Clinical, Endoscopic, and Histologic Characteristics of Patients with Solitary Rectal Ulcer Syndrome at a Tertiary Care Center

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Abstract

Objective Solitary rectal ulcer syndrome (SRUS) is a chronic disorder of defecation presenting with bleed per rectum, mucorrhea, tenesmus, perianal discomfort, etc. We aim to report clinical, endoscopic, and histologic features of SRUS in patients who underwent either colonoscopy or sigmoidoscopy at our center.

Materials and Methods We performed retrospective analysis of all patients diagnosed with SRUS on endoscopy and confirmed by histopathologic examination at our center between January 2010 and June 2022. Data retrieved included demographic details, clinical features along with endoscopic, and histopathological findings of SRUS patients.

Results The study included 132 patients with SRUS with mean (± standard deviation) age of 45 (± 20.6) years and male to female ratio of 1.5:1. While most of the patients presented with a combination of symptoms, the most common clinical presentation was bleeding per rectum (82%). Rectal ulcers, either single or multiple, were the predominant findings on endoscopy followed by polypoidal lesions. Histology showed fibromuscular obliteration and crypt distortion in all patients. Biofeedback training, lifestyle changes, and sucralfate enema were successful in about 87% of the patients at the end of 6 weeks and about 76% at the end of 12 weeks with surgery for rectal prolapse being performed in two of our patients.

Conclusion SRUS presents with a myriad of symptoms and requires a high index of suspicion by the treating physician(s). While the most common presenting symptom in our study was rectal bleed; ulcers, either solitary or multiple, were the commonest endoscopy findings. Endoscopic findings along with histopathology confirm the diagnosis.

Keywords ► bleeding ► constipation ► mucorrhea ► rectal ulcer ► tenesmus

Introduction

Solitary rectal ulcer syndrome (SRUS) is a benign defecatory disorder characterized by incomplete evacuation, passage of blood and/or mucus per rectum, altered bowel habits, etc. The term solitary rectal ulcer is a misnomer as only about 40% of the patients have a true rectal ulcer, and the endoscopic findings vary from a single or multiple rectal ulcers of varying size to localized hyperemic rectal mucosa to polypoidal lesions.1 SRUS has an estimated prevalence of 1 in 100,000 per year and has been described across all age groups. It is seen mostly in the third decade in men and a decade later in women with men and women being affected equally, although few studies have shown a slight female

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preponderance.\textsuperscript{2,3} Pathogenesis of SRUS is thought to be from ischemia secondary to pressure of impacted stools and uncoordinated puborectalis contractions along with local trauma from repeated digital evacuation.\textsuperscript{4} The choice of treatment depends upon the severity of symptoms and presence or absence of rectal prolapse.

This study aimed to describe the clinical, endoscopic, and histopathologic findings of patients who underwent either colonoscopy or sigmoidoscopy and were ultimately diagnosed with SRUS at our center.

Materials and Methods

Medical and endoscopic records of all patients who underwent colonoscopy or sigmoidoscopy between January 2010 and June 2022 were retrieved from our hospital database. A total of 132 patients who presented with various symptomatology (\textsuperscript{►}Table 1) and were diagnosed both endoscopically and histologically as having SRUS were included in our study. Endoscopic features were further classified as ulcerative lesions, either solitary or multiple ulcers, polypoidal lesions, and focal rectal hyperemia. Typical histopathologic features included fibromuscular obliteration with crypt distortion and disarray of smooth muscle cells. The categorical variables were expressed as number with percentages and continuous variables as mean, median, and standard deviation. Given its retrospective nature, institutional ethical clearance was not deemed necessary for the study.

Briefly, biofeedback training in our study included the following. Patients were educated on how to strain effectively by using a propulsive force while bracing their abdominal musculature. They were educated on normal defecatory behavior and bowel habits including restricting the number of visits to the toilet for patients who made frequent defecatory attempts during the day or increasing the number of visits to the toilet for those patients with infrequent defecation. Biofeedback included reducing excessive straining with defecation by correcting abnormal pelvic floor behavior and by attempting to stop the aid of laxatives, enemas, and suppositories. The amount of time spent and posture in the toilet were also specified.

Results

A total of 24,905 patients underwent either sigmoidoscopy or colonoscopy during the study period extending between January 2010 and June 2022 and 132 patients were diagnosed as having SRUS (based on a combination of both endoscopic and histologic features) with a prevalence of around 0.0053 (132/24905). The mean age of our study group was 45 ± 20.6 (range: 4–89 years). There were 88 males and 44 females.

Bleed per rectum was the most common presentation, although many presented with a combination of symptoms.

Based on endoscopic appearance, lesions were classified as ulcerative, polypoidal, and localized erythematous/hyperemic rectal patch (\textsuperscript{►}Table 2 and \textsuperscript{Fig. 1}). Of note, a total of 14 (10.6%) patients had associated hemorrhoids.

All patients received biofeedback training as well as dietary modifications involving a high-fiber diet along with a bulk laxative, and avoidance of straining during defecation and anal digitation. A total of 116 patients also received sulfasalazine enema twice daily, while 10 patients were treated with mesalamine suppository twice daily and 6 patients were treated with budesonide enema once daily, and these patients were followed up at 6 and 12 weeks of therapy. Symptomatic improvement was observed in 87% at the end of 6 weeks and 76% at the end of 12 weeks in patients treated with sulfasalazine enema, while the response rate was around 61% and 46% at the end of 6 and 12 weeks in patients who received budesonide and mesalamine enemas, respectively. Repeat sigmoidoscopy was performed in only 23 of the treatment-responsive patients between 6 and 12 weeks of therapy, and it showed more than or equal to 50% reduction in the index lesions in all these patients with 8 patients showing complete endoscopic resolution of SRUS. Two patients with rectal prolapse and symptoms refractory to conservative treatment were referred for rectopexy procedure with complete resolution of symptoms at the end of 6 months.

<table>
<thead>
<tr>
<th>Presenting symptoms</th>
<th>Number of patients</th>
<th>Percentage of patients</th>
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<tbody>
<tr>
<td>Bleed per rectum</td>
<td>108</td>
<td>81.8</td>
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<tr>
<td>Abdominal pain</td>
<td>42</td>
<td>31.8</td>
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<tr>
<td>Digital evacuation</td>
<td>40</td>
<td>30.3</td>
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<td>Mucorrhea</td>
<td>37</td>
<td>28.03</td>
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<tr>
<td>Perianal discomfort</td>
<td>21</td>
<td>15.9</td>
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<tr>
<td>Anemia</td>
<td>15</td>
<td>11.4</td>
</tr>
<tr>
<td>Alternating bowel habits</td>
<td>15</td>
<td>11.4</td>
</tr>
<tr>
<td>Rectal prolapse</td>
<td>13</td>
<td>9.8</td>
</tr>
<tr>
<td>Tenesmus</td>
<td>8</td>
<td>6.1</td>
</tr>
<tr>
<td>Asymptomatic</td>
<td>3</td>
<td>2.3</td>
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</tbody>
</table>

Abbreviation: SRUS, solitary rectal ulcer syndrome.

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<tr>
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<th>Percentage of patients</th>
</tr>
</thead>
<tbody>
<tr>
<td>Bleed per rectum</td>
<td>42</td>
<td>91.2</td>
</tr>
<tr>
<td>Abdominal pain</td>
<td>30</td>
<td>31.8</td>
</tr>
<tr>
<td>Digital evacuation</td>
<td>20</td>
<td>20.8</td>
</tr>
<tr>
<td>Mucorrhea</td>
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<td>Perianal discomfort</td>
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<td>5.2</td>
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<tr>
<td>Anemia</td>
<td>5</td>
<td>5.2</td>
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<tr>
<td>Alternating bowel habits</td>
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<td>5.2</td>
</tr>
<tr>
<td>Rectal prolapse</td>
<td>3</td>
<td>3.1</td>
</tr>
<tr>
<td>Tenesmus</td>
<td>2</td>
<td>2.1</td>
</tr>
<tr>
<td>Asymptomatic</td>
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<td>1.1</td>
</tr>
</tbody>
</table>

Abbreviation: SRUS, solitary rectal ulcer syndrome.
Discussion

SRUS is a chronic disorder of defecation that can present with varied clinical and endoscopic findings. The incidence of SRUS in one study was estimated to be 1 in 1,00,000 per year; however, some authors have suggested that SRUS may not be as rare as reported in literature.\(^5\)–\(^7\) To our knowledge, this study enrolled one of the largest series of patients. The mean age of our study population was 45 years with a male to female ratio of 1.5:1. Although most of the studies have shown a slight female predominance, few case series concur with our findings of male preponderance.\(^7\)–\(^10\) Rectal bleed was the most common symptom noted in our study with as many as 15 patients presenting with anemia with 3 patients requiring repeat blood transfusions before presenting to our center. Other common symptoms include abdominal pain, digital evacuation, mucorrhea, and perianal discomfort that are in concurrence with other studies.\(^11,12\)

SRUS must be differentiated from other chronic and potentially fatal diseases such as inflammatory bowel disease, malignancy, chronic ischemic colitis, infectious colitis, and stercoral ulcers. Typical endoscopic features of SRUS include a solitary ulcer located over the anterior wall of the rectum at 5 to 10 cm from the anal verge.\(^5,13\) Ulcerative lesions were noted in 78% of our patient with solitary ulcer noted in only 31% of patients. Polypoidal lesions were noted in 18.2% that are often misdiagnosed as rectal malignancy by naïve endoscopists. The typical histological findings include fibromuscular obliteration of the lamina propria with splaying of muscularis mucosa, thickened mucosa, and glandular distortion.\(^14\)–\(^16\) The characteristic fibromuscular obliteration was seen in all our patients with a majority also showing crypt distortion and distortion of the mucosal glands. Other findings reported include surface ulcerations and hyperplastic crypts.\(^12\) Few studies have shown association of SRUS with rectal adenocarcinoma, although our study failed to show such an association.\(^17\)–\(^19\)

Treatment options for SRUS range from behavioral modification to topical treatment, biofeedback training, and surgery as there are no definitive treatment recommendations for SRUS and treatment depends on the severity of symptoms and presence or absence of underlying rectal prolapse. Behavioral modification with high-fiber diet, avoiding straining during defeation and anal digitation are effective in patients with mild-to-moderate symptoms with no rectal prolapse. Sucralfate enemas have shown good clinical response in some case series.\(^20,21\) In our own study, biofeedback training and lifestyle changes along with sucralfate enemas given as 2 g twice daily resulted in symptomatic improvement in as many as 87% (102/118) of patients at the end of 6 weeks and 76% at the end of 12 weeks. Most of these patients were, however, lost to follow-up after 12 weeks and hence long-term resolution of symptoms could not be ascertained in these patients. The response rate in patient who received either mesalamine or budesonide enemas was 61% and 46% at the end of 6 and 12 weeks, respectively. Surgical treatments such as ulcer excision, diversion, and rectopexy are reserved for those with refractory symptoms and full-thickness rectal prolapse. Two of our patients with full-thickness rectal prolapse and with refractory symptoms
despite conservative management were subjected to recto-
pexy with resolution of symptoms at the end of 6 months. Budesonide and sulfasalazine enemas were effective in only a small proportion of our patients. Novel experimental thera-
pies reported as isolated case reports include fibrin glue injection and endoscopic mucosectomy of SRUS; however, these need further validation through randomized control trials.\textsuperscript{22,23}

Although anorectal manometry was not performed in our study, anorectal physiology studies have shown that 25 to 82% of patients with SRUS may have dyssynergia with paradoxical anal contraction.\textsuperscript{24} Studies have confirmed that uncoordinated defection with excessive straining over time plays a key role in SRUS. Up to 82% of subjects exhibited dyssynergia along with prolonged balloon expulsion time. Also, SRUS patients exhibited rectal hypersensi-
tivity that in turn may lead to a persistent desire to defece and/or feeling of incomplete evacuation and excessive straining.\textsuperscript{25} Studies have found that SRUS patients have more frequent increase in anal pressure and paradoxical pubor-
ectalis contraction during straining.\textsuperscript{26}

Our study highlights that in addition to laxatives and enemas, gut-specific biofeedback training results in signifi-
cant improvement in the overall symptomatology of patients with SRUS, albeit in the short-term. Biofeedback is thought to improve symptoms by increased rectal mucosal blood flow and altering the efferent autonomic path-
ways to the gut.

The main strengths of our study were the large number of patients enrolled in it. Majority of our patients were followed up at the end of 6 and 12 weeks of therapy. The main limitations of our study were its retrospective design and that none of our patient underwent endoscopic ultrasound or anorectal manometry unlike other studies in literature. Also, majority of our patients were lost to follow-up after 12 weeks of therapy and hence, the long-term resolution of symptoms could not be ascertained in our study.

Conclusions
SRUS is a chronic defecatory disorder affecting the rectum. Patients present with a myriad of symptoms, and SRUS is a misnomer as the lesions are not always ulcerated. The diagnosis of SRUS is based on clinical, endoscopic, and histologic characteristics, and endoscopic appearance can mimic other potentially life-threatening diseases. Majority of patients benefit from biofeedback therapy and lifestyle changes including a high-fiber diet, avoidance of straining at stools, and anal digitation along with sucralfate retention enemas. Patients with rectal mucosal prolapse benefit from rectopecty.

Authors’ Contributors
The author contributed to conceptualization, design, def-
inition of intellectual content, literature search, clinical studies, experimental studies, data acquisition, data anal-
ysis, statistical analysis, manuscript preparation, editing, and review. The author is guarantor of this study.

The manuscript has been read and approved by all the authors, that the requirements for authorship as stated earlier in this document have been met, and that each author believes that the manuscript represents honest work, if that information is not provided in another form.

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Conflict of Interest
None declared.

References