Implantation Of Sigmoid Adenocarcinoma Into Intersphincteric Anal Fistula Detected Three Months After Anterior Resection

Hajir Nabi* Daniel Kozman†

*Bankstown-Lidcombe Hospital, New South Wales, Australia, hajirnabi@yahoo.com.au
†Bankstown-Lidcombe Hospital, New South Wales, Australia
Implantation Of Sigmoid Adenocarcinoma Into Intersphincteric Anal Fistula Detected Three Months After Anterior Resection

Hajir Nabi and Daniel Kozman

Abstract

Adenocarcinomas associated with perianal fistulas are very uncommon. Two different pathological pathways have been described to underlie their development. The more commonly described pathway relates to a chronic inflammatory process and subsequent dysplasia in the absence of any associated colorectal malignancy. The second –and much more uncommon- mechanism results from the implantation of viable exfoliated tumour cells from a synchronous remote colorectal cancer.

We report the case of a 68 year old man who had an adenocarcinoma identified in a perianal fistula three months after a high anterior resection performed for a pT3 N0 sigmoid adenocarcinoma. Pathological assessment of this deposit revealed that this had identical tumour characteristics to his original sigmoid adenocarcinoma confirming that exfoliated viable malignant cells had likely become implanted in a pre-existing clinically dormant perianal fistula.

KEYWORDS: Implantation, Anal Fistula, Colorectal cancer
Implantation of sigmoid adenocarcinoma into intersphincteric anal fistula detected three months after anterior resection

Hajir Nabi MBBS, FRACS
Daniel Kozman MBBS, FRACS
Bankstown-Lidcombe Hospital, New South Wales, Australia.

Abstract

Adenocarcinomas associated with perianal fistulas are very uncommon. Two different pathological pathways have been described to underlie their development. The more commonly described pathway relates to a chronic inflammatory process and subsequent dysplasia in the absence of any associated colorectal malignancy. The second —and much more uncommon- mechanism results from the implantation of viable exfoliated tumour cells from a synchronous remote colorectal cancer.

We report the case of a 68 year old man who had an adenocarcinoma identified in a perianal fistula three months after a high anterior resection performed for a pT3 N0 sigmoid adenocarcinoma. Pathological assessment of this deposit revealed that this had identical tumour characteristics to his original sigmoid adenocarcinoma confirming that exfoliated viable malignant cells had likely become implanted in a pre-existing clinically dormant perianal fistula.

Introduction

Adenocarcinomas arising in anal fistulas are rare. There are two mechanisms which have been identified which can lead to this unusual pathology. Firstly, Adenocarcinomas have been detected in the absence of any other colonic malignancy1-3. It has been postulated that the chronic inflammatory process which is associated with this otherwise benign pathology can give rise to dysplasia, and subsequent malignant transformation. Approximately 150 case reports of this uncommon pathology exist in the literature.

Alternatively -and less commonly- exfoliated viable adenocarcinoma cells from a separate colorectal malignancy can implant into a concurrent anal fistula. Implantation of exfoliated colorectal adenocarcinoma into an anal fistula was first recognised and reported by Guiss4 in 1954. Since then less than twenty cases of this rare phenomenon have been documented in English literature5-18.

Through the same mechanism viable exfoliated colorectal adenocarcinoma cells have been implanted in staple lines, biopsy sites and haemorrhoidectomy wounds19-21.

Case Report
A 68 year old man with few other medical co-morbidities presented to his general practitioner with a one month history of new onset constipation. A colonoscopy was arranged to evaluate this change in bowel habits. This revealed a moderately differentiated adenocarcinoma in the sigmoid colon 20cm from the anal verge. The tumour was hemi-circumferential, non-obstructing and extended over a distance of 5 cm. Staging computer tomography (CT) scans of the patient’s chest, abdomen and pelvis revealed no evidence of disseminated disease (cM0) and no evidence of tumour perforation or local invasion.

The patient subsequently underwent a laparoscopic high anterior resection, and made an unremarkable recovery. The procedure was uneventful with tumour handling kept to a minimum and no specimen perforation at the time of the resection. A povidone-iodine antiseptic washout of the rectal stump was performed prior to the creation of a double-stapled end-to-end anastomosis using a circular stapler to prevent the implantation of exfoliated tumour cells into the staple line.

Histopathology confirmed the presence of a moderately differentiated sigmoid adenocarcinoma with mucinous foci and invasion through the muscularis propria for an estimated distance of up to 8mm (pT3c). Extramural venous invasion immediately deep to the tumour was noted. The resection was complete (R0) - with a 5cm distal clearance, 15cm proximal clearance and 27mm non-peritonealised circumferential clearance margin. Twenty-five lymph nodes were identified in the specimen and none were involved (pN0). No extramural tumour deposits were identified.

The patient’s pathology and imaging were presented at a multidisciplinary tumour board meeting and a subsequent referral was made for medical oncology outpatient review. The patient elected for no adjuvant therapy after having a discussion with the medical oncologists.

On clinical review at three months post-resection the patient complained of perianal discomfort. On examination a perianal lump was identified and an examination under anaesthesia (EUA) was arranged.

At the time of the EUA a 5mm nodule was identified on the perianal skin 2 cm from the anal verge. On probing, this lump was found to be a nodule at the external opening of an intersphincteric fistula in ano. The nodule was excised and sent for pathological review (although at the time the nodule was thought to represent an area of inflammation associated with the fistula). The fistula was subsequently laid open as minimal internal sphincter was involved. No other lesions were identified in the anal canal and there was no evidence of local recurrence at the anastomosis.

Pathological examination of the perianal nodule confirmed the presence of a moderately differentiated adenocarcinoma- with identical pathological characteristics to the recently excised sigmoid carcinoma (Figure 1).

The patient went on to have repeated imaging to exclude the presence of disseminated disease which included repeated CT chest, abdomen and pelvis as well as a proton emission tomography (PET) scan. The PET scan demonstrated intensely increased uptake in the region of the laid open fistula track, which was reported to be in keeping
with metastatic disease in the anal canal (Figure 2). No other recurrent or metastatic
disease was demonstrated on the CT or PET scans.

The patient went on to have an abdominoperineal resection (Figure 3) with final
pathology confirming the presence of residual adenocarcinoma in the previously laid
open fistula tract. The circumferential resection margins were complete (R0
resection). No lymph node involvement was identified.

Discussion

There are two types of adenocarcinomas which can develop in anal fistulas.
Adenocarcinomas can arise de-novo in chronic anal fistulas or more rarely exfoliated
viable colorectal tumour cells can implant into pre-existing fistula tracts.

The first report of carcinoma originating in a chronic anal fistula was described by
Rosser in 1931. The following diagnostic criteria for primary cancers from anal
fistulas were established: 1. suffering from anal fistula for more than ten years, 2.
indurations and severe pain at an anal fistula, 3. mucus secretion, 4. stoma aperture in
the anal canal and anus crypt and 5. no tumour at the oral side of an anal fistula.

The implantation of viable exfoliated colorectal cancer cells on staple lines, biopsy
sites, other benign anal lesions and wounds is well established. Likewise, viable
malignant cells have been isolated from the bowel lumen, surgical gloves and
instruments after colorectal surgery.

Ours review of English literature identified fifteen other reports of exfoliated
colorectal tumour cells leading to malignancy in pre-existing anal fistulas. Like our
case, authors of previous reports confirmed identical pathology between the deposits
in the fistulas and the synchronous or metachronous colorectal cancers. All cases
reported have been males. From the fifteen other case reports the site of the primary
colorectal malignancy was recorded in the rectum in six, sigmoid colon in seven and
left colon in two. Of those recorded: three were Dukes A cancers, seven were Dukes
B and three were Dukes C. Interestingly no other disseminated disease was recorded
in any reported cases. Seven of the cases like ours went on to have abdominoperineal
resections, while eight patients had local resections (one of whom had neoadjuvant
chemoradiotherapy prior to local excision). Only one of the fifteen other case reports
documented that the patient had died from carcinomatosis 10 months after a local
resection (original pathology Dukes C).

Our case highlights the need for a high index of clinical suspicion when faced with
unusual clinical presentations in people with recent colorectal cancer resections. The
issues pertaining to the implantation of exfoliated malignant cells into benign tissue
during colorectal cancer operations remains an area of concern for clinicians.
References

2. Getz SB Jr, Oogh YD, Patterson RB, Kovalcik PJ. Mucinous adenocarcinoma developing in chronic anal fistula: report of two cases and review of literature.

Figure 1. Histopathology slides confirming that the implanted area in the anal fistula was identical to the sigmoid adenocarcinoma excised 3 months earlier.
Figure 2. PET scan demonstrating increased uptake in region of laid open fistula track (indicated by white arrow).

Figure 3. Pre-operative photograph with patient in lithotomy position showing the area of malignancy in the laid open perianal fistula (indicated by white arrow).