SOLITARY RECTAL ULCER SYNDROME – A CASE REPORT AND LITERATURE REVIEW – DIAGNOSTIC AND THERAPEUTIC PITFALLS

SINDROM SOLITARNOG REKTALNOG ULCERA – PRIKAZ SLUČAJA I PREGLED LITERATURE – DIJAGNOSTIČKA I TERAPIJSKA ZAMKA

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Case report
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Abstract

Introduction. Solitary rectal ulcer syndrome typically presents as painful or difficult defecation and rectal bleeding. The aim of this case report is to highlight how misinterpretation of histopathological findings following colonoscopy can lead to unnecessary and potentially mutilating surgical interventions. **Case Report.** A 58-year-old man was diagnosed with rectal cancer based on histopathological findings obtained after colonoscopy and supported by pelvic magnetic resonance imaging. A transanal excision of the suspected tumor mass was performed. However, definitive histopathological analysis of the surgical specimen revealed features consistent with solitary rectal ulcer syndrome. Subsequent revision of the initial colonoscopic biopsy by an experienced colorectal pathologist confirmed the diagnosis of solitary rectal ulcer syndrome. Follow-up colonoscopy performed three months postoperatively showed no evidence of tumors, ulcerations or polyps in the colon. **Conclusion.** Careful integration of clinical presentation, endoscopic findings, imaging, and expert histopathological evaluation is essential to avoid misdiagnosis and overtreatment. In suspected cases of solitary rectal ulcer syndrome, biopsy specimens should be reviewed by pathologists experienced in colorectal pathology do prevent unnecessary radical surgical procedures.

Key words: Rectal Diseases; Ulcer; Rectum; Gastrointestinal Hemorrhage; Colonoscopy; Biopsy; Treatment Outcome; Diagnostic Errors; Diagnosis

Sažetak

Uvod. Solitarni rektalni ulkus karakteriše bolna i otežana defekacija praćena rektoragijom. Cilj ovog prikaza slučaja je da pokaže kako pogrešna dijagnoza zasnovana na patohistološkom nalazu dobijenom nakon kolonoskopije može dovesti do izuzetno mutilantnih hirurških intervencija. **Prikaz slučaja.** Pedesetosmogodišnji pacijent sa inicijalnom dijagnozom karcinoma rektuma te nakon obavljene onkološke komisije za tumore kolorektuma donesena odluka o izvođenju amputacije rektuma, s obzirom na nalaze patohistološke analize i magnetne rezonance male karlice. Urađena je transanalna ekscizija tumora, a nakon definitivne patohistološke analize postavljena je dijagnoza sindroma solitarnog rektalnog ulkusa. Nakon revizije patohistološkog nalaza preparata dobijenog na kolonoskopiji, iskusni patolog je potvrdio dijagnozu dobijenu na operativnom materijalu. Kontrolna kolonoskopija je urađena tri meseca nakon operacije, a pacijent je bio bez prisustva tumora, ulceracija ili polipa u kolonu. **Zaključak.** Pažljivim pristupom svim anamnestičkim podacima kao i fizikalnim pregledom, te uvidom u ostale nalaze, može se izbeći prekomerni hirurški tretman pacijenata. U slučajevima rektalnog ulkusnog sindroma

Ključne reči: bolesti rektuma; uklus; rektum; gastrointestinalno krvarenje; kolonoskopija; biopsija; ishod lečenja; dijagnostičke greške; dijagnoza

Introduction

Solitary rectal ulcer syndrome (SRUS) was first described by Cruveihier in 1820s as a chronic disorder affecting of the rectal mucosa [1]. Despite its name, SRUS does not always present as a solitary lesion or an ulcer. The clinical and endoscopic spectrum ranges from erythematous mucosal changes to polypoid lesions and well-developed ulcers. These lesions are most commonly located 4-10 cm from the anocutaneous line (ACL) [1–4].

The pathogenesis of SRUS remains incompletely understood; however, several predisposing factors have been implicated, including prolonged straining during defecation, chronic constipation, and anatomical abnormalities of the rectum [4,5]. The estimated incidence is approximately one case per 100,000 individuals per year, with predominance in females [1–3]. SURS most commonly affects patients in the third and fourth decades of life.

The term *mucosal prolapsed syndrome* has been proposed as a more accurate designation, as the condition is not invariably rectal, ulcerative, or solitary. Clinically, SRUS is characterized by painful or difficult defecation, a sensation of incomplete evacuation, constipation, and rectal prolapse. Notably, up to 26% of patients may be asymptomatic. Lower gastrointestinal bleeding occurs in approximately 56%

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Abbreviations

SRUS – solitary rectal ulcer syndrome
ACL – anocutaneous line
HP – histopathological
EUS – endoscopic ultrasound

of cases, excessive straining during defecation 28%, and pelvic fullness in 23%, all of which may lead to misdiagnosis as inflammatory bowel disease or rectal malignancy [1–6].

Diagnosis relies on a combination of clinical presentation, endoscopic appearance, and histopathological (HP) evaluation. As clinical and endoscopic findings are often nonspecific, definitive diagnosis depends on characteristic HP features, including surface serration, fibromuscular obliteration, crypt distortion, and vascular abnormalities [5,7].

The aim of this case report is to demonstrate how an incorrect histopathological diagnosis following colonoscopic biopsy can result in unnecessary and potentially mutilating surgical treatment.

Case Report

A 58-year-old man was admitted to the Department of Surgery at the Oncology Institute of Vojvodina with a three-month history of progressively worsening symptoms, including intermittent abdominal pain, rectal bleeding, and a persistent sensation of incomplete evacuation. The rectal bleeding was painless, fresh, and mixed with stool. The patient reported chronic constipation and regular use oral laxatives. He had experienced an unintentional weight loss of approximately 6 kg over the preceding six months. The patients had no personal history of diabetes mellitus, hypertension, allergic reactions, or previous anorectal surgery. Family history was negative of gastrointestinal malignancies.

Digital rectal examination revealed no palpable rectal tumors, hemorrhoids, or fissures. A nonspecific, slightly protruding firmness was palpated on the posterior rectal wall.

Colonoscopy was recommended and subsequently performed at an external institution. Retroversion revealed a tumor-like mass measuring approximately 3 cm in diameter, located 2 cm from the ACL on the posterior rectal wall. Biopsy samples were obtained, and HP reported rectal adenocarcinoma (G2) with a mucinous component of approximately 20% **(Figure 1).** Neither the colonoscopy not the histopathological analysis was performed at the Oncology Institute of Vojvodina.

Further staging included chest and abdominal computerized tomography, which showed no evidence of distant metastases. Pelvic magnetic resonance imaging demonstrated a 15 mm lesion infiltrating the perirectal tissue, located 2 cm from ACL, adjacent to but not infiltrating the anal sphincters (radiological stage T1N0M0). Several perirectal lymph nodes were enlarged but without radiological features suggestive of malignant involvement. Tumor markers (CEA and CA 19-9) were within reference ranges. Based on these findings, the multidisciplinary oncology team initially recommended surgical treatment in the form of rectal amputation.

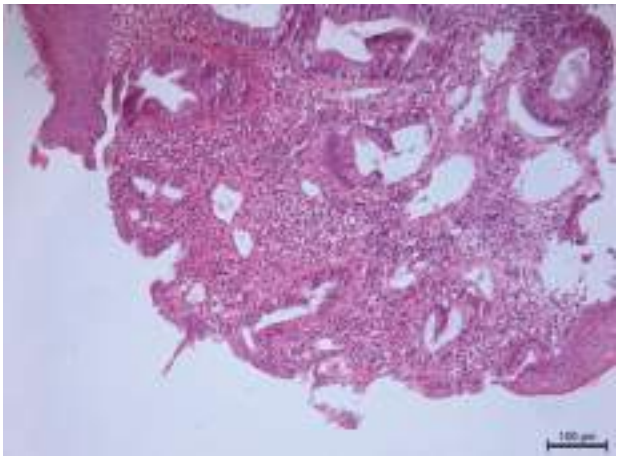

Figure 1. Histopathological findings of colonoscopic biopsy
Solitary rectal ulcer syndrome, H&E x 20. (Biopsy specimen demonstrating ulcerated surface of anorectal mucosa. Cryptal hyperplasia with dilatation is present, with moderate ectatic capillaries and inflammatory infiltrate.)

A transanal excision of the lesion was performed, and the surgical specimen was submitted for frozen-section analysis to assess the deep resection margin. HP examination revealed low- and high- grade dysplasia of the rectal mucosa. Gross examination showed a flattened, ulcerated mucosal area. Microscopic analysis demonstrated ulceration with prominent fibromuscular hyperplasia and thickening of the muscularis mucosae. Crypt distortion was mild, inflammatory infiltrate was minimal, and ectatic capillaries were conspicuous. These findings were consistent with solitary rectal ulcer syndrome **(Figures 2A and 2B).**

Following this result, the original colonoscopic biopsy was evaluated by an experienced colorectal pathologist. Revision of the specimen confirmed the diagnosis of SRUS. Histology revealed an ulcerated anorectal mucosal surface with distorted crypts lined by hyperplastic colonic epithelium. The lamina propria was thickened, with discrete fibromuscular hyperplasia and ectatic blood vessels. A moderate inflammatory infiltrate composed of lymphocytes and plasma cells was present, with no evidence of dysplasia **(Figure 1).** Consequently, the diagnosis of rectal cancer was definitely excluded.

Follow-up colonoscopy performed three months postoperatively showed no evidence of tumors, ul-

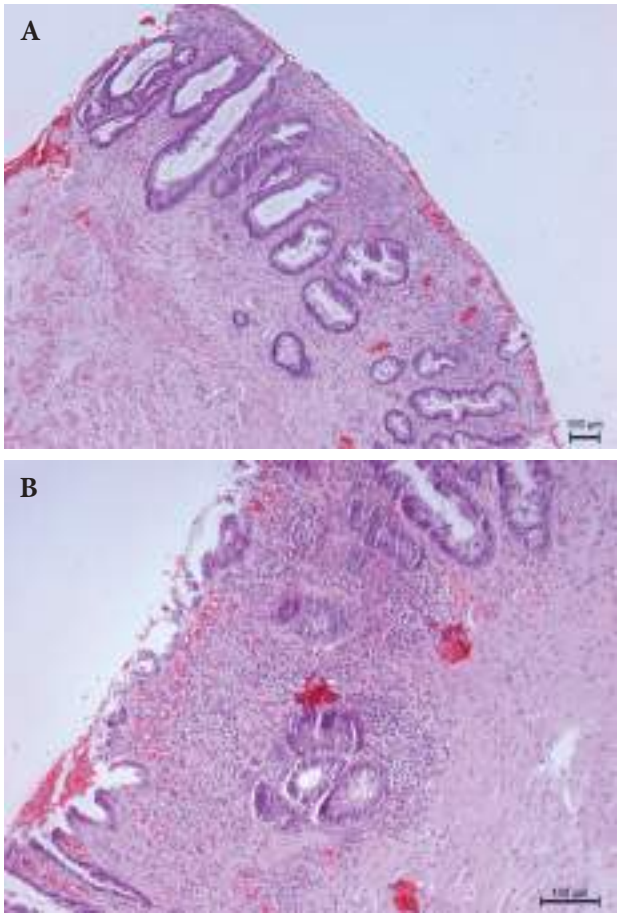

Figure 2 (A, B). Histopathological findings of the surgical specimen

Solitary rectal ulcer syndrome, H&E x 10. (Operative specimen confirming ulcerated surface with irregular crypts lined with hyperplastic epithelium. Muscularis propria is thickened and fibromuscular hyperplasia is present. Ectatic capillaries are marked and inflammation is mild.)

cerations, or polyps. At one-year follow-up, the patient remained asymptomatic.

Discussion

The etiology and pathophysiology of this rare benign disorder remain incompletely understood, and the clinical presentation of SRUS is notably heterogeneous. Three main hypotheses have been proposed to explain its pathophysiology. The first suggests that chronic mechanical trauma and local ischemia of the rectal mucosa - resulting from prolonged straining, constipation, tenesmus, intussusception, and chronic inflammation may play a central role in mucosal injury and ulcer formation [6–8]. The second hypothesis implicates paradoxical contraction of the puborectalis muscle during defecation, a feature commonly observed in patients with pelvic floor dyssynergia. This abnormal contraction may contribute to overt or occult rectal prolapse over time, leading to repetitive ischemic injury and mucosal ulceration. Although paradoxical puborectalis contraction can also be observed in healthy individuals, anorectal physiology studies have demonstrated dyssynergia in approximately 25–82% of patients with SRUS, supporting its role in disease development [8]. The third hypothesis proposes abnormal defecation dynamics caused by a reversed pressure gradient generated by inappropriate contraction of the external anal sphincter during evacuation [6–8].

SRUS is a well-recognized clinical and endoscopic mimicker of serious colorectal diseases, particularly rectal carcinoma and inflammatory bowel diseases. Endoscopic findings vary widely and may include erythematous patches, solitary or multiple ulcers, and non-ulcerative polypoid or mass-like lesions that closely resemble rectal malignancy of inflammatory bowel disease [1–3,6,8]. Lesions vary in size and location but are most commonly up to 4 cm in diameter and located on the anterior rectal wall [8]. In contrast, the lesion in our patient measured 15 mm and was located on the posterior rectal wall, highlighting the variability of SRUS presentation. Clinically, the classic symptom triad consists of rectal bleeding, constipation, and abdominal pain.

Several studies have suggested that endoscopic ultrasound (EUS) may be superior to MRI in the diagnostic evaluation of SRUS [9,10]. Typical EUS findings include thickening of the rectal wall and internal anal sphincter, mucosal intussusception, submucosal cysts, hyperechogenic fibrotic bands within the submucosa, and regional lymph node enlargement. Additionally, a reduced ratio of external to internal anal sphincter thickness has been reported in patients with SRUS [11].

HP examination remains the cornerstone of diagnosis. Characteristic features include fibromuscular obliteration of the lamina propria, hypertrophy and disorganization of the muscularis mucosae, and distorted crypt architecture. The most commonly reported HP findings include superficial ulceration (59%), crypt distortion (17%), and inflammatory infiltrated (33%) [3,8]. These changes reflect chronic mucosal injury and regenerative processes. Importantly, collagen disposition within the lamina propria is a key feature distinguishing SRUS from inflammatory bowel disease and chronic ischemic colitis [12]. Malignant lesions may initially present as superficial ulcers or polypoid masses; therefore, adequate biopsy and expert pathological interpretation are essential to exclude neoplastic infiltration. Although colonoscopy is indispensable for diagnosis, our case clearly demonstrated that misdiagnosis of biopsy specimens can lead to an incorrect diagnosis and potentially devastating overtreatment.

Management of SRUS begins with patient education and behavioral modification, which represent the foundation of therapy. Patients should be instructed in proper defecation techniques, including relaxation of the pelvic floor and external anal sphincter during evacuation. In selected cases, Kegel exercises may be beneficial [3]. Additional behavioral measures include a high-fiber diet, smoking cessation, adequate hydration (at least two liters of water daily), regulation of toilet habits, treatment of underlying psychological disorders, and avoidance of excessive straining or digital evacuation [13]. Patients with persistent or severe symptoms may require medical or surgical intervention. Topical therapies such as sucralfate, 5-ASA, sulfasalazine, and corticosteroid enemas have been reported to reduce inflammation and preventing irritant injury [1–15]. Surgical treatment is reserved for refractory cases and includes procedures such as rectopexy for rectal prolapse, the Delorme procedure for mucosal resection, or perirenal proctectomy (Altemeier procedure) [1–3,6,8,16].

Conclusion

A careful and systematic evaluation of clinical, endoscopic, radiological, and histopathological data is essential to avoid diagnostic errors and unnecessary surgical overtreatment in patients with solitary rectal ulcer syndrome. This case underscores the clinical importance of expert histopathological assessment by experienced colorectal pathologist.

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