

Adenocarcinoid tumor of the rectum: a rare finding in a patient with recurrent fistulizing perianal Crohn's disease

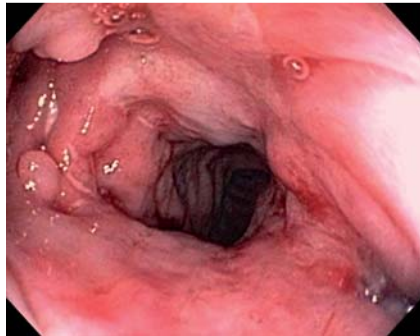


Fig. 1 Endoscopic image of the rectum in a 48-year-old man with Crohn's disease showing a fistulous tract and distal inflammation.

Malignancy in patients with Crohn's colitis is well described; however, adenocarcinoid tumor associated with Crohn's disease is very rare with only four cases reported in the literature [1]. This is the first reported case of an adenocarcinoid tumor arising in a fistulous tract.

A 48-year-old man presented with diarrhea and a chronic perirectal fistula. Colonoscopy showed pancolitis with aphthous ulcers and pseudopolyps. Clinical and endoscopic findings were consistent with Crohn's disease. Treatment with immunosuppressive agents was recommended, but the patient refused because of the potential side effects and sulfasalazine was therefore prescribed. He initially achieved clinical remission but experienced a recurrent perirectal abscess and fistulous drainage, for which he received a few short courses of antibiotics. A repeat colonoscopy showed mild pancolitis with inflammation of the distal rectum and a fistulous tract (▶ Fig. 1). Rectal biopsies showed moderately differentiated adenocarcinoma with extensive mucin and signet-ring cells present (▶ Fig. 2). Staging showed no evidence of metastases and he underwent abdominoperineal resection followed by adjuvant chemotherapy. Pathology from the surgical specimen showed active colitis, cryptitis, and transmural inflammation with noncaseating granulomas, further substantiating the diagnosis of Crohn's disease.

Two years later, he developed small-bowel obstruction, which required ileocecectomy. Small-bowel mesenteric deposits

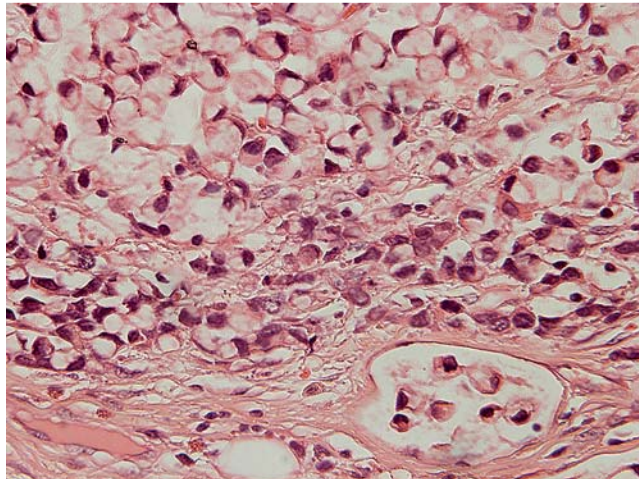


Fig. 2 Histopathological appearance of the original rectal biopsy showing a signet-ring cell pattern in an apparent moderately differentiated adenocarcinoma (hematoxylin and eosin [H&E] stain, magnification $\times 400$).

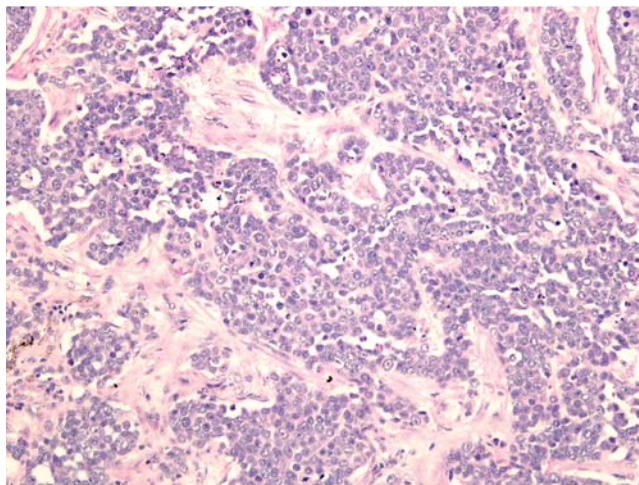


Fig. 3 Histopathological appearance of the mesenteric deposits found during surgery 2 years later showing an organoid and trabecular pattern consistent with metastatic adenocarcinoid (hematoxylin and eosin [H&E] stain, magnification $\times 100$).

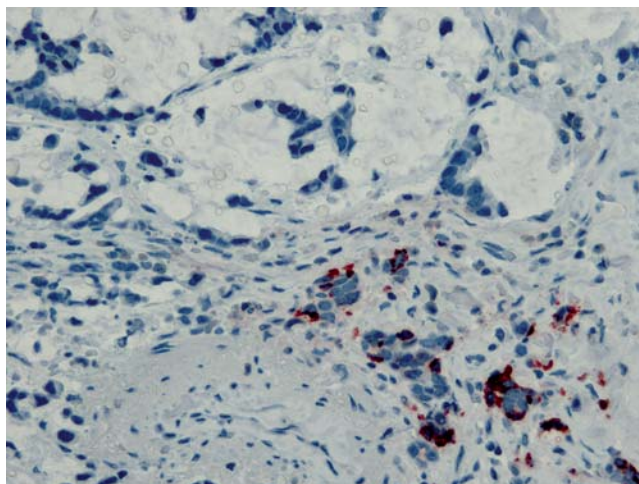


Fig. 4 Appearance of the original specimen stained immunohistochemically for chromogranin showing positive staining of small nests adjacent to the signet-ring cells, which confirmed the diagnosis of an adenocarcinoid tumor (magnification $\times 200$).

were seen during laparotomy; pathological examination of these showed metastatic adenocarcinoid tumor with signet-ring cell features (● Fig. 3); immunohistochemical stains for both CK20 and chromogranin were positive. Adenocarcinoid had not been suspected after his initial surgery, but immunohistochemical staining of tissue from the original rectal tumor confirmed this had been the diagnosis (● Fig. 4).

Adenocarcinoid tumor, or goblet-cell tumor, is a rare malignancy that comprises features of both adenocarcinoma and carcinoid. Most adenocarcinoid tumors arise in the appendix and only six cases have previously been reported in the rectum, none of which were associated with Crohn's disease [2]. Most patients with rectal adenocarcinoid present with rectal bleeding; symptoms of carcinoid syndrome are rare [3]. The management and prognosis of adenocarcinoid tumors are most dependent on the glandular component [3].

Malignancy arising in fistulous tracts in Crohn's disease is rare, with only 61 cases reported from 1950 to 2008 [4]. We hypothesize that malignant transformation arises from chronic bacterial colonization and fistulous tract inflammation. This is the first reported case of adenocarcinoid arising in a fistulous tract in Crohn's dis-

ease. Malignancy, including adenocarcinoid, should be considered in patients with recurrent fistulizing disease.

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Competing interests: None

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