Is Hartmann's Procedure Safe in Crohn's Disease?

Report of Three Cases

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INTRODUCTION: Crohn's disease-associated colorectal cancer may occur in an area of defunctioning bowel. Some patients with Crohn's colitis undergo subtotal colectomy, ileostomy, and low Hartmann's procedure in an effort to preserve the rectum. This procedure has also been advocated for patients with severe anorectal Crohn's disease, in whom nonhealing of the perineal wound after proctectomy occurs with alarming frequency. The authors present a review of the literature and three cases of cancer developing in the defunctioning rectal stump despite surveillance proctoscopy.

METHODS: Twenty-five patients underwent low Hartmann's procedure for severe anorectal Crohn's disease. Surveillance proctoscopy was performed as follow-up. Development of cancer in the rectal remnant or anus or recurrence of symptoms was managed by resection and adjuvant therapy.

RESULTS: One patient developed squamous-cell carcinoma of the anal canal, underwent resection and adjuvant therapy, and was disease free at the time of this study. Two patients developed adenocarcinoma of the rectum. Both underwent resection and adjuvant therapy. One patient died and the other developed a recurrence.

CONCLUSIONS: The authors recommend interval perineal proctectomy in all patients undergoing low Hartmann's procedure for severe anorectal Crohn's disease in whom rectal preservation is not possible. Regularly scheduled interim surveillance proctoscopy performed every two years, with biopsies of macroscopically normal-appearing and abnormal-appearing rectal mucosa and curetting of fistulous tracts, is also recommended to decrease the possibility of missing occult malignancies. (Key words: Hartmann's procedure; Crohn's disease; Colitis)


The development of colon cancer in patients with Crohn's disease has been reported since the 1940s. Patients with Crohn's disease of the colon have undergone subtotal colectomy, ileostomy, and Hartmann's procedure, after failure of medical therapy, with good results. This operation was modified by Sher, et al., who performed a low Hartmann's procedure (LHP) in 25 patients with severe, septic, fistulizing anorectal Crohn's disease. In these patients, where nonhealing of the perineal wound had been a significant complication, they advocated leaving a short rectal stump rather than performing abdominoperineal resection.

Several cases of carcinoma developing in a retained rectal stump after surgery for Crohn's disease have been reported. The authors report two cases of rectal cancer and one case of anal cancer developing in patients with Crohn's disease. Two of these patients were part of the original 25 previously mentioned patients. One patient underwent his initial operation at another institution and was referred to the authors for management of his postoperative course. The lesions in these three patients developed despite proctoscopic surveillance of the rectal stump and resolution of most disease activity.

REPORT OF CASES

Case 1

A 36-year-old female diagnosed with Crohn's disease in 1982 at age 21 had been treated medically with 6-mercaptopurine for several years with good results. She sustained a colonic perforation after a motor vehicle accident in 1987 and required a right hemicolectomy, ileocolic anastomosis, and proximal loop ileostomy, which was closed two months later. She developed a high rectovaginal fistula in 1991 that required repair and a diverting loop ileostomy. Biopsy specimens of the fistulous tract were benign. The ileostomy was closed four months later. The fistula recurred shortly thereafter, and in 1992 she underwent an end ileostomy and LHP after again having negative biopsy results. Her perianal symptoms subsided for two years. Yearly surveillance proctoscopy was unremarkable. In 1994 she developed recurrence of the rectovaginal fistula and fistulas to the perineum and buttocks. At the time of total perineal proctectomy, a poorly differentiated squamous-cell carcinoma with transmural invasion of the rectum was found. Despite chemotherapy and...
radiation therapy, she developed a pelvic mass within months of the proctectomy. Small-bowel suspension and further radiation therapy to the pelvis were performed. A CT scan done one year later revealed two lesions in the right lobe of the liver for which she underwent a liver resection. At the time of surgery, inspection of the pelvis revealed no residual mass. Later in 1996 she was noted to have another mass in the area of the liver. She was re-explored and this mass was noted to be a celiac node involved with tumor, which was excised. Inspection of the liver and pelvis showed no evidence of gross disease. She was disease free at the time of this study.

**Case 2**

The patient was a 35-year-old female who was diagnosed with Crohn’s colitis in 1981 at age 19. She was treated medically with high-dose corticosteroids, adrenocorticotropic hormone, 6-mercaptopyrine, and intermittent antibiotics for four years. In 1984, after an exacerbation of her colitis, a subtotal colectomy, ileostomy, and Hartmann’s procedure was performed. She complied with surveillance proctoscopy approximately every two years. Her examinations revealed a normal rectum, which was suitable for ileorectal anastomosis. She had complete resolution of her symptoms until she became pregnant in 1993. At that time she began experiencing nonspecific pelvic pain, but no perianal disease. Her postpartum proctoscopy revealed an ulcerated lesion in the residual rectum. Biopsy revealed adenocarcinoma, and she underwent a total perineal proctectomy. Pathology revealed a Dukes A lesion. She did not receive any postoperative adjuvant therapy. Two years later she developed left hip pain and magnetic resonance imaging revealed a pelvic soft-tissue mass. A metastatic workup revealed multiple lung nodules. She underwent chemotherapy with methotrexate, 5-fluorouracil, and leucovorin and radiation therapy to the pelvis. Despite several cycles of chemotherapy the lung masses continued to enlarge and she ultimately died in 1996.

**Case 3**

The patient was a 54-year-old male who was diagnosed with Crohn’s disease in 1967 at age 25. He was treated with adrenocorticotropic hormone for ten years and then switched to prednisone for unknown reasons. During the next 15 years he continued to have episodic diarrhea but was essentially well controlled with medication. In 1992 he developed pneumaturia, fecaluria, and rectal pain and was diagnosed with a colovesical fistula. Medical management with high-dose steroids and antibiotics was unsuccessful. In 1994 he underwent a subtotal colectomy, ileostomy, and LHP. He remained asymptomatic after the surgery. In 1997, during surveillance proctoscopy, an adenocarcinoma of the rectum was discovered and abdominoperineal resection was performed. Pathology revealed moderately differentiated adenocarcinoma, with mucinous features and invasion of the perirectal fat. Despite adjuvant radiation and chemotherapy, he developed a recurrence in the pelvis.

**DISCUSSION**

Severe Crohn’s colitis unresponsive to medical therapy requires surgical treatment. When the rectum is free of disease or only minimally involved, subtotal colectomy and ileorectal anastomosis can be performed. Patients with severe anorectal Crohn’s disease have been shown to benefit from an LHP when used to avoid the problem of a nonhealing perineal wound. One problem with primary proctectomy in the presence of perineal sepsis is that nonhealing of the perineum is a common complication and may create more severe symptoms than before surgery. The incidence of nonhealing of the perineal wound 6 to 12 months after proctectomy ranges from 12 to 80 percent. It is for this reason that Sher et al. recommended performing a low Hartmann’s procedure, leaving behind a 3-cm to 5-cm rectal stump. This procedure was performed in 25 patients with severe, fistulizing anorectal Crohn’s disease, with an excellent rate of perineal healing. Fifteen of these patients required no further surgery. Of the ten remaining patients who underwent delayed perineal proctectomy, seven of them had a completely healed perineum within 12 months. The authors advocate performing LHP specifically in patients with perineal sepsis because of the improvement over abdominoperineal resection in perineal healing.

Crohn’s disease, like ulcerative colitis, carries a risk of developing a colorectal malignancy. Previous articles have noted the increased relative risk of colorectal carcinoma in Crohn’s patients to be as high as 4 to 20 times the general population. Some similarities can be observed in the cases reported in the literature. Most patients developed Crohn’s disease at a very