

Ileostomy Adenocarcinoma

A Case Report

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Abstract

Proctocolectomy is the standard treatment for patients suffering from ulcerative colitis which is long-standing, refractory to pharmacotherapy or otherwise associated with complications. An ileal pouch with pouch-anal anastomosis is fashioned in the majority of cases; however, in some instances terminal ileostomy is preferable. Ileostomy cancer is rare but a number of cases have been reported over the past twenty years suggesting a rising incidence. We report a case that developed thirty years post-operatively.

Case History

A 48 year old lady presented to our department with a one year history of a painless lesion arising in her ileostomy. The patient's main complaint was difficulty in securing the wafer of her ileostomy appliance. Thirty years previously our patient had a proctocolectomy with ileostomy for ulcerative colitis refractory to medical treatment.

On examination, the ileostomy was functioning properly. There was a hard polypoid lesion measuring 1.5 by 0.5cm arising from the inferomedial mucocutaneous junction of the ileostomy with an area of adjacent skin induration (measuring 0.5 by 0.5cm). Physical examination was otherwise unremarkable, the liver was not palpable and there were no enlarged inguinal

lymph nodes.

Excision biopsy of the lesion and the adjacent indurated skin was performed. Histologically, the lesion was a carcinoma made up of colonic-type glands, which invaded deep into the submucosa and infiltrated the adjacent skin. Stromal pools of mucin were a prominent feature. Immunohistochemical staining confirmed that the lesion was of local origin rather than a metastasis. Therefore, the pathological diagnosis was of a primary well-differentiated mucinous adenocarcinoma with local skin invasion.

A CT scan of the abdomen and pelvis was performed to stage the disease. It confirmed the absence of metastases. The patient underwent wide local excision of the stomal site and resection of the terminal 12cm of ileum. An ileostomy was refashioned. She made an uneventful recovery and was discharged home six days after surgery.

Histology of the excised skin and bowel revealed a background of chronic inflammation and colonic metaplasia in the mucocutaneous area. There was no evidence of residual neoplasia.

Discussion

Ileostomies are associated with an array of complications which are well documented. The development of primary cancers in ileostomies is considered rare. The interval between operation and neoplasia ranges from 2 to 39 years in the reported cases, with a mean of 22 years.¹⁻³ Only two cases were reported in the world literature before 1980. To our knowledge another thirty four⁴ such cases have been reported over the past twenty years, with the highest number occurring after operations performed for ulcerative colitis.^{1,5-7} A handful of cases have been reported following similar procedures performed for familial adenomatous polyposis.²⁻⁴ Two cases have been reported following ileostomy for Crohn's disease.⁸ This complication is not limited to the traditional Brooke ileostomy; cases have also been reported in continent ileostomies.⁷

It appears that the causative sequence starts with a chronic inflammatory process leading to a colonic-type epithelial metaplasia.^{1,9,10} It is thought that cytological atypia and architectural abnormalities may ensue in a process of dysplasia. This may ultimately lead to frank carcinoma. These tumours are usually slow-growing mucinous adenocarcinomas. They may present with a variety of symptoms including bowel obstruction and more trivial complaints such as ileostomy site irritation,

Keywords

Ulcerative colitis, ileostomy, adenocarcinoma, colonic metaplasia

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pain and bleeding.

It is important to note that not all polypoid lesions at an ileostomy site are cancerous. Attanoos and colleagues studied a series of 60 ileostomy polyps occurring in seven patients who received an ileostomy for ulcerative colitis.¹¹ Fifty of these polyps were inflammatory polyps associated with ileostomy prolapse. Another six polyps consisted of granulation tissue. Four polyps proved to be neoplastic - two were adenomas, one was an invasive adenocarcinoma and the other a mucinous adenocarcinoma. The authors recommend careful histological examination because neoplastic features may be overshadowed by inflammatory change.

The appropriate treatment for ileostomy carcinoma appears to be wide stomal site excision with re-fashioning of the ileostomy. Proper staging is necessary because these tumours have a significant recurrence and metastatic potential.^{12,13}

Conclusion

The population of patients with long-standing ileostomies is increasing. These patients are at risk of developing ileostomy adenocarcinoma. For this reason, we advocate a strategy of annual ileostomy surveillance, supplemented by more frequent examination by the patients themselves. Suspicious symptoms and lesions should prompt thorough examination with biopsy and careful histology. Early detection of malignant lesions leads to minimal surgical treatment.

References

1. Berman JJ, Ullah A. Colonic metaplasia of ileostomies: Biological significance for ulcerative colitis patients following total colectomy. *Am J Surg Pathol* 1989;13:955-60.
2. Gilson TP, Sollenberger LL. Adenocarcinoma of an ileostomy in a patient with familial adenomatous polyposis. Report of a case. *Dis Colon Rectum* 1992; 35:261-5.
3. Johnson JA 3rd, Talton DS, Poole GV. Adenocarcinoma of a Brooke ileostomy for adenomatous polyposis coli. *Am J Gastroenterol* 1993; 88:1122-4.
4. Iizuka T, Sawada T, Hayakawa K, et al. Successful local excision of ileostomy adenocarcinoma after colectomy for familial adenomatous polyposis: report of a case. *Surgery Today* 2002; 32:638-41.
5. Carter D, Choi H, Otterson M, et al. Primary adenocarcinoma of the ileostomy after colectomy for ulcerative colitis. *Dig Dis Sci* 1988; 33:509-12.
6. Bedetti CD, DeRisio VJ. Primary adenocarcinoma arising at an ileostomy site. An unusual complication after colectomy for ulcerative colitis. *Dis Colon Rectum* 1986; 29:572-5.
7. Cox CL, Butts DR, Roberts MP, et al. Development of invasive adenocarcinoma in a long-standing Kock continent ileostomy: report of a case. *Dis Colon Rectum*; 1997; 40:500-3.
8. Sherlock DJ, Suarez V, Gray JG. Stomal adenocarcinoma in Crohn's disease. *Gut* 1990; 31:1329-32.
9. Gadacz TR, McFadden DW, Gabrielson EW, et al. Adenocarcinoma of the ileostomy: the latent risk of cancer after colectomy for ulcerative colitis and familial polyposis. *Surgery* 1990; 107:698-703.
10. Reissman P, Avroutis O, Cohen DY, et al. Ileostomy-site colonic metaplasia with adenocarcinoma after proctocolectomy for ulcerative colitis. *Am J Gastroenterol* 1997; 92:1932-3.
11. Attanoos R, Billings PJ, Hughes LE, et al. Ileostomy polyps, adenomas, and adenocarcinomas. *Gut* 1995; 37:840-4.
12. Longo WE, Stephan RN, True LD. et al. Recurrent ileal mucinous adenocarcinoma in an ileostomy. *J Clin Gastroenterol* 1986; 8:192-4.
13. Sigler L, Jedd FL. Adenocarcinoma of the ileostomy occurring after colectomy for ulcerative colitis: report of a case. *Dis Colon Rectum* 1969; 12:45-8.

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