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Pseudotumoral Rectal Malakoplakia: A Case Report

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Abstract

Malacoplakia is an uncommon acquired granulomatous disease, with a prevalence in females that is quadruple that of males, and a peak incidence in middle-aged individuals. This report describes the case of a young patient presenting with iliac fossa pain, weight loss, and urinary symptoms. Imaging and endoscopic ultrasound revealed an irregular pararectal tissue mass alongside multiple adenopathies. Excisional biopsies of selected adenopathies performed via laparoscopy indicated suppurative adenitis upon histological examination. A histological re-evaluation of the initial series of endoscopic biopsies confirmed the diagnosis of rectal malacoplakia. The patient received antibiotic therapy with ciprofloxacin at a dosage of 1 g per day for six weeks. Subsequent endoscopic assessment revealed two fistulous orifices, one on the anterior surface of the lower rectum and another at the bladder. Histological analysis of bladder biopsies was consistent with malacoplakia.

Keywords: *Malacoplakia, urinary localization, rectal localization, endoscopic appearance, antibiotic therapy.*

Introduction

Malacoplakia is a rare chronic granulomatous inflammatory condition that may affect multiple organs. The gastrointestinal tract is the second most frequent site of involvement after the genitourinary tract, with a prominent predilection for the colorectum, and less commonly, the terminal ileum and appendix. Contributing factors include localized infection, immunosuppression, neoplasia, and systemic disorders. Initially described by Michaelis and Gutmann in 1902 following its observation in the bladder, the term "malacoplakia" was subsequently introduced by Van Hansemann in 1903. The term originates from the Greek words "malakos" (soft) and "plakos" (plaque), signifying its characteristic endoscopic manifestation as yellowish and friable mucosal lesions.

Observation

A 28-year-old patient with no significant medical history presented for evaluation with right iliac fossa pain radiating to the right lower limb and concomitant constipation and dysuria, occurring alongside a weight loss of approximately 7 kg within one month. Physical examination revealed unremarkable findings. Abdominopelvic ultrasound and subsequent pelvic MRI demonstrated a large, irregular pararectal tissue mass measuring 77 x 98 x 107 mm, exerting extrinsic compression on the rectum. A computed tomography scan of the thorax, abdomen, and pelvis illustrated a pararectal mass associated with multiple mesenteric, intrathoracic, cervical, axillary, and left inguinal adenopathies.

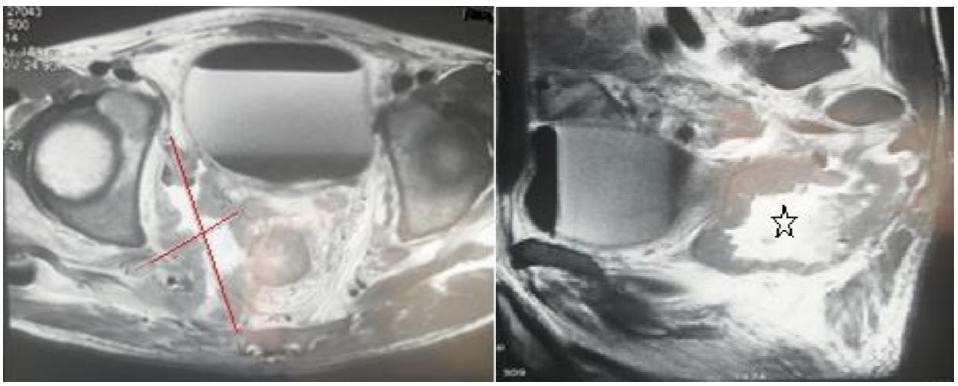

Figure 1: Pelvic MRI showing a large, irregular pararectal mass measuring 77x98x107 mm.

Ileocolonoscopy revealed evidence of extrinsic compression on the mid-rectum extending over 5 cm, with mucosal infiltration in certain areas; endoscopic biopsies suggested histiocytic inflammatory tissue without definitive specificity. Rectal echoendoscopy identified a significant solid-cystic mass of indeterminate origin. Micro-biopsies were performed, with histological and immunohistochemical analyses indicating a hemolymphangioma characterized by CD34+/CD31– expression.

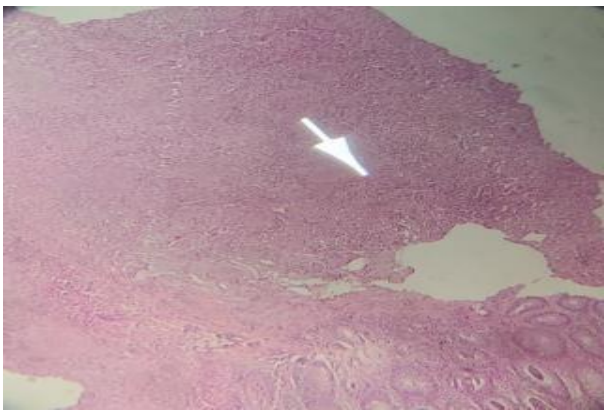

Figure 2: Rectal mucosa demonstrating a dense histiocytic infiltrate.

The case was discussed during a multidisciplinary consultation, leading to a decision to pursue exploratory laparotomy. Intraoperative inspection via laparoscopy revealed a large tumor mass deemed inextirpable; excisional biopsies of select adenopathies were conducted, which upon histological examination demonstrated suppurative adenitis. Given the diagnostic impasse, a histological reassessment of the initial endoscopic biopsies revealed rectal mucosa containing foamy histiocytes with finely granular cytoplasm, including rounded intracytoplasmic basophilic inclusions known as Michaelis-Gutmann bodies. This cellular population coexisted with eosinophilic polymorphonuclear cells and regular lymphocytes, consistent with rectal malacoplakia.

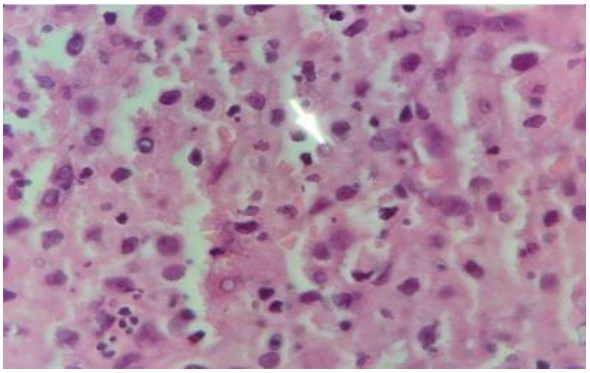

Figure 3: Presence of Michaelis-Gutmann bodies.

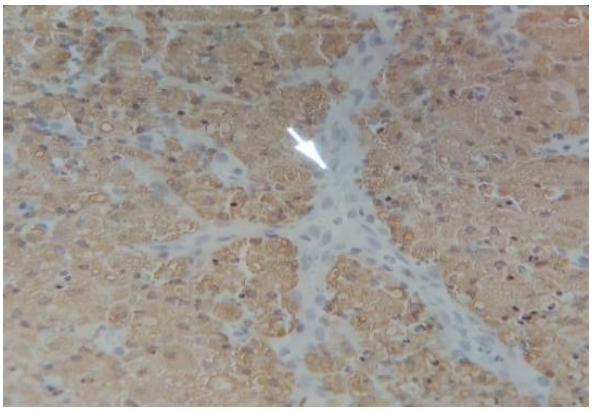

Figure 4: Immunohistochemistry showing diffuse CD68 expression.

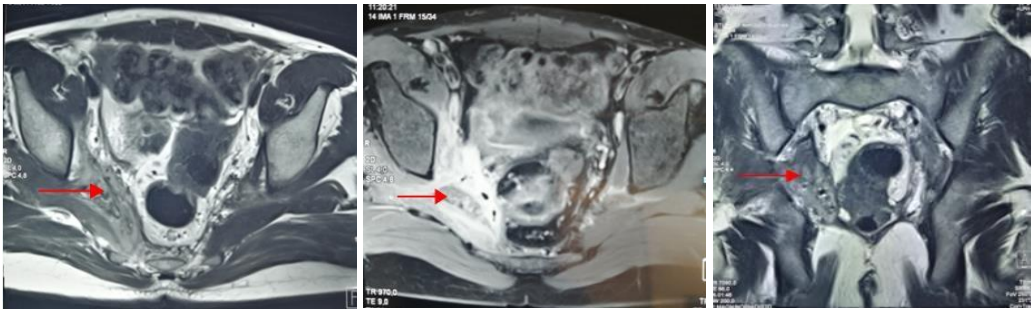

Figure 5: Inflammatory remnant observed at the end of treatment.

Antibiotic therapy with ciprofloxacin at a regimen of 1 g daily for six weeks resulted in favorable clinical outcomes, evidenced by marked improvement in the patient's overall condition, weight gain of 14 kg over two months, alleviation of abdominal pain, pain-free mobilization of the lower limb, and radiological regression of the pararectal mass, albeit with persistent circumferential rectal thickening and inflammatory residue. Follow-up rectoscopy identified a fistulous orifice on the anterior surface of the lower rectum. A ureterocystoscopy further revealed a non-productive right posterolateral fistulous orifice. Histological assessment of bladder biopsies corroborated the diagnosis of malacoplakia. Given the persistence of mild dysuria (with sterile urine on bacteriological examination), a temporary left colostomy was considered, alongside the potential for colonic lowering.

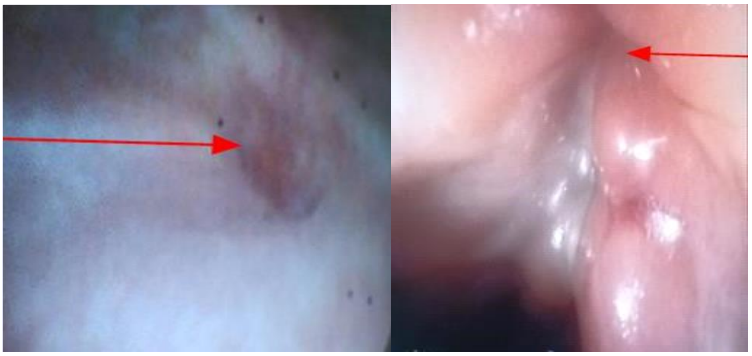

Figure 6: Fistulas involving the lower rectum and bladder.

Discussion

Malacoplakia is a rare, acquired granulomatous condition that is four times more frequent in women than in men, with a peak incidence in middle-aged individuals. However, the disease may also occasionally occur in children [2]. Clinical presentation of rectal malacoplakia is nonspecific; it can be asymptomatic or manifest as diarrhea, abdominal pain, gastrointestinal bleeding, constipation, or even intestinal obstruction [3, 4].

In our patient, clinical symptoms included abdominal pain radiating to the right lower limb, limiting mobility, fecaluria, burning during urination, and weight loss.

Endoscopically, the lesions can be focal or diffuse and vary in size and shape. Three types of lesions are described: erythematous, multinodular, or polypoid mucosa, which may resemble polyps and cancers, or appear as a large mass [3, 5-7]. Malacoplakia usually presents as pale to brown mucosal plaques in the early stages, and later as gray-brown lesions with surrounding congestion and central depression in advanced stages [4].

Endoscopy in our patient revealed extrinsic compression of the lower and middle rectum with a small ulceration. Histologically, malacoplakia is characterized by infiltration of histiocytes (called Von Hansemann cells or Hansemann histiocytes) with abundant basophilic granular inclusions measuring 5 to 15 mm, positive with periodic acid-Schiff (PAS) staining and resistant to diastase. Michaelis-Gutmann bodies (MG bodies), which are incompletely degraded bacterial products mineralized with iron and calcium deposits within phagolysosomes, are positive with Von Kossa staining for calcium or iron staining [3, 8-10].

Given the rarity of the disease, there is no standardized treatment. However, management typically involves antibiotic therapy and surgical resection when medical treatment is ineffective. [9] Antibiotics such as quinolones or trimethoprim-sulfamethoxazole achieve high concentrations in macrophages and have effective bactericidal activity [11-13].

Other therapeutic options include:

- Cholinergic agonists like bethanechol, which may increase the GMPc/AMPc ratio in macrophages, thereby improving lysosomal bactericidal function [13].
- Reduction or discontinuation of immunosuppressive therapy.
- Good control of associated immunosuppressive pathology.
- Endoscopic resection of localized lesions.
- Surgical resection in cases of multiple organ involvement or suspected malignancy.

The prognosis depends on the location and extent of the disease as well as the patient’s condition. In our young patient, the persistence of residual inflammation and the presence of a rectovesical fistula led to consideration of surgical treatment, with the planned creation of a temporary left colostomy,

followed by surgery to repair the fistula and perform colonic lowering [14].

Conclusion

Rectal malacoplakia with lymph node involvement and extension to adjacent organs is often misdiagnosed in clinical practice, mimicking malignancy radiologically. It is crucial for radiologists to consider malacoplakia in the differential diagnosis of large rectal lesions, whether benign or malignant, even if radiological findings are nonspecific. Endoscopic evaluation and histopathological study of a biopsy are recommended for accurate diagnosis, which helps avoid unnecessary surgical resection.

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