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A report of 112 cases of solitary rectal ulcer syndrome from Iran

Research Article

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Abstract: Solitary rectal ulcer syndrome (SRUS) is a syndrome with symptoms such as rectal bleeding, obstructed defecation, straining at stool and incomplete evacuation, and rectal polyps. In the present study, the clinical features of SRUS among Iranian patients is reviewed. Records of 112 patients diagnosed with SRUS between 1997 and 2007 admitted to a special coloproctology clinic have been analyzed retrospectively. Of 112 patients with SRUS, 61 were male (54.4%) and 51 female (45.6%) with a mean age of 32.2 years (range, 16–64 years). The mean interval between onset of symptoms and final diagnosis of SRUS was 3.8 years (range, 1–14 years). Rectal bleeding (67%) was the most common symptom in both genders, while a feeling of fullness was the least common symptom, observed only in one woman (2%). Incontinence was observed in 4 women (7.8%) and 3 men (4.9%), comprising 6.25% of the total clinical presentations. There were 38 patients (33.9%) with mucosal prolapse and 2 patients (1.8%) with total rectal prolapse. In conclusion, rectal bleeding in the presence of constipation or diarrhea is the main sign for diagnosis of SRUS. This syndrome is usually misdiagnosed; it is frequently confused with inflammatory bowel disease (IBD) because the rectal macroscopic and microscopic lesions and true polyps of rectum are similar. We suggest that most of patients who are treating for IBD and true polyps without any response are suffering from SRUS. Regarding misdiagnosis between SRUS and IBD or rectal polyp, the exact prevalence of SRUS has been mistakenly underestimated.

Keywords: *Solitary rectal ulcer syndrome • Inflammatory bowel disease • Rectal bleeding • Rectal polyp*

1. Introduction

Solitary rectal ulcer syndrome (SRUS) appears to be an uncommon disease because of misdiagnosis as inflammatory bowel disease (IBD) or rectal polyps that results in an underestimation of its true prevalence. SRUS is usually accompanied by symptoms of obstructed defecation that cause straining at stool and incomplete evacuation. For complete evacuation, patients must perform a water enema or digitation; these maneuvers cause trauma to the rectal wall and then ischemia, and eventually, ulceration [1-3]. In the ulcerative stage, the patient's symptoms of rectal bleeding, mucous discharge, and frequency of defecation necessitate differential diagnosis with inflammatory bowel disease (IBD) [4,5]. In advanced cases, a polypoid appearance develops, causing misdiagnosis with true polyps of the rectum [4,6]. Fibromuscular obliteration of the lamina propria and distortion of mucosal architecture are seen upon pathology screening [7].

There are different treatments for SRUS that include enema of corticosteroid or salicylates, rectopexy, rectal mucosectomy, and biofeedback. However, biofeedback should be the main treatment, while for those who do not respond to regular treatments or those with total rectal prolapse, surgery should be considered [8]. In the present study, clinical features of SRUS among Iranian patients have been evaluated to help determine the prominent criteria that differentiate this syndrome from IBD.

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Table 1. Demographic data of included patients.

Sex	(n)	(%)	Mean Age (years)	Mean symptoms duration (years)
Male	61	54.4	28.8	3.5
Female	51	45. 5	35.5	4
Total	112		32.2	3.8

Table 2. Clinical presentation of SRUS in 112 patients.

Symptoms	Male		Female		Total	
	(n)	(%)	(n)	(%)	(n)	(%)
Rectal bleeding	40	65.6	35	68.6	75	67
Wait in Toilet	39	64	27	53	66	59
Mucosal discharge	41	67.2	24	47	65	58
Self digitation	31	50.8	32	62.7	63	56.2
Anorectal pain	30	49.2	24	47	54	48.2
Constipation	28	45.9	24	47	52	46.4
Obstructive defecation	22	36	18	35.3	40	35.7
Tenesmus	12	19.7	19	37.2	31	27.7
Incomplete evacuation	16	26.2	10	19.6	26	23.2
Increasing number of bowel movement	14	23	12	23.5	26	23.2
Diarrhea	10	16.4	6	11.7	16	14.3
Bowel habit disturbance	5	5	10	19.6	15	13.4
Straining	5	8.2	6	11.7	11	9.8
Incontinency	3	4.9	4	7.8	7	6.2
Feeling of fullness	0	0	1	2	1	2

2. Material and Methods

We retrospectively reviewed the records of 112 patients diagnosed with SRUS between 1997 and 2007 in the Baharan Coloproctology clinic in Tehran, Iran. Patients who presented with complaints of rectal bleeding, constipation, waiting in toilet suggestive of SRUS, and who were diagnosed having SRUS after confirmation by sigmoidoscopy and rectal pathology, were included in the study. During sigmoidoscopy, the distance of the lesion from the anal verge was measured. Rectal manometry was done for 33 patients and rectal endosonography was done for 5 patients. Patients with incomplete data, those who had been treated by biofeedback or surgery, and those having a history of psychiatric disorders were excluded.

Data were analyzed for age, gender, clinical presentations, as well as the results of physical examination, sigmoidoscopy, histopathology, rectal manometry, and rectal endosonography.

The histopathological criteria for confirmation of SRUS were a preserved architectural pattern and fibromuscular proliferation of lamina propria negative for dysplasia in glandular cell. On the basis of these criteria and macroscopic appearance, SRUS was subdivided to active, ulcerative, and polypoid stages.

3. Results

Of 112 documented patients with SRUS, 61 were male (54.4%) and 51 female (45.6%) with a mean age of 32.2 years (range, 16–64 years). The mean interval between onset of symptoms and final diagnosis of SRUS was 3.8 years (range: 1 month–14 years) (Table 1).

The clinical presentation of the patients included rectal bleeding, delayed evacuation of stool, mucosal discharge, self-digitation, anorectal pain, constipation, obstructive defecation, tenesmus, incomplete evacuation, frequency, diarrhea, disturbed bowel habit, straining, incontinency, and feeling of fullness. Among these signs and symptoms, rectal bleeding (67%) was the most common, particularly in females (68.6%), while mucosal discharge was the most prominent (67.2%) in males. In contrast, a feeling of fullness was the least frequent symptom, observed only in one woman (2%). Waiting in toilet, anorectal pain, obstructive defecation, incomplete evacuation, and diarrhea were often observed in males but other signs were more prominent in women. Incontinence was observed in 4 women (7.8%) and 3 men (4.9%) comprising 6.25% of the total clinical presentations (Table 2). Rectal prolapse was observed in 17 women and 23 men. There were 38 patients (33.9%) with mucosal prolapse and 2 patients

Table 3. Result of rectal manometry in 33 patients with SRUS.

No. of cases	Paradoxical straining	Squeezing	Resting Pressure
7	N	N	N
9	N	N	↑
4	N	N	↓
2	Abnormal straining	N	N
7	Abnormal straining	N	↑
1	Abnormal straining	N	↓
2	N	Low duration	N
1	N	Low duration	↓

Abbreviations: N: Normal, ↑: Increase, ↓: Decrease

(1.8%) with total rectal prolapse. The mean distance of lesion from anal verge in both women and men were 6.5, 8, and 7.3 cm, respectively.

Rectal manometry was performed in 33 patients; 7 of the results were normal. Nine patients showed an increase in the resting pressure, and 4, a decrease. Paradoxic abnormal strain was observed in 2 patients; 7 patients had paradoxic abnormal strain with high resting pressure. In contrast, low resting pressure coexisting with paradoxic abnormal strain was observed in only 1 patient. Three patients had a low duration in squeezing phase of manometry, while 1 of them showed low resting pressure (Table 3).

Rectal endosonography was done for 5 patients. Three patients had injury in the internal anal sphincter (IAS) consisting of rupture, thickness, and destruction. One patient was normal, and another had polyp in external anal sphincter (EAS). Rigid sigmoidoscopy was done for all patients, but the position of the ulcer was determined only in 12 patients, indicating 8 in anterior, 3 in posterior, and 1 in right lateral positions.

According to the previously mentioned histopathological criteria for SRUS, all 112 cases were diagnosed as suffering from SRUS. On the basis of the morphologic appearance of the lesions, there were 91 patients (81%) in active, 14 (14%) in polypoid, and 7 (6%) in ulcerative stages.

4. Discussion

The present case series is one of the largest series of SRUS ever conducted because patients were collected from a referral center for colorectal diseases in the country. The first reason for the high prevalence of SRUS in Iran seems to be the use of the Turkish-style toilet. Using the Turkish toilet requires sitting a long time in a squatting position, which may be a predisposing factor for descent of the perineum and induction of SRUS. Interestingly, all patients included in this study were using that style of toilet.

In fact, SRUS can be misdiagnosed as IBD by endoscopists, particularly in the inflammatory phase of the disease; while in the polypoid stage, SRUS is very similar to true polyps [3,4,6]. A similar rate of misdiagnosis of SRUS has been already reported by another group of researchers [3]. In most series of SRUS reported previously from other countries, the male to female ratio was one [11,12] but in our series, the number of males was higher than that of females (61 vs. 51). Rectal bleeding was the most common symptom in the present study. We believe that SRUS should be considered as the main cause of rectal bleeding. The present study revealed that more than half of our patients experienced obstructed defecation, long standing on toilette, and self-digitation. Thus, insufficient relaxation of the puborectalis muscle, trauma to the rectal mucosa, and ischemia of the rectal mucosa are the main pathogeneses of rectal ulcer. Actually, the SRUS-causal effect of self-digitation has not gained much support in the literature, especially considering signs of non-relaxing puborectalis muscle and symptoms of rectal mucosal ulcer. Therefore, biofeedback that relaxes the puborectalis muscles seems the fundamental treatment for SRUS; it is frequently used for the treatment of constipation [9,10].

Among our study cases, only 2 patients had total rectal prolapse, while more then 30% had rectal mucosal prolapse and were improved by surgery. In a recent study from Italy, treatment of mucosal prolapse with PPH (procedure for prolapse and hemorrhoids) showed good results [8]. In addition, intra-anal Delorme's surgery indicated good results in a few studies [14-17].

Pathologic reports in our series revealed that 81% of patients were in active phase of SRUS, 14% in the polypoid phase, and 6% in ulcerative phases. In latter stage, SRUS is very similar to IBD and true polyps of rectum, and thus it can be misdiagnosed and mistreated. Our study revealed that the distance of the ulcer from the anal verge is about 8 cm, which should be an indication for the endoscopist to consider this diagnosis when the ulcer is seen at this distance during endoscopy.

Manometric results showed abnormal or paradox straining alone in less than 30% of our patients, which is not compatible with number of patients with symptoms of a non-relaxing puborectalis muscle. Our belief is that manometry is not an effective instrument for exploring this symptom. Endoanal ultrasound was done in some of the present study patients, showing internal anal sphincter injury that is a result of previous anal surgery in those patients. In other studies, endoanal ultrasound revealed thickening of internal anal sphincter due to total rectal prolapse. Our patients had different modalities of treatment for SRUS, such as biofeedback, enema with mixed components, intra-anal Delorme's surgery; further studies are necessary to review their results.

We suspect that psychiatric problems, particularly mood disorders, are among of the reasons for SRUS that need to be evaluated in the future. One of the consequences of SRUS is infection; that should be taken under consideration. Recently, the role of microbes in inflammatory lesions have raised much interest; the effectiveness of antibiotics and the benefit of probiotics in amelioration of inflammatory conditions in the colon have been confirmed by meta-analyses [18-24].

In conclusion, this is the first report of SRUS among Iranian patients. Non-relaxing puborectalis muscle is the main cause of this syndrome and treatment should focus on treating it. On the other hand, the frequency of diagnosis of this syndrome is underestimated, and physicians and surgeons should consider this diagnosis in patients with rectal bleeding and constipation or diarrhea. Referring suspected samples in IBD and SRUS for confirmation by another pathologist is suggested.

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