Case Report

DOI: 10.7860/JCDR/2022/51074.15991

An Unusual Case of Rectal Bleed in an Adolescent: Solitary Rectal Ulcer Syndrome Mimicking Rectosigmoid Malignancy

Internal Medicine Section

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ABSTRACT

Solitary Rectal Ulcer Syndrome (SRUS) is an uncommon rectal disorder. Severe rectal bleeding, anaemia and a mass on evaluation are a rare presentation of SRUS, and can pose as an endoscopic challenge in differentiating from other causes of rectal bleeding. The incidence of SRUS is 1 in 100,000 people per year. Hence, a high degree of suspicion and timely diagnosis is necessary. The present case is about a 17-year-old female who presented with rectal bleeding. Physical evaluation revealed a large polypoidal rectal mass mimicking malignancy which later turned out to be SRUS. The patient improved with medical management. On outpatient follow-up, she had no complaints after a month and follow-up lower endoscopy and imaging later revealed complete healing within three months. Although uncommon, SRUS should be considered as a potential cause of lower gastrointestinal bleeding after sinister causes have been ruled out.

Keywords: Endoscopy, Female, Lower gastrointestinal bleed

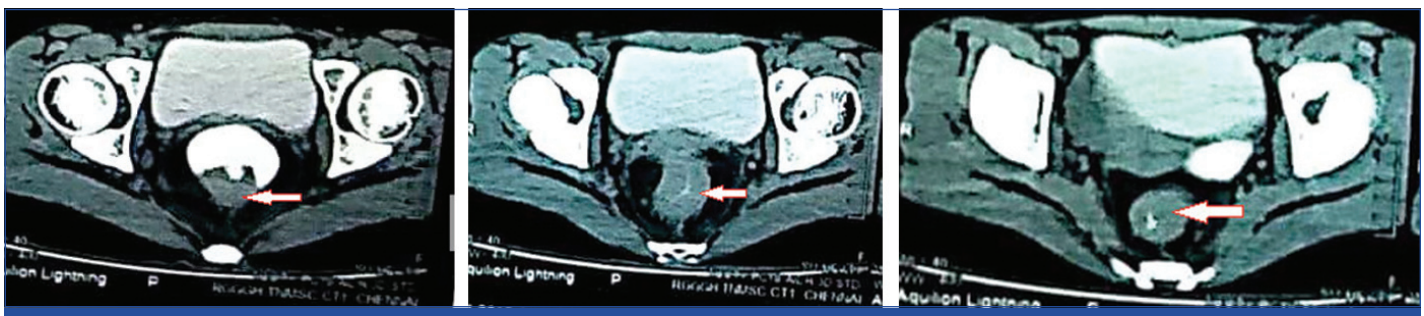
CASE REPORT

A 17-year-old female presented to the Outpatient Department (OPD) with history of bleeding per rectum of six month duration, lower abdominal pain, straining on defecation and occasional episodes of digital rectal evacuation. The patient also complained of easy fatigability and loss of appetite one month prior to admission. There was no history of tenesmus, mass descending per rectum or passage of mucus per rectum. There were essentially no significant systemic symptoms.There was no history of malignancy or chronic illnesses in the family. Examination revealed pallor and tachycardia. Per abdomen examination was normal. Digital rectal examination was painless and revealed a firm nodular swelling on the anterior wall of the rectum, 4 cm from the anal verge. Fresh blood was noticed mixed with stool on withdrawing the gloved finger.

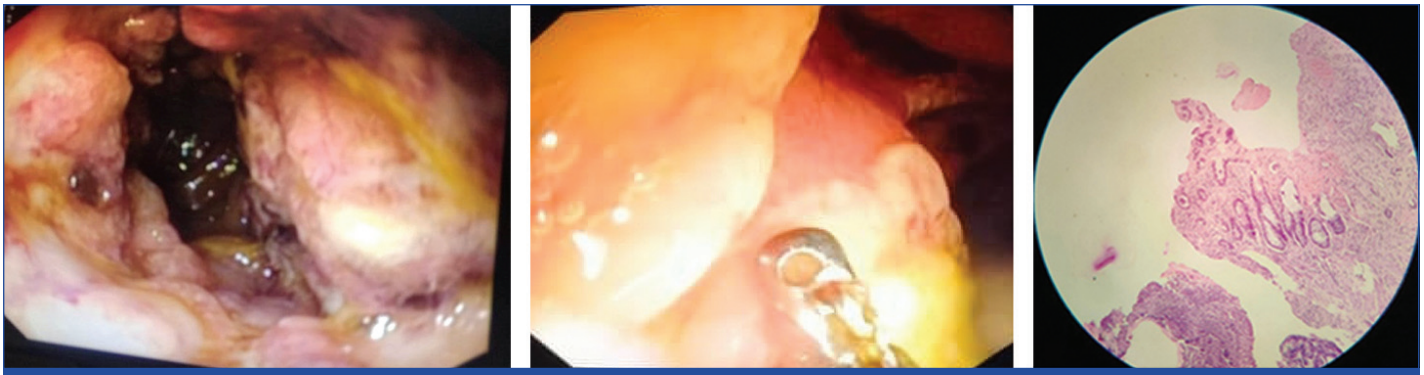
Laboratory investigations showed microcytic hypochromic anaemia (haemoglobin: 5 gm/dL, White Blood Count (WBC): 5540/dL, platelet: 4.94 lacs/dL). Renal and liver function tests were normal. Serum carcinoembryonic antigen level was less than 0.5 ng/mL. Contrast Enhanced Computerised Tomography (CECT) abdomen showed an irregular circumferential enhancing lesion 45×40 mm in size within the rectum extending to involve the rectosigmoid junction. The lesion began 5.4 cm from the anal verge and extended 5.5 cm craniocaudally. The surrounding fat planes were preserved. Enlarged lymph nodes were noticed in the right internal iliac, perirenal, presacral and para-aortic regions [Table/Fig-1]. A malignant lesion involving the rectosigmoid region, stage IIIA (T2N1M0), was considered as a probable diagnosis. Being a young girl with rectal bleeding, benign aetiologies like rectosigmoid polyps, inflammatory bowel disease, SRUS and infectious colitis had to be ruled out. Two units of packed red cells were transfused and haemoglobin of 8 g/dL was achieved and patient was subjected to upper and lower gastrointestinal endoscopy.

Colonoscopy under sedation revealed a circumferential polypoidal ulceroproliferative lesion extending from 5 cm of anal verge to 10 cm [Table/Fig-2,3]. Rest of the colon was normal. Multiple biopsies were taken from the lesion and sent for histopathological studies {Haematoxylin and eosin (H&E) staining method}. Upper gastrointestinal endoscopy revealed pan gastritis [Table/Fig-4,5].

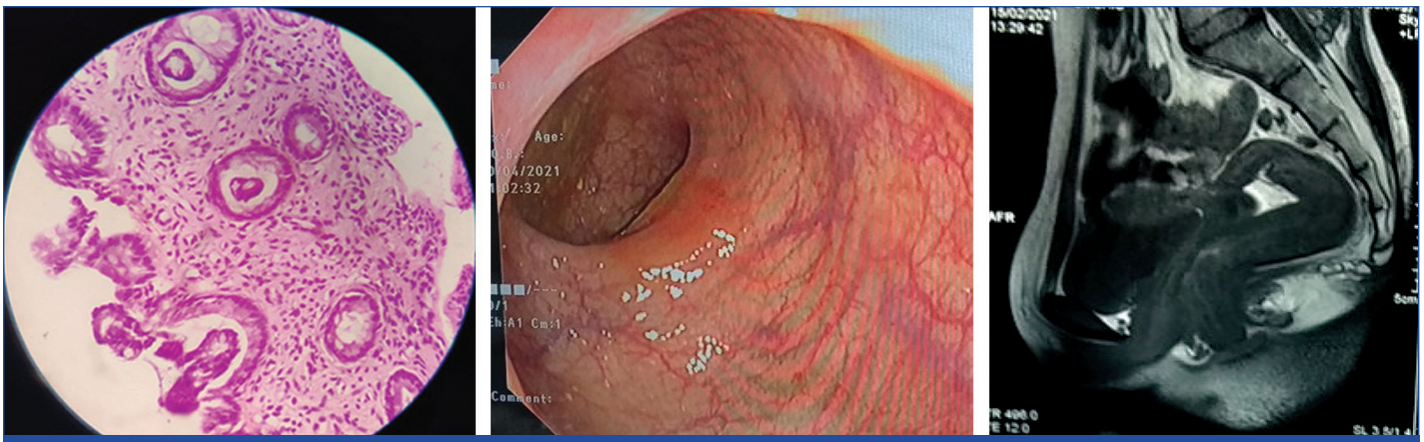
The patient was treated with oral bulk laxative (psyllium powder 10 grams twice a day) and sucralfate enema. Her symptoms got better. Three days later she was discharged and advised high fibre diet, stool softeners and sucralfate suppository. She was also advised to avoid prolonged straining at defecation and rectal digitation. Follow-up in the OPD showed symptomatic improvement after four weeks. Magnetic Resonance Imaging (MRI) of the pelvis was done to assess the lesion after two months. MRI was normal with no residual lesion in the rectosigmoid region. Sucralfate suppository was stopped and she was advised to continue only oral psyllium powder. Sigmoidoscopy done three months after discharge showed complete healing of the lesion, and a normal vascular pattern of the mucosa [Table/Fig-6,7].



[Table/Fig-1]: CECT abdomen showing irregular circumferential mass lesion (arrows) involving the rectum and sigmoid colon. (Images from left to right)



[Table/Fig-2]: Colonoscopy image showing circumferential polypoidal ulceroproliferative lesion in the rectosigmoid area. **[Table/Fig-3]:** A closer view showing biopsy being taken from rectosigmoid lesion during colonoscopy. **[Table/Fig-4]:** Histopathological examination under light microscopy showing breach in the mucosa, obliteration of lamina propria by fibroblasts and muscle fibres, and neutrophilic infiltrates {H&E, low power (10x)}. (Images from left to right)



[Table/Fig-5]: Histopathological examination under light microscopy showing breach in the mucosa, obliteration of lamina propria by fibroblasts and muscle fibres, and neutrophilic infiltrates {H&E, high power (40x)}. **[Table/Fig-6]:** Sigmoidoscopy repeated after three months showing normal mucosal and vascular pattern. **[Table/Fig-7]:** MRI pelvis done after two months showing normal rectosigmoid anatomy. (Images from left to right)

DISCUSSION

The SRUS is essentially a defecatory disorder. The exact aetiology of SRUS is still unknown. It is a misnomer as solitary ulcer of the rectum is seen only in a minority of patients. It is an infrequent and underdiagnosed disorder and has an incidence of 1 in 100,000 individuals per year[1].It is more commonly seen in men in the third decade and in women in the fourth decade[1]. There are case reports of children as young as five years diagnosed with SRUS [2].

The underlying aetiology and pathogenesis of SRUS are not fully understood. Direct trauma or local ischaemia can be the predisposing factors. Mucosal prolapse is the most common mechanism of SRUS[3], which was not the case in the index patient. Abnormal contraction of the puborectalis during straining on defecation could compresses the anterior wall of the rectum leading to venous congestion, oedema and ischaemia leading to ulceration [4]. The history of prolonged defecation and the habit of rectal digitation might be the predisposing factors in the present patient. However, the hypothesis of rectal digitation remains unproven in SRUS.

The clinical manifestations of SRUS may vary from patient to patient. The disease can be asymptomatic in about 25% of the patients. Chong VH and Jalihal A describe rectal bleeding as the most common symptom of SRUS, which can occur in up to 86% of patients. Other symptoms are abdominal pain (36%), mucus per rectum (25%), straining at defecation (25%), diarrhoea (14%) and constipation (14%) [5]. This patient had bleeding per rectum, lower abdominal pain, straining during defecation, digital evacuation and symptoms of iron deficiency anaemia.

The endoscopic spectrum of SRUS was reviewed by Sharara AI et al., [6]. Endoscopic appearance can vary from discrete ulcerations to polypoidal lesions especially on the anterior wall. Polypoidal lesion is seen in 25% of the cases. Lesions are located mostly anteriorly (38.5%) but can also be seen posteriorly (30.7%) or circumferentially (31.8%) with size ranging from 0.5 to 4 cm [5]. Mucosal erythema and multiple lesions are seen in 18% and 30% of the cases, respectively [6]. The circumferential polypoidal presentation in this case was atypical as lesions are usually located anteriorly, within the rectum, and are broad based. The appearances may be confused with an inflammatory polyp, hyperplastic polyps or rectal carcinoma resulting in a delay of diagnosis and treatment.

In an analysis by Blanco F et al., rectal cancer diagnosed and staged in 14 patients by endorectal ultrasonography and MRI were later proven as SRUS by biopsy [7]. The diagnosis of SRUS should be confirmed by histological examination. Classical characteristics include fibromuscular obliteration of the lamina propria, hypertrophied muscularis mucosa with extension of muscle fibres between the crypts, crypt distortion and glandular crypt abnormalities [3]. In this case, a probable diagnosis and staging of rectal carcinoma was initially considered based on CECT findings but the benign aetiology was later proven by histopathology.

The choice of treatment for SRUS depends on the severity of symptoms and whether there is an underlying rectal prolapse. In a systematic review of 20 studies, Gouriou C et al., described the various treatment options for SRUS [8]. Conservative management includes patient education, high fibre diet, bulk laxative agents and topical therapies including steroid, sucralfate and 5-aminosalicylate enemas. Jarrett MED et al. and Rao SSC et al., have described the beneficial role of biofeedback therapy in SRUS [9,10]. Surgical options for treatment of SRUS are reserved for patients who do not improve with conservative measures, and those with rectal prolapse. In a systematic review by Qari Y and Mosli M, medical management resulted in resolution of SRUS in 57% of patients [11]. There is no difference in clinical response between polypoidal and ulcerative lesions, 39% of patients can have persistent bleeding per rectum [5]. This patient became symptomatically better with conservative management (bulk laxative and sucralfate enema). The rectosigmoid lesion resolved over three months which was confirmed radiologically and by colonoscopy.

CONCLUSION(S)

Whenever, a patient presents with rectal bleeding, mucus discharge and history of rectal digitation, the possibility of SRUS should be considered after ruling out sinister pathologies. Appropriate diagnosis can help to avoid unnecessary interventions as most cases resolve with conservative management. In this case, even though malignant aetiology was considered initially based on imaging, conservative management after histological confirmation of SRUS lead to resolution of symptoms. Maintaining a high index of suspicion is thus required with a multidisciplinary approach.

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PLAGIARISM CHECKING METHODS: [Jain H et al.]
- Plagiarism X-checker: Aug 31, 2021
- Manual Googling: Dec 06, 2021
- iThenticate Software: Dec 13, 2021 (15%)

ETYMOLOGY: Author Origin

AUTHOR DECLARATION:
- Financial or Other Competing Interests: None
- Was informed consent obtained from the subjects involved in the study? Yes
- For any images presented appropriate consent has been obtained from the subjects. Yes

Date of Submission: **Aug 29, 2021**
Date of Peer Review: **Nov 01, 2021**
Date of Acceptance: **Dec 11, 2021**
Date of Publishing: **Feb 01, 2022**