



## Solitary Rectal Ulcer Syndrome In Children And Adolescents

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### Abstract

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### Introduction

Solitary rectal ulcer syndrome is a rare disorder characterized by nonspecific symptoms, a variety of endoscopic findings and a few key histological features. SRUS has a prevalence of 1 in 100000 adults per year. It is described as a disease of young adults, commonly reported in third decade of life in males and fourth decade in females. Rarely, children and elderly may be affected. (1)

SRUS may present with rectal pain, bleeding, tenesmus, diarrhea or constipation and rectal prolapse. The symptoms of SRUS are nonspecific which makes it difficult to diagnose, especially in children, without a strong clinical suspicion. A few patients may even be asymptomatic. (2) It may be caused by chronic constipation with associated straining and rectal prolapse. Clinically it might simulate Ulcerative colitis, Crohn's disease or neoplastic etiologies. Therefore, endoscopy and biopsy are often needed for a definitive diagnosis of the disease. Several other investigations like MRI, defecography, barium enema is available for diagnosis but not usually preferred. Treatment often focuses on conservative management which stresses on behavioral and diet changes. Further treatment with sucralfate enema, corticosteroids and sulfasalazine maybe given followed by surgical intervention. (3,4)

### Materials And Methods

We report a case series of Solitary rectal ulcer syndrome in 8 children after a retrospective analysis of colonoscopy biopsy specimens which were studied in

our tertiary care center- DMMC, Wayanad. All the cases were diagnosed in our institution, over past 3 years, from January 2022– December 2024. A review of cases submitted to the department of Pathology was done. All cases diagnosed as Solitary rectal ulcer syndrome based on colonoscopy and histopathological examination were selected. The details of age, gender, clinical features with presenting complaint, colonoscopy findings and histopathological examination were collected from the hospital information system. Histopathological evaluation of the archived slides was done in the department of pathology.

### Results

The total number of adolescents who were diagnosed with Solitary rectal ulcer syndrome were 8 during these 3 years. The median age of presentation was 15 years. The patients ranged in age from 10 to 18 years. The male to female ratio was 3:1. The predominant complaint was bleeding per rectum (62.5%). Mucorrhoea was noted as the next common associated symptom (37.5%) followed by altered bowel habits (25%) and abdominal pain (25%). Excessive straining during defecation and tenesmus were present in 25% of children. A clinical suspicion of Inflammatory bowel disease was made in one patient. Another patient was asymptomatic and had undergone screening colonoscopy owing to strong family history of carcinoma colon. The median duration of symptoms from onset to a final diagnosis was 6 months (IQR 2.0-

12.0). Anemia of less than 10g/dl was recorded in one patient. USG abdomen was done in 25% of cases. The findings were unremarkable except for mesenteric lymphadenitis. Colonoscopy was done in all the patients. Colonoscopy revealed that 87.5% of patients had ulcer in the anterior wall of rectum. Most of the ulcers were in the rectum and one in anal canal (14%). The ulcers were 1-1.5cm in size. 12.5% of the cases had an associated nodularity of the mucosa. Internal hemorrhoids (25%) were noted in a few patients. Histopathological examination showed 75% of the cases with fibromuscular hyperplasia and followed by the finding of ulceration (62.5%) and distorted glandular crypts (50%). Hyperplastic muscularis mucosa with splayed muscle fibers (37.5%) was also noted. Ectatic capillaries were identified (37.5%). Occasional biopsies showed granulation tissue with

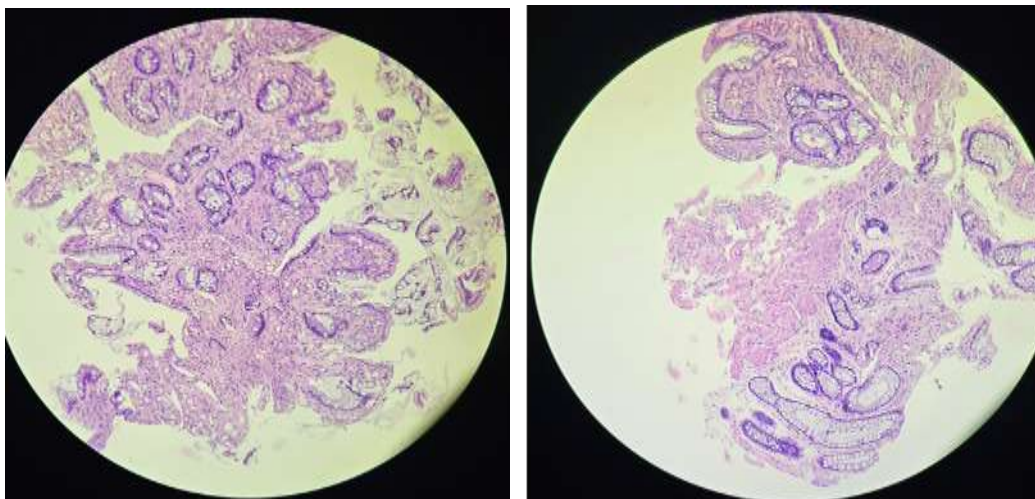
mixed inflammatory infiltrate (37.5%). Characteristic findings of inflammatory bowel disease like crptitis, crypt abscess, crypt distortion or features of chronicity were not identified in any of the biopsies.

All children were advised to have high fiber diet with plenty of fluids. The patients and their families were counselled about the benign nature of the disease. The patients were given toilet training and were put on laxatives. 87.5% of the cases were symptomatically better at one year of follow up. One patient continued to have occasional bleeding per rectum and was referred to biofeedback therapy. The patient with strong family history of cancer was advised to remain under surveillance colonoscopy. None of the patients were managed with surgical interventions.

**Coloscopic findings: 1) Erythematous mucosa with ulceration 2) Rectal ulcer with slough**



**Histological findings: 1) Colonic mucosa with fibromuscular hyperplasia 2) Mucosal ulceration with hyperplastic muscularis mucosa**



Sl No	Age (yrs)	Gender	Presenting symptoms	Colonoscopy findings	Histological findings
1.	13	Female	Bleeding per rectum	Anorectal nodule	Fibromuscular hyperplasia, ulceration
2.	15	Male	Screening colonoscopy	Ulcer in rectum	Hyperplastic mucosa with splaying of muscularis mucosa
3.	14	Male	Bleeding per rectum, mucorrhoea	Ulcer in rectum	Hyperplastic mucosa with splaying of muscularis mucosa, ectatic capillaries
4.	10	Male	Bleeding per rectum, mucorrhoea	Ulcer in rectum	Fibromuscular hyperplasia, ulceration
5.	15	Female	Bleeding per rectum, anemia	Ulcer in rectum	Fibromuscular hyperplasia, ulceration, ectatic capillaries
6.	18	Male	Diarrhoea, abdominal pain, tenesmus	Ulcer in rectum	Fibromuscular hyperplasia, Hyperplastic mucosa with splaying of muscularis mucosa
7.	18	Male	Bleeding per rectum, abdominal pain	Ulcer anal mucosa, internal hemorrhoids	Fibromuscular hyperplasia, ulceration
8.	18	Male	Diarrhoea, mucorrhoea	Ulcer in rectum, internal hemorrhoids	Fibromuscular hyperplasia, ulceration, ectatic capillaries

## Discussion

Cruveilhier (5) published details of four cases of rectal ulcer in 1829 and nearly hundred years later in 1930s Lloyd -Davis described the disease as solitary ulcers of the rectum. Madigan et al (6) and Rutter et al (7) furthered the understanding of the disease in their respective papers. SRUS is a chronic disorder of defecation which is benign in nature. Even though the disease is described as an ulcerative disease, the term is a misnomer. Only 40% of the patients present with ulcerated mucosa and the rest might present with polypoidal or hyperemic mucosa. SRUS is widely described in adults but is often underdiagnosed in children and adolescents.

The pathogenesis of SRUS is poorly understood. It is proposed that the rectal ischemia and pressure due to the paradoxical contraction of the puborectalis pushing the rectum might be the cause of this disease. This trauma to the rectal mucosa leads to ischemia and further ulceration of the rectal mucosa. In the present study the median age of the patients was 15 years with the youngest being the age of 10 years and oldest 18 years. This is in concordance with the study by Perito et al (2) and Abusharifah et al (8). Suresh et al (10) and Kennedy (11) et al noticed a male predominance in the patients

In our series most of the patient presented with bleeding per rectum as in most of the case series. Paradoxically, instead of constipation two of our

patients presented with diarrhea. This was also noticed by Torres et al (9) in their research on the disease. Inflammatory bowel disease can be a differential diagnosis for patients presenting with similar symptoms (12). Hence a complete work up of the patient including colonoscopy and biopsy should be done for a proper diagnosis.

Colonoscopy might show ulcerations which can be solitary or multiple and care often seen in rectum or in the anal verge. The presentation in colonoscopy can also be polypoidal mucosa or erythematous area. (13) In our case series 87.5% cases showed ulceration in rectum. Occasional case showed nodular rectal mucosa.

Defecography, barium enema, MRI are other modalities available for diagnosis. In the present case series these investigations were not preferred. We proceeded directly with biopsy of the mucosa of the affected area. Common histological findings in solitary rectal ulcer syndrome are ulceration with fibromuscular hyperplasia of lamina propria, splaying of muscularis mucosa, branching and distorted crypts. The changes in inflammatory bowel disease like cryptitis, crypt abscess, architectural distortion and features of chronicity are absent in cases of SRUS. (1-10) In our series fibromuscular hyperplasia of lamina propria was the most common finding with ulceration and followed by hyperplastic muscularis with splaying of fibers.

All the patients in this series were managed conservatively with toilet training and dietary changes. High fiber diet and increased fluids were advised. Treatment of SRUS in literature also recommends high fiber diet with significant improvement in symptoms (15) Our patients were advised to refrain from straining during defecation. One patient had refractory symptoms and was recommended for biofeedback therapy. Other treatment modalities with surgical options including excision of the ulcer, rectopexy, repair of puborectalis and colostomy maybe preferred in patients with symptoms refractory to medical management (16). However, none of our patients underwent surgical interventions. Our patients were followed up for 1 year and showed improvement with conservative management.

## Conclusion

Solitary rectal ulcer syndrome is often underdiagnosed in children. Colonoscopy and histological features can clinch the diagnosis and conservative management is effective in children and adolescents. Hence a strong clinical suspicion at an early stage is needed for diagnosis and can significantly improve the symptoms of the patient.

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