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Effectiveness of Serial Home Rectal Irrigation among Neonates and Early Infants Suffering from Hirschsprung's Disease

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ABSTRACT

Background: Hirschsprung's disease (HSCR) is a congenital disorder characterized by absence of ganglion cells in the distal intestine, leading to functional obstruction. Although rectal irrigation is a standard preoperative measure, the feasibility and effectiveness of home-based serial rectal irrigation in neonates and early infants remains underexplored. Objective: This study aimed to evaluate the effectiveness of serial home rectal irrigation in neonates and early infants with HSCR prior to definitive surgery Bangladesh. Methods: This comparative longitudinal study was conducted at the Department of Pediatric Surgery, Mymensingh Medical College Hospital, Bangladesh, including 30 neonates and early infants with short-segment HSCR (Group A) and 30 age- and sex-matched controls (Group B). Parents of Group A patients were trained to perform home rectal irrigation for 2–3 months. Clinical outcomes were evaluated through serial body weight, serum albumin, and total protein measurements. All HSCR patients subsequently underwent single-stage pull-through (SSPT) surgery, and immediate and delayed postoperative complications were recorded. Results: Parents successfully performed home rectal irrigation in 90–100% of cases, with no reported complications. Group A patients showed significant weight gain across follow-up visits (p=0.001), with serum albumin and total protein levels comparable to controls. Postoperative complications were minimal, including perianal excoriation (16.66%), intestinal obstruction (10%), persistent constipation (3.33%), and anastomotic stricture (3.33%), all managed conservatively. Conclusion: MSK Home rectal irrigation is a safe and effective preoperative management strategy for neonates and early infants with HSCR. Combined with SSPT, it ensures satisfactory outcomes with minimal morbidity, offering a viable alternative to colostomy in early life.

1. INTRODUCTION:

Hirschsprung's disease (HSCR) is a multigenetic disease which can be characterized by the absence of intrinsic ganglionic cells in the submucosal and myenteric plexus of the intestinal tract. The aganglionic segment starts distally at the internal anal sphincter and extends proximally to variable lengths of the intestine which results in aperistalsis in affected bowel length and causes functional intestinal obstruction[1].

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Incidence of HSCR is 1:5000 live births with the ratio of male: female is 4:1[2]. Hirschsprung's disease (HSCR) is classified by the extent of aganglionosis into rectosigmoid (short-segment), long-segment, total colonic, and very short-segment (vssHSCR) types[3]. In early infants, presentations may differ which are constipation, abdominal distension and bilious vomiting. Early infants may also present with features of complication of HSCR like enterocolitis, recurrent constipation with intervening diarrhea, failure to thrive and sometimes may even present with fever with features of bowel perforation[4].

Treatment modalities of HSCR may be single stage or multi stage depending on age of onset of the disease, severity, relevant investigations, imaging studies and tissue[5]. But to this date it has yet not been established the ideal time to perform SSPT in neonates or in early infants. In most cases, this surgery is performed in infancy or in the neonatal period as presentation in older children and adulthood is uncommon. Rectal irrigation is a conventional and effective means for pre-operative preparation which can relieve the bowel obstruction caused by an aganglionic segment in patients with HSCR[3].

The goal of serial rectal irrigation (2-3 months) is to prevent enterocolitis and to reduce colonic distension. Neonates and early infants who do not have colostomy may develop functional intestinal obstruction and ultimately enterocolitis, therefore, in order to prevent such complications, rectal irrigation (10 – 20 ml per kg body weight) with 0.9 % NaCl solution should be perform at hospital and at home to facilitate easy passage of stool and to keep the rectum decompressed[6]. Home rectal irrigation during neonatal and early infancy followed by delayed and planned SSPT can be an alternative to colostomy in the neonatal period and early infancy to treat HSCR[3]. The present study is primarily designed to evaluate the effectiveness of home rectal irrigation in neonates and early infants prior to definitive treatment. In this study, HSCR child whose age and sex were matched with non-HSCR child, then at the end of 2- 3 months of irrigation schedule their mean weight, mean serum total protein and mean serum albumin have been compared to find out regarding the success of home rectal irrigation.

MATERIALS AND METHODS:

This comparative longitudinal study was conducted in the Department of Pediatric Surgery, Mymensingh Medical College Hospital, Bangladesh, following approval by the Institutional Review Board (Memo No. MMC/IRB/2020/336) on 30 November 2020. The study population included neonates and early infants with short-segment Hirschsprung's disease (HSCR) admitted or attending the outpatient department. Age- and sex-matched non-HSCR infants served as controls.

The calculated sample size was 84 per group based on standard formulae; however, due to COVID-19 restrictions and limited patient availability, 30 patients were recruited per group using purposive sampling. Inclusion criteria were neonates and early infants with short-segment HSCR after successful rectal irrigation, while preterm, low-birth-weight infants, long-segment HSCR, neonates with other intestinal obstructions, or life-threatening congenital anomalies were excluded.

Group A patients underwent clinical evaluation, contrast enema after one month of age, and punch biopsy when indicated. Home serial rectal irrigation was performed for 2–3 months, and monthly follow-ups assessed clinical outcomes, including body weight, serum albumin, and total protein. Group B (controls) were assessed for the same parameters. Definitive surgical treatment was performed using the modified Soave pull-through procedure (SSPT), with standard pre- and postoperative care.

Participants Home rectal irrigation involved inserting a lubricated rubber catheter 10–15 cm into the rectum, allowing saline to flow by gravity, and gently moving the catheter to and fro for 2–3 minutes, repeated until effluent was clear. Parental performance and adherence were assessed using a structured questionnaire.
Data were collected via structured questionnaires and analyzed using SPSS v20 and Microsoft Excel 2019. Continuous variables were expressed as mean ± SD, categorical variables as percentages, and $p < 0.05$ was considered statistically significant. Written informed consent was obtained from all parents/guardians.

RESULTS:

Table 1 demonstrates that the majority of the study population were neonates (3–28 days), comprising 63.33% (19/30) of the patients, while early infants (30–42 days) accounted for 36.67% (11/30). The male predominance is notable, with 23 males (76.67%) and 7 females (23.33%), yielding a male-to-female ratio of 3.29:1. This indicates that Hirschsprung's disease in this cohort was more common in male neonates compared to females and slightly less frequent in early infants.

Table-1. Age and Sex Distribution of the Study Population (n=30)

Age Group (days)	Male	Female	Total	Percent (%)

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3–28 (Neonates)	15	4	19	63.33
30–42 (Early infants)	8	3	11	36.67
Total	23	7	30	100.0

Figurer I shows the number and percentage of patients came to the hospital with different clinical presentations. Here, we can see all the 30 patients that is 100% patients presented with 2 problems that are history of delayed passage of meconium and constipation with assisted defecation. So, these 2 were the most common problem among the patients. Besides these, patients also presented with abdominal distention, vomiting and dehydration. Statistically 66.66% patients presented with these three complains. Fever was exhibited by 33.33% patients and only 16.33% patients were presented with history of enterocolitis.

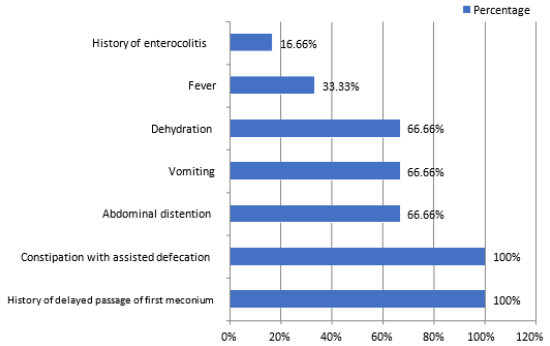

Figure 1. Clinical presentation of the patients

Table 2 shows the difference between mean body weights of the patients during different visits. We can see from the table that, here the p value for single factor Anova test is 0.001 which is less than 0.05. There is significant differences among the mean body weight of patients with Hirschsprung disease at the p<.05 level on different visits. (p=0.001).

Table 2. Mean body weight of the study population with Hirschsprung's disease on different visits during successful home rectal irrigation (n=30)

Visit	Weight (Kg) Mean±SD	p value (ANOVA)
1st	3.06±0.17	0.001
2nd	3.23±0.18	
3rd	3.39±0.21	

Table 3 shows that Group B (control) had slightly higher mean values for body weight (3.52 ± 0.22 kg), serum albumin (4.20 ± 0.35 g/dL), and serum total protein (6.95 ± 0.26 g/dL) compared to Group A (HSCR patients), whose corresponding values were 3.39 ± 0.21 kg, 4.01 ± 0.33 g/dL, and 6.81 ± 0.23 g/dL. The calculated t-values for all variables were less than the t-table value (±2.001), and the p-values ranged from 0.024 to 0.033, indicating statistically significant differences between the groups. These results suggest that although differences exist, HSCR patients maintained comparable nutritional and growth parameters to age- and sex-matched controls following home rectal irrigation.

Table 4 shows the percentage of complications that were faced by the patients in the immediate post-operative period. 16.66% presented with perianal excoriation. Intestinal obstruction presented by 10 % of the patients post operatively. None of the patients presented with the complication of post-operative enterocolitis, intestinal perforation, anastomotic leakage.

1 case of intestinal obstruction developed on 3rd post-operative day and 2 cases of intestinal obstruction developed on 4th post-operative day which were treated by N.P.O, N.G suction, I.V fluid, I.V antibiotic, correction of electrolytes and blood transfusion. All the cases improved after 2 days of conservative treatment. Perineal excoriation was managed by perineal exercise and rectal exercise by gentle introduction of lubricated finger, twice daily. All cases improved 4 weeks after discharge.

Table 5 shows the delayed complications that were faced by the patients after operation. It describes persistent constipation occurred among 10% of the patients in the 4th week after operation, at the same time anastomotic stricture occurred among 3.33%. In 6th week persistent constipation occurred among 6.66% of the patients and anastomotic stricture occurred among 3.33% of the patients. In 8th week persistent constipation occurred among 3.33 % and anastomotic stricture among 3.33% of the patients.

Table- 3. Comparison of Body Weight, Serum Albumin, and Serum Total Protein Between Group A and Group B (n=30 each).

Variable	Group A Mean ± SD	Group B Mean ± SD	t (calculated)	t (table)	p value
Body Weight (Kg)	3.39 ± 0.21	3.52 ± 0.22	-2.32	±2.001	0.024
Serum Albumin (g/dL)	4.01 ± 0.33	4.20 ± 0.35	-2.18	±2.001	0.033
Serum Total Protein (g/dL)	6.81 ± 0.23	6.95 ± 0.26	-2.28	±2.001	0.026

Table 4. Immediate post-operative complications of the study population (n= 30).

Complications	Number of patients	Percentage (%)
Intestinal obstruction	03	10

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Post operative Enterocolitis (Present/ Absent)	00	00.00
Intestinal perforation	00	00.00
Anastomotic leakage	00	00.00
Perianal excoriation	05	16.66

Table 5. Delayed post-operative complications of the study population (n= 30).

Complications	4th week		6th week		8th week	
	Number of patients	Percentage (%)	Number of patients	Percentage (%)	Number of patients	Percentage (%)
Persistent constipation	03	10	02	6.66	01	3.33
Anastomotic stricture	01	3.33	01	3.33	01	3.33

DISCUSSION:

A total 30 patients consisting of neonates and early infancy were included for the final stage of the study. All of these 30 cases were diagnosed and confirmed with HSCR by radiological contrast study and punch biopsy. At the same time these selected candidates underwent serial rectal irrigation via lubricated plain rubber catheter and normal saline solution both at hospital and at home for a period of 2 to 3 months. Home serial rectal irrigations 2 times daily were conducted via parents who were trained by experienced investigators before the patient was discharged from hospital. At the end of serial rectal irrigation schedule a delayed but planned single stage pull through operation (SSPT) was performed for each of the patients. At the same time a healthy non HSCR group whose age and sex matched as HSCR group was selected for the comparison of growth and development before the definitive surgery.

In this study, the mean age at diagnosis was 20.77 ± 13.15 days, which is consistent with Sauer [7] (16.5 ± 20.9 days) and Ongeti et al. [8] (<30 days). The male-to-female ratio in this study was 3.29:1, indicating male predominance. Similar findings were reported by Green et al. [6] and Ambartsumyan et al. [2] with a ratio of 4:1, and by Shahjahan et al. [9]. However, Sakurai et al. [10] reported a female-to-male ratio of 2.4:1. Overall, Hirschsprung's disease shows a male predominance, which is consistent with the findings of this study.

Also, in this study, the mean body weight of Group A during the last visit (3rd visit) at the end of serial rectal irrigation schedule was 3.39±0.21. There was no significant difference between the mean body weight of Group A 3rd visit and mean body weight of Group B. In Vũ, P.A study, mean body weight at the end of rectal irrigation schedule were 3.05±0.49KG, which was consistent with Group A study population of this study [11]. However, in Lu C. study, mean body weight at the end of successful rectal irrigation was 6.77±0.66KG. It can be speculated that this difference is due to variation in the age group of study population and conduction of home rectal irrigation for a longer period of time which lasted for up to 4 months in Changgui Lu study[3].

This study states, serum total protein and serum albumin during last visit after successful rectal irrigation of patients with HSCR (group A) were found to be 6.81±0.23 gm/dl and 4.01±0.33 gm/dl respectively. On the other hand, serum total protein and serum albumin of same age group of non HSCR (group B) were 6.95±0.26 gm/dl and 4.20±0.35gm/dl respectively. There was no significant difference of serum total protein and serum albumin between Group A and Group B in this study. In Lu C. study, estimation of serum albumin was 4.18 ± 0.24 gm/dl and they didn't included estimation of serum total protein[3].

In this study, clinical presentations were history of delayed passage of meconium, constipation, assisted defecation with intermittent intestinal obstruction and enterocolitis. These clinical features were consistent with Kessmann study[12].

Post operative complications both immediate and delayed of the study population include intestinal obstruction, perianal excoriation, persistent constipation and anastomotic stricture. These were treated by conservative measures and improved. In Pham Anh study, post operative complication were enterocolitis, persistent constipation and anastomotic stricture, which were treated conservatively and improved. One of them required a redo pull through for persistent constipation having residual aganglionosis proven by frozen biopsy [11].

In current study, immediate post operative complications were intestinal obstruction (10%), perianal excoriation (16.66 %). On the other hand, delayed post operative complications were persistent constipation (3.33%) & anastomotic stricture (3.33%). None of the patients presented with the complication of post operative enterocolitis, intestinal perforation, anastomotic leakage which indicates minimal number of immediate and delayed post- operative complications. In Shahjahan M et al. study, post operative complications were perianal

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excoriation and post operative anastomotic stenosis[9]. Therefore, it can be said that post operative complications of this study were within a satisfactory level when compared with other studies [9,10]

Present study was conducted with small sample size in a teaching hospital which showed satisfactory outcome of home rectal irrigation in neonates and early infants with HSCR prior to definitive surgery with minimal complications. Persistent constipation occurred 4 weeks after discharge which were treated by gentle saline irrigation daily for the next 8 weeks and improved. Anastomotic stricture also occurred 4 weeks after discharge and treated by gentle anal dilatation by lubricated anal dilator and improved.

CONCLUSION:

This study demonstrates that home rectal irrigation is a safe, and effective preoperative management for neonates and early infants with Hirschsprung's disease, with high parental compliance (≥90%) and no reported complications. It was associated with significant improvement in body weight (p=0.001) and maintenance of nutritional status comparable to controls. Postoperatively, single-stage pull-through surgery yielded favorable outcomes with minimal morbidity, where immediate complications were limited to perianal excoriation (16.66%) and intestinal obstruction (10%), and delayed complications included mild constipation and anastomotic stricture in a small proportion of patients. Overall, both home rectal irrigation and SSPT proved effective, safe, and practical in the management of Hirschsprung's disease.

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