https://doi.org/10.29309/TPMJ/2022.29.10.7168

The Professional Medical Journal
www.theprofesional.com
2022, Volume, 29 Issue, 10

ORIGINAL ARTICLE

Solitary rectal ulcer syndrome: Role of hydrocortisone enema in pediatric patients.

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Article Citation: Hammad A, Aziz MT, Khosa GK, Sheikh MA. Solitary rectal ulcer syndrome: Role of hydrocortisone enema in pediatric patients. Professional Med J 2022; 29(10):1563-1568. https://doi.org/10.29309/TPMJ/2022.29.09.7168

ABSTRACT… Objective: To evaluate the effectiveness of adding hydrocortisone enema in the traditional management of Solitary rectal ulcers (SRU) in pediatric patients. **Study Design:** Randomized Controlled Trial. **Setting:** Department of Pediatric Gastroenterology, Children's Hospital and The Institute of Child's Heath, Multan, Pakistan. **Period:** January, 2021 till December, 2021. **Material & Methods:** A total of 52 children (15 years or younger) diagnosed to have SRU were included. Demographic data, clinical and colonoscopic findings were collected. Children were randomly divided into two groups. Group-A received only conventional therapy while Group-B received hydrocortisone enema in addition to conventional therapy. After starting treatment, patients were followed up every 15 days up to duration of 6 months and response to the treatment in terms of complete resolution of symptoms was noted. **Results:** The Mean age of 52 children was 9 + 1.8 years. Twenty eight (53.8%) were boys. The mean duration of symptoms was 5 + 1.4 months. Most common presenting features were per-rectal bleed 48 (92.3%) and mucus in stool 27 (51.9%). It was found that 14/25 (56.0%) children in Group-A reported complete resolution of symptoms in comparison to 20/24 (83.3%) in Group-B (p=0.0380) at 2-months post treatment follow up. Forty one children completed the 6-months follow up period and included in the final analysis. It was found that 10/21 (47.6%) children reported completed resolution of symptoms while Group-B, 18/20 (90.0%) children had complete resolution of symptoms (p=0.0036) at 6-months follow up. **Conclusion:** In children with SRUS, hydrocortisone enemas along with conventional treatment were found to be significantly more useful in resolution of pretreatment symptoms in comparison to conventional treatment alone. Further studies involving large sets of population are needed to verify the findings of present study.

Key words: Colonoscopy, Conventional, Hydrocortisone enema, Solitary Rectal Ulcer.

INTRODUCTION

Solitary Rectal Ulcer Syndrome (SRUS) is a benign condition, characterized by a combination of symptoms, findings of colonoscopy and histology.[1] This condition was first described by Cruveihier in 1829, when he reported few cases of rectal ulcers.[2] Lloyd-Davis coined the term "solitary ulcers of the rectum" and later in 1969 the disease became widely recognized as Madigan et al reviewed 68 cases, and afterwards, Rutter et al gave a more comprehensive understanding of pathogenesis of the disease.[3,4] SRUS is less frequent, under-recognized and often misdiagnosed disorder, with an estimated prevalence of 1 in 100 000 persons per year.[5,6]

SRUS is essentially due to prolapse and resulting trauma of the rectal mucosa.[7] Possible mechanisms may include inappropriate pubo-rectalis muscle contraction, abnormal perineal descent, and overt rectal prolapse and rectal mucosal ischemia.[7] The etiology of SRUS remains obscure. Studies done on adult patients showed that excessive straining and uncoordinated defecation, resulting from dys-synergy of pelvic floor muscles lead to development of SRUS.[8] However, in children, various factors may contribute in causing this lesion including chronic severe constipation, improper toilet training.

Management of SRUS is often challenging because of lack of standardized treatment protocols. Various medical and surgical options have been tried with variable success.[9,10] Many surgeons opt for the surgical option.[11] Considering

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Article received on: 09/06/2022
Accepted for publication: 12/08/2022

the rarity of the condition in children, to the best of our knowledge, not much work has been done especially in Pakistan. So this prospective study was conducted at our center to determine the etiology of SRUS in children and study the impact of adding hydrocortisone enema to the conventional treatment strategy and its impact on the outcome of SRUS in children.

MATERIAL & METHODS

This single center Randomized controlled trial was conducted at The department of Pediatric Gastroenterology and Hepatology, Children's hospital and Institute of child health Multan from January 1, 2021 till December 31, 2021 after approval from hospital ethical review committee. Children of both genders aged up to 15 years, presenting with per-rectal bleeding or dyschezia and diagnosed having SRU were included. Children having any bleeding disorder/coagulopathy or those in whom gross colonoscopy findings or histopathology of colonic mucosal biopsy specimen report was suggestive of inflammatory bowel disease or malignancy were excluded. Confirmation of SRU was made on the basis of histopathological examination of biopsy specimen revealed crypt distortion, hypertrophied muscularis mucosa and fibro muscular obliteration of lamina propria. Demographic factors like age and gender were documented. Possible etiologic factors were also noted.

After detailed clinical history and physical examination, as per institutional protocol, basic blood tests were done. After taking informed written consent from parents/ care givers and preparation as per protocol, patients underwent colonoscopy under general anesthesia. Biopsy specimens were taken from the ulcer and surrounding mucosa and samples were sent for histopathology. After retrieving the histopathology report and confirmation of SRUS, patients were randomly divided into two groups. Group-A received conventional therapy only including behavioral modification including bio feedback training, bulk laxative (psyllium husk) and Group-B received hydrocortisone enema (hydrocortisone acetate, 10% w/w; 125 mg/ application) twice daily for 1 month and then once daily for 1 more month)[12] in addition to conventional therapy. Children were advised to stay in left lateral position for 30 minutes after taking steroid enema. Patients were educated about normal bowel habits and toilet training (avoid excessive straining, rectal digitation and prolonged sitting in toilet). All children were sent home and called on follow ups after every 2 weeks in outdoor for a total duration of 6 months. Upon follow-up compliance to therapy was checked along with complete resolution of symptoms. Figure-1 is showing methodology flow chart

SPSS version 24.0 was used for statistical analysis. Frequencies and percentages were calculated for qualitative variables, while Mean and standard deviations were noted for quantitative variables. Chi-square test was used to compare the outcome of the two treatment protocols. P value of < 0.05 was considered statistically significant.

RESULTS

In a total of 52 children, 28 (53.8%) were male while 24 (46.1%) were females. Overall, mean age was 9.1 ± 4.1 years while 7 (13.4%) children were aged <5 years, 19 (36.5%) between 6 to 10 years while remaining 26 (50%) were aged between 11 to 15 years. Mean duration of symptoms was 5.2 ± 1.4 months. Rectal bleeding, mucus in stool, abdominal pain, diarrhea, small infrequent stools and incomplete evacuation were noted in 48 (92.3%), 27 (51.9%), 12 (23%), 2 (3.8%), 2 (3.8%) and 1 (1.9%) children. Regarding the possible etiology, 49 (94.2%) patients had constipation, 46 (88.4%) had history of excessive straining, 39 (75%) had prolonged sitting while defecation, 8 (15.3%) patients had history of rectal digitation. Thirty (57.6%) children were well thriving, while 22 (42.3%) were under-weight having weight Z score $<$ -2.0 of median weight for age. Anemia (hemoglobin $<$ 10 G/dl) was found in 47 (90.3%) children. On colonoscopy, 41 (78.8%) children had single rectal ulcer while 11 (21.1%) had more than 1 ulcers. Table-I is showing comparison of various demographic and clinical parameters among children of both study groups.

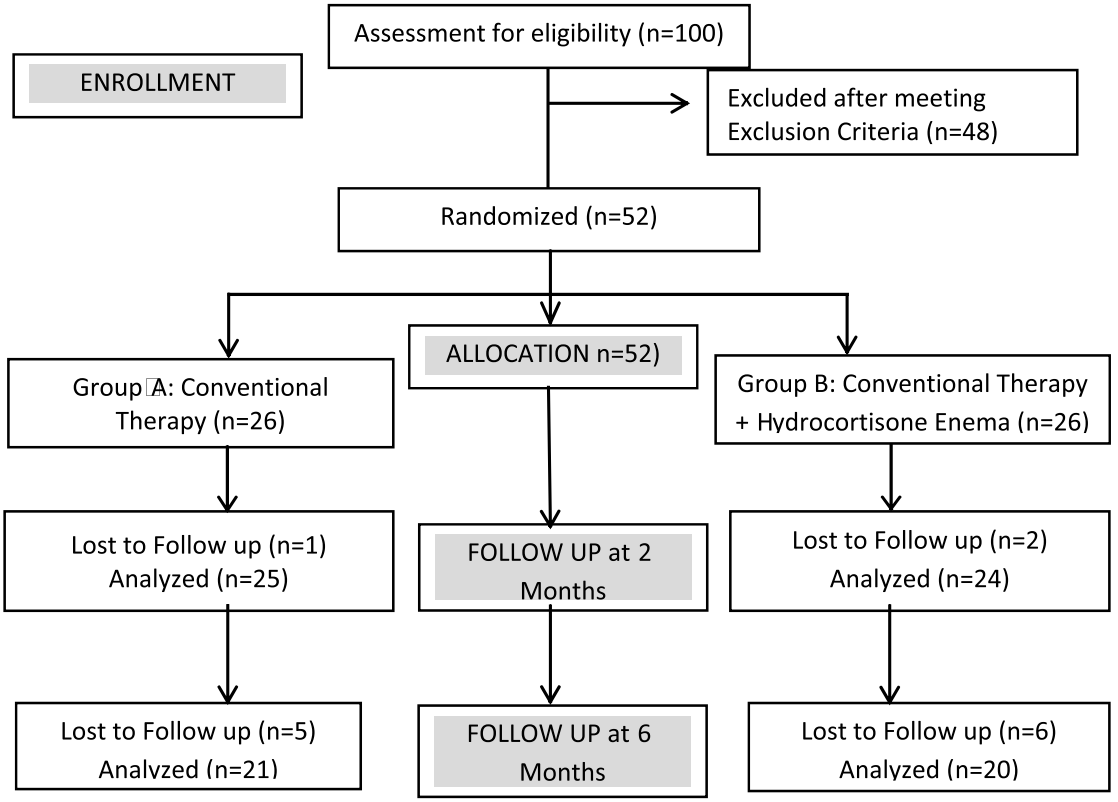

Figure-1. Methodology flow chart

Demographic and Clinical Parameters		Group-A (n=26)	Group-B (n=26)	P-Value
Gender	Male	15 (57.7%)	13 (50.0%)	0.5780
	Female	11 (42.3%)	13 (50.0%)	
Age (years)	<5	3 (11.5%)	4 (15.4%)	0.6803
	6-10	11 (42.3%)	8 (30.8%)	
	11-15	12 (46.2%)	14 (53.8%)	
Duration of Symptoms (Mean±SD)		5.0±1.1	5.5±1.7	0.2138
Anemia		22 (84.6%)	25 (96.1%)	0.1582
Underweight		10 (38.5%)	12 (46.2%)	0.5745
Frequency of Presenting Symptoms	Rectal Bleeding	22 (84.6%)	26 (100%)	0.4363
	Mucus in Stool	12 (46.2%)	15 (57.7%)	
	Abdominal Pain	8 (30.8%)	4 (15.4%)	
	Diarrhea	1 (3.8%)	1 (3.8%)	
	Small Infrequent Stools	-	2 (7.6%)	
	Incomplete Evacuation	1 (3.8%)	-	
Frequency of Possible Etiology of SRU	Constipation	23 (88.5%)	26 (100%)	0.6636
	History of Excessive Staining	24 (92.3%)	22 (84.6%)	
	Prolonged Sitting While Defecation	23 (88.5%)	16 (61.5%)	
	History of Rectal Digitation	5 (19.2%)	3 (11.5%)	
Number of Rectal Ulcers*	Single Rectal Ulcer	23 (88.5%)	18 (69.2%)	0.0895
	>1 Rectal Ulcer	3 (11.5%)	8 (30.8%)	

Table-I. Demographic and clinical parameters of children in both study groups (n=52)
Group-A: Conventional therapy; Group-B: Conventional therapy and hydrocortisone enema; *As per colonoscopic findings

Complete Resolution of Symptoms after 2-months of Treatment	**Group-A (n=25)**	**Group-B (n=24)**	**P-Value**
	14 (56.0%)	20 (83.3%)	0.0380
Complete Resolution of Symptoms after 6-months of Treatment	**Group-A (n=21)**	**Group-B (n=20)**	**P-Value**
	10 (47.6%)	18 (90.0%)	0.0036

Table-II. Comparison of treatment outcomes in both study groups
Group-A: Conventional therapy; Group-B: Conventional therapy and hydrocortisone enema

After 2 months of treatment, 49 patients returned for follow up and analyzed for treatment outcomes. It was found that 14/25 (56.0%) children in Group-A reported complete resolution of symptoms in comparison to 20/24 (83.3%) in Group-B (p=0.0380) at 2-months post treatment follow up. Forty one children completed the 6-months follow up period and included in the final analysis. It was found that 10/21 (47.6%) children reported completed resolution of symptoms while Group-B, 18/20 (90.0%) children had complete resolution of symptoms (p=0.0036) at 6-months follow up. None of the patients reported non-adherence to the treatment protocols in both study groups. Table-II is showing comparison of post-treatment outcomes at 2-months and 6-months follow up.

DISCUSSION

The exact pathophysiology behind SRUS is not fully understood but hypersensitivity linked to rectum may be contributing to consistent need to defecate as is also thought that partial evacuation could also have an important role to play in SRUS cases.

In this study, 53.8% children were male. Suresh N et al reported male to female ratio of 1.4:1 among children with SRUS which is somewhat similar to what we observed.[13] Some researchers analyzing adult patients with SRUS have reported female predominance but among children with SRUS, majority of the cases tend to be male.[14,15] We found that bleeding per rectum (92.3%) was the commonest clinical finding among children with SRUS. The literature reports bleeding per rectum to be the most frequent clinical finding which correlates well with our observations.[16] Mucus in stool (51.9%) and abdominal pain (23.0%) were other most frequent clinical findings in this research. Local data from Lahore analyzing children with per-rectal bleeding and SRUS reported 66.7% children to have mucus in stool while tenesmus and perineal discomfort were recorded among 47.6% and 42.9% children.[17]

In the present work, anemia was found in 90.3% children with SRUS. Local data has found 85.7% children with SRUS to accompany anemia which correlates well with our findings and shows that anemia is a common entity among children with SRUS.[17] Per-rectum bleeding in regular intervals among children with SRUS could be the most obvious reason behind high prevalence of anemia in these children.

Laxatives, avoidance of constipation and proper use of commode are some of the main compartments of SRUS treatment while hydrocortisone enema as adopted in the present study has been used at our center for some time.[18,19] Researchers have found behavioral therapy was found useful.[20] Another study found pharmacological therapy including corticosteroid, salicylate and sucralfate enema more beneficial.[21] Most of the centers follow more conservative/conventional approach including patient/parental reassurance and behavioral therapy.[22] We found addition of hydrocortisone enema along conventional treatment of SRUS to be significantly more successful in resolution of pretreatment symptoms in comparison to conventional treatment along. Regarding the best treatment option, the opinion differs from center to center. Experts advise steroid enemas to be useful as was found in this study along with conventional treatment options for SRUS.[23-25]

The present study is the 1st local research analyzing effectiveness of hydrocortisone enemas along with conventional therapy versus conventional therapy alone for the treatment of SRUS which makes the findings of the present study very interesting.

Our study had some limitations. As this was a single center study, our findings cannot be generalized. We were unable to compare frequency of bleeding per rectum in the post-treatment phase due to missing data by the parents which would have given us further insight into usefulness of study drugs.

CONCLUSION

In children with SRUS, hydrocortisone enemas along with conventional treatment were found to be significantly more useful in resolution of pretreatment symptoms in comparison to conventional treatment alone. Further studies involving large sets of population are needed to verify the findings of present study.

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AUTHORSHIP AND CONTRIBUTION DECLARATION

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1	Ali Hammad	Study design, Data collection & interpretation.	
2	Muhammad Tariq Aziz	Statistical analysis.	
3	Ghazi Khan Khosa	Literature search.	
4	Muhammad Aslam Sheikh	Manuscript preparation.	