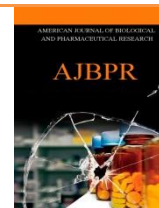




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EFFECTS OF NON-SURGICAL TREATMENT OF 42 CASES WITH SOLITARY RECTAL ULCER- A RETROSPECTIVE STUDY FROM A SINGLE CENTRE OF EASTERN INDIA

Biswarup Bose¹, Amit Kumar Samanta², Kakali Roy Basu³, Abhimanyu Basu^{4*}

¹ Assistant Professor in General Surgery, ICARE Institute of Medical Sciences and Research, Haldia, W.B, India.

² RMO-cum-Clinical Tutor in Surgery, NRS Medical College, Kolkata, India.

³ Professor of Physiology, Institute of Post Graduate Medical Education & Research, Kolkata, India.

⁴ Professor of Surgery. Institute of Post Graduate Medical Education & Research, Kolkata, India.

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ABSTRACT

Solitary rectal ulcer is a rare disorder characterized by erythema or ulceration of the rectal wall, associated with typical histological features, and disturbed defecatory behavior with the passage of blood and mucus. The treatment is difficult and the patients often frustrated. For further characterization, retrospective, a hospital-based review of diagnosed SRU and then the effect of treatment were conducted. 42 cases were diagnosed at Institute of Post Graduate Medical Education & Research and SSKM Hospital, Kolkata between 2000 and 2011 were reviewed and evaluated. Review of literature search (Medline) and manual cross-referencing were also done. 42 cases were identified: 28 males and 14 females. The age range was 17-70 years. Blood and mucus discharge, constipation / tenesmus and pain were the most common presenting symptoms and were seen, either alone or in various combinations. The pathogenesis is likely to vary in different patients: it include trauma from straining, direct digital trauma, and possibly primary neuromuscular pathology. The histopathological findings of extension of the muscularis mucosa between crypts and muscularis propria disorganization on full thickness specimens are characteristic. Behavioral therapy, including habit training, can lead to symptom improvement and return to work in majority of patients. Application of Topical *Sucralfate* long term symptom improvement in 70% Of our patients. Rectal ulceration may persist after any treatment, even if the symptomatology improved. SRU is a rare disorder and only 42 cases were diagnosed at our Institution at Kolkata which mainly serves the population of Eastern India. The clinical pattern of our patients was variable and often mimics clinical presentation of rectal cancer patients. Behavioral therapy, topical Sucralfate and carefully considered surgical interventions offer the best treatment results. Further work on psychological factors and neuromuscular and vascular pathology is required.

INTRODUCTION

The solitary rectal ulcer (SRU) is a benign lesion

predominantly of the adults of either sex, although pediatric populations [1,2] may be affected. Owing to its rarity, SRU is often misdiagnosed as malignant ulcer, or ulcer associated with inflammatory bowel disease. The characteristic appearance of the disease is a “neither *being always ulcerate, nor always solitary*” lesion, but often with

Corresponding Author

Abhimanyu Basu

Email:- ovmanyubasu@yahoo.co.in



polypoid or granular features, typically localized in anterior rectal wall, a few inches from anal canal. Rectal bleeding, constipation and tenesmus, pain were the most common presentations [3]. The histological examination is the key to the diagnosis of SRU. Full thickness rectal biopsy should be taken and histology reveals architectural derangement of the muscularis propria in some patients [4]. The condition is uncommon and difficult to treat [5]. Conservative treatments including laxatives, fiber supplementation and attempted reduction of straining are of no proven benefit and there are no topical agents known to improve this condition, except topical Sucralfate has shown some good results [6].

The cause of this condition is unknown. However, physiological and histological studies suggest a spectrum of disease, and result from more than one cause. In clinical practice some patients seem to have a behavioral disorder with excessive straining, whereas in others there is no history of straining. The encouraging results from the use of behavioral therapies for defecation disorders [7, 8] led us to explore whether patients might benefit from behavioral retraining and topical Sucralfate.

In this study, we evaluate retrospectively the use of behavioral therapy including habit training and topical Sucralfate for SRU in a consecutive unselected group of adults along with review of literatures.

PATIENTS AND METHODS

The purpose of the study is to evaluate clinical presentations and the effect of treatment in patients with SRU. Review of literature search (Medline) and manual cross-referencing were also done. From January 2000 to December 2011, forty two patients (fourteen females, median age 47 (M) and 39(F), age range 17-70 years) were treated and followed up in our Institution. All were undergone colonoscopic biopsies (it might take repeat biopsies and review of slides by two pathologists) and histologically proven SRUs were evaluated and included in this series. All cases were managed with behavioral therapy, bulk laxative and topical application of Sucralfate. Out of them; three cases were undergone surgical intervention (who had associated with rectal prolapsed).

Behavioral therapy (Habit training)

Patients were taught how to strain effectively by using a propulsive force through bracing with their abdominal muscles. They were advised on normal defecatory behavior and bowel habits. This included restricting the number of visits to the toilet (Max three) for patients who made frequent defecatory attempts during the day, or increasing the number of visits to the toilet for those patients with infrequent defecation. The amount of time spent (Max 15min) and posture in the toilet were also specified. They were also told to avoid digitations of rectum.

Bulk laxatives and topical Sucralfate

We prefer Isabgol husk as bulk laxative; 10gm with 500ml of water twice daily (in the morning and evening, not at bed time). Sucralfate (preferably as gel form) to be applied 4-6 times daily (one medium squeeze at a time) with rectal applicator for at least 4-6 weeks.

Questions asked for evaluation of symptoms (before and after therapy started)

Number of visits to toilet / amount of time spent in the toilet / Passage of blood or mucus per rectum / Ineffectual straining at stool / Need to digitate per rectum / Presence of pain / Ability to work.

RESULTS

Clinical findings

Most of our patients with SRU presented with rectal bleeding, perianal pain, tenesmus, and mucus discharge; either alone or in combinations.

Diagnosis

Meticulous history taking (specific questions as described) is very important to reach the diagnosis. Long duration of symptoms without much morbidity is another important aspect in suspecting SRU (Median duration of symptoms was 26 months). History of defective toilet training at childhood may be a cause for developing SRU.

Digital rectal examination and proctoscopy to find shallow ulcer (and /or granularity) on the anterior wall of low rectum is the usual finding. Colonoscopy and biopsy were done in all cases for confirmation of diagnosis and to exclude malignancy; as these cases are often misdiagnosed clinically as rectal cancers.

Therapeutic approach

We assessed our patients, initially in 2 weeks interval until there was symptomatic relief and also to boost habit training. Subsequently these patients were followed up at three to six month interval. The median follow up period after conservative therapy (Behavioral therapy+ Laxative+ Sucralfate) was sixty months. The intense therapy period was continued for 4-6 weeks and results were evaluated. Any recurrence of symptoms may be dealt with in the same fashion. The results are shown in Table 2.

So, there was significant improvement of symptoms (esp. Bleeding, Pain, and tenesmus) and there was also healing of ulcers (? Sucralfate effect). There were recurrences of symptoms but in a less severe manner. Recurrences were also advised again with behavioral therapy, bulk laxatives and Sucralfate.

DISCUSSION

SRU is an uncommon benign condition and the pathogenesis is still unclear. Possibly, intense and prolonged straining at stool (which is almost universal in



patients with SRU) results in trauma and ischemic ulceration of the anterior wall of rectum [11,12]. SRU is actually a misnomer because in many patients, no ulceration is present (only mucosal granularity is seen) and occasionally multiple ulcerations are evident. In general, up to 70% of SRU lesions are located in the anterior rectum [9]. The clinical presentation of SRU is variable. In addition, colonoscopic findings are also diverse and can be of ulcer, polyps or only erythema and granularity. Therefore, SRU can be great mimicker of other serious disorders e.g. IBD and cancers. The diagnosis is almost invariably established by biopsy. The pathognomonic features of SRU reveal muscle fibers are seen streaming out into the lamina propria below. The characteristic features were surface serration, crypts' distortion, and fibromuscular obliteration of the lamina propria [10-14]. The management of SRU is difficult and the patients obtained poor symptomatic relief [15].

In order to study the prevalence of SRU in our hospital population (mostly people are from Eastern India) to further characterize the clinical presentations and management options, we conducted this hospital based retrospective study of SRU in IPGME&R and SSKM Hospital, Kolkata. Also, we felt that the study will make clinicians more aware of this syndrome so that it is less likely to be confused with other conditions.

Between January 2000 and December 2011, we evaluate 42 patients with SRU. This is a small number and indicates the rarity of this condition. IPGME&R and SSKM Hospital, Kolkata is the most prominent Government Teaching hospital of Eastern India and serves a large population (annually more or less 30000 new patients attend at Department of Surgery). In our series, there was male preponderance which was also seen in other reported series from Pakistan, Tunisia and Kuwait [16]. Series from Western countries showed, either equal sex distribution or female preponderance [3, 17]. There was a wide age range (17-70years); other series also demonstrated of wide age range. 15-85 years [16], 14-76 years [3] and 25-86 years [17]. The youngest patient in our series is aged 17 years. There are only a few reports of patients with SRU in the younger age groups [1,2,18]. All patients in our series reported to the hospital because of their clinical symptoms. Rectal bleeding / mucus discharge, ineffectual straining at stool (tenesmus), and perianal pain was the most common finding (Table 1). The symptomatology are consistent with the other reported series also [3, 9,10,14,17]. The bleeding is likely due to ulceration of the mucosa. Surprisingly, some of the other studies have reported a higher proportion of asymptomatic patients, where SRU was diagnosed incidentally during colonoscopy done for cancer screening or polyp surveillance. Tjandra reported that 26% of his series were asymptomatic. Rectal digitations and self-inflicted injury has been claimed to contribute to rectal injury [19] and this has been reported in

up to 28% of the patients in some other series [14]. In our observation and careful clinical history, it was revealed that up to 70% of our patients had practiced rectal digitations (Table 1).

Colonoscopic findings are important for the diagnosis of SRU. All (n=42) of the patients in our series had rectal ulceration or granularity. Some of them had multiple ulcers and some presented with polyps. Thus, it is obvious that the designation 'solitary ulcer' is a misleading. Other series, such as the one published by Torres *et al.*, [20] had similar findings, with 65.3% of the patients reported to have ulceration. In contrast, Tjandra *et al.* [3] reported that 29% of their series had ulcers and 44% presented with polyps. Transrectal ultrasound (TRUS) is also used in patients with SRU who present with chronic constipation to exclude other causes e.g., sphincter defects [21]. Two of our cases had undergone TRUS, who were clinically diagnosed of rectal cancers.

Histopathology is the key to the diagnosis of SRU. A combination of fibromuscular obliteration of the lamina propria, crypts' distortion, and surface serration can establish the diagnosis in most cases. Some combination of these features was seen in all the patients in this study. Other authors have also reported that these features are the most common. In the series reported by Tendler *et al.*, [22] some reported 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. Different vascular changes have been noted in biopsies of SRU. Lonsdale in his series [23], reported that ectasia with congestion was seen in 95% of cases. Another common feature he noted was muscularized capillaries, which were seen in 50% of his cases. Less common features were thrombosis, fibrin deposition, and atherosclerosis. Lee and his colleagues, also reported that thickened internal anal sphincter was a typical finding in SRUS. Our study revealed similar findings [22].

The treatment of SRU is very difficult and symptomatic relief of the patients is poor. Medical management should be attempted in all cases except those patients with complete rectal prolapse. Avoidance of straining, bulk laxatives, stimulating suppositories /enema – one study had shown very impressive result (70% of the patients were improved with healing of ulcers). [24]. Topical medications have had variable success in treating SRU. Although glucocorticoids applied topically and sulfasalazine enema are not efficacious; Sucralfate (as topical & retention enemas) has shown symptomatic improvements and healing of ulcers [6, 25]. We follow the



treatment of bulk laxative, Sucralfate topical cream / gel and behavioral therapy (bowel habit training) for 4-6 weeks in intensely and our results are encouraging. Overall, about 80% of our patients were symptomatically improved; especially, rectal bleeding, tenesmus and pain are diminished and there is significant healing of ulcers too (24 out of 42 cases) (Table2). Study from Mississippi Medical Center, USA also shown good results with this conservative regimen [26]. Gut directed biofeedback therapy also shown

promising results in refractory cases of SRU [7,8] but long term follow-up revealed poor symptomatic relief [14,27]. Argon plasma coagulation (APC) may represent a therapeutic approach for bleeding SRU, even if controlled studies are necessary before recommending it as an acceptable treatment [28]. Somani *et al.* from Lucknow, has shown that APC controls bleeding in patients with SRU and it also improves the healing of these ulcers [29].

Table 1. Presenting Features (%) IPGME&R (2000-11)

Presenting Features (%)	IPGME&R (2000-11)	Madigan & Morson (1969) [9]	Britto et al.(1987) [10]
Rectal bleeding	90	91	80
Pain	60	42	25
Tenesmus	80	24	30
Mucus discharge	85	68	45
Need to digitate	70	---	---
Ulceration /Granularity	100	---	---

Table 2. Healing of ulcer

Symptomatology	On first visit	At subsequent visit
Number of visits to toilet	Median 5	Median 3
Time spent in the toilet	Median 25 min	Median 15min
Blood or mucus per rectum	38	3 (infrequent)
Ineffectual straining at stool	33	15
Need to digitate per rectum	30	10
Presence of pain	25	10 (infrequent)
Healing of ulcer	42 (Ulcer present)	24 (Ulcer healed)

Fig 1. Trans rectal US

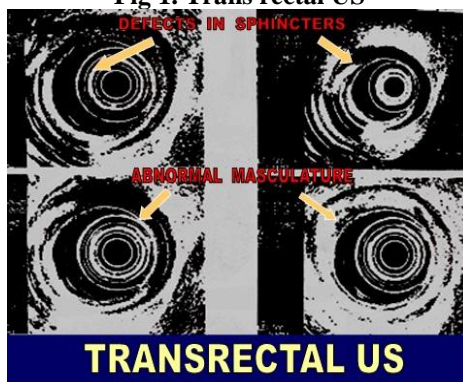


Fig 2. Solitary rectal ulcer (Colonoscopy)

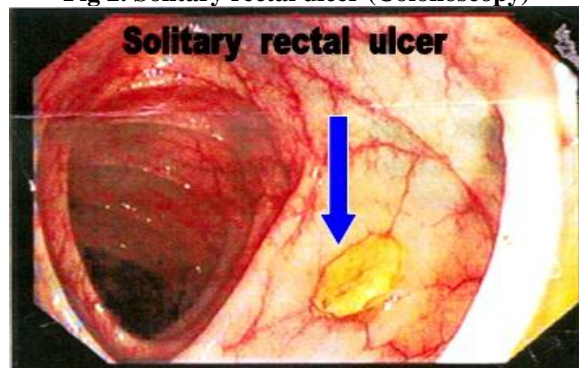


Fig 3. Areas of erythema and granularity (Colonoscopy)

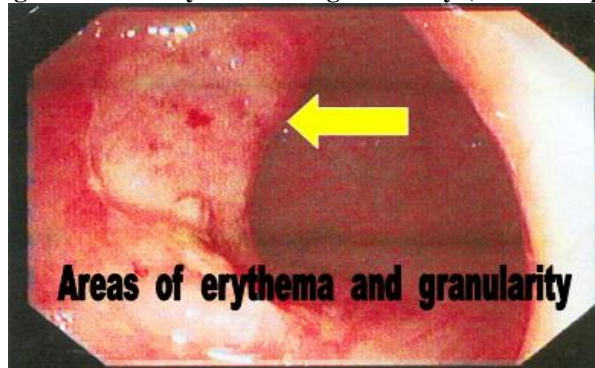
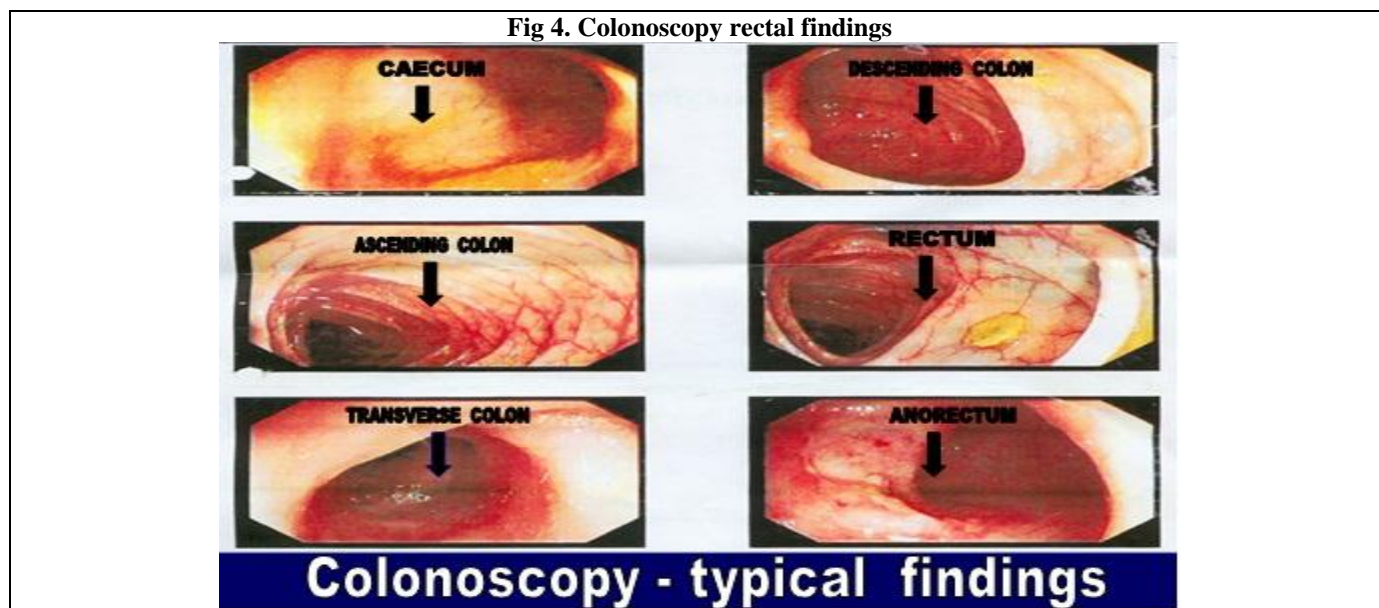


Fig 4. Colonoscopy rectal findings



CONCLUSION

In conclusion, this study, to the best of our knowledge, the first study of this rare disease from Eastern India and shows that SRU in our area has similar epidemiological, clinical, pathological characteristics to SRU from other areas (esp. Asian countries). The clinical presentations are variable but the combinations of perianal pain, blood /mucus discharge and ineffectual straining at stool are most common. Most of the time, these cases are misdiagnosed clinically as IBD or malignancy. So, careful and meticulous history and digital rectal examination is very much necessary. Colonoscopic biopsy and

histopathology is mainstay of diagnosis. Though the treatment is difficult and often unsatisfactory, but the combinations of habit training, bulk laxative and topical Sucralfate are encouraging with healing of ulcers and symptomatic improvements.

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None

CONFLICT OF INTEREST

The authors declare that they have no conflicts of interest.

REFERENCES

1. Dehghani SM, Malekpour A, Haghtghat M. (2012). Solitary rectal ulcer syndrome in children: a literature review. *World J Gastroenterol*, 18, 6541-6545.
2. Keshtgar AS. (2008). Solitary rectal ulcer syndrome in children. *Eur J Gastroenterol Hepatol.*, 20(2), 89-92.
3. Tjandra JJ, Fazio VW, Church JM, Lavery IC, Oakley JR, Milsom JW. (1992). Clinical conundrum of solitary rectal ulcer. *Dis Colon Rectum*, 35, 227-34.
4. Kang YS, Kamm MA, Engel AF, Talbot IC. (1996). Pathology of the rectal wall in solitary rectal ulcer syndrome and complete rectal prolapse. *Gut*, 38, 587-90.
5. Qing Z, Rong R, Yu W, et al. (2014). Solitary rectal ulcer syndrome: clinical features, pathophysiology, diagnosis and treatment strategies. *World J Gastroenterol*, 20(3), 738-744.
6. Zagar S, Khuroo M, Mahajan R, et al. (1991). Sucralfate retention enema in solitary rectal ulcer. *Dis Colon Rectum*, 34, 455.
7. Rao SS, Ozturk R, De Ocampo S, Stessman M. (2006). Pathophysiology and role of biofeedback therapy in solitary rectal ulcer syndrome. *Am J Gastroenterol.*, 101(3), 613-18.
8. Jarrett ME, Emmanuel AV, Vaizey CJ, Kamm MA. (2004). Behavioural therapy (biofeedback) for solitary rectal ulcer syndrome improves symptoms and mucosal blood flow. *Gut.*, 53(3), 368-70.
9. Madigan MR, Morson BC. (1969). Solitary ulcer of the rectum. *Gut*, 10, 871.
10. Britto E, Borges AM. (1987). Solitary rectal ulcer: 20 cases. *Dis Colon Rectum*, 30, 381-86.
11. Rutter KRP, Riddell RH. (1975). Solitary rectal ulcer syndrome of the rectum. *Clinic Gastroenterol*, 4, 505.
12. Haray PN, Morris-Stiff GJ, Foster ME. (1997). Solitary rectal ulcer syndrome-An underdiagnosed condition. *Int J*



- Colorectal Dis*, 12, 313-5.
13. Abbasi A, Bhutto AR, Taj A *et al.* (2015). Solitary Rectal Ulcer Syndrome: Demographic, Clinical, Endoscopic and Histological Panorama. *J Coll Physian Surg Pak*, 25(12), 867-9.
 14. Ennaifer R, Elleuch N, Hefaiiedh R, Romdhana H *et al.* (2015). Solitary rectal ulcer syndrome. A Tunisian monocentric survey. *Tunis MO*, 93(1), 6-10.
 15. Meurette G, Siproudhis L, Regenet N, Frampas E, Proux M, Lehur PA. (2008). Poor symptomatic relief and quality of life in patients treated for "solitary rectal ulcer syndrome without external rectal prolapse. *Int J Colorectal Dis.*, 23(5), 521-6.
 16. Al-Brahim N, Al-Awadhi N, Al-Enezi S, Alsurayei S, Ahmad M. (2009). Solitary rectal ulcer syndrome: A clinicopathological study of 13 cases. *Saudi J Gastroenterol*, 15, 188-92.
 17. Marchal F, Bresler L, Brunaud L, Adler SC, Sebbag H, Tortuyaux JM, *et al.* (2001). Solitary rectal ulcer syndrome: A series of 13 patients operated with a mean follow-up of 4.5 years. *Int J Colorectal Dis*, 16, 228-33.
 18. Dehghani SM, Haghghat M, Imanieh MH, Geramizadeh B. (2008). Solitary rectal ulcer syndrome in children: A prospective study of cases from southern Iran. *Eur J Gastroenterol Hepatol*, 20, 93-5.
 19. Wong WM, Lai KC, Shek TW, Lam SK. (2002). Self-inflicted rectal ulcer with exuberant granulation tissue: A lesion that mimics carcinoma. *Gastrointest Endosc*, 55, 951-95.
 20. Torres C, Khaikin M, Bracho J, Luo CH, Weiss EG, Sands DR, *et al.* (2007). Solitary rectal ulcer syndrome: Clinical findings, surgical treatment, and outcomes. *Int J Colorectal Dis*, 22, 1389-93.
 21. Bharucha AE. (2006). Update of tests of colon and rectal structure and function. *J Clin Gastroenterol*, 40, 96-103.
 22. Lee TH, Hang ST, Lee JS. (2015). Thickened internal anal sphincter has been reported to be a typical finding in SRUS. *J Neurogut*, 21(1), 140-1.
 23. Lonsdale RN. (1993). Microvascular abnormalities in the mucosal prolapse syndrome. *Gut*, 34, 106-9.
 24. Brandt- Gradel V, Tytgat GNJ. (1984). Treatment of solitary rectal ulcer syndrome with high- fiber diet and abstention from straining at defecation. *Dig Dis Sci*, 29, 1005.
 25. Tavakkoli H, *et al.* (2008). Efficacy of Sucralfate Enema in Solitary Rectal Ulcer Syndrome. *Gastroenterology*, 134(4), 408-409.
 26. Bishop PR, Nowicki MJ. (2002). Non surgical therapy for solitary rectal ulcer syndrome. *Current Treatment Options in Gastroenterol*, 5, 215-223.
 27. Rao SS, Benninga Ma, Bharucfa AE, Chiarioni G *et al.* (2015). ANMS-ESNM position paper and consensus guidelines on biofeedback therapy for anorectal disorder. *Neurogastroenterol Motil*, 27(5), 594-609.
 28. Waniczek D, Rdes J, Rudzki MK *et al.* (2014). Effective treatment of SRUS using argon plasma coagulation. *Prz Gastroenterol*, 9(4), 249-53.
 29. Somani SK, Ghosh A, Avasthi G *et al.* (2010). Healing of solitary rectal ulcers with multiple sessions of argon plasma coagulation. *Digestive Endoscopy*, 22(2), 107-111.

