Case Report

Solitary Rectal Ulcer Syndrome in a 12-Year Bangladeshi Boy: an Unusual Cause of Rectal Bleeding

Md.Benzamin[1], Kaniz Fathema[2], Dipawnita Saha[3], Bodhrun Nahar[4], Sharmin Akther[5], Khan Lamia Nahid[6].

Abstract

Solitary rectal ulcer syndrome (SRUS) is an uncommon cause of per rectal bleeding in children. Due to it's wide variety of presentation and rarity, it is frequently misdiagnosed as other clinical condition like inflammatory bowel disease, rectal polyp, amoebiasis or malignancy. Here we presenting a case initially misdiagnosed as ulcerative colitis, latter after thorough evaluation diagnosed as a case of SRUS.

Key words: *Solitary rectal ulcer syndrome, Children, Per Rectal Bleeding.*

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Introduction

Solitary rectal ulcer syndrome (SURS) is an benign, rare, rectal disorder in children.[1-4] In 1829, Cruveilhier described four unusual cases of rectal ulcers.[5] The term "solitary ulcers of the rectum" was used by Lloyd-Davis in the late 1930s.[6] The term solitary rectal ulcer syndrome is a misnomer because i) ulcers are found in 40% of patient, others having single or multiple ulcers including hyperemic mucosa to broad-based polypoid or mass lesions ii) 20% of patients have a solitary ulcer, other lesions are different in shape and size and iii) may involve the sigmoid colon.[7,8] Regarding pediatric age group, there are very limited data. From adult study, SRUS is an infrequent, with an estimatedprevalence ofabout 1 in 100000 persons per year.[1] The pathogenesis is incompletely understood. Various factors may be involved in its disease process. Most accepted theories are factors associated with direct trauma or causes of local ischemia.[9]

Case Report

XX, a 12 year old immunized boy, 5th issue of nonconsanguineous parents, presented with painless fresh per rectal bleeding for last 3 years. Bleeding occur drop by drop after defecation or occasionally mixed with stool with variable amount. He also mentioned about something coming out per rectum for last 1 year (Figure 1) He had no history of abdominal pain, fever, tenesmus, constipation, manual disimpaction of stool, diarrhea, joint pain, bleeding manifestation from other site, contact with tuberculosis patient or family history of colonic polyp. But he had history of feeling of incomplete defecation and prolonged straining during defecation. With these complaints he visited to several physicians and underwent colonoscopy for 3 times. Colonoscopy findings was, mucosa of rectum and sigmoid colon looked granular, loss of vascular pattern, covered with whitish mucous like substances, friable and was diagnosed as a case of ulcerative colitis.

1. MD Resident, Department of Pediatric Gastroenterology and Nutrition, Bangabandhu Sheikh Mujib Medical University, Dhaka, Bangladesh.
2. MD Resident, Department of Pediatric Gastroenterology and Nutrition, Bangabandhu Sheikh Mujib Medical University, Dhaka, Bangladesh.
3. FCPS student, Department of Pediatric Gastroenterology and Nutrition, Bangabandhu Sheikh Mujib Medical University, Dhaka, Bangladesh.
4. FCPS student, Department of Pediatric Gastroenterology and Nutrition, Bangabandhu Sheikh Mujib Medical University, Dhaka, Bangladesh.
5. MD Resident, Department of Pediatric Gastroenterology and Nutrition, Bangabandhu Sheikh Mujib Medical University, Dhaka, Bangladesh.
6. Assistant Professor, Department of Pediatric Gastroenterology and Nutrition, Bangabandhu Sheikh Mujib Medical University, Dhaka, Bangladesh.

Correspondence: Md.Benzamin, MD Phase B Resident, Department of Pediatric Gastroenterology and Nutrition, Bangabandhu Sheikh Mujib Medical University, Dhaka, Bangladesh. e-mail: drmd.benzamin@yahoo.com, Mobile: +8801719183948

He was treated with oral prednisolone, oral mesalamine and oral sulfasalazine for variable duration without any improvement. He received 5 units blood transfusion during the course of illness.

On examination, he was found ill looking, severely pale, anicteric, vitals and anthropometry were within normal limit. There was no lymphadenopathy, BCG mark was present. Abdominal examination revealed no organomegaly, no ascites. On perianal region examination showed first degree rectal prolapse and on digital rectal examination no mass lesion or growth found. Other systems examination found no abnormality.

Investigation reports showed severe anaemia (Hb 2.8 gm/dl), inflammatory marker (ESR, CRP, platelet) were normal, coagulation profile, Meckel's scan and serum albumin were normal. Immediate blood transfusion was done and when patient become stable (Hb 12.6 gm/dl) colonoscopy was done. Colonoscopy showed diffuse erythema and erosion in rectal mucosa (Figure 2). Vascular pattern and mucosa of rest of the colon were normal. Multiple biopsieswere taken from lesion and histopathology showed surface erosion and moderate infiltration of chronic inflammatory cells in the lamina propria (Figure 3). Crypts hyperplasia and disarrayed muscularis mucosa were noted. No granuloma or malignancy is seen. From the previous history, colonoscopic findings and histological descriptions, finally we diagnosed this case as a solitary rectal ulcer syndrome. After diagnosis he was treated with behavioral therapy to avoid straining, per rectal hydrocortisone enema for one month and bulk laxative. On follow up after 2 weeks there was no per rectal bleeding for 5 days and he was otherwise stable.

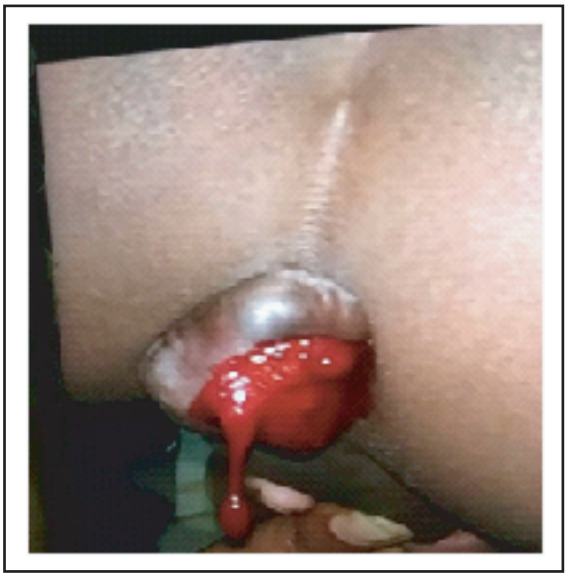

Figure 1: Rectal prolapsed with bleeding.

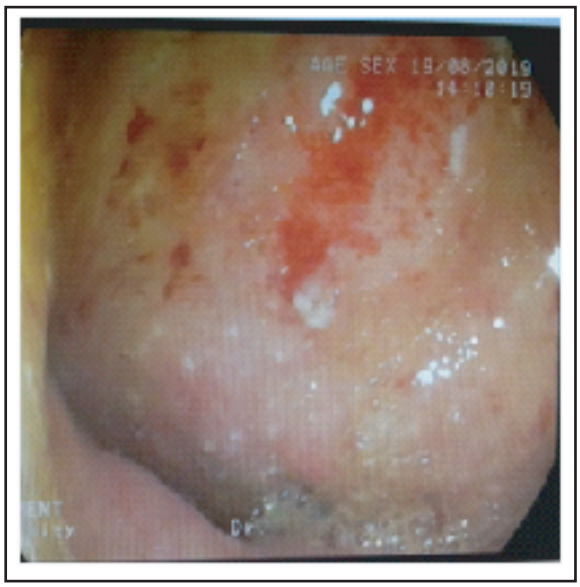

Figure 2: Mucosal erosion

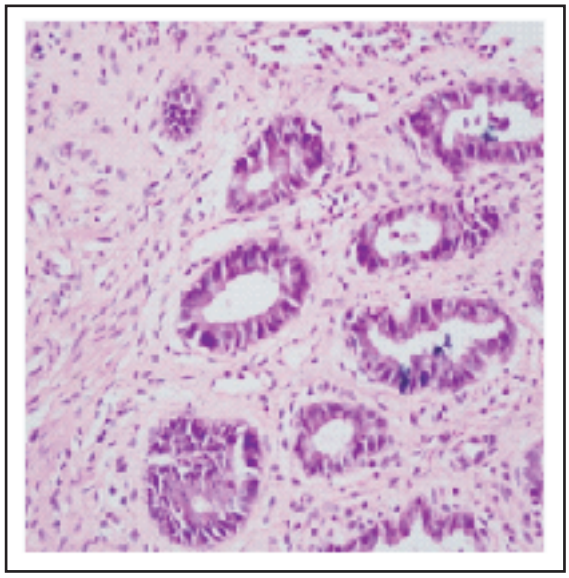

Figure 3: Chronic inflammatory cells in lamina propria and disarrayed muscularis mucosa

Discussion

SRUS have wide range of presentation. Patient may asymptomatic up to 26% cases and symptoms were mostly rectal bleeding (56%), straining with defecation (28%) and pelvic fullness (23%) mucous discharge, incontinence, tenesmus and rectal pain, abdominal pain, constipation, and rectal prolapse, sense of incomplete evacuation.[7,8,10-15] Our case presented with per rectal bleeding, rectal prolapse, prolong straining with defecation and sense of incomplete defecation.

In children median age presentation of is 10 years among them maximum are above 8 years and range from 18 months to 18 years. It has somewhat male predominance with male-female ratio 1.4:1.0.[10] Our case is a male child and his symptoms began at 9 years of age and diagnosed at 12 years.

In pathogenesis of SRUS, trauma or local ischemiaare responsible etiology. Some factors contribute to local trauma or ischemia like (1) prolonged straining during defecation in the patient who suffers from constipation may result in a direct trauma to the mucosa, (2) Direct trauma during attempts at manual disimpaction, (3) Paradoxical contraction of puborec talis muscle (4) Rectal prolapse and intussuscep tions.[9,16,17] Our case had history of prolonged straining during defecation and rectal prolapsed.

Rectal hypersensitivity causing persistent desire to defecate and sensation of incompleteevacuation may also have a role in SRUS.[9] This case had a sense of incomplete defecation.

It often goes unrecognized or easily misdiagnosed with inflammatory bowel diseases (IBD), amoebiasis, malignancy and juvenile polyp.[3,7,18-20] Our case was misdiagnosed as IBD (ulcerative colitis).

Diagnosis of SRUS is done via combination of symptom with colonoscopic and histological findings.[21]

Macroscopic findings on colonoscopy ranges from mucosal erythema to single or multiple ulcers,small or giant ulcers and broadbased polypoid/mass lesions of different sizes.[7,22] Ulcers are usually superficial and 1 to 1.5 cm in diameter, but may 0.5 to 4 cm. Lesions usually are located in the anterior rectal wall within 10 cm of the anal verge, but they can also be located in the anal canal or the sigmoid colon.[7,23] In our case, diffuse erythema and erosion in rectal mucosa was seen.

On histology, fibromuscular obliteration of the lamina propria that leads to hypertrophy and disorganization of the muscularis mucosa, streaming of fibroblasts and muscle fibers between crypts, branching and distorted glandular crypts and diffuse collagen infiltration of the lamina propria are diagnostic finding.[6] In our case, surface erosion and moderate infiltration of chronic inflammatory cells in the lamina propria. Crypts hyperplasia and disarrayed muscularis mucosa were noted.

Treatment of SRUS is difficult and mostly practiced management includes general measures, bio-feedback therapy, pharmacotherapy and surgery for selected cases.[24-29] Topical application of sucralfate enema can be effective for treatment of SRUS in some patients.[30,31] Medications that are useful in inflammatory bowel disease patient also have been tried in thosewith SRUS, such as sulfasalazine and topical glucocorticoids, and mesalamine has been described in small series of patients with varying responses.[18,21,25-27] Our case was treated with behavioral therapy to avoid straining, per rectal hydrocortisone enema for one month and bulk laxative and was improved.

Conclusion

High index of suspicion for the possibility of SRUS in young children is the key for diagnosis. Proper evaluation and treatment give an early recovery of such cases.

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