

Biofeedback efficacy to improve clinical symptoms and endoscopic signs of solitary rectal ulcer syndrome

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Abstract

Solitary rectal ulcer syndrome (SRUS) is often resistant to medical and surgical treatment. This study assessed the effect of biofeedback in decreasing the symptoms and the healing of endoscopic signs in SRUS patients. Before starting the treatment, endoscopy and colorectal manometry was performed to evaluate dyssynergic defecation. Patients were followed every four weeks, and during each visit their response to treatment was evaluated regarding to manometry pattern. After at least 50% improvement in manometry parameters, recipients underwent rectosigmoidoscopy. Endoscopic response to biofeedback treatment and clinical symptoms were investigated. Duration of symptoms was 43.11 ± 36.42 months in responder and 63.9 ± 45.74 months in non-responder group ($P=0.22$). There were more ulcers in non-responder group than responder group (1.50 ± 0.71 versus 1.33 ± 0.71 before and 1.30 ± 0.95 versus 0.67 ± 0.50 after biofeedback), although the difference was not significant ($P=0.604$, 0.10 respectively). The most prevalent symptoms were constipation (79%), rectal bleeding (68%) and anorectal pain (53%). The most notable improvement in symptoms after biofeedback occurred in abdominal pain and incomplete evacuation, and the least was seen in mucosal discharge and toilet waiting as shown in the bar chart. Endoscopic cure was observed in 4 of 10 patients of the non-responder group while 8 patients in responder group experienced endoscopic improvement. It seems that biofeedback has significant effect for pathophysiologic symptoms such as incomplete evacuation and obstructive defecation. Improvement of clinical symptoms does not mean endoscopic cure; so to demonstrate remission the patients have to go under rectosigmoidoscopy.

Key Words: Solitary rectal ulcer syndrome, endoscopy, colorectal manometry, dyssynergic

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The solitary rectal ulcer syndrome (SRUS) is a benign chronic disorder in young adults.¹ SRUS is characterized by a combination of symptoms, clinical findings and histological abnormalities. Ulcers are only found in 40% of the patients; 20% of the patients have a solitary ulcer, and the rest of the lesions vary in shape and size, from hyperemic mucosa to broad-based polypoid. Men and women are affected equally, with a small predominance in women. SRUS has also been

described in children and in the geriatric population. The pathogenesis of the solitary rectal ulcer (SRU) has not been yet clearly identified. As previous studies have shown, there is a reduction of hindgut mucosal blood flow in patients with functional constipation that can be increased with successful biofeedback therapy.² SRU is uncommon and its treatment is difficult.³ For treating SRU, a wide range of therapeutic interventions from behavioral modification to surgery have been proposed. Generally there are two categories of treatment: surgical

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and nonsurgical.⁴ Surgical modalities were reported to be effective for patients whose bleeding was so severe that made it necessary to transfuse them 10 pints of blood in a month.⁵ They are also effective for treating rectal prolapse (either with or without resection of the lesion)⁶ and those who are resistant to conservative therapies.⁷ There is no proven benefit for conservative treatments like fiber supplementation, laxatives, and attempted reduction of straining.⁸ While no history of straining has been found in some patients, a behavioral disorder seems to be present in others, which is characterized by excessive straining. The good results achieved by behavioral therapies for defecation disorders made us to investigate the probable benefits of biofeedback retraining for SRU patients.⁹ There are only a few therapeutic control trials, none of them has led to a definitive treatment.¹⁰ Some previous studies have used biofeedback and behavioral approaches as the first line of treatment.¹¹ This approach has led to about 54-75% improvement of symptoms and sigmoidoscopic resolution of ulcer in 30% of cases.¹²⁻¹⁵ In another study, complete ulcer healing was observed in about 28% of patients with SRU and 33% of them were resistant to rectopexy.⁹ Regarding the facts that SRU has no definite treatment and the mean age of patients is 48 years old and it can disturb one's diurnal function chronically and it causes absence of work, this treatment seems to be cost effective. The latest studies in this field have just investigated the symptoms of patients after biofeedback therapy, so this study aimed to evaluate both clinical symptoms and endoscopic signs during the treatment period to find out a staging and scoring for this therapeutic approach.¹⁵

Materials and Methods

In this prospective study, 19 patients who suffered with gastroenterology and liver diseases with diagnosis of

SRU were referred to the Motility Department of Taleghani Hospital, Tehran, Iran from June 2013 to July 2014. These patients, evaluated for dyssynergic defecation, had been analyzed by endoscopy and pathology. A questionnaire on the symptoms and biofeedback therapy complaints, i.e., bowel frequency, increased time spent on toilet, straining, rectal blood loss, rectal mucus loss, need to anal digitation, sensation of incomplete emptying, abdominal pain, constipation, diarrhea, obstructive defecation and tenesmus was filled out for all patients by a trained nurse. Inclusion criteria included having bowel frequency, increased time spent on toilet, straining, rectal blood loss, rectal mucus loss, need to anal digitation, sensation of incomplete emptying, abdominal pain, constipation, diarrhea, obstructive defecation and tenesmus, who were diagnosed having SRU after confirmation by colonoscopy and rectal pathology and showed evidence of dyssynergic defecation in rectal manometry. They had no other problem in colonoscopy. Patients who candidate for anal surgery, having history of psychiatric disorders [Hospital Anxiety & Depression Scaling (HADS) scores 11 or more], haemorrhoid grade 3 or 4, under medical treatment, constipation, acute or chronic anal fissure, overt rectal prolapse (by taking photographs with their cellphones), no response to biofeedback after 12 sessions based on manometry parameters, were excluded from the study. All patients after medical examination by a physician were submitted to routine blood and stool tests (microscopy and culture) for infections.

Demographic and clinical data included age, sex, pharmacological treatment, clinical presentation, constipation scoring system form,^{16,17} ROME3 and SRUS symptoms (table1), duration of symptoms, HADS.¹⁸ The CSS consists of seven items that are scored using a five-point scoring system that ranges

Table 1. Comparison based on the response to biofeedback.

	Responder	Non-Responder	p-value
Male gender	5	4	0.509
Age(yr)	30.67 ±13.59	32.6 ±13.97	0.54
Duration(m)	43.11 ±36.42	63.9 ± 45.74	0.22
Number Of Ulcers before	1.33 ±0.71	1.50 ± 0.71	0.604
Number of Ulcers after	0.67 ± 0.50	1.30 ±0.95	0.1
Type of ulcers before			0.672
Endoscopic Improvement			0.027

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from 0 (none of the time) to 4 (all of the time) and one item that is rated on a 0–2 scale. The range of total score is from 0 (normal) to 30 (severe constipation). A cutoff score of 15 suggests constipation.¹⁶ Before starting therapeutic interventions, endoscopy and colorectal manometry (with eight channel high resolution manometry device from MMS company) was performed (by an expert gastroenterologist) to confirm dyssynergic defecation. The number of lesions, their type and location were recorded during endoscopy. The criteria investigated via manometry included: maximum squeeze pressure, sustained squeezing pressure and push maneuver. Definition of dyssynergic defecation was based on manometry: if 2/3 of manometric criteria were present, the diagnosis was confirmed. The patients filled consent forms after a complete description of the methodology. Management began with instructing all the patients by a resident of internal medicine. Patients were strongly advised to maintain regular bowel habits, and to avoid excessive straining during bowel evacuation. They were also asked to desist from doing manual evacuation. Patients then were treated with biofeedback. Biofeedback is a process, which allows an individual to learn how to alter his body's physiological activity in order to improve health and performance. Biofeedback (verbal, visual) was done by a trained person. The visual biofeedback was provided by watching changes in pressure activity on the computer monitor. During biofeedback training sessions the patients were asked to lie down in right lateral position while being covered with a sheet. Initially the patients were ordered to relax and then to squeeze or strain gently for 10 seconds and repeat this process for many times to observe the changes. During biofeedback sessions we trained the patients to perform pelvic exercises and modifying their defecation habits via verbal instructions. Each biofeedback session lasted approximately 30-45 minutes. Patients were followed every 4 weeks and during each visit their response to treatment was measured via manometry. Response to

biofeedback was defined as an improvement of at least 50% in manometric criteria. After detecting a response to biofeedback, clinical symptoms (CSS and SRU symptoms) and endoscopic response were investigated. Patients who did not respond to biofeedback after a maximum of 12 sessions were excluded from the study. Response to treatment was categorized into one of four groups: major improvement (at least 80% reduction in symptoms), fair improvement (symptoms decreased more than 50%), mild improvement (symptoms decreased less than 50%), and none (less than 30% symptom reduction). Then the statistical analysis was performed separately in one of the two groups: responder group (with major or fair improvement) and non-responder group (with mild or no improvement). Endoscopic improvement was defined as going one stage back (changing from ulcer to polyp or from ulcer/polyp to erythema) and/or at least 50% reduction in size of the lesion. Endoscopic response was defined into two groups: improved group included one stage back (changing ulcer to polyp or ulcer/polyp to erythema) or decrease at least 50% in size of lesion. Unimproved group included otherwise conditions.

Statistical analysis

Test of normality for distribution of variables was performed using the Kolmogorov–Smirnov test. Qualitative variables were analysed by chi-square test and quantitative variables with a student t- test. Data are expressed as mean \pm SD. Mann-Whitney u test was used to compare differences between both groups. We used default selection criteria of SPSS 19.0 for Windows (SPSS, Chicago, IL).

Results and Discussion

In this study were enrolled 19 patients from a total of 48 SRU patients (mean age was 31.68 ± 13.44 years). Average number of biofeedback sessions was 6.9 (5 to 10). Patients with at least a 50% improvement in the manometric parameters were more than 80% of enrolled

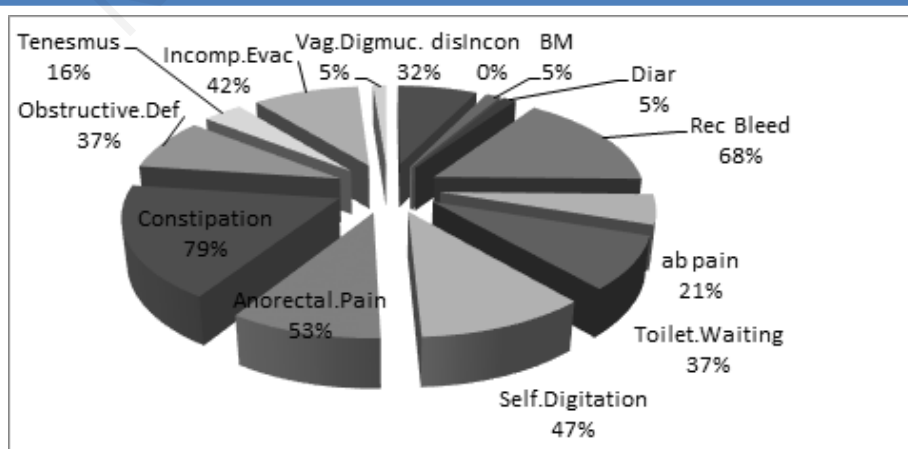


Fig 1: Prevalence of symptoms

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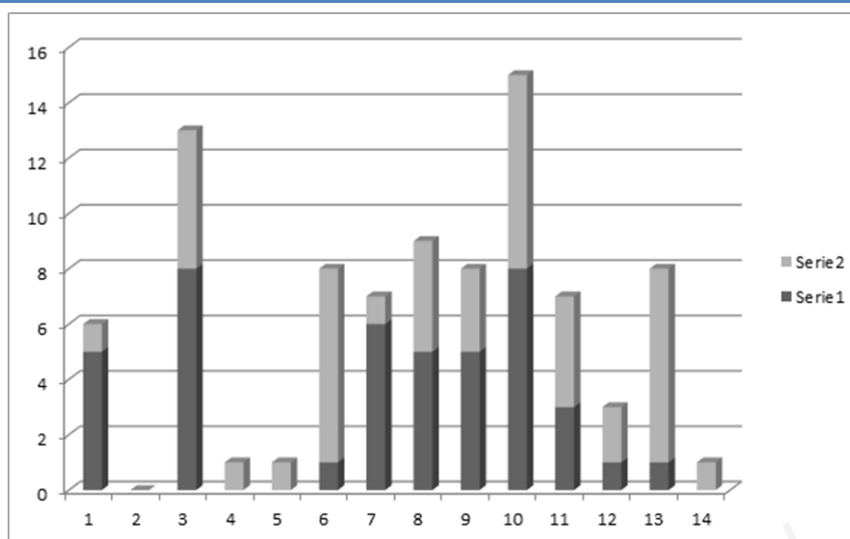


Fig 2. Improvement in symptoms after biofeedback

subjects. In 50% of cases decrease in size of the lesion has been observed. Demographic characteristics of the study population is presented in table 1. After completion of biofeedback sessions, patients were divided into two groups (responder and non-responder) based on clinical improvement (CSS form, SRU symptoms that were reported by the patients). Improvement in symptoms consider as; major (> 80%) and fair (50% to 80%) improvement in patients' symptoms as "responder" and mild (30% to 50%) or no improvement (<30%) as "non-responder". There is no significant difference in sex and age distribution ($p = 0.5$). Duration of symptoms was 43.11 ± 36.42 months in responder and 63.9 ± 45.74 months in non-responder group ($P=0.22$). There were more ulcers in non-responder group than responder group (1.50 ± 0.71 versus 1.33 ± 0.71 before and 1.30 ± 0.95 versus 0.67 ± 0.50 after biofeedback), although the difference was not significant ($P=0.604, 0.10$ respectively) (table 1). Our results showed three types of solitary rectal ulcers, defined erythema, polyp, and ulcer. Before biofeedback therapy 6 patients had polyp and 13 had ulcer. Two of the polyps changed to erythema, one of them had no change and others healed. Figure 1 shows that the most prevalent symptoms were constipation (79%), rectal bleeding (68%) and anorectal pain (53%). Less common symptoms included incontinency, diarrhea and increased bowel motion. The most notable improvement in symptoms after biofeedback occurred in abdominal pain and incomplete evacuation, and the least was seen in mucosal discharge and toilet waiting as shown in the bar chart (Figure 2). The average number of biofeedback sessions was 6.5 times in responder group and 7.1 times in the non-responder group. There was any significant difference between the two groups (p -value = 0.38). Significant difference between responder and non-responder groups was endoscopic improvement

as shown in the cross tabulation below. Endoscopic improvement was observed in 4 of 10 patients of the non-responder group while in 8 of 9 patients in responder group, which implied a significant difference between them ($p = 0.027$). The data of this study show that biofeedback is more effective in polypoid type lesion than in ulcerative type. Most of the previous studies have investigated only clinical improvement without endoscopic evaluation. In our study there was significant difference in endoscopic findings between responder group and non-responder group (p -value = 0.027) but no significant difference found in clinical responses between the two groups in regard to the duration of symptoms, type and number of ulcers and age; although in non-responder group the number of ulcers and duration of symptoms was greater and age of patients was older. It means that real SRUS improvement resulted from endoscopic improvement and not merely a consequence of symptom therapy. SRUS is an uncommon, benign and chronic disorder often diagnosed in young adults and middle-aged, and is usually related to straining or abnormal defecation. Several treatments have been suggested for this disorder, including topical medications, behavioral modification in combination with fiber supplements and biofeedback, and surgery. Patient education is required for good management of this disorder along with a stepwise conservative approach individualized for each patient.¹⁹

Patients who only suffer from mild symptoms without any rectal prolapse are considered appropriate for conservative management in which stool bulking agents are used in conjunction with biofeedback therapy for retraining the patients' bowel habits. Tjandra et al claimed that preventing the paradoxical contraction of puborectalis muscle and managing constipation are the main goals of therapy.²⁰ Whereas, Halligan showed in

patients suffering from rectal prolapse, conservative management alone is rarely effective and hence surgery (usually abdominal rectopexy) may become necessary.²¹ Palsson and Jorge in separated studies reported the efficacy of biofeedback therapy in managing functional anorectal disorders like functional constipation and functional fecal incontinence.^{22, 23} Previous studies have confirmed that for many SRUS patients gut directed biofeedback could be an effective behavioral treatment.^{12, 24,25} In those studies about 75% of patients reported subjective improvement.²⁴ Vaizey et al. presenting endoscopic evaluation of these patients showed that the rectal ulcer had healed in 31% of them after biofeedback therapy.²⁵ Behavioral treatment could change the bowel function and its blood flow by several mechanisms. Improvement of mucosal microcirculation and bowel transit may result from changes in autonomic innervations of the bowels via some cerebral mechanisms. Previous studies have shown that extrinsic autonomic innervations to the bowels exhibit changes in their activity level which are correlated to psychological factors.²⁶ The increased blood flow to the rectal mucosa may also be attributed to the improved psycho-social functioning as a consequence of behavioral treatment. Another possible mechanism for enhanced blood flow may be the improved rectal motor function.²⁷ Furthermore, during the last few years multiple studies have been published similar to those described above.²⁸⁻³⁰ A behavioral approach seems to be of therapeutic benefit for some SRU patients. It often makes them feel subjectively better, improves many symptoms associated with bowel function, and allows some patients to go back to work again. Since in some patients SRUS may be related to chronic straining, it seems rational to use retraining toileting behavior as a treatment strategy. Biofeedback therapy does not solely include retraining co-ordination of pelvic floor muscles. In this approach patients are also taught the appropriate posture and correct use the abdominal muscles during defecation, and they are made to follow a discipline about the amount of time being spent in the toilet, number of visits to the toilet, self digitation, and using laxatives. It also brings them psychological support. Therefore the term “biofeedback” in this context entails a complex entity composed of several complementary parts such as behavioral conditioning, paying more attention to the defecatory process and likely other psychological factors. The most important limitation of this study was the study population. Another limitation was related to the last of follow-ups. This problem was resolved with calling back the participants and explaining the importance of follow-up visits in the treatment process, though ultimately some cases had to be excluded from the study. In conclusion, it seems that Biofeedback is more effective in polypoid type of lesion than in ulcerative type, though to reach conclusive results require a larger sample size.

List of acronyms

SRU - solitary rectal ulcer
SRUS - solitary rectal ulcer syndrome
HADS - Hospital Anxiety & Depression Scaling
ROME3 - Diagnostic Criteria for Functional Gastrointestinal Disorders.
CSS - Constipation scoring system.

Author's contributions

Authors equally contributed to the manuscript.

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Conflict of Interest

The authors report no conflicts of interests.

Ethical Publication Statement

We confirm that we have read the Journal's position on issues involved in ethical publication and affirm that this report is consistent with those guidelines.

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References

1. Martin CJ, Parks TG, Biggart JD. Solitary rectal ulcer syndrome in Northern Ireland. 1971–1980. *Br J Surg* 1981; 68:744–7.
2. David J Kim MD, Peter A L Bonis MD. Solitary rectal ulcer syndrome; Up to date 2014; available at URL: <http://www.uptodate.com>.
3. Haray PN, Morris-Stiff GJ, Foster ME. Solitary rectal ulcer syndrome — an under-diagnosed condition. *Int J Colorectal Dis* 1997;12:313–5.
4. Marshall JK, Irvine EJ. Rectal aminosalicylate therapy for distal ulcerative colitis: a meta-analysis. *Aliment Pharmacol Ther* 1995;9:293–300.
5. Madigan MR, Morson BC. Solitary ulcer of the rectum. *Gut* 1969;10:871–81.
6. Meurette G, Regenet N, Frampas E, Sagan C, Le Borgne J, Lehur PA. The solitary rectal ulcer syndrome. *Gastroenterol Clin Biol* 2006;30:382–90.
7. Emmanuel AV, Kamm MA. Response to a behavioural treatment, biofeedback, in constipated

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- patients is associated with improved gut transit and autonomic innervation. *Gut* 2001;49:214–19.
8. Vaizey CJ, van den Bogaerde JB, Emmanuel AV. Solitary rectal ulcer syndrome. *Br J Surg* 1998;85:1617–23.
 9. Lam TC, Lubowski DZ, King DW. Solitary rectal ulcer syndrome. *Baillieres Clin Gastroenterol* 1992;6:129–43.
 10. Kargari S, Salmanroughani H. Laparoscopic Rectopexy in Solitary Rectal Ulcer. *Acta Medica Iranica* 2011;49:810-3.
 11. Malouf AJ, Vaizey CJ, Kamm MA. Results of behavioral treatment (biofeedback) for solitary rectal ulcer syndrome. *Dis Colon Rectum* 2001;44:72-6.
 12. Jarrett MED, Emmanuel AV, Vaizey CJ, Akamm M. Behavioral therapy (biofeedback) for solitary rectal ulcer syndrome improves symptoms and mucosal blood flow. *Gut* 2004;53:368-73
 13. Vaizey C, Roy A, Kamm M. Prospective evaluation of the treatment of solitary rectal ulcer syndrome with biofeedback. *Gut* 1997;41:817-20
 14. Times ML, Craig A, Reickert MD. Functional Anorectal Disorders. *Clin Colon Rectal Surg* 2005;18:109-15.
 15. Rao SS, Ozturk R, De Ocampo S, Stessman M. Pathophysiology and role of biofeedback therapy in solitary rectal ulcer syndrome. *Am J Gastroenterol* 2006;101:613-8.
 16. Sharma SH, Agarwal BB. Scoring Systems in Evaluation of Constipation and Obstructed Defecation Syndrome (ODS). *JIMSA* 2012;25:57-9
 17. Agachan F, Chen T, Pfeifer J, Reissman P, Wexner SD. A constipation scoring system to simplify evaluation and management of constipated patients. *Dis Colon Rectum* 1996;39:681-5.
 18. Zigmond AS, Snaith RP. The hospital anxiety and depression scale. *Acta Psychiatr Scand* 1983;67(6):361-70
 19. Zhu QC, Shen RR, Qin HL, Wang Y. Solitary rectal ulcer syndrome: Clinical features, pathophysiology, diagnosis and treatment strategies. *World J Gastroenterol* 2014; 20(3):738–44.
 20. Tjandra JJ, Fazio VW, Church JM, et al. Clinical conundrum of solitary rectal ulcer. *Dis Colon Rectum* 1992;35:227.
 21. 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.
 22. Palsson OS, Heymen S, Whitehead WE. Biofeedback treatment for functional anorectal disorders: a comprehensive efficacy review. *Appl Psychophysiol Biofeedback* 2004;29:153–74.
 23. Jorge JM, Habr-Gama A, Wexner SD. Biofeedback therapy in the colon and rectal practice. *Appl Psychophysiol Biofeedback* 2003;28:47–61
 24. Malouf AJ, Vaizey CJ, Kamm MA. Results of behavioral treatment (biofeedback) for solitary rectal ulcer syndrome. *Dis Colon Rectum* 2001;44:72–6.
 25. Vaizey CJ, Roy AJ, Kamm MA. Prospective evaluation of the treatment of solitary rectal ulcer syndrome with biofeedback. *Gut* 1997; 41:817–20.
 26. Emmanuel AV, Mason HJ, Kamm MA. Relationship between psychological state and level of activity of extrinsic gut innervation in patients with a functional gut disorder. *Gut* 2001;49:209–13
 27. Murray CDR, Emmanuel AV, Kamm MA. Biofeedback, not laxatives, improves symptoms, transit and autonomic tone in functional constipation. *Gut* 2003;52(suppl 1):A15.
 28. Haghghi S, Molaei M, Foroughi F, et al. Role of endoscopic ultrasound in evaluation of pancreatic neuroendocrine tumors - report of 22 cases from a tertiary center in iran. *Asian Pac J Cancer Preven* 2012;13:4537-40.
 29. Hosseinzadeh ST, Poorsaadati S, Radkani B, Forootan M. Psychological disorders in patients with chronic constipation. *Gastroenterol Hepatol Bed Bench* 2011;4:159-63.
 30. Foroutan M, Loloie B, Irvani S, Azargashb E. Accuracy of rapid urease test in diagnosing *Helicobacter pylori* infection in patients using NSAIDs. *Saudi J Gastroenterol* 2010;16:110-2.

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