Solitary Rectal Ulcer Syndrome: A Biopsychosocial Assessment

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Case Study

Abstract

Background: Solitary rectal ulcer syndrome (SRUS) is a chronic disorder of the gastrointestinal tract and its etiology is not well understood. There is no specific treatment for this syndrome and patients with SRUS may, for years, experience many complications. The aim of the present research was the biopsychosocial study of patients with SRUS.

Methods: The study participants consisted of 16 patients with SRUS (7 men and 9 women). Their medical records were reviewed retrospectively to evaluate the clinical spectrum of the patients along with the endoscopic and histological findings. Moreover, psychiatric and personality disorders [based on Diagnostic and Statistical Manual of Mental Disorders, 4th ed, Text Revision (DSM IV-TR)], psychosocial stressors, early life traumas, and coping mechanisms were assessed through structured interviews.

Results: At presentation, mean age of the patients was 39 years (16 to 70). Common symptoms reported included rectal bleeding (93.8%), rectal self-digitations (81.2%), passage of mucous (75%), anal pain (75%), and straining (75%). Endoscopically, solitary and multiple lesions were present in 9 (60%) and 4 (26.7%) patients, respectively, and 87% of lesions were ulcerative and 13.3% polypoidal. The most common histological findings were superficial ulceration (92.85%) and intercryptic fibromuscular obliteration (87.71%). Common psychosocial findings included anxiety disorders (50%), depression (37.5%), obsessive-compulsive personality disorder (OCPD) or traits (62.5%), interpersonal problems (43.75%), marital conflicts (43.75%), occupational stress (37.5%), early life traumas, physical abuse (31.25%), sexual abuse (31.25%), dysfunctional coping mechanisms, emotional inhibition (50%), and non-assertiveness (37.5%).

Conclusion: Given the evidence in this study, we cannot ignore the psychosocial problems of patients with SRUS and biopsychosocial assessment of SRUS is more appropriate than biomedical evaluation alone.

Keywords: Solitary rectal ulcer syndrome (SRUS), Biopsychosocial assessment, Psychosocial factors

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Introduction

Solitary rectal ulcer syndrome (SRUS) is an uncommon, chronic, benign anal disorder, with an estimated prevalence of one in 100,000 persons per year (Martin, Parks, & Biggart, 1981). It occurs most commonly in the third decade in men and in the fourth decade in women (Kennedy, Hughes, & 1977; Sharara, Azar, Masterton, Haddad, & Eloubeidi, 2005). Often, SRUS is underdiagnosed due to its wide spectrum of clinical presentation and variable endoscopic findings (Al-Brahim, Al-Awadhi, Al-Enezi, Alsurayei, & Ahmad, 2009). Rectal bleeding, constipation, mucous discharge, straining on defecation are the most common presentations (Abbasi et al., 2015; Al-Brahim et al., 2009; Tjandra et al., 1992). Diarrhea and abdominal pain are also sometimes observed (Chong & Jalihal, 2006). In addition, 26% of patients are asymptomatic (Tjandra et al., 1992). It is essential to differentiate SRUS from other chronic or malignant diseases. Although the name suggests the presence of a solitary ulcer in the rectum, some studies have reported SRUS patients with polyps, multiple lesions, or even ulcer in the colon. Thus, the disease is also known as the three-lies disease" (Crespo, Moreira,, Redondo, Lopez San, & Milicua Salamero, 2007).

The diagnosis of SRUS is based on symptomatology in combination with histological endoscopic and findings (Al-Brahim et al., 2009). Its characteristic features include fibrosis or obliteration of lamina propria, thickening or hypertrophy of mucosa, and crypt architecture distortion (Al-Brahim et al., 2009; Bahadori Hesari, Gouhari Moghadam, Derakhshani, & Vafaei, 2006; Suresh, Ganesh, & Sathiyasekaran, 2010).

The pathogenesis of SRUS is not well known, but it is believed that multiple factors contribute to its development. The most accepted causes of this disease are direct trauma or local ischemia due to mucosal prolapse, straining during bowel movements, or self-digitations (Chong & Jalihal, 2006;

Kuijpers, Scheve, & Ten Cate, 1986; El-Hemaly et al., 2012).

There is no specific cure for SRUS. Only the symptoms may be improved by current treatments that include the use of bulking agents, laxatives, sucralfate, bowel retraining with or without biofeedback, and surgery (Meurette et al., 2008; Zhu, Shen, Qin, & Wang, 2014).

In recent years, it has generally been accepted that illness and health are the result interaction between biological, psychological, and social factors (Sarafino & Smith, 2014). The biopsychosocial model is now widely accepted as the most important approach to chronic illnesses. In this model, aspects of biological, psychological, and social domains are most important to understanding and promoting the patient's health (Engel, 1980). Previous studies have stated that psychosocial factors including chronic life stressors, life traumas (e.g., abuse history), psychiatric or psychological co-morbidities, and the patient's coping mechanisms may influence the nature and severity of the symptoms of chronic gastrointestinal (GI) disease (Gastroenterology and Endoscopy News, 2003; Drossman, Talley, Leserman, Barreiro, 1995). Thus, & biopsychosocial model has been increasingly used for functional GI diseases such as irritable bowel syndrome (IBS) (Tanaka, Kanazawa, Fukudo, & Drossman, 2011).

SRUS is a disease with almost unknown etiology and a chronic disabling condition that affects the quality of life (QOL) (Meurette et al., 2008). Thus, it seems, this model is suitable for the evaluation and treatment of these patients. In the literature review, we found no study on psychosocial aspects in patients with SRUS. Therefore, the aim of this study was an initial assessment of the biopsychosocial aspects of SRUS.

Methods

After obtaining the approval of the ethics committee of Isfahan University of Medical Sciences, Iran, based on the computer data, all patients with SRUS who referred for treatment to two medical centers affiliated to Isfahan University of Medical Sciences from 2004 to 2014 were identified. As a result, 30 patients were identified, 16 of whom agreed to participate in the study. The aim of the study was explained to the participants and a written informed consent was obtained from each.

The clinical records of these patients and laboratory, colonoscopy, and histological findings were retrospectively reviewed. Subsequently, they were interviewed by a psychiatrist for the evaluation of psychiatric and personality problems through structural interviews based on Diagnostic and Statistical Manual of Mental Disorders, 4th ed, Text Revision (DSM IV-TR) and a psychologist for the assessment of early life traumas (physical and sexual abuse), psychosocial stressors in and dysfunctional coping recent vears, mechanisms through open response questions.

The statistical analysis was performed using SPSS software (version 20, SPSS Inc., Chicago, IL, USA) and frequency analysis was also performed.

Results

In total, 16 patients with SRUS were studied. There were 7 men (43.8%) and 9 women (56.2%) and 69% of them were married. Their age ranged from 16 to 70 years (mean: 39 years). The duration of their symptoms ranged from 1 to 21 years (mean: 8 years). In addition, 7 patients had undergone abdominal and anorectal surgery (procedures such as hemorrhoidectomy, sphincterotomy, rectocele, herniorrhaphy, and ppendectomy).

Eight patients (50%) had a comorbidity of functional GI diseases [IBS and functional dyspepsia (FD)]. Moreover, 8 patients (50%) had a history of psychiatric disorder and 4 patients (25%) had rectal sex.

Table 1 shows the clinical manifestations of the patients. Rectal bleeding (15, 93.8%), rectal self-digitations (13, 81.2%), passage of mucous (12, 75%), anal pain (12, 75%), and straining (12, 75%) were the most common symptoms. All patients had more than one symptom.

In laboratory evaluations, only 1 patient was diabetic (BS > 126), 3 patients had anemia (HB < 12), and the results of the remaining tests, including erythrocyte sedimentation rate (ESR), C-reactive protein (CRP), antineutrophil cytoplasmic antibody (ANCA), perinuclear neutrophil antibodies (PANCA), platelet count (PLT), white blood cell count (WBS), and stool exam, were normal.

Furthermore, rectoscopic and histological findings have been summarized in table 2.

Table 1. Frequency of clinical manifestations of patients with SRUS (n = 16)

Symptoms*	Number	%
Rectal bleeding	15	93.8
Rectal self-digitations	13	81.2
Passage of mucous	12	75.0
Anal pain	12	75.0
Straining	12	75.0
bloating	11	68.8
Constipation	11	68.8
Abdominal pain	11	68.8
Long- time defecation	10	62.5
Frequent bowel movements	10	62.5
Diarrhea	7	43.8

*All patients have more than one symptom at presentation.

Table 2. Rectoscopic and histological findings of patients with solitary rectal ulcer syndrome (SRUS) (n = 16)

Rectoscopic findings*	Number	%
Site of lesion < 6 cm above anal verge	6	33.3
Site of lesion > 6 cm above anal verge	10	66.7
Single ulcer	9	60.0
Multiple ulcers	4	26.7
Polypoidal/nodular	2	13.3
Erythematous mucosa	4	26.7
Hemorrhoids	3	20.0
Histological findings**		
Superficial ulceration, congestion, or ecstatic venues	13	92.85
Intercryptic fibromuscular obliteration	12	85.71
Hypertrophy/thickening of muscularis mucosae	7	50.00
Fibrosis of lamina propria	4	28.75
Mucosal architecture distortion	4	28.75

^{*}A patient might have more than one type of lesion; ** Histological findings of two cases were not available.

Colonoscopy findings revealed that the site of lesions was less than 6 cm above anal verge in 66.7% of cases and more than 6 cm above anal verge in 33.3% of cases. On the basis of appearance, single and multiple ulcers were observed in 9 (60%) and 4 patients (26.7%), respectively, while polypoidal/nodular lesions were observed in 2 patients (13.3%). In addition, 4 patients (26.7%) had erythematous mucosa and 3 patients had hemorrhoids.

Histological findings of patients showed superficial ulceration, congestion, or ecstatic venues (13, 92.85%), and intercryptic fibromuscular obliteration (12, 87.71%) were respectively the most common findings.

Table 3 illustrates the frequency of psychiatric disorders, psychosocial stressors in recent years, and early life traumas (physical and sexual abuse) among the patients. Almost half of the patients (50%) were suffering from anxiety disorders including general anxiety disorder (GAD), obsessive compulsive disorder (OCD), posttraumatic stress disorder (PTSD), and social phobia. Unipolar mood disorders such as major depression disorder (MDD) and dysthymia were the next most common (37.5%) disorders among the patients. Interpersonal problems (43.75%), marital conflicts (43.75%), occupational stress (37.5%), and death of a family member (31.25%) were, respectively, the most common psychosocial stressors experienced by patients in recent years. Regarding early life trauma, 5 patients had experienced physical abuse (31.25%) and sexual abuse (31.25%).

An overview of the biopsychosocial assessment of each patient is presented in table 4. The results show that almost all patients had many clinical symptoms. All of them, except 3 patients, suffered from one or more psychiatric disorders specially anxiety disorders. The majority of patients had traits or disorders of obsessive compulsive personality (n = 10). Ineffective coping mechanisms such as emotional inhibition (n = 6) and non-assertiveness (n = 8) were common among the

patients. Psychosocial stressors were experienced by all patients. Most patients experienced more than one stressor and half of them complained of interpersonal and marital conflicts. Frequency of early life traumas such as physical and sexual abuse (31.25%) among these patients was significant.

Table 3. Frequency of psychiatric disorders, psychosocial stressors in recent years, and early life traumas (physical and sexual abuse) among patients with solitary rectal ulcer syndrome (SRUS) (n = 16)

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Psychiatric disorders *	Number	%
Anxiety disorders	8	50.00
Depressive disorders	6	37.50
Adjustment disorders	4	25.00
No diagnosis	3	18.75
Psychosocial stressors **		
Interpersonal problems	7	43.75
Marital conflicts	7	43.75
Occupational stress	6	37.50
Death of a family member	5	31.25
Illness	3	18.75
Family problems	3	18.75
Financial problems	2	12.50
Captivity in war	2	12.50
Addiction of offspring	1	6.25
Love problems	1	6.25
Educational problems	1	6.25
Early life trauma		
Physical abuse	5	31.25
Sexual abuse	5	31.25

^{*}Patients may have had more than one psychiatric disorder.

Discussion

The purpose of this study was an initial assessment of biopsychosocial aspects of patients with SRUS.

Biomedical findings of the study were notable. Although, some studies have reported SRUS in young adults (Chong & Jalihal, 2006; Bahadori Hesari et al., 2006; Amaechi, , Papagrigoriadis, Hizbullah, & Ryan, 2010), mean age of participants of the present study was 39 years. Almost half of patients had pervious surgeries especially anorectal surgery that may be indicative of underdiagnosis of SRUS. Moreover, half of the patients had coof functional GI diseases morbidity especially IBS.

^{**} Patients may have had more than one stressor.

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Table 4. Summary of biopsychosocial assessment of solitary rectal ulcer syndrome (SRUS) cases (clinical presentation, psychiatric and personality

disorders, ineffective coping mechanisms, and psychosocial stressors (n = 16)

Age	Sex	Clinical presentation	Psychiatric disorder	Personality disorder or trait and ineffective coping mechanisms	Psychosocial stressors and early life traumas
70	F	Rectal bleeding, straining, Constipation, Bloating, Abdominal pain, Self-digitations	Adjustment disorder	Emotional inhibition	Interpersonal problems
42	M	Rectal bleeding, Straining, Mucous discharge, Anal pain, Constipation, Long- time defecation, Frequent bowel movements, Abdominal pain, Bloating, Self-digitations	GAD PTSD	OCPD	Marital conflict, Family problems, Death of a child (son), Captivity in war, Physical abuse, Sexual abuse
16	F	Rectal bleeding, straining, Mucous discharge, Constipation, Long-time defecation, Frequent bowel movements, Abdominal pain, Bloating, Self-digitations	MDD Social phobia	Non-assertiveness	Interpersonal problems, Sexual abuse, Physical abuse
26	F	Rectal bleeding, mucous discharge, anal pain, abdominal pain	GAD	OCPD	Marital conflict, Illness
24	F	Rectal bleeding, Straining, Mucous discharge, Anal pain, Constipation, Long- time defecation, Frequent bowel movements, Abdominal pain, Bloating, Self- digitations	Adjustment disorder	OCPD	Interpersonal problem, Educational problems
24	M	Rectal bleeding, Straining, Mucous discharge, Anal pain, Constipation, Long- time defecation, Frequent bowel movements, Abdominal pain, Bloating, Self- digitations	Dysthymia GAD	OCPD	Occupational stress, Family problems
52	F	Rectal bleeding, Straining, Mucous discharge, Anal pain, Constipation, Long- time defecation, Frequent bowel movements, Abdominal pain, Bloating, Self- digitations	GAD	OCP trait Non-assertiveness, Emotional inhibition	Occupational stress, Family problems
34	F	Rectal bleeding, Straining, Mucous discharge, Anal pain, Constipation, Long- time defecation, Frequent bowel movements, Abdominal pain, Bloating, Self- digitations	OCD	OCP trait (Perfectionism) Non-assertiveness	Marital conflict, Interpersonal problems, Physical abuse

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Table 4. Summary of biopsychosocial assessment of solitary rectal ulcer syndrome (SRUS) cases (clinical presentation, psychiatric and personality disorders, ineffective coping mechanisms, and psychosocial stressors (n = 16) (Continue)

Age		Clinical presentation	Psychiatric disorder	Personality disorder or trait and ineffective coping mechanisms	Psychosocial stressors and early life traumas
62	F	Rectal bleeding, Straining, Mucous discharge, Anal pain, Constipation, Long- time defecation, Frequent bowel movements, Abdominal pain, Diarrhea, Bloating, Self-digitations	Adjustment disorder	-	Marital problems, Death of father and brother, Financial problems
29	F	Rectal bleeding, Straining, Mucous discharge, Anal pain, Constipation, Longtime defecation, Self-digitations	-	OCPD Non-assertiveness Emotional inhibition	Financial problems, Interpersonal problems, Marital conflict
26	M	Rectal bleeding, Straining, Mucous discharge, Constipation, Long-time defecation, Frequent bowel movements, Bloating, Self-digitations	MDD	OCP trait (Perfectionism) Non-assertiveness	Occupational stress, Interpersonal problems, Physical abuse, Sexual abuse
38	M	Rectal bleeding, Straining, Mucous discharge, Anal pain, Long-time defecation, Self-digitations	Adjustment disorder	Non-assertiveness Emotional inhibition	Occupational stress
64	F	Rectal bleeding, Straining, Mucous discharge, Anal pain, Long-time defecation, Frequent bowel movements, Abdominal pain, Diarrhea, Bloating, Self-digitations	Dysthymia OCD	-	Family problems, Marital conflict, Death of a son, Sexual abuse
57	M	Rectal bleeding, Anal pain, Constipation, Long-time defecation, Self-digitations	-	OCP trait (Perfectionism)	Occupational stress death of two brothers
25	F	Rectal bleeding, Anal pain, Constipation, Abdominal pain, Bloating	MDD OCD	Dependent personality Non-assertiveness Emotional inhibition	Interpersonal problems, Illness
44	M	Mucous discharge, Anal pain, Frequent bowel movements, Diarrhea, Bloating, Self- digitations	-	OCP trait (Perfectionism) Non-assertiveness Emotional inhibition	Occupational stress, Death of a friend, Captivity in war, Sexual abuse

F: Female; M: Male; GAD: General anxiety disorder; PTSD: Post-traumatic stress disorder; MDD: Major depression disorder; OCD: Obsessive compulsive disorder; OCPD: Obsessive-compulsive personality disorder

In terms of clinical presentations, the frequency of all symptoms in our patients was significant (43-93%). According to most studies, rectal bleeding, passage of mucous, anal pain, and straining were the most common symptoms (Abbasi et al., 2015; Abid et al., 2012; Al-Brahim et al., 2009; Chong & Jalihal, 2006; Crespo et al., 2007; Bahadori Hesari et al., 2006; Chiang, Changchien, & Chen, 2006). Disturbances of bowel function including constipation (68.8%) and diarrhea (43.8%) were relatively common. Nearly all patients reported problems with defecation and the most common of them was excessive straining with the feeling of incomplete evacuation (75%).

They needed to spend a long time in the toilet (62.5%) or go to the toilet frequently (62.5%). Rectal self-digitations usually followed failures to relieve tenesmus after straining for long periods of time (Abid et al., 2012). This is an abnormal defecation behavior that was more common in patients in the present study (81.2%) than other studies (Chong & Jalihal, 2006; Bahadori Hesari et al., 2006). There are different hypotheses about the etiology of SRUS and direct trauma is one of the most important of them. Self-digitation due to difficult defecation may lead to direct trauma and ulcer formation (Crespo et al., Contractor & Contractor, 2003). Moreover, history of rectal sex as a probable factor of inducing direct trauma was reported by 25% of patients. Therefore, direct trauma may be a major cause of SRUS.

In the present study, other medical conditions such as diabetes, ulcerative colitis, parasites, inflammatory diseases, platelet problems, and anemia were assessed through laboratory tests. According to our data, the incidence of these conditions was not considerable.

Among rectoscopic findings, a considerable number of cases of lesions were ulcerative (87%), but only two of the lesions were polypoidal/nodular. Although, 60% of the lesions were single ulcers, 5 patients had

multiple ulcers. The presence of polyps, erythematous lesions, and multiple ulcerations in patients provides further evidence that the term SRUS is misleading. This finding was consistent with the studies by Al-Brahim et al. (2009), Chong & Jalihal (2006), and Crespo et al. (2007).

Histological features are key diagnostic factors in SRUS. The diagnosis is confirmed through the presence of a combination of surface serration, fibromuscular obliteration, hypertrophy/thickening muscularis mucosae, fibrosis of lamina propria, mucosal architecture distortion (Suresh, et al, 2010; Madigan & Morson, 1969). In the current study, superficial ulceration, fibromuscular obliteration, and hypertrophy/thickening of muscularis mucosae were the most common features.

The psychosocial assessment of patients with SRUS revealed interesting findings. For instance, comorbid psychiatric disorders, especially anxiety disorders and depression, were the most common among these patients. Most patients (83%), were suffering from a psychiatric disorder. This is consistence with studies that reported 42% to 61% anxiety disorders and depression in gastrointestinal patients (Drossman & Chang, 2003; Olden & Drossman, 2000).

As personality aspects, OCPD and OCP traits, especially perfectionism, cause a high level of stress, 10 patients (62, 5%) had these personality problems. On the other hand, dysfunctional coping mechanisms such as emotional inhibition and non-assertiveness were common among them. Therefore, it is understandable that these patients experience psychological problems.

According to the biopsychosocial model, psychological distress also affects somatic symptoms and their outcome (Drossman et al., 2003). The findings of the study also indicted that psychosocial stressors in these patients were significant. Relationship difficulties, including marital conflicts or interpersonal problems, and occupational stress were the most commonly observed

psychosocial stressors among them. Furthermore, history of physical and sexual abuse in early life was prevalent among the patients. Early life traumas in GI patients may strongly influence the severity of the symptoms, illness behaviors, daily function, and treatment outcome (Drossman et al., 1996; Drossman et al., 2011). In some studies, patients of a GI clinic who had a history of abuse reported more severe pain and greater psychological distress. Moreover, they spent more days in bed with poorer health status, more frequent visits to the physician, and more surgical procedures (Drossman et al., 1995, Drossman et al., 1996). Therefore, an evaluation of these psychological traumas may also be important in the treatment of SRUS.

Several limitations in this study needed to be addressed. This study was a retrospective study on a small number of patients without a control group. It was not possible to draw any conclusions on treatment modalities. Obtained psychosocial data on the patients were only based on interviews and no standard selfadministered questionnaires were used. Thus, we should be cautious in generalizing the findings. However, this study is a new assessment of SRUS and may be the first biopsychosocial assessment patients.

In conclusion, SRUS is a chronic disorder with a spectrum of clinicopathological abnormalities. Rectal bleeding, passage of mucous, anal pain, and straining are the most common symptoms of SRUS. Direct trauma through self-digitations or rectal sex may be important in the etiology of Furthermore, the assessment of psychosocial aspects including psychiatric comorbidities, personalities, dysfunctional maladaptive coping mechanisms, chronic life stressors, and early life traumas, especially physical or sexual abuse, may be important in the treatment of patients with SRUS. Therefore, it seems that biopsychosocial assessment of SRUS is more appropriate than biomedical evaluation alone.

Conflict of Interests

Authors have no conflict of interests.

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