

Colon Adenocarcinoma Metastasis Through Ileocolic Fistula to Small Bowel in the Setting of Crohn's Disease

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Abstract

Purpose: Patients with Crohn's disease (CD) are at higher risk of developing colorectal cancer (CRC) and gastrointestinal fistula. We report an unusual case of sigmoid colon adenocarcinoma in a CD patient that metastasized to the small bowel through an ileocolic fistula tract.

Methods: This case report was written after patient was seen in the clinic and reviewing overall hospitalization including clinically relevant data including imaging and pathology reports associated to our focus and presentation. Prior cases of metastatic CRC via fistula tract were reviewed and compared as well.

Results: We described an unprecedented case of a sigmoid adenocarcinoma metastasized to ileum via ileal-sigmoid fistula. Patient received surgical treatment and systemic chemotherapy and currently in remission.

Conclusion: CD is associated with a higher risk of fistula development. Few cases in the past described CRC metastasized within the gastrointestinal tract through a fistula. Intriguingly in our case, sigmoid adenocarcinoma developed and further metastasized to the ileum via the ileal-sigmoid fistula in the setting of CD. In addition to presenting a unique pathological phenomenon in these patients, this case raises awareness of the importance of regular follow-up and early initiation of IBD therapies.

Introduction

Colorectal cancer (CRC) is the third most common type of cancer worldwide, with colon cancer more frequent than rectal cancer [1]. Inflammatory bowel disease (IBD) patients are 2-6 times more likely to develop CRC than the general population [2,3]. IBD-related CRC, is primarily seen in patients with ulcerative colitis (UC); therefore, guidelines recommend starting surveillance colonoscopy after 8-10 years of diagnosis [4]. The risk of CRC associated with Crohn's Disease (CD) is more contentious, and CD diagnosis by itself does not indicate a high risk of CRC; therefore, surveillance colonoscopy is recommended only if more than one third of the colon is involved [4-6]. The most common site of colon cancer metastasis is in the liver due to its anatomical location and portal circulation [7]. When metastasized, the most common route is hematogenous or localized spread [8]. We report an unprecedented case of adenocarcinoma of the sigmoid colon in a CD patient that metastasized to the small bowel through an ileocolic fistula tract.

Case Presentation

A 37-year-old Caucasian male with a history of ileocolic CD presented with abdominal pain associated with nausea, vomiting, and diarrhea. The patient was diagnosed with CD at the age 19 but was not on treatment due to non-compliance. On admission, the patient was hemodynamically stable and afebrile, with a blood pressure of 130/80 mmHg, heart rate of 90 beats per minute, and respiratory rate of 14 per minute. Physical exam was remarkable for diffuse abdominal tenderness and hypoactive bowel sounds. The patient had computed tomography (CT) of the abdomen that revealed prominent wall thickening of the terminal ileum consistent with active inflammation suggestive of CD flare with a fistulous tract noted between the terminal ileum and sigmoid colon. (Figure 1) The patient was started on budesonide with a plan to start infliximab as an outpatient for fistulizing CD. Colonoscopy before initiating infliximab, showed a severe rectosigmoid stricture that could not be traversed with a colonoscopy. (Figure 2) Biopsies from the stricture revealed adenocarcinoma of the colon. The patient underwent open total proctocolectomy; however, at the time of surgery, there was involvement of the terminal ileum through the sigmoid colon-ileum fistulous tract, and therefore, a proximal portion of the terminal ileum was also surgically resected. The

pathology showed adenocarcinoma of the sigmoid colon with the involvement of the terminal ileum at the fistula site. (Figure 3 and 4) The patient recovered well from surgery and is currently undergoing adjuvant chemotherapy.

Discussion

We have reported an unusual case of colon cancer in a young patient with CD, that metastasized through a fistulous tract to the terminal ileum. Colon cancer in the setting of CD is an infrequent occurrence as compared to UC [5,6]. Metastatic CRC through a fistula is a very rare disease presentation with no reports in the literature of implantation into the small bowel through an ileal-sigmoid fistulous tract. Table outlines the reported cases of CRC metastasis within the gastrointestinal tract through fistulas. Chronic intestinal inflammation such as IBD leads to a higher incidence of gastrointestinal malignancy [9,10]. A study by Axelrad et al., described an incidence ratio of 5.7 on CRC, 21 of small bowel adenocarcinoma in IBD patients; and notable for 20 to 30 fold increase risk especially for ileal carcinoma in CD compared to general population [11]. Despite his young age, unfortunately, our patient developed colon cancer, which was likely secondary to ongoing accelerated inflammation in setting of non-compliance with medical therapy.

Patients with CD are more prone to fistula formation with occurrence reported in 17-50% of CD patients. The cumulative incidence of fistulizing CD is 21% one year after diagnosis that is increased to 50% after 20 years [12]. An ileal-sigmoid fistula is a rare presentation described in a surgical case report by Hurwitt and Lantino in 1957 [13]. Other etiologies of ileocolic fistulas development include surgical complications and radiotherapy [14]. Common metastatic sites of colon cancers include the liver, lungs, nervous system, and peritoneum [8]. Intra-gastrointestinal tract metastasis are also described in few cases in the past, such as a sigmoid adenocarcinoma discovered in the duodenal bulb by Iwamuro et al. [15]. The mechanism of the metastasis is suspected to be via hematogenous, translocation of tumor cells, or peritoneal spread [16]. As described in our case, CRC metastasized through a fistulous tract is an unusual mechanism of metastasis. Reported cases of intra-intestinal tract metastasis via fistula are almost always through an anal fistula or closer proximity anatomical position, as listed in table. However, there is no prior case of metastatic adenocarcinoma of sigmoid origin into ilea-sigmoid fistula reported. Treatment of the ileocolic fistula is often surgical, as they are less likely to resolve spontaneously. However, treatment is individualized for each patient, as the morphology of the fistula and course are often complicated. In our patient, in addition to surgical treatment, his extensive disease warranted chemotherapy.

In conclusion, CD is associated with a higher risk of fistula development. Few cases in the past described CRC metastasized within the gastrointestinal tract through a fistula. Intriguingly in our case, sigmoid adenocarcinoma developed and further metastasized to the ileum via the ileal-sigmoid fistula in the setting of CD. In addition to presenting a unique pathological phenomenon in IBD patients, this case raises awareness of the importance of regular follow-up and early initiation of IBD therapies.

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Tables

Table: Cases of colon cancer metastasis to other sites in the gastrointestinal tract through fistulous tracts

Author	Age, Sex	IBD (Yes/No), type	Primary cancer site	Metastatic site	Fistula (Yes/No)	Cancer implanted fistula (Yes/No)
Ikeda [17]	68, Male	No	Rectosigmoid adenocarcinoma	Anal fistula/Anus	Yes, Anal fistula	Yes
Benjelluon [18]	55, Male	Y, unspecified	Rectosigmoid adenocarcinoma	Anal sphincter/perianal area	Yes, Anal fistula	Yes
Benjelluon [18]	68, Male	No	Rectosigmoid adenocarcinoma	Perianal mass	Yes, perineal fistula	Yes
Gomes [19]	65, Male	No	Sigmoid adenocarcinoma	Anus	Yes, Anal fistula	Yes
Spiridaki [20]	79, Male	No	Mucinous rectosigmoid adenocarcinoma	Anus	Yes, Anal fistula	Yes
Balcabasa [21]	61	No	Synchronous adenocarcinoma	Colon	Yes, Gastrocolic fistula	No
Zhou [22]	59, Male	No	Colon tubular adenocarcinoma	Gastral antral fistula; omentum, lymph node, serosa.	Yes, Gastrocolic fistula	Yes
Lee [23]	82, Female	No	Gastric carcinoma	Colon at splenic flexure	Yes, Gastrocolic fistula	Yes
Tejedor [24]	49, Male	No	Colon carcinoma	Stomach, jejunum	Yes, Gastrojejunocolic fistula	Yes
Nakazawa [25]	84, Male	No	Sigmoid colon cancer	Bladder, Urethral	Yes, Colovesical fistula	Yes
Present Case	37, Male	Yes	Sigmoid colon cancer	Ileum	Yes, Ileosigmoid fistula	Yes

* IBD; inflammatory bowel disease

Figures



Figure 1

The patient had computed tomography (CT) of the abdomen that revealed prominent wall thickening of the terminal ileum consistent with active inflammation suggestive of CD flare with a fistulous tract noted between the terminal ileum and sigmoid colon. (Figure 1)

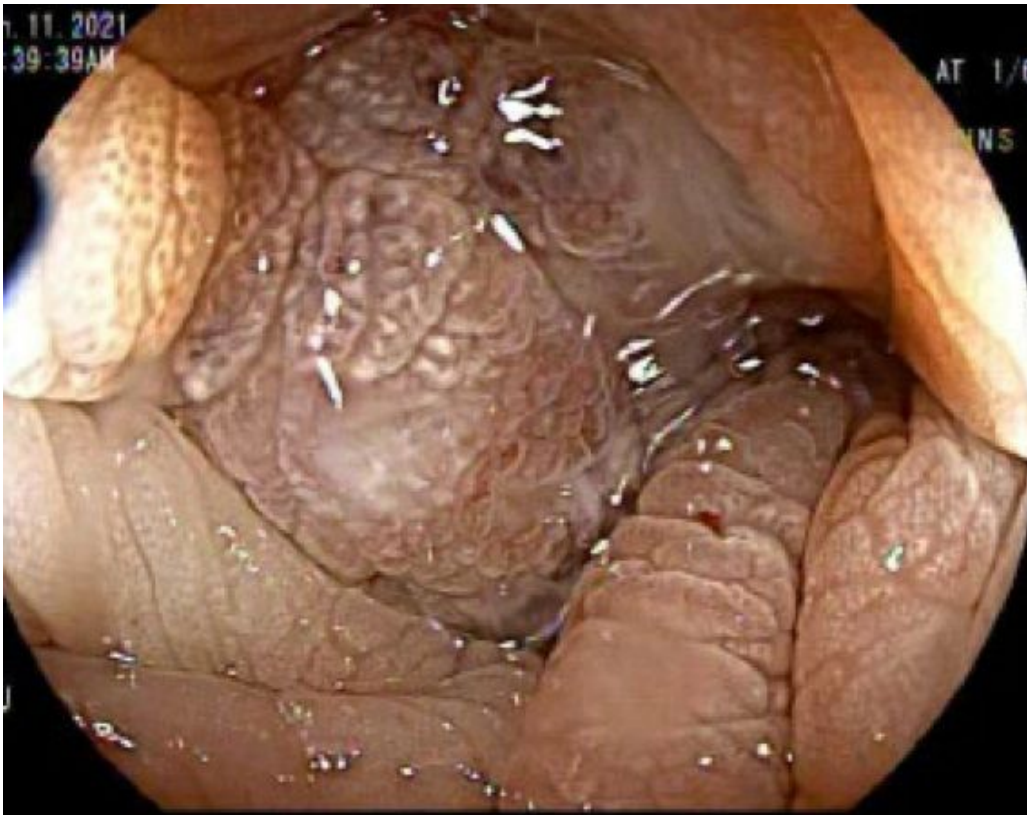


Figure 2

The patient was started on budesonide with a plan to start infliximab as an outpatient for fistulizing CD. Colonoscopy before initiating infliximab, showed a severe rectosigmoid stricture that could not be traversed with a colonoscopy. (Figure 2)

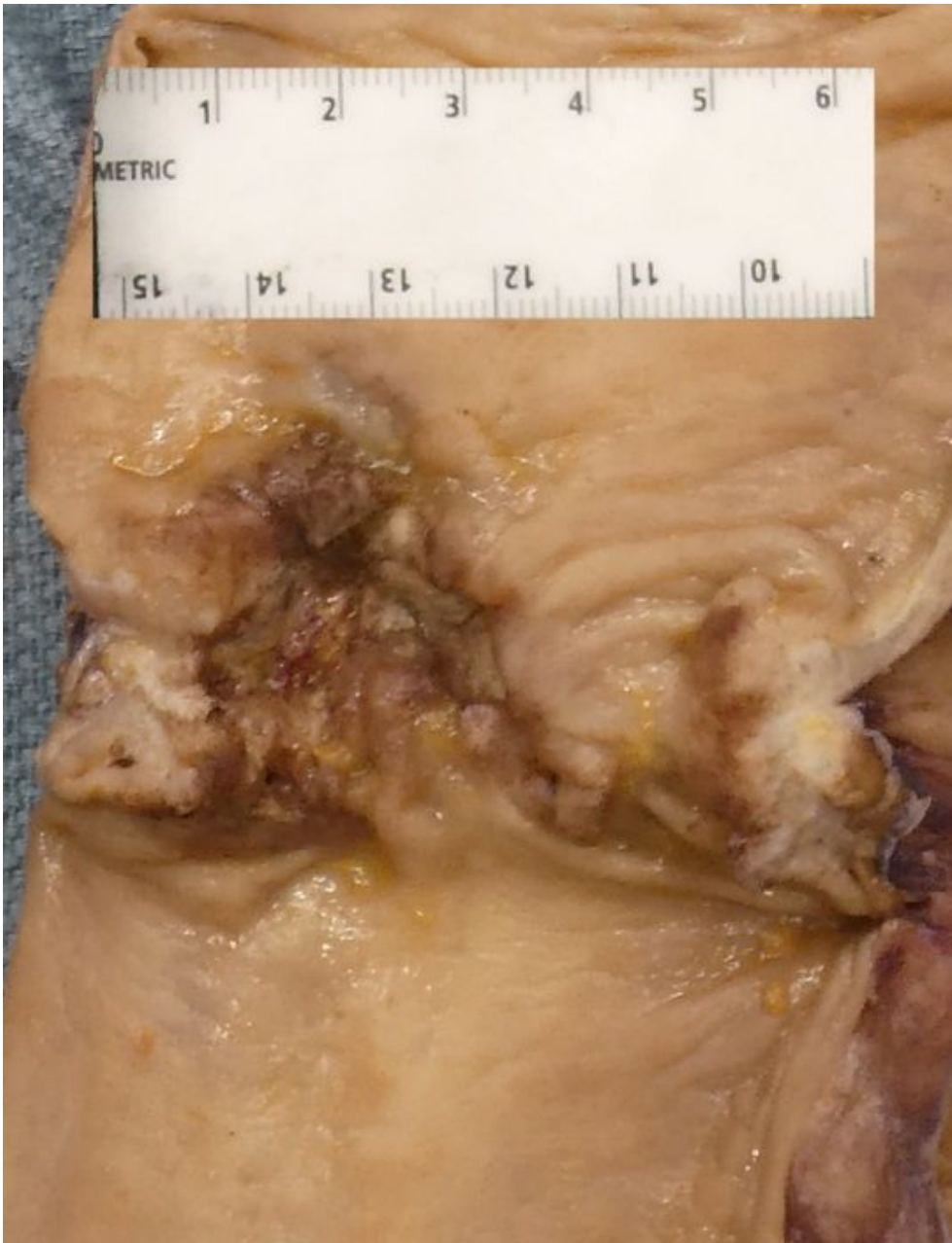


Figure 3

The pathology showed adenocarcinoma of the sigmoid colon with the involvement of the terminal ileum at the fistula site. (Figure 3 and 4)

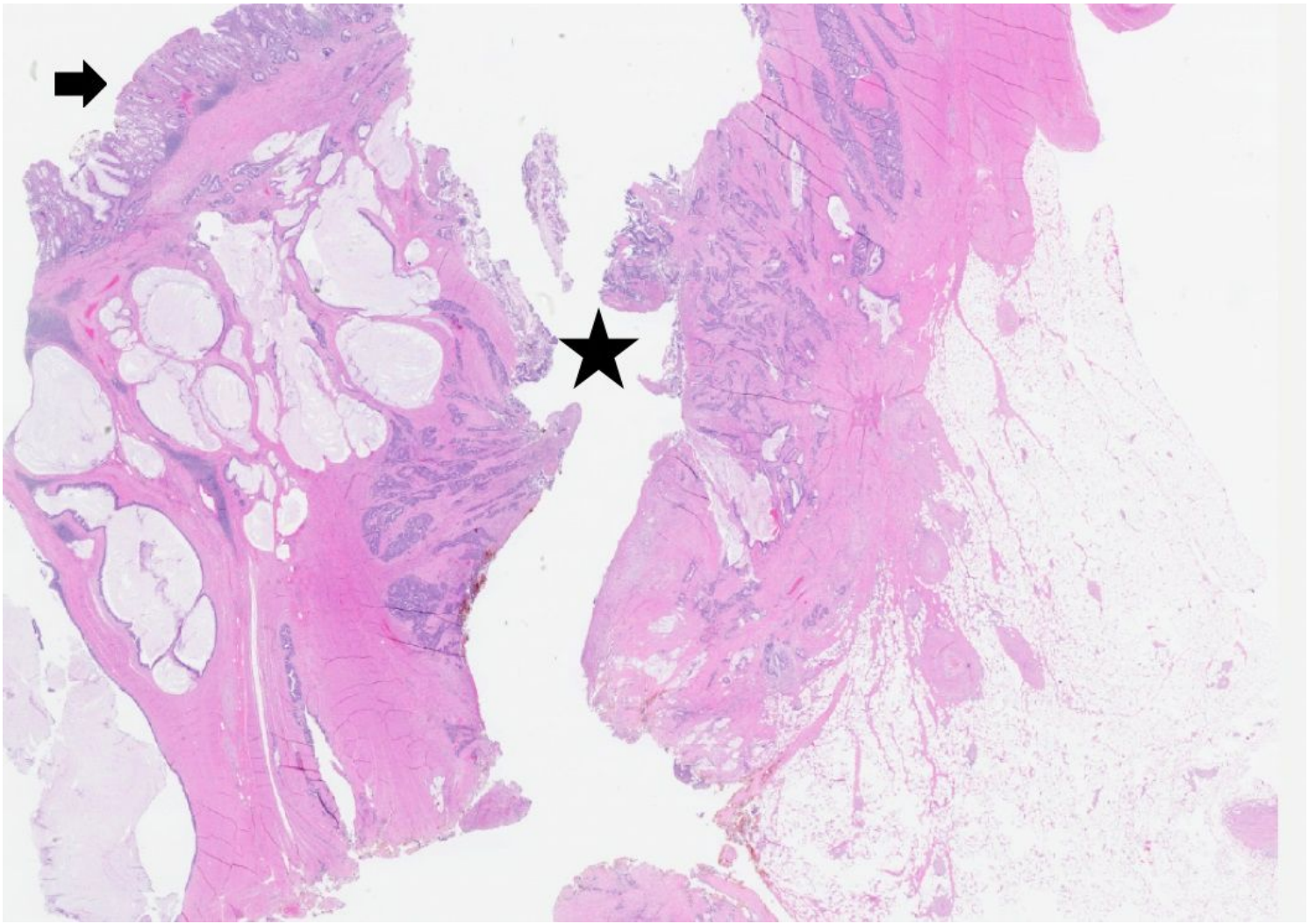


Figure 4

The pathology showed adenocarcinoma of the sigmoid colon with the involvement of the terminal ileum at the fistula site. (Figure 3 and 4)