Mucinous adenocarcinoma at the ileostomy site after 35 years, in ulcerative colitis

Balachandra C. Uppin, Suryakant D. Mandolkar, Sudhakar R. Notu, Sharanabasavesh B. Alur

Dept of Surgery, B.L.D.E.A's Shri B. M. Patil Medical College and Research Centre, Bijapur - 586 103, Karnataka, India

For correspondence:
Suryakant D. Mandolkar, Plot No. 125, Rajendra Nagar, Opp. Manickbag Automobiles, Solapur Road, Bijapur – 586 103, Karnataka, India. E-mail: drsuryakant2001@yahoo.com

ABSTRACT

Mucinous adenocarcinoma at the ileostomy stump after 25 yrs following proctocolectomy for ulcerative colitis, has been documented rarely. We report a case of ileal adenocarcinoma at the mucocutaneous junction of an ileostomy, after 37 yrs. The incidence of this late complication of surgery for ulcerative colitis is increasing, as reviewed through previous reports.

Key words: Ileostomy, mucinous adenocarcinoma, mucocutaneous junction stoma, ulcerative colitis

How to cite this article:

Since the 1950s, the treatment for ulcerative colitis was colectomy combined with eversion ileostomy, revolutionized by Brooke. Numerous complications are associated with the ileostomy stoma. But the development of a carcinoma at the mucocutaneous junction of an ileostomy is an exceedingly rare complication; only few cases have been reported.

CASE REPORT

A 65 yrs old man attended the outpatient department occasionally in August-2002, for growth at the ileostomy stoma since 6 months, associated with blood discharge at the tumor site and pain. The patient had already undergone a pan proctocolectomy with ileostomy in 1965, for bleeding of diagnosed ulcerative colitis. Since then, the patient was keeping good health. On examination of the abdomen, there was a 4 x 3 x 3 cm. polypoid, friable, tumor mass adjacent to the ileostomy stoma superior border, encroaching onto the mucocutaneous junction. The lesion was seen to be arising from the junctional area of the ileal mucosa and skin. However, the ileal lumen appeared free. [Figure 1]. Hematological and radiological investigation of the abdomen revealed no abnormality. Edge biopsy histology of the lesion revealed infiltrating adenocarcinoma.

Wide excision of the mass with full thickness abdominal wall up to the peritoneum, was done. Refashioning of ileostomy was performed after removing about 6 cms. of ileum. The post-operative course was uneventful. The resected specimen contained terminal ileum with full thickness abdominal wall.

Histology of the tumor revealed pleomorphic tumor composed of abundant mucoid material with scattered glands lined by columnar and cuboidal epithelium, and large hyperchromatic nuclei with coarse chromatin pattern and prominent nucleoli. The mucoid pool showed signet ring cells, and stroma showed lymphohistiocytic infiltration suggestive of mucin-secreting adenocarcinoma of ileum at the ileostomy site [Figure 2].

DISCUSSION

Total colectomy with an eversion ileostomy was revolutionized in 1950s by Brooke, as surgical treatment in ulcerative colitis. The same may be undertaken in
Various complications are known to arise from an ileostomy, including skin excoriation, stenosis, intestinal obstruction, retraction, prolapse of the stoma, abscess, fistula formation, and ileitis. Conventional teaching does not recognize carcinoma as a complication of a long-standing ileostomy.

Development of a primary adenocarcinoma arising from a mucocutaneous junction of ileostomy without stomal complication is a rare occurrence, and our case brings the total reported cases around to 20.\(^1\) Of the cases reported previously, 16 had ulcerative colitis.

The interval between the formation of ileostomy and presentation of adenocarcinoma was 3-38 yrs. Most of them presenting after 25 years had an exophytic growth or induration, predominantly at the mucocutaneous junction. Most of the tumors were moderately well-differentiated mucinous adenocarcinoma. These tumors tend to be locally invasive and slow-growing, without great potential for early metastatic dissemination.\(^2\)

Previous reports reveal 3 cases with widespread metastasis, 10, 13, and 26 months after the initial diagnosis. Cuesta and Donner reported a well-differentiated adenocarcinoma of abdominal wall skin, 31 yrs after surgery, with no tumor in ileum and no metastasis.\(^2\)

Estimated incidence of 2 to 4 carcinoma per 1000 ileostomies, shows it to be a rare problem.\(^3\) In India, contrary to the earlier belief, ulcerative colitis is not considered any longer to be a disease of developed countries. It is responsible for 1% of total hospital admissions in a tertiary care hospital, and incidence of primary adenocarcinoma is unknown to us.\(^4\)

Numerous factors are responsible for cause of this rare condition, like preexisting backwash ileitis, placing an ileal mucosa in an abnormal environment with repeated physical trauma, and chemical irritation from ileostomy appliance. Intestinal flora in patient with ileostomies come to resemble that of colon, in bacteria-type and concentration.\(^3\)

Epithelial squamous metaplasia and hyperplasia of the skin have been described a common feature, when epithelial cells encroach on stomal mucosa. Evidence of colonic metaplasia and dysplasia have been found in mucosa adjacent to adenocarcinoma. Mucin studies have indicated a focal change to large bowel type mucin within the ileostomy mucosa, and within the tumor.\(^5\)

Although rare, it must be recognized that carcinoma is a late complication of ileostomy in ulcerative colitis.

Multiple and repeated biopsies should be undertaken around ileostomy of patients with fresh symptoms.

The increasing incidence is worrying, hence; implimentation of screening of long-standing ileostomies by biopsy may be necessary for early diagnosis.

Advanced surgical techniques of present times, like stapler ileo-anal anastomosis following total colectomy, have eliminated the ileostomy procedure, which was practiced since the 1950s. This technique would reduce incidence or eliminate the reported complication. The future concern would be regarding the probable change in character of presentation of this rare complication as adenocarcinoma, at the ileo-anal anastomotic site.\(^1\)

En bloc resection of ileostomy, wide resection of the adjacent anterior abdominal wall, and transposition of the stoma to a new site, has been shown to provide the
Mucinous adenocarcinoma at ileostomy site in ulcerative colitis

best prognosis for an adenocarcinoma arising from an ileostomy.\cite{1}

**REFERENCES**