

Implantation metastasis of rectosigmoid cancer in an anal fistula

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ABSTRACT

Implantation metastasis of colorectal cancer in an anal fistula is very rare. We report a case of a 61 years old male who underwent fistulectomy for an anal fistula. Histopathology unexpectedly revealed adenocarcinoma in the fistula track, however the patient refused further treatment. Sixteen months later he presented with an obstructing locally advanced rectosigmoid cancer found to be fixed into the pelvic wall. An ileosigmoid bypass was fashioned and he was treated with neoadjuvant chemoradiotherapy followed by high anterior resection. Histopathology confirmed a colorectal adenocarcinoma, Immunohistochemistry of the tumors from both sites was CK7-/CK20+. The patient died 34 months later with liver and lung metastasis however no perianal recurrence occurred. Local resection with or without radiotherapy, instead of abdominoperineal resection, was feasible for control of perianal metastatic lesion implanted from colorectal cancer if local extended resection was possible.

KEY WORDS: Implantation metastasis; Colorectal cancer; Anal fistula.

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INTRODUCTION

Adenocarcinoma in an anal fistula is rare. It can arise as a complication of long-standing chronic inflammation or by metastatic implantation from a colorectal cancer. We present a case of anal fistula implantation metastasis from a rectosigmoid adenocarcinoma successfully managed with local excision and review the literature. The evidence suggests local excision appears to provide good local control if negative margins can be achieved.

CASE REPORT

A 61-year-old man with a six month history of perianal pain and purulent discharge underwent fistulectomy in June 2006 for a subcutaneous anal fistula. The internal and external openings of the fistula were clearly identified at the operation and the track was completely excised. Histopathology revealed an unexpected diagnosis of moderately differentiated adenocarcinoma near the external opening (Fig.1). However the patient refused further investigation. The incision was completely

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healed five weeks later. Sixteen months later he presented with fecal urgency and PR bleeding. Colonoscopy revealed a circumferential ulcerating lesion 18cm from the anal verge, confirmed on biopsy to be a moderately differentiated adenocarcinoma. The perianal scar was soft with no palpable mass. Computerized tomography (CT) showed irregular thickening of rectosigmoid wall and a locally invasive mass (Fig.2). Laparotomy revealed an irresectable 10×12cm mass fixed to the retroperitoneum, the terminal ileum and the right psoas muscle. Side-to-side ileo-ileostomy and sigmoid colostomy were fashioned and the patient was subsequently treated with seven cycles of sandwich FOLFOX4 chemotherapy and a total of 50-Gy of radiotherapy. Six weeks following completion of treatment CT showed downsizing of the mass (Fig.2) and the patient underwent anterior resection, small bowel resection with defunctioning ileostomy. Histopathology revealed a ypT3N0 moderately differentiated adenocarcinoma. Immunohistochemistry stained CK7-/CK20+ (Fig.1). The patient made a good recovery and

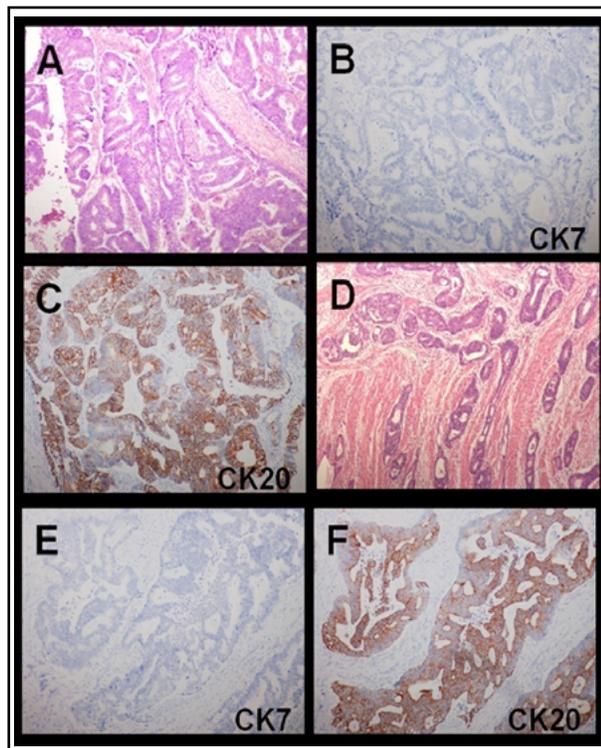


Fig.1: A, B and C shows primary rectosigmoid cancer, D, E and F shows perianal tumor. In hematoxylin and eosin staining, both tumor were moderate-differentiated adenocarcinoma. In immunohistochemistry of CK7 and CK20, both tumor showed CK7 negative (B and E) and CK20 positive pattern (C and F).

was treated with five further cycles of FOLFOX4 chemotherapy. In July 2009, screening CT revealed widespread liver and lung metastases. However there was no evidence of recurrent perianal disease. The patient died from pulmonary failure 17 months later.

DISCUSSION

Adenocarcinoma can arise in an anal fistula through two mechanisms. Malignant change can occur secondary to longstanding chronic inflammation or malignant implantation of cancer cells may occur from a remote site. Ryall¹ was the first to report implantation metastasis in 1907. This is now a well accepted mechanism for the metastatic recurrences observed in surgical wounds and port sites. Shed cancer cells are unable to implant on intact epithelial membranes, however, de-epithelialised surfaces such as hemorrhoidectomy wounds^{2,3}, biopsy tracks⁴ and anastomotic site⁵ provide a more fertile environment for implantation. Implantation metastasis from colorectal adenocarcinoma to anal fistula is rare. Guiss⁶ reported the first case in

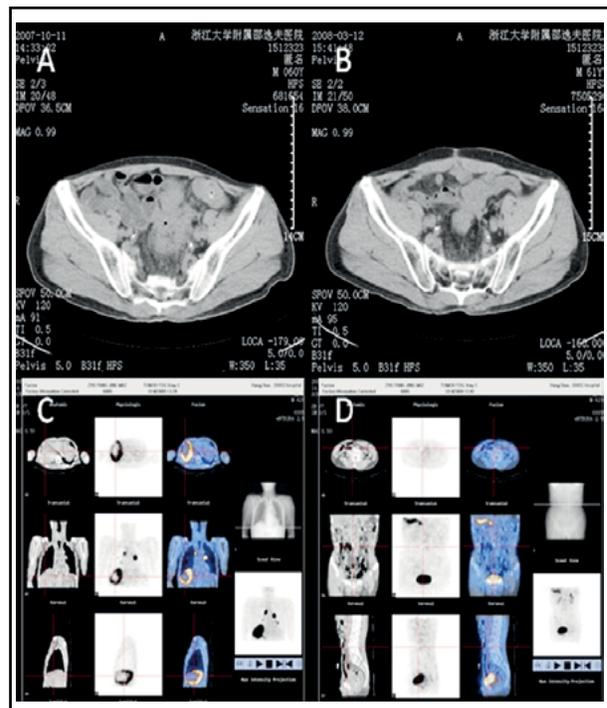


Fig.2: Computed tomography (CT) of pelvis showed a big rectosigmoid mass invading to surrounding area (A). After seven cycles of FOLFOX4 chemotherapy and synchronous pelvic radiotherapy, the mass was shrunken (B). 18F- FDG SPECT/CT revealed multiple pulmonary metastasis, and a big liver metastasis (C) fifteen months after tumor resection, however, there was no perianal recurrent sign (D).

Table-I: Reported cases of implantation metastasis from colorectal cancer to an anal fistula.

Author, year	Age	Sex	Loc	Dukes	Dur	Histology	Operation	Status	LR	DM
Guis,1954	47	M	S	A	2m	mod	APR	14m(A)	NA	NA
Killingback,1965	63	M	S	A	8y	well	APR	NA	NA	NA
Rollinson,1984	65	M	Rs	NA	20y	well	APR	10m(A)	NA	NA
Thomas,1992	68	M	S	B	1y	mod	APR	NA	NA	NA
Isbister,2000	47	M	Rs	C	2y	mod	NA	NA	NA	NA
	39	M	S	NA	1y	well	LoR	NA	NA	NA
Shinohara,2001	36	M	R	C	16y	mod	LAR, LoR	6m(A)	No	Liver
Kouraklis,2002	75	M	S	B	1y	mod	APR	NA	NA	NA
Hyman,2003	66	M	S	B	15y	mod	APR	1y(A)	No	No
Gupta,2005	44	M	L	C	NA	mod	LH+LoR	3y(A)	No	No
Madoka,2005	53	M	S	B	7y	well	AR, LoR	1y(A)	No	No
Satoshi,2006	53	M	R	C	20y	mod	LoR, LAR	10m(D)	No	Peritoneum
Sandiford,2006	72	M	Rs	B	2y	mod+muci	AR+LoR	14m(A)	No	No
Wakatsuki,2008	57	M	Rs	C	7y	mod	AR, LoR	43m(A)	No	Liver
Gravante,2008	64	M	L	C	8m	mod	LR, APR	12m(A)	No	No
Our case	61	M	Rs	B	6m	mod	LoR, AR	54m(D)	No	Liver, lung

Loc, location; Dur, duration of anal fistula; Diff, differentiation; M, male; S, sigmoid colon; Rs, rectosigmoid region; R, rectal; L, left colon; m, month; y, year; mod, moderate- differentiated adenocarcinoma; well, well-differentiated adenocarcinoma; mucin, mucinous adenocarcinoma; APR, abdominoperineal resection; LoR, local resection; LAR, low anterior resection; AR, anterior resection; LH, left hemicolectomy; 'LAR, LoR', LAR followed by LoR; 'LH+LoR', homeochronous LH and LoR; A, alive; D, dead; NA, not available, LR, local recurrence; DM, distant metastasis.

1954. Only fifteen cases have been reported so far. All these cases occurred in patients with a history of anal fistula for two months - 16 years duration and diagnosis was confirmed by confirming similar histopathological characteristics between the primary cancer and the perianal lesion. Considering the possibility of a primary adenocarcinoma developing in an anal gland, Satoshi⁷ defined the presence of a mucinous component on pathology as an exclusion criterion for diagnosing an implantation metastasis. However, Sandiford⁸ later reported a case of implantation metastasis from a moderate-differentiated adenocarcinoma with extensive mucinous component. Immunohistochemical staining of CK7 and CK20 is helpful to differentiate cells of colorectal origin from anal origin. Ranalingam⁹ reported that 87% of rectal adenocarcinomas had a CK7-/CK20+ immunophenotype, which is seen in colon adenocarcinomas. However anal gland and anal transitional mucosa has is CK7+/CK20-. The similar immunophenotypes of the malignancies in the case we report confirms the metastatic nature of the malignancy in the fistula.

There are no recommendations for the management of colorectal cancer implantation metastases to anal fistulae. Although abdominoperineal resection was performed in 7/15 of the reported cases, more recent reports have reported successful manage-

ment with sphincter sparing surgery and combined with local resection with or without radiotherapy to manage the implantation metastasis, without local recurrence. In our case, there was no evidence of perianal recurrence after four years following management with local excision and chemoradiotherapy. The evidence suggests local excision with or without radiotherapy appears to provide good local control for perianal metastatic colorectal adenocarcinoma if negative margins can be achieved. However, others have advocated a more aggressive approach. Gravante¹⁰ has suggested that even when a benign anal lesion was excised in the presence of a colorectal malignancy, abdominoperineal resection should be strongly considered.

Metastatic cancer has different biological characteristics and different surgical stratagem compared to its primary cancer in a certain organ. For example, the surgical margin of zero to four mm is acceptable for a hepatic lesion metastasized from colorectal cancer.¹¹ However for primary hepatocellular carcinoma, surgical margin was advised more than 10mm.¹² Local resection of liver metastatic lesions with generous margin has been achieved encouraging outcome, which highlight the feasibility of local resection, instead of APR, for implantation metastasis of anal fistula from colorectal adenocarcinoma, to avoid a colostomy.

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