

CASE REPORT

Laparoscopic abdominoperineal resection for the treatment of a mucinous adenocarcinoma associated with an anal fistula

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Abstract

Mucinous adenocarcinoma associated with an anal fistula is a rare oncologic entity which may pose diagnostic and therapeutic challenges for Surgeons and Medical Oncologists. Few reported cases without definite therapeutic guidelines exist. It represents 2–3% of all gastrointestinal malignancies and arises from chronic anal fistulas, ischioirectal or perianal abscesses. We report a case of perianal mucinous adenocarcinoma in a 65-year-old male initially surgically treated multiple times for a recurrent fistula in ano of 5 years duration. He presented with an ischioirectal and a perianal fistula. Incisional biopsy from fistulotomy revealed mucinous adenocarcinoma. Contrast enhanced computed tomography scan and magnetic resonance imaging showed a localized perianal growth of a tumor which was further evaluated with colonoscopy. With no evidence of metastasis, we performed a laparoscopic abdominoperineal resection (APR). Two years follow-up after APR and without adjuvant chemotherapy there is not any evidence of recurrence or distant metastasis.

INTRODUCTION

Perianal fistulas are a very common clinical entity. However, the development of mucosal adenocarcinoma in them, is extremely rare and it is more likely to be related to chronic inflammation, with more than 10 years duration. These carcinomas represent ~2–3% of large bowel cancers [1]. There are few case series in the literature, for instance in Japan they represent 6.9% of all anal carcinomas [2]. Clinical suspicion is of paramount importance in diagnosis. Moreover, they may present at an advanced stage.

They are often misdiagnosed as for commonly a benign disease and this may lead to a delay in diagnosis. Therefore, early detection is challenging. An incisional biopsy under anesthesia is often needed for histological confirmation of the tumor. Magnetic resonance imaging (MRI) can facilitate the diagnosis and is essential to determine the range of the surgical resection. Abdominoperineal resection is usually performed as the surgical treatment of choice. Combined chemoradiation therapy may improve the outcome of locally advanced tumor [2–4].

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Figure 1: The specimen of abdominoperineal resection and the wound deficit covered with a flap from the gluteus maximus muscle.

CASE REPORT

We report the case of a 65-year-old patient who was treated after multiple attempts to drain an ischiorectal abscess. Three procedures followed, to surgically repair a recurrent suprasphincteric fistula by mobilizing mucosal flaps along with fistulotomies. At last fistulotomy, the histology revealed the development of a mucinous adenocarcinoma of moderate differentiation.

Staging the disease, revealed a 3 cm tumor with mucin-like discharge on the right side of the anus. His CEA level was high, at 55.71 ng/mL, and T2-weighted MRI revealed a multilocular tumor with high signal intensity.

The patient was subjected to laparoscopic abdominoperineal resection (APR). During the operation rectosigmoid was laparoscopically mobilized and resected with an ENDOGIA stapler and an end colostomy was performed. The patient was then placed in a prone 'jackknife position' and a wide resection of the anus was performed out of the pelvis with the coccyx excised in order to enter the presacral space. Afterwards, the rectosigmoid was dissected from the perianal incision and the APR was completed by an anterior dissection from the Denonvillier fascia. The deficit was covered by mobilization of a myocutaneous flap from the gluteus maximus muscle (Fig. 1).

The final diagnosis was confirmed as mucinous adenocarcinoma, pT2b, NO, pPM0, pDM0, pRM0, pStage II. The patient was discharged 20 days after surgery. The patient had a good post-operative course and has no recurrence nearly 2 years after the operation. The resection was on clear margins and due to the absence of lymph node metastasis, no adjuvant therapy was recommended.

Abdominoperineal resection, is the most frequent accepted treatment of these rare neoplasms. The extralavator APR can achieve a better oncologic outcome, since the fistula in which the cancer is developed has a predominantly extrasphincteric course. Although neoadjuvant radiotherapy is recommended in these carcinomas, it can damage the healing of a wide perineal deficit and prevent the mobilization of a well vascularized flap.

DISCUSSION

Perianal mucinous adenocarcinoma is a rare entity accounting for only 2–3% of all gastrointestinal tumors [1]. Early diagnosis of perianal adenocarcinoma is extremely difficult as the tumor has a very slow growth and it is almost hidden within the perianal region. However, it should be suspected in a case of a persistent fistula in ano. Staging is of paramount importance. A complete colonoscopic examination to assume the diagnosis is

the first approach. Imaging modalities like endoscopic ultrasound, computed tomography scan and MRI aim to determine the disease extension to adjacent tissues

Risk factors that are associated with the etiopathogenesis of the disease are benign inflammatory conditions like chronic anal fistula, perianal abscesses, syphilis, diabetes, tuberculosis and lymphogranuloma venereum with an incidence of 0.7% in patients with perineal Crohn's disease [4]. The association of perianal carcinoma with a long standing anal fistula is a fact. The clinical depiction of this disease may be perianal pain or itching, painful defecation, stool mixed with blood or mucus, bleeding per rectum, obstruction due to the growth obscuring the anal opening or an ulcer or mass in the perianal region with bloody or mucoid discharge. In other cases in the literature, more than 10 years pass to develop such a carcinoma.

Concerning the therapeutic approach although surgery plays the principal role, the exact role of radiotherapy and chemotherapy in perianal adenocarcinoma either in neoadjuvant or adjuvant setting is still under investigation.

In conclusion, due to the rarity of this tumor and the lack of sufficient patients for controlled trials, there is no consensus regarding diagnosis and treatment strategies.

CONFLICT OF INTEREST STATEMENT

None declared.

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