

Implantation metastasis from adenocarcinoma of the sigmoid colon into a perianal fistula: a case report

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Abstract

Implantation metastasis from a colorectal cancer into a perianal fistula is very rare. Such lesions are commonly mistaken as benign perianal abscesses or fistulas and diagnosed only after pathological analysis of surgically excised fistulas. Once diagnosed, the management of this condition remains controversial. We report a case of perianal fistula that was unexpectedly found to harbor adenocarcinoma on biopsy. Further investigation by colonoscopy and computed tomography scan revealed a sigmoid adenocarcinoma. Abdominoperineal resection was performed. Histology and immunohistochemical staining was identical in both primary and metastatic tumors. We herein review the literature on the metastasis of colorectal cancer to a benign perianal fistula presumably acquired through implantation of viable malignant cells shed from the primary tumor and discuss the approach to this rare scenario in colorectal cancer surgery.

Keywords Rectosigmoid cancer, anal fistula, implantation metastasis, abdominoperineal resection

Ann Gastroenterol 2014; 27 (3): 276-279

Introduction

Charles Ryall in 1907 first reported implantation metastasis of solid cancers and described this phenomenon as 'cancer infection'. Spread of colorectal cancers can be lymphatic, hematogenous, transperitoneal, or by direct extension. Colorectal cancer spreading by implantation though rare has been described in hemorrhoidectomy wounds [1], colonoscopic biopsy sites [2], laparoscopic port sites [3], at site of perianal injury site at introduction of EEA stapler at anterior resection [4] and Lonestar retractor insertion site after coloanal anastomosis [5]. The first case of cancer implantation to a perianal fistula was reported by Guiss in 1954. Since then, there have been several cases reported and the concept of colorectal cancer implantation metastasis into a benign perianal fistula has become widely accepted. The management of this condition however still remains a matter of controversy.

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Conflict of interest: None

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Received 30 November 2013; accepted 15 December 2013

Case report

A 65-year-old man with a two-year history of a perianal fistula and recent worsening of symptoms underwent fistulectomy with the track completely excised. Histopathology revealed an unexpected diagnosis of moderately differentiated adenocarcinoma. A contrast-enhanced computed tomography of the abdomen and pelvis was performed which revealed a sigmoid tumor (Fig. 1). Colonoscopy revealed a normal rectum, but a



Figure 1 Axial section of a contrast-enhanced computed tomography scan of the abdomen and pelvis showing the sigmoid tumor (marked with arrow)

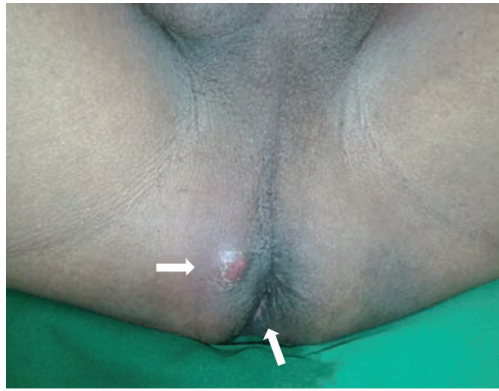


Figure 2 Intraoperative photograph showing the perianal scar at the site of excised fistula and the perianal abscess (marked with arrows)

circumferential ulcerating lesion 18 cm from the anal verge, confirmed by histology to be a moderately differentiated adenocarcinoma. He was referred to us for further management. The perianal scar was soft with no palpable mass. A complete staging work-up found no other evidence of metastatic spread. A day before the surgery he developed another perianal abscess (Fig. 2). Surgery consisted of abdominoperineal resection (APR) with excision of the sigmoid mass, perianal mass and the scar site of excised fistula (Fig. 3). Pathological results of the sigmoid tumor revealed moderately differentiated adenocarcinoma with focal signet ring cell morphology up to subserosal fat without lymph

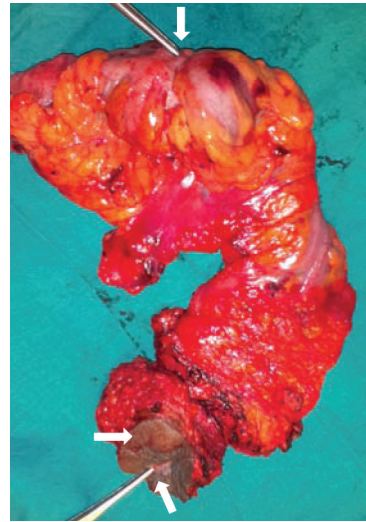


Figure 3 Photograph of the abdominoperineal resection specimen showing the perianal scar at site of the excised fistula, the perianal abscess and the sigmoid tumor (marked with arrows)

node involvement of 12 lymph nodes dissected (T3N0M0) with MSI-H histology (Fig. 4 A). The perianal fistula tract (Fig. 4 B,C) also revealed moderately differentiated adenocarcinoma with extracellular mucin amidst acute inflammation. Tumor was seen infiltrating the perianal skeletal muscle; however lateral and distal resection margin were free of tumor. The site of perianal abscess showed

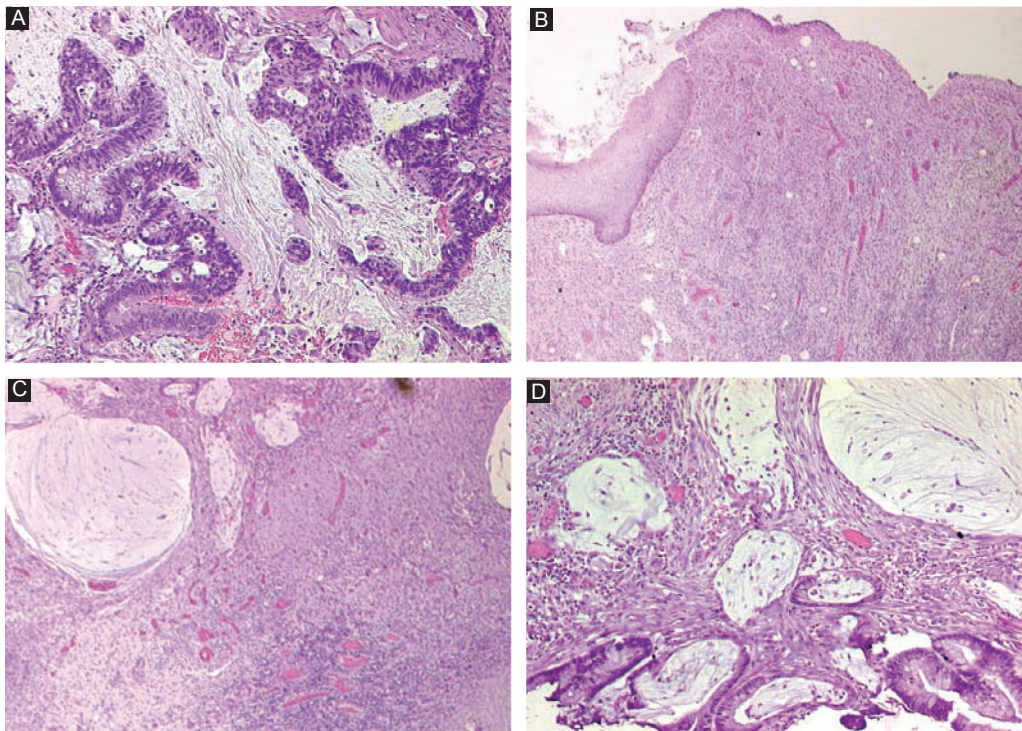


Figure 4 (A) Sections showing moderately differentiated adenocarcinoma of the sigmoid (H& E x 100x); (B) and anal fistula tract lined by inflammatory granulation tissue (H& E x 40x); (C) with tiny foci of adenocarcinoma in the fistulous tract along with pools of extracellular mucin (H& E: 40x); (D) and high magnification showing similar tumour in anal fistula tract as in the sigmoid (H& E: 100x)

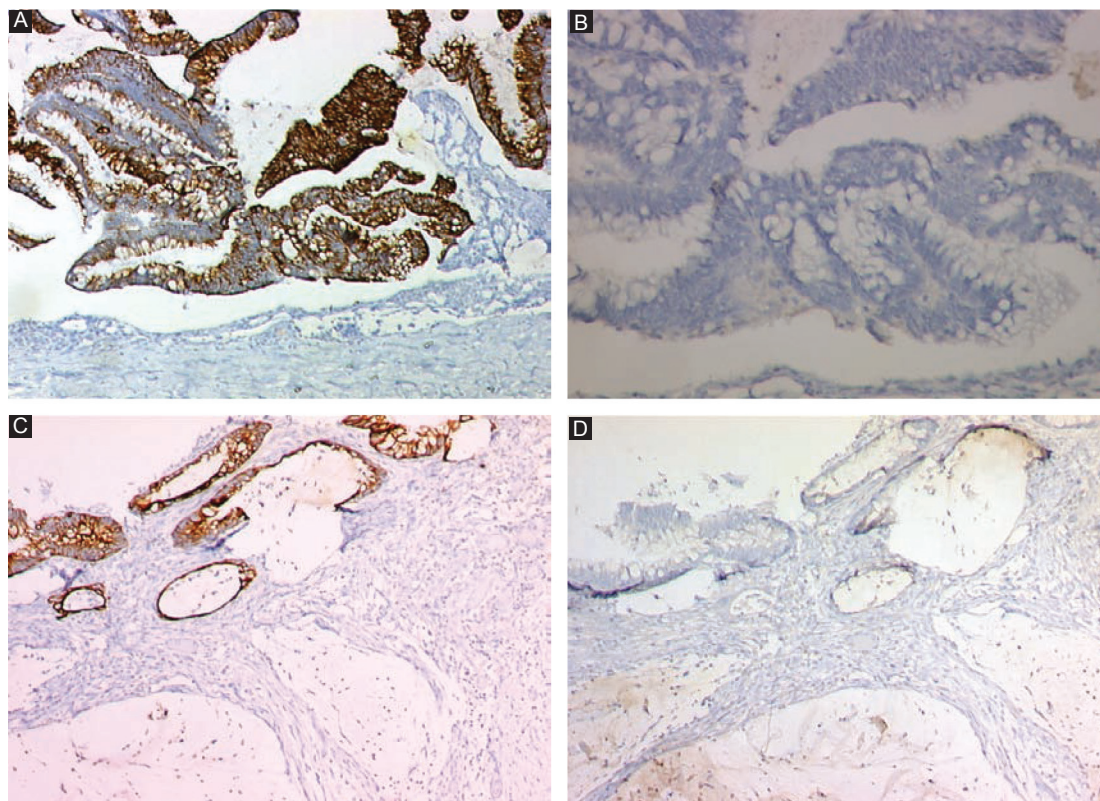


Figure 5 (A) Sections showing sigmoid adenocarcinoma immunopositive for cytokeratin 20; (B) while immunonegative for cytokeratin 7; (C) and perianal adenocarcinoma immunopositive for cytokeratin 20; (D) while immunonegative for cytokeratin 7

dense acute organizing inflammation with no evidence of tumor. The histologic appearances of the two malignancies were similar (Fig. 4 A,D). Immunohistochemistry for cytokeratin (CK) 7 and CK20 was performed to distinguish colorectal adenocarcinoma from anal gland carcinoma. Both the sigmoid cancer and the perianal tumor were CK7- and CK20+ and showed a similar pattern (Fig. 5A-D). The patient recovered uneventfully from surgery and no adjuvant chemotherapy or radiotherapy was planned. The patient remains well at 3 months of follow up, with no local recurrence or distant metastases identified.

Discussion

Implantation metastasis from a colorectal cancer to a perianal fistula is a very rare entity. As in our case report, the symptoms in such patients are often attributed to their perianal disease and the primary carcinoma is missed until pathological examination of fistula is reported to harbor adenocarcinoma. It is therefore important that all cases of fistula and anorectal abscess, tissue should be examined pathologically. Also of importance would be a high level of suspicion if there is a change in the behavior of a chronic anal fistula noted by increased discharge, bloody discharge, nodule formation, slow healing or scar induration making biopsy and further colonic investigation mandatory.

The other possibility of finding adenocarcinoma in a perianal fistula is a malignant degeneration occurring within a long-standing anal fistula. Such malignant degeneration can be either squamous or mucinous and though uncommon has been also reported. Rosser first described in 1931 the 3 basic criteria to determine that a fistula has undergone malignant transformation [6]. These include that the fistula should have been present for a minimum of 10 years, there should be no evidence of tumor within the rectal or anal canal mucosa and the internal opening of the fistula should be devoid of malignancy.

Implantation metastasis also has to be distinguished from a rectal carcinoma presenting as a fistula or as acute perirectal sepsis wherein the primary carcinoma itself has fistulated.

Lastly, very rarely can a mucinous carcinoma can occur within an unrecognized rectal duplication presenting as a perianal mass or discharge [7].

Immunohistochemical staining of CK7 and CK20 is used to distinguish colonic tumor tissues from tumors originating from the anal glands. Anal glands are strongly immunoreactive with antibodies to CK7 but not to CK20, whereas colorectal adenocarcinomas show strong immunoreactivity for CK20 but not to CK7 [8].

In our reported case there was a lack of dysplasia in the surrounding rectal mucosa or anal glands with a distant location of the primary tumor in sigmoid colon. As both the sigmoid cancer and the perianal tumor showed the same type of tumor, histological differentiation and were CK7- and CK20+ we considered the tumor of the anal fistula as implantation

metastases from the sigmoid adenocarcinoma.

Controversies remain in the management of metastatic anal fistula. There are no specific recommendations for the management of colorectal cancer implantation metastases to perianal fistulae. Some authors report to have performed APR, while others have chosen local resection. However, increasing recent reports have shown successful management with sphincter sparing surgery combined with local resection with or without radiotherapy or combined with local resection after neoadjuvant chemoradiotherapy to manage the implantation metastasis without local recurrence [9,10]. An APR was performed in this reported case because of associated perianal sepsis which was probably after inadequate local excision of fistula. It is important to perform a wide excision with confirmation of negative margins if only a local excision is performed. A clinical examination revealing a soft supple scar after a patient has undergone surgery for presumably benign disease may not be representative as in our case as residual tumor was demonstrated on reoperation.

In conclusion, any excised perianal fistula should be pathologically examined. A change in the behavior of a chronic perianal fistula mandates biopsy and further colonoscopic examination. It is difficult to make a conclusion about the best approach to implantation metastasis to a benign perianal fistula because of the rarity of this condition and due to the short follow up of the reported cases. Sphincter-saving surgery may be an option.

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