

Fistulizing Mucinous Anal Carcinoma: A Case Report

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Abstract

Introduction: Mucinous anal adenocarcinoma is a rare and often misleading entity, because its presentation may mimic suppurative perineal disease [abscess/fistula]. We report a case of fistulizing mucinous anal carcinoma diagnosed by endoscopic ultrasound and fine-needle aspiration, and discuss the diagnostic and therapeutic aspects. **Case presentation:** A 73-year-old man with a history of surgery for an anal fistula [2000] presented with purulent anal discharge associated with mild anal incontinence, without rectal bleeding or bowel habit disorders, and with preserved general condition. Proctological examination found fistulous openings and sphincter hypotonia. Pelvic MRI showed a mass in the left ischioanal fossa fistulizing toward the anal canal. Rectal endoscopic ultrasound showed a heterogeneous pararectal mass measuring 33 × 47 mm, with a fluid component and sparing the rectal wall; histological examination of the fine-needle aspiration sample confirmed mucinous adenocarcinoma. The staging work-up found no secondary lesions. A multidisciplinary decision was made to proceed with chemoradiotherapy, followed by reassessment and surgical discussion according to the clinical course. **Conclusion:** In the presence of a chronic, recurrent, or complex anal fistula, particularly when associated with a mass, induration, or functional impairment, malignant degeneration should be considered and documented by tissue sampling. MRI and endoscopic ultrasound with fine-needle aspiration/biopsy guide the diagnosis. Management is often multimodal and resembles that of rectal adenocarcinomas, with discussion of neoadjuvant treatment followed by radical surgery such as abdominoperineal resection.

Keywords: Mucinous adenocarcinoma; Anal canal; Anal fistula; Pelvic MRI; Endoscopic ultrasound; Chemoradiotherapy; Surgery; Abdominoperineal resection.

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INTRODUCTION

Anal canal cancers are predominantly squamous cell carcinomas, whereas adenocarcinomas of the anal canal are rare. Among them, mucinous adenocarcinoma may arise in the setting of a chronic anal fistula and present as persistent perineal suppuration, leading to delayed diagnosis. The literature emphasizes that these fistulizing forms, although uncommon, should be suspected in any atypical fistula [long-standing, recurrent, complex, associated with repeated abscesses, induration, a mass, or sphincter impairment].

CLINICAL OBSERVATION

Patient: 73-year-old man.

Past History: Surgery for an anal fistula in 2000; no arterial hypertension or diabetes; no chronic liver disease; no known inflammatory bowel disease; no reported tuberculosis exposure; no smoking or alcohol use.

History of Present Illness: For three months, purulent anal discharge associated with mild anal incontinence, without bowel habit disturbance or visible bleeding, with preserved general condition and no fever.

Clinical Examination:

Abdominal Examination: Epigastric tenderness without guarding or palpable mass.

Proctological Examination: Two external openings at 8 o'clock and one internal opening at 11 o'clock; digital rectal examination showed sphincter hypotonia.

Lymph-node areas were free.

Biological Work-up: Hb 13.3 g/dL; WBC 3,886/mm³; platelets 236,000/mm³; CA 19-9 and CEA tumor markers were normal.

Imaging [pelvic MRI]: It described extrinsic compression of the anal canal, which was deviated by a

mass of the left ischioanal fossa, tissue-like in appearance with a fluid center [probable superinfection], with several fistulous tracts [including one toward the anal canal], extending over approximately 5 cm.

Short Colonoscopy: Normal-appearing rectal mucosa; aspect of extrinsic compression just above the pectinate line.

Rectal Endoscopic Ultrasound and Fine-needle Aspiration: Pararectal mass in contact with the lower rectum, round, heterogeneous, well circumscribed, with fluid content in places, measuring 33 × 47 mm, sparing the rectal wall; fine-needle aspiration was performed.

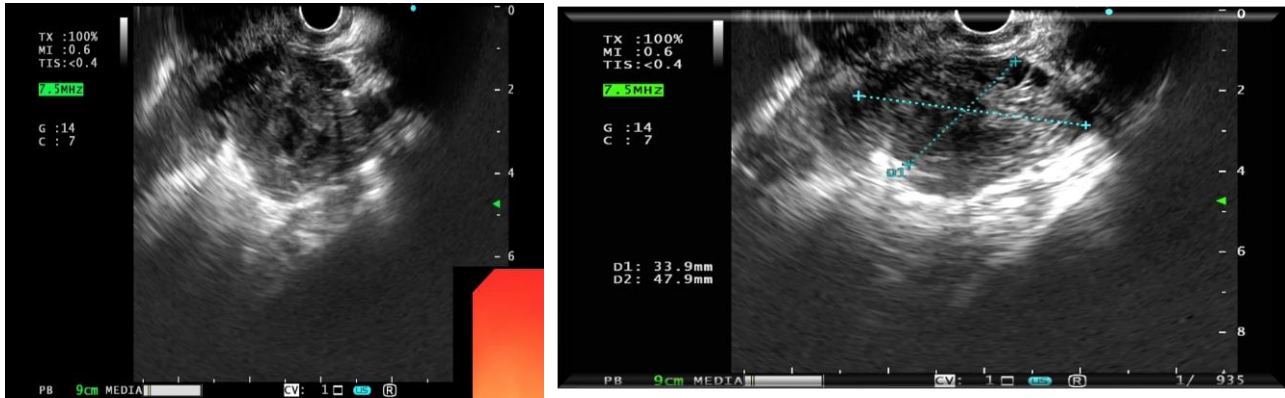


Figure 1: Rectal endoscopic ultrasound: heterogeneous pararectal mass

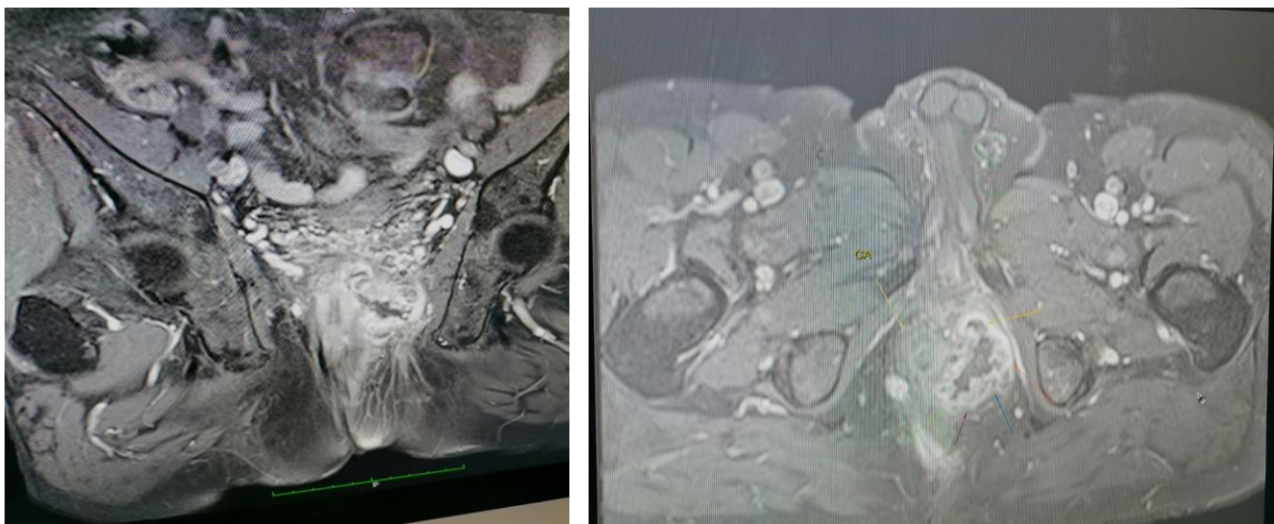


Figure 2: Pelvic MRI: extrinsic compression of the anal canal deviated by a mass of the left ischioanal fossa

Anatomopathological Study of the Mass:

The specimen consisted of several mucoid fragments from a clearly malignant tumor formation. It was characterized by broad pools of mucus within which fragments of mucin-secreting columnar epithelium with moderate cytonuclear atypia were identified. The stroma was scant and limited to a few inflammatory areas. The appearance favored mucinous adenocarcinoma.

Staging Work-up: No secondary lesion was reported.

Management:

Discussion at a multidisciplinary team meeting; indication for neoadjuvant chemoradiotherapy with reassessment and discussion of surgery according to the clinical course.

DISCUSSION

Mucinous adenocarcinoma [MA] is an invasive adenocarcinoma composed of malignant glandular cells containing intracytoplasmic mucin. Usually, infiltrating glandular structures are associated with a mucoid stromal formation [1].

MA can appear in several segments of the digestive tract [colon, stomach, pancreas, gallbladder, etc.], but also in other sites such as the breast, thyroid, or even the skin. However, the perianal location of MA is very rare. It has been suggested that malignant degeneration of a long-standing perineal fistula [PF] may be related to mucosal regeneration, while other authors believe that malignant cells may colonize fistulous

granulation tissue from proximal gastrointestinal neoplasms [2]. Repeated friction, scarring, and inflammatory reactions may be predisposing factors for the development of perianal MA [3,4].

Initial symptoms generally include perianal pain, pruritus, mucinous discharge and/or abscess, associated with an ulceroproliferative lesion or a palpable mass in the perianal region [3]. Because rectal mucosal infiltration is rare, neither bowel obstruction nor rectal bleeding is a frequent symptom [5,7,8]. The most common clinical presentation is related to a chronic anal fistula [1]; tumor progression may lead to tissue destruction, causing perianal abscesses and anal fistulas, with the development of perianal oozing and/or mucinous discharge [6].

Early diagnosis of perianal adenocarcinoma is difficult because the tumor does not cross the rectal mucosa and its course is usually indolent [9].

Digital rectal examination reveals a thickened and indurated area involving the fistulous tract [6], whereas colonoscopy generally shows no visible anorectal lesion. Imaging modalities such as MRI and endoscopic ultrasound [EUS] help establish the diagnosis, assess extension to adjacent tissues, and guide the surgical strategy [10-11].

Thus, pelvic MRI is the best imaging technique, because the abundance of mucin gives these tumors a characteristic radiological appearance, with marked hyperintensity on T2-weighted sequences [12]. However, MRI findings may be difficult to distinguish from other fluid-containing lesions, such as cysts, fluid collections, or necrotic tumors. Involvement of the fistulous tract and anal canal by the mass is suggestive of MA rather than a benign perineal fistula [13,14].

Histological diagnosis remains the gold standard. The presence of extracellular mucin lakes surrounded by well-differentiated, dilated, and tortuous glands, as well as nerves and vessels, confirms the diagnosis. In many cases, the definitive diagnosis is established by anatomopathological examination of the surgical specimen, since preoperative biopsies may fail to reveal invasive carcinoma [15,16].

Surgery is the cornerstone of treatment for this malignant entity. Most surgeons recommend abdominoperineal resection [APR] with wide excision as the treatment of choice because it allows negative margins to be obtained and reduces the risk of local recurrence [17]. In locally advanced tumors, ischioanal APR or extralevator abdominoperineal excision [ELAPE] may be necessary to reduce the rate of positive margins. In such situations, pelvic floor reconstruction is required, involving a longer operating time and increased morbidity.

However, the role of chemotherapy and radiotherapy, in the neoadjuvant or adjuvant setting, remains controversial. Yang *et al.*, Suggested that combined chemoradiotherapy is a valid option when the tumor is not completely resectable or when the patient refuses surgery [18]. Hongo *et al.*, in a study of 11 patients, reported better recurrence-free survival in patients who received preoperative chemoradiotherapy [19]. In a retrospective study including 82 patients, Belkacemy *et al.*, Concluded that T and N stage, histological grade, and treatment modality are independent prognostic factors, with better outcomes in patients treated with chemoradiotherapy; they recommend surgery as salvage treatment [20]. Thus, in locally advanced tumors, preoperative chemoradiotherapy allows tumor downsizing, helps eliminate disseminated tumor cells, increases the chances of R0 resection, and decreases the risk of local recurrence [21].

Moreover, the mucinous component identified on pretreatment MRI has been reported to be associated with a poorer response to chemoradiotherapy than non-mucinous rectal tumors [22].

Distant metastases are rare in MA; dissemination is mainly lymphatic, with inguinal lymph nodes being the most common metastatic site [23]. According to published data, prognosis appears less favorable when the tumor exceeds 5 cm, when carcinoembryonic antigen is elevated, or when lymph-node or hematogenous metastases are present at diagnosis [24,25], with reported survival ranging from 2 to 48 months [26].

Long-term survival rates remain unsatisfactory in locally advanced disease.

CONCLUSION

Fistulizing mucinous anal adenocarcinoma is rare and may mimic benign suppurative disease. Suspicion should be systematic in the presence of a complex or atypical fistula, especially when there is a mass or functional symptoms. MRI and endoscopic ultrasound with tissue sampling are decisive. Management is most often multimodal and resembles rectal cancer strategies, with discussion of neoadjuvant treatment and radical surgery according to stage and response.

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