

# Incidence of Solitary Rectal Ulcer Syndrome in Patients Undergoing Colonoscopy

 **Ufuk Kutluana**

Department of Gastroenterology, Usak University Faculty of Medicine, Usak Training and Research Hospital, Usak, Turkey

## Abstract

**Introduction:** This study aimed to investigate the incidence of solitary rectal ulcer syndrome in patients who underwent colonoscopy or sigmoidoscopy and were ultimately diagnosed with solitary rectal ulcer syndrome at our center.

**Methods:** Solitary rectal ulcer syndrome is a chronic benign condition that commonly presents with rectal bleeding, mucous discharge, and constipation. The diagnosis of this syndrome is based on clinical symptoms and endoscopic and histological features. The incidence rate of the syndrome has been noted to be 1 in 100.000 per year.

**Results:** We performed a retrospective analysis of patients who underwent colonoscopy or sigmoidoscopy by the same gastroenterologist at our center between 2012 and 2017. Data retrieved included demographic details, comorbidities, clinical presentations, laboratory, endoscopic, and histopathological findings, treatment procedures, and clinical outcomes.

**Discussion and Conclusion:** The incidence rate of solitary rectal ulcer syndrome in patients who underwent colonoscopy was 0.68%. The most common symptoms included rectal bleeding, mucous discharge, and constipation.

The incidence rate observed here was significantly higher than that in the previous report. Although the syndrome was described as a benign disease, it can result in serious complications including rectovaginal fistula, which was observed in one patient.

**Keywords:** Colonoscopy; solitary rectal ulcer syndrome; sigmoidoscopy.

Solitary rectal ulcer syndrome (SRUS) is a chronic benign condition that commonly presents with rectal bleeding, mucous discharge, constipation, rectal discomfort, and the use of digital maneuvers to defecate. The underlying etiology is not well understood, but secondary ischemic changes and trauma in the rectum associated with paradoxical contraction of the pelvic floor and external anal sphincter muscles have been implicated in the pathogenesis [1]. SRUS diagnosis is based on clinical symptoms combined with endoscopic and histological features. Endoscopic findings vary. Single or multiple ulcers, hyperemic mucosa, or poly-

loid lesions in the rectum or the other sides of the colon can be observed. Therefore, SRUS is known as “the three lies disease” [2]. The lesion may mimic other rectal pathology and may result in a misdiagnosis, such as infectious, malignancy, or inflammatory bowel disease (IBD) [1]. The incidence of SRUS is uncertain, but in one demographic study, it has been noted to be 1 in 100.000 per year [3]. Some authors suggest that this condition may not be as rare as previously reported [4-6]. This study aimed to investigate the incidence of SRUS in patients who underwent colonoscopy or sigmoidoscopy and were ultimately diagnosed with SRUS at our center.

**Correspondence (İletişim):** Ufuk Kutluana, M.D. Department of Gastroenterology, Usak University Faculty of Medicine, Usak Training and Research Hospital, Turkey

**Phone (Telefon):** +90 505 502 47 70 **E-mail (E-posta):** drufukkanas@gmail.com

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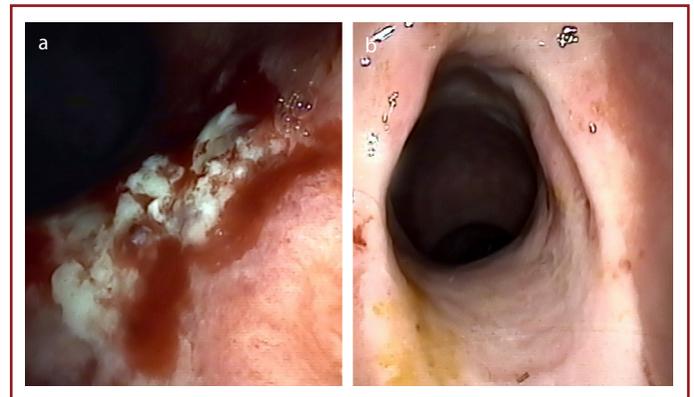
## Materials and Methods

We performed a retrospective analysis of patients who underwent colonoscopy or sigmoidoscopy by the same gastroenterologist at the gastroenterology department at our hospital between March 2012 and September 2017. The inclusion criteria of the patients included presence of SRUS. The diagnosis of SRUS was based on a combination of symptoms, endoscopic findings, and histopathological features [7]. Data retrieved included demographic details, comorbidities, clinical presentations, laboratory, endoscopic, and histopathologic findings, management procedures, and clinical outcomes. Hemoglobin (Hb) level less than 11 g/dl was defined as anemia. Lesions on endoscopic findings were categorized based on appearance as ulcerated, polypoidal, or erythematous; based on numbers as solitary or multiple; and based on location of rectal wall as anterior, anterolateral, or circumferential. The histological criteria for the diagnosis of SRUS included fibromuscular obliteration, in which the lamina propria is replaced with smooth muscle and collagen. This leads to hypertrophy and a thickened mucosal layer with distortion of the crypt architecture and disorganization of the muscularis mucosa [8]. The study was approved by the ethics board of Usak University Faculty of Medicine dated March 13, 2018 Nr.28-02. The statistical analysis of the data was performed using SPSS version 23. The descriptive distribution statistics of the study group (mean values, frequency, standard deviation) were recorded by calculations. For categorical variables, data was expressed using frequencies. To compare cases with qualitative variables, an analysis of contingency tables was carried out using Pearson's Chi-squared test. Fisher's exact test was used where appropriate. The quantitative variables were expressed as mean±standard deviation. Normal distribution of the variables was checked by the Saphiro–Wilk test.

## Results

All patients with SRUS who underwent colonoscopy or sigmoidoscopy between March 05, 2012 and September 20, 2017 were included in this study. During the same period, 1.317 colonoscopies or sigmoidoscopies were performed by the same gastroenterologist. In total, nine patients (0.68%) comprising six women and three men were diagnosed with SRUS. The mean age of the nine patients was 52 years (range, 17–83 years). The mean duration of onset of the symptoms to diagnosis was 21 months (range, 11–26 months). Two patients had diabetes mellitus, and one patient had hypertension. Three patients had irritable bowel syndrome (IBS). Three patients had anemia. Three patients'

history included rectal digitation, and four patients' history included straining. Most patients presented with rectal bleeding and mucous discharge, followed by constipation, tenesmus, abdominal pain, and perianal pain. Although most of the patients presented with constipation, none had diarrhea. The mean Hb levels were 11.8 g/dl (range, 6.7–16.8 g/dl). In all patients, the lesions were located at the rectum. Single or multiple ulcers were observed in seven patients (single, five; multiple, two). In terms of shape, ulcers were round in four, linear (Fig. 1a) in one, and oval (Fig. 1b) in two patients and measured 6–20 mm in diameter (mean, 12.4 mm). Polypoidal lesions (Fig. 2a) were observed in one patient. Erythematous area was observed in one patient. The distance of lesions to the anal canal varied from 5 to 12 cm (mean, 7.3 cm). Eight lesions involved the anterior rectal wall, whereas one lesion involved the anterolateral wall. One patients had rectal prolapse. Rigidity at the edge of the lesion and difficulty during biopsy acquisition were recorded in seven patients. In this study, rectovaginal fistula (RVF) was observed in one patient (Fig. 1b).



**Figure 1.** (a) Endoscopic appearance of a single oval ulcer with surrounding erythema observed in the rectum; (b) an endoscopic appearance of a linear ulcer.

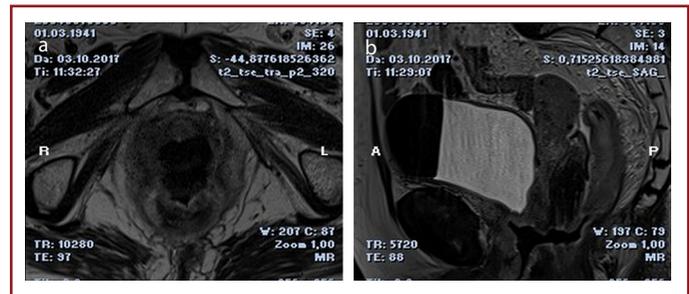


**Figure 2.** (a) Endoscopic appearance of a solitary rectal ulcer syndrome with polypoidal lesion; (b) Endoscopic appearance of a large oval solitary rectal ulcer complicated with rectovaginal fistula.

**Table 1.** Clinical characteristics and laboratory findings of patients with solitary rectal ulcer syndrome

| Case | Sex    | Age (yr) | Rectal bleeding | Symptoms Duration (month) | Mucous discharge | Constipation | Tenesmus | Abdominal pain | Straining | Anal pain | Digitation | IBS | Ad.Dis. | Anemia | Hb (g/dl) |
|------|--------|----------|-----------------|---------------------------|------------------|--------------|----------|----------------|-----------|-----------|------------|-----|---------|--------|-----------|
| 1    | Female | 17       | Yes             | 24                        | Yes              | Yes          | No       | No             | No        | Yes       | No         | Yes | No      | No     | 13.8      |
| 2    | Female | 83       | Yes             | 26                        | Yes              | Yes          | Yes      | Yes            | Yes       | No        | Yes        | No  | No      | No     | 11        |
| 3    | Female | 30       | No              | 14                        | Yes              | No           | No       | Yes            | No        | Yes       | No         | No  | No      | No     | 11.5      |
| 4    | Female | 51       | Yes             | 24                        | Yes              | Yes          | No       | No             | No        | No        | No         | No  | No      | Yes    | 8.8       |
| 5    | Male   | 48       | Yes             | 24                        | Yes              | No           | No       | Yes            | Yes       | No        | No         | Yes | No      | No     | 16.8      |
| 6    | Female | 76       | Yes             | 18                        | Yes              | Yes          | Yes      | Yes            | Yes       | Yes       | Yes        | Yes | Dm      | Yes    | 6.7       |
| 7    | Female | 47       | Yes             | 11                        | No               | No           | No       | Yes            | No        | No        | No         | No  | Ht      | Yes    | 8.8       |
| 8    | Male   | 56       | Yes             | 26                        | Yes              | Yes          | Yes      | No             | Yes       | No        | No         | No  | Ht      | No     | 13        |
| 9    | Male   | 60       | Yes             | 22                        | No               | Yes          | No       | No             | No        | No        | Yes        | No  | No      | No     | 14        |

IBS: Irritable bowel syndrome; Ad. dis: Additional disease; DM: Diabetes mellitus; HT: Hypertension; Hb: hemoglobin.



**Figure 3.** (a) Pelvic magnetic resonance imaging of a patient with solitary rectal ulcer showing a rectovaginal fistula in axial plane and (b) sagittal plane.

Tables 1 and 2 summarize the clinical characteristics, laboratory and endoscopic findings, and management of nine patients.

Conservative treatment involving a high fiber diet, using bulk laxatives, and avoiding straining and anal digitation was also used. All patients were conservatively treated along with topical medical treatment with sucralfate (2 g twice daily for 6 weeks). Five patients were treated with mesalamine (500 mg twice daily for 6 weeks) suppository, and three patients were treated with budesonide (2 mg once daily for 6 weeks) enema. Symptomatic response was observed in 66.7% of patients treated with sucralfate and in two patients treated with budesonide. Partial symptomatic response was observed in only one patient treated with mesalamine. Further, two patients with IBS received additional oral rifaximin (1200 mg daily for 2 weeks). Three patients with refractory symptoms were referred for biofeedback training and/or further therapies. Biofeedback treatment undertaken using the manometry method was applied to one patient. Rectopexy was performed to one patient with rectal prolapse. Local excision and fistula closure were applied to one patient with RVF.

## Discussion

SRUS is a chronic, benign, and rare condition. The pathogenesis of SRUS has been identified as chronic mucosal trauma and ischemia [1]. In one demographic study, the incidence of SRUS is 1 in 100,000 per year [3]. In the literature, some authors suggest that SRUS may not be as rare as previously reported [4-6]. Based on their experience, Haray et al. [5] believe that this condition may not be as rare as previously suggested. Tjandra, Knoepf et al. reported that the incidence of SRUS could not be so rare [4, 6]. The incidence rate of patients who underwent colonoscopy was 0.68% in this study. We could not find any data regarding this issue in the literature. In our study, a higher proportion of female patients were identified with a wide age range (17-83

**Table 2.** Endoscopic findings and management of patients with solitary rectal ulcer syndrome

| Case | Lesion type          | Ulcer type | Location on rectal wall | Distance of lesion from anal verge (cm) | Lesion Dia Meter (mm) | Difficulty biopsy | Rectal Prolapse | Recto Vaginal fistula | Conservative treatment | Response to Sucralfate treatment | Mesalamine treatment         | Steroid treatment            | Rifaximin treatment  | Clinical outcomes    |
|------|----------------------|------------|-------------------------|---|-----------------------|-------------------|-----------------|-----------------------|------------------------|----------------------------------|------------------------------|------------------------------|----------------------|----------------------|
| 1    | Polypoid lesions     | -          | anterior                | 12                                      | 15                    | Yes               | No              | No                    | Yes                    | Partial symptomatic response     | -                            | -                            | symptomatic response | clinical improvement |
| 2    | Solitary ulcer       | rounded    | anterior                | 6                                       | 10                    | Yes               | Yes             | No                    | Yes                    | Partial symptomatic response     | No                           | No                           | -                    | Further therapies*   |
| 3    | Multiple ulcers      | ovale      | anterior                | 8                                       | 15                    | No                | No              | No                    | Yes                    | symptomatic response             | -                            | -                            | -                    | clinical improvement |
| 4    | Multiple ulcers      | rounded    | antrolateral            | 5                                       | 12                    | Yes               | No              | No                    | Yes                    | Partial symptomatic response     | No                           | symptomatic response         | -                    | clinical improvement |
| 5    | Solitary ulcer       | rounded    | anterior                | 7                                       | 16                    | No                | No              | No                    | Yes                    | Partial symptomatic response     | No                           | -                            | symptomatic response | clinical improvement |
| 6    | Solitary ulcer       | ovale      | anterior                | 6                                       | 20                    | Yes               | No              | Yes                   | Yes                    | Partial symptomatic response     | No                           | -                            | -                    | Further therapies*   |
| 7    | Solitary ulcer       | linear     | anterior                | 6                                       | 10                    | Yes               | No              | No                    | Yes                    | bad symptomatic response         | Partial symptomatic response | Partial symptomatic response | -                    | Further therapies*   |
| 8    | Erythematous lesions | -          | anterior                | 6                                       | 6                     | Yes               | No              | No                    | Yes                    | symptomatic response             | -                            | -                            | -                    | clinical improvement |
| 9    | Solitary ulcer       | rounded    | anterior                | 10                                      | 8                     | Yes               | No              | No                    | Yes                    | symptomatic response             | -                            | -                            | -                    | clinical improvement |

See the text for further therapies\*

years), similar to four previous studies [1, 9-11]. In our study, rectal bleeding, mucous discharge, and constipation were the most common symptoms. Mucous discharge is not often the predominant symptom; usually, it is overshadowed by tenesmus and constipation in SRUS [1, 9, 11]. However, in our study, mucous discharge was observed as frequently as rectal bleeding. In this study, three patients' history included rectal digitation. Rectal digitation has been hypothesized as a possible cause of SRUS [12, 13].

In this study, the endoscopic spectrum was a wide variety. In contrast to the name of disease, multiple ulcers, polypoidal lesions, and erythematous area were observed in two, one, and one patient, respectively. Remarkably, SRUS, which is known as a benign disease, was complicated with a serious condition such as RVF in one patient (Fig. 3 a, b). In the recent study, rigidity at the edge of the lesion and difficulty while taking a biopsy specimen was observed in seven patients. Difficulty during biopsy acquisition may be indicative of SRUS. Histopathological assessment is key to establishing SRUS diagnosis. To confirm SRUS diagnosis, histopathological assessment of biopsy material was performed in all our patients.

Treatment of SRUS typically depends on the severity of symptoms and whether there is an underlying rectal prolapse or not. The management of SRUS includes conservative treatment, bulking agents (lactulose), enemas (sucralfate, steroid, and mesalamine), oral 5-ASA, bowel retraining with or without biofeedback, endoscopic steroid injection, and surgery in refractory cases not responding to conservative treatments [14]. The initial conservative treatment remains the cornerstone of the treatment of SRUS [15]. Topical treatments, including sucralfate, mesalamine, and corticosteroids are reportedly effective [16, 17]. In this study, all the patients were treated with an initial conservative treatment and topical medical treatment with sucralfate (2 g twice daily for 6 weeks). Partial and total symptomatic responses were observed in five and one patient, respectively. Five patients were treated with mesalamine (500 mg twice daily for 6 weeks) suppository. Partial symptomatic response was observed in only one patient treated with mesalamine. Of three patients treated with budesonide (2 mg once daily for 6 weeks) enema, partial and total symptomatic responses were observed in one patient each. One patient was unresponsive to budesonide treatment. Rifaximin was effective in patients with IBS [18]. To our knowledge, there is no data regarding rifaximin treatment for SRUS in the literature. In this study, two patients with IBS received additional oral rifaximin (1200 mg daily for 2 weeks). These patients displayed a dramatic symptomatic response to rifaximin

following inadequate topical and conservative therapy.

Consequently, in our series, clinical improvement was achieved in a majority of patients. Based on our observation, sucralfate, budesonide, and rifaximin are relatively effective agents for the treatment of SRUS. Three patients with refractory symptoms were referred for further therapies. Argon plasma coagulation (APC) has been recently reported to control bleeding and improve the healing of lesions in SRUS [19]. APC can be selected for patients with refractory to medical treatments before considering surgical procedures. Our study has several limitations. First, the number of cases were relatively few. Second, the study had a retrospective design. Third, important investigations such as defecography and anal manometry were not performed and accordingly, could not be included in the study.

In conclusion, the incidence rate of SRUS in patients who underwent colonoscopy was 0.68%. This condition may not be as rare as previously suggested. The endoscopic findings were various. Difficulty during biopsy acquisition may be indicative of SRUS. Conservative and topical treatment is relatively successful in SRUS. Furthermore, rifaximin may be a treatment option for patients with SRUS and IBS. Although SRUS is described as a benign disease, it can cause serious complications such as RVF. Further prospective studies are required to clarify these observations.

**Ethics Committee Approval:** The study was approved by the ethics board of Usak University Faculty of Medicine dated March 13, 2018 Nr.28-02.

**Peer-review:** Externally peer-reviewed.

**Conflict of Interest:** None declared.

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