Original Article

Solitary Rectal Ulcer Syndrome: A Single-center Case Series

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ABSTRACT

Background/Aim: Solitary rectal ulcer syndrome (SRUS) is a benign, chronic defecation disorder with varied presentations. The aim of this study is to summarize the clinical features, endoscopic findings, histological appearance, and treatment strategies associated with SRUS. **Patients and Methods:** This is a retrospective study of all patients diagnosed with SRUS at the King Faisal Specialist Hospital and Research Centre in Riyadh from January 2003 to December 2013. Cases were identified using the Department of Pathology database. Data were obtained from medical records that included clinical manifestation, endoscopic findings, and histopathological features. **Results:** Twenty patients were identified. The mean age was 42.5 years (±18.5) and 55% were females. Most of the patients presented with bleeding per rectum (85%), constipation (75%), and straining (50%), with a mean symptom duration of 26.7 months. The most common associated factors identified were constipation (75%), history of rectal surgery (25%), digital rectal manipulation (20%), and rectal prolapse (20%). Endoscopic findings included a single ulcer (50%) and multiple ulcers (30%); 55% had a polypoidal appearance. On histopathology, there was surface ulceration (95%), fibrosis of the lamina propria (60%), distorted architecture (55%), and muscle hypertrophy with increased mucin production (50%). Patients were treated conservatively and none required surgery. **Conclusion:** SRUS is a rare disorder with variable clinical presentations. Stool softeners, a high fiber diet in addition to topical mesalamine, and biofeedback proved to be effective in this patient population.

Key Words: Case series, Saudi Arabia, solitary rectal ulcer

Received: 24.04.2016, Accepted: 17.06.2016
How to cite this article: AlGhulayqah AI, Abu-Farhaneh EH, AlSohaibani FI, Almadi MA, AlMana HM. Solitary rectal ulcer syndrome: A single-center case series. Saudi J Gastroenterol 2016;22:456-60.

Solitary rectal ulcer syndrome (SRUS) is an uncommon chronic benign disorder mostly seen in middle-aged and young adults and less often in children, with an incidence of 1 in 100,000 adults, and can be misdiagnosed in up to 26% of patients.[1] The median age of presentation is 48 years, equally affecting both the sexes. It is characterized by a single or multiple ulcers in the rectum with specific histological changes. Individuals present with recurring anal or rectal discomfort, rectal bleeding, straining, mucous discharge, constipation, and the use of digital maneuvers to defecate.[2] Several risk factors have been implicated in the pathogenesis of SRUS including constipation, direct digital trauma, or surgery.[3] The diagnosis is based on the presence of symptoms with a combination of endoscopic and histological features. The aim of this study is to describe a single center experience with regard to the clinical presentation, diagnosis, and treatment of SRUS in Saudi Arabia.

Access this article online

Quick Response Code:

Website: www.saudijgastro.com

DOI: 10.4103/1319-3767.195555

PATIENTS AND METHODS

This is a retrospective study of all patients diagnosed with SRUS at the King Faisal Specialist Hospital and Research Centre in Riyadh. Patients were identified using the Department of Pathology database, utilizing pathology reports, paper, and electronic charts of patients labeled as having SRUS were retrieved. Data retrieved included demographics, comorbidities, presenting symptoms, endoscopic findings, histopathologic reports,

and management strategies. We included all adult patients aged 14 years or older who were diagnosed in our hospital from January 2003 to December 2013. We excluded any patients with unconfirmed histopathology or those with missing data. All the specimens were reviewed by a single pathologist (HM) to confirm the diagnosis, and were examined for specific histopathological characteristics. The statistical analysis of the data was done using the software package SAS, version 9.3 (SAS Institute Inc., Cary, NC, USA). Descriptive statistics for continuous variables are reported as means and standard deviations (SD) whereas categorical variables are summarized as frequencies and percentages. Continuous variables were compared using the Student's *t*-test, whereas categorical variables were compared by the Chi-square test. The level of statistical significance was set at a *P* value of < 0.05.

RESULTS

Twenty patients met our inclusion criteria; 9 patients were males (45%). The mean age was 42.5 years (95% confidence interval (CI); 33.8–51.2 years), with no statistical difference between males and females. The mean body mass index (BMI) for males was 23 (95% CI; 17.3–28.7) and for females was 31.7 (95% CI; 26.2–37.2), with a mean of 27.8 for both sexes (95% CI; 23.7–31.9) [Table 1a and b]. Thirty percent of the patients had diabetes mellitus. Other significant comorbidities included hypertension (25%), chronic liver disease (15%), chronic renal failure (10%), and colonic diverticulosis (10%).

Most of the patients presented with bleeding per rectum (85%), followed by constipation (75%), straining (50%), and abdominal pain (30%). Tenesmus was found in 30%, passing of mucus in 25%, and perianal pain in 15% [Figure 1]. Although 75% of the patients presented with chronic constipation, 15% had mild chronic diarrhea. Only 1 patient was asymptomatic as the condition was discovered incidentally during a screening colonoscopy. The mean duration for the symptoms was 24.6 months with no significant difference between the sexes ($P = 0.28$). Associated factors were significant for constipation (75%); previous surgery (25%), such as lateral internal sphincterotomy, sigmoidectomy, and hemorrhoidectomy; digital rectal manipulation (20%); and rectal prolapse (20%).

Endoscopic appearances were polypoidal lesions in 55%, a single ulcer in 50%, whereas 30% had multiple ulcers. Associated findings were hyperemic mucosa surrounding the lesion in 30%, erosions in 25% whereas a stricture was seen in 1 patient [Figure 2a-c]. On histopathology, surface ulcerations (95%), fibrosis of the lamina propria (60%), distorted architecture (55%), muscle hypertrophy with increased mucin production (50%), and serrated crypts were seen in 15% [Figure 3a-c].

All the patients were treated conservatively. Most received stool softeners, a high-fiber diet, or laxatives, however, 4 patients were treated with mesalamine either oral or suppository. Two patients received additional budesonide foam enema. Three patients with refractory symptoms were referred for biofeedback training.

Table 1a: Demographics of 20 patients

Variable	*N*	Mean	SD	Lower 95% CL for Mean	Upper 95% CL for Mean	Minimum	Maximum
Age	20	42.5	18.5	33.8	51.2	14.0	72.0
BMI	20	27.8	8.8	23.7	31.9	15.4	51.0
Symptoms Duration*	19	24.6	29.6	10.4	38.9	1.0	120.0

*One of the patients was asymptomatic

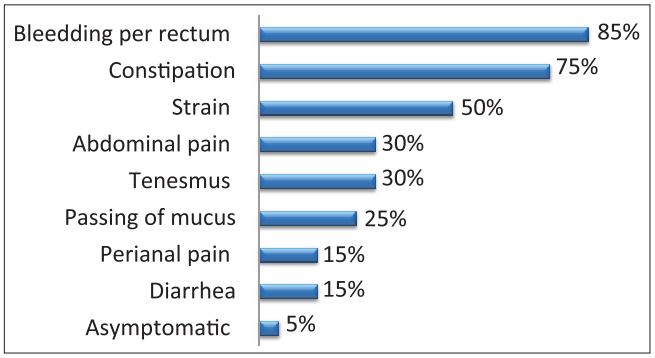

Figure 1: Presenting symptoms

Table 1b: Demographics of 20 patients

Gender	*N*	Variable	Mean	SD	Lower 95% CL for Mean	Upper 95% CL for Mean	Minimum	Maximum
Female	11	Age (year)	49.4	15.4	39.0	59.7	16.0	72.0
		BMI	31.7	8.1	26.2	37.2	22.0	51.0
		Symptoms Duration (months)	31.6	38.8	3.8	59.4	1.0	120.0
Male	9	Age (year)	34.1	19.3	19.3	48.9	14.0	67.0
		BMI	23.0	7.4	17.3	28.7	15.4	34.1
		Symptoms Duration (months)	16.9	12.3	7.4	26.4	2.0	36.0

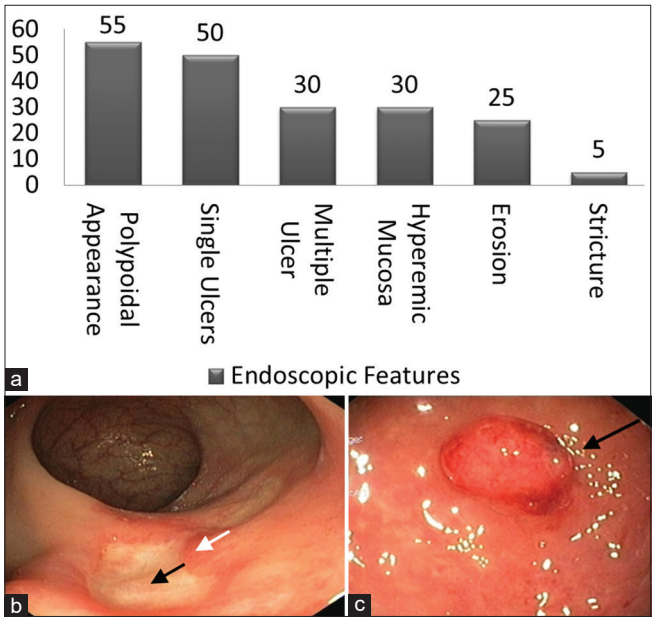

Figure 2: (a) Endoscopic appearance; (b) endoscopic appearance of a single ulcer (black arrow) with surrounding erythema (white arrow) seen in the rectum; (c) Endoscopic appearance of a polypoidal lesion (black arrow) in the rectum that was proven to be a solitary rectal ulcer on histopathology

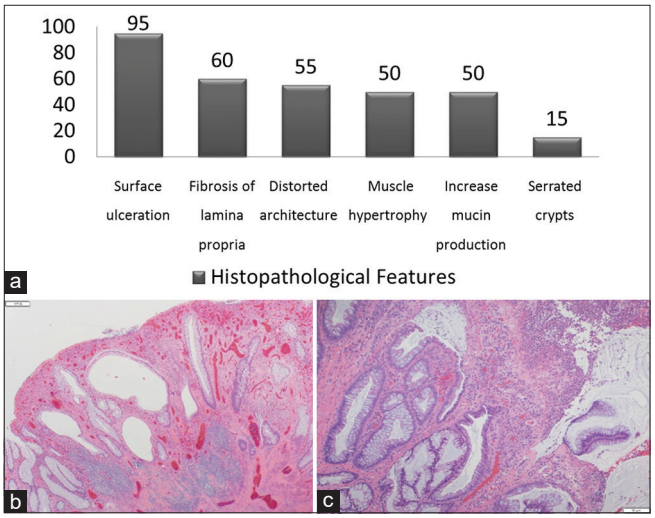

Figure 3: (a) Histopathological appearance; (b) Histological appearance of a solitary rectal ulcer demonstrating glands that are mucin-dense and acute inflammatory cells infiltrating the muscle strands between the glands; (c) Histological appearance of a solitary rectal ulcer demonstrating surface ulceration with irregularly dilated glands and the lamina propria depicting dense inflammation and crypt hyperplasia

DISCUSSION

SRUS is an uncommon benign and chronic disorder of defecation. It is a well-recognized entity in adults. It was first described by Cruveilhier in 1829 as unusual rectal ulcer.[4] In 1969, clinical manifestation and histopathological features were described.[5] The pathogenesis of SRUS is incompletely understood. However, a common observation in a number of reports is rectal prolapse and paradoxical contraction of the puborectalis muscle, which can result from rectal trauma or chronic constipation as well as excessive straining that generates a high intrarectal pressure, which in part pushes the anterior rectal mucosa onto the contracting puborectalis muscle, resulting in pressure necrosis of the mucosa, congestion, edema and ulceration.[6,7] This study, to the best of our knowledge, is the first retrospective study of SRUS in the kingdom of Saudi Arabia, which was conducted in a tertiary care facility. In this study, most of the cases were from the central region (65%) probably due to the location of the institution. Our study showed a slight female predominance (55%); however, other studies have shown a male predominance.[8,9] The most common associated factor was constipation which was found in three-fourth of our patients. A limitation of the study is that we could not validate nor quantify constipation in our study population due to the retrospective nature of the study. One-fourth of the patients had surgery in the rectal area before having the symptoms; the surgeries conducted included sigmoidectomy, lateral internal sphincterotomy, ileocecal resection, and hemorrhoidectomy. Another association was rectal prolapse, which can lead to congestion and poor blood flow, edema, and ischemia resulting in ulceration. Self-digitation was found in 20%, which can contribute to rectal injury and ulceration because it is attempted by the patients to reduce rectal prolapse or to evacuate impacted stools; this has been reported in up to 28% in other studies.[10]

The most common symptoms were bleeding per rectum and constipation. In other series, abdominal pain was as common as rectal bleeding, however, in our study, only 30% had this presentation.[8] Some patients had an unusual presentation of diarrhea rather than constipation, accounting for 15% of our patients; this was reported in 22% of patients in another study.[11] Three patients had alternating bowel habits between diarrhea and constipation. Bleeding per rectum might be a confounder associated with constipation rather than a manifestation of SRUS per se, however, discerning that was not possible. SRUS can be mistaken with other conditions such as inflammatory bowel disease (IBD), adenomatous polyps, malignancy, or a nonspecific ulcer. In our study, 2 patients were initially diagnosed as IBD upon clinical symptoms and colonoscopy features, in which treatment was started for them; later, biopsy showed features of SRUS rather than IBD. SRUS might be a misnomer because patients can present with lesions that are neither solitary nor ulcerated and not necessarily restricted to the rectum.[2] In our series, polypoidal lesions on endoscopy (55%) were more frequent than single ulcers (50%). Other studies found polyps in approximately 25% of patients; 30% of the patients can have multiple ulcers.[3,9,12] Most of the

lesions were located 4–15 cm from the anal verge, with an ulcer size ranging from 0.3–3.2 cm.[1,3,12] It is not clear what factors are associated with the variable phenotypic features associated with SRUS nor the number of lesions that are found on endoscopic evaluation. Our endoscopists were able to diagnose SRUS grossly with its variable appearance in 50% of the cases whereas the rest were only evident after histological examination. Three of our patients had a defecography performed, and 1 demonstrated a mild degree of intussusception at the distal rectum and rectal prolapse with strain. Only 1 patient underwent anorectal manometry and a barium enema, which were both normal.

Histopathological examination is considered to be the cornerstone for diagnosing SRUS. Although the use of these characteristics vary between studies, they have a reasonable reproducibility. In our series, surface ulceration, lamina propria fibrosis, and distorted architecture were the most common findings with a frequency of 95, 60, and 55%, respectively. Furthermore, 50% had muscle hypertrophy and increased mucin production; others have reported crypts distortion and surface serration in all patients.[8]

All our patients were managed without the need for surgical intervention. Stool softeners and high-fiber diet were used in all patients except for those who presented with diarrhea. Avoidance of any rectal trauma or digital manipulation was advised for all the patients. In literature, the response rate to bulking agents and dietary fiber was reported in the range 19–70%.[1,2] In 21 patients treated with high-fiber diet and habit training to reduce straining, 14 patients reported symptomatic improvement, with sigmoidoscopic evidence of ulcer healing in 8.[13] Four of our patients were given additional treatment with mesalamine suppository and 2 received budesonide foam, especially in patients where IBD was suspected or in patients who had ulcerations with surrounding erythema. The majority of the patients (85%) showed a dramatic response to conservative therapy, with some of them recovering completely; however, others (15%) showed a mild response even with different modalities of topical and systemic therapies. Three patients were referred for biofeedback after they failed medical therapy with some improvement but not complete recovery. Unfortunately, because of the retrospective nature of the data and the small sample size, we could not perform any meaningful analyses that would differentiate those who had a clinical and endoscopic response from those who did not. In patients presenting with diarrhea or with diarrhea and constipation alternatively, mesalamine and budesonide foam showed significant improvement of symptoms. Topical treatments, including sucralfate, salicylate, corticosteroids sulfasalazine, and topical fibrin sealant, have been reported to be effective with varied responses and improvement of the symptoms.[14] In a small study, among 6 patients who were treated with sucralfate enemas (2 g twice daily for 3–6 weeks), symptomatic improvement and microscopic healing occurred in all although histological changes persisted.[15] Surgery remains an option for patients not responding to conservative measures and biofeedback. Surgical treatment includes excision of the ulcer and treatment of internal or overt rectal prolapse and rarely defunctioning colostomy. In a series of 66 adult patients with SRUS, 49 patients opted for rectopexy, 9 Delorme's, 4 primary colostomy, 2 restorative anterior resection, and 2 postanal repair division of puborectalis.[14,16]

A limitation of the study is that the number of cases included is relatively small and are from a single center, thus limiting its generalizability; however, at the same time, the study reflected the infrequent nature of the disease and its possible under-appreciation by practicing endoscopists. Nonetheless, this study describes in detail the associated factors as well as the response, and lack thereof, to various interventions in this uncommon disease.

CONCLUSION

SRUS is an uncommon benign defecation disorder with variable clinical presentation, mainly bleeding per rectum. Endoscopic features are variable and conservative management is successful in a majority of patients in our series.

Financial support and sponsorship

The authors extend their sincere appreciation to the Deanship of Scientific Research at King Saud University for its funding of this research through the Research Group Project number RGP-279.

Conflicts of interest

There are no conflicts of interest.

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