

ORIGINAL ARTICLE

CLINICAL SPECTRUM OF SOLITARY RECTAL ULCER IN CHILDREN PRESENTING WITH PER-RECTAL BLEED

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Background: Solitary rectal ulcer syndrome (SRUS) is a benign and chronic disorder well known in young adults and less common in children. The objective of this study was to determine the frequency and clinical spectrum of solitary rectal ulcer in children with bleeding per rectum. **Methods:** This study was conducted in the Department of Paediatric Gastroenterology Hepatology & Nutrition; The Children's Hospital & The Institute of Child Health, Lahore, from January–December 2015. Total 187 children presenting with per-rectal bleeding who underwent colonoscopy were entered in the study. Demographic and presenting clinical features; colonoscopy and histopathology findings were recorded. Data was analysed using SPSS-20. **Results:** Out of a total of 187 children with bleeding per rectum, 21 (11.23%) were diagnosed with solitary rectal ulcer. Males were 15 (71.43%) and females were 6 (28.57%) with age range 8–12 years. Mucus in stool 14 (66.7%), constipation 12 (57.1%) and tenesmus 10 (47.6%) were the most common clinical presentations. Colonoscopic finding are solitary erythematous ulcerative lesion was seen in 8 (38.09%) children, multiple ulcerative lesions in colon 6 (28.57%), multiple ulcerative lesions in rectum 5 (23.81%), polypoidal growth in colon and hyperaemic rectal mucosa in 1 (4.76%) each. Histopathological findings were consistent with SRUS in all the cases. **Conclusion:** The frequency of SRUS was high (19.6%) in patients with per-rectal bleed. Mucus in stool, constipation and tenesmus were the most common clinical presentations. Colonoscopic and histopathological findings were helpful in the confirmation of the underlying aetiology.

Keywords: Solitary Rectal Ulcer Syndrome; Bleeding per Rectum; Constipation; Tenesmus

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INTRODUCTION

Bleeding per rectum in children is a worrisome symptom which can lead to significant parental anxiety. It is a common complaint and accounts for about 10–20% referral to Paediatric Gastroenterology unit.¹ Most of the causes are benign and self-limiting. However, sometimes, lower gastrointestinal bleed may be due to a significant pathology or may lead to life-threatening haemorrhage.

Solitary rectal ulcer syndrome (SRUS) is a chronic but benign condition. It is well recognized in adults but much less common in paediatric age group that is often misdiagnosed. Annual prevalence of SRUS in adult population is estimated to be 1 in 100,000 but its prevalence in children is still unknown. SRUS usually affects adults in the third or fourth decade of life. There is slight female preponderance reported in the literature.² Solitary rectal ulcer in children is usually found between age 2–18 years and more than 80% of children are older than 8 years at presentation.³ Demographic data in paediatric age group is very deficient and limited only to few case reports and case series.

Solitary rectal ulcer has a wide range of symptomatology which may simulate inflammatory bowel disease; infectious colitis; neoplasm and rectal polyp. Bleeding per rectum is the most common

presentation followed by copious mucoid discharge associated with abdominal and perineal discomfort; straining, constipation, rectal prolapse; tenesmus and even diarrhea.⁴ The amount of blood loss varies from little fresh blood to significant haemorrhage that may require blood transfusion.⁵ Up to 20% of patients can be asymptomatic and digital manipulation to assist with a bowel movement is reported in 5–6% of children.⁶

The diagnosis of solitary rectal ulcer relies on colonoscopy and histopathology. The lesion may be solitary, multiple, ulcerative, polypoidal, nodular or erythematous mucosa.⁷ Histopathology is the gold standard for diagnosis. The histological findings include thickened mucosal layer with crypt distortion, hypertrophied muscularis mucosa with extension of muscle fibres in to lamina propria.⁸

Therapeutic regimes include high fibre diet, stool softeners, debulking agents, bowel training, Sucralfate, mezilamine and steroids enemas. Surgery is indicated in patients with persistent bleeding not amenable to treatment and includes rectopexy, excision of ulcer and rarely colostomy.⁹

The data on clinical spectrum and its frequency in children is scarce in Pakistan and limited to few case reports. Therefore, this study was

planned to determine the clinical spectrum and frequency of this rare entity in children in our setting.

MATERIAL AND METHODS

This was a descriptive case series study conducted in The Gastroenterology Department of the Children Hospital and the Institute of Child Health, Lahore, from January–December 2015

Sample size was calculated using WHO sample size calculator, considering anticipated population proportion (P) 7.5%¹⁰ and keeping confidence interval (1 - α) 95% and absolute precision (d) 0.05, sample size (n) was 107 patients. Consecutive, non-probability sampling technique was used. All children between ages of 2–18 years of either sex, presenting with bleeding per rectum having duration of symptoms more than 2 months were included. Critically ill children unfit for colonoscopy or children with some known cause of bleeding per rectum were excluded. Parents unwilling for their children to be included for the study were also excluded. History was taken from the parents and/or patient and detailed clinical examination was done. Data including age, gender, duration of bleeding per rectum; mucous discharge; perineal discomfort, constipation and manual evacuation was recorded. Complete blood counts and stool complete was done from hospital laboratory. Colonoscopy for all patients was done at Gastroenterology Department by a senior Paediatric Gastroenterologist. All biopsies taken were reported by consultant histopathologist. Data was entered in specially designed proforma and analysed by SPSS 20. All children of solitary rectal ulcer were analysed for gender, clinical features, Histopathological findings and aetiology in terms of frequencies and percentages. Quantitative data such as age, haemoglobin was presented as mean and standard deviation in these children.

RESULTS

Out of a total of 187 children with bleeding per rectum, 21 (11.23%) were having solitary rectal ulcer. Other aetiologies of Per-rectal bleeding encountered were Rectal polyp in 115 (61.50%), Ulcerative colitis in 19 (10.16%), Lympho-nodular hyperplasia in 26 (13.90%) and anal fissure in 6 (3.21%) patients.

In the 21 children with SRUS, 16 (76.19%) were male and 5 (23.81%) female. Age range was between 6–15 years while mean age was 10.29±2.3 years (Table-1). Majority of the children (16;76.2%) were between 8–12 years of age.

In children with solitary rectal ulcer, 14 (66.67%) were having mucus in stool, tenesmus 10 (47.6%), abdominal pain 2 (9.5%), perineal discomfort 9 (42.9%), constipation 12 (57.1%),

digital evacuation 4 (19%) and diarrhoea 2 (9.5%). (Table-2) Mean haemoglobin was found to be 8.87±1.24 g/dl in children with solitary rectal ulcer with a range of 5.5–10.7 g/dl. Haemoglobin less than 10g/dl was seen in 18 (85.71%) patients while 02(9.52%) had haemoglobin less than 7g/dl. Only 01 patients required blood transfusion.

Common findings on colonoscopy finding were single erythematous ulcerative lesion in rectum seen in 8 (38.1%) children, multiple ulcerative lesions in colon were seen in 6 (28.6%) while multiple ulcerative lesion in rectum were seen in 5 (23.8%). Polypoidal growth in colon and hyperaemic rectal mucosa was seen in 1 (4.8%) patient each. (Table-3)

Histopathological examination of biopsy specimen revealed crypt distortion, hypertrophied muscularis mucosa and fibromuscular obliteration of lamina propria, and these findings were seen in all cases of solitary rectal ulcer.

Table-1: Age distribution in children with SRUS

Age (years)	Frequency (%age) (n=21)
<8	3 (14.3)
8–12	16 (76.2)
>12	2 (9.5)
Total	21

Table-2: Clinical presentation of children with SRUS

Clinical Presentation	Frequency (%age) (n=21)
Mucus in stool	14 (66.7)
Tenesmus	10 (47.6)
Abdominal pain	2 (9.5%)
Perineal discomfort	9 (42.9)
Constipation	12 (57.1)
Digital evacuation	4 (19)
Diarrhoea	2 (9.5)

Table-3: Colonoscopic findings of children with SRUS

Colonoscopy Findings	Frequency (%age) (n=21)
Single erythematous ulcerative lesion in rectum	8 (38.1)
Multiple ulcerative lesions in colon	6 (28.6)
Multiple ulcerative lesion in rectum,	5 (23.8)
Polypoidal growth in colon	1 (4.8)
Hyperaemic rectal mucosa.	1 (4.8)

DISCUSSION

The Department of Paediatric Gastroenterology, Hepatology and Nutrition is the dedicated teaching institute in the field of Paediatric Gastroenterology, we receive referral from all over the country. Although SRUS is considered a very rare ailment; the substantial number of patients seen can be attributed to a very wide catchment area.

Frequency of SRUS was found to be 11.23% in our study which is higher than study conducted in Southern Iran where 4.7% children had SRUS,

whereas a study from Chennai¹⁰, 7.5% children were having SRUS.

In present study, 76.2% children with SRUS were male and male to female ratio was 3.2:1 this male predominance is in concordance to study conducted by Suresh *et al*¹⁰ where male to female ratio of 2.4:1 was noted.

Mean age at presentation in SRUS was 10.29±2.3 years that is similar to Suresh *et al*¹⁰ who also noted the mean age to be 10.38±2.4 years. The youngest patient with SRUS, reported in literature by Suresh N¹⁰ was a child of 1.5 years. Gabra *et al*¹³ also reported two boys with SRUS of ages 2 and 3 years; while in our study, the youngest patient was 5 years old male.¹⁰

The clinical features of SRUS in present study are largely similar to contemporary literature. All patients had bleeding per-rectum. Most common form of clinical presentation was mucus along with blood in stool (66.7%), followed by constipation (57.1%) and tenesmus (47.6%). Interestingly, a study done in India,¹⁰ the common mode of clinical presentations in children with SRUS were mucus discharge in 77.3%, constipation 63.3% and tenesmus 59% respectively. In our study, abdominal pain and diarrhoea was found in (9.5%) of patients which is lower than study conducted by Abid S who showed abdominal pain in half (49%) and diarrhoea in one fourth (26%) of the patients. Digital evacuation was found in (19%) in our study which is higher than comparable literature. History of digital evacuation in patients with SRUS has also been documented in literature. In a study done by Suresh N¹⁰, 27.2% of patients with SRUS had history of digital evacuation which is in close concordance with the results of present study. History of digital evacuation was less frequently elicited in the study by Abid S¹¹; in which only 8% patients were found to have history of digital evacuation.

In present study, a significant proportion of patients had anaemia with haemoglobin less than 10 g/dl seen in 18 (85.71%) patients. This finding is in slight disagreement with existing literature. In a case series presented by Urganci N¹², only 50% of the patients had anaemia while in study done by Perito ER¹³, only 1 patient out of 15 (6.67%) had anaemia.

Besides, bleeding from solitary rectal ulcer is usually mild and does not lead to profound anaemia; although, cases with severe bleeding per-rectum due to SRUS have been reported in literature. High frequency of anaemia in our children with SRUS could be a reflection of the

poor nutrition status of child population in general with high prevalence of nutrient deficiencies.¹²

Clinical features of SRUS in children are largely comparable to adult patients. In a study done in adult patients with SRUS by Abbasi A, bleeding per rectum (93.2%) and mucus (88.6%) were the commonest symptoms followed by straining (77.3%), constipation (72.7%) and tenesmus (11.4%).¹⁴

Endoscopy is a key investigation in the diagnosis of SRUS. However, endoscopic findings are diverse and variable. It is imperative for the clinician and endoscopist to have high index of suspicion in order to arrive at the diagnosis. Most common endoscopic finding in present study was single erythematous ulcerative lesion in rectum seen in 8 (38.1%) patients. Multiple ulcerative colonic lesions were observed in 6 (28.6%) patients while multiple ulcerative lesion in rectum were seen in 5 (23.8%) patients. Less common findings were polypoidal growth in colon and hyperaemic rectal mucosa seen in 1 (4.8%) patient each.

Wide spectrum of endoscopic findings has been well documented in literature. In a review by Zhu QC, commonest finding is an ulcerative lesion on anterior rectal wall, 3–10 cm from the anal verge. Size of ulcer was between a range of 0.5–4 cm. Lesion varied from mucosal erythema to established ulcer with slough. Polypoidal lesion was present in 25% of patients. Multiple erythematous areas were seen in 18% of patients; while 30% had multiple lesions. These findings are closely concordant with the results of present study.⁵ In a case series of 6 patients by Urganci N¹², 3 (50%) had single ulcerative lesion while 2 (33.33%) had circumferential ulcerated lesion. Polypoidal growth was seen in 1 (16.67%) patient. These results are also loosely concordant with the results of present study. Rare endoscopic presentations have been reported in literature. One such case report describes massive spurting bleed from ulcer crater, successfully treated with argon laser coagulation.¹⁵ Histopathology constitutes the most important step in diagnosis of SRUS. Histopathologic findings were very much consistent in all the patients included in the study. Crypt distortion, hypertrophy of muscularis mucosae and invasion of muscularis in the lamina propria were seen in all the patients. In a study done by Al-Brahim N⁴, most consistent histopathological findings seen in all patients were mucosal serration, lamina propria invasion by fibromuscular tissue and crypt distortion. Emerging diagnostic modalities include endoscopic ultrasound examination or the rectal mucosa.

Characteristic findings include thick hyperechoic mucosa and hypoechoic, thick muscularis propria. However, extensive research is needed until the modality can be recommended for routine diagnostic workup.¹⁶ SRUS is a benign lesion however it is difficult to treat and complete cure is uncommon. Patient counselling and reassurance constitutes an important part of treatment. Treatment aims at achieving clinical as well as microscopic healing. First step of therapy is dietary modification with a high fibre diet and bulk laxatives. Some studies have suggested sucralfate enema as the second step of pharmacotherapy. In patients not responding to these measures, defecography to assess puborectalis muscle dysfunction and occult prolapse of rectal mucosa. Presence of these pathologies makes patients a candidate for bio feed back and surgery respectively. Rectopexy and Delorme's procedures have been found to offer the best results.¹⁷

Pharmacotherapy is considered second line therapy. Sulphasalazine and botulinum toxin injection have been used with acceptable results. Behavioural therapy has been found very beneficial in patients with excessive straining. Bio feed back therapy includes behavioural change with respect to the process of defecation. However poor adherence to the treatment may limit effectiveness. Argon plasma coagulation alone or in combination with other modalities has also shown to be effective in reducing size and depth of solitary rectal ulcer lesion.¹⁸

CONCLUSION

The frequency of SRUS in this study was high (11.23%). Mucus in stool, constipation and tenesmus were the most common clinical presentations. Colonoscopic and histopathological findings were helpful in the confirmation of the underlying aetiology. Children with SRUS must be assessed carefully to define clearly the underlying pathophysiology, and to select the appropriate treatment strategies.

AUTHORS' CONTRIBUTION

MNA: Designed the study, did literature search, data analysis, interpretation of results and wrote the manuscript. HA: Contributed in study design, interpretation of results and supervised writing of manuscript. HS: Contributed in literature search and acquisition of data. MAH: Contributed in statistical analysis of data.

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