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Isolated Carcinoma in Colostomy Site Following Proctectomy for Recurrent Rectal Cancer

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Background	Metachronous or recurrent cancer at a stoma site following resection for colorectal cancer is extremely rare, as only a handful of case reports can be found in the literature.
Summary	We present a review of the literature and case report of isolated carcinoma at colostomy site two years after abdominoperineal resection for recurrent rectal cancer.
Conclusion	Whenever stomal enlargement, persistent bleeding, bruising or stomal obstructions present, the surgeon must have high level of suspicion for cancer recurrence. These symptoms should be carefully investigated and there should be a low threshold for stomal mucosal biopsy.
Keywords	Recurrent rectal cancer, isolated stoma site recurrence, recurrence in colostomy

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Case Description

Metachronous or recurrent cancer at a stoma site following resection for colorectal cancer is extremely rare, as only a handful of case reports can be found in the literature. We present a review of the literature and case report of recurrent cancer at a colostomy site two years after abdominoperineal resection (APR) for recurrent rectal cancer.

A 66-year-old female with no significant past medical history underwent neoadjuvant chemotherapy followed by low anterior resection with J pouch and diverting loop ileostomy for rectal cancer in another institution. The pathologic specimen revealed T3N1 rectal cancer and the patient subsequently underwent adjuvant chemotherapy. The loop ileostomy was reversed 3 months later.

Four years later, surveillance colonoscopy revealed a posterior rectal ulcer, just distal to the anastomosis, positive for moderately differentiated adenocarcinoma of the rectum. Pelvic magnetic resonance imaging (MRI) demonstrated circumferential rectal wall thickening. The patient underwent APR with creation of end sigmoid colostomy. The pathology report revealed a 4.1 cm, T3N0, moderately differentiated adenocarcinoma. The mesorectal excision appeared to be incomplete, the tumor was focally presented at the circumferential resection margin, and focal perineural invasion was identified. The tumor had pushing growth border, the distal resection margin was free of tumor, no vascular invasion noted, there were no tumor deposits, and there were 4 benign lymph nodes. The patient underwent adjuvant chemotherapy. The patient had a normal follow-up colonoscopy at one-year post-APR.

Two years after the second operation the patient had complaints of stomal bruising and enlargement, requiring larger pouches. On physical examination, the stoma had raised borders circumferentially, with diffuse granularity and induration (Figure 1).

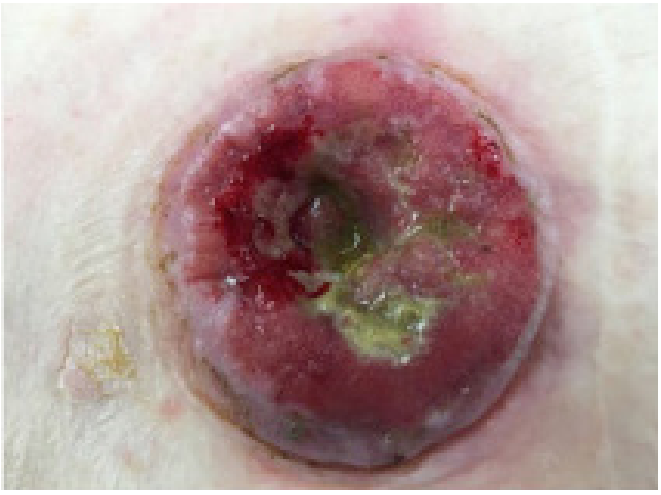

Figure 1. Stoma recurrence, preoperative

Bedside biopsy of the colostomy mucosa was obtained. Pathology report showed moderately differentiated adenocarcinoma. Positron emission tomography (PET)/computer tomography (CT) scan, pelvic MRI, and colonoscopy did not show metastatic disease or synchronous lesions. The patient underwent exploratory laparotomy with wide local excision of the colostomy and distal 5 cm of colon with its mesocolon. The surrounding abdominal wall tissue was excised en bloc and the colostomy was relocated to the contralateral side. The fascia and skin were closed primarily (Figure 2 and Figure 3).

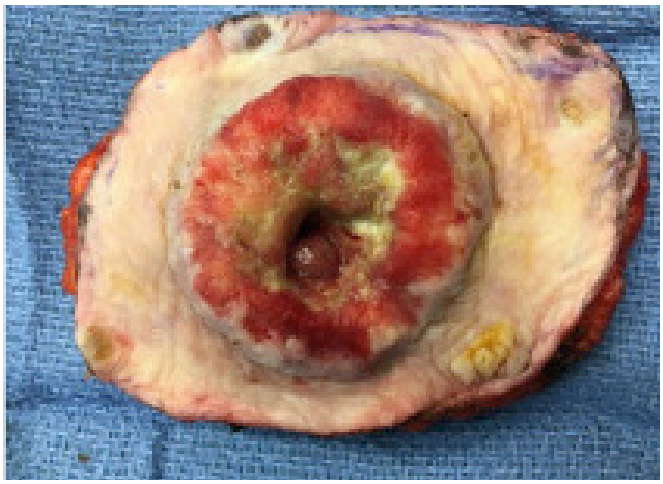

Figure 2. Local excision of stoma and surrounding tissues

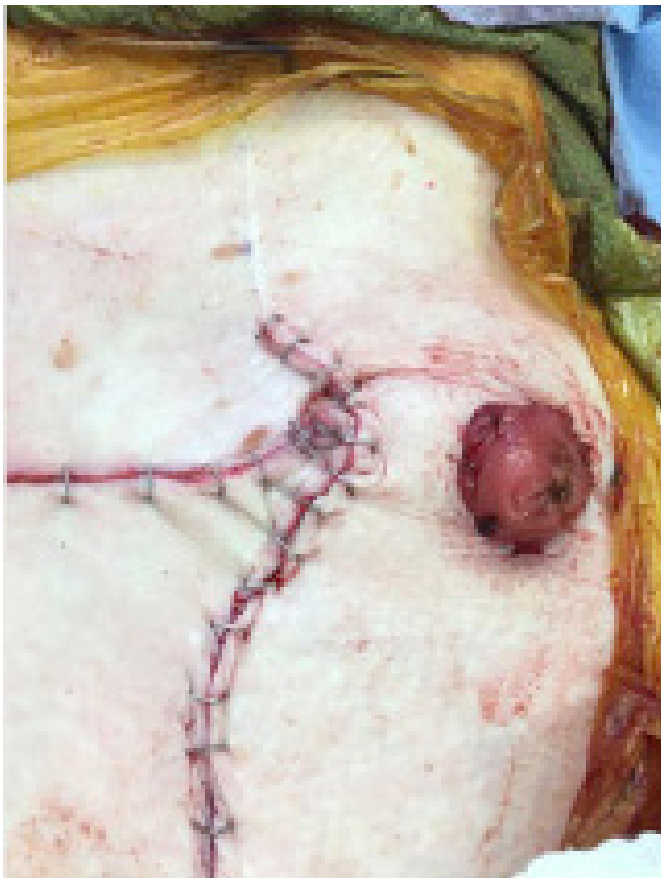

Figure 3. Stoma relocation and reconstruction

There was no other evidence of metastatic disease in the peritoneal cavity. The pathology report was consistent with moderately differentiated colonic adenocarcinoma involving the colonic wall and adjacent skin and subcutaneous tissue. All margins were tumor-free.

Discussion

Although recurrent colon and rectal cancer is common, rectal cancer with isolated recurrence at the colostomy site is rarely encountered. We generated a PUBMED and MEDLINE search for recurrence of colorectal cancer in a stoma site. Only a few case reports exist in the English literature. The origin of this tumor may be either a metachronous colonic lesion, metastasis to the peristomal skin, or implantation of tumor cells during the initial cancer operation. Both metastatic disease and local recurrence typically present within the first 2 to 4 years after surgery for colorectal cancer.[1,2] In previously described case reports, the stoma site cancer was discovered much later (Table 1). Metachronous or synchronous colorectal cancer occurs in 5 to 10% of colorectal cancer cases and are defined as tumors of the colon that do not occur in the anastomotic site and are not suspected for metastasis or extension of other tumors.[3]

Author	Year of publication	Time from primary resection to recurrence (years)
Takami et al [8]	1983	19
Saegusa et al [9]	1986	5
Nakano et al [10]	1987	22
Ohta et al [11]	1991	9
Ishikawa et al [12]	1994	30
Ohtsuka et al [13]	1996	4
Shibuya et al [6]	1997	8
Chintamani et al [5]	2007	6
Chintamani et al [5]	2007	5
Greenberg et al [14]	2007	0.5
Vijayasekar et al [15]	2008	14
Okamoto et al [16]	2009	15
Kuo et al [4]	2012	0.5
Iwamoto et al [17]	2015	27

Table 1. Previous case reports of recurrent cancer in colostomy site

In our case report, the fact that the recurrence occurred two years after the initial procedure in combination with the pathology report showing threatened circumferential margins and incomplete total mesorectal excision (TME), increases the likelihood that this is an isolated metastasis. Recurrence at a previous colo-colonic anastomosis is common and is likely due to intraluminal malignant cells that are present at the time of resection and implant the healing anastomotic site. We suspect that malignancy found at a colostomy site may have the same mechanism (implantation of malignant cells at healing site) and therefore suspect that this is more likely a recurrence rather than metachronous malignancy.[4] Unfortunately, we do not have genetic testing of either specimen. If on genetic testing the specimens were similar, it would be obvious that this is a recurrence rather than metachronous malignancy.

Stoma site cancer can manifest symptoms such as stoma site bruising, mucosal bleeding, gradual enlargement of the stoma, or stoma site obstruction. Previous case reports describe an extraordinarily long delay in diagnosis due to the slow, progressive nature of the symptoms. Chintamani et al described a case in which the patient presented with stoma obstruction managed with regular finger dilations by his family physician for over one year before a biopsy was performed and the cancer diagnosis was made.[5]

Shibuya et al also described a case with a delayed diagnosis. The patient had presented multiple times to the local emergency room with symptoms of obstruction. The patient was discharged and treated with manual dilations for over one year. In another case report, the patient suffered from a growing peristomal mass for more than five years until he decided to see a physician. At the time of his initial presentation, the size of the tumor was more than 16 cm at its greatest dimension.[6] Our patient presented with complaints of an "enlarging stoma" and peristomal bleeding, which we initially thought was secondary to over granulation and hyperplasia at the edge of the stoma. A biopsy was finally taken, and the diagnosis confirmed only after one year of persistent symptoms.

There are no specific guidelines for the treatment of this rare condition and it is our practice to discuss all rectal cancer cases in our multi-disciplinary team (MDT) conference, which is represented by at least one member of each of the following disciplines: surgery, pathology, radiology, medical oncology and radiation oncology.[7] Treatment in this case was individualized in accordance with the National Comprehensive Cancer Network (NCCN) guidelines for recurrent rectal cancer to achieve a wide local resection of the stoma including skin, subcutaneous tissues, and the colon with clear margins. Adjuvant chemotherapy will be administered. The surveillance strategy will include history and physical examination with specific focus on inspection of the colostomy, CEA levels and chest/abdomen/pelvis CT every three to six months for the first two years and then every six months for a total of five years. Colonoscopy will be performed at one year after surgery and follow-up colonoscopy will depend on findings.

Conclusion

Rectal cancer recurrence at the colostomy site is extremely rare, as demonstrated by the limited amount of published case reports and supporting literature. Whenever stomal enlargement, persistent bleeding, bruising, or stomal obstructions present, the surgeon must have a high level of suspicion for cancer recurrence. These symptoms should be carefully investigated and there should be a low threshold for stomal mucosal biopsy.

Lessons Learned

Carcinoma in the colostomy site following proctectomy for rectal cancer is rare and mandates a high level of suspicion from the clinician. Possible mechanisms of stoma site carcinoma are recurrence, metachronous malignancy, or metastases. Treatment is based on multi-disciplinary team approach and is patient tailored.

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