

Bowel management programme in patients with anorectal malformations

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Abstract

Objective: To establish an effective and practical bowel management protocol to achieve social continence in children operated for anorectal malformations.

Method: The quasi-experimental study was conducted from January to December 2021 at the Colorectal Clinic of the National Institute of Child Health, Karachi, and comprised patients aged >3 years who had completed surgical management for anorectal malformations. Tailored modifications were made to a one-week bowel management boot camp to adapt to institutional constraints and the practical needs of the patient population. History, examination, abdominal X-ray and incontinence score were recorded before and after the commencement of high-volume saline enemas. Data was analysed using SPSS 22.

Results: Of the 80 patients, 46(57.5%) were boys with mean age 6.74 ± 3.24 years and 34(42.5%) were girls with mean age 5.41 ± 1.84 years. Among the boys, rectobulbar fistula was the most common anomaly 26(56.5%), whereas rectovestibular fistula was the most frequent anomaly in girls 20(58.8%). The mean incontinence score at presentation was 5.375 ± 1.72 , which improved significantly to 3.637 ± 1.74 following bowel management ($p < 0.001$). For the soiling component, the mean score difference was 1.275 ± 0.45 ($p < 0.001$).

Conclusion: Modified bowel management programme was effective in keeping the incontinent and constipated patients after anorectal malformation surgical treatment.

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Introduction

Anorectal malformations (ARMs) are common congenital anomalies managed in paediatric surgery departments. Many patients continue to have faecal incontinence and/or constipation after corrective surgical procedures.^{1,2} Bowel management programmes (BMPs) are designed to keep these patients socially clean that may affect functional and psychosocial wellbeing.^{3,4} Several management protocols and methods have been devised for these patients, including diet, laxatives, enemas, rectal washouts, biofeedback and surgical procedures, like Malone's antegrade appendicostomy, with variable results.⁵⁻⁷ The BMP devised by Pena et.al. is reported to achieve socially acceptable continence and cleanliness, and is in practice at paediatric colorectal centres all over the world.⁸⁻¹² The studies on BMPs include diseases, like functional constipation, ARMs, Hirschsprung, spina bifida etc.

ARMs patients behave differently,^{13,14} and there is a need to have a modified BMP to suit the local context in terms of resource constraints. The current study was planned to establish an effective and practical BMP with the aim of

achieving social continence in children operated for ARMs.

Patients and Methods

The quasi-experimental study was conducted from January to December 2021 at the Colorectal Clinic of the National Institute of Child Health (NICH), Karachi. After approval from institutional ethics review board, the sample size was calculated with 95% confidence interval (CI) and 80% power, taking baseline and post-intervention faecal incontinence scores from a study by Lombardi et al.¹² The sample was raised using non-probability consecutive sampling technique.

All patients with incontinence on follow-up at the clinic, primarily operated at least 6 months earlier for imperforate anus, were approached, including both staged and single-surgery procedures. Patients of both genders aged at least 3 years were selected to ensure that every child was beyond potty training age. Patients with any postoperative complication, such as incomplete dilatation protocol, stricture/stenosis or mucosal prolapse, were excluded. The initial screening comprised completed dilatation protocol, performing Henicke Mikulick's anoplasty for anal stricture, and mucosectomy for mucosal prolapse.¹⁵

Written informed consent was taken from the parents in their native language. On the first visit, complete history, including demographics and previous surgical record, was obtained. This was followed by a digital rectal examination

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Table-1: Krickenbeck's incontinence score.

Subdomain	Grade	Score
Voluntary bowel movement	Hold the bowel movement	1
	Capacity to verbalise	2
	Feeling of Urge	3
Soiling	No	0
	Occasionally (once or twice per week)	1
	Everyday no social problem	2
Constipation	Constant social problem	3
	No	0
	Manageable by changes in diet	1
	Requires laxatives	2
	Requires enema	3

(DRE) and a general physical exam. In the DRE, the tone of the anal canal and the ability to squeeze the sphincter were assessed. A baseline X-ray was done to see faecal loading. If there were any palpable faecalomas or stool impaction, they were first evacuated with the help of phosphate enemas and saline washouts. Parents were advised to adhere to a diet plan at fixed timings that were designed according to the child's tendency towards constipation or diarrhoea.

Before starting BMP, the Krickenbeck's continence score, as modified by Pena et al., was noted (Table 1). The urinary component was skipped because of non-relevance to the subject of study. Voluntary bowel movement, constipation and soiling components were separately and cumulatively scored. The score ranged 1-9, with lower score indicating improved condition and higher score indicating miserable incontinence.

The original BMP⁸⁻¹² requires patients to be admitted for a week-long hospital stay involving history-taking, examination under anaesthesia (EUA), contrast enema studies, and daily abdominal radiographs to titrate high-volume saline enemas for effective colonic evacuation. However, due to resource limitations, provisions of inpatient accommodation and daily radiological assessments were not feasible. Therefore, a modified protocol was implemented. DRE was performed instead of EUA, and a baseline abdominal and pelvic X-ray was used in place of a contrast enema. Patients were managed through fortnightly outpatient clinic visits, with abdominal radiographs taken at each visit to guide enema adjustments. Additionally, Nelaton catheters were used instead of Foley catheters, as they are simpler to use, more affordable and better suited for a predominantly low-literacy patient population. The bowel management protocol consisted of large-volume saline enemas, initiated at 20ml/kg and adjusted individually based on patient response using a hit-and-trial approach. Enemas were administered via a Nelaton catheter. The procedure was

first demonstrated to the parents by doctors in the minor surgical procedure room. The patients were placed in the prone jack-knife position, and the lubricated Nelaton catheter was gently inserted. The patients were maintained in this position for five minutes before being allowed to evacuate. Glycerin, up to 30ml per wash, was added once daily if the regulation of bowel movements was difficult. Normal saline was prepared at home by mixing one teaspoon of table salt in 500ml of tap water. The patients were scheduled for fortnightly outpatient visits to reassess and adjust the irrigation volume based on the degree of faecal evacuation and the number of accidents. Abdominal radiographs were obtained during visits to evaluate residual faecal loading in the colon. Communication regarding any issues was maintained through a dedicated WhatsApp group, or, where internet access was limited, via phone calls. After three fortnightly visits, a fourth follow-up was scheduled three months later to reassess continence scores and perform a repeat abdominal radiograph to evaluate colonic clearance.

Data was analysed using SPSS 22.0. Paired sample t-test or Wilcoxon signed-rank test, as appropriate, was applied to compare the baseline and post-intervention scores. Stratification was done on the basis of age, diagnosis and the definitive surgery. Two-sided $p \leq 0.05$ was considered statistically significant.

Results

Of the 80 patients, 46(57.5%) were boys with mean age 6.74 ± 3.24 years and 34(42.5%) were girls with mean age 5.41 ± 1.84 years. The overall age range was 3-16 years.

Among the boys, rectobulbar fistula was the most common anomaly 26(56.5%), whereas rectovestibular fistula was the most frequent anomaly in girls 20(58.8%). Others included perineal fistula 10(12.5%), rectoprostatic fistula .8(10%), cloaca 6(7.5%), rectovesical 4(5%), rectovaginal 2(2.5%), pouch colon 2(2.5%) and there were 2(2.5%) cases without any fistula. The choice of procedure was posterior sagittal ano-rectoplasty (PSARP) in 40(50%) cases, followed by anterior sagittal ano-rectoplasty (ASARP) 18(22.5%) in females with grossly visible vestibular and perineal fistulas, abdominoperineal pull through 16(20%) and anoplasty 6(7.5%).

The mean incontinence score at presentation was 5.375 ± 1.72 , which improved significantly to 3.637 ± 1.74 post-intervention, with the mean difference being 1.74 ± 0.84 ($p < 0.001$). Individual patient scores were noted separately (Figure). For the soiling component, the mean baseline score was 2.75 ± 0.54 which improved to 1.47 ± 0.59 post-intervention. The mean difference was 1.275 ± 0.45 ($p < 0.001$). For the constipation component, the mean

Table-2: Baseline and post-intervention mean Krickenbeck and soiling scores in various types of anorectal malformations (ARMs).

Type of Anorectal Malformation	Pre-BMP Krickenbeck score	Post-BMP Krickenbeck score	p-value_ mean difference Krickenbeck score	Pre-BMP Soiling score	Post-BMP Soiling score	p-value mean difference Soiling score
Perineal fistula	3.4	2	<0.005	2.4	1	<0.005
Rectovaginal fistula	5	4	<0.005	3	2	<0.005
Rectobulbar fistula	5.85	3.92	<0.005	2.85	1.54	<0.005
Rectoprostatic fistula	5	4	<0.005	2.75	1.75	<0.005
Rectovesical fistula	5	3.5	0.014	3	1.5	0.014
Rectovestibular fistula	5.6	3.8	<0.005	2.8	1.5	<0.005
Cloaca	4.67	3.67	<0.005	2.33	1.33	<0.005
No fistula	6	5	<0.005	3	2	<0.005
Perineal fistula	5	3	<0.005	3	1	<0.005

BMP: Bowel management programme.

baseline score was 5.22 ± 1.93 which improved to 3.62 ± 1.75 post-intervention. The mean difference was 1.6 ± 0.84 ($p < 0.001$).

The baseline and post-intervention soiling component scores were stratified on the basis of the type of procedure and the primary diagnosis. In both cases the difference was significant (Table 2).

In terms of baseline scores, despite having incontinence, some degree of ability to hold a voluntary bowel movement was seen in 28(50%) patients with rectoperineal, rectobulbar and rectovestibular fistula. These types of fistulas had a greater tendency towards constipation as seen in 10(40%) patients with rectobulbar fistula and 10(50%) with rectovestibular fistula. Overall, 22(27.5%) patients had constipation with pseudo incontinence. Among them, 2(9%) were manageable with diet, 8(36.4%) needed laxatives and 12(54.6%) were resistant to both laxatives and diet. After three month of bowel management, 18(82%) of these patients still had constipation. Among them, 2(11%) were manageable with diet, and 16(89%) responded to laxatives.

Soiling was a constant social problem in 64(80%) cases, daily soiling episode was noted in 12(15%) and occasional soiling problem was found in 4(5%). Post-intervention, none of the participants had constant social soiling. Median wash quantity was 300ml (IQR:150) normal saline, and glycerine was added in 35% patients to regularise bowel habits.

Abdominal pain was the most common complaint, initially experienced by 10(12.5%) patients. It was resolved by making the saline slightly warmer than room temperature and adding antispasmodics in 4(5%) patients. Further, 4(5%) patients experienced loose stools after washes, so the quantity was decreased. Finally, 24(30%) patients reached the

desired quantity of wash on the second visit, and the rest on the third visit.

Discussion

After a successful ARMS surgery, the rate of faecal incontinence remains high. Qazi et al. reported a 12% incontinence rate in patients with ARMs.¹⁶ True continence cannot be achieved, but it can be managed to keep one socially continent. So, the true measure of success would be the quality of life that can be provided to a patient. Baxter et al. concluded that

establishing a successful programme was, indeed, a cumbersome task.¹⁷ Costigan et al. mentioned different systems involving rectal drain for bowel wash.¹⁸ Although many methods and techniques exist and are being applied to these patients for bowel control, they are physically and financially demanding. Some modifications to suit a certain population based on their socioeconomic construct will certainly help the patients to get along with their healthy peers.¹⁹ The introduction of a bootcamp programme for bowel management is a practical model adopted by many institutions worldwide. The current study modified some aspects of assessment, gadgets and follow-up frequency to suit the institutional setting, and assessed this modification quantitatively using Krickenbeck's incontinence score.

A better programme has also been proposed for managing bowel by using ultrasound to determine the quantity of saline needed to evacuate large gut.²⁰ But, then again, in a resource-limited setting like NICH, adopting this is not possible. Instead, a baseline plain abdominal X-ray was used to document faecal loading.

Multiple scoring systems are present for quantifying incontinence.^{11,21,22} The current study used the Krickenbeck

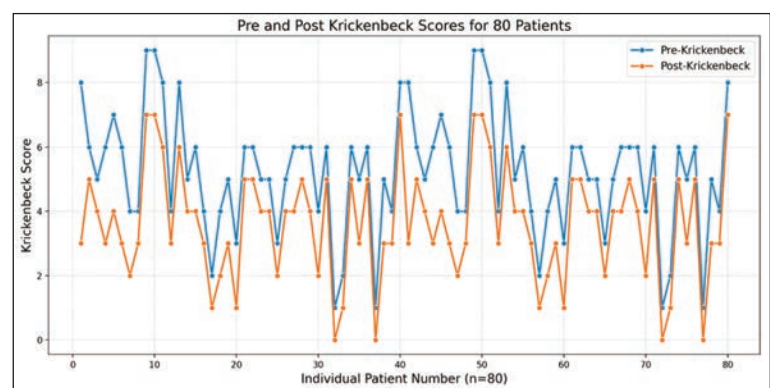


Figure: Baseline and post-intervention Krickenbeck scores.

score because it focusses on the soiling component specifically, which is the major issue in the context of the disease. It has been acknowledged by Bischoff et al., the creators of BMP, who also made slight modifications to convert that into a scoring system which can be re-evaluated from time to time.²³

Levitt et al. described that those with lower anomalies had a better control, but were more prone to constipation.²⁴ The current results were in agreement as 20 of the 22 patients with constipation had rectovestibular and rectourethral fistula. The school-going children took the enemas in the morning and had a near-normal school experience. Almost all of them added glycerine to fix their bowel movement in the morning. Those who stayed at home were given washes in the evening as per the mother's convenience. Baxter et al. conducted a multi-institutional review, and reported a similar impression that school-going children and their parents were looking for more control over soiling.¹⁶ The problem persists in adolescence, and it does not settle with age.⁴ With the newer concept of transition of care in mind, the parents start involving the patient actively in the process.²⁵ Nine of the current patients aged >10 years were actually able to manage the whole process without adult help, and it helped them manage their symptoms.

The current study has limitations as it had no control group in which the standard bowel management protocol would have been used. In the absence of the control group, it is difficult to label it equally efficient despite having statistically significant results. Further studies comparing the quality of life in these patients, using different modalities of bowel control, are recommended.

Conclusion

BMP with localised modifications was found to be efficient in improving incontinence in environments where costly investigations and protocols are not feasible due to financial constraints. The key is to understand every child's individualised needs and social setup.

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BF: Concept, data collection, interpretation, drafting, final approval and agreement to be accountable for all aspects of the work.

NZ: Concept, supervision of research, revision, final approval and agreement to be accountable for all aspects of the work.

TK & SP: Data analysis, interpretation, drafting, final approval and agreement to be accountable for all aspects of the work.

SA & AJB: Data acquisition, drafting, final approval and agreement to be accountable for all aspects of the work.