OncoTargets and Therapy

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CASE REPORT

Synchronous Multiple Primary Malignant Adenocarcinoma of the Descending Colon and Fungating Bleeding Adenocarcinoma of the Terminal Ileum Presenting Massive Rectal Bleeding: A Trap for the Unwary

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Abstract: Primary cancer of the ileum is rare, and when it occurs in conjunction with primary colon cancer, it becomes even more infrequent and challenging to diagnose prior to surgical intervention. Primary small bowel cancers can be overlooked and may be misidentified as small bowel mesenchymal tumours or advanced metastases from colon cancer. We present an exceedingly uncommon case of ruptured primary ileal cancer combined with primary descending colon cancer presenting with gastrointestinal bleeding. Based on our understanding, instances of dual tumours concurrently occurring are exceedingly infrequent. In this patient, there was a preoperative suspicion of bleeding from colon cancer in the descending region. However, intraoperative exploration revealed that the location of the bleeding was a terminal ileal mass. Following the surgical intervention, the patient recovered satisfactorily. Intraoperative exploration of the entire gastrointestinal tract is therefore necessary in patients with gastrointestinal haemorrhage, especially in those who require urgent surgery without adequate preoperative investigations. If a mass is detected at the end of the ileum, intraoperative pathology should be performed if feasible. Subsequently, if the diagnosis reveals an adenocarcinoma, terminal ileocolic resection and right hemicolectomy are necessary for appropriate resection.

Keywords: multiple primary malignant neoplasms, gut bleeding, adenocarcinoma of the small bowel, descending colon cancer, gastrointestinal tumours, case report

Introduction

Multiple primary malignant neoplasms (MPMNs) are multiple primary tumours that occur simultaneously or heterochronously at different sites in the same patient and may occur throughout the body.[1] For the diagnosis of multiple primary tumours, it is very easy to miss a diagnosis when the tumours occur at the same time, and it is easy to misdiagnose metastatic cancer when they occur at different times. The duodenum and jejunum are the most common sites for small bowel adenocarcinoma, whereas small bowel adenocarcinoma in the distal ileum is rare.[2] Due to the infrequency of small bowel cancer cases, lesions of the small bowel occasionally become overlooked during gastrointestinal surgery due to lack of examination of the small intestine during surgery, leading to the failure to diagnose small bowel cancer.[3] The diagnosis of ileal adenocarcinoma was almost completely missed in this patient.

This case report describes a patient with primary ileal cancer combined with primary descending colon cancer that was associated with ileal cancer rupture and bleeding. In this patient, there was a preoperative suspicion of bleeding from a colon cancer lesion in the descending region. However, intraoperative exploration revealed that the location of the

Received: 4 January 2024
Accepted: 9 April 2024
Published: 2 May 2024

bleeding was a terminal ileal mass. The aim of this report is to help clinicians avoid a missed diagnosis of this condition during emergency surgery due to a lack of in-depth preoperative assessments.

Case Presentation

A 72-year-old woman (BMI of 25.6 kg/m2) presented at the emergency clinic with persistent fresh blood in her stool for a duration of 2 hours, resulting in a total blood loss volume of approximately 2500 mL. Cardiac examination conducted in the emergency resuscitation room revealed a heart rate of 94 bpm, blood pressure of 90/45 mmHg, and haemoglobin level of 55 g/L. The patient was diagnosed with haemorrhagic shock.

Preoperative CT suggested that there was thickening of the end of the descending colon–sigmoid colon–rectum intestinal tube with multiple areas of significant exudate and small lymph nodes. There were slightly hyperdense foci in the terminal sigmoido-rectal mesentery of the descending colon (Figure 1). Emergency surgical treatment and preoperative colonoscopy revealed a large amount of bright red blood in the rectum. The examination further revealed a cauliflower-shaped mass that had infiltrated and grown into in the descending colon and was located 20 cm away from the anus, suggesting the possibility of bleeding in combination with descending colon cancer, and the decision was made to perform radical surgery for descending colon cancer.

Exploration of the abdomen revealed a descending colonic mass; on continued exploration, we found a mass in the lumen of the ileum that was 5 cm from the ileocecal valve, and it was considered that the mass could possibly be an ileocecal mesenchymal tumour. No blood was found in the proximal bowel of the ileocecal mass. However, the distal region and entire colon were filled with blood, which strongly suggested that the bleeding site was likely within the ileocecal region. Intraoperatively, we considered the small bowel mass to be a mesenchymal tumour, so we first resected the small bowel mass and performed lateral anastomosis of the two severed ends of the small bowel. Radical surgery for colon cancer located in the descending colon was subsequently performed. Simultaneously, a descending colostomy was performed due to the lack of bowel preparation. The patient had a total of 100mL of blood loss, and the whole operation took 2 hours and 30 minutes. After surgery, the patient returned to the general ward. Postoperative tumour marker testing revealed no abnormalities; CEA was 0.67 ng/mL, CA125 was 8.10 U/mL, and CA19-9 was less than 1.20 U/mL. With nutritional support, the patient had no apparent complications and was discharged from the hospital twelve days after surgery with a good recovery.

Postoperative pathology revealed moderately differentiated adenocarcinoma (descending colon tumour) with infiltration of the peritoneal fat membrane (pT3); cancer metastasis was observed in 1/17 of the peritoneal lymph nodes, and 2 other cancerous nodules were observed, staged as pT3N1M0. Immunohistochemistry showed the following: Tumour cells MLH1 (no deletion), MSH2 (no deletion), MSH6 (no deletion), PMS2 (no deletion), HER2[4B5] (20%+), p53 (I), Ki67 (90% positive).(Small bowel tumour) Moderately differentiated adenocarcinoma had infiltrated into the submucosal layer (pT1); no cancerous metastases were observed in the peristomal lymph nodes 0/2, staging pT1N0M0. Surprisingly, postoperative pathology revealed that the small bowel mass was an adenocarcinoma of the small bowel, and gross pathology (Figure 2) and paraffin sectioning (Figure 3) revealed that the adenocarcinoma of the small bowel and

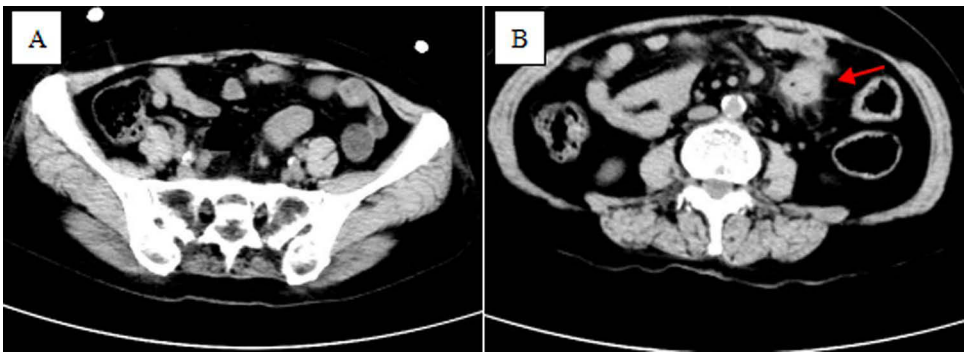

Figure 1 (**A**) The terminal ileum shows no obvious occupancy. (**B**) Preoperative CT image showing the morphology of the end of the descending colon–sigmoid colon–rectum intestinal tube thickening with multifocal areas of exudation and small lymph nodes. There were slightly hyperdense foci in the terminal sigmoido-rectal mesentery of the descending colon.(The descending colon tumour is indicated by the red arrow in the figure).

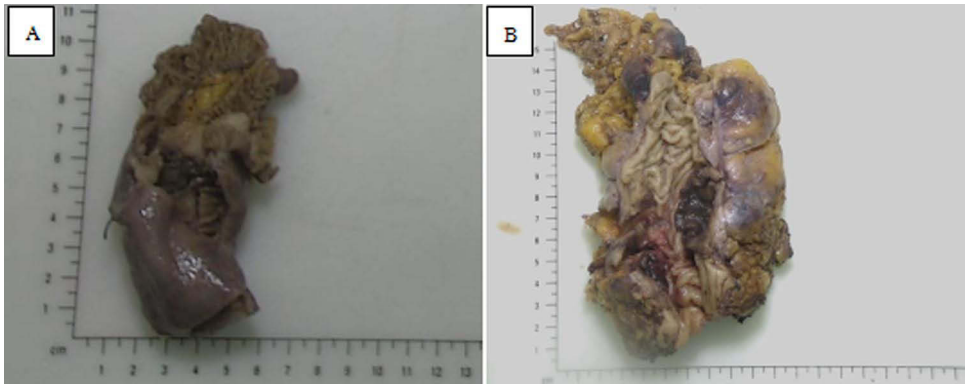

Figure 2 (**A**) Gross pathology of a tumour of the terminal ileum. (**B**) Gross pathology of a tumour of the descending colon.

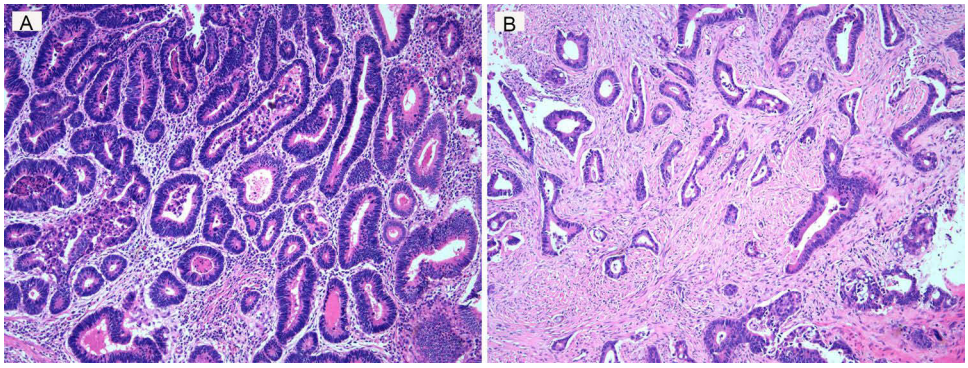

Figure 3 (**A**) The picture shows ileal adenocarcinoma cells found in sections of the resected tissue. (**B**) The picture shows the descending colon intestinal adenocarcinoma cells found in the resected tissue sections.

adenocarcinoma of the descending colon were nonhomologous. After a histopathological diagnosis of ileal adenocarcinoma, we advised the patient to undergo right hemicolectomy, but due to weakness and lack of financial support, the patient chose to undergo chemotherapy first. The patient was started on 1–6 cycles of chemotherapy with the XELOX regimen 4 weeks after surgery, followed by 1–2 cycles of chemotherapy with the capecitabine regimen. At 1, 3 and 6 months postoperatively, we recommended that the patient undergo outpatient imaging, haematological examinations and endoscopy. Postoperative follow-up endoscopy revealed four 0.3–0.6 cm polyps in the small and large bowel, which were endoscopically resected as soon as they were detected. No neoplastic tumour was found during follow-up. Therefore, the patient did not undergo a right hemicolectomy. We have advised the patient to have regular check-ups, and if a recurrence of the tumour is detected, we will perform additional surgical treatment.

Discussion

This report outlines the presence of two adenocarcinomas that were growing concurrently in the bowel of a patient who suffered from gut haemorrhage. This is an extremely rare case. First, this was a rare case because adenocarcinoma of the ileum and adenocarcinoma of the descending colon are primary tumours that occur in patients with multiple primary malignant neoplasms. Second, because a colonic mass was identified on the preoperative colonoscopy, it was concluded that the patient's gastrointestinal haemorrhage was caused by this mass. However, after intraoperative exploration of the entire gastrointestinal tract, a clear diagnosis of an ileal mass was made.

The prevailing small bowel neoplasms are still secondary colorectal tumours, but primary lymphomas and adenocarcinomas also occur but less frequently. In this case, the adenocarcinoma of the small intestine was located in the distal ileum, an exceptionally unusual occurrence. Each year, only 2% of gastrointestinal cancers occur in the small bowel.[4] Owing to the infrequency of small bowel cancer cases, lesions of the small bowel occasionally become overlooked

because examination of the small bowel during gastrointestinal surgery either does not occur or is inadequate, leading to the failure to diagnose small bowel cancer. Therefore, intraoperative exploration of the entire gastrointestinal tract is necessary in patients with a bowel obstruction or bleeding in the gastrointestinal tract, particularly patients who require emergency surgery or who have not been adequately examined preoperatively.

Gastrointestinal polyposis syndrome is distinguished by the occurrence of numerous polyps in the gastrointestinal tract, predominantly in the colon.[5] Among these, familial adenomatous polyposis,[6] Lynch syndrome,[7] Cowden syndrome,[8] and NTHL1 tumor syndrome[9] are associated with an elevated risk of colorectal cancer. There is a lack of literature documenting an increased risk of small bowel adenocarcinoma in individuals with gastrointestinal polyposis syndromes. In addition, a hypothesis has been reported in the literature that some of the genetic mechanisms that lead to colorectal cancer may also be involved in the development of small bowel adenocarcinoma. Both tumours can arise from pre-existing tumours; patients with colorectal adenocarcinoma have an increased risk of developing small bowel adenocarcinoma; and patients with small bowel adenocarcinoma also have an increased risk of colorectal adenocarcinoma.[10] Moreover, the microsatellite instability-high (MSI-H)/mismatch repair gene defect (dMMR) rates are greater in small bowel adenocarcinoma than in colorectal cancer.[11] No significant irregularities were observed in the postoperative MSI immunohistochemistry or MMR images of this patient.

The clinical presentation of primary cancer of the small bowel tends to be nonspecific, with the majority of its manifestations appearing during later stages of the disease. The predominant manifestation is gastrointestinal haemorrhage, while a minority of patients exhibit abdominal discomfort or alterations in bowel movements.[12] Preoperative diagnosis of primary small bowel cancer is mainly performed by endoscopy or enterography, both of which complement each other. Many studies have reported that double-balloon endoscopy is very effective in diagnosing small bowel cancer. Specifically, this approach focuses on patients with small bowel stenosis. For small bowel cancer, magnetic resonance enterography is highly accurate.[13–20]

Surgery for small bowel tumours is currently the mainstay of treatment. Initially, we suspected that the small bowel mass in this patient could be an ileocecal mesenchymal tumour. Due to preoperative haemorrhagic shock, the patient's vital signs were unstable, and she could not tolerate prolonged surgery, so only partial resection of the small intestine was performed. Unfortunately, the postoperative pathology of the small bowel mass revealed primary small bowel adenocarcinoma. In accordance with the National Comprehensive Cancer Network (NCCN) 2023 Guidelines for Surgery for Adenocarcinoma of the Small Bowel, jejunum/Ileum segmentectomy and margins of at least 5–10 cm on either side of the tumour were obtained. Terminal ileal resection with right hemicolectomy should be used for distal ileal tumours. Lymph nodes were identified and resected down to the origin of the feeder vessels. Clinically suspicious nodes outside the field of resection should be biopsied or resected whenever possible, and the evaluation of at least 8 lymph nodes should be the goal for all resections.[21–26] Therefore, if intraoperative exploration of the entire gastrointestinal tract detects a mass in the terminal ileum and if the patient's vital signs allow, it is advisable to perform intraoperative pathological examination whenever feasible. If the pathological findings show ileal adenocarcinoma, the extent of surgical resection should be terminal ileocolic resection and right hemicolectomy. We recommend reoperation with right hemicolectomy in similar cases. Some studies have shown that small bowel adenocarcinoma is sensitive to chemotherapy. Therefore, chemotherapy should be given after surgery in strict accordance with the National Comprehensive Cancer Network (NCCN) guidelines for small bowel adenocarcinoma. It is also important for the patient to have regular follow-ups after surgery, include imaging, haematological examinations or endoscopy.

Treating primary small bowel cancer is a major challenge for doctors because patients are usually diagnosed during a later stage of their disease.[27] Some researchers have used databases to analyse the factors that affect the prognosis of small bowel adenocarcinoma patients. Studies have shown that tumour location correlates with survival, with patients with duodenal cancer having the worst cancer-specific survival rates and patients with ileal tumours having the best survival rates. Patients treated with chemotherapy also have better survival than patients not treated with chemotherapy.[28] Despite significant advances in imaging and treatment, the prognosis for primary small bowel cancer patients remains poor, with an overall survival rate of 30% at 5 years. Although the prognosis beyond 5 years remains poor, surgical resection offers some survival benefit.

Conclusion

Therefore, a comprehensive intraoperative exploration of the entire gastrointestinal tract is necessary in patients with a bowel obstruction or bleeding in the gastrointestinal tract, particularly in patients who require emergency surgery or who have not been adequately examined preoperatively. If a tumour is found in an unexpected location, intraoperative pathology should be completed to provide a rational and standardized strategy to guide the next step in treatment.

Abbreviations

MPMNs, multiple primary malignant neoplasms; MSI-H, microsatellite instability-high; dMMR, mismatch repair gene-deficient; NCCN, National Comprehensive Cancer Network.

Data Sharing Statement

The paper includes all data related to the study.

Ethics Approval and Consent to Participate

Ethical approval for this study was obtained from the Ethics Committee of the Institutional Review Board of the Second Affiliated Hospital of Dalian Medical University. Written informed consent for this case report was obtained from the patient, who consented to the publication of the relevant details and imaging data in her case.

Funding

There is no funding to report.

Disclosure

The authors report no conflicts of interest related to this work.

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