Solitary rectal ulcer syndrome
A systematic review

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Abstract

Background: Solitary rectal ulcer (SRUS) may mislead the inflammatory bowel disease (IBD) or rectal polyps, which may reduce the actual prevalence of it. Various treatments for SRUS have been described that can be referred to therapeutic strategies such as biofeedback, enema of corticosteroid, topical therapy, and rectal mucosectomy. Nevertheless, biofeedback should be considered as the first stage of treatment, while surgical procedures have been offered for those who do not respond to conservative management and biofeedback or those who have total rectal prolapse and rectal full-thickness.

Methods: A systematic and comprehensive search will be performed using MEDLINE, PubMed, Scopus, EMBASE, AMED, the Cochrane Library, and Google Scholar.

Results: The results of this systematic review will be published in a peer-reviewed journal.

Conclusion: To our knowledge, our study discusses the factors involved in the pathogenesis, clinical symptoms, diagnosis, treatment, and management of patients. This review can provide recommended strategies in a comprehensive and targeted vision for patients suffering from this syndrome.

Abbreviations: CT = computed tomography, FS = flexible sigmoidoscopy, IBD = inflammatory bowel disease, MRI = magnetic resonance imaging, PET = positron emission tomography, SRUS = solitary rectal ulcer.

Keywords: diagnosis, management, prevalence, solitary rectal ulcer, treatment

1. Introduction

Solitary rectal ulcer syndrome (SRUS) is an unusual rectal disorder that does not necessarily end with an ulcer and may affect different parts of the rectum and other site of gastrointestinal tract.[1,2]

The cause of this syndrome is unclear and may have various factors in causing a lesion simultaneously, including rectal prolapse, chronic, and severe constipation. SRUS is often caused by chronic constipation, which can be associated with straining during defecation, rectal bleeding, tenesmus, mucoid secretion from the rectum, rectal pain, and a sense of incomplete evacuation.[3-5]

The occurrence of symptoms affects the probability of the disease, and its diagnosis is by direct examination of the lesion by colonoscopy and histological study of lesion. However, the etiology, pathophysiology, and clinical manifestations of SRUS are not fully understood. Given different clinical symptoms and endoscopic findings, SRUS may be confused with disorders such as inflammatory bowel disease (IBD) and neoplasms.[6]

Therefore, this paper attempts to evaluate the pathogenesis, clinical signs, diagnosis, and management of patients. Evaluating the causes and therapeutic strategies will be helpful in future therapies and prevention strategies.

2. Prevalence

The prevalence of SRUS is not exactly clear, but it is estimated as 1 in 100,000 people per year.[7,8] SRUS have been reported more often in men in the third decade and in the fourth decade of women’s lives; however, several cases have been reported previously.[9-12] However, the prevalence of SRUS in men and women is almost the same and can occur at any age. A large number of SRUS patients have been reported from a specialized gastroenterology center in Iran over the past 5 years, indicating a high prevalence in Iran.[13] One prospective study has also reported cases of children with SRUS in southern Iran.[12]

3. Symptoms (Clinical presentation)

SRUS is often known as rectal ulcer within 10 cm, which is often misdiagnosed in many cases as IBD. Clinical signs of this syndrome based on reports are as follows.

Patients typically complain of rectal pain, rectal prolapse, bleeding, pain, tenesmus, mucus, chronic and severe constipation,
lengthened straining on defecation, pelvic discomfort as well as a sense of incomplete evacuation. Nonetheless, it has been suggested that up to 26% of patients may be asymptomatic. The most common clinical symptom is reported to be rectal bleeding. The amount of hemorrhage varies in this condition, and the direct bleeding of the blood vessels varies a little too severe bleeding where there is a need for blood transfusion. Severe rectal hemorrhage, which requires emergency endoscopy to diagnose the underlying cause, is rarely reported. The history of repeated use of laxatives has been reported in many patients. Self-induced trauma has been reported in people who have been trying to remove stools by rectal digitation. 

4. The pathogenesis of SRUS

The pathogenesis of SRUS is not well known; various factors may be involved in its creation and development, which should be considered. It has been stated that the most important theories are associated with direct trauma or causes of local ischemia. 

(1) Straining: Lengthened straining during bowel movements in the patient who suffers from constipation may result in a direct trauma to the mucosa.

(2) Self-induced trauma: Self-instrumentation can be occurred when individuals attempt to remove impacted stool by rectal digitation.

(3) Paradoxical contraction of puborectalis muscle: Uncoordinated muscle contraction in the puborectalis muscle has been indicated to be associated with increased intrarectum pressure and anal canal, resulting in ischemic production and ulceration.

(4) Rectal prolapse and intussusception: Rectal intussusception can lead to localized vascular trauma and consequently the onset of solitary local ulceration.

5. Diagnosis

SRUS is already well-known, but easily misdiagnosed condition, where proper diagnosis and treatment of SRUS is still an important challenge. It should be noted that its rare occurrence usually leads to the fact that it is not properly diagnosed with other diseases due to the lack of knowledge or lack of experience of doctors. There are clinicopathologic similarities between SRUS and IBD or constipation. Specialists also believe that the concept of SRU in some cases may coincide with misleading interpretations, so that lesions may not be solitary or ulcerated. In other words, the emergence of SRUS in endoscopy can be largely due to well-demarcated ulcers to cauliflower-looking tumors or edema swelling. This can be posed as the most common childhood conditions, such as IBD or constipation, which lessens the management of lower gastrointestinal symptoms. The diagnosis of SRUS can usually be performed by combination of symptomatology, endoscopy, sigmoidoscopy, and histology.

The syndrome is characterized by histological features, the importance of which can be summarized according to the following characteristic appearance:

(1) Thickening of the mucosal layer along with crypts distortion;
(2) Fibromuscular obliteration in the lamina propria has been reported to be the cornerstone for diagnosing SRUS.
(3) The extension of muscle fibers is also seen as an upward movement between crypts.
(4) Thickening of the mucosal layer along with distorting crypt architecture;
(5) Glandular crypt abnormalities were revealed in this syndrome.
(6) Surface ulceration;
(7) Mucous cell proliferation, hyperplastic, and serrated mucosa
(8) Mucosal glands distortion;
(9) Mild inflammation and reactive epithelial atypia.

Evaluation of internal or full-thickness rectal prolapse is also strongly recommended in this syndrome. Flexible sigmoidoscopy (FS) is a method in which a sigmoidoscope is inserted into the rectum by which the rectum and part of colon can be examined and each diagnostic or therapeutic maneuver is accordingly implemented. FS or colonoscopy is used to determine the unknown cause of mucosal lesions, rectal ulcers, IBD, etc.

Medical imaging technique such as magnetic resonance imaging (MRI), defecating proctography, transrectal and endoanal ultrasound, and barium enema have been reported to be most important diagnostic methods for imaging evaluation.

Regarding reports, a series of characteristics are described for the transrectal and endoanal ultrasonography, including an absence of distinction between the mucosa and the muscularis propria, thickened muscularis propria, considerable thickening of the internal anal sphincter, thickening evidence in external sphincter, as well as thickened submucosal layer. A study has reported that ultrasound is very helpful in evaluating the thickness of the anal sphincter in patients suffering from SRUS.

Thickening of internal anal sphincter has been described previously to be associated with high-grade rectoanal intussusception. It has been reported that thickening of the submucosal layer may be secondarily linked to the rectal mucosal prolapsing in the anus and edema in the rectum wall. Defecography is a radiological imaging in which different stages of defection can be visualized by a fluoroscope by which anorectal prolapse, external prolapse of rectum, intussusception in-relaxing puborectalis muscle are diagnosed as well as defection difficulties. Nevertheless, due to easier access to endoscopy and biopsy, defecography is most commonly used for underlying pathophysiology, as well as preoperative evaluation.

Magnetic resonance (MR) defecography can show pelvic muscles action and accordingly rectum function and sphincter. This method can show the cause of constipation and other problems such as lower limb prolapse.

MRI is not routinely used in the diagnosis and management of patients suffering from SRUS. MRI has been used for patients suspected of having malignancy where examined by endoscopy. MRI has been introduced as a differential procedure of mural thickening of the rectum and could indicate SRUS by adequate clinical information. SRUS mimicking rectal cancer based upon use of various diagnostic methods, including endoscopy, positron emission tomography (PET)-CT, MRI, and abdominopelvic computed tomography (CT), has been shown in some cases.

Barium enema as a type of X-ray imaging method can be used for examination of muscle function and its coordination, as well as prolapse. It is capable of showing thickening of the rectal folds, polypoid lesions and ulcers, as well as stricture formation, but these observations can result in a misdiagnosis where the results are markedly similar to malignant lesions.

6. Treatment

Treatment for SRUS is based on its symptoms (the severity of the disease) and presence of rectal prolapse. Asymptomatic patients
may usually require behavioral changes, and other types of treatment may not be considered. It should be noted that a conservative, stepwise, patient education, and behavioral modification approach are the first proposed strategies.[37–39] Patients who are asymptomatic or minimally symptomatic may be treated with bulk laxatives, bowel retraining, and reassurance.

7. Conservative treatment and biofeedback therapy
At the time of diagnosis, patients should be advised to use a high-fiber diet and bulk laxatives. They also need to be trained for prevention of straining and anal digitation. The toilet habits (time spent in the toilet) should be adjusted and defecation training should be noted. It is noteworthy that dietary and behavioral changes, especially in patients with mild to moderate symptoms, can be dramatically effective in the absence of mucosal prolapse, which can help in the improvement and prevention of disease progression.[20,40,41] Conservative treatment may be no longer effective if the disease is more advanced, especially in cases where there is a high degree of intussusception in rectum, and, fibrosis, or external prolapse. In these cases, the resistance to conservative treatment may occur; subsequently, biofeedback can be promising in these patients for improving symptoms. Biofeedback is known as a variety of behavioral changes that are effective in reducing excessive straining with defecation through correction of abnormal pelvic floor behavior and stopping the use of suppositories and laxatives.[20,42] Compliance with behavioral modalities has been reported to have had an effective outcome in childhood SRUS, which may be due to short duration of this syndrome compared with adults.[13]

Studies have suggested that biofeedback is an appropriate and useful treatment for most patients with SRUS and an appropriate result has been achieved as a result of increased rectal mucosal blood flow.[43]

However, problems have also been addressed for this treatment. Of these problems, the lower number of patients who can be treated with this type of treatment can be noted, which leads to failure of treatment.[45] In addition, over time, the effects of this type of treatment may be reduced in some patients.[45] In fact, its short-term effects are beneficial because it is not effective in the long term.[25,40] Durable efficacy is uncertain and may therefore be necessary to repeat treatment.

8. Topical therapy
Topical therapy has been reported to be effective in some cases. Sucralfate enema, corticosteroids, and sulphasalazine enemas have been reported to be effective in improving the symptom in uncontrolled case series; however, their long-term effectiveness needs further evaluation.[20,25,46,47] Moreover, topical glucocorticoids, salicylates, and botulinum toxin have also been used, but they do not seem to be suitable for treatment.[46,48] Of course, the botulinum toxin is expected to last for about 3 months, which may be more effective than biofeedback therapy.[49]

9. Surgery
Surgical treatment is recommended for patients who suffer from full-thickness or rectal mucosal prolapse or for those who are resistant to conservative management and biofeedback treatment.[23,50]

Options that are recommended for surgery include rectopexy, perineal proctectomy (Altemeier procedure), excision, diversion, as well as Delorme procedure as mucosal resection.[15,51,52] Removing lesions or local excision has been successful, but remains unclear with long-term effects. Rectopexy is also intended to correct anal prolapse.[53] Long-term results of antiprolapse surgery have been reported to substantially improve the resolution of symptoms in patients with resistant SRUS to medical treatment.[54]

In general, antiprolapsal surgery has led to a promising long-term outcome of about 60% of patients undergoing surgery.[54] Previous studies have shown that rectopexy has been very effective in improving the rectal configuration and the success of rectal prolapse treatment in SRUS.[34] Mucosal resection or perineal proctectomy has been previously introduced in a full thick prolapse.[4,25] Surgical procedures such as transanal mucosal sleeve resection along with coloanal pull-through (P-T) or diverting colostomy has been described to be available for when the above-mentioned methods fail.[25]

The fecal diversion approach has also been effective in improving the symptoms of patients and can be performed in patients who have failed other surgical methods.[55] Surgeries, including rectopexy, excision of ulcer, and rarely colostomy, are used in children with continuous hemorrhage per rectum that was not curable.[12,51,54]

10. Conclusion
This syndrome is misleading, where simply erythema, mucosal ulcerations, and polypoid lesions can be present in patients by endoscopy. Moreover, there are clinicopathologic similarities between SRUS and IBD or constipation. The pathogenesis of SRUS is not adequately described, but various factors can be involved. The diagnosis of SRUS is usually done by analyzing the outcomes of symptoms, endoscopy, sigmoidoscopy, and histology. As already mentioned, the treatment for SRUS is based on its symptoms (severity) and the presence of anal prolapse. Asymptomatic patients are usually advised to change their behavior and other types of treatment may not be taken into consideration in these cases. It is noteworthy that conservative management, patient education, fiber consumption, and behavioral modification are the first strategies that can be applied at an early stage. Behavioral modification or biofeedback treatment has been shown to be effective in improving both rectal blood flow and symptoms. Surgical treatment is recommended for patients with certain symptoms who have complete prolapse or full-thickness or those who do not respond to conservative and biofeedback.

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References
[1] Marchal F, Bresler L, Brunaud L, et al. Solitary rectal ulcer syndrome: a series of 13 patients operated with a mean follow-up of 4.5 years. Int J Colorectal Dis 2001;16:228–33.
[2] Rosai J. Ackerman’s surgical pathology, 8th ed. 1996; Mosby–Year Book, St. Louis, 751.
Dehghani SM, Haghighat M, Imanieh MH, et al. Solitary rectal ulcer syndrome: a single-center experience of 116 cases. BMC Gastroenterol 2012;12:72.

Tjandra JJ, Fazio VW, Church JM, et al. Clinical conundrum of solitary rectal ulcer syndrome: the role of autofluorescence colonoscopy. Photodiagnostics Photodyn Ther 2007;4:179–83.

Dehghani SM, Haghighat M, Imanieh MH, et al. Solitary rectal ulcer syndrome: a prospective study of cases from southern Iran. Eur J Gastroenterol Hepatol 2008;20:93–5.

Dehghani SM, Malekpour A, Haghighat M. Solitary rectal ulcer syndrome in children: a literature review. World J Gastroenterol 2012;18:6541–5.

Tjandra JI, Fazio VW, Church JM, et al. Clinical conundrum of solitary rectal ulcer. Dis Colon Rectum 1992;35:227–34.

Kato K, Nagase A, Iwasaki Y, et al. Massive bleeding from visible vessels within a solitary rectal ulcer. Surgery 2014;155:969–75.

Geramizadeh B, Bagherzad H, Jahanshani Afsar A. Solitary rectal ulcer: a literature review. Ann Colorectal Res 2015;3:e33500.

Erdem D, Acar Y, Karaa EK, et al. A rare and often unrecognized cause of hematochezia and tenesmus in childhood: solitary rectal ulcer syndrome. Pediatrics 2002;110:79.

Contractor TQ, Contractor QQ. Traumatic solitary rectal ulcer in Saudi Arabia. A distinct entity? J Clin Gastroenterol 1995;21:298–300.

Martin de Carpi J, Vilar P, Varea V. Solitary rectal ulcer syndrome in childhood: a rare, benign, and probably misdiagnosed cause of rectal bleeding. Report of three cases. Dis Colon Rectum 2007;50:534–9.

Sharara AI, Azar C, Amr SS, et al. Solitary rectal ulcer syndrome: endoscopic spectrum and review of the literature. Gastrointest Endosc 2005;62:753–62.

Jarrett ME, Emmanuel AV, Vaizey CJ, et al. Behavioural therapy (biofeedback) for solitary rectal ulcer syndrome: a case-matched series. Dis Colon Rectum 1992;35:641–5.

Zhu QC, Shen RR, Qin HL, et al. Solitary rectal ulcer syndrome: clinical findings, surgical treatment, and outcomes. Int J Colorectal Dis 2006;21:348–56.

Yamagawa H. Pronounced variants in solitary rectal syndrome. Acta Pathol Jpn 1998;48:471–8.

Zhu QC, Shen RR, Qin HL, et al. Solitary rectal ulcer syndrome: clinical features, pathophysiology, diagnosis and treatment strategies. World J Gastroenterol 2014;20:738–44.

Tendler DA, Aboudala S, Zack JF, et al. Prolapsing mucosal polyps: an unrecognized form of colonic polypl:a clinicopathologic study of 15 cases. Am J Gastroenterol 2002;97:370–6.

Simsek A, Yagci G, Gorgulu S, et al. Diagnostic features and treatment modalities in solitary rectal ulcer syndrome. Acta Chir Belg 2004;104:92–6.

Rao SS, Go JT. Update on the management of constipation in the elderly: new treatment options. Clin Interv Aging 2010;5:163–71.

Feczko PJ, O’Connell DJ, Riddell RH, et al. Solitary rectal ulcer syndrome: radiologic manifestations. AJR Am J Roentgenol 1980;135:499–506.

Amaeche I, Papagrigoriadis S, Hurbullah S, et al. Solitary rectal ulcer syndrome mimicking neoplastic on MRI. Br J Radiol 2010;83:e221–4.

Van Outryve MJ, Pekelmans PA, Fiersens H, et al. Transrectal ultrasound study of the pathogenesis of solitary rectal syndrome. Gut 1993;34:1422–6.

Cola B, Cucchi D, Dalla via B, et al. Endosonographic pattern of solitary polyoid rectal ulcer. Tech Coloproctol 2005;9:71–2.

Gopal DV, Young C, Katon RM. Solitary rectal ulcer syndrome presenting with rectal prolapse, severe mucorrhea and eroded polyoid hyperplasia: case report and review of the literature. Can J Gastroenterol 2001;15:479–83.

Halligan S, Nicholls RJ, Bartram CI. Proctographic changes after rectopexy for solitary rectal ulcer syndrome and preoperative predictive factors for a successful outcome. Br J Surg 1995;82:314–7.

Halligan S, Sultan A, Rottenberg G, et al. Endosonography of the anal sphincters in solitary rectal ulcer syndrome. Int J Colorectal Dis 1995;10:79–82.

Choi YM, Song HJ, Kim MJ, et al. Solitary rectal ulcer syndrome mimicking rectal cancer. Ewha Med J 2016;39:28–31.

Millward SF, Bayjoo P, Dixon MF, et al. The barium enema appearances in solitary rectal ulcer syndrome. Clin Radiol 1985;36:185–9.

Swatton A. Solitary rectal ulcer syndrome: physiology and treatment options. Br J Nurs 2009;18:1312–5.

Ignaotovic A, Saunders BP, Harbin L, et al. Solitary ‘rectal’ ulcer syndrome in the sigmoid colon. Colorectal Dis 2010;12:1163–4.

Malouf AJ, Vaizey CJ, Kamm MA. Results of behavioral treatment (biofeedback) for solitary rectal ulcer syndrome. Dis Colon Rectum 2001;44:72–6.

van Den Brandt-Gradel V, Hubregtsen K, Tygat GN. Treatment of solitary rectal ulcer syndrome with high-fiber diet and abstention of straining at defecation. Dig Dis Sci 1984;29:1005–8.

Vaizey CJ, Roy AJ, Kamm MA. Prospective evaluation of the treatment of solitary rectal ulcer syndrome with biofeedback. Gut 1997;41:817–20.

Jarrett ME, Emmanuel AV, Vaizey CJ, et al. Behavioural therapy (biofeedback) for solitary rectal ulcer syndrome improves symptoms and mucosal blood flow. Gut 2004;53:368–70.

Daniel F, Siproudis I, Tohme C, et al. Solitary rectal ulcer: another view of the management algorithm. Gastrointest Endosc 2006;63:738–9.

Binne NR, Papachryssotomou M, Clare N, et al. Solitary rectal ulcer: the place of biofeedback and surgery in the treatment of the syndrome. World J Surg 1992;16:836–40.

Zargar SA, Khoroo MS, Mahajan R. Sucralfate retention enemas in surgery for solitary rectal ulcer syndrome. Br J Surg 1998;85:5120.

Ederle A, Bulighin G, Orlandi PG, et al. Endoscopic application of botulinum toxin in solitary rectal ulcer syndrome mimicking rectal neoplasm on MRI. Br J Radiol 2010;83:62.

Keshtgar AS, Ward HC, Sanei A, et al. Botulinum toxin, a new treatment option A. Br J Surg 1995;82:1246.

Katter B, Ward HC, Sanei A, et al. Long-term outcome of laparoscopic mesh rectopexy: a follow-up study of the pathogenesis of solitary rectal syndrome. Gut 1993;34:1422–6.

Keshtgar AS, Ward HC, Sanei A, et al. Botulinum toxin, a new treatment option A. Br J Surg 1995;82:1246.

Katter B, Ward HC, Sanei A, et al. Long-term outcome of laparoscopic mesh rectopexy: a follow-up study of the pathogenesis of solitary rectal syndrome. Gut 1993;34:1422–6.

Keshtgar AS, Ward HC, Sanei A, et al. Botulinum toxin, a new treatment option A. Br J Surg 1995;82:1246.