Two Cases of Malignant Transformation in Crohn’s Disease with Perianal Involvement

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Abstract
This report includes 2 cases of Crohn’s disease with perianal involvement in which adenocarcinoma developed in the setting of a fistula. The cases illustrate the importance of considering the development of neoplasia in the presence of fistula, treatment-resistant symptoms, and rectal discharge.

Keywords: Crohn’s disease, malignancy, perianal disease

INTRODUCTION
The frequency of intestinal and extraintestinal malignancies in patients with Crohn’s disease is higher than that in the normal population.1 In perianal involvement, there are difficulties in the detection of malignancies due to the severity of inflammation, similarity of symptoms to those seen in infection, and limited sensitivity of imaging methods. Special attention regarding malignant transformation should be paid for patients with Crohn’s disease who have long-standing anal fistulas, especially when they complain of exacerbated symptoms (i.e., increased pain, fluid discharge) from a perianal lesion.2

In this report, we present 2 cases of Crohn’s disease with perianal involvement who subsequently developed mucinous rectal adenocarcinoma.

CASE 1
A 38-year-old male patient was admitted to our clinic with rectal pain and discharge. Eighteen years ago, he was diagnosed with fistulizing and stenosing Crohn’s disease, and a right hemicolectomy was performed due to ileus at the time of diagnosis.

The patient was followed up with mesalamine and azathioprine treatments and was lost from follow-up for the next 7 years as he had no complaints. One year ago, a perianal abscess was detected in the patient due to rectal pain and discharge; drainage and seton procedures were performed; treatment was planned as 40 mg of adalimumab every other week and azathioprine 150 mg per day.

Pelvic magnetic resonance imaging (MRI) performed at his first presentation due to rectal pain and discharge revealed an appearance compatible with an abscess in the perirectal area with a size of 46 × 17 × 50 mm and intersphincteric fistula tracts located at the 4, 6, and 9 o’clock levels. Colonoscopy showed an anastomosis line and distal ileum, deep ulcers in the rectum, and perianal high-flow fistulas (Figure 1). Neoterminal ileum biopsy was consistent with mild active chronic enteritis with crypt distortion, and rectal biopsy was consistent with chronic inflammatory mucosa.

The patient underwent perianal abscess drainage and a seton procedure. After 1 month of ciprofloxacin and metronidazole antibiotic therapy, the current treatment was revised as infliximab 400 mg for 8 weeks and AZA 150 mg/day. On the pathological examination of the perianal abscess drainage material, which was repeated due to the persistence of rectal pain and discharge complaints, abscesses in the fatty tissue as well as dense mucin pools and single-row mucin pools without obvious atypia within these mucin pools and epithelial cell groups arranged basally were observed, and mucinous adenocarcinoma was suspected.

Positron emission tomography–computed tomography showed focal nonhomogeneity (maximum standardized uptake value (SUVmax): 10.0) in the area extending from the rectum to the anal canal and skin level and pathological fluoro 2-deoxyglucose (FDG) uptake (SUVmax: 7.8) in several foci in the right lung. It was reported that Miles operation was performed after neoadjuvant FOLFOX and radiotherapy.
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