Isolated colostomy site recurrence in rectal cancer-two cases with review of literature

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Abstract

Background: Colostomy site carcinomas are rare with only eight cases reported in the world literature. Various etiological factors like adenoma-cancer sequence, bile acids, recurrent and persistent physical damage at the colostomy site by faecal matter due to associated stomal stenosis have been considered responsible. Two such cases are being reported and in both cases there was no evidence of any local recurrence in the pelvis or liver and distant metastasis. Both patients had received adjuvant chemotherapy following surgery.

Case presentation: First case was a 30-year-old male that had reported with large bowel obstruction due to an obstructing ulcerative growth (poorly differentiated adenocarcinoma) at the colostomy site after abdomino-perineal resection, performed for low rectal cancer six years previously. Wide local excision with microscopically free margins was performed with a satisfactory outcome. Four years later he presented with massive malignant ascites, cachexia and multiple liver metastasis and succumbed to his disease.

Second case was a 47-year-old male that presented with acute large bowel obstruction due to an annular growth (well differentiated adenocarcinoma) in the upper rectum. He was managed by Hartmann’s operation and the sigmoid colostomy was closed six months later. Five years following closure of colostomy, he presented with two parietal masses at the previous colostomy site scar, which, on fine needle aspiration cytology were found to be well-differentiated adenocarcinomas of colorectal type. Surgery in the form of wide local resection with free margins was performed. He presented again after five years with recurrence along the previous surgery scar and an incisional hernia and was managed by wide local excision along with hernioplasty. Follow-up of nine years following first surgery is satisfactory.

Conclusion: Colostomy site/scar recurrence of rectal carcinoma is rare and could be due to various etiological factors, although the exact causative mechanism is not known. Surgery with microscopically free margins is recommended in the absence of metastatic disease. Stenosis of the stoma is considered as one of the most important contributory factors and should be followed carefully.

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This is an Open Access article distributed under the terms of the Creative Commons Attribution License (http://creativecommons.org/licenses/by/2.0), which permits unrestricted use, distribution, and reproduction in any medium, provided the original work is properly cited.
Background
Metachronous carcinomas rarely occur at the colostomy site and only eight cases have been reported previously. Various factors like adenoma-cancer sequence, stenosing stoma or bile acids have been implicated. Colon cancer presenting as cutaneous metastasis in an old operative scar has also been reported [1-6]. Possible etiological factors include an alteration in the microscopic anatomy around the scar, perhaps in the lymphatic channels, altered adhesion molecule profile or altered local immunosurveillance mechanisms leading to change in the local environs of the scar which become more receptive to metastatic tumor cells [7]. The occurrence being so rare, no definite etiology and management protocol is known. Management in the form of curative surgery along with adjuvant chemotherapy is recommended.

Case presentation
Case-1
A 30-year old male had been operated in March 1996 for low rectal cancer (T4N0M0) in the form of classical abdomino-perineal resection with microscopically free (R0) margins and satisfactory outcome. He was on a regular follow-up for three years and was subsequently lost to follow-up. In April 2002 he presented with acute large bowel obstruction due to complete stenosis and infiltration of the stoma site by an ulcerco-proliferative growth, which was extending into the surrounding skin and the abdominal muscles (figure 1). He had been having increasing narrowing of the stoma for the past one year and was managed by his family doctor by regular finger dilatations. Biopsy from the growth was suggestive of poorly differentiated adenocarcinoma. The ultrasound and computed tomography scan ruled out any local recurrence in the pelvis or metastases in the liver or ascites. Blood biochemistry including liver function tests was within normal limits except for anaemia (Hb = 10 gm%) and raised carcino-embryonic antigen (CEA = 10.7 ng/ml, normal being up to 2.5 ng/ml). He was managed by wide local excision including the stoma with wide margin, the entire descending colon, 2/3rd of the transverse colon in bloc with the involved para and pre-aortic lymph nodes. The left ureter, which was encased within the lymph node mass, was dissected free and right-sided transverse colostomy was done (figure 2). The histopathological examination revealed poorly differentiated adenocarcinoma of the rectum (T4N1M0). The colostomy was closed after eight months and the regular follow-up was satisfactory. He received adjuvant chemotherapy in three weekly (total 12 cycles). He was again lost to follow-up and presented subsequently in August 2006 with cachexia, massive malignant ascites, and liver metastasis and succumbed to his disease.

Case-2
A 45-year old male presented to the surgical emergency in Aug 1997 with features of acute large bowel obstruction of three days duration due to an annular growth in the upper rectum without any evidence of liver or peritoneal metastasis. Due to poor general condition (anemia, deranged electrolytes) and the unprepared bowel, he was managed by Hartmann’s operation. The histopathological examination of the resected specimen revealed well-differentiated adenocarcinoma of the rectum (T4N1M0). The colostomy was cut after eight months and the regular follow-up was satisfactory. He received adjuvant chemotherapy in...
the form of Levamisol and 5 fluorouracil. He was subse-
sequently lost to follow-up and reported back five years later
in 2002 with two parietal lumps in the left lumbar region
and the fine needle aspiration cytology showed them to
be well-differentiated adenocarcinoma of colorectal vari-
ety. The Ultrasound and contrast enhanced computed
tomographic scan (CECT) showed infiltration in to the
muscles of the anterior abdominal muscles and the peritoneum. A wide local excision including the abdominal
muscles was done and the wound was closed in layers. He
reported back in May 2005 with a recurrent 2 × 3 cm pari-
etal nodule along the scar of the previous colostomy and
an incisional hernia. The scar recurrence was excised with
an R0 margin and the hernia was repaired using an onlay
prolene mesh. The histopathological examination of the
specimen revealed well-differentiated adenocarcinoma of
colorectal type (figure 3). The postoperative recovery was
good. He was discharged on the seventh day and a regular
follow up of one year (nine years following first surgery)
is satisfactory.

Discussion
Colon is not an uncommon site for synchronous and met-
achronous malignancies. The criteria for multiple carcino-
mas of the colon are well laid out [6]. In a study, multiple
carcinomas accounted for 4.3% of all colorectal carcino-
mas, and for 12.9% of colorectal carcinomas and poly-
poid cancers of the colon (synchronous 4%; metachronous 2.0%) [6-8].

However, adenocarcinoma occurring at the colostomy site
with no recurrence in the pelvis is very rare. Only eight

Figure 2
Eight months after second surgery with transverse colostomy on the right side. The well-healed transverse scar on left side of
abdomen may be seen.
Table 1: Cases reported in the literature earlier with two present cases

| Investigator          | Year of report | Past history (Age of first surgery in years) | Age at presentation with colostomy site adenocarcinoma(years) | Sex | Time of development of carcinoma at the colostomy site |
|-----------------------|----------------|---------------------------------------------|----------------------------------------------------------------|-----|--------------------------------------------------------|
| Takami et al. [3]     | 1983           | Rectal cancer(19)                          | 38                                                             | M   | 19 years after APR                                     |
| Saegusa [10]          | 1986           | Rectal cancer(55)                          | 60                                                             | M   | 5 years after APR                                      |
| Nakano et al. [1]     | 1987           | Rectal cancer(31)                          | 53                                                             | F   | 22 years after APR                                     |
| Takeyuchi et al. [4]  | 1990           | Rectal cancer(44)                          | 56                                                             | M   | 12 Year after APR                                      |
| Ohta et al. [2]       | 1991           | Rectal cancer(68)                          | 77                                                             | F   | 9 Years after APR                                      |
| Ishikawa et al. [1]   | 1994           | Rectal cancer(46)                          | 76                                                             | F   | 30 Years after APR                                     |
| Ohtsuka et al. [5]    | 1996           | Rectal cancer(77)                          | 81                                                             | M   | 4 Years after APR                                      |
| Shibuya et al. [1]    | 1997           | Rectal cancer(73)                          | 81                                                             | M   | 8 Years after APR                                      |
| Our patient case-1    | 2006           | Rectal cancer(30)                          | 36                                                             | M   | 6 years after APR                                      |
| Our patient case-2    | 2006           | Rectal cancer(45)                          | 50                                                             | M   | 5 years after LAR                                      |

APR: Abdominoperineal resection
LAR: Low anterior resection with Hatmann’s operation

Figure 3
The histopathological examination of the resected recurrent colostomy site specimen showing well-differentiated adenocarcinoma (40×).
Presence of enterobacteria and bile acids in the stools has also been implicated and reported and the carcinomas occurring at a stoma with which stools were briefly in contact appeared to be rare [2]. Amongst the eight known cases (table 1), three cases including one of the two reported cases may have been affected by bile acids and changes in enterobacteria that resulted from prolonged contact with stools.

There could be a possible similarity in the mechanisms involved in the recurrence at colostomy site to that of recurrence of cancer in a scar of previous surgery and various causative mechanisms have been suggested. There may be a direct extension of the disease, hematogenous spread, lymphatic spread or implantation of exfoliated tumor cells if the specimen had been retrieved through the incision [7]. This is however unlikely to be the scenario in both the presented cases particularly in the first case with recurrence occurring at the site of colostomy. In the second case, the recurrence developed along the scar of previously performed colostomy and this case could resemble in presentation to an incisional scar. Predilection to metastasis at incision site has also been postulated to be due to alteration in the microscopic anatomy around the scar, perhaps in the lymphatic channels, making it more receptive to metastatic tumor cells, possibly due to altered adhesion molecule profile or altered local immunosurveillance mechanisms. The exact pathogenesis however remains a matter of speculation awaiting further recognition and investigation [1,7-10].

**Conclusion**

Carcinomas may occur coincidentally at a stoma, but the stricture of the stoma and the physical stimulation like regular pressure of tight clothing, could promote their occurrence. The stricture of a stoma should be followed carefully and patient informed about the possibility of development of carcinoma at the colostomy site. When and if detected, surgery with a curative intent (R0 resection) is associated with a good outcome if there are no liver, peritoneal or distant metastases.

**Competing interests**

The author(s) declare that they have no competing interests.

**Authors’ contributions**

CM was the chief surgeon in charge of the case and VS was the first surgical assistant; AB and SS were the pathologists responsible for the histopathological examination, DB assisted in the preparation of the manuscript. All authors read and approved the manuscript.

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Informed consent was obtained from the relative of the first case, and the patient in the second; regarding permission to publish this case in the *World Journal Of Surgical Oncology*.

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