



Case Report

A Rare Case of Anal Squamous Cell Carcinoma Recurrence in Parastomal Hernia Sac and Review of the Literature

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Abstract

Background: Anal Squamous Cell Carcinoma (ASCC) represents roughly 90% of anal cancers. Recurrence of ASCC after successful diversion and Chemoradiotherapy (CRT) is rare and has been reported in prior literature in the liver, lungs, regional lymph nodes, bone and kidneys. However recurrence in a parastomal hernia sac after negative surveillance imaging and the possible efficacy of metastasectomy has not been reported.

Objective: To present a rare case of the first known incidence of recurrent anal squamous cell carcinoma of the anus in a parastomal hernia sac after successful CRT and negative surveillance imaging.

Case Summary: A 55-year-old male who presented with abdominal pain, constipation and a large fungating anal mass underwent laparoscopic diverting loop sigmoid colostomy and Nigro protocol for ASCC. Post-treatment MRI (magnetic resonance imaging) and PET/CT (positron emission tomography/computerized tomography) scans were negative for residual disease. The patient underwent exploratory laparotomy, colostomy takedown, large bowel resection with primary anastomosis, small bowel resection with primary anastomosis, parastomal hernia repair with excision of parastomal hernia sac and abdominal wall reconstruction. His postoperative course was uneventful and the patient was discharged on postoperative day 7. Final surgical pathology of the parastomal hernia sac reported recurrence of invasive squamous cell carcinoma. The patient was further evaluated in the outpatient setting and remained disease free, in remission.

Conclusion: This case highlights the importance of further studies to investigate recurrence patterns of ASCC and may also help to recognize a possible role for selective salvage metastasectomy with surveillance in select patients with prior identified disease.

Keywords: Anal squamous cell carcinoma; Anus neoplasms; Colorectal surgery; Parastomal hernia

Introduction

Anal Squamous Cell Carcinoma (ASCC) is the malignant transformation of the transitional or squamous mucosa. It is a rare form of cancer that represents approximately 4% of anorectal cancers [1], but 90% of anal cancers [2]. It is most often associated with Human Papilloma Virus (HPV) infection, but the risk of occurrence is also elevated in patients with Human Immunodeficiency Virus (HIV) infection [1]. After diagnosis, staging is used to determine the appropriate treatment course, with the original standard for stages I-III being the Nigro protocol (fluorouracil (5-FU) and radiation), and more recently, the addition of mitomycin [3]. The 5-year incidence of recurrence after successful treatment with Chemoradiotherapy (CRT) is 20% and the five-year net survival is 81% [4]. Of the recurrent cases, 55% were local, and of those that were distant metastases, the main site was the liver 48%, followed by the lung 10% [4]. Additionally, less frequent distant recurrence sites included non-regional lymph nodes such as the common iliac and para-aortic, bone and kidney [5]. Throughout our review no prior literature was found documenting recurrence of ASSC in a hernia sac/parastomal hernia sac.

Case Description

This case describes a 55-year-old male with past medical history of HIV on antiretroviral therapy, hepatitis C virus (Hep C) and heroin abuse who presented with abdominal pain and constipation for five days. The patient was found to have a large fungating anal mass extending from anal verge circumferentially out, growing well beyond the gluteal folds laterally to posterior gluteal region in the midline with no palpable inguinal lymph nodes (Figures 1,2) Image showing large fungating anal mass on initial consultation.



Figure 1: Fungating anal mass.

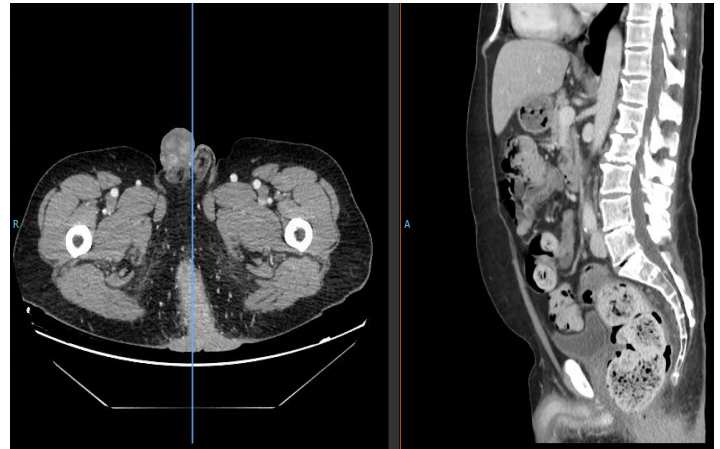


Figure 2: Initial CT abdomen/pelvis with intravenous contrast

Computerized Tomography (CT) scan of the abdomen and pelvis with intravenous contrast showed mucosal thickening of the rectum with either a mucosal lesion/mass and/or inflammatory process associated with overlying skin thickening, exophytic lesion and/or inflammatory process surrounding the rectum and anus, as well as medially in the buttocks (Figure 2). This patient underwent elective laparoscopic diverting loop sigmoid colostomy and anal biopsy. Biopsy pathology provided a diagnosis of moderately differentiated invasive squamous cell carcinoma with pathological staging by Hematology and Oncology as locally advanced anal CA, Stage IIIC (pT4N1c). The patient's PET scan showed a hypermetabolic anorectal mass with inguinal and external iliac lymph nodes (Figure 3). Image showing transverse (Left) and sagittal (Right) views of the pelvis illustrating the overlying skin thickening and exophytic lesion surrounding the rectum, anus as well as medially in the buttocks.

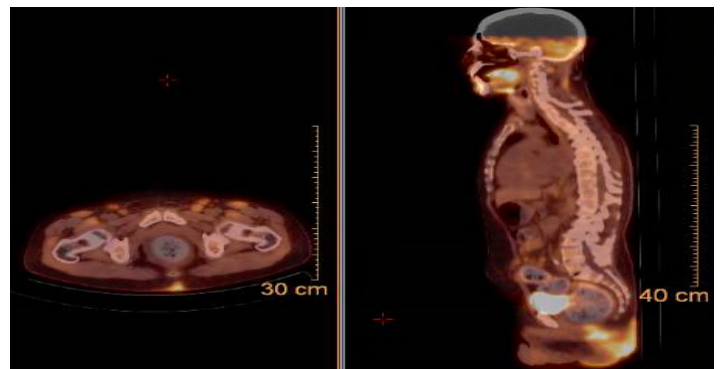


Figure 3: Initial PET/CT Skull base - Mid Thigh.

Image showing hypermetabolic large irregular anorectal mass extending externally to involve the bilateral medial gluteal clefts. Hypermetabolic enlarged bilateral inguinal and external iliac chain

lymph nodes. Three months postoperatively at routine follow up a large non-obstructing parastomal hernia was noted. On completion of chemoradiation with Nigro protocol physical examination did not reveal any recurrent palpable masses and MRI pelvis and PET/CT (Figures 4,5) were negative for residual disease with no pelvic adenopathy. Colostomy reversal was planned and preoperative colonoscopy identified no new or persistent lesions. Patient underwent successful exploratory laparotomy, extensive lysis of adhesions, small bowel resection with primary anastomosis, colostomy takedown with large bowel resection and anastomosis and parastomal hernia repair with excision of parastomal hernia sac and abdominal wall reconstruction and closure. Left image showing diverting colostomy with associated large parastomal hernia. Right image showing no pelvic lymphadenopathy, no pelvic soft tissue masses with small bilateral fat containing inguinal hernias. Postsurgical changes with left-sided colostomy with interval complete resolution of the previously seen hypermetabolic activity seen in the anorectal region and overlying gluteal clefts. Resolved pelvic adenopathy. Complete Response.

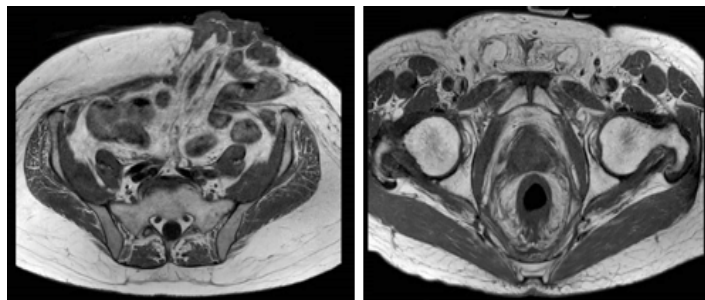


Figure 4: MRI Pelvis with and without contrast post CRT.



Figure 5: PET/CT Skull base - Mid Thigh post CRT.

Surgical specimens were collected and sent and are detailed in Table 1. The patient had an uneventful postoperative course and was discharged home on postoperative day 7 for follow up and review of surgical pathology. Notably, all sites from gross specimens taken intraoperatively were negative for malignancy except Hernia sac specimen 1 (Figures 6,7). Image showing two aspects of the parastomal hernia sac submitted as surgical pathology.



Figure 6: Gross surgical specimens.

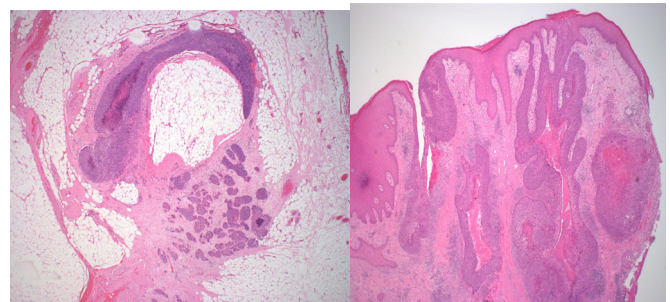


Figure 7: Histopathological slides of parastomal hernia sac.

Left image shows a low power area of interest from the parastomal hernia sac specimen. Right image shows a high power view of the parastomal hernia sac demonstrating invasive squamous cell carcinoma with necrosis, moderately differentiated in fibro-adipose. After discharge the patient returned to The Colorectal Surgery Clinic for follow up at 2 weeks, 3months. At this time the patient reports no symptoms and remains in clinical remission with plans for surveillance for his ASCC via physical examinations, imaging, biopsies and colonoscopy in the future.

Gross Specimen Site	Pathological Findings
Hernia Sac 1	Invasive squamous cell carcinoma with necrosis, moderately differentiated in fibro-adipose tissue
Hernia Sac 2	Negative for malignancy
Small bowel	Negative for malignancy
Small bowel staple line	Negative for malignancy
Colon	Negative for malignancy
Colon staple line	Negative for malignancy
Skin from ostomy site	Negative for malignancy

Table 1: Pathology from surgical specimens.

Discussion

Anal Squamous Cell Carcinoma (ASCC) is rare malignancy, representing 2% of all gastrointestinal cancers, however roughly 90% of anal cancers [2,5,6]. The management of anal cancer has undergone numerous developments in recent decades, with an even stronger emphasis on local control and sphincter preservation. Definitive Chemoradiation Therapy (CRT) using 5-fluorouracil and mitomycin C constitutes the standard treatment for localized disease [7]. Three randomized controlled trials validated CRT as a standard of care for locally advanced disease [8-10]. Recent data reported 17% local recurrences, 11% distant and 4% both after a median follow-up of 4 years s/p CRT [11,12]. As seen in our patient, locally advanced disease, including T3/4 tumor or node-positive disease seems to provide a high risk of recurrence [12]. While multiple studies have documented recurrence at various sites, to our knowledge after review of the literature, this is the first reported case at a parastomal hernia site. In this case, in retrospect, resection of the hernia sac during the colostomy reversal procedure resulted in identification of the only foci of recurrence. This foci was unknown at the time of surgery and not suspected after surveillance pelvic MRI and PET/CT conveyed no local or metastatic disease. While the mechanism of spread to the peritoneal surface of the hernia sac remains unknown, excision and abdominal wall reconstruction appears to have served as a successful curative treatment for this oligometastatic foci of ASCC. Currently, common practice for locally recurrent ASCC involves Abdominoperineal Resection (APR) [7] and systemic therapy for recurrent metastatic disease [13]. Recent data suggest that salvage surgery for recurrence could achieve local pelvic control in about 60% of cases. A recent review of 39 observational studies, including 1388 patients, reported a 5-year

disease free survival rate (DFS) rate of 38.3% and overall survival rate of 45% following salvage surgery for recurrent disease [14]. Overall, 10–20% of patients tend to experience distant relapse after chemoradiation therapy (CRT) [15]. The para-aortic nodes, liver, lungs, and skin are among the most common metastatic sites in anal cancer [12]. Currently, there is limited data, mostly retrospective, regarding the benefit of metastasectomy in stage IV anal cancer [12,13]. A review of the National Cancer Database of more than 2000 patients with metastatic anal cancer from 2004 to 2014 revealed better outcomes for patients with liver metastasis who underwent metastasectomy with a median overall survival of 34 versus 16 months ($P < 0.0001$) [13]. There was however no difference in survival for metastasectomy of other sites. Other retrospective studies have also reported better outcomes for patients with metastatic ASCC if they had received multimodal treatment, including metastasectomy and/or ablation [12].

Conclusion

While the presence of recurrent distant ASCC was unknown preoperatively, our case highlights the importance of continued research into recurrence patterns and the potential benefit of salvage surgery and metastasectomy for selective recurrence sites to improve the chances of complete, sphincter-preserving resection and increase DFS and overall survival rates.

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