

# Mucinous Adenocarcinoma Arising from Chronic Perianal Fistula

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## Summary

Perianal mucinous adenocarcinoma is a rare tumor which may be associated with long-standing chronic perianal sepsis. Early diagnosis is challenging and is based on a high index of clinical suspicion and specific histological features. Definitive treatment is surgical, in the form of an abdomino-perineal resection. We hereby describe a case of a perianal mucinous adenocarcinoma arising from long-standing recurrent perianal fistula and complement this with a brief review of the literature pertaining in particular to the management of this condition.

**Key Words:** Mucinous adenocarcinoma, Perianal fistula

## Introduction

Operative exploration of all patients with perianal sepsis is recommended principally for the control of sepsis and to rule out inflammatory and malignant cause or association. Perianal mucinous adenocarcinoma is very rarely associated with chronic perianal sepsis. Due to its rarity, the condition is often overlooked.

## Case report

A 61 year-old man presented with a chronic discharging sinus affecting the right gluteal region. This was his third presentation within the last two years. In the previous two episodes, incision and drainage of the abscesses were performed. On both occasions, the histology specimens were reported to be consistent with abscesses.

Examination revealed frank pus discharging from the two previous operation scars. *Per rectal examination* was essentially normal. A diagnosis of perianal sepsis was made and an examination under anaesthesia with drainage of abscess was planned. Intraoperatively a large abscess cavity inter-communicating the right and

left ischiorectal fossa was noted. The cavity was filled with pus admixed with some gelatinous material. The cavity was drained and specimens were sent for histological assessment.

Histology of the specimen confirmed the diagnosis of mucinous adenocarcinoma. The tumor cells had marked pleomorphic nuclei with some prominent nucleoli within pools of mucin (Figure 1). Colonoscopy and a computer tomography (CT) scan of the abdomen and pelvis were performed. Colonoscopy was essentially normal whilst CT scan demonstrated a large extensive tract and cavity involving the right and left ischiorectal fossa, the gluteal region and the obturator internus muscles (Figure 2). Surgery was deemed unsuitable due to the locally advanced state of the tumor. Palliative treatment in the form of local radiotherapy was discussed and hence the patient was referred to the oncologist.

## Discussion

Perianal mucinous adenocarcinoma arising from long-standing chronic perianal sepsis is rare<sup>1</sup>. Most reported

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cases of perianal mucinous adenocarcinoma arise in relation to chronic inflammatory bowel disease, specifically Crohn's disease<sup>2</sup>.

The origin of perianal mucinous adenocarcinoma is still debatable. Some has been shown histologically to arise from an anal gland or duct. More recently, the tumor has been accepted as a distinct clinical entity. It has been recognized that the tumor may arise *de novo*, present as a mass, as a newly developed fistula-in-ano, as a sequelum of a long-standing chronic fistula-in-ano or in association with perianal sepsis, with or without intraluminal involvement<sup>3</sup>.

In a five year-review by Nelson RL *et al*, eight patients were diagnosed with perianal mucinous adenocarcinoma arising from chronic perianal sepsis. Of the eight, only two underwent abdomino-perineal resection and post-operative radiotherapy whilst a single patient underwent a pelvic exenteration. However, there was no mention of the extent of the tumor. Due to the locally advanced state of the tumor at the point of presentation, the rest of the patients (five in total) were managed only with colostomies and palliative radiotherapy. The outcome, as expected, was better in the surgical group of patients<sup>1</sup>.

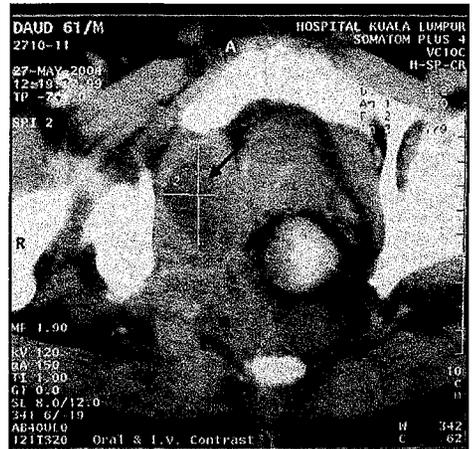
Surgery in the form of a pelvic exenteration was not an option in our patient due to the locally advanced nature of the tumor. The aim of treatment was therefore palliative in the form of radiotherapy.

The difficulty in early diagnosis is principally related to the interpretation of histological specimens from drainage procedures. The presence of mucin lakes and globules in specimens drained from a perianal sepsis has been suggested to provide important histological clues that could point to the diagnosis<sup>3</sup>. More importantly a high index of suspicion is required and any evidence of increasing induration, anal stenosis, an intraluminal mass or a mucinous discharge should alert the attending physician to the diagnosis.

This tumor although slow-growing will ultimately metastasize, most commonly to the inguinal lymph nodes. To date there has been no therapy more effective than total extirpation of the disease with an abdomino-perineal resection or even a pelvic exenteration. Thus to miss this diagnosis at the initial opportunity to make it, that is, when we are draining the abscess, really is to miss that golden opportunity of rendering the patient tumor free.



**Fig. 1: Histological analysis of the specimen from the drainage procedure demonstrating malignant adenocarcinoma cells in a mucinous pool.**



**Fig. 2: Computer tomography scan showing a well-defined collection separate from the rectum.**

## CASE REPORT

### References

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