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SOLITARY RECTAL ULCER SYNDROME - A RETROSPECTIVE HOSPITAL BASED STUDY IN KERALA - SOUTH INDIA.

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ABSTRACT

Background: Solitary rectal ulcer syndrome (SRUS) is a rare disorder that has a wide spectrum of clinical presentation and variable endoscopic findings. Solitary rectal ulcer syndrome can present with rectal bleeding, straining during defecation, and a sense of incomplete evacuation. The term solitary rectal ulcer syndrome is a misnomer. Endoscopic findings in patients with SRUS, can range from mucosal erythema alone to single or multiple ulcers and polypoid/mass lesions.The aim of this study was to evaluate the variable clinical, endoscopic and histological features of SRUS. **Methods**: Patients diagnosed with SRUS histologically from March 2014 to April 2016 at Medical Trust hospital , a tertiary care referral centre in Kerala, were included in this study. The medical records were reviewed retrospectively to evaluate the clinical spectrum of the patients along with endoscopic and histological findings. **Results:** A total of 37 patients were identified, within the age group (range: 10–80) years, with slight female prepondance, 20 (54%) as compared to males 17(45%). Rectal bleeding was the most common presenting symptom, seen in 30 patients (81%), followed by constipation (72.9%) ,straining at stools (64.8%) and perianal pain in 45.9%.Endoscopic findings revealed solitary and multiple lesions in 27 (79.2%) and 10 (27%) patients, respectively. On the basis of appearance, rectal ulceration was the most common finding seen in 25 patients. (67.5%)., while 9 (24.3%) had polypoidal/nodular lesions. Two (5.4 %) patients had erythematous mucosa only, while one patient had telengectatic spots. Associated conditions were hemorrhoids, in 8 patients (21.6%), hyperplastic polyps 5 (13.5%), adenomatous polyps in 3 (8.1%). Histologically fibromuscular obliteration and crypt distortion were seen in all cases (100%), surface ulceration in 94.5%. Vascular ectasia and mild chronic inflammation was a common findings. **Conclusion:** Solitary rectal ulcer syndrome (SRUS) is an uncommon benign disease, with a wide spectrum of clinical presentation and variable endoscopic findings, therefore early diagnosis requires a high index of suspicion from both the Clinician and the pathologist especially, because the term "solitary rectal ulcer" is a misnomer.

KEYWORDS;- Constipation, rectal, solitary, ulcers.

INTRODUCTION

Solitary rectal ulcer syndrome (SRUS) is a rare benign disorder characterized by a combination of symptoms, endoscopic findings, and histological abnormalities.[1] It was first described by Cruveihier.[2] in 1829, when he reported four unusual cases of rectal ulcers. The term "solitary ulcers of the rectum" was used by Lloyd-Davis in the late 1930s and in 1969 the disease became widely recognized after a review of 68 cases by Madigan *et al.*[3] and few years later, a more comprehensive pathogenic concept of the disease was reported by Rutter *et al.*[4] SRUS is an infrequent and underdiagnosed disorder, with an estimated annual prevalence of one in 100000 persons. It is a disorder of young adults, occurring most commonly in the third decade in men and in the fourth decade in women. Men and women are affected equally, with a small predominance in women.[5] However, it has been described in children and in the geriatric population.[6] Solitary rectal ulcer is a misnomer because ulcers are found in 40% of patients, while 20% of patients have a solitary ulcer, and the rest of the lesions differ in shape and size, including hyperemic mucosa to broad-based polypoid lesions.[7] There is even a suggestion that the disease process also may involve the sigmoid colon.[8] In addition, the etiology is not known but may involve a number of mechanisms. For example, ischemic injury from pressure of impacted stools and local trauma due to repeated self- digitation may be contributing factors.[9]

Tjandra and colleagues reported the largest series of SRUS in the literature from Cleveland Clinic.[7] Rectal bleeding, constipation, and straining at stool was the most common presentation, the combination of these symptoms suggesting a local disease in the rectum. However, 20% of the patients presented with diarrhea and 26% were asymptomatic. The characteristic histological features include surface serration, fibromuscular obliteration, and crypts' distortion.[10-12] In addition, different vascular changes, such as ectasia, congestion, and hyalinization, can be seen.[13] It is important to note that these features are not pathognomonic of SRUS. This wide spectrum of clinical , endoscopic findings, and histological features make SRUS a great mimicker of other serious conditions, including adenocarcinoma , inflammatory bowel disease, dysplasia, and adenomatous polyp.[14]

To the best of our knowledge the data on clinical and endoscopic spectrum of SRUS is scarce in this region of the world. Therefore, we carried out this study to deduce the clinical and endoscopic spectrum in patients with SRUS in our setting and to increase the awareness of clinicians and Pathologists regarding this entity as it is a great mimicker of other conditions and ours is the first study of this entity reported from Kerala, south India.

MATERIALS AND METHODS

A Retrospective hospital based study, was conducted at Gastroenterology Department, Medical Trust hospital, a tertiary referral centre , in Kochi - Kerala over a period of 2 years , from March 2014 – April 2016, following its approval by the Institutional Review Board and ethical committee. The study subjects included male and female patients of all ages, who were admitted in our Gastro units with clinical, endoscopic features & histologically confirmed cases of SRUS . The medical record of patients diagnosed with SRUS were retrieved from the electronic database of Medical trust hospital . We analyzed only those patients whose complete records (clinical notes, endoscopic findings and histology) were available. Diagnosis of SRUS was based on characteristic endoscopic and histological findings. Lesions on endoscopic findings were divided on the basis of numbers, as solitary or multiple and on the basis of appearance as ulcerative, polypoidal /nodular or erythematous mucosa only. The histological criteria included fibromuscular obliteration, surface ulceration, crypts and mucosal gland distortion and hyperplasia which may lead to polypoidal appearance.

RESULTS

A total of 37 cases, were evaluated within the the age group (range: 10–80) years for clinical, endoscopic and histological findings that had the complete information available in their medical records. Most of them were females, 20 (54%) as compared to males 17 (45%)**.** Majority of patients,20 (54%) were in the age group of 18-60 years .(Table 1).

Rectal bleeding was the most common presenting symptom, seen in 30 patients (81%), followed by constipation (72.9%), straining at stools (64.8%) and perianal pain in 45.9%, Mucus per rectum (40.5%) . Manual digital evacuation was reported in 12 (32.4%) patients and in 13.5% patients, were an incidental findings. (Table 2).

Endoscopic findings revealed solitary and multiple lesions in 27 (79.2%) and 10 (27%) patients, respectively. On the basis of appearance, 25 (67.5%) of the lesions were ulcerative, while 9 (24.3%) were polypoidal/nodular, Two (5.4%) patients had erythematous mucosa only, while one patient had telengectatic spots. Associated conditions were hemorrhoids, in 8 patients (21.6%), hyperplastic polyps 5 (13.5%) and adenomatous polyps in 3 (8.1%). (Table 3).

Histologically fibromuscular obliteration of lamina propria and crypt distortion were found in all cases (100%). surface ulceration in 94.5%. Vascular ectasia and mild chronic inflammation was seen in 72.9% and 48.6%. (Table 4).

Table 1: Demographic characteristics of patients with SRUS (n=37) .

Age (years)	Number of patients	Percentage (%)
< 10	0	0.0
10 -18	11	29.7
18- 60	20	54.0
60-80	6	16.2
Sex		
Male	17	45.9
Female	20	54.0

Table 2 ;- Symptomatology of patients with SRUS (n = 37) .

Presenting symptoms	Number of patients	Percentage (%)
Bleeding per rectum	30	81.0
Mucus per rectum	15	40.5
Constipation	27	72.9

Straining at stools	24	64.8
Perianal pain	17	45.9
Tensmus	3	8.1
Altered bowel habits	9	24.3
H/o Self digitation.	12	32.4
Asymptpmatic (incidental finding)	5	13.5

Table 3 ;- Endoscopic Findings of patients with SRUS (n=37).

Endoscopic findings	Number of patients	Percentage (%)
Solitary lesion	27	72.9
Multiple lesions	10	27.0
Ulcerative	25	67.5
Polypoid / Nodular lesion	9	24.3
Erythematous mucosal patch	2	5.4
Telengectatic spots	1	2.7
Hemorrhoids	8	21.6
Hyperplastic polyp	5	13.5
Adenomatous polyps	3	8.1

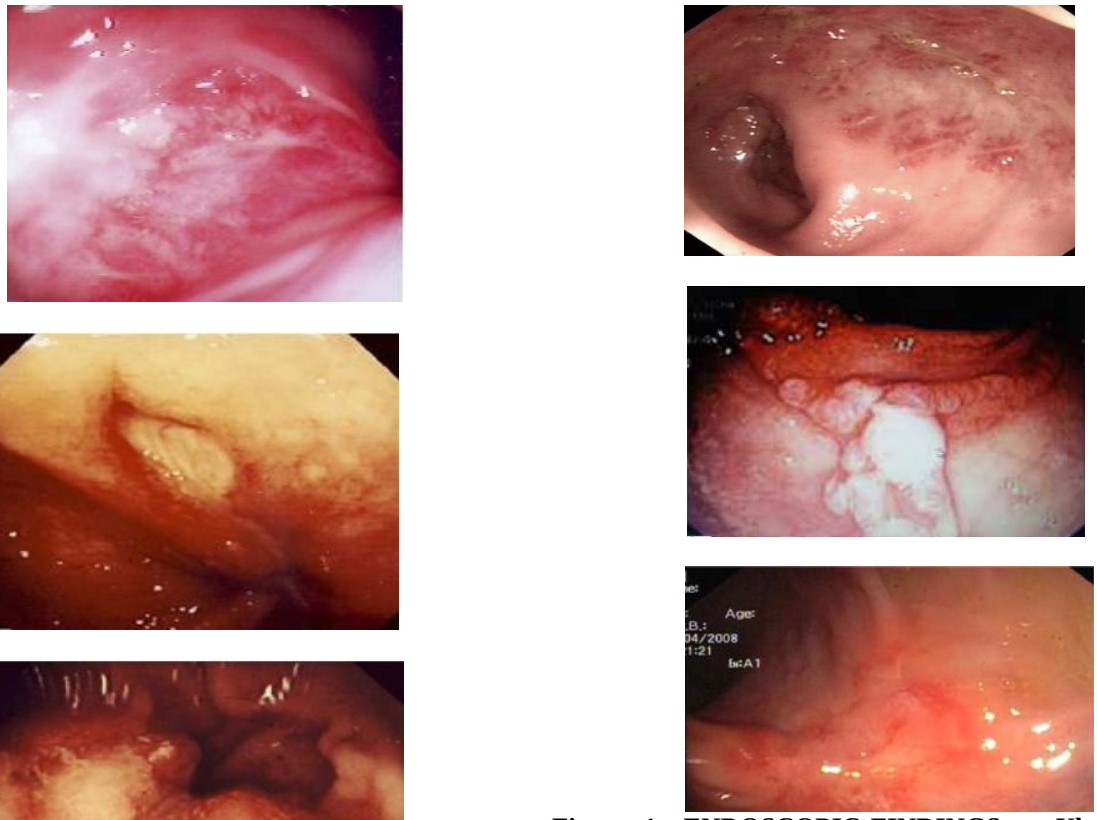
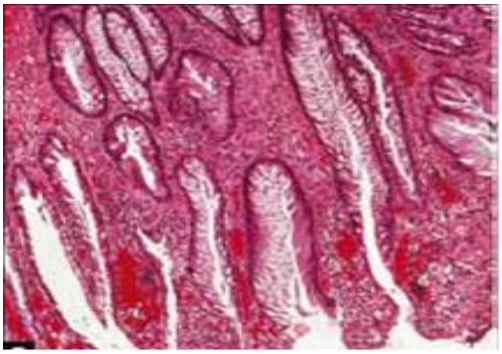


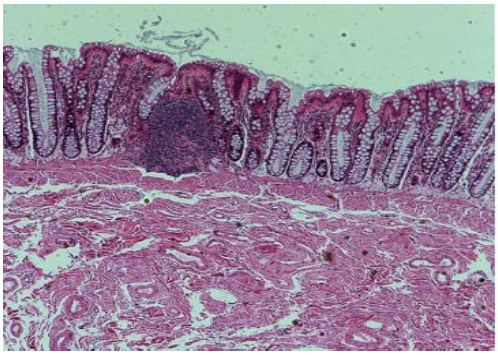
Figure .1;- ENDOSCOPIC FINDINGS ; - Ulcerated variant of SRUS. (A). Flat shallow ulcer with abundant mucus & hyperemic base. (B). Punched out isolated ulcer in base of the rectum. (C). Large shallow ulcer with multiple adjacent areas of erythema, edema & superficial ulcerations. (D). SRUS, as multiple telengectatic spots (E). SRUS, presenting as a large polypoidal /Nodular lesion. (F) SRUS, as erythematous rectal mucosa

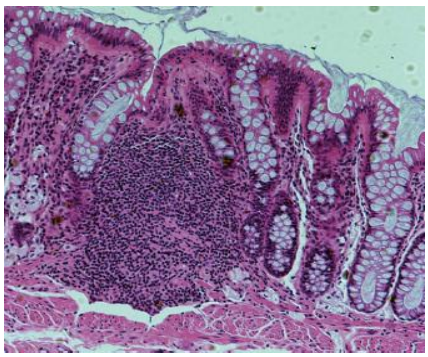
Table 4 ;- Histological findings of patients with SRUS (n = 37) .

Histological finding	Number	Percentage (%)
Fibromuscular obliteration	37	100
Surface ulceration	35	94.5

Crypts distortion	37	100
Mucosal glands distortion	20	54.o
Crypts hyperplasia	10	27.0
Vascular changes		
Ectasia	27	72.9
Congestion	10	27.0
Thrombosis	0	0
Hyaline changes	0	0
Inflammation		
Acute	3	8.1
Chronic	18	48.6
Mixed	6	16.2
None	10	27.0







Figures. 2 ;- Histological features of SRUS: (A) surface serration with fibromuscular obliteration and crypts' distortion. (B) Histology (hematoxylin and eosin) shows smooth muscle hyperplasia in the lamina propria between colonic glands and surface ulceration with associated chronic inflammatory infiltrates. Magnification: × 40 (Figure B), × 100 (Figure C).

DISCUSSION

SRUS is a chronic disorder which can present with diverse endoscopic findings. Since diagnosis on the basis of clinical symptoms alone is difficult, it is imperative for the clinicians to keep this entity in their differentials on endoscopic examination to reach the correct conclusion. Incidences of under and mis-diagnosed cases have been reported in literature.[15,16-18]. Much of the lapse in diagnosis is ascribed to the lack of familiarity of clinicians with endoscopic revelations and the actual condition associated with SRUS. A typical solitary rectal ulcer is a shallow based ulcerating lesion encircled by hyperemic mucosa.[19, 20]. This study to the best of our knowledge is the first study of patients with SRUS in Kerala –South india . It met with intriguing diversity in the appearance of these lesions from being plain ulcerative to polypoidal and from presenting as an erythematous mucosa to multiple ulcerative lesions. Other interesting findings included multiple telengectatic spots, an erythematous mucosal patches and in some asymptomatic patients, SRUS was an incidental finding.

The findings in our study correspond with the literature in terming SRUS as a misnomer. In our Study, there was a slightly higher proportion of female patients. Tjandra etal and Marchal etal in their studies have also demonstrated similar, a slight female preponderance.[1,11]. In our study, there was a wide age range (10-80 years). The series from Cleveland Clinic.[7] demonstrated a similar wide age range of 14-76 years as also did another study by Marchal et al., .[21] which reported an age range of 25-86 years. The triad of rectal bleeding, constipation, Straining at stools and perianal pain was the most common finding. Rectal bleeding and constipation were the most common presentation in other series also.[7–11,21] The bleeding is likely due to ulceration of the mucosa. Another possibility is that the bleeding was due to associated conditions, such as hemorrhoids (which was seen in eight of our patients) or diverticular disease.[11] In our Study almost 5 (13.5%) patients were asymptomatic, where SRUS was diagnosed incidentally during colonoscopy done for cancer screening or polyp surveillance. Tjandra reported that 26% of his series were asymptomatic.[7] Rectal digitations and self-inflicted injury have been claimed to contribute to rectal

injury,[22] and this has been reported in up to 28% of the patients in some other series.[8] . However in our study history of self digitation was seen in 32.4% patients.

Colonoscopic findings are important for the diagnosis of SRUS. Twenty five (67.5%) of the patients in our study had rectal ulceration & 24.3% had polypoid lesion. Solitary and multiple ulcers were seen in 72.9% and 27% respectively. The lesions were 7-20 cm from the anal verge. Thus, it is obvious that the designation 'solitary ulcer' is a misleading. Other series, such as the one published by Torres et al.,[23] had almost similar findings, with 65.3% of the patients reported to have ulceration. In contrast, Tjandra et al.[7] reported that 29% of their series had ulcers and 44% presented with polyps. Tendler et al .[11] reported that all of his 15 patients had polypoid lesions. Based on these findings, it is obvious that all kinds of rectal lesions can be expected in patients with SRUS, from mild erythema of the mucosa to a solitary ulcer, multiple ulcers, and polyps. Clinicians should be aware of this fact and, in the right clinical setting, should consider SRUS in the differential diagnoses of all kinds of rectal lesions.

Histopathological examination is the key to the diagnosis of SRUS. A combination of fibromuscular obliteration of the lamina propria, crypts' distortion, and surface serration can establish the diagnosis in most cases**. (Figure 2).**

In our study, fibromuscular obliteration and crypt distortion were seen in all cases (100%), surface ulceration in 94.5%. Other authors have also reported that these features are the most common. In the series reported by Tendler et al.[11] crypts' distortion and surface serration was seen in 100% of cases, and fibromuscular obliteration of the propria was seen in 93% of the cases. These changes are seen due to ongoing degenerative–regenerative process occurring in the mucosa. It should be mentioned that these changes can also be seen in inflammatory bowel diseases. However, the absence of other features such as cryptitis, crypt abscess, and granuloma, as well as the clinical setting, can help to differentiate between the two conditions.

Lonsdale in his series, [13] reported that ectasia with congestion was seen in 95% of cases. Tendler and his colleagues,[11] also identified similar mucosal capillary abnormalities, including dilatation, congestion, and thrombosis, in 87% of their patients. Our study revealed findings, with ectasia and congestion being seen in 72.9% and 27% of cases, respectively.

CONCLUSION

In conclusion this study to the best of our knowledge, is the first study of this rare syndrome in Kerala-South india and shows that SRUS in our area has similar clinical and pathological characteristics to SRUS from other areas. This study reaffirms that the SRUS is a misleading term for this condition. The clinical presentation is variable, but the combination of rectal bleeding, constipation, straining at stools and perianal pain should alert the clinician to this diagnosis. The endoscopic finding commonly is a solitary ulcer, but other findings can also be seen, such as polyp or erythema. Histological examination is the gold standard for establishing the diagnosis of SRUS. The presence of fibromuscular obliteration of the lamina propria, crypts distortion and surface serration, are diagnostic features. Clinicians and pathologists should be aware of the features of SRUS so that it is not confused with other conditions.

DISCLOSURE The authors report no conflicts of interest in this work.

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