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Mystery Behind Hidden Long Standing Perianal Fistula-mucinous Adenocarcinoma

Uzun Süreli Gizli Perianal Fistül-Musinöz Adenokarsinomun Arkasındaki Gizem

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ABSTRACT

Mucinous adenocarcinomas (MA) of the perianal region is a rare entity and have an uncertain etiopathogenesis. They are mostly contributed by, or MA arising from chronic perianal fistula (PF) has an indolent growth with locoregional spread, and increased rate of disease recurrence. Here, we report a rare case of a 58-year-old male presenting with MA of a left perianal exophytic growth for 20 years and hidden PF tract. Though abdomino-perineal resection remains mainstay of treatment, we were able to achieve effective response by doing wide local excision of malignant lesion with fistulous tract and primary reconstruction of the defect by V-Y flap and adjuvant chemotherapy.

Keywords: Perianal fistula, mucinous adenocarcinoma, V-Y flap

Öz

Perianal bölgenin müsinöz adenokarsinomları (MA) nadir görülen bir durumdur ve etiopatogenezi belirsizdir. Çoğunlukla kronik perianal fistülden (PF) kaynaklanan MA, yavaş büyüyen, bölgesel yayılım gösteren ve hastalık tekrarlama oranının yüksek olduğu bir hastalıktır. Burada, 20 yıldır sol perianal ekzofitik büyüme ve gizli PF traktına sahip MA ile başvuran 58 yaşında nadir bir erkek olguyu bildiriyoruz. Abdomino-perineal rezeksiyon tedavinin temelini oluşturmaya devam etse de, malign lezyonun fistül traktıyla birlikte geniş lokal eksizyonu ve defektin V-Y flap ve adjuvan kemoterapi ile primer rekonstrüksiyonu ile etkili yanıt elde edebildik.

Anahtar Sözcükler: Perianal fistül, musinöz adenokarsinom, V-Y flap

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INTRODUCTION

Perianal fistula (PF) is a common disease in proctological diseases. However, rare cases have reported the development of a mucinous adenocarcinoma (MA) from chronic anal fistula. Rosser et al. (1) in 1934 reported seven cases of fistula that had undergone malignant transformation. The pathogenesis of this malignancy remains a mystery. It exhibits slow growth, and although inguinal lymph nodal involvement is seen observed in advanced stages, distant metastases are uncommon (2-4). The aim of this study is to share our experience on diagnostic and therapeutic challenges, as there is a lack of data and uncertainty of disease presentation.

CASE REPORT

A 58-year-old male presented to the surgical out patient department with complaints of a mass over the left perianal region for 20 years, which has increased in size past over the 6 months (Figure 1). He did not reveal any history of pain, mucoid discharge, difficulty in passing stools, rectal bleeding, or weight loss. Inspection of the perianal region revealed a single ulcer proliferative lesion measuring 10x8 cm with no fixity to the underlying structure. The lesion did not bleed on touch, and no discharge was noted. Per rectal examination, there was no induration or fistulous opening noted. No inguinal lymphadenopathy. The patient underwent a colonoscopy, which had normal findings, and the histopathological report of the incisional biopsy suggested dysplastic glands in granulation tissue. A contrast-enhanced computed tomography of the Abdomen and Pelvis was done and reports are suggestive of a well-defined, heterogeneously enhancing exophytic lobulated lesion measuring 6.7x9.9x8.8 cm (Anterior-posterior x Transverse x Craniocaudal) with few internal calcific foci and hypodense areas noted. This lesion arises from the surface of the left gluteal region, with associated minimal skin thickening, obliterating the gluteal cleft, and extending to the anal verge and anorectal junction along the left lateral wall, with suspicious involvement of the anal canal and anorectal junction. The lesion is seen extending along the left ischioanal fossa measuring



Figure 1. Clinical picture.

6.2x2.2 cm, suspicious of malignancy; hence, the patient underwent wide local excision of the swelling, and a fistula tract was noted (site was marked) which was ligated (Figures 2, 3). Histopathological examination was suggestive of Mucin secretory adenocarcinoma in a fistulous tract; all margins were uninvolved (Figures 4, 5). The patient was scheduled for abdomino-perineal resection (APR). However, the patient was not willing to undergo the same procedure, and hence was planned for closure of the defect by V-Y flap (Figure 6, 7) with adjuvant chemotherapy (capecitabine and oxaliplatin). The post-operative period was uneventful. The follow-up was done for one year and was uneventful. Advised follow-up magnetic resonance imaging (MRI) of the abdomen and pelvis, but the patient refused due to financial constraints.

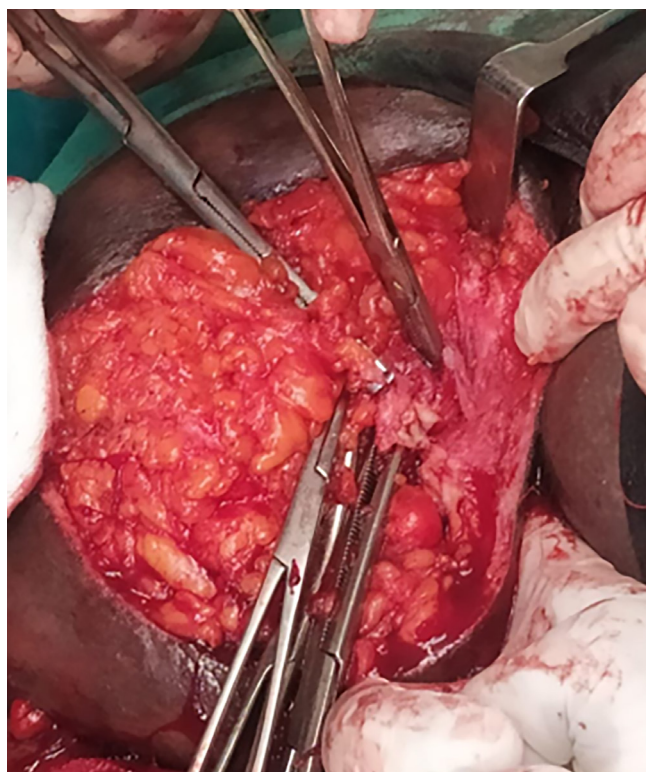


Figure 2. Intra operative fistulous tract picture.



Figure 3. Specimen picture.

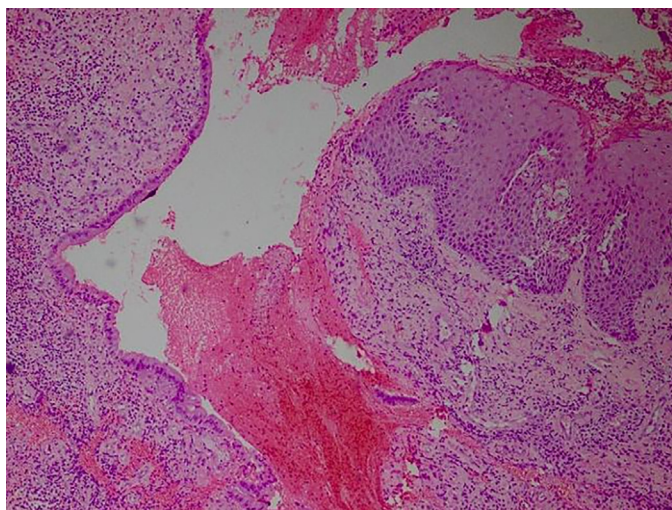


Figure 4. Histopathological picture.

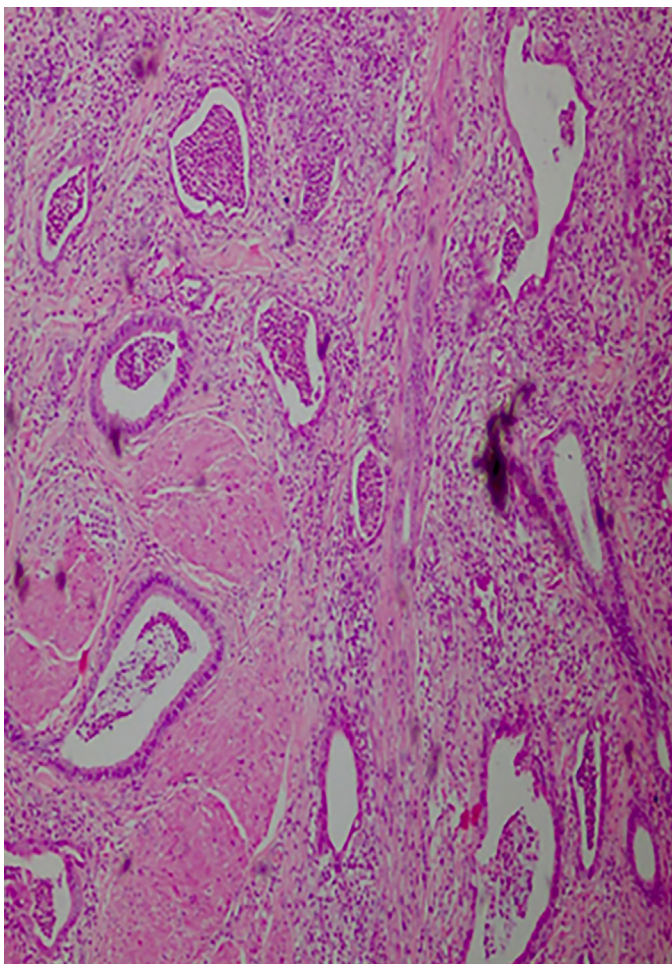


Figure 5. Histopathological picture.

DISCUSSION

It is been reported in the World Health Organization, MA is an invasive adenocarcinoma consisting of malignant glandular cells which contain intracytoplasmic mucin and the infiltrating glandular



Figure 6. Post operative picture.



Figure 7. Post operative picture.

structures are associated with mucoid stromal formation (5). Though few articles state that malignant transformation of a long-standing PF is attributed to mucosal regeneration, but few set of authors think that malignant cells settle in the fistulous granulation tissue arising from proximal gastrointestinal malignancy (6). Associated symptoms are pain in the perianal region, itching, mucinous discharge or pus discharge, and perianal ulcero-proliferative growth. However, rare involvement of the rectal mucosa, intestinal obstruction, or rectal bleeding are uncommon symptoms (7-9). While biopsy is remains the initial essential investigation for diagnosis, endoscopic ultrasound, colonoscopy, computed tomography, and MRI help in diagnosing perianal MA (PMA), with MRI being the most sensitive in demonstrating the mucinous structures (10). Many studies on PMA

indicate that the treatment of choice is radical surgical excision, with APR.

The role of neoadjuvant/adjuvant chemoradiotherapy in PMA is still controversial as it lacks proven data. However, The Journal of Surgical Oncology has provided data suggesting that neoadjuvant chemoradiotherapy can be a part of the treatment of PMA in order to reduce local recurrence and improve survival rate (11).

CONCLUSION

In this case report, we would like to highlight our patient did not present with typical symptoms and signs of malignancy which could have delayed the diagnosis and treatment, but with prompt investigation it is possible to come to the diagnosis at the earliest. Though APR is the definitive treatment for PMA, we were able to get good result with wide local excision and V-Y flap.

Ethics

Informed Consent: Informed consent it was obtained.

Footnotes

Authorship Contributions

Surgical and Medical Practices: K.S.R., S.K.S., G.G.M., J.K.R., K.P.S., J.S., Concept: K.S.R., S.K.S., K.P.S., Design: K.S.R., J.K.R., Data Collection or Processing: G.G.M., Analysis or Interpretation: S.K.S., Literature Search: J.K.R., J.S., Writing: K.S.R.

Conflict of Interest: No conflict of interest was declared by the authors.

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