

## Original Article

# Solitary rectal ulcer syndrome in children and adolescents: a descriptive clinicopathologic study

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Received September 4, 2020; Accepted December 2, 2020; Epub April 15, 2021; Published April 30, 2021

**Abstract:** Solitary rectal ulcer syndrome (SRUS) is an uncommon disorder of the rectum. While benign, it can cause concern for patients and affect quality of life. Reported studies on SRUS worldwide are scarce. The aim of this study is to describe the clinicopathologic characteristics of SRUS in a cohort of children based in Saudi Arabia. In this study, children with a confirmed diagnosis of SRUS at King Abdulaziz University Hospital (KAUH) were included, during the period November 2003 to November 2017. Data were collected from hospital medical records. The study comprised twenty-one patients: 17 males (81%) and 4 females (19%); the median age was 11.4 years (range, 5.43-17.9 years). The most common presenting symptoms were rectal bleeding in 21 patients (100%), passage of mucus in 16 (76.1%), abdominal pain in 14 (66.6%), constipation in 13 (61.9%), straining in 9 (42.9%), and rectal prolapse in 5 (23.8%). The most common finding at initial colonoscopy was a single ulcer in 7 patients (33.3%), multiple ulcers in 6 (28.5%), polypoid lesions in 5 (23.8%), and hyperemic mucosa in 3 (14.2%). All patients received medical treatment and 14 (81%) continued to manifest one or more of the symptoms following treatment, which required subsequent modification of the treatment course. None of the patients required surgery. In conclusion, the study found rectal bleeding to be the most common presentation, with a single ulcer being the most prevalent lesion in endoscopy. Treatment response was variable, but almost half of patients reported relief of symptoms following treatment.

**Keywords:** Solitary rectal ulcer, mucosal prolapse syndrome, mesalamine, sucralfate, enema, children, Saudi Arabia

## Introduction

Solitary rectal ulcer syndrome (SRUS) is an uncommon and benign disorder that affects the rectum, characterized by a combination of symptoms, endoscopic findings, and histologic abnormalities that do not necessarily culminate in ulceration or a singular ulcer [1, 2]. Indeed, the term *solitary rectal ulcer* may be inaccurate in specific cases because the endoscopic spectrum of the syndrome varies from simple hyperemic mucosa, to small or giant ulcers, to broad-based polypoid lesions [3]. Also, it has been suggested that SRUS might not be limited to the rectum and may involve the sigmoid colon [4].

Although relatively uncommon, it is a common condition in the adult population, with an esti-

mated prevalence of 1 in 100,000 adults per year [5, 6]. In children, however, SRUS is rare, and despite the symptoms being similar to those in adults it is still underdiagnosed or misdiagnosed owing to its infrequency, especially compared to more common and serious conditions, such as inflammatory bowel disease [7, 8].

Data on SRUS in children are scarce, with the majority of key publications being case reports [8, 9] or case series on small patient cohorts [7, 10-12]. The condition is also well-recognized in Middle Eastern children [13-15], with the largest local case series being conducted in Iran [16]. The most recent case series to the authors' knowledge is based in India, and is the largest to date in both adults and children, reporting on 140 children with SRUS [17].

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**Table 1.** Clinical and demographic characteristics of the study cohort (n=21) at diagnosis

| Variable                | mean $\pm$ SD, n (%) |
|-------------------------|----------------------|
| Demographic feature     |                      |
| Mean age, years         | 12 $\pm$ 3.9         |
| Male gender             | 17 (80.9%)           |
| Clinical manifestations |                      |
| Rectal bleeding         | 21 (100%)            |
| Passage of mucus        | 16 (76.1%)           |
| Abdominal pain          | 14 (66.6%)           |
| Constipation            | 13 (61.9%)           |
| Straining               | 9 (42.9%)            |
| Rectal prophase         | 5 (23.8%)            |
| Diarrhea                | 4 (19%)              |
| Blood transfusion       | 2 (9.5%)             |
| Perianal discomfort     | 1 (4.8%)             |
| Habits                  |                      |
| Use of squat toilet     | 10 (47.6%)           |
| Digitation              | 1 (4.8%)             |
| Laboratory data         |                      |
| Hemoglobin (g/dL)       | 11.7 $\pm$ 2.9       |
| Albumin (g/L)           | 34.1 $\pm$ 11.9      |

The aim of this study is to summarize the clinical, endoscopic, and histologic features of SRUS in children and to share the experience of the treatment of this rare disease.

## Patients and methods

### Study population

Children with a confirmed diagnosis of SRUS were identified on the Department of Pathology database in King Abdulaziz University Hospital during the period between November 2003 and November 2017. Only patients investigated by colonoscopy or sigmoidoscopy that were 18 years or younger at the time of diagnosis were included. Patients with SRUS identified incidentally during anal or rectal surgery were excluded.

### Diagnostic criteria for SRUS

The diagnosis of SRUS was established according to standard histopathologic criteria. The histopathologic examination was performed by an experienced certified gastrointestinal pathologist (one of the authors). Histologic specimens of rectal tissue at the time of diagnosis were

re-reviewed to confirm the diagnosis and were compared with subsequent biopsies.

The data were collected using the patient medical charts, endoscopy unit records, and the hospital digital information system, comprising information regarding age, clinical manifestations, laboratory investigations, endoscopic examination, and treatment given. Follow-up telephone calls were made to parents to ascertain patient symptoms.

### Statistical analysis

The Statistical Package for the Social Sciences (SPSS), version 20 (SPSS, Inc., Chicago, Ill) was used to analyse the data. Descriptive statistics, specifically scores and percentages, were used for categorical data, and means with standard deviation (SD) or medians with range were used for continuous variables when appropriate.

### Ethical oversight

The study was approved by the Research Committee of the Biomedical Ethics Unit at King Abdulaziz University (Reference No. 230-20). Informed consent was obtained from all participants. The study was performed in accordance with the Declaration of Helsinki.

## Results

### Clinical and demographic data

Twenty-one patients were included in the analysis. The median age was 11.4 years (range, 5.43-17.9 years). Males constituted 81% of the cohort (n=17). The most common presenting symptoms were rectal bleeding, recorded in all 21 patients (100%), with passage of mucus in 16 (76.1%), constipation in 13 (61.9%), abdominal pain in 14 (66.6%), straining in 9 (42.9%), and rectal prolapse in 5 (23.8%) (**Table 1**). Ten patients (47.6%) reported using squat toilets.

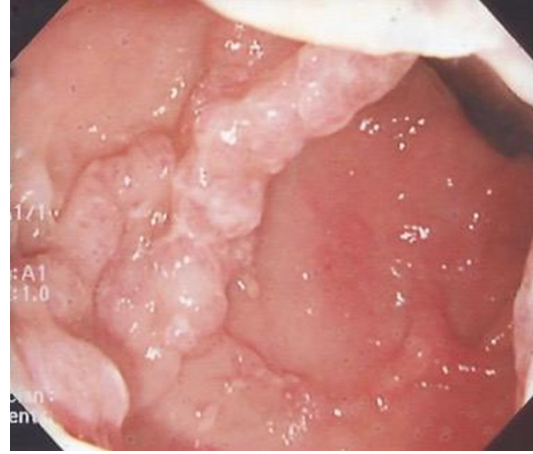
### Endoscopic findings

During the initial endoscopy, SRUS was described as a single ulcer in 7 patients (33.3%) (**Figure 1**), multiple ulcers in 6 (28.55) (**Figure 2**), polypoid lesions in 5 (23.8%) (**Figure 3**), or hyperemic mucosa in 3 (14.2%). Fourteen patients (66.7%) underwent repeated elective sigmoidoscopy or colonoscopy for monitoring

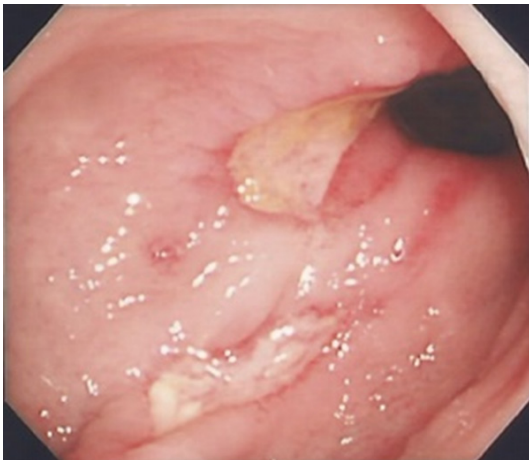
## Solitary rectal ulcer syndrome in children



**Figure 1.** Endoscopic image showing a large wide based single ulcer covered with exudate (whitish area, left), located on a transverse rectal fold surrounded by mucosal erythema.



**Figure 3.** Finding of a rectal polypoid mass, non-obstructing and involving left side of the rectum.



**Figure 2.** Two adjacent small rectal ulcers separated by an area of relatively healthy mucosa.

response to therapy. The average number of endoscopy procedures for each patient was 4.

### *Histopathologic data*

Histological examination of colonic mucosal samples was performed on all patients to confirm the diagnosis of SRUS. All patient samples had fibromuscular hyperplasia of the lamina propria. Thickening, splaying, and vertical extension of the muscularis mucosae (MM) into mucosa was seen in 9 of 10 patients where the muscularis mucosa was included in the sampled tissue. MM was not included in the biopsy specimens of 11 patients. Crypt abnormalities were seen in 13 patients (61.9%), villiform surface in 5 (23.8%), and ulceration in 9 (42.8%).

**Table 2** and **Figures 4-6** demonstrate the various findings seen in the patients.

### *Treatment outcomes*

All patients were prescribed stool softeners and advised against straining during defecation. Two patients were lost in follow-up, following initial endoscopy. Of the remaining patients, 10 required multiple therapy, either sequentially or in combination, and 7 received monotherapy. In patients that received multiple therapies, there was either a combination of mesalamine and sucralfate enema (n=5), mesalamine and corticosteroid enema (n=1), or a combination of these three therapies (n=4). One patient (4.8%) was on monotherapy, and oral mesalamine was given, sucralfate enema was given for 3 patients (14.2%), and likewise corticosteroid enema for 3 patients (14.2%) (**Tables 3** and **4**). None of the patients required surgery.

Blood transfusions were required for two patients (9.5%); 5 patients (23.8%) received oral iron therapy alone; and one patient (4.8%) required parenteral iron after failure of oral iron therapy.

Of the 19 patients who were seen subsequent to initial treatment, 14 continued to manifest one or more symptoms and required repeated endoscopy and adjustment of treatment.

### **Discussion**

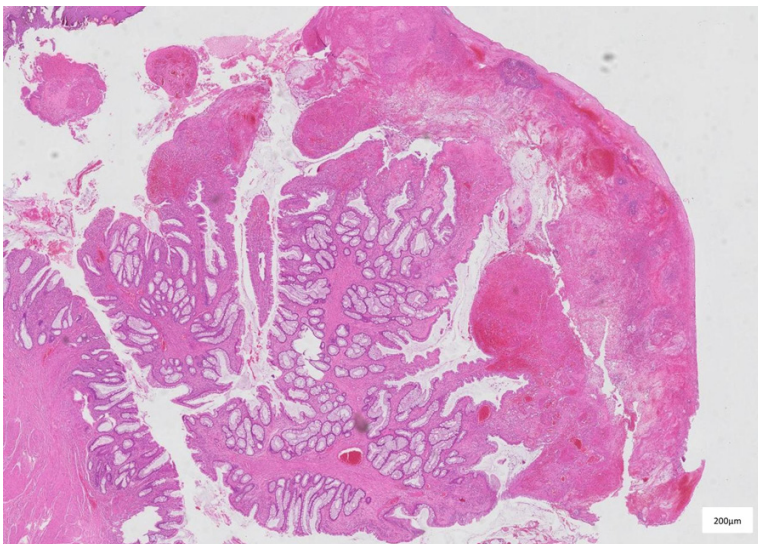
Solitary rectal ulcer syndrome is an uncommon gastrointestinal disorder in children but occurs more commonly in adult patients. The first clini-

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**Table 2.** Histopathologic features of patients with SRUS in the first histologic examination (n=21)

| Patient number | LP fibromuscular hyperplasia | Thickening, splaying & vertical extension of MM | Crypt abnormalities | Villiform surface  | Ulceration           |
|----------------|------------------------------|---|---------------------|--------------------|----------------------|
| 1              | +                            | MM not included                                 | +                   | -                  | -                    |
| 2              | +                            | +   | +                   | +                  | -                    |
| 3              | +                            | MM not included                                 | -                   | -                  | +                    |
| 4              | +                            | MM not included                                 | -                   | -                  | -                    |
| 5              | +                            | MM not included                                 | +                   | -                  | +                    |
| 6              | +                            | -   | -                   | +                  | +                    |
| 7              | +                            | +   | +                   | -                  | -                    |
| 8              | +                            | MM not included                                 | +                   | -                  | +                    |
| 9              | +                            | +   | +                   | -                  | -                    |
| 10             | +                            | +   | -                   | -                  | -                    |
| 11             | +                            | MM not included                                 | +                   | +                  | -                    |
| 12             | +                            | +   | +                   | -                  | +                    |
| 13             | +                            | MM not included                                 | +                   | -                  | +                    |
| 14             | +                            | +   | +                   | -                  | +                    |
| 15             | +                            | MM not included                                 | +                   | -                  | -                    |
| 16             | +                            | +   | +                   | -                  | -                    |
| 17             | +                            | MM not included                                 | -                   | +                  | +                    |
| 18             | +                            | MM not included                                 | -                   | -                  | -                    |
| 19             | +                            | +   | -                   | +                  | -                    |
| 20             | +                            | +   | +                   | -                  | -                    |
| 21             | +                            | MM not included                                 | -                   | -                  | +                    |
| Total, n (%)   | Positive N=21 (100%)         | Positive n=9 (42.8%)<br>Negative n=1 (4.7%)     | Positive n=13 (61%) | Positive n=5 (23%) | Positive n=9 (42.8%) |

LP = lamina propria, MM = muscularis mucosae.



**Figure 4.** Low power overview of one of the cases showing a polypoid architecture of the lesion with surface erosion and overlying fibrinopurulent exudate. The crypts are distorted, hyperplastic, and elongated with some showing focal dilation near surface (hematoxylin and eosin stain, 40× magnification).

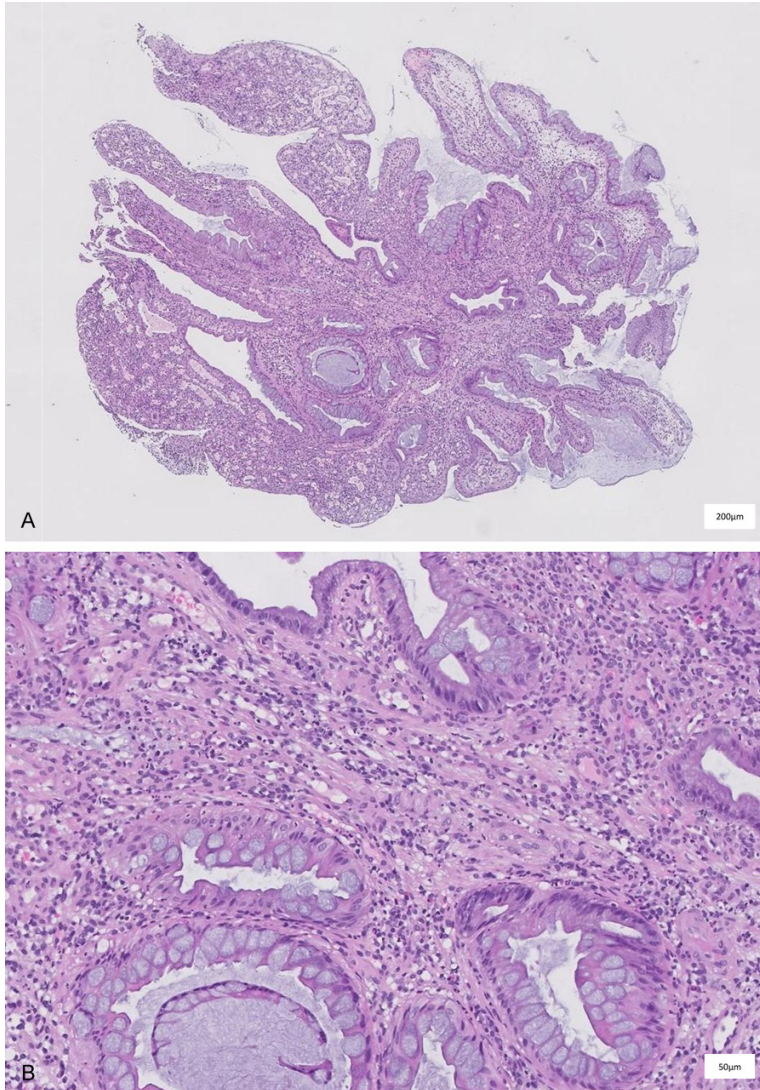
was introduced. In 1969, a more comprehensible description of the disease was reported, in the context of 68 adult cases, where the definition of “solitary rectal ulcer syndrome” was proposed, and the term has broadly retained its meaning until the present day [18].

The exact etiology of SRUS is not known, but it seems to involve a number of mechanisms. Direct mucosal trauma or local ischemia are the most accepted theories, causing trauma inflicted by defecation straining [19]. This may occur during descent of the perineum with abnormal contraction of the puborectalis muscle during straining on defecation, resulting in traumatic

copathologic description reported by Lloyd-Davies, when the term “solitary rectal ulcer”

compression of the anterior wall of the anal canal and development of internal intussuscep-

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**Figure 5.** A: The lesion displays a villiform surface architecture (hematoxylin and eosin stain, 40× magnification). B: A close look at another case illustrating fibromuscular obliteration of the lamina propria (hematoxylin and eosin stain, 200× magnification).

tion or prolapse of the rectum [6, 20-22]. Studies of anorectal physiology have confirmed that uncoordinated defecation with excessive straining over time plays an important role in the development of SRUS. Furthermore, self-induced trauma to the rectum by digitation to remove the impacted stool in cases of constipation, or during attempted reduction of rectal prolapse may cause direct trauma to the rectal mucosa [20, 21].

Of the total cohort in the present study, 81% were males, with a male to female ratio of 4.3:1. This degree of male predominance has

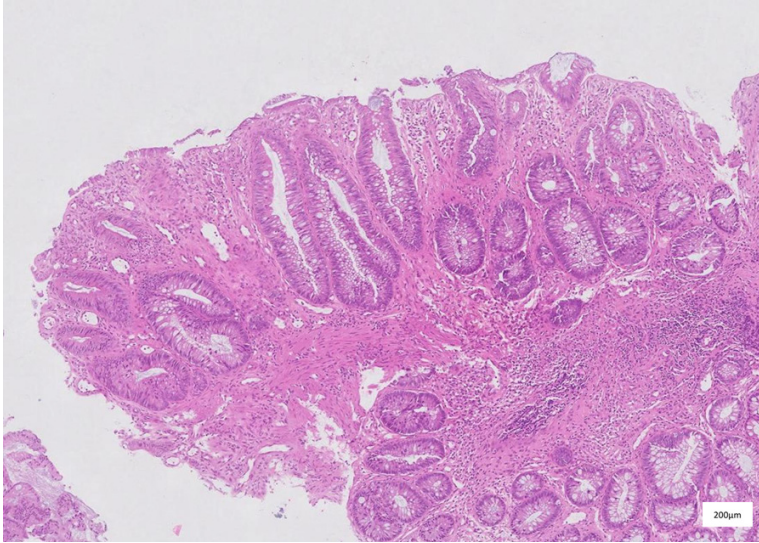
been found in other studies [3]. Anjum et al. reported male gender in 76.19% of patients, with a male to female ratio 3.2:1 [23], and Dehghani et al. reported male gender in 74.5% of patients, with a male to female ratio of 2.9:1 [16]. However, this male gender predominance has been observed only in pediatric studies; studies in adults report either equal gender distribution or a small predominance of the female gender [5].

The youngest patient included in the present study was 5.43 years, while the oldest was 17.9 years, with a median age of 11.4 years. Other case series reported children with SRUS presenting at a significantly younger age than those in the present study [12, 23]. Both studies concerned the reported occurrence of SRUS in a child of approximately 1.5 years.

In the present study, the first and most common presenting symptoms were rectal bleeding, followed by passage of mucus, which is in accordance with the findings of Kowalska-Duplaga et al. [10]. Poddar et al. [17] reported rectal bleeding in 93.6% of

cases (64.3% bleeding alone and 29.3% with mucus) and passage of mucus in 5% [17]. Further reported symptoms in the present study were abdominal pain and constipation, but obtaining history of dyssynergic defecation, such as straining, prolonged sitting on the toilet, and manual evacuation of stools, was difficult, perhaps owing to fear of social stigmatization. Approximately half of the patients in the present study (n=10, 47.6%) reported the use of squat toilets. Defecation in the squatting position may force the anterior rectal mucosa into the anal canal, instigating local strangulation, which can lead to congestion, edema, and

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**Figure 6.** In this case the thickened muscularis mucosae with splayed fibers is apparent (hematoxylin and eosin stain, 40× magnification).

ulceration. The use of this method of defecation could be a contributing factor to the increased occurrence of SRUS in children of the Middle East and South Asia [24].

Anemia may develop in children with SRUS as a result of continuous rectal bleeding, especially in those with delayed diagnosis. The degree of severity of anemia depends on the amount of bleeding, the disease duration, and other local factors related to the vascularity of the mucosa in the rectum. The need for blood transfusion in acute hemorrhage has been reported by some previous studies [25, 26]. The bleeding may be profuse enough to require an emergency endoscopy to control the bleeding area [27]. Almost one-third of patients in the present cohort had a hemoglobin level less than 11 gm/dL; two patients had a hemoglobin level less than 6 gm/dL, which required a blood transfusion. The remaining patients were treated with iron supplements.

A single ulcer was the most common form of SRUS reported in the literature [12, 16, 17, 23]. Almost one-third (n=7) of the present cohort presented with a single ulcer. This is often described as a shallow ulcerated lesion on a hyperemic surrounding mucosa, generally located on the anterior wall of the rectum [28]. Multiple ulcers may present in some patients [29]; indeed, the present study found multiple ulcers in 6 patients. Occasionally, SRUS may present with an ulcerated mass (polypoid

SRUS), which can mimic tumors or rectal sessile polyps [14].

Histopathologic examination of the rectal mucosal biopsy is required to confirm the diagnosis of SRUS and to exclude alternative diagnoses, such as IBD, rectal polyps, or tumors. The presence of fibromuscular obliteration in the lamina propria is the cornerstone of a diagnosis of SRUS [30]. Other important features associated with this syndrome is hypertrophied muscularis mucosae with extension of muscle fibers upward between the crypts, which cannot be seen in small

or superficial biopsy specimens, in addition to granular crypt abnormalities. The presence of surface erosion, mild inflammation, distorted crypts, and reactive epithelial atypia may create some conflation with the features of IBD [31]. However, the presence of diffuse collagen deposition in the lamina propria and abnormal smooth muscle fiber extensions are reliable features that distinguish SRUS from other possible diagnoses [32].

The management of SRUS is sometimes challenging because of a lack of standardized treatment protocol and the often-reported unfavorable response to various treatment modalities [24, 33, 34]. It is recommended to reassure patients of the benign nature of the condition, and to motivate patients to regulate toilet habits and avoid straining during defecation. In the present study, 58.3% of patients reported symptom relief with sucralfate enema, either alone or in combination with other modalities. This finding is consistent with the results reported in Dehghani et al. [24]. Pharmacological options for the treatment of inflammatory bowel disease, such as sulfasalazine and corticosteroids, have been reported with variable success [35, 36]. Furthermore, a report on the application of argon plasma coagulation (APC) in the management of SRUS in randomized controlled clinical trials was recently published, showing promise in comparison with conventional therapies [37].

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**Table 3.** Endoscopic findings of patients with SRUS and treatment response

| Patient | No. of endoscopic examination | 1 <sup>st</sup> Colonoscopy  | Rx regimens                              | Last colonoscopy        | Symptoms' relief |
|---------|-------------------------------|--|--|-------------------------|------------------|
| 1       | 10                            | Single ulcer   | Mesalamine + budesonide/Sucralfate enema | Multiple small ulcers   | YES              |
| 2       | 6                             | Polypoid mass  | Mesalamine + Sucralfate enema            | Polypoid mass           | NO               |
| 3       | 5                             | Multiple ulcers  | Sucralfate enema                         | Multiple small ulcers   | YES              |
| 4       | 4                             | Single ulcer   | Sucralfate enema                         | Hyperemic mucosa        | YES              |
| 5       | 7                             | Multiple ulcer   | Mesalamine + Sucralfate enema            | Hyperemic mucosa        | YES              |
| 6       | 1                             | Single ulcer   | Sucralfate enema                         | -                       | NO               |
| 7       | 7                             | Single ulcer   | Mesalamine + Sucralfate enema            | Hyperemic mucosa        | NO               |
| 8       | 8                             | Single ulcer   | Mesalamine + budesonide/Sucralfate enema | Hyperemic mucosa        | YES              |
| 9       | 5                             | Single ulcer   | Budesonide enema                         | Single ulcer            | NO               |
| 10      | 6                             | Multiple ulcers  | Mesalamine + budesonide/Sucralfate enema | Hyperemic mucosa        | YES              |
| 11      | 2                             | Ulcerated polypoid mass  | Mesalamine + Sucralfate enema            | Ulcerated polypoid mass | YES              |
| 12      | 2                             | Hyperemic mucosa   | Stool softeners alone                    | Multiple ulcers         | NO               |
| 13      | 1                             | Ulcerated polypoid mass  | Mesalamine + Sucralfate enema            | -                       | NO               |
| 14      | 2                             | Multiple ulcers  | Budesonide enema                         | Multiple ulcers         | NO               |
| 15      | 6                             | Polypoid mass  | Mesalamine + budesonide enema            | Polypoid mass           | NO               |
| 16      | 1                             | Hyperemic mucosa   | -  | -                       | -                |
| 17      | 1                             | Single ulcer   | Stool softeners alone                    | -                       | NO               |
| 18      | 1                             | Multiple ulcers  | Mesalamine                               | -                       | NO               |
| 19      | 3                             | Hyperemic mucosa   | Mesalamine + budesonide enema            | Polypoid mass           | NO               |
| 20      | 1                             | Multiple ulcers  | -  | -                       | -                |
| 21      | 1                             | Polypoid mass  | budesonide enema                         | -                       | NO               |
| TOTAL   | 79                            | Single ulcer n=7 (33.3%)<br>Multiple ulcers n=6 (28.5%)<br>Polypoid mass n=5 (23.8%)<br>Hyperemic mucosa n=3 (14%) |  |                         |                  |

**Table 4.** Therapeutic modalities applied in the management of SRUS in 19 patients

| Treatment  | Number of patients (n) | Percentage (%) |
|--|------------------------|----------------|
| Mesalamine + sucralfate enema                        | n=5                    | 26.3%          |
| Mesalamine + Corticosteroid enema + Sucralfate enema | n=4                    | 21.0%          |
| Corticosteroid enema                                 | n=3                    | 15.7%          |
| Sucralfate enema                                     | n=3                    | 15.7%          |
| Stool softeners alone                                | n=2                    | 10.5%          |
| Mesalamine   | n=1                    | 5.2%           |
| Mesalamine + Corticosteroid enema                    | n=1                    | 5.2%           |

This study may be limited by its retrospective design, a relatively small sample size, and a lack of long-term follow-up, but sheds significant light on the clinical, endoscopic, and histologic characteristics and practiced treatment modalities.

In conclusion, our study of children with SRUS has found rectal bleeding to be the most common presentation. A single ulcer was the most prevalent form of the condition, followed by multiple ulcers and polypoid lesions. A squatting position during defecation was reported in

almost half of patients. Treatment response was variable, but almost half of patients reported symptom relief following treatment.

### Acknowledgements

The authors acknowledge Dr. Trevor Rawbone, Cardiff, UK for English editing and proofreading of the manuscript.

### Disclosure of conflict of interest

None.

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