



Successful Treatment of Double Malignant Fistulas in Advanced Rectosigmoid Junction Tumor with Neoadjuvant Chemoradiation and Surgery: A Case Report

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This case report describes a successful treatment of rare double malignant fistulas in an advanced rectosigmoid junction tumor using neoadjuvant chemoradiation and surgery. An 81-year-old woman presented with poor appetite, abdominal fullness, dizziness, and bloody stool. She was diagnosed with an advanced rectosigmoid junction tumor with double malignant fistulas (ceco-sigmoid and ileo-sigmoid fistulas). The patient received neoadjuvant chemoradiation therapy followed by surgical resection of the tumor and the fistulas. The patient's postoperative recovery was uneventful. Her pathological examination showed a partial pathological response to the neoadjuvant therapy. The patient had no evidence of local recurrence or distant metastasis at her 1-year follow-up. This case demonstrates the importance of a multidisciplinary approach in managing complex cases and suggests that neoadjuvant chemoradiation therapy combined with surgery may be an effective treatment for advanced rectosigmoid junction tumors with double malignant fistulas.

Key words: Double malignant fistulas, neoadjuvant chemoradiation, rectal-sigmoid colorectal cancer

INTRODUCTION

The incidence of malignant gastrointestinal tract fistulas is infrequent, and the presentation of double fistulas at one time is even rarer. This condition lacks a well-established treatment approach, making management challenging for healthcare providers.

Here, we report a case of an elderly woman with an advanced rectosigmoid junction tumor complicated with double fistulas. Concurrent chemoradiotherapy (CCRT) was delivered initially to the unresectable pelvic tumors, and surgical resection was successfully undertaken after favorable tumor regression.

CASE REPORT

An 81-year-old female presented to our Emergency Department with a 3-month history of poor appetite, abdominal fullness, dizziness, bloody stool, frequent midnight urination, and general fatigue. Her medical and surgical histories were

unremarkable. Her primary care physician found severe anemia, and she was referred to our hospital. Her physical examination revealed abdominal fullness and a palpable mass in the hypogastric area with localized tenderness but without muscle guarding. There was no fever or other infectious sign or symptom. Her blood work revealed a hemoglobin of 4.5 g/dL, a serum potassium of 2.8 mmol/L and C-reactive protein (CRP) of 8.44 mg/dL. Other routine laboratory investigations were within normal limits. She was given a blood transfusion and potassium supplementation. Her chest and abdominal X-rays showed no apparent findings. Her colonoscopy demonstrated a circumferential ulcerative mass larger than 10 cm, irregular folds, and a lumen that narrowed in the rectosigmoid junction. In addition, a fistula between the sigmoid colon and the ilium was noted. A well-differentiated adenocarcinoma of colonic origin was histologically proven. Her abdominal computed tomography (CT) revealed a long segmental mass in the pelvic

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inlet involving the sigmoid colon, ileum, and cecum with two fistula tract formations (ceco-sigmoid and ileo-sigmoid fistulas) [Figure 1a-c]. No distant metastases were found in the other imaging studies. The clinical staging for this rectosigmoid junction tumor was cT4bN1bM0, Stage IIIB.

The patient received conservative treatments to address unbalanced metabolism, primarily focusing on hypokalemia, and poor nutritional status. Since there were no symptoms of bowel obstruction present, a colostomy was not performed. During cancer staging, the patient's CRP level decreased to 2.87 mg/dL without the use of antibiotics. The elevated CRP level was suspected to be cancer-related, as there were no signs or symptoms of infection observed. Given the unresectable status judged by our surgeon, neoadjuvant 5-fluorouracil-based CCRT was administered. The radiotherapy regimen was 45 Gy to the pelvis in conventional fractionation [Figure 2]. The patient tolerated CCRT well

except for Grade 2 body weight loss and fatigue, which improved after supportive care.

Follow-up colonoscopy and radiologic studies showed marked tumor regression [Figure 1d]. The ceco-sigmoid fistula was not visible on the follow-up CT, but the ileo-sigmoid colon fistula remained. Exploratory laparotomy with a low anterior resection, right hemicolectomy, total abdominal hysterectomy, and bilateral salpingo-oophorectomy was performed 12 weeks after she completed CCRT. The pathological staging was ypT4bN0M0, Stage IIC with a tumor regression score of 2 by the modified Ryan scheme [Figure 3]. Oral tegafur-uracil was administered as adjuvant chemotherapy. One year after surgery, the patient is alive without the development of recurrence.

DISCUSSION

Fistulas between the gastrointestinal tract are uncommon and can be caused by benign diseases, such as diverticulitis (the most prominent causative factor), Crohn's disease,¹ and ulcerative disease. Malignant fistulas are rare and can be due to various origins, including intestinal lymphoma,² carcinoid tumors, and carcinomas.³ Various types of malignant fistula have been reported, including enterocutaneous fistulae,⁴ colo-vesical or colo-vaginal fistulae,⁵ ileo-rectal fistulae,³ ceco-sigmoid fistulae,⁵ colo-duodenal fistulae,⁶ sigmoid-duodenal fistulae,⁷ or gastro-colic fistulae.⁸ Most cases present with a single fistula between hollow organs, resulting in their clinical manifestation. The presence of double fistulas is a very unusual complication of malignant disease and is rarely described in the literature.

Malignant fistulas can form in two ways. The first is that the neoplasm growth directly invades an adjacent organ(s).³ The other is caused by inflammation secondary to the tumor-related deep ulceration, resulting in adherence to a nearby hollow organ(s), and finally, perforation.^{3,8} Symptoms of gastrointestinal fistula are usually nonspecific and include weight loss, diarrhea, gastrointestinal bleeding, and abdominal pain. Fecal vomiting is a classic symptom if the fistula

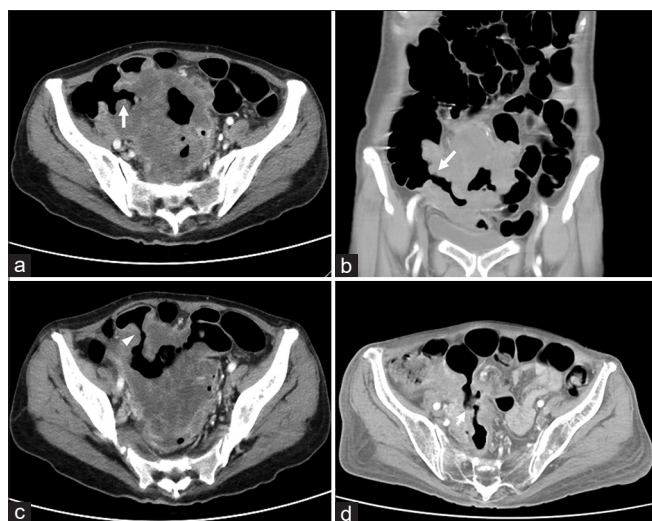


Figure 1: A computed tomography scan revealed fistula formations between the cecum and sigmoid colon (arrow) (a and b) and between the ileum sigmoid colon (c) (arrowhead). The tumor shrinkage compatible with a partial response was achieved after neoadjuvant concurrent chemoradiotherapy (d). Note that the ileo-sigmoid colon fistula remains despite tumor regression (arrow)

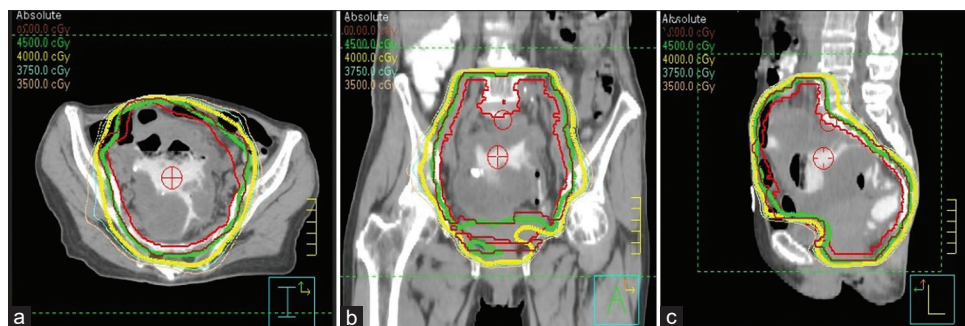


Figure 2: Radiation isodose curves in axial (a). Coronal (b). And sagittal views (c). A dose of 45 Gy is prescribed to cover the pelvic tumor and lymphatic drainage area with an appropriate margin



Figure 3: Surgical specimen. Please note that the rectosigmoid junction tumor (arrowhead) is closely attached to the ileum (arrow) and cecum (asterisk)

involves the duodenum.^{6,7} Malignant gastrointestinal fistula is an unusual presentation of colorectal cancer. Symptoms may be related to the primary tumor and fistula or the metabolic disease it caused.⁶ In our case, the symptoms of the malignant bowel obstruction by the advanced primary tumor are partially relieved by double fistulas, which decompressed the raised intraluminal pressure to a certain degree, resulting in her subtle gastrointestinal symptoms and delayed diagnosis. A radiologic examination is an excellent diagnostic tool, including a barium contrast study and CT. Endoscopies provide an opportunity for direct visualization of the fistula tract and biopsy for histology.^{3,6,8}

Treating malignant gastrointestinal fistula is usually challenging, and their management depends on the patient's underlying medical condition, disease stage, and fistula types. It is best discussed within a multidisciplinary tumor board setting.⁷ Correction of an unbalanced metabolism and poor nutrition status, followed by immediate *en bloc* resection of the fistula, were conducted in most presented cases.^{3,5,7,8} In our case, we initially administered concurrent chemoradiation to the unresectable pelvic tumor and involved lymph nodes. The following radiologic and endoscopic studies showed significant tumor regression, and curative surgery was successfully conducted 12 weeks after chemoradiation.

The timing of surgical intervention is worthy of discussion. The down staging benefit of CCRT in the neoadjuvant setting is proven for advanced rectal cancer.⁹ Total neoadjuvant therapy (TNT), a new paradigm involving the delivery of chemoradiotherapy plus neoadjuvant chemotherapy before surgery, had a higher pathologic complete response rate (29.9% vs. 14.9%) and a potentially disease-free survival benefit.¹⁰ In this case, the down staging effect might be better if TNT was administered before surgery. Considering the persistence of fistulas after CCRT and the associated electrolyte and

nutritional imbalance, surgical resection of the residual tumors and fistulas followed by adjuvant chemotherapy were used instead of TNT. A multidisciplinary approach is essential for successfully treating these malnourished and complicated patients.

CONCLUSION

This case report highlights a rare complication of colorectal cancer, the formation of double fistulas between the sigmoid colon, ileum, and cecum. Managing malignant gastrointestinal fistula is challenging and depends on the patient's underlying medical condition, disease stage, and fistula types. In this case, concurrent chemoradiation was administered to the unresectable pelvic tumor and involved lymph nodes, followed by curative surgery 12 weeks after chemoradiation. One year after surgery, the patient was alive without recurrence. The timing of surgical intervention, particularly in the context of TNT, requires further investigation. Early detection and management of malignant gastrointestinal fistulas are crucial for better outcomes.

Ethical approval statement

This study proposal was approved by the Institutional Review Board of Tri-Service General Hospital, National Defense Medical Center, B202315037, 2023/03/21.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given her consent for her images and other clinical information to be reported in the journal. The patient understands that her name and initials will not be published and due efforts will be made to conceal her identity, but anonymity cannot be guaranteed.

Data availability statement

Data sharing not applicable to this article as no datasets were generated or analyzed during the current study.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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