

Solitary rectal ulcer syndrome: characteristics, outcomes and predictive profiles for persistent bleeding per rectum

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ABSTRACT

Introduction: Solitary rectal ulcer syndrome (SRUS) is a rare disorder of defaecation and persistence of symptoms is common, particularly bleeding per rectum (BPR). This study assessed the clinical, endoscopic characteristics and predictive profiles for persistent BPR.

Methods: 28 patients (14 males) with biopsy-proven SRUS were identified from 1989 to 2003, and the clinical and endoscopic findings were retrospectively reviewed.

Results: At presentation, the mean age was 29.5 +/- 16.1 (range ten to 81) years. Common symptoms reported included BPR (86 percent), abdominal pain (36 percent), mucus per rectum (25 percent), straining at defaecation (25 percent), diarrhoea (14 percent) and constipation (14 percent). Digital manual evacuation was reported by 11 percent. 68 percent were anaemic and 57 percent required blood transfusion. Lesions were located anteriorly (38.5 percent), posteriorly (30.7 percent) and circumferentially (31.8 percent). The lesions were multiple (34 percent), ulcerative (64.3 percent) and polypoidal/nodular (32.1 percent). At a mean follow-up of 43.5 +/- 36 months, 64 percent (n=18) had multiple admissions (mean 3.1, range one to 12), mainly for transfusion (mean 7.4 units, range two to 27). There was no difference in clinical responses between patients with polypoidal/nodular or ulcerative lesions (p-value is 0.653). Follow-up endoscopies showed improvement (58 percent), progression (21 percent) and no change (21 percent) in the lesions. Four patients had surgery for concerns of neoplasms (n=2) and persistent BPR (n=2). BPR was persistent in 39 percent. Presence of abdominal pain (p-value is 0.008) and passage of abnormal stool (p-value is 0.002) were predictive of persistent BPR.

Conclusion: SRUS occurs predominantly in young patients and despite being a benign condition, morbidity remains a problem. Patients' profiles are predictive of persistent BPR.

Keywords: haematochezia, rectal prolapse, rectal ulcer, solitary rectal ulcer syndrome

Singapore Med J 2006; 47(12):1063-1068

INTRODUCTION

Solitary rectal ulcer syndrome (SRUS) is a rare disorder of defaecation that has typical histological findings. It was first described in 1968⁽¹⁾. This syndrome commonly presents with bleeding per rectum (BPR) in association with other symptoms such as abdominal pain, passage of mucus, straining at defaecation and feeling of incomplete evacuation. A history of manual evacuation can be elicited by direct questioning^(2,3). Unless recognised, the diagnosis can be delayed and be mistaken for non-specific ulcer, inflammatory bowel disease or neoplasm⁽²⁾. This can lead to inappropriate treatment being given. However, despite a correct diagnosis, the outcome can be unsatisfactory, leading to non-healing of the rectal lesions and persistence of symptoms. Persistence of symptoms such as BPR is common and can be distressing for the patients. Current treatment includes the use of bulking agents, laxatives, sucralfate, bowel retraining with or without biofeedback and surgery⁽⁴⁻¹⁰⁾. However, currently available therapies have major limitations. This study assessed the clinical, endoscopic characteristics and outcomes, and the factors that predict persistence of BPR in SRUS patients treated at a referral centre in Brunei Darussalam.

METHODS

Raja Isteri Pengiran Anak Saleha (RIPAS) Hospital is a 550-bed hospital and is the main referral centre in Brunei Darussalam, situated in the capital, Bandar Seri Begawan. The hospital caters to three of the four districts in the country, namely:

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Brunei-Muara, Tutong and Temburong. The population catchment for the unit is approximately 270,000. The other district (Kuala Belait) is served by a smaller endoscopy unit.

Evaluation of patients included routine blood investigations, stool tests (microscopy and culture) for infections, and endoscopy (sigmoidoscopy or colonoscopy). Diagnosis of SRUS was based on the characteristic endoscopic findings in the rectum, namely: ulcerations (Fig. 1) or polypoidal/nodular (Fig. 2), exclusion of other causes of the rectal lesions (neoplasm, infection, inflammatory bowel disease, and trauma), and a consistent histology. The histology should have a characteristic appearance that includes a thickened mucosal layer with distortion of the crypt architecture. The lamina propria is replaced with smooth muscle and collagen leading to hypertrophy and disorganisation of the muscularis mucosa, an appearance that has been referred to as "fibromuscular obliteration"⁽¹⁾.

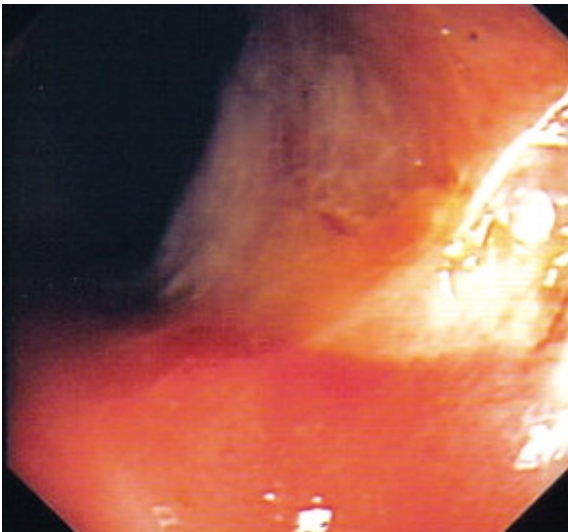


Fig. 1 Endoscopic photograph shows a ulcerative lesion.

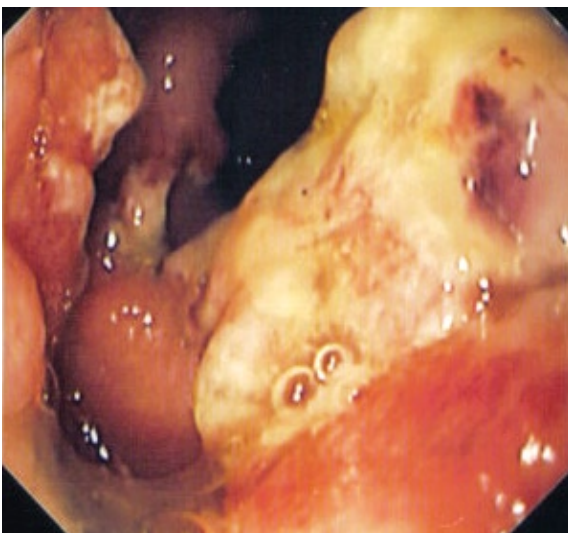


Fig. 2 Endoscopic photograph shows a polypoidal/nodular lesion.

Specialised investigations such as ano-rectal physiological testing and defaecography are not available at our institution. Hence, no differentiation or evaluation for any prolapse and the degree of prolapse were done. Types of treatment locally available consist of bulking agents (Normacol, Metamucil and Lactulose), enemas (Steroid [Predsol enema] and Mesalamine), oral 5-aminosalicylate (5-ASA), endoscopic steroid injection and surgery. Intra-lesional steroid injections were used due to the unavailability of other effective therapies for those patients who remained symptomatic despite treatment and who were not keen on surgical alternatives.

Patients diagnosed with SRUS were informed of their underlying condition and the overall management plan. Management began with education and fibre supplements, which were given to all patients (n=28). Patients were advised to be regular with their bowel habits, and to avoid excessive straining during bowel evacuation. They were also advised to refrain from performing manual evacuation. Iron replacement was given (n=19) if there was anaemia. In addition, short courses (one to two weeks) of daily enema (steroid-Predsol enema and/or 5-ASA, Mesalamine enema [n=15]) treatment was given to those who have large rectal lesions (usually polypoidal or nodular), especially after episodes of significant bleeding. Oral 5-ASA (n=6) was given to those patients who responded to 5-ASA enema. Therapies were tailed down once symptoms have improved.

Endoscopic steroid injections (n=7) (Solu-Cortef®, Pharmacia, 100 mg diluted in 10 ml, injected in 1 to 2 ml aliquots) were performed for those who had large lesions with persistent bleeding. All of these seven patients had already been given enema treatment. Surgery (n=4) was only considered after thorough discussions with the patients or guardians, particularly for those with suspicious histology or persistent symptoms such as BPR, and difficult stool evacuation. Patients were routinely followed-up and assessed for further bleeding and difficulty with bowel evacuation. Blood investigations (complete blood count) were routinely carried out to assess for anaemia. Transfusions were given if there was symptomatic anaemia. Repeat endoscopies were not routinely carried out unless patients had persistent symptoms, particularly bleeding. Patients whose symptoms had resolved after a period of follow-up were discharged to the outpatient clinic for follow-up.

Between 1989 and 2003, 32 patients with histology-proven SRUS were treated. However, four patients' records were not available, leaving 28 patients for the analysis. Demographical data (age, gender and race), clinical presentations (BPR,

abdominal pain, diarrhoea, constipation, passage of mucus, manual evacuation, stool characteristics and description of bowel habits), treatment (medications and transfusions), and endoscopic and histopathology findings were retrieved from the case records and computer laboratory records. Clinical and endoscopic outcomes of patients were also reviewed.

Data was entered into the Statistical Package for Social Sciences (SPSS) version 10.0 (Chicago, IL, USA) programme for analysis. Continuous data was presented as mean, standard deviations and range. The Mann-Whitney test and Fisher's Exact test were used to assess the differences between patients who still had bleeding on follow-up and those who did not have bleeding. Patients who had surgery (n=4) were excluded from analysis, leaving 24 patients for comparison. Level of statistical significance was considered when $p < 0.05$ (two-tailed).

RESULTS

The patient demographics are shown in Table I. Only three patients had comorbid conditions. One was wheelchair-bound with associated osteoarthritis, ischaemic heart disease and congestive cardiac failure. One patient had schizophrenia and the other has diabetes mellitus, hypertension, gout and epilepsy. The indications for evaluation were: BPR 46.4% (13), anaemia evaluation 25% (7), BPR/anaemia 25% (7) and elevated tumour marker (carcinoembryonic antigen) 3.6% (1). The prevalence of symptoms is shown in Table II. The frequency of bowel evacuations ranged from one to seven per day, often passing a small amount of stool. Only three patients described their bowel habits as changed before investigations. 19 (68%) patients were anaemic, with 16 (57%) patients requiring blood transfusion.

The lesions were located at a mean distance of 5.7 cm from the anal verge (ranging from within anal verge to 10 cm proximally). The lesions were located anteriorly (38.5%), posteriorly (30.7%) and circumferentially (31.8%). 66% had a solitary lesion and 34% had multiple lesions. Lesions were ulcerative in 64.3%, polypoidal/nodular in 32.1%, with friable mucosa in one patient. At a mean follow-up of 43.5 ± 36 months, 64% of patients (18) had multiple admissions (mean 3.1, range 1-12), mainly for blood transfusion (mean 7.4 units, range 2-27) in 15 patients. Equal proportions of patients with ulcerative and polypoidal/nodular lesions were treated with fibre and combination therapies. Ten (35.7%) patients [ulcerative (7) and polypoidal (3)] were treated with only bulking

Table I. Demographical and haematological data at presentation.

Age (years) [†]	29.5 ± 16.1 (10-81)
Gender (male:female)	14:14
Race	
Malay	27 (96.4%)
Indigenous	1 (3.6%)
Comorbid conditions	3 (10.7%)
Haemoglobin (g/dL) [†]	8.7 ± 3.9 (4.0-14.9)
MCV (fL) [†]	69.9 ± 14.8 (50.4-94.0)
Haematocrit (%) [†]	27.1 ± 11.0 (13.5-46.5)
Documented iron deficiency [‡]	13 (46.4%)

[†] In mean, standard deviation and range (in parentheses)

[‡] In absolute number and percentage (in parentheses)

MCV: mean corpuscular volume

Table II. Prevalence of clinical symptoms.

Symptoms [†]	n (%)
Abdominal pain	
Lower	8 (28.6)
Non-specific	2 (7.1)
Bleeding per rectum	24 (86)
Constipation	4 (14)
Diarrhoea	4 (14)
Digital manual evacuation	3 (11)
Passage of mucus	7 (25)
Stool	
Hard	7 (25)
Loose	4 (14)
Normal	17 (61)
Straining during bowel evacuation	7 (25)

[†]Some patients have more than one symptom at presentation.

agents and had remained well without significant symptom relapse.

Four patients [polypoidal (2), ulcerative (1) and friable mucosa (1)] underwent surgery [trans-anal mucosectomy (3) and anterior resection of the rectum (1)] due to concerns of underlying malignancies (1) and patients' decision for surgical treatment after failure of medical therapies (2). Histology showed some adenomatous features without dysplasia (2) and features consistent with SRUS (2). One patient's progress was complicated by sepsis and adult respiratory distress syndrome post-surgery. This was later complicated by a chronic fistula requiring colostomy in an attempt to allow the fistula to close.

Two patients still experienced BPR post-surgery secondary to haemorrhoids.

80% (12/15) of patients who had experienced enema treatment still had intermittent BPR on follow-up. Six of these patients had also been treated with steroid intra-lesional injection. One patient proceeded to surgery due to persistent bleeding. One patient died of metastatic breast carcinoma. She had been discharged from follow-up when her symptoms had completely resolved without further relapse. Overall, patients with bulkier (polypoidal or nodular) lesions had less clinical response (66.7% versus 77.8%, $p=0.653$) compared to those with ulcerative lesions at follow-up, but this was not statistically significant.

Two-thirds (19/28) of patients had multiple endoscopies (mean 3.7/patient, range 2-8). There were no significant differences in the improvement (60% versus 63%, $p=1.000$) and progression (10% versus 25%, $p=0.396$) rates between patients with ulcerative and polypoidal/nodular lesions, respectively. These findings are shown in Table III. Exacerbation of symptoms correlated with endoscopical progression. None of these patients had complete resolution of their underlying lesions. Follow-up biopsies showed consistent features of SRUS and did not show any evidence of malignant changes.

Table III. Outcomes of lesions in patients at follow-up endoscopy (n=19).

Type of lesion	Improvement		Progression		No change	
	n	n (%)	n (%)	n (%)	n (%)	n (%)
Ulcerative	10	6 (60)	1 (10)	3 (30)		
Polypoidal/nodular	8	5 (62.5)	2 (25)	1 (12.5)		
Friable mucosa	1	-	1 (100)	-		
Overall	19	11 (58)	4 (21)	4 (21)		

Overall, 46% (13/28) still had BPR on follow-up. However, bleeding secondary to SRUS was seen in 39% (11/28). Two patients had bleeding secondary to haemorrhoids. Table IV shows the differences in patients with or without BPR on follow-up. Patients who had persistent BPR had significantly more abdominal pain (64% versus 8%, $p=0.008$) and passage of abnormal stool, either hard or liquid (73% versus 8%, $p=0.002$). Patients were also younger and tended to have more constipation but these were not statistically significant.

Table IV. Profiles that were predictive of persistent BPR.

Parameters compared	BPR (n=11) n (%)	No BPR (n=13) n (%)	p-value
Age at presentation (years) [†]	24.1 ± 14.2	35.7 ± 18.1	0.111
Haemoglobin at presentation (g/dL) [†]	8.5 ± 4.3	8.5 ± 3.7	0.451
Passage of mucus	4 (36)	1 (8)	0.142
Straining at defaecation	4 (36)	2 (15)	0.357
Abdominal pain	7 (64)	1 (8)	0.008
Constipation	3 (27)	0 (0)	0.082
Diarrhoea	2 (18)	2 (15)	1.000
Digital manual evacuation	2 (18)	1 (8)	0.576
Abnormal stool	8 (73)	1 (8)	0.002
Endoscopical findings			
Ulcerative lesions (n=17)	7 (41)	10 (59)	0.659
Polypoidal/nodular (n=7)	4 (57)	3 (43)	

Four patients who had surgery were excluded from comparison.

[†] Mean and standard deviation (Mann-Whitney test)

F-test was used for comparison of other parameters.

DISCUSSION

Our study showed that SRUS predominantly occurs in young population with no gender predilection. In addition, SRUS commonly occurs in patients without other significant comorbid conditions. The mean age at presentation is younger than those previously reported^(2,3,6,7,11). However, this varies between reports, ranging from as young as 4.5 years in the paediatric literature⁽¹²⁾ to 81 years in our study. The clinical spectrum of presentations in this study is similar to published reports with BPR being the commonest presentation. It is difficult to explain why only three patients admitted that their bowel habits had changed prior to presentation. Perhaps this reflected the nature of the condition and also the fact that the disorder processes may have actually been present longer before presentation. Hence, patients may not have considered their bowel habits as changed or abnormal.

In agreement with many authors^(5,11,13), the term SRUS is a misnomer, as 34% of the endoscopic findings in this study showed multiple lesions. Similarly, lesions are not necessary ulcerative. A spectrum of endoscopical findings suggests that SRUS may be part of a defaecation disorder^(14,15).

Associations with malignancy have been reported and this is a major concern^(16,17). In the present study, there was no evidence of malignant transformation in any of the patients who had undergone repeated endoscopies and histological examinations. However, there were adenomatous changes seen in the resected specimens of two patients but there was no evidence of dysplasia. Malignant transformation therefore is possible but this requires a longer follow-up study of a larger cohort of patients.

As shown in this and many other studies^(5,6,11,18), currently-available treatment options are suboptimal. One study showed that symptomatic improvement with bulk laxatives and bowel retraining was only seen in 19% of the patients⁽¹¹⁾. In our study, more patients with bulkier polypoidal or nodular lesions have persistent BPR and less clinical improvement on follow-up compared to patients with ulcerative lesions. Despite this, approximately two-thirds of patients reported symptomatic improvement. Follow-up endoscopies did not show any significant differences in both the improvement and progression rates of the lesions. This is in agreement with studies that have shown improvement in clinical symptoms even without significant endoscopic improvement⁽⁴⁾. Similarly, symptoms can persist despite endoscopic healing⁽¹¹⁾.

Intra-lesional steroidal injections and enemas were tried, due to the unavailability of other treatment modalities. These were given to patients who had bulkier lesions and significant bleeding. Our initial experience with steroid injection showed that some patients benefited from this mode of therapy. However, almost all patients had persistent BPR on follow-up, suggesting its ineffectiveness. Similarly, this was also the case with enema therapy. Although it could be argued that trauma to the rectal mucosa from the enema delivery device could worsen the condition, it is unlikely that these contributed significantly to the persistence of symptoms as these were only given intermittently for one to two weeks. Furthermore, none of the patients were on long-term enema treatment.

Of note, there were ten patients who were only treated with fibre supplement and did not have further relapse. This suggests that a subgroup of patients' conditions may be self-limiting or responsive to fibre supplement and bowel retraining (based on advice given by the treating clinicians). Specialised treatment modality, such as biofeedback therapy, has been reported to benefit patients by correcting the defaecation process. However, the benefit tended to decline with longer follow-up⁽⁶⁾. The same group also showed that biofeedback can improve the mucosal blood flow, leading to lesion improvement⁽¹⁹⁾,

suggesting that ischaemia is involved in the underlying pathogenesis.

As SRUS is believed to involve a defaecatory disorder associated with prolapse^(3,11,20,21), biofeedback and surgery (particularly abdominal rectopexy) seem to be ideal strategies as they aim to correct the underlying disordered processes^(4,6,7). However, these were not always reflected by published reports utilising these two methods. Since only four patients underwent surgery in this study, it is not possible to draw any conclusion on this treatment modality, particularly with the potential complications as well as the fact that two patients still had BPR after surgery. Although bleeding were due to haemorrhoids, persistence of BPR can be distressing to the patients.

Although this disorder can be recognised easily, there is still the possibility of misdiagnosis. Even with a correct diagnosis, there is still a significant proportion of patients who will continue to have symptoms as reflected in this and other studies. More needs to be done to evaluate different treatment strategies or better selection criteria for the various treatment modalities that are currently available. Prolonged preoperative evacuation time and lesion characteristics have been shown to predict poor outcomes^(11,21). Polypoidal or nodular lesions have been shown to predict better symptomatic response compared to the ulcerative variety⁽¹¹⁾. Our study showed that the bulkier polypoidal or nodular lesions had a less favourable response, although this was not statistically significant.

Patients' symptom profiles can also predict persistence of symptoms such as BPR. Presence of abdominal pain and abnormal stool (hard or loose) at presentations and follow-up were significant predictive factors. The patients were also younger and tended to have more constipation. Presence of abdominal pain and altered stool most likely indicated that the underlying processes were still present, rather than the cause of the on-going pathogenesis. Although not statistically significant, finding of constipation as a predictive factor for symptom persistence is not unexpected as it is often associated with difficulty with defaecation. Constipation can perpetuate the underlying pathology due to the straining process involved and it can also lead to mucosal prolapse. The underlying condition itself can lead to constipation. Younger age is known to be associated with more noncompliance with treatment. Therefore, patients with such profiles should be managed more aggressively to reduce the associated morbidities.

In many centres, specialised investigations and treatment modalities, such as biofeedback or bowel retraining programmes, are not widely available. It is possible that with these forms of treatment, the results might have been better. We were not able to make much comparisons between the various treatment types due to the limited options that were available in our local setting. However, our study is representative of the "real life situation" where investigation modalities and treatment options are often limited. Therefore, our results provide some insight into the management of SRUS and the factors that help to identify patients who are likely to fail with the treatment options that are currently available.

In conclusion, this study has shown that SRUS occurs predominantly in young patients, presenting mainly with BPR and anaemia. Despite being a benign condition, morbidity remains a problem, as reflected by persistence of symptoms, especially BPR requiring multiple admissions. All the difficulties with treating patients should ideally be identified and managed in specialised centres with an interest in these areas. Presence of abdominal pain and passage of abnormal stool at presentation and follow-up are predictive factors. Like any other disorders, especially those where available treatment modes have limitations, patients' understanding and involvement are paramount. More studies are required to define the selection criteria and treatment modalities for the management of this condition.

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