Occult small bowel adenocarcinoma complicating Crohn’s disease: a report of three cases

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Summary
Three patients with Crohn’s disease were treated by ileal resection for intestinal obstruction. Histological examination of the resected specimens in each case established the diagnosis of adenocarcinoma of the small intestine complicating Crohn’s disease. This diagnosis should be considered in patients with longstanding macroscopic Crohn’s disease who present with severe or recurrent symptoms. The diagnosis may not be apparent on routine radiological examination or even macroscopically at laparotomy.

Keywords: Crohn’s disease, small bowel, adenocarcinoma

Introduction
The increased risk of colorectal cancer in extensive Crohn’s colitis is well recognised, and is of the same order as in ulcerative colitis. Small bowel adenocarcinoma complicating Crohn’s disease is uncommon but well recognised. Sixty one cases were published in the literature to 1982 and 60 cases have been summarised in a series of publications subsequently. A total of 121 examples in all. It is thus an important complication to consider in the management of patients with longstanding macroscopic small bowel Crohn’s disease. In this paper we describe three examples of adenocarcinoma of the ileum complicating Crohn’s disease which were only apparent on histological examination of the resected specimens.

Case reports
Case 1
A diagnosis of ileal Crohn’s disease in this white man, born in June 1949, was established on radiological examination in 1969. He had further abdominal symptoms in 1975 when radiological studies confirmed the changes of terminal ileal Crohn’s disease with a localised ileocecal fistula. He re-presented in July 1992 with a three month history of right-sided colicky abdominal pain, nausea, intermittent constipation and 10 kg weight loss. Further radiological assessment confirmed an irregular 8 cm of terminal ileum with abnormal mucosa and several areas of narrowing with mucosal irregularity adjacent to the ileo-caecal valve. At laparotomy the appearance macroscopically was described as typical of Crohn’s disease and he was treated by distal ileal resection. The resected specimen comprised an 11 cm length of small bowel with a 4 cm diameter ulcerated area with some transmural thickening. There were three distinct areas of mucosal irregularity. Macroscopically, the changes were considered typical of Crohn’s disease. However, histological examination revealed a moderately well differentiated adenocarcinoma, extending
through the bowel wall and invading muscle (figure 1). One group of dysplastic glands was identified in the small bowel mucosa adjacent to the tumour and there was evidence of perineural tumour spread and foci of submucosal lymphatic invasion. The lymph nodes examined from the first operative specimen were clear. The irregular mucosa contained foci of submucosal scarring consistent with quiescent Crohn's disease but there was no evidence of active disease.

In view of the histological evidence of adenocarcinoma of the ileum a further laparotomy was undertaken when a right hemicolectomy was performed. Histological examination of the resected specimen showed that two of the 11 lymph nodes examined were replaced by carcinoma.

The patient made an excellent post-operative recovery and received adjuvant therapy with 5-fluorouracil and levamisole. Unfortunately, the patient now (February 1994) has hepatic and pelvic metastases.

Case 2
The patient was a white woman, born October 1915, with a 30-year history of mild intermittent bouts of vomiting, diarrhoea and abdominal distention which had been attributed to the irritable bowel syndrome. She developed further symptoms in 1988 when a barium enema examination was reported as normal (?filling of distal ileum). She re-presented in October 1992 with a four year history of similar symptoms. Physical examination revealed a tender right iliac fossa mass. A barium follow-through examination showed several strictures in the distal ileum and an inflammatory mass consistent with Crohn's disease. At laparotomy there were matted loops of small bowel in the right iliac fossa with an ileocecal fistula and an ileal stricture 15 cm proximal to the matted loops. Histological examination revealed a Duke's B adenocarcinoma of the distal ileum. There were no metastases in the resected lymph nodes. The patient made an uncomplicated recovery. In view of her age, re-operation was not undertaken and she remains well (February 1994).

Case 3
The patient was a white man, born in September 1946, in whom Crohn's disease of the distal ileum and right colon was diagnosed in 1967 and treated conservatively. He was admitted acutely in October 1992, aged 46 years, with a one month history of abdominal pain, diarrhoea, and vomiting. Barium enema examination showed changes consistent with a carcinoma at the hepatic flexure and multiple strictures in the distal ileum consistent with Crohn's disease. A laparotomy revealed extensive Crohn's colitis with a mobile colonic carcinoma just distal to the hepatic flexure with changes of Crohn's disease in the distal ileum. There were no hepatic or peritoneal metastases.

The distal ileum and right colon were resected and continuity restored with an ileocolonic anastomosis. Histological examination of the resected specimen confirmed Crohn's colitis, complicated by Duke's B adenocarcinoma of the colon. In addition there was an ileal adenocarcinoma and histological evidence of Crohn's disease (figure 2). There was no vascular invasion or lymph node involvement.

Figure 1 Adenocarcinoma of the ileum invading muscle (H&E)

Figure 2 Ileal Crohn's disease with fissuring ulceration (H&E)
Despite post-operative complications which included pulmonary embolus, subclavian venous thrombosis and an enteroenterogenous fistula, he improved with conservative management and was fit and well when last reviewed in February 1994.

Discussion

Since Ginzburg et al. published the first report of adenocarcinoma of the ileum complicating Crohn’s disease, 61 examples had been published in 1982 and there have been 60 examples since, a total of 121. The largest reported series described 20 small bowel cancers in 19 patients with Crohn’s disease between 1960 and 1989. Since bypass surgery is no longer in vogue and diseased ileal segments are often resected, fewer may be seen in future. No examples of small bowel adenocarcinoma complicating Crohn’s disease were identified in a recent view of 1008 Crohn’s patients in the Aberdeen area.

These three examples of occult adenocarcinoma complicating Crohn’s disease emphasise that routine radiological assessment and macroscopic evaluation at laparotomy may overlook this complication since in all three examples, the adenocarcinoma of the small intestine was only identified on histological examination. It is of interest that one case presented with synchronous adenocarcinoma of the ileum and colon.

To ensure optimal management of patients with longstanding macroscopic Crohn’s disease of the small intestine who present with recurrent symptoms after a long asymptomatic interval, the possibility of malignancy should be considered and resection advised, since exclusion of adenocarcinoma of the small bowel on radiological or macroscopic grounds alone remains extremely difficult.

Summary

- Consider bowel carcinoma in a Crohn’s patient with recurrent symptoms after a long asymptomatic interval
- The adenocarcinoma may not be apparent on radiological examination