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Mucinous Adenocarcinoma Arising From Chronic Fistula-In-Ano: A Case Report

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ORIGINAL

Abstract
Mucinous adenocarcinoma of the fistula tract is a rare consequence of fistula-in-ano accounted for approximately 2-3% of perianal cancers. Due to its rarity, no specific guidelines are available on diagnostic modality and treatment of choice. Early diagnosis carries a good prognosis, but most of the time, patients are presented with advanced stages of disease. Chronic and recurrent perianal fistula and abscess often mask the malignant transformation, especially when the index of suspicion is low. Diagnosis is ascertained by tissue biopsy and supported by imaging modality such as MRI. Oncological resection with neoadjuvant or adjuvant radiotherapy and systemic therapy is proven to have a benefit in a patient's overall survival. This article reports a case of mucinous adenocarcinoma of the fistula tract in a 61-year-old gentleman who had a previous history of recurrent unresolved perianal sepsis and fistula.

Keywords: perianal fistula, anorectal adenocarcinoma, fistula-in-ano

Introduction

Complex fistula-in-ano (FIA) is a common surgical condition in anorectal surgery; the cases of malignant transformation are rare. Complex FIA are commonly due to Crohn's disease, radiation treatment, and recurrent infection (1). The pathophysiology of carcinoma transformation from chronic FIA is possible due to chronic inflammation with recurrent epithelial regeneration (2). Early diagnosis is challenging as the symptoms often mimic recurrent benign conditions with tissue biopsy usually fails to obtain infiltrating malignant tissue. Late diagnosis of the condition led to a delay in treatment and a poor prognosis in advanced disease, which proved a good prognosis in aggressive surgical and neoadjuvant or adjuvant chemoradiation therapy. This case highlights a gentleman with recurrent FIA diagnosed with malignant transformation of complex FIA.

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Case presentation

A 61-year-old man presented with recurrent perianal discharge and buttock swelling for 5 years. He had multiple visits to various centres and was treated as a complex FIA. He underwent multiple surgeries over a period of 5 years. However, the FIA kept recurring and complicated with abscess (Figure 1). A computed tomography (CT) of the pelvis and CT-guided percutaneous drainage for recurrent perianal abscess were performed. Subsequent magnetic resonance imaging (MRI) revealed circumferential low rectal wall thickening with a fistulous tract and multiloculated collection with associated pelvic and inguinal lymphadenopathy as well as an extensive bony lesion. Colonoscopy showed proctitis, and the histological assessment revealed inflammatory

lesions. Repeated biopsy from a wedged biopsy reported as adenocarcinoma with mucinous differentiation. He was planned for neoadjuvant chemotherapy followed by abdominoperineal resection (APR). However, the patient's condition was further deteriorating, and he passed away before initiating the treatment.

Figure 1. Physical examination showed bilateral gluteal swelling with multiple fistula tract with growth.

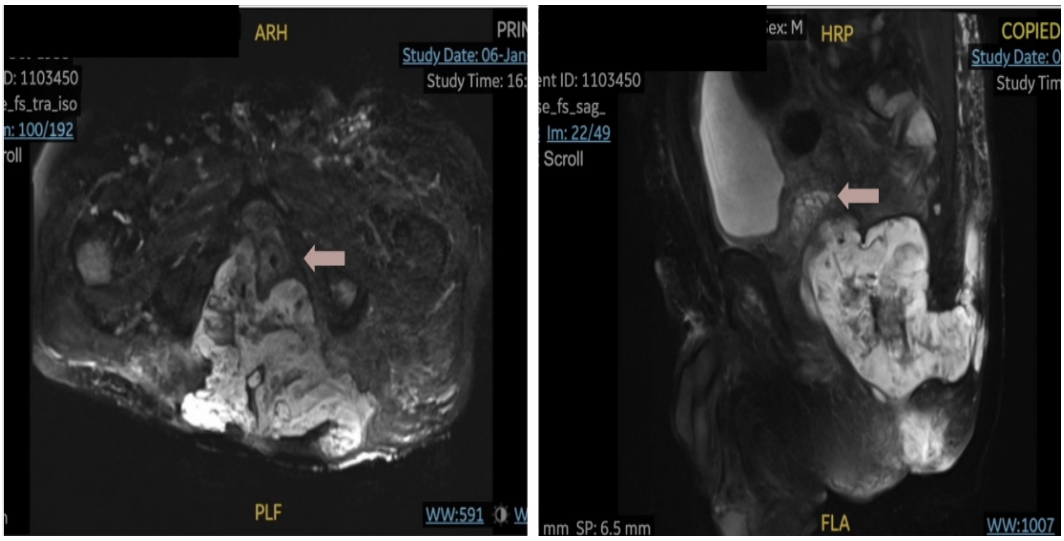

Figure 2. Axial and sagittal view magnetic resonance imaging of pelvis showed circumferential low rectal wall thickening with multiloculated collection and pelvis lymphadenopathy.

Discussion

In coloproctology, anal fistula is a common benign disease; however, perianal adenocarcinoma associated with chronic FIA is an extremely rare occurrence, making up to 6.9% of all cancers in the anal canal (3). Meanwhile, mucinous adenocarcinoma accounted for approximately 2-3% of perianal cancers. Inflammatory bowel disease and Lynch syndrome are the most frequent causes of mucinous adenocarcinoma, and patients with radiation-induced colorectal cancers have a higher incidence of this condition. There have been a few cases where chronic FIA has been linked to mucinous adenocarcinoma. The pathophysiology of malignant transformation remains

unclear, but persistent inflammation accompanied by frequent epithelial regeneration has been implicated (2).

Clinical presentation often revealed symptoms of perianal pain, swelling, and discharge, and examination showed multiple fistula tracts with abscesses. A complete examination must include digital rectal examination and anoscope, which is usually conducted under anaesthesia due to discomfort and pain (1). A case series reported by McIntyre JM in 1948 concluded that one should consider the possibility of carcinoma in the anal fistula when the fistula has been present for many years, the fistula becomes more painful or indurated and the drainage becomes more mucinous in character (4). Definite diagnosis is based on biopsy and histopathology report, although obtaining adequate and right tissue for biopsy is challenging because the mucinous component of the tumor is much larger than the carcinomatous component, which is comparatively small (5). Imaging studies like MRI and CT scans will provide adequate information regarding the invasion of the disease into the adjacent tissue. MRI is the most sensitive modality (>90%) for diagnosing the condition. A notable hyper-intense signal can be seen on T2-weighted images, making pelvic MRI a valuable imaging modality in this case (6). However, a combination of two imaging modalities, such as MRI, endoanal ultrasound, fistulography, and CT scan are proven to provide diagnostic accuracy up to 100% (1).

The standard treatment is oncological resection. The most recommended surgical technique is APR; in more complex cases, securing a negative resection margin requires extensive excision of surrounding tissues, including the overlying skin. Neoadjuvant or adjuvant therapy like chemotherapy, radiotherapy, or chemoradiotherapy can be considered; however, the efficacy is still debatable. A study conducted by Gaertner et al, 8 out of 14 patients with anal fistula malignancy showed no evidence of disease after neoadjuvant or adjuvant therapy with APR (7). In other reported cases by Inoue et al in Japan, 3 out of 9 patients who underwent combination treatment have shown a good response. A total of 2 of the patients had local recurrence during follow up at 56 months despite receiving CCRT and APR with a good margin (8). Diaz-Vico et al reported 3 cases of mucinous adenocarcinoma arising from chronic perianal fistula from their centre; all 3 cases received neoadjuvant CRT and primary surgery, 2 of them received adjuvant oral chemotherapy, which showed good response and no recurrence; however, another 1 did not receive postoperative chemotherapy due to a medical condition and developed nodal metastasis at 26-month follow-up (9).

Distant metastases are uncommon in mucinous adenocarcinoma, and tumor spread is typically lymphatic, with inguinal nodes being the most common location of metastases as seen in our case (10). Higher suspicion of malignancy should be aroused in view of a long-standing chronic fistula and recurrent abscess. Prognosis is worse in mucinous adenocarcinoma when the tumor is larger than 5cm (>T3), or nodal or hematogenous metastases are present at times of diagnosis (7; 8).

Conclusion

A malignant transformation of complex FIA, although rare, should be considered in patients with long-standing, recurrent perianal fistulas, especially when clinical features such as persistent pain, induration, and mucinous discharge are present. Accurate diagnosis requires a combination of imaging and repeated histopathological assessment, as initial biopsies may fail to detect malignancy due to the tumor's mucinous nature. Early detection and aggressive treatment with oncological resection and appropriate chemoradiotherapy offer the best chance for improved outcomes, although the overall prognosis remains guarded in advanced cases.

Conflict of Interest

All authors declare no conflict interest of any kind.

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