

CASE REPORT

Late Malignancy after 26 Years of Evolution on an Untreated Perianal Fistula

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Abstract

Squamous cell carcinoma and basal cell carcinoma are two types of neoplasms that rarely affect the perianal region, and their etiology is still a matter for debate. We present the case of a 75 year old patient with a 26 year history of perianal fistula, who presents with purulent and fecal perianal discharge and swelling at this level. Physical examination and anoscopy detected low transsphincteric fistula. The biopsy revealed the diagnosis of squamous cell carcinoma, for which a local excision was performed followed by adjuvant radiotherapy. Two years after this event, the patient presented with another perianal lesion, which according to the histopathological result was a basal cell carcinoma; local excision was the only treatment performed for this malignancy.

Keywords: perianal, fistula, fistulotomy, squamous, basal, carcinoma.

Rezumat

Carcinomul scuamos și carcinomul bazocelular reprezintă două tipuri de neoplazii care afectează foarte rar regiunea perianală, iar etiologia lor este încă în dezbateri. Prezentăm cazul unui pacient în vârstă de 75 ani, cu istoric de 26 de ani de fistulă perianală, care se prezintă pentru scurgeri perianale purulente și fecaloide și tumefacție la acest nivel. Examenul fizic și explorarea anoscopică a decelat o fistulă transsfincteriană joasă. Biopsia relevă carcinom scuamos, pentru care s-a efectuat excizie locală urmată de radioterapie adjuvantă. La 2 ani de la acest eveniment pacientul se prezintă cu altă formațiune tumorală perianală, care în urma rezultatului histopatologic se constată a fi carcinom bazocelular; excizia locală a reprezentat tratamentul unic pentru cea de-a doua neoplazie.

Cuvinte cheie: perianal, fistula, fistulotomie, scuamos, bazal, carcinom.

INTRODUCTION

Anal and perianal tumors represent a rare pathology, and the presentation can vary greatly given the complex anatomy of this region. The most common form of cancer at this level is squamous cell carcinoma, the frequency at the perianal level being three to five times lower than at the anal canal level¹. Perianal squamous cell carcinoma, unlike anal canal carcinoma, is staged and treated like any other squamous cell carcinoma of the skin². In spite of being the most common neoplasm

of the skin, basal cell carcinoma with perianal localisation is extremely rare.

CASE PRESENTATION

We present the case of a 75 year-old patient, with long history of perianal fistula for which he did not receive specific treatment, who presents at our clinic for purulent and fecal perianal discharge and a small swelling at 9 o'clock left lateral decubitus. The associated pathologies of this patient are essential hypertension, prostate

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adenoma diagnosed in 2008, and hemorrhoidal disease for which hemorrhoid banding was performed in 2008.

Clinical and anoscopic examination reveals low transspincteric perianal fistula and grade II hemorrhoids. The other paraclinical exams and colonoscopy were normal. The patient tested negative for HPV infection. Fistulotomy was performed and a partially epithelialized fistulous tract with an ulceration area were discovered. A biopsy from the the ulceration was performed and revealed a well differentiated, keratinized exulcerated squamous cell carcinoma. After the diagnosis, a CT examination was performed which did not show the presence of metastasis. Shortly after the biopsy, the excision of the perianal lesion was performed, and the initial diagnosis of SCC on the perianal fistula was confirmed. Following the excision, the patient underwent adjuvant radiotherapy treatment as recommended by the oncologist. During the adjuvant treatment the patient presented with abdominal bloating and diarrheal stools (10 stools/day) with onset approximately 3 weeks after the initiation of radiotherapy. This manifestations were suggestive for severe root enteritis, which is why radiotherapy treatment was stopped. Subsequently the evolution was favorable and without complications.



Figure 1. Squamous cell carcinoma on fistulous tract

The patient was monitored periodically, and two years after the initial operation, a 15/7 mm tumor at 7 o'clock left lateral decubitus is discovered. A complete resection of the tumor is performed, and the histopathological result of the resected specimen reveals

basal cell carcinoma, with nodular shape, with lateral and deep margins of resection free of tumor tissue.

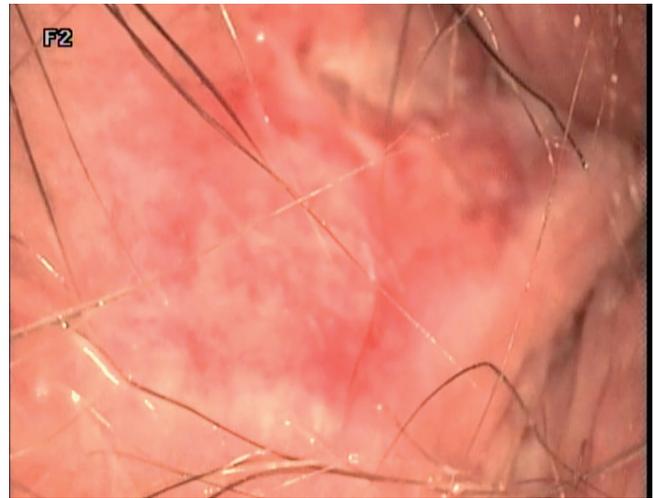


Figure 2. Perianal basal cell carcinoma

Postoperative follow-up was performed by clinical and MRI examinations, which showed no tumoral presence 5 years after surgery.

DISCUSSIONS

A strong link has been discovered between anal squamous cell carcinoma and HPV infection (especially type 16 and 18), this aspect also being shared with cervical cancer ³. Additional risk factors are smoking, anal sex, HIV infection and AIDS⁴. In our case, the patient hasn't recalled any of the above mentioned practices, and has been tested negative for HPV.

The possibility of anal cancer being associated with fissures, fistulas, perianal abscesses and hemorrhoids is still under debate. Some authors suggest that irritation, long term inflammation and repeted epitelial regrowth generated by these conditions are contributing factors to cancer development^{5,6}; on the other hand case control type studies stated that the association is minimal or nonexistant⁷. Chronic fistulas can represent a site of implantation for colo-rectal cancer or endometriosis ^{8,9}. Given the patients history of over 26 years of perianal fistula, without the rest of his underlining conditions bearing any significance regarding his perianal and anal pathology,a cause-efect relationship is hipotesised between his long term perianal and anal condition, and his more recently developed cancer.

Local tumor resection is the elective treatment in 60-70 % of anal verge squamous cell carcinoma cases,

while abdomino-perineal resection is reserved for the advanced cases^{10,11}. Moderate and high differentiated carcinomas with a maximum diameter of <2cm, without lymph node dissemination and without the involvement of the anal sphincter benefit from local excision^{12,13}. Given the reduced size and superficial localisation of the tumor in our case, tumor resection was performed followed by adjuvant radiotherapy treatment.

Basocellular carcinoma is the most common skin tumor, its main cause being UV radiation exposure¹⁴. Genital and perianal areas are affected in less than 1% of the cases, while the most common localisation of this neoplastic pathology (>80%) is on the most exposed areas to sunlight¹⁵. The perianal basal cell carcinoma found in our case appeared two years after the first cancer; taking into account the patient's history of radiotherapy for his first cancer and also the important timeframe between the irradiation and cancer onset it is safe to assume that radiotherapy is a probable cause. Since this tumor type is non-aggressive we considered local excision as the best therapeutic option.

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CONCLUSIONS

Perianal fistula with prolonged evolution can present itself as a risk factor for malignant lesions at this level, which is why treatment of the fistula alone is not enough when the fistulous tracts present modifications. The recommended approach involves multiple biopsies from fistulous tracts plus biopsies targeting suspicious lesions followed by histopathological examination; repeated clinical and imaging evaluations also bear great importance.

Compliance with ethics requirements: The authors declare no conflict of interest regarding this article. The authors declare that all the procedures and experiments of this study respect the ethical standards in the Helsinki Declaration of 1975, as revised in 2008(5), as well as the national law. Informed consent was obtained from all the patients included in the study.