in fat planes. Fluid collections, and inflammatory changes within muscles. MRI was deemed no longer necessary in our patient given the results of the colonoscopy the CT scan and the transvaginal ultrasound [21–27].

In some reports, barium enema and colonoscopy have been used but they are not expected to demonstrate the fistula. They may however be useful in documenting diverticular disease as possible etiology of the fistula and to rule out colonic malignancy. Some authors have reported a low-cost method of documenting a colouterine fistula by having the patient ingest charcoal and tracking its flow out of the cervical os [22]. While this may be highly suggestive of a colouterine fistula, it will not demonstrate the tract or the site of the fistula.

Surgery is the definitive treatment for colouterine fistula and usually entails the resection of the sigmoid, being the origin of the fistula [13–20]. The management of the sigmoid is thus fairly straightforward but the uterus is a bit more complicated. The fistula usually involves the fundus of the uterus and the sigmoid colon [17], as is the case with our patient. Various options regarding the management of the uterus have been proposed. In cases where a malignant etiology cannot be excluded, or where the colon and uterus cannot be separated because of chronic inflammation, an en-bloc hysterectomy can be performed. On the other hand, if the colon can be easily dissected off the uterus, and the suspicion for a malignant process is low given the preoperative work up and intra-operative assessment, the uterine fistulous opening can be closed and the uterine cavity drained via the os. Since diverticular disease usually occurs in the older age groups, the atrophic uterus may usually be removed, avoiding the need for further drainage post operatively [13–20,28–30].

In our patient, surgery was performed and, at laparotomy, a chronic inflammatory mass was observed. This mass included the whole sigmoid colon and the posterior uterine wall. The initial dissection was carried out with an attempted en-bloc resection. On manipulation however, the sigmoid was separated from the uterus and purulent material was drained from the phlegmon. Having separated the sigmoid from the uterus, the surgical team proceeded with a Hartman's procedure. The team decided against primary anastomosis due to the presence of a localized abscess, the questionable overall nutritional status of the patient, the need for expedited treatment during the pandemic, and an aversion to monitoring an elderly patient where an anastomotic complication may prove catastrophic during the pandemic. In the presence of colonic obstruction, paracolic abscess, or severe inflammatory reaction, a two stage approach is safer, but a single-stage resection and anastomosis is reasonable given favorable conditions. Since colonoscopy has established the absence of intraluminal masses or lesions, colonic malignancy as the cause of the fistulization has been virtually ruled out. Although there were no gross uterine tumors in

the CT scan, the gynecologic team decided to proceed with a hysterectomy to manage the uterine side of the fistula. This was to ensure proper histopathologic examination of the diseased uterus and obviate the need of leaving behind an infected uterus requiring suturing of the perforation and possible percutaneous or surgical drainage. Some authors have claimed that excising the source of the pathology, the diseased sigmoid colon is necessary but not the uterus. Others even suggested simple percutaneous drainage of the uterus after colonic diversion. Some even advocate just simply primarily suturing the colon after separating the viscera closing the uterine perforation. More surgeons prefer resection to examine the complete specimen for the purpose of diagnosing potential malignancies, both on the sigmoid and the uterine side [30–32]. Some authors have pointed out that fistulas involving the colon in elderly patients are more likely to be managed conservatively than in younger patients. This may be the case particularly given conditions like the pandemic. In some cases reported in the literature however, conservative management has led to death. Some institutions report successfully managing even complicated diverticulitis with fistula using laparoscopic surgery [31–33]. Lack of clear guidance on the safety of laparoscopic surgery and the aerosol generating nature of the procedure during the early part of the pandemic dissuaded the surgical team from this approach.

4. Conclusion

Colouterine fistulas as a complication of colonic diverticular disease are extremely rare. Accurate diagnosis of a colouterine fistula requires correlation of clinical presentation, properly selected radiologic examination and endoscopic evaluation. Colouterine fistulas from other causes must be considered to properly select appropriate management. Colouterine fistulas presenting similarly to the case presented must raise the possibility of a colonic or gynecologic malignancy and thus the surgical team must be prepared to perform an en bloc resection for malignant disease. The surgical treatment of choice will be dictated by patient factors, local tissue conditions and available expertise. This case was presented to add to the scarce body of literature on this rare entity and will hopefully assist surgeons handling similar cases beyond the pandemic.

The patient expressed gratitude for the relief of symptoms afforded by the surgery. Having been made to understand the decision involved resulting in a colostomy, she is amenable to undergo the reversal when risks associated with the pandemic are lowered to acceptable levels.

Declaration of Competing Interest

The authors report no declarations of interest.

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Ethical approval

Ethics approval obtained from University of the Philippines Ethics Review Board.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution

Anthony R. Perez: Study concept, writing the paper.

Mary Ellen Perez: Data collection, study design.

Crisostomo Arcilla Jr.: Writing and editing the paper.

John Isaac Merin: Data collection, review of literature, final draft.

Registration of research studies

Not applicable.

Guarantor

Anthony R. Perez.

Provenance and peer review

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