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Ulcerated Lesions are not always Present in Solitary Rectal Ulcer Syndrome: A Case Report

Faisal Mujib[1*], Basim Saad Alharbi[2], Dhilal Issaoui[3], Yasir Saeed Aldughar[4], Juhaynah Abdullah Alshehri[5] and Shrooq Saad Alharbi[6]

[*1]Registrar Internal Medicine Imam Abdulrahman Alfaisal Hospital., cluster1, Riyadh, Saudi Arabia
[2]Internal medicine and gastroenterology consultant Head of internal medicine department Imam Abdulrahman Alfaisal Hospital., cluster1, Riyadh, Saudi Arabia
[3]Consultant Gastroenterologist Imam Abdulrahman Alfaisal Hospital., cluster1, Riyadh, Saudi Arabia
[4]HistopathologistAnatomic Pathology Srmrctio. Regional laboratory, Riyadh shared Services Ministry of Health Saudi Arabia
[5]Resident Internal Medicine Imam Abdulrahman Alfaisal Hospital., cluster1, Riyadh, Saudi Arabia
[6]Intern Internal Medicine Imam Abdulrahman Alfaisal Hospital., cluster1, Riyadh, Saudi Arabia

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Abstract

Solitary rectal ulcer syndrome (SRUS) is a benign chronic disorder that usually occurs in young adults. It is characterized by various symptoms, such as rectal bleeding, copious mucus discharge, prolonged excessive straining, perineal and abdominal pain, a feeling of incomplete defecation, constipation, and, rarely, rectal prolapse. The etiology of this syndrome remains unclear, and its diagnosis is easily confused with that of other diseases, contributing to difficulties in treatment. Here, we present a case of an 18-year-old male with complaints of constipation, weight loss and lower abdominal pain. Colonoscopy showed a polypoidal erythematous rectal lesion grossly resembling rectal cancer. Histopathology of biopsy specimen from lesion showed no evidence of malignancy and showed features of SRUS.

***Corresponding author:** Faisal Mujib, Registrar Internal Medicine Imam Abdulrahman Alfaisal Hospital., cluster1, Riyadh, Saudi Arabia.

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Learning Points

- SRUS can present diagnostic difficulties and may mimic a malignant neoplasm on endoscopy.
- The clinician and pathologist should keep a high index of suspicion to diagnose SRUS once the more sinister causes have been excluded.

Introduction

SRUS is a rare and enigmatic disorder affecting the rectum and anal canal. A previous article called SRUS "the three-lies disease", as it is not always a single lesion, it does not occur exclusively in the rectum, and ulcers are not present in every case [1].

SRUS occurs in 1 in 100,000 people and is more common in men than in women. It is characterized by a spectrum of clinical symptoms and endoscopic findings, posing diagnostic and therapeutic challenges. The condition primarily affects young and middle-aged adults and involves multifactorial etiologies, including abnormal defecation dynamics, increased intra-rectal pressure, and mucosal prolapse [2].

Case Description

This report describes the case of an 18-year-old male with a 1-year history of intermittent lower abdominal pain, constipation, and incomplete bowel evacuation. He had a history of COVID-19 1 year prior, and following recovery, he began to experience the above-mentioned symptoms. He also had a history of weight loss of approximately 6 kg. The patient had no history of special sexual behaviour (such as anal intercourse) and no family history of cancer. His lab results were unremarkable, with a haemoglobin (Hb) level of 15.7 g/dL and normal LFT, urea, and creatinine levels. A computed tomography (CT) scan of the abdomen showed a long segment of diffuse annular thickening involving the rectum that spanned from the S2–3 level to 5 cm from the anal verge, almost obliterating the lumen. The lesion was moderately and heterogeneously enhanced with contrast. Later, a colonoscopy revealed a 3–4 cm polypoidal erythematous lesion with whitish spots 5 cm from the anal verge. (Figure 1).

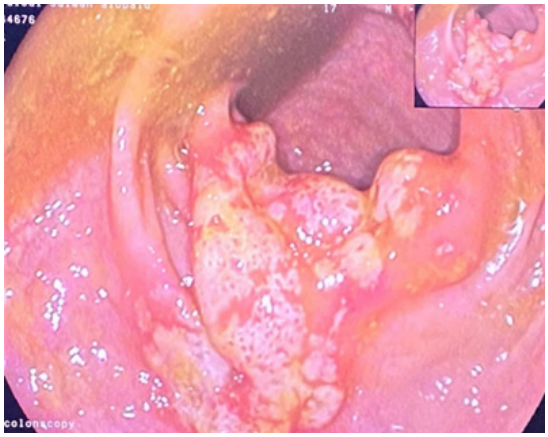

Figure 1: Endoscopic view of the rectal lesion: a 3–4 cm polypoidal erythematous lesion.

Malignancy was suspected from naked eye inspection. Biopsies showed superficial colonic mucosa with ulceration and granulation tissue but no malignancy. The colonoscopy was repeated, and additional biopsies were taken, leading to a diagnosis of solitary rectal ulcer syndrome (SRUS) with no evidence of dysplasia or metaplasia (Figure 2).

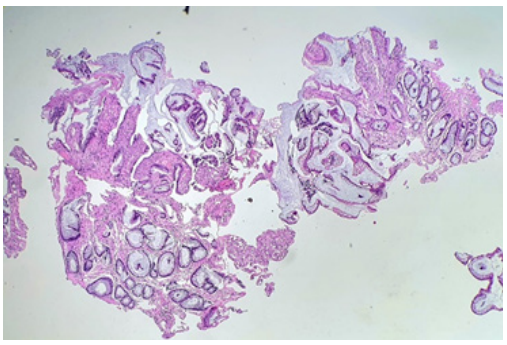

Figure 2a: Distorted glandular architecture, paucity of inflammatory cell infiltrate, and fibromuscular disarray of the lamina propria.

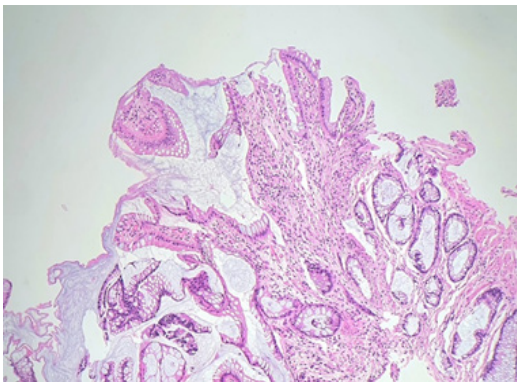

Figure 2b: Surface ulceration and disruption of several glands.

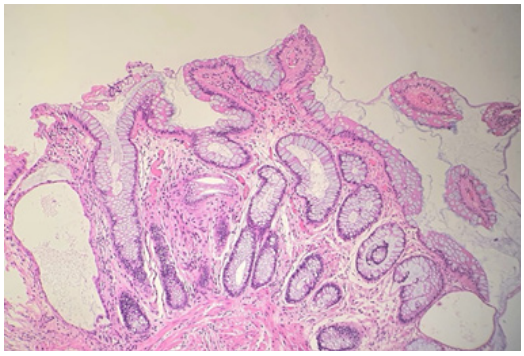

Figure 2c: On high magnification, smooth muscle bundles were observed in the lamina propria. Note the congested capillaries in the stroma.

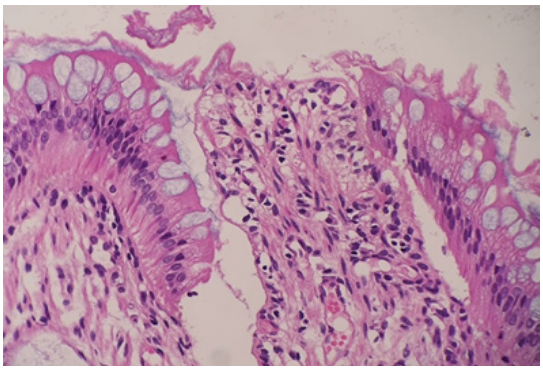

Figure 2d: Fibroplasia and proliferation of ectatic capillary-sized vasculature beneath the ulcerated epithelium with bland-appearing nuclear morphology of the epithelium.

Discussion

SRUS often presents with multiple symptoms, such as rectal bleeding, mucorrhea, and varying degrees of defecation difficulties. Patients may also experience anorectal pain and a sensation of incomplete evacuation. However, the clinical presentation can be heterogeneous, making diagnosis challenging [3].

Endoscopy serves as the gold standard for diagnosing SRUS. It typically reveals single or multiple ulcerations of varying size and appearance in the rectal mucosa [3]. It can also reveal a polypoidal mass, which was the case in our patient. Such lesion often mimics a neoplastic process. Endoscopic ultrasound can help elucidate the involvement of the rectal wall layers. Anorectal manometry may reveal abnormal patterns of rectal contractions and relaxation [4]. Dynamic imaging with defecography can determine the presence of rectal prolapse and excessive perineal descent during defecation [5].

Under a microscope, solitary ulcers exhibit distinct histopathological features. Mucosal ulcers are central to SRUS pathology, and they vary in size and depth in most cases. However, mucosal ulcers are not seen in other cases that may exhibit intact polypoid or flat configuration. Often, concomitant fibromuscular hyperplasia is observed, an indication of thickened rectal muscular layers. Furthermore, disarray within the rectal wall's smooth muscle fibers is consistently observed. Additionally, histological examination commonly unveils fibrotic changes, distorted glandular architecture, and a paucity of inflammatory cell infiltrate. The absence of cellular atypia and architectural dysplasia exclude invasive adenocarcinoma. These discernible histopathological traits serve as critical diagnostic indicators, complementing clinical and endoscopic assessments to ensure a comprehensive and precise diagnosis of SRUS [6].

SRUS management aims to alleviate symptoms and enhance patients' quality of life. Strategies adopted depends on the severity of symptoms. Conservative management including dietary modifications, fiber supplementation, stool softeners, and Biofeedback improve bowel habits and reduce symptoms in many patients [7]. Medications such as sucralfate enemas and topical steroids are efficacious in some cases. Sucralfate enemas are thought to promote the healing of rectal ulcers [8]. Surgical intervention is considered for refractory cases or in patients with complications such as rectal prolapse. Surgical options include rectopexy and trans-anal mucosectomy [9]. Other modalities used in some case of SRUS include endoscopic mucosectomy and argon plasma coagulation. SRUS generally has a favourable prognosis, particularly when it is diagnosed and managed promptly. Most patients experience substantial symptom relief with appropriate treatment [10].

Conservative management strategy including high fiber diet and Lactulose was adopted in this patient and he showed marked symptomatic improvement.

In this case, SRUS presented in the form of a large polypoidal mass, which was suspected as a malignant polyp by its endoscopic appearance. Histopathologic evaluation did not show any features suggestive of a malignant process and confirmed the diagnosis of SRUS.

Conflict of Interest Statement

The authors have no conflicts of interest or financial ties to disclose.

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