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Synchronous mucinous adenocarcinoma of the recto sigmoid revealed by and seeding an anal fistula. (A case report and review of the literature)

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ABSTRACT

INTRODUCTION: There are few cases of synchronous rectal adenocarcinoma revealed by an anal fistula. The diagnosis of synchronous mucinous adenocarcinoma of the recto sigmoid and anal canal remains difficult. The chronic anal fistula can be mistaken as the common manifestation of a benign perianal abscess or fistula.

CASE PRESENTATION: We present a rare case of a Greek Caucasian 79 year old male patient with anal fistula and a recurrent perianal abscess who subsequently was found to have developed synchronous rectosigmoid and perianal mucinous adenocarcinoma on biopsy. The histological exam revealed mucinous adenocarcinoma in two sites, representing two tumors, cells were immunopositive for cytokeratin 20 and negative in cytokeratin 7. The patient underwent "laparoscopic extralevator abdominoperineal excision " with both lesions being resected. There is no recurrence after four years of follow up.

CONCLUSIONS: This case highlights the importance of high suspicion, further investigation and the need of biopsy in all anal fistulae.

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1. Introduction

Synchronous rectosigmoid mucinous adenocarcinoma seeding an anal fistula is a very rare condition. Diagnosis is difficult especially when primary tumor remains unknown and the chronic lesion of anal fistula seems to be a benign condition. The management of these cases remain controversial. We present a rare case of chronic anal fistula with the presence of a reccurent perianal abscess with synchronous rectosigmoid mucinous adenocarcinoma, treated with laparoscopic extraelevator abdominoperineal resection. A persistent anal fistula with recurrent inflammation is known to lead to primary cancer, and such cancer develops in 0,1% of all anal fistulae [1,2]. This case highlights the importance of further investigation and the need of biopsy in anal fistulae. Our case has been reported in line with the SCARE criteria [3], a 14-item checklist that was formulated to help improve the reporting quality of case reports.

2. Case presentation

A Caucasian Greek 79 years old male presented in our general hospital for investigation of an anal fistula with a recurrent perianal abscess that was treated as a benign condition. (Fig. 1) Clinical examination revealed the presence of a longstanding perianal fistula which appeared to be transsphincteric. The patient underwent pelvis Magnetic Resonance Imaging (Fig. 2) and colonoscopy. Biopsy was taken the histopathology examination revealed a well differentiated mucinous adenocarcinoma in two synchronous tumors with CK20 markers positive and CK7 markers negative.The possibility of primary cancer in the rectosigmoid with metastatic seeding in the perianal fistula could not be excluded.

The patient underwent laparoscopic abdominoperineal resection with complete excision of both lesions (Fig. 3) and the pelvic floor was strengthened by a biological mesh(Fig. 4) The histologi-

Abbreviations: CK7, Cytokeratin 7; CK20, Cytokeratin20.

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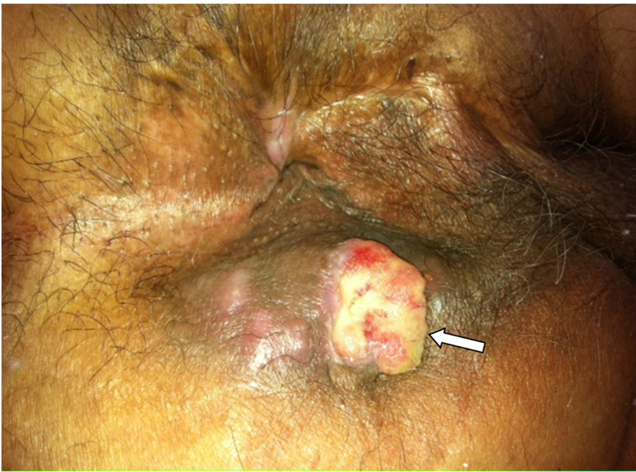

Fig. 1. Peri-anal fistula before surgery.

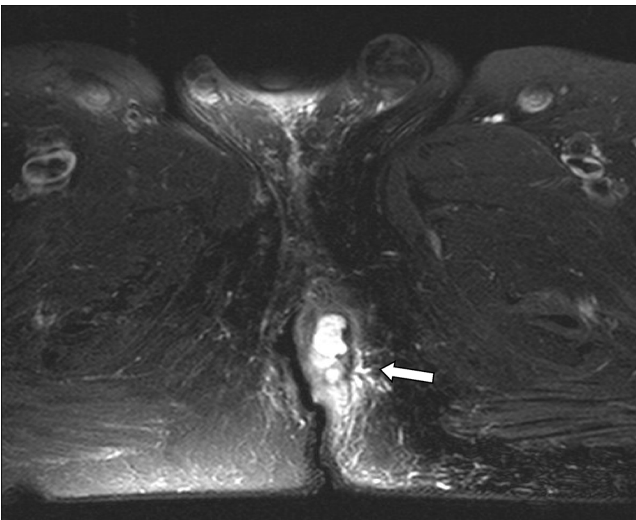

Fig. 2. Fistula presenting in pelvic MRI.

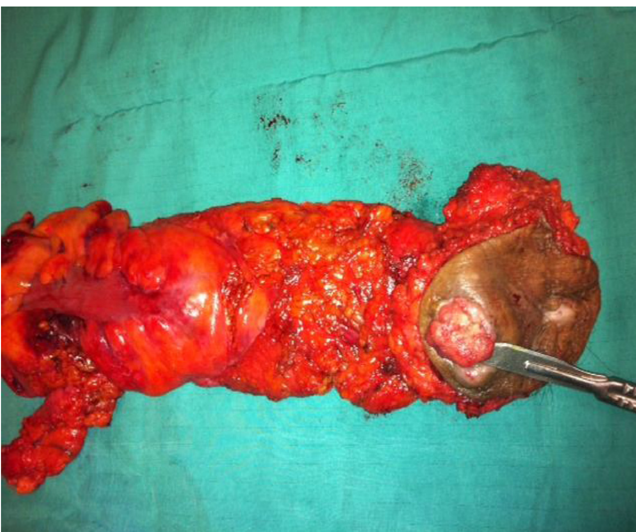

Fig. 3. Surgical specimen.

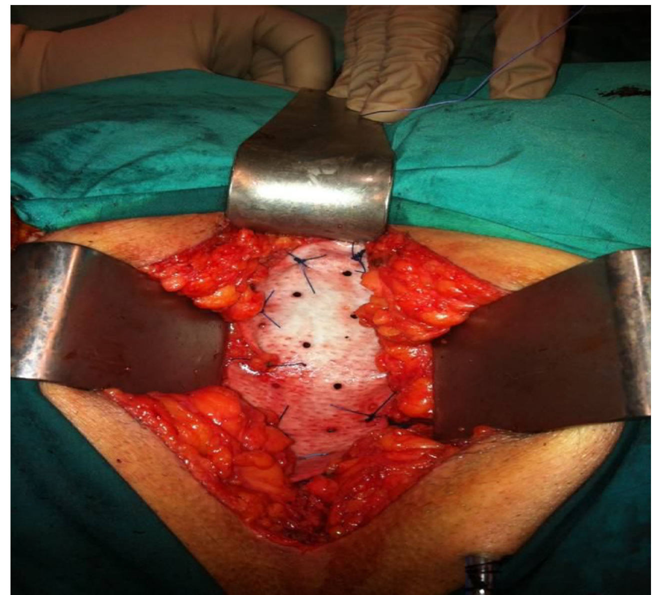

Fig. 4. Pelvis floor construction with biological mesh.

cal examination of the resection margin confirmed the diagnosis of well diffentieted mucinous adenocarcinoma with 34 lymph nodes in the pericolonic fat with maximum diameter of 0,2–0,7 cm, all negative for cancerous infiltration (T3N0M0). The resection margin was classified as R0. The postoperative course of the patient was normal. He was discharged on the 6th postoperative day.

3. Discussion

Synchronous adenocarcinoma of the rectosigmoid, seeding in an anal fistula, presenting as a recurrent perianal abscess is extremely rare. Symptoms in these patients appear due to their perianal disease. That's why the coexisting carcinoma is missed. There is an important observation about sex prevalence, all cases reported were male except only one female. In all cases of fistula and perianal abscess it is very important that histological examination is performed [4,5,6].

For the first time in 1907 Charles Ryall reported implantation metastasis of solid cancers and described this phenomenon as cancer infection [5,7,8].

The first case of cancer implantation to a perianal fistula was reported in 1954 by Guiss and al. Since then, more cases have been reported and metastasis of colorectal cancer into a perianal fistula has become accepted even if the management remains a matter of controversy [7]. In our case, the symptoms of our patient was attributed to his perianal disease and the primary carcinoma was missed. It is important that clinicians who manage patients with anal fistulae and recurrent perianal abscesses to referee them for further colonic investigation (colonoscopy and biopsies) in order to exclude the possibility of primary carcinoma.

Rosser in 1931 first described the following basic criteria which determine if a fistula has undergone malignant transformation. These include persistence of anal fistula for more than ten years, presence of mucous secretion, no evidence of tumor in the rectal or anal canal mucosa and internal opening of the fistula negative of malignancy [9,10].

Sumikoski, Skir and McIntyre et al. and Rundle [10–12] established the diagnostic criteria for primary cancer in anal fistula:

1) recurring inflammation of the fistula for at least ten years
2) Secretion of mucous

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