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Solitary Rectal Ulcer Syndrome in Children

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Abstract

To describe clinical features, demographic data, and complications of the patients with SRUS, which is a rare cause of rectal bleeding in children. Eleven patients diagnosed with Solitary Rectal Ulcer Syndrome (SRUS) were evaluated. The patients assessed by colonoscopy and the biopsies were investigated. The data evaluated in SPSS Program. The exact Method of the Chi-square test was used to compare groups according to qualitative variables. P <0.05 value was considered statistically significant. The most common symptom of the patients was rectal bleeding followed by abdominal pain and constipation. Lesions were mostly ulcerative in the endoscopic examination. There was a statistically significant relationship between the admission symptom and the response to treatment. Patients with abdominal pain and rectal bleeding had poor responses to treatment. In conclusion, SRUS is not uncommon than is thought in pediatric patients with the symptoms of rectal bleeding and constipation. SRUS should be considered in patients with or without rectal prolapse, with any complaints of any lesions in the rectum, hematochezia, and tenesmus.

Keywords: Solitary rectal ulcer, rectal bleeding, constipation

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Introduction

Solitary Rectal Ulcer Syndrome (SRUS) is a rare condition that manifests with rectal pain and bleeding described mostly in young adults.[1] Generally, the presentation complaints are rectal bleeding with pain, constipation, and sometimes diarrhea. Cruveilhier first described it in 1830; later, in 1969, clinical and pathological features were revealed by Madigan and Morson. The underlying pathophysiology is multifactorial, and its annual prevalence is estimated to be 1/100.000.[4] Although the etiology of SRUS is not fully clear, direct trauma (self-digitation) and ischemia are two main mechanisms held responsible in pathogenesis.[3] The most annoying course of SRUS is the difficulty in treatment; experiences have demonstrated that numerous treatment options are inadequate. There are little data on treatment and outcome in children with SRUS. Enema, laxatives, and surgical options have been used in the literature.[4]

Highlights

- Constipation is common in children.
- Solitary Rectal Ulcer Syndrome is a clinicopathological abnormality.
- Solitary Rectal Ulcer Syndrome should be considered in pediatric patients with the symptoms of rectal bleeding and constipation.
- Enema, laxatives, and surgical options are used in the treatment of SRUS.

The present study, aimed to contribute to the literature by evaluating the clinical features, demographic data, and complications of our patients with SRUS, which is a rare cause of rectal bleeding in children.

Material and Method

In this study, SRUS was diagnosed in 16 cases, 11 of which were evaluated in the determination of clinical findings and demographic features. The study was conducted in Erciyes University in 2010 and the enrolled patients had been followed between 1998 and 2007. All the patients were male. There was a fresh blood story on the patients' stools' surface or mixed with the stool. The growth of all patients was average. None had a problem explained by bleeding diathesis, bacterial or parasitic infection, and any systemic disease. A pathologist evaluated biopsies taken from the colon in all patients. The data were assessed on SPSS Program. Average, standard deviation, median, minimum and maximum values were given as descriptive statistics. The Exact Method of Fisher's Exact test was used to compare groups according to qualitative variables. P<0.05 value was considered statistically significant.

Results

The patients' mean age was 12.02±4.57 years (Min-Max: 16 months - 18 years), and all were male. The most common symptom seen in patients was rectal bleeding (100%) followed by abdominal pain (45.5%), constipation (27.3%), and mucus (27.3%) (**Table 1**).

Table 1
Symptoms observed in patients

Symptom	Number (n)	Frequency (%)
Rectal bleeding	11	100.0
Abdominal pain	5	45.5
Constipation	3	27.3
Mucus	3	27.3
Diarrhea	0	0.0

Bleeding tests of all patients were normal. The mean hemoglobin level of patients at the time of admission was 11.31±1.08 (min-max: 9.50-12.80) gr/ dl, MCV value 74.64±8.09 fl (min-max: 58.70-83.30). Iron deficiency anemia was detected in four of the patients (36.4%). In the follow-up, one patient (9.1%) received a blood transfusion.

In the endoscopic examination, 63.6% of the lesions were multiple. Macroscopically, 72.7% were ulcerative, and 27.3% were polypoidal/nodular. Lesions were detected at a distance of 2-10 cm from the anal edge (**Table 2**). All of the biopsies were compatible with SRUS, and none developed malignancy.

There was a statistically significant relationship between the admission symptom and treatment response (p=0.027). Patients with abdominal pain and rectal bleeding had an inadequate response to treatment (**Table 3**).

In the treatment, constipation therapy was used with local sucralfate, local steroid (oral or rectal). Rectal bleeding continued in seven patients receiving treatment, and two patients required surgery for bleeding. Endoscopy was not repeated routinely, except in four patients with ongoing rectal bleeding. Factors affecting the response to treatment in patients are shown in **Table 4**.

Table 3.
Initial symptom - Response to treatment

Symptom	Clinical Answer		P*
	Yes	No	
Rectal bleeding	5	0	0,027
Rectal bleeding + constipation	1	0	
Rectal bleeding + abdominal pain	0	2	
Rectal bleeding + mucus	1	2	

* According to Fisher's Exact test

Table 4.
Factors affecting clinical response in patients

Variables		Clinical Answer		P*
		Yes	No	
Number of lesions	Single	3	1	1
	Multiple	4	3	
Lesion type	Ulcerated	5	3	1
	Ulcer inflammatory / polypoid	2	1	
Treatment	Sucralfate	0	1	0,727
	Mesalazine	1	0	
	Sucralfate, mesalazine	0	1	
	Laxative	1	2	
	Sucralfate, mesalazine, emptying enema	0	1	
	Laxative, emptying enema	2	2	
Iron deficiency	Yes	3	1	1
	No	4	3	

* According to Fisher's Exact test

Table 2.
Endoscopic findings of the patients and the treatments applied

Patient Number	Rectoscopy Findings	Number of Lesions	Lesion Type	Treatment	Surgical	Number of Recurrent Biopsies	Clinical Answer
1	Nonspecific Proctitis	Multiple	Ulcerated	Sucralfate, Mesalazine	NO	-	YES
2	Nonspecific Proctitis	Single	Ulcerated	Laxative	NO	-	YES
3	Nonspecific Proctitis	Single	Ulcerated	Laxative, Emptying enema	NO	-	YES
4	Ulcerated areas at 7-8 cm	Multiple	Ulcerated	Sucralfate	NO	-	NO /EXITUS
5	Active bleeding	Multiple	Ulcerated inflammatory polyp	Laxative, Emptying enema	NO	-	YES
6	Mutual hyperemia, minor erasions	Multiple	Ulcerated inflammatory polyp	Laxative, Emptying enema	NO	-	NO
7	2 cm proximal anterior localized ulcer	Single	Ulcerated	Laxative, Emptying enema	YES	-	NO SURGICAL
8	Ulcer lesion at 7-8cm and 1cm	Single	Ulcerated	Laxative	NO	2	NO
9	3-4 ulcers at 10 cm	Multiple	Ulcerated	Laxative	NO	2	NO
10	Ulcers at 8 cm	Multiple	Ulcerated	Mesalazine	NO	2	NO
11	Hyperemic edema, 5-10 cm edema, some covered with white exudate, one polypoid lesion	Multiple	Ulcerated inflammatory polyp	Sucralfate Mesalazine Emptying enema	YES	3	NO SURGICAL

Discussion

Solitary Rectal Ulcer Syndrome is a collection of clinicopathological abnormalities that express a solitary ulcer or rectal wall thickening in the rectum and is characterized by rectal bleeding, stool with mucous, prolonged straining, tenesmus, and local pain in the perineum. The actual pathogenetic mechanism is not fully understood and is likely multifactorial. However, it is known that there is a defecation defect in patients, and this is thought to occur with two main mechanisms:[5,6] (i) Relaxation defect of the puborectal muscles during defecation (or paradoxical contraction) and (ii) rectal prolapse. SRUS is seen mostly in young adults, and the incidence in women and men is almost equal. SRUS is seen mainly in older children (>10 years of age).[7-9]

The diagnosis of SRUS is based on the findings of manometer and electromyography, clinical features, rectal examination, proctosigmoidoscopy, histological examination, nutritional habits, defecation habit, dynamic MRI, and anorectal functional studies.[4,6] While no symptoms are found in 1/4 of the disease, symptomatic cases typically complain of the feeling of straining during defecation, being in the toilet for a long time, but still not fully emptying.[10] The predominant presentation is rectal bleeding (as in the present study), in a study reported in Iran (the most extensive pediatric series in the literature with 256 rectal bleeding cases), and in another study in which 140 SRUS cases were examined in 2020.[4,7] In our study, the relationship between presentation symptoms and response to treatment was found to be significant. Patients with rectal bleeding alone, constipation with rectal bleeding, and rectal bleeding with mucus-containing defecation responded positively to the treatment. Mucus discharge, tenesmus, perineal/abdominal pain, incomplete defecation sensation, and fecal incontinence are also among the symptoms. While there was no difference between genders in previous studies, the disease was significantly higher in boys.[1,4]

Histologically, in laminated propria, fibromuscular obliteration is characterized by disorientation in muscle fibers. This condition is thought to develop secondary to chronic mechanical and ischemic trauma with intussusception in the rectal mucosa. Although this syndrome is well defined in adults, its pediatric forms are generally not well defined, misidentified, or limited in number.[9,12]

Treatment can be divided into two groups, conservative or surgical.[13,14] In the absence of overt rectal prolapse, high-fluid and high-fiber diet, laxative, and avoiding difficult defecation are the basis of conservative treatment. Sucralfate, salicylate, corticosteroid, sulfasalazine, mesalazine, and topical fibrin concealing agents, are used in medical treatment.[15] Surgical treatment should be considered in the presence of rectal prolapse. We found that the number of lesions, lesion type, treatment type, and iron deficiency presence did not affect the clinical response.

In pediatric case series, laxatives, enemas, and surgical approaches were used more often than behavioral modification as biofeedback therapy teaches how to relax pelvic floor muscles and the external anal sphincter during bowel movements, especially in adults.[16,17] As shown in many other studies, good results cannot be obtained with the disease's current treatment options. In some patient groups, the disease is treated with only fibrin glues, while in others, the patient goes to surgery. Also, there are patients in whom rectal bleeding continues despite surgery. For this reason, it is suggested that some patients can recover on their own.

A study conducted by Dehghani et al.[4] on 12 children recommended avoiding difficulty in defecation, excess fluid, and fiber diet to all patients. They observed that all symptoms improved in seven patients (58.3%) with sucralfate enema. Salicylate enema (50 mg/kg/day six weeks) and corticosteroids were given to those

who did not respond to sucralfate. Three patients were treated with corticosteroids injected around the ulcer during medical treatment and colonoscopy; two were asymptomatic, and one underwent surgery. Two of our seven cases that could not respond to medical therapy were operated on, and these patients were asymptomatic after surgery. In their pediatric SRUS study of 140 cases, Poddar et al.[7] noted that approximately 60 percent of children without significant rectal prolapse showed a clinical response to behavioral change with a corticosteroid enema.

Kırıştıoğlu et al.[1] reported a series of four cases. They used defecation training, laxative, sulfasalazine, and rectal sucralfate in medical treatment. Ertem et al.[6] reported a series of two cases, and one patient who did not respond to medical therapy underwent rectopexy. The patient was asymptomatic 1.5 years after the operation. In our study, patients who underwent surgery were asymptomatic two years after the operation.

Adaptation to treatment is difficult in younger patients, so follow-up of these patients is more critical, and therapy should be more aggressive. Most pediatric patients with SRUS have a satisfying outcome using a simple behavioral modification approach. Continuity of follow-up is vital to reinforce behavioral modifications and can prevent prolonged, treatment-resistant illness into adulthood.[4] Special studies cannot be conducted in many centers, and biofeedback or large intestine training programs cannot be performed in treatment. Treatment success is likely to increase with the use of these treatment options.[14,18] Keshtgar et al.[19] have developed a treatment method by injecting botulinum toxin into the internal anal sphincter of children with chronic idiopathic constipation. In the SRUS cases, external anal sphincter botulinum toxin injection may have a therapeutic role.[20] Due to our hospital's limited treatment options, it was impossible to compare different treatments in our study.

In patients with abdominal pain and stool of eccentric character (soft or hard), the symptoms last longer. In our study, patients with abdominal pain and rectal bleeding had more prolonged symptoms and did not respond to treatment. Constipation is significant, especially for the persistence of the disease.

Conclusion

Contrary to what is stated in the literature, the frequency of SRUS is not low in children who present with fresh rectal bleeding. The reason for this is not known precisely. However, the most common pathogenetic factor is thought to be mucosal injuries associated with rectal prolapse. It requires a good clinician and an experienced pathologist for early diagnosis. Therefore, patients with rectal bleeding should be questioned in terms of SRUS, and SRUS should be considered in the differential diagnosis. SRUS also should be considered in patients with or without rectal prolapse, with any complaints of any non-natural-looking lesions in the rectum, hematochezia, or tenesmus.

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Ethics Committee Approval: The study was conducted in Erciyes University in 2010 and the enrolled patients had been followed between 1998 and 2007. An ethical approval was not necessary for retrospective studies at the time of this study.

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