Adenocarcinoma Arising from a Strictureplasty Site in Crohn’s Disease
Report of a Case

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The occurrence of small-bowel cancer in Crohn’s disease (CD) is a rare event. The risk seems to be greatest in patients with long-standing disease. Strictureplasty has proved to be a valuable alternative in the management of Crohn’s strictures of the small-bowel. Critics and proponents of strictureplasty for selected patients with small-bowel Crohn’s disease have voiced their concerns about cancer risk in the strictured or strictureplasty site. To date, there has been no clear or detailed report of such an occurrence. The authors report the first case of small-bowel adenocarcinoma arising at the site of a previous strictureplasty. In this patient, biopsies of the strictures at the original operation confirmed CD and excluded both cancer and dysplasia. Malignancy occurred seven years later at a strictureplasty site. The main clinical sign associated with the adenocarcinoma was severe, persistent anemia. The authors conclude that the risk of adenocarcinoma developing at the site of a previous strictureplasty for CD, although small, is real. [Key words: Crohn’s disease; Strictureplasty; Small-bowel adenocarcinoma]


D uring the last decade, strictureplasty has proved to be a useful and effective surgical option in selected patients with Crohn’s disease (CD). Studies have indicated that there is no statistically significant difference in terms of reoperation and complication rates between strictureplasty and resective procedures in Crohn’s disease. One of the major concerns raised by this technique, however, is that the segment of inflamed bowel is left in situ. Thus, critics have feared that early relapse and possible malignancy may occur at strictureplasty sites. The relative risk of developing small-bowel cancer in Crohn’s disease is reported to vary from 6 to 320 times that of the general population. Despite this, its actual incidence is low. Since the first report of small-bowel cancer occurring in Crohn’s disease in 1956, 124 cases have been described in the literature. Although Alexander-Williams and Haynes reported a case of jejunal carcinoma occurring in the vicinity of a previous strictureplasty, no case of cancer arising at a strictureplasty site has been described in the literature to date. We report the first case of adenocarcinoma arising at a previous strictureplasty site for Crohn’s disease.

REPORT OF A CASE

The patient is a 78-year-old woman with a reported history of celiac disease since 1966. She had been following a gluten-free diet with good symptomatic response. In 1986, she started to experience nausea and vomiting, abdominal pain, diarrhea, weakness, fatigue, reduced appetite, and weight loss (7 lb). A small-bowel series performed at that time was reported as being “typical of sprue,” with a dilated mid small bowel. On evaluation at the Cleveland Clinic in 1987, a firm mass (8 × 10 cm) was palpable in the hypogastrium. A previous computed tomographic (CT) scan had shown some dilated loops of bowel but failed to identify the palpable mass (Fig. 1). The patient underwent laparotomy for chronic obstructive symptoms and intra-abdominal mass. At surgery, she was found to have jejunal and ileal Crohn’s disease. The mass was found to be a dilated loop of bowel filled with a large enterolith located between two strictures in the proximal ileum (Fig. 2), 15 cm apart. In addition, a fistula was present between the terminal ileum and the sigmoid. The more proximal stricture was 2 to 3 cm long and 20 mm in diameter. It appeared fibrotic, with no signs of acute inflammation.
Figure 1. Computed tomographic scan showing dilated loops of small intestine with a filling defect.

Figure 2. A. An enterotomy is made at the site of the stricture proximal to the enterolith. B. The enterolith is removed through the enterotomy.

Figure 3. No evidence of strictures or obstructing lesions with a barium contrast study one year before diagnosis of small-bowel adenocarcinoma.

The distal stricture was 4 to 5 cm long and 8 mm in diameter. This latter segment showed some degree of acute inflammation: both the bowel wall and mucosa appeared edematous and erythematous. No ulcers were seen.

An ileocecal resection and two proximal Heineke-Mikulicz strictureplasties were performed, and the sigmoid fistula was oversewn. Biopsies from the strictures were taken before strictureplasty, and no signs of cancer or dysplasia were present. In the more proximal stricture, focal pyloric-gland metaplasia and fibrosis were found, whereas in the other one, edema and focal acute inflammation were reported.

The patient remained well until late 1991, when she again began to experience weakness and fatigue. Laboratory studies showed a hemoglobin of 7 g/dl and a positive guaiac stool test. Blood count failed to respond to iron supplementation and to intermittent blood transfusions. An esophagogastroduodenoscopy showed only some mild gastritis. A small-bowel series at this time revealed recurrent, nonobstructing CD of the distal ileum (i.e., no narrowing or stricture), and a barium enema and a colonoscopy found no abnormality that could explain the severe anemia (Fig. 3). In the belief that a relapse of CD was the source of the bleeding, oral steroids were introduced.

In January 1993, the patient was referred back to the Cleveland Clinic with persistent abdominal cramps and anemia (hemoglobin, 7.2 g/dl). Also, an elevated carcinoembryonic antigen (CEA) of 10.9 ng/ml was noted. Laparotomy revealed a 8.5 × 5 × 1.2 cm neoplastic mass at the proximal strictureplasty site, suspicious for adenocarcinoma. The other strictureplasty site and distal small-bowel anastomosis were identified and found to be normal (the distal strictureplasty site could be identified from the metallic clips). A wide small-bowel resection was performed. Frozen