



Research Article

Prevalence and characteristics of the Solitary Rectal Ulcer Syndrome among a Cohort of Iraqi Patients with Lower Gastrointestinal Bleeding

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ABSTRACT

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Background: Solitary rectal ulcer syndrome (SRUS) is a benign, uncommon rectal disorder characterized by combination of symptoms, clinical findings, and histological abnormalities. It has wide range of presentations and variable endoscopic findings.

Objectives: to study the prevalence of SRUS in patients presenting with lower gastrointestinal bleeding and to further evaluate this syndrome in the affected patients.

Subjects and Methods: A retro-prospective descriptive study conducted in Basrah Gastroenterology and Hepatology Hospital involved revision of 350 colonoscopic reports performed for the period from January 2022–June 2023 for patients presented with bleeding per rectum. Fifteen patients fulfilled the diagnostic criteria of SRUS were reviewed and followed up.

Results: the prevalence rate was 4.28%; nine male (60%), seven female (40%), mean age (22 ±9.3) years (12-45years). Mean duration of symptoms until diagnosis (8.22 ±4) weeks (3-17) weeks, females had shorter diagnosis time compared to males (5.5±2.3) (8.2±5.3) weeks respectively. The most frequent single associated symptom with the bleeding per rectum was constipation (96.3%), 20% had multiple associated symptoms.

On endoscopic examination:11 (73.4%) had ulcerated lesions,4 (26.6%) non ulcer lesions [3(20%) had polypoidal lesion,1(6.6%) had only hyperemic mucosa] Majority of rectal lesions located anteriorly 13 (86.6%). At follow up; symptoms improved in 7 (46.6%), endoscopic improvement in 8 (53.3%) and histological improvement in 11(73.4%), 2(13.3%) underwent endoscopic treatment and 3(20%) ended with surgery.

Conclusions: SRUS is chronic, benign disorder related to straining or abnormal defecation. It has variable clinical presentations and variable endoscopic findings rather than solitary ulcer as the name imply with different therapeutic options are available.

Introduction

Solitary rectal ulcer syndrome (SRUS) is an uncommon type of rectal disease that is defined by a mix of symptoms, clinical findings, and histological abnormalities. Patients may exhibit lower gastrointestinal hemorrhage, mucous passing, straining during defecation, and a feeling of incomplete evacuation (1).

The name of the syndrome is misleading, since patients can often present with lesions that are neither solitary nor ulcerated. The lesions are located in the anterior rectal wall within 10 cm of the anal verge in the majority of patients (2).

Endoscopic and radiologic findings can vary and include mucosal ulcerations, polypoidal or mass-like lesions that mimic rectal cancer, or just hyperemic mucosa (3). Because of this, misinterpretation is

frequent; in one study, up to 26% of patients had received an inaccurate initial diagnosis, which was most frequently a nonspecific ulcer, inflammatory bowel disease, or adenomatous changes (4).

Symptoms are variable or may be absent. In an interpretive series, the most common symptoms were rectal bleeding (56%), straining (28%), and pelvic fullness (23%) while mucous discharge, incontinence, tenesmus and pain were less frequently described (5).

The pathogenesis of the solitary rectal ulcer is incompletely understood, however; a number of factors appeared to have a causative role. It is possible that different etiologies may contribute to the development of the final lesion.

A common observation in a number of reports is rectal prolapse and paradoxical contraction of the puborectalis muscle, which can result in rectal trauma by two different mechanisms (6):

1. The pressures produced by the rectum during defecation force the prolapsed rectal mucosa downward. The opposing force of the puborectalis muscle's paradoxical contraction can create high pressures inside the rectum and cause mucosal ischemia, which makes ulceration more likely.

2. The contraction of the puborectalis muscle results in shear stresses on the rectal mucosa.

However, not all SRUS patients have excessive puborectalis contraction. Additionally, it is unclear whether prolapse leads to ulceration or if they are both different symptoms of the same disease process. Research in which surgical repair of rectal prolapse had no appreciable effect on patients' symptoms supports the concept that prolapse is an associated condition rather than a causal one (7).

In comparison to control groups, patients with solitary rectal ulcer syndrome more commonly experienced paradoxical puborectalis contraction, prolapse of the inner circular smooth muscle of the rectum, and rising anal pressure at strain. They also experienced less complete rectal emptying. Mean resting and compressing anal pressures were substantially higher in these patients compared to those with overt rectal prolapse (6). Direct digital trauma has also been implicated, since many patients have a history of constipation and report attempts at manual disimpaction. However, a number of lesions have been described that were beyond the reach of an inserted finger (8).

A possible hormonal cause has also been proposed. A case report documented a woman with solitary rectal ulcer syndrome that resolved during two pregnancies but recurred when she was not pregnant (9).

Histologically, the pathognomonic triad of fibrous obliteration of lamina propria, Disorientation of muscularis mucosa, extension of muscle fiber into the lamina propria must be present to confirm the diagnosis (10).

Various treatment strategies have been advocated, ranging from conservative management, medical therapy with sucralfate enema, sulfasalazine enema or simply with xylocaine gel, endoscopic therapy with argon plasma coagulation (APC) to a variety of surgical procedures, but the optimal treatment for the condition remained unclear (5).

This study aims to study the prevalence of the solitary rectal ulcer syndrome in patients presenting with bleeding per rectum and referred

to the endoscopy unit in this hospital for sigmoidoscopic/ colonoscopic examinations and to study the characteristics findings of this syndrome in the affected patients.

Subjects and Methods

This study was a retro-prospective descriptive, single center study conducted in the Basrah Gastroenterology and Hepatology hospital /southern Iraq. In this study lower endoscopic reports for patients presented with lower gastrointestinal bleeding had been reviewed.

During the period of the study 1578 lower endoscopy (sigmoidoscopy, colonoscopy) had been performed in this hospital , from this number , 350 lower endoscopy had been performed for complain of bleeding per rectum , the diagnosis of solitary rectal ulcer syndrome was in 15 patients out of those 350 patients that complain from bleeding per rectum , those 15 patients were enrolled in this study , full history had been taken, full physical examination had been done , type of treatment offered for them had been reviewed with follow up response of six months for each patient had been considered.

The detailed history taken from the patients included: duration of symptoms before the diagnosis, disorder of defecation (constipation, diarrhea, straining at defecation, digital evacuation, incontinence), perianal and abdominal pain, rectal bleeding, mucus discharge, rectal prolapse, weight loss, use of medications, tenesmus, and other points in the history include backache, skin rash and mouth ulcer.

All patients were submitted to abdominal and rectal examination looking for evidence of blood or prolapse.

After explaining and taking informed written consent from the patient for lower endoscopy and to enrolled in the study, Sigmoidoscopic or total colonoscopic examination were performed using (variable stiffness EC760R-V/I, Fujifilm, Tokyo, Japan) endoscope. The examination was performed under conscious sedation or under deep sedation for paediatric patients (using: propofol with midazolam or fentanyl) and under the supervision of an anesthetist specialist.

These endoscopic procedures were carried out after full bowel preparation by use of the preparation protocol adopted in our endoscopy unit (using split doses of osmotic laxatives with Coloclean (polyethylene glycol) sachets, dietary advice and sometimes with use of add on stimulant laxative (bisacodyl tablets) and normal saline enemas for presumed difficult preparation individuals.

These endoscopic procedures were carried out by different endoscopists at the endoscopy unit of this hospital. During the endoscopic procedure, multiple biopsies (4-6-4) were taken from the edge of lesion, center of the lesion and from the surrounding mucosa respectively and put in three different diluted formalin test tubes, the tubes were labeled for patient's name and site of biopsy taken before subjected to preparation and final examination ,these samples were prepared, stained with eosin and hematoxylin staining ,then were studied and reviewed by one and sometimes by more than one expert gastrointestinal pathologists .

The diagnosis of SRUS was established according to the histological criteria which described by Madigan and Morison [10] which necessitate the presence of the following three findings:

- 1- Fibrous obliteration of lamina propria.
- 2- Disorientation of muscularis mucosa.

3- Extension of muscle fibers into the lamina propria. In addition, all involved patients had general stool examination, biochemical and hematological investigations. Regarding the treatment of the affected individuals, patients with constipation were advised to avoid straining at defecation, avoid the habit of rectal digitation and they were advised to take a high roughage diet or fiber supplementation. All patients were subjected to different medications that are appropriate according to the recommended guidelines and include: Sucralfate enema (2gm/day for 6 weeks), Sulfasalazine enema (1-2gm/day for 3-6 months) and/or xylocaine gel. The clinical, endoscopic and histological state was assessed at time of presentation and during follow up at six months, results were graded as no symptoms ,partially improved ,unchanged or worse according to the patients' assessment of symptoms. All the fifteen patients continued and completed the follow up period, so complete medical records and follow up data were available for the fifteen patients.

As a Statistical analysis, prevalence rate had been calculated for the studied condition. The studied variables were arranged as numbers and percentages for categorical data while numerical data were represented as mean and standard deviation. Independent samples t-test was used to compare the means of two samples, with the P-value of < 0.05 was the criterion of statistical significance. The data were coded and analysed using the Statistical Package for the Social Sciences (SPSS) version 26.

Results

The prevalence rate of the SRUS among patients attending the Basrah Gastroenterology and Hepatology hospital and complained of bleeding per rectum was 4.28% .15 (4.28%) patients out of 350 patients presented with bleeding per rectum and underwent lower endoscopy in this hospital had fulfilled the diagnosis of SRUS. 9(60%) were male and 6(40%) were female (1.5:1 ratio). The mean age at the time of presentation was (22 ±9.3) years, with a range from (12-45years).

The mean duration of symptoms until confirmed the diagnosis in our hospital was (8.22 ±4) weeks, ranging from (3-17) weeks, with the female patients diagnosed earlier than male with the mean duration of (5.5±2.3) weeks and (8.2±5.3) weeks respectively and this was statistically significant (P value= 0.03). Table 1.

Despite all the patients had bleeding per rectum (100%) , but this study also categorized patients according to the associated symptoms with the bleeding per rectum and was as the following : associated constipation 14 (93.3%), associated mucous discharge 13 (86.6%), associated straining at defecation 12 (80%), associated anorectal pain 11 (73.3), associated tenesmus 9 (60 %), associated digital evacuation 7 (46.6%), associated altered bowel habits 5 (33.3%), associated lower abdominal pain 4 (26.6%) , associated diarrhea 3 (20%) and associated rectal prolapse 2 (13.3 %) , this study also demonstrated that 12 patients(80%) had multiple associated symptoms in addition to the bleeding per rectum while only 3 patients (20%) had only one associated symptom . Table2.

Table 1: Study characteristics of the patients

Number of Patients(n.)	15
Prevalence rate	4.28%
Age (year)	
Mean ± SD	22±9.3
Range	12-45
Sex (n., %)	
Male	9(60%)
Female	6(40%)
Male to Female ratio	1.5:1
Duration of symptoms (weeks)	
Mean ± SD	8.22±4
Range	3-17
Sex difference in duration of symptoms(weeks)	
Male (Mean ± SD)	5.5±2.3
Female (Mean ± SD)	0.03
p-value	

Table2: Study of the presenting symptoms

Symptoms	Number (n.)	Percentage (%)
Bleeding per rectum	15	100%
Constipation	14	93.3%
Mucous discharge	13	86.6%
Straining at defecation	12	80%
Anorectal pain	11	73.3%
Tenesmus	9	60%
Normal bowel habit	8	53.3
Digital evacuation	7	46.6%
Altered bowel habits	5	33.3
Abdominal pain	4	26.6%
Diarrhea	3	20%
Rectal prolapse	2	13.3%
Multiple associated symptoms	12	80%
Single associated symptom	3	20%

From the 15 patients, 7(46.6%) patients underwent imaging study before underwent lower endoscopy in form of dynamic computed tomography (CT scan) of the abdomen and the results interpreted as: suspicious for carcinoma in 4 patients, ulcerative colitis in one patient and nonspecific finding in the remaining 2 patients. While the hematological investigations of the involved patients revealed 7 (46.6 %) patients had iron deficiency anemia with haemoglobin less than 10 g/dl and serum ferritin less than 30 ng/ml, 4(26.6%) patients required oral iron therapy, 2(13.3%) patients required parenteral iron and only 1 (6.6%) patient required blood transfusion. A general stool examination had been performed for all fifteen patients: 1 (6.6%) patient had an Entamoeba histolytic trophozoite, 3 (20%) had an Entamoeba histolytic cyst only and 11(73.4%) had normal general stool examination. Table3.

Table 3: Study investigations of the patients

Number of patients (n.)	15
Imaging study (n., %):	7(46.6%)
Suspicious of carcinoma (n., %)	4(26.6%)
Ulcerative colitis like (n., %)	1(6.6%)
Nonspecific findings (n., %)	2(13.4%)
Iron deficiency anemia (n., %):	7(46.6%)
Received oral iron therapy (n., %)	4(26.6%)
Received parenteral iron (n., %)	2(13.4%)
Received blood transfusion (n., %)	1(6.6%)
General stool examination (n., %):	15(100%)
Normal (n., %)	11(73.4%)
Entamoeba histolytica trophozoite (n., %)	1(6.6%)
Entamoeba histolytica cyst (n., %)	3(20%)

Endoscopic examination demonstrated that 11 (73.4 %) of patients had ulcerated lesions (range from one large ulcer to multiple small ulcers), the remaining 4(26.6%) patients had non ulcer lesions {3(20%) had polypoidal lesion and 1 (6.6%) patient had just erythematous mucosa}.

The majority of the rectal lesions were located anteriorly 12/15(80 %) , circumferential lesion in 2(13.4%)patients ,while only 1(6.6%) patient had posteriorly located lesion.

The median distance of the rectal lesion was (7.6±1.78 SD) cm from anal verge (range 6-12 cm). During endoscopic examination, 12 (80%) of the examined patients had prolapse of anterior rectal wall.

Table 4

Table 4 :Endoscopic findings of the patients

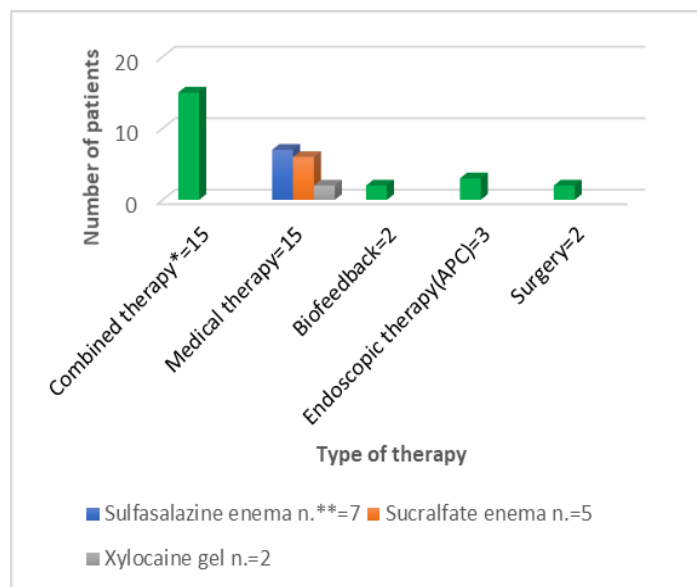
Endoscopic finding	Number(n.)	Percentage(%)
Ulcerated lesion	11	73.4%
Polypoidal lesion	3	20%
Hyperemic mucosa	1	6.6%
One ulcer	6	54.5%
Multiple ulcers	5	45.5%
Distance of lesion from anal verge:	7.6±1.78	
Mean±SD (cm)	6-12	
Range (cm)		
Location of rectal wall ulcer:		
Anterior	12	80%
Circumferential	2	13.4%
Posterior	1	6.6%
Anterior rectal wall prolapses	12	80%
Other colonoscopic finding:		
Hemorrhoids	3	20%
Sigmoid diverticulosis	1	6.6%
Fissure in Ano	1	6.6%

According to the histological abnormalities, this study showed that all the patients 15/15(100%) had fibrous obliteration of lamina propria ,hypertrophy of muscularis mucosa and regenerative changes in crypts, while associated granulation tissue were seen in 12 (80%) of

the affected patients and 9 (60%) showed associated ulcerations and /or erosions, cystic changes of mucous glands in 3(20%) patients and 4(26.6%) showed neutrophilic infiltration. Table5

Table 5: Histological findings of the patients

Histological finding	Number(n.)	Percentage (%)
Fibrous obliteration of lamina propria	15	100%
Hypertrophy of muscularis mucosa	15	100%
Regenerative crypts changes	15	100%
Granulation tissue	12	80%
Ulcerations and/or erosions	9	60%
Cystic changes of mucous glands	3	20%
Neutrophilic infiltration	4	26.6%



*Combined therapy: Conservatives treatment + Medical treatment, n.**=Number of patients

Figure1: Study of the patients according to the type of therapy

According to the modalities of therapies offered for the affected patients, this study demonstrated that all the 15(100%) patients were subjected to combined treatment modalities at time of confirmed diagnosis that consist of conservative treatment (i.e. reassurance of the patient that the lesion is benign, encouragement of a high-fiber diet, avoidance of straining, regulation of toilet habits, and attempt to

discuss any psychosocial factors, diet and bulking agents) combined with medical therapy in form of Sulfasalazine enema (1-2gm/day for 3-6 months) used by 7(46.6%) patients ,sucralfate enema (2gm/day for 6weeks) used by 6 (40%) patients and /or xylocaine gel used by 2 (13.4%) patients. 2(13.3%) patients underwent biofeedback therapy, 3(20%) underwent endoscopic therapy in form of Argon Plasma Coagulation (APC) while only 2(13.3) patients underwent surgery in form of rectopexy. Figure1

The average follows up period for the studied patients with SRUS in this study were about 6 months duration and accordingly this study showed that 7 (46.6) patients demonstrated improved symptoms, disappearance of symptoms in 5 (33.4%) while persist in 3(20%).

At the end of the follow up period all the studied patients subjected to sigmoidoscopic examination and biopsy were taken. The endoscopic examination showed that endoscopic healing of the lesion was documented in 5 (33.3%) patients, improved in 8(53.3%) while persisted in 2 (13.4%) patients, while the histopathological results showed that were no changes in histological examination in 2 (13.3%), improved in 11 (73.4%) while disappearance of changes (normal histopathology) in 2 (13.3%) patients. Table6

Table 6: Follow up study of the patients

Follow up (6 months)	Number(n.)	Percentage (%)
Symptom:		
Disappear of symptoms	5	33.4%
Improving of symptoms	7	46.6%
Persistence of symptoms	3	20%
Sigmoidoscopy:		
Disappear of lesion	5	33.4%
Improvement of lesion	8	53.3%
No change in endoscopy	2	13.3%
Histopathology:		
Normal histopathology	2	13.3%
Improving histopathology	11	73.4%
No change in histopathology	2	13.3%

Discussion

Although there is no so much studies to compare the prevalence of the SRUS with it, the Morio O et al showed that the incidence of SRUS is 1/100000/year(11), this study showed increase in the prevalence of the SRUS as this is a single center study , this can be explained by that the endoscopies were done in specialized tertiary hospital and done by expert endoscopists that had increased awareness to this syndrome, and the specimens had been examined by expert pathologists with high awareness to this uncommon syndrome.

There was male predominance of this syndrome in this study (male: female ratio is 1.5:1) , this is similar to a study done by Abusharifah O et al , that showed male predominance for this syndrome(12) , although AlGhulayqah AI et al showed female predominance(13) ,

while Forootan M, Darvishi M reported in their study equal prevalence in men and women (14).

This study showed that the mean age of the affected patients was (22 ±9.3) years, with range from (12-45years) , this is younger than the age presentation in other studies like that done by Abid S et al in Pakistan and Behera MK et al in India (15,16) , and this can be explained by the increase awareness to this syndrome in our hospital. Regarding the mean duration for the diagnosis of SRUS in Basrah Gastroenterology and Hepatology(from the onset of symptoms and presentation to the diagnosis in this hospital), this study demonstrated that the mean duration was (8.22 ±4) weeks, ranging from (3-17)weeks and this is shorter than the mean duration in Zubair E et al, that show that the mean duration was (11.5 ± 4.3) weeks with a range from (1-23 weeks) (5) , and this is explained by the adoption of the strategy in our hospital that bleeding per rectum is considered an urgent indication for endoscopy that result in short waiting list in addition to the increase the experience to this syndrome among endoscopists , radiologists and pathologists in this hospital.

The female patients in this study diagnosed earlier than the male, this can be explained by the attitude of the patients in our country; in that women usually seek medical help earlier than the men as with other diseases. Although no study elsewhere is available to compare this result with it.

In this study the association of bleeding per rectum , constipation and mucous discharge were the most common associated presented symptoms , this is similar to the finding by Urganci N et al (17) , while Dehghani SM et al showed that than combination of bleeding per rectum, straining during defecation or forceful defecation and Sense of incomplete evacuation(98.2%, 90.9% and 61.8%)respectively were the most common presenting symptoms(18) ,and this can be explained by the horror of bleeding per rectum for the patient and the neglect of the other associated symptoms in presence of bleeding per rectum.

This study showed that the imaging study in case of SRUS can be confusing as what demonstrated by the finding of suspicious rectal carcinoma, ulcerative colitis and nonspecific finding , similar to that; Bhusal U founded SRUS mimicking rectal tumor on CT scan(19),Powell CR et al founded SRUS mimicking Perianal Crohn's Disease(20) , this is can be explained by the characteristic similarity of the SRUS with the neoplasm and the inflammatory bowel disease on imaging study and the difficulty of differentiation between them based on imaging study only.

Regarding the laboratory investigation , this study show that anemia is a relatively common finding in the affected patients(46.6%) and this can be explained by the ongoing blood loss that sometimes can be massive and by the relatively long duration till reaching the diagnosis , Abusharifah O et al showed similar results in their study that showed iron deficiency anemia in (42.1%) of the affected patients(12).

In this study most of the studied patients had ulcerated lesions ranging from one to multiple ulcers of varying sizes , three patients had polypoidal like lesion and only one patient had hyperemic mucosal lesion , these results similar to the results demonstrated in other studied like that done by Behera MK et al in India and showed that ulcerative lesions were seen in 83% of the patients , Polypoidal

lesions in 17.4% ,erythematous mucosa in 2.2% and rectal polyps in 5.4%(16) , other study conducted in Baghdad by Lafta KB et al showed comparable results as ulcerated lesions (87.5%) were the most common endoscopic findings(21).

This study as other studies conducted for similar reason like that performed by Waniczek D et al in Poland, Kumagai H et al in Japan and showed that most of the rectal lesions located anteriorly, very less commonly the lesions were located circumferentially or posteriorly (22,23).

The histological abnormalities of the studied affected patients showed that all the patients 15 (100%) had fibrous obliteration of lamina propria with hypertrophy of muscularis mucosa and regenerative changes in crypts and this is the typical histopathological diagnostic finding in SRUS ,while associated granulation tissue were seen in 12 (80%) of the affected patients and 9 (60%) showed associated ulcerations and /or erosions, this is identical to the finding in the similar studies like that done by Al-Brahim N et al in Kuwait and that conducted by Suresh N et al in India (24,25).

Different modalities of therapy are available for the SRUS ranging from the conservatives' therapy (dietary modifications, toilet education, avoidance of digitation), single or combined medical treatment with sucralfate enema, sulfasalazine enema and xylocaine gel application, there is also endoscopic treatment with argon plasma coagulation and finally there are different surgical modalities. There is no agreement about the management of SRUS, numerous modalities have not been proved successfully. Patient education and behavioral modifications are the first steps in the treatment of SRUS. In this study all patients received combined conservatives and medical therapy ,in addition there were two patients that received biofeedback therapy, three patients received endoscopic therapy and two patients underwent surgical therapy with rectopexy , these treatment approaches also had been adopted by Forootan M et al that showed the effective role of biofeedback in SRUS, Gouriou C et al in France that showed the effectiveness of the conservative and the medical treatment and Bulut T et al that explored the treatment options available for the SRUS (26,27,28).

On follow up of the patients , this study showed that symptoms improved or disappeared in most of the affected patients (80%) , while symptoms persist in (20%) of patients , with most of the affected patients showed endoscopic improvement or healing of the lesion(86.6%) and only (13.4%) showed persistent lesion on follow up sigmoidoscopic examination , while (86.7%) of the affected patients showed histological improvement and/or healing of the lesion and only (13.3%) showed no histological changes , and as most of those patients received combined conservatives and medical treatment , so the high percentage of symptoms , endoscopic and histological improvement and/or healing can be explained by the effectiveness of the management strategy adopted in this hospital which included the encouragement of a high-fiber diet, avoidance of straining, regulation of toilet habits, and attempt to discuss any psychosocial factors, in combination with stool softeners and bulking laxatives, along with the effect of the topical agents(sucralfate and sulfasalazine enema), in which the sucralfate enema contains aluminum complex salts which coat the rectal ulcer and form a barrier against irritants, allowing the ulcer to heal, while the sulfasalazine

enemas has anti-inflammatory properties that help ulcer healing by reducing the inflammatory responses, these results supported by studies done by Blackburn C et al , El-hemaly M et al and Gouriou C et al for similar purposes(29,30,31).

Anorectal physiological testing and defecography are investigative tools that may provide further insight into the pathogenesis of this condition were not performed in this study due to unavailability of these modalities in this hospital, in addition this is a single center study, and these can be considered as limitations for this study.

Conclusion

SRUS is an uncommon benign defecation disorder with diverse clinical presentations, with rectal bleeding being the most common presenting symptom. The endoscopic findings can be variable with the histological examination being the gold standard for establishing the diagnosis.

Different treatment options are available and the majority of patients in our study respond well to non-surgical therapy.

Confusion with other conditions like rectal cancer and inflammatory bowel diseases is not uncommon. Physicians, surgeons and pathologists should be aware of the features of SRUS so that it is not confused with other conditions.

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Conflict of Interest

Authors have declared that no competing interests exist

Data availability

The datasets used and/or analyzed during the current study are available from the corresponding author on reasonable request.

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