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Solitary Rectal Ulcer Syndrome: Exploring Possible Management Options

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Solitary rectal ulcer syndrome (SRUS) is a rare condition with various causes that results in ischemic injury. The aim of this study was to assess the clinical findings, diagnosis, and outcomes of treatment in patients with SRUS. Between 1992 and 2006, a retrospective review was undertaken for all patients diagnosed with SRUS. Fifty-eight patients were diagnosed with SRUS. Among patients with paradoxic rectal spasm (PRS), lesions disappeared in 1 of 3 given applied biofeedback treatment, and in 2 of 4 injected with Botulinum toxin (Botox™). Twenty-three patients underwent appropriate surgical treatment. Overall, postoperative improvement was seen in 18 patients (78.2%). In conclusion, every patient with SRUS must be assessed for causative disease. Treatment should include conservative approaches such as Botox injection; in patients with pelvic floor disorders, surgical treatment should be considered.

Key words: Solitary rectal ulcer syndrome – SRUS – STARR – Botulinum toxin – Paradoxic rectal spasm

Solitary rectal ulcer syndrome (SRUS) is a rectal disorder associated with reduced blood perfusion of the rectal mucosa, leading to local ischemia and ulceration that can present with pelvic chronic pain, mucous discharge, rectal bleeding, straining during defecation, and a sense of incomplete evacuation.[1] The disorder was first reported in 1829 by Cruveilhier,[2] but the distinctive histopathologic characteristics were defined in 1969 by Madigan and Morson.[3]

SRUS diagnosis is delayed in many cases because of its rarity, nonspecific signs, and symptoms and various causes. However, chronic constipation, strenuous defecation, rectal bleeding and mucous secretions from the rectum, and nonspecific pelvic pain are the major complaints encountered by physicians.[1,4,5] Diagnosis can be performed by clinical examination and confirmed by endoscopy with biopsies that show fibromuscular obliteration of the lamina propria to exclude malignant lesions.[1] Endoscopic findings vary

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Table 1 Clinical features of the 58 patients[a]

Symptoms	Patients, n (%)
Rectal bleeding	42 (77.7)
Constipation	20 (37)
Mucous discharge	5 (9.2)
Digitation	6 (11.1)
Anal pain	13 (24)
Tenesmus	4 (7.4)
Anemia	1 (1.8)

[a]Each patient may have had more than 1 symptom.

and include mucosal ulcerations, polypoid and mass lesions, and erythema.

Anorectal manometry and defecography can be useful for recognizing a concomitant internal rectal prolapse or pelvic dyssynergia.[6,7]

Different treatment strategies ranging from conservative management to a variety of surgical procedures have been advocated for SRUS. The results of conservative treatments (diet, drugs, biofeedback)[8,9] and/or surgery[10] are controversial. To date, no consensus has been reached regarding optimal treatment for SRUS.

The aim of the present study was to assess the clinical findings, treatments, and outcomes of patients with SRUS.

Patients and Methods

We retrospectively reviewed the records of 58 patients diagnosed with SRUS and treated between 1992 and 2006 in the Department of General Surgery in Istanbul Medical Faculty of Istanbul University, Turkey. Data analyzed included age, gender, clinical presentation, past surgical history, preoperative workup, operative procedure, complications, and outcomes. A successful symptomatic outcome was defined as a subjective report by the patient that symptoms had substantially resolved. Healing was assessed with endoscopic evaluation. Treatment failure was defined as no changes in and no lessening of symptoms.

Results

During the study period, 58 patients (28 male and 30 female patients) were diagnosed with SRUS. Patient age ranged from 17 to 77 years (mean, 39 years). Median follow-up was 72 months (range, 48–96 months). Patient follow-up consisted of clinical visits, endoscopic examinations, and/or telephone conversations.

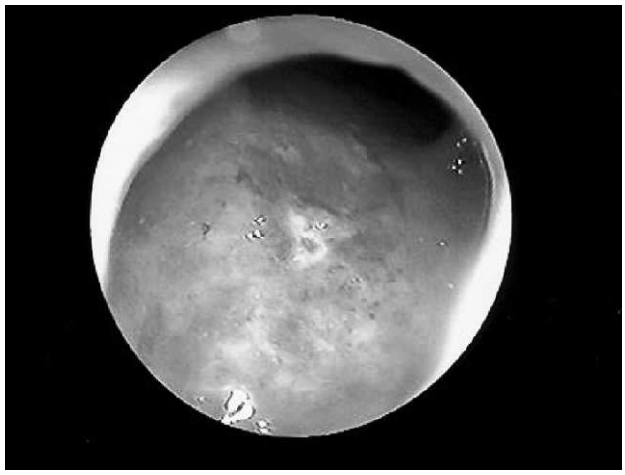

Fig. 1 Endoscopic features of SRUS as small cicatricial lesions.

Symptoms

The mean interval between onset of symptoms (Table 1) and final diagnosis of SRUS was 8.5 years (range, 1 month–34 years). Before receiving care at our institution, 6 patients (11%) had undergone anorectal surgery for anal fissure (n = 1), hemorrhoidal disease (n = 3), and rectal prolapse (n = 2), outcomes of which were inconclusive.

Diagnostic measurements

Endoscopic features of SRUS varied from small cicatricial lesions to wide tumor-like granulating tissues (Figs. 1 and 2). The mean distance of the ulcer from anal verge was 6.2 cm (range, 2–13 cm), and mean ulcer diameter was 2.8 cm. Circumferential involvement of the rectal wall was detected in 3 cases. Defecography was performed in 29 patients (Table 2). Mean anorectal angles were 100 $\pm$ 10 degrees at rest and 140 $\pm$ 38 degrees during defecation. Paradoxic rectal spasm (PRS) at the level of the ulcer area was the most prominent finding in 16 of 29 patients (68.9%) who underwent defecography (Fig. 3). Other defecographic findings included intussusception of the proximal rectum through the contracted segment in 7 patients (24.2%), internal prolapse in 6 patients (20.7%), and total rectal prolapse (20.7%) and pelvic descensus in 3 (10.3%).

Anal manometry investigation was carried out in 6 patients; mean resting tone was 58 $\pm$ 12 mmHg, and mean maximum squeeze pressure was 101 $\pm$ 20 mmHg. First sensation and resting and squeeze pressures were low in 3 patients.

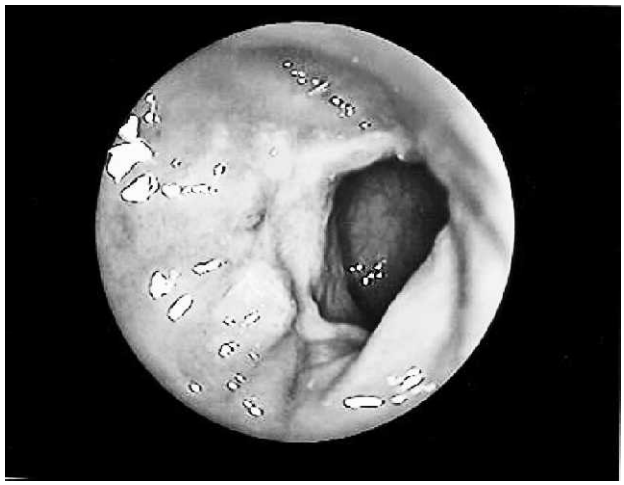

Fig. 2 Endoscopic features of SRUS showing wide tumor-like granulating tissues.

Fig. 3 Defecography of patients with paradoxic rectal spasm.

Treatment and outcomes

Types of treatment given to patients with SRUS are given in Table 3. Ten patients received medical treatment consisting of a high-fiber diet, in combination with stool softeners and bulking laxatives. Spontaneous healing occurred in 2 of 10 patients (20%). Eighteen patients received anti-inflammatory sulfasalazine enemas (mesalazine 4 gr enema, once daily), in addition to medical treatment. Lesions improved or healed completely in 5 of 18 patients (27.7%).

Biofeedback treatment undertaken using the manometry method (Biofeedback Monitor, Biosearch Medical Products Inc, Somerville, New Jersey) was applied to 3 patients with PRS. It was found to be effective in 1 patient and was discontinued thereafter when the ulcer had healed. However, 2 patients did not report ulcer healing or relief of complaints.

For the first time, Botulinum toxin (Botox™, 100 IU/flacon, Abdi Ibrahim, Istanbul, Turkey) injection to the area of PRS was applied to 4 patients in whom PRS had been detected with defecography; patients' complaints were relieved, and their condition returned to normal even though lesions were not regressed in 2 patients. Complete healing occurred in 2 patients given Botox injection.

Surgical treatment was provided in 23 cases with no response to medical treatment or with severe symptoms. Anterior resection, low anterior resection, rectopexy, stapled transanal local excision (STARR) was performed on the basis of defecography findings. Results of these procedures are shown in Table 3.

Overall postoperative symptomatic improvement was seen following 18 (78.2%) of 23 procedures performed in patients with SRUS. Patients with PRS and complete internal prolapse were treated with STARR. Healing was detected in all patients. Four patients with PRS were treated with Botox™, and 3 of those were treated with biofeedback therapy (Table 4). Two of 4 patients injected with Botox™ and 1 of 3 patients for whom biofeedback treatment was applied were completely healed.

Table 2 Results of defecography in patients (n = 29)[a]

Radiologic abnormalities	Patient number (n = 29)	Percentage of patients
Paradoxic rectal spasm	16	55
Intussusception	10	34
Internal prolapse	6	20
Total rectal prolapse	6	20
Pelvic floor descensus	3	10
Sigmoidocel	1	3

[a]Patients may have had more than 1 abnormality.

Discussion

SRUS is a benign, uncommon, often underdiagnosed condition that is categorized as a functional evacuatory disorder rather than as an independent entity.[1,11] The pathogenesis of SRUS has been clearly identified as chronic mucosal trauma and ischemia.[3,12–14] In our series, a slightly higher proportion of female patients were identified with a wide age range (17–77 years), as occurred in 3 other studies.[8,15,16] In our series, the triad of rectal bleeding, constipation, and anal pain was the most common

Table 3 Treatments including surgical procedures in patients with SRUS

Type of treatment	Number of patients	Healing	Postoperative complications (number of patients)
Medical treatment	10	2	–
Medical treatment and sulphasalazine enema	18	5	–
Biofeedback treatment	3	1	–
Botox	4	2	–
Low anterior resection	10	7	Sexual dysfunction (2) Incontinence (2) Bleeding (2)
STARR	6	6	Bleeding (2)
Rectopexy	6	2	Impotence (1) Graft migration (1) Incontinence (1)
Anterior resection	1	1	

finding. Rectal bleeding and constipation were reported as the most common presentation in other series.[8,15,16]

Retrospective studies on the treatment of SRUS have been published.[5,9,17] Conservative treatments may improve symptoms but are unlikely to achieve endoscopic and histologic normality.

Behavioral techniques may help patients with SRUS and may be part of the treatment regimen. Dietary changes to a high-fiber diet to avoid straining have appeared successful in a number of cases.[11,15,18] In our study, 10 patients received medical treatment consisting of a high-fiber diet, in combination with stool softeners and bulking laxatives, and 20% of patients showed resolution of the ulcer with only medical treatment; in 2 series, improved symptoms were reported in 19% to 70% of those who had received bulking agents or dietary fiber.[5,15]

In our study, local treatment with sulfasalazine enemas, in addition to medical treatment, was applied to 18 patients; symptoms improved or healed in 5 patients. Previously, it had been reported that local treatment with steroids and sulfasalazine was not effective in all patients,[1] and their long-term benefits are still unclear.

Biofeedback, which usually is associated with correction of pelvic floor defecatory behavior, can precede surgery.[1,8,11,20] In the series of Vaizey *et al*,[1] none of the patients treated with biofeedback therapy showed complete healing of the ulceration, even though they had improved symptoms of SRUS. Some studies reported better results with 36% healing,[21] which were similar to those obtained after biofeedback therapy for obstructed defecation and for PRS at defecography.[9,22] In our series, 1 of 3 patients treated with biofeedback therapy showed healing of the ulceration.

Table 4 Treatment options for patients with PRS

Type of treatment	Number of patients	Healing
Biofeedback	3	1
Botulinum toxin	4	2
STARR	6	6

In our study, manometric findings provided little additional information because we examined a limited number of patients. Six patients had findings of normal resting pressure and low squeeze pressure. Contrary results were described in other studies.[5,23]

In a high percentage of patients with SRUS, paradoxic contraction of the puborectalis muscle during defecation is reported.[18,24] In our study, 16 of 29 patients who underwent defecography presented with paradoxic contraction of the puborectalis muscle.

We applied Botox injection into the paradoxic contraction area of the puborectalis muscle in 4 patients with PRS. Symptoms were improved by Botox injection in all patients, and complete histologic and macroscopic healing of the ulcer occurred in 2 of 4 patients.

Since its introduction in the 1980s for the treatment of strabismus and blepharospasm,[25] Botox has been used increasingly in the interventional treatment of several other disorders characterized by excessive or inappropriate muscle contractions. Use of this pluripotential agent has been extended to a plethora of conditions, including (1) inappropriate contraction in most sphincters of the body, such as those associated with esophageal achalasia and gastroesophageal reflux disease,[26] (2) chronic anal fissure,[27] (3) genitourinary disorders, such as overactive and neurogenic bladder, and (4) prostatic and pelvic floor disorders.[28] In addition, Botox is being investigated for its use in control of pain and in

management of myofascial pain syndrome[29] and postcholecystectomy biliary pain.[30] In our study, for the first time, Botox was applied into the paradoxic contraction area of the puborectalis muscle in 4 patients with PRS and perianal pain. Even though the sample size was small, our preliminary results are promising for Botox intervention in the treatment of SRUS. Botox injections offer several advantages over medical and surgical therapies in the management of intractable or chronic disease. Systemic pharmacologic effects are rare, and permanent destruction of tissue does not occur. Graded degrees of relaxation may be achieved by varying the dose injected; most adverse effects are transient. Finally, patient acceptance is high.

The mechanism of healing of the puborectal muscle contraction could be explained by stating that application of Botox released the blockage in glyceryl trinitrate bioactivation in smooth muscle cells and suppressed basal continuous sympathetic activity, causing modulation of contraction that was assumed to be responsible for healing of the ulcer and ceasing of anorectal pain, as explained for healing of chronic anal fissure.[27]

Outcomes of surgical treatment for SRUS are inconclusive because studies have included small numbers of patients, various procedures were applied, and the mean follow-up period has not been fully considered.

Previously, it was reported that SRUS patients were treated surgically by rectopexy (n = 26), rectal mucosectomy (n = 4), segmental colon resection (n = 2), and local excision and colostomy (n = 1 patient each from Turkey).[17] Total regression and healing of the ulcer occurred in 32 of 34 patients. Partial regression of symptoms was noted in 2 patients who underwent rectopexy and rectal mucosectomy, but complete healing was not achieved.[17] We have not performed local excision because of reported data indicating that local excision was unsuccessful.[1,8,15,31] On the other hand, it has been reported that three-quadrant mucosal excision was performed successfully for three-quadrant hemorrhoidal prolapse with SRUS.[32] In this study, the most common resection performed was low anterior resection (n = 10) caused by large rectal ulcer or intussusception or rectocele.

It has been stated that low anterior resection as a primary procedure has yielded successful outcomes.[15,16] In our series, anterior resection was performed in the only patient in whom healing occurred. Anterior resection was carried out as a primary procedure in 2 patients in the study of Sitzler *et al.*[16] However, anterior resection was proffered after failure of rectopexy or Delorme's operation in other studies.[3,15,19] Postoperative sexual dysfunction and bleeding are possible after colorectal surgery.[17] In our study, 3 patients presented with sexual dysfunction and 4 patients presented with bleeding.

The STARR operation was performed in 6 patients with complete internal rectal prolapsus and complete healing was achieved in all patients. STARR has been demonstrated to successfully cure internal rectal prolapsus with rectocele[33] and SRUS,[22] as well as prolapsed hemorrhoids.[22]

The results of this study show that each patient with SRUS has to be assessed individually. Patients of wide age ranges are diagnosed with SRUS. Defecography has to be performed for the diagnosis of SRUS associated with pelvic floor disorder for selection of an appropriate therapeutic approach. Patients without pelvic floor disorders may be managed through a conservative approach. Botox intervention seems to be promising in SRUS patients with PRS and perianal pain in the absence of rectal prolapse. If complete rectal prolapse occurs, STARR seems to be the most appropriate technique. Rectopexy can be performed in incomplete prolapse; anterior and low anterior resections are preferable techniques in cases of SRUS associated with pelvic floor disorders.

Conclusion

Our retrospective data imply that SRUS develops secondary to functional anorectal evacuatory disorders rather than through an independent pathogenesis. Furthermore, results confirm the positive role of surgical treatment in pelvic floor disorders and Botox™ injection in PRS. However, a prospective randomized trial with larger sample sizes will help to elucidate the therapeutic algorithm of SRUS.

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