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ORIGINAL ARTICLE

Improvement in bladder function in children with functional constipation after a bowel management program

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Abstract

Purpose We sought to determine if children with functional constipation (FC) would have an improvement in bladder function with treatment of constipation with a bowel management program (BMP).

Methods A single-institution review was performed in children aged 3–18 with FC who underwent a BMP from 2014 to 2020. Clinical characteristics, bowel management details, and the Vancouver Symptom Score for Dysfunctional Elimination Syndrome (VSS), Baylor Continence Scale (BCS), and Cleveland Clinic Constipation Score (CCCS) were collected. Data were analyzed using linear mixed effect modeling with random intercept.

Results 241 patients were included with a median age of 9 years. Most were White 81 and 47% were female. Univariate tests showed improvement in VSS (− 3.6, $P < 0.0001$), BCS (− 11.96, $P < 0.0001$), and CCCS (− 1.9, $P < 0.0001$) among patients having undergone one BMP. Improvement was noted in VSS and CCCS among those with more than one BMP (VSS: − 1.66, $P = 0.023$; CCCS: − 2.69, $P < 0.0001$). Multivariate tests indicated undergoing a BMP does result in significant improvement in VSS, BCS, and CCCS ($P < 0.0001$).

Conclusions There is significant improvement in bladder function in children with FC who undergo a BMP. For patients with bowel and bladder dysfunction and FC, a BMP is a reasonable treatment strategy for lower urinary tract symptoms.

Keywords Bowel and bladder dysfunction · Dysfunctional elimination syndrome · Urinary dysfunction · Urotherapy

Abbreviations

ARM	Anorectal malformation
BBD	Bowel and bladder dysfunction
BCS	Baylor continence scale
BMP	Bowel management program
CCCS	Cleveland clinic constipation score
DES	Dysfunctional elimination syndrome
FC	Functional constipation
PROM	Patient-reported outcomes measure
VSS	Vancouver symptom score for dysfunctional elimination syndrome

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Introduction

Bowel and bladder dysfunction (BBD), previously known as dysfunctional elimination syndrome (DES), is a common clinical entity that describes concomitant lower urinary tract symptoms and issues with constipation and/or fecal incontinence [1–3]. An increased fecal load in the rectum can cause mechanical compression of the bladder and affect the shared neural pathways of the bladder, bowel, and pelvic floor, resulting in decreased bladder capacity, urge incontinence, frequency, a decreased urge to evacuate, insufficient emptying, bladder spasms, and high post-void residual volumes [4, 5]. This may also result in vesicoureteral reflux and recurrent urinary tract infections with potential subsequent renal failure [4, 6].

The Vancouver Symptom Score for Dysfunctional Elimination Syndrome (VSS) is a patient-reported outcomes

measure (PROM) that has been validated to diagnose BBD in pediatric patients [7, 8]. Urotherapy, which consists of hydration, timed voiding regimens, pelvic floor biofeedback, clean intermittent catheterization, and pharmacologic therapy, has been shown to significantly improve the VSS and quality of life in children and adolescents with BBD [9]. Several studies have also shown improvement in BBD after treatment of constipation [6, 10, 11]. A bowel management program (BMP) is a formal, tailored program that employs a variety of treatment strategies to improve severe pediatric constipation and fecal incontinence when standard medical management has failed [12–14]. Strategies employed during the bowel management week include dietary changes, oral laxatives and other medication, rectal enemas, and antegrade continence enemas. Progress is monitored throughout the BMP week using daily stooling diaries and abdominal X-rays and adjustments to each patient's regimen are made as needed.

A recent study examining continence outcomes in patients with anorectal malformation was able to show that undergoing a BMP improved urinary symptoms in these patients with underlying anatomic abnormalities of the urologic system [15]. While not specific for improvement in BBD, the Baylor Continence Scale (BCS) and Cleveland Clinic Constipation Score (CCCS) are also PROMs that are used to assess changes in bowel and bladder symptoms after intervention, which the aforementioned study examined as well [16, 17]. It has not been established whether a BMP alone improves urinary symptoms in patients with functional constipation (FC) and BBD with no underlying anatomic abnormalities. The objective of this study was to determine whether a BMP can improve urinary symptoms in patients with FC and BBD by examining changes in the VSS and other PROMs before and after the program.

Methods

Participants

After local Institutional Board Review approval (STUDY00001799), a single-institution, retrospective cohort study was performed among patients with FC, aged 3–18 years, who underwent a formal BMP at these authors' institution from April 2014 to August 2020. Study dates encompass the first year of our comprehensive multidisciplinary colorectal clinic up to present day. Functional constipation was defined using the Rome IV criteria [18]. Patients who did not have FC or did not complete a BMP at our center were excluded, as were those who met the Rome IV criteria for irritable bowel syndrome. All patients who underwent BMP underwent a prospective consent process and were contacted via email by nurse clinicians for follow-up questionnaire completion.

Study measures

Given the difficulty of objectively describing urinary symptoms such as enuresis, urgency, and frequency in this patient population, we sought to find a standardized method to evaluate urinary symptoms. The primary outcome of interest was the change in scores for the VSS, the BCS, and the CCCS before and after BMP [7, 16, 17]. The VSS is an instrument aimed at capturing DES or BBD. All 14 items use a 5-point Likert scale, with a score of greater than or equal to 11 indicating the presence of DES or BBD—improvement to a score of 10 or less is ideal. Of the VSS, only three questions pertain to bowel symptoms and the remaining questions pertain to urinary symptoms. The BCS has been validated in children with anorectal malformation and is used to assess social continence. All 23 items use a Likert scale with final scores ranging from 2 to 84; lower scores reflect better social continence. Lastly, the CCCS is often used as a measure for determining the extent and severity of constipation. Scores in this instrument range from 0 to 30, with higher scores indicating more severe constipation. These questionnaires are completed electronically via the Research Electronic Data Capture tool by parents prior to the BMP and at the completion of the BMP at 1-month and 3-month follow-up.

Bowel management program

The BMP at the authors' institution consists of a week-long program that can either be completely in-person or partially/fully remote via telemedicine. The telemedicine option was introduced after the onset of the COVID-19 pandemic. Patients receive individually tailored bowel regimens that are adjusted throughout the week based on abdominal films, symptoms, and daily stooling charts to achieve social fecal and urinary continence. Examples of different regimens include combinations of oral laxatives and fiber, rectal enemas, and antegrade continence enemas via Malone appendicostomy or cecostomy.

Cohort information

Sociodemographic information was collected on all patients. Clinical characteristics collected on patients consisted of whether the child was potty-trained or catheterized, BMP date(s), BMP regimen (oral laxatives/medication, suppository, antegrade or rectal enema, combination), and PROMs before and after undergoing the BMP. Outcome scores were included in the analysis if the scores were collected at least 9 months prior to BMP during the intake process. Follow-up scores up to 1-year post-BMP were included in

analyses. Modeling was conducted on one cohort consisting of patients who underwent only one round of BMP. The second cohort analyzed consisted of those with multiple rounds of BMP of which only the most recent BMP program was assessed.

Statistical analysis

Continuous data were reported as medians and interquartile range while categorical data were reported as frequencies and proportions. Univariate tests were carried out using paired *t* tests to detect differences in scores. To account for possible differences between patients who underwent one BMP from those that underwent multiple BMPs, univariate tests were conducted separately on patients with one BMP from those that had multiple BMPs. Linear mixed effect regression modeling with a random intercept was conducted after checking that all assumptions were satisfied and correlation among variables explored. Model building was conducted via the backward selection process with variables removed if partial F *P* values were greater than or equal to 0.10. Individual groups with the highest count were selected as the reference group in most models. Final model selection was conducted after lowest Akaike information criterion and Bayesian information criterion were determined. Adjustments for multiple comparisons were done using Tukey's multiple comparison test. Findings were determined to be significant at $P < 0.05$. All statistical tests were conducted on SAS Enterprise version 8.1.

Results

A total of 241 patients met inclusion criteria (Table 1). Most patients were White (81%) and potty-trained (91%). Females comprised 47% of the cohort. The majority of patients did not have a behavioral disorder (66%) and over half had no spinal abnormalities (56%). The median age of patients at the time of the most recent BMP was 9 years (IQR: 7, 13). 72 percent of patients required only one BMP. The most common BMP regimen was oral laxatives/medication (61%) followed by enemas (30%).

Median time at which the follow-up VSS was recorded after the BMP was 92 days (IQR: 31, 153). Univariate tests indicate significant improvement in VSS (mean drop in score of 3.6, (95% CI 2.72, 4.48), $P < 0.0001$), BCS (mean drop in score of 11.96, (95% CI 9.41, 14.5), $P < 0.0001$), and CCCS (mean drop in score 1.9, (95% CI 1.06, 2.73), $P < 0.0001$) among patients having undergone one BMP (Table 2). Significant improvement in scores was also noted in the VSS and CCCS among those who underwent more than one BMP (mean drop in score 1.66, (95% CI: 0.23, 3.09), $P = 0.023$ for VSS; mean drop in score 2.69, (95% CI: − 0.91, 6.28), $P < 0.0001$ for CCCS). (Table 3). However, improvement in the BCS was observed but not found to be significant (mean drop in score 2.69, (95% CI − 0,91, 6.28), $P = 0.14$). Multivariate tests indicate that even after adjusting for all covariates, undergoing a BMP does result in significant

Table 1 General cohort characteristics

	Total ($n = 241$)	
	n	%
Age at BMP, years (median, IQR)		9 (7. 13)
Gender ($n = 232$)		
Male	119	49.38
Female	113	46.89
Race ($n = 214$)		
White	196	81.33
Bi-racial or Multi-racial	7	2.9
Other	6	2.49
Black	5	2.07
Ethnicity ($n = 67$)		
Hispanic	4	1.84
Catheterized ($n = 237$)	11	4.56
Behavioral disorder ($n = 240$)		
None	158	65.65
More than one	36	14.94
ADHD	27	11.2
Autism	9	3.73
Other	8	3.32
OCD	1	0.41
Developmental delay ($n = 240$)	40	16.6
Spinal status ($n = 240$)		
Normal	136	56.43
Unknown	86	35.68
More than one	8	3.32
Fatty filum	4	1.66
Tethered cord	3	1.24
Myelomeningocele/spina bifida	3	1.24
Number of BMPs		
One	173	71.78
Two	58	24.07
Three	9	3.73
Four	1	0.41
BMP regimen ($n = 232$)		
Oral laxative/medication	146	60.58
Enema	72	29.88
Oral laxative/medication & enema	11	4.56
Oral laxative/medication & suppository	2	0.83
Enema & suppository	1	0.41

Age at BMP is at most recent BMP encounter

IQR interquartile range, *ADHD* Attention-deficit/hyperactivity disorder, *OCD* obsessive–compulsive disorder, *BMP* bowel management program

Table 2 Univariate analysis of difference in score pre- and post-BMP among patients having undergone one BMP

Measure	Pre-BMP		Post-BMP		Mean diff	95% CI		*P* value
	Min/Max	Median (IQR)	Min/Max	Median (IQR)		Lower	Upper	
VSS	2/37	14 (9. 20)	1/30	10 (6. 16)	3.6	2.72	4.48	< 0.0001
BCS	11/78	30 (26. 42)	5/68	20 (13. 28)	11.96	9.41	14.5	< 0.0001
CCCS	2/25	12 (9. 16)	0/21	10 (8. 13)	1.9	1.06	2.73	< 0.0001

IQR interquartile range, *CI* confidence interval, *BMP* bowel management program, *VSS* vancouver symptom score for dysfunctional elimination syndrome, *BCS* baylor continence scale, *CCCS* cleveland clinic constipation score

Table 3 Univariate analysis of difference in score pre- and post-BMP among patients having undergone more than one BMP

Measure	Pre-BMP		Post-BMP		Mean diff	95% CI		*P* value
	Min/Max	Median (IQR)	Min/Max	Median (IQR)		Lower	Upper	
VSS	1/33	11.5 (6, 20)	1/29	10 (5, 16)	1.66	0.23	3.09	**0.0233**
BCS	6/51	19.5 (12, 28)	6/41	16.5 (11, 23)	2.69	−0.91	6.28	0.14
CCCS	3/22	12 (8.5, 13)	2/16	8.5 (5, 11)	2.93	1.74	4.11	**< .0001**

Date of bowel management program is most recent encounter

$P < 0.05$ are in bold

IQR interquartile range; CI, confidence interval, *BMP* bowel management program, *VSS* vancouver symptom score for dysfunctional elimination syndrome, *BCS* baylor continence scale, *CCCS* cleveland clinic constipation score

improvement in the VSS, BCS, and CCCS ($P < 0.0001$) (Table 4). Patients who underwent multiple BMPs also showed improvement in their scores, though this improvement was only significant for BCS ($P = 0.0001$).

Marginal means estimates looking at changes in scores pre- and post-BMP by individual regimen indicate decreases in scores among regimens involving oral laxatives/medication or enemas (Table 5). Regimens involving oral laxatives/medication revealed significant decreases in all three scores ($P = 0.0001$ for VSS, $P = 0.0002$ for BCS, and $P < 0.0001$ for CCCS). Regimens using enemas resulted in larger mean differences in both the VSS and BCS (mean difference -4.05 (95% CI: − 6.51, − 1.58), $P < 0.0001$ for VSS; mean difference − 14.67 (95% CI: − 20.43, -8.91), $P < 0.0001$ for BCS).

Discussion

This study shows that there is significant improvement in urinary symptoms in children with FC who undergo a BMP. This is evidenced by the significant improvement in VSS. For challenging patients with BBD and FC, a BMP is a reasonable treatment strategy to treat lower urinary tract symptoms.

The VSS is a 14-item questionnaire that assesses urinary continence and also the severity of symptoms associated with BBD and fecal incontinence [7]. A score of 11 or higher is indicative of nonneurogenic lower urinary tract dysfunction/dysfunctional elimination syndrome, with a sensitivity of 80% and specificity of 91%. The median pre-BMP VSS score in our cohort was 14, which would characterize the patients as having DES. After the BMP, the median VSS decreased to a median of 10, thereby showing enough improvement in patient symptoms to the degree that they no longer were considered dysfunctional eliminators. This was true in patients who performed only one BMP and those who underwent multiple BMP. Both of these groups ultimately scored a median of less than 11 which suggests that improvement and resolution of BBD is possible with BMP alone.

It has been shown previously in patients with anorectal malformation who have underwent a BMP that the VSS improves as fecal continence in the population improves [15]. Children with anorectal malformation have known associated urologic abnormalities and commonly have difficulty with urinary and bowel elimination. Additionally, children with these malformations often have multiple surgical procedures that may impact bowel and bladder function. In contrast, children with FC have normal urologic and colorectal anatomy and rarely, if ever, has surgery been performed. Our finding that BMP alone significantly improves VSS in these patients reinforces the well-accepted concept that fecal retention can cause urinary symptoms, even in children with no underlying neurologic or anatomic abnormality. Our study is the first to our knowledge that shows that BMP alone can improve the VSS in children with FC. Additionally, we saw improvement in the BCS and CCCS scores as well. The BCS is a scoring measure used to assess

Table 4 Linear mixed effect regression modeling for each measure

	VSS			BCS			CCCS		
	Estimate (95% CI)	Standard Error	*P* value	Estimate (95% CI)	Standard Error	*P* value	Estimate (95% CI)	Standard Error	*P* value
Intercept	24.38 (20.04, 28.73)	2.2	**<.0001**	37.11 (30.56, 43.66)	3.31	**<.0001**	11.33 (8.68, 13.98)	1.34	<.0001
Catheterized									
Yes	5.76 – 0.34, 11.85)	3.09	0.064	9.51 (– 4.48, 23.50)	7.11	0.1819	– 0.06 (– 5.73, 3.82)	2.42	0.6921
No	Reference			Reference			Reference		
Number of BMPs									
One	Reference			Reference			Reference		
Two	– 1.41 (– 3.87, 1.04)	1.24	0.2571	– 7.22 (– 10.87, – 3.57)	1.85	**0.0001**	– 1.30 (– 2.77, 0.16)	0.74	0.0811
Three	– 4.56 (– 14.14, 5.02)	4.85	0.3487	– 11.85 (– 26.83, 3.13)	7.6	0.1205	– 0.95 (–6 .86, 4.96)	2.99	0.7505
BMP regimen									
Oral	Reference			Reference			Reference		
Enema	1.34 (– 1.19, 3.88)	1.29	0.2968	5.57 (1.36, 9.79)	2.14	**0.0098**	0.95 (– 0.66, 2.56)	0.82	0.2474
Oral & enema	– 2.41 (– 8.13, 3.30)	2.9	0.406	4.42 (– 4.21, 13.15)	4.44	0.3199	0.01 (– 3.35, 3.36)	1.7	0.9976
Oral & suppository	– 0.44 (– 12.15, 11.27)	4.6	0.4405	13.20 (– 9.08, 35.59)	11.33	0.2448	– 1.78 (– 10.03, 6.47)	4.19	0.6719
Enema & suppository	–	–	–	–	–	–	–	–	–
Time									
Pre-BMP	Reference			Reference			Reference		
Post-BMP	– 3.20 (– 4.09, – 2.30)	0.45	**<.0001**	– 6.41 (– 9.16, – 3.67)	1.39	**<.0001**	– 3.08 (– 4.04, – 2.13)	0.49	<.0001
Age at BMP	– 0.61 (– 0.89, – 0.33)	0.14	**<.0001**	0.45 (– 0.88, – 0.02)	0.22	**0.0409**	0.16 (-0.01, 0.33)	0.08	0.0585
^BMP regimen and pre/post-BMP									
Oral/pre-BMP	Reference			Reference			Reference		
Oral/post-BMP	Reference			Reference			Reference		
Enema/pre-BMP	Reference			Reference			Reference		
Enema/post-BMP	– 1.36 (– 3.31, 0.59)	0.99	0.17	– 8.26 (– 12.88, – 3.64)	2.34	**0.0005**	1.69 (0.12, 3.29)	0.8	0.035
Oral & enema/pre-BMP	Reference			Reference			Reference		
Oral & enema/post-BMP	– 1.03 (– 5.66, 3.61)	2.35	0.6634	1.30 (– 9.42, 12.02)	5.44	0.8116	3.71 (– 0.05, 7.48)	1.91	0.0532
Oral & suppository/pre-BMP	Reference			Reference			Reference		
Oral & suppository/post-BMP	– 4.0 (– 13.19, 5.19)	4.66	0.3919	– 13.11 (– 35.59, 9.37)	11.41	0.2518	6.18 (– 1.56, 13.92)	3.93	0.1167

All findings here were made with reference group being those on a medication regimen

$P < 0.05$ are in bold

CI confidence interval, *BMP* bowel management program, *VSS* vancouver symptom score for dysfunctional elimination syndrome, *BCS* baylor continence scale, *CCCS* cleveland clinic constipation score, *Oral* oral laxative/medication

social continence in children who have underwent repair of an anorectal malformation [17]. The patients in this cohort had FC, not an anorectal malformation, but both populations have been known to have concomitant urinary dysfunction

Table 5 Estimates of scores based on pre- and post-BMP regimen and comparison of means

VSS						
BMP Regimen	Pre-BMP	Post-BMP	Comparison of mean			
	Estimate (95% CI)	Estimate (95% CI)	Estimate ([a]95% CI)	Standard Error	*P* value	Adjusted *P* value
Oral	17.40 (12.92, 21.89)	14.72 (10.24, 19.19)	−2.69 (−4.43, −0.94)	0.57	< .0001	**0.0001**
Enema	18.75 (14.25, 23.24)	14.70 (10.32, 19.08)	−4.05 (−6.51, −1.58)	0.81	< .0001	**< .0001**
Oral & enema	14.99 (8.69, 21.28)	11.28 (4.91, 17.64)	−3.71 (−10.70, 3.28)	2.28	0.1055	0.7349
Oral & suppository	16.96 (4.56, 29.37)	10.28 (0.16, 20.39)	−6.69 (−20.84, 7.47)	4.63	0.1499	0.8353
[x]Enema & suppository	–	–	–	–	–	–
BCS						
BMP regimen	Pre-BMP	Post-BMP	Comparison of mean			
	Estimate (95% CI)	Estimate (95% CI)	Estimate ([a]95% CI)	Standard Error	*P* value	Adjusted *P* value
Oral	29.75 (21.05, 38.45)	23.33 (14.63, 32.04)	−6.41 (−10.68, −2.15)	1.39	< .0001	**0.0002**
Enema	35.32 (26.86, 43.79)	20.65 (11.85, 29.45)	−14.67 (−20.43, −8.91)	1.88	< .0001	**< .0001**
Oral & enema	34.17 (23.16, 45.17)	29.05 (16.90, 41.21)	−5.12 (−21.21, 10.98)	5.26	0.332	0.978
Oral & suppository	42.95 (19.23, 66.66)	23.42 (6.66, 40.18)	−19.52 (−54.14, 15.09)	11.32	0.0861	0.6715
[x]Enema & suppository	–	–	–	–	–	–
CCCS						
BMP regimen	Pre-BMP	Post-BMP	Comparison of mean			
	Estimate (95% CI)	Estimate (95% CI)	Estimate ([a]95% CI)	Standard Error	*P* value	Adjusted *P* value
Oral	11.41 (8.24, 14.59)	8.33 (5.15, 11.50)	−3.08 (−4.57, −1.60)	0.49	< .0001	**< .0001**
Enema	12.36 (9.20, 15.51)	10.96 (7.80, 14.13)	−1.40 (−3.33, 0.54)	0.63	0.028	0.3499
Oral & enema	11.42 (7.32, 15.51)	12.05 (7.50, 16.59)	0.63 (−5.03, 6.29)	1.85	0.734	1
Oral & suppository	9.63 (0.87, 18.40)	12.73 (6.24, 19.23)	3.10 (−8.82, 15.02)	3.9	0.427	0.9932
[x]Enema & suppository	–	–	–	–	–	–

$P < 0.05$ are in bold

CI confidence interval, *BMP* bowel management program, *Oral* oral laxative/medication

Adjusted *P* value from Tukey adjustment

[a]95% CI = Tukey adjusted confidence interval

[6, 11, 15]. The CCCS, while validated for adults, is a scoring system that assesses the severity of symptoms of constipation [16]. Improvement in both of these scores, along with the VSS, supports the interrelatedness of constipation and urinary dysfunction.

It has been reported that nearly 50% of patients seen in pediatric urology clinics have issues with BBD [1, 19]. Bowel and bladder dysfunction can cause negative physical and psychosocial effects on children and their families [4]. Given the economic and psychosocial effects of BBD, it is important to identify adequate treatments that may prevent additional morbidity for patients suffering from it. Additionally, urologic testing commonly requires catheterization, radiation exposure, and sometimes sedation. The objective improvement in VSS seen in our study suggests that if a formal BMP is initiated as a first-line treatment for dysfunctional elimination, symptoms may improve enough to eliminate the need for such testing or any additional intervention aimed at bladder control and relief of urinary symptoms, such as medication.

This study does have several limitations. First, we were not able to accurately describe symptomatic improvement of urinary symptoms, so utilized various scoring metrics to study this population in a standardized manner. The VSS itself demonstrates 80% sensitivity and 91% specificity for dysfunctional elimination syndrome/BBD, meaning we may not be identifying all patients with BBD [7]. While the VSS focuses mainly on urinary symptoms, it is possible that an improvement in bowel symptoms may have led to an improved score. We did not assess the association of these scores with stool and urinary continence. The BCS and CCCS were not validated for our population of pediatric patients with FC and BBD. The median time at which the VSS was measured post-BMP was 92 days and, therefore, we are unable to describe the longevity of the impact of a BMP on BBD, though long-standing effects may be

difficult to interpret due to the normal waxing and waning disease course of FC and the relationship between stool habits with diet and bowel regimen compliance. Future studies should identify long-term results. Lastly, this is a single-institution study performed at a specialized, high-volume center, so results may not be generalizable.

We conclude that in children with functional constipation and concomitant bladder dysfunction, a bowel management program significantly improves urinary symptoms. Thus, in this patient population, a bowel management program should be undertaken prior to any other measures to control urinary symptoms.

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Declarations

Competing interests The authors declare no competing interests.

Conflict of interest None.

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