

# Bowel management program for pediatric postoperative fecal incontinence in China

## A surgeon's experience

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### Abstract

The purpose of this article is to report the status of the efficacy of and long-term adherence to the Bowel Management Program (BMP) for fecal incontinence (FI) postoperation in China.

Children over 3 years of age with FI postoperation referred to our medical center were included in the study. Evaluations were performed before and 2 years after their clinic visit. The cost of bowel care, improvement in incontinence, health-related quality of life, and family functioning with the BMP were analyzed.

A total of 48 children with FI were included in our study, of whom 38 were boys. The median treatment fee was 660.1 dollars. The complications included abdominal pain (4 patients, 8%), occasional vomiting (2 patients, 4%), and hypoglycemia (1 patient, 2%). The incontinence status and health-related quality of life improved significantly after the BMP. Despite the good outcome of the BMP, half of the patients discontinued the program.

The BMP is an effective approach to manage FI and improve the patients' quality of life. Poor long-term adherence is currently the main challenge affecting the BMP application in China.

**Abbreviations:** ARM = anorectal malformation, BMP = Bowel Management Program, FI = fecal incontinence, HD = Hirschsprung disease, IQR = interquartile range.

**Keywords:** bowel management program, efficacy, fecal incontinence, long-term adherence

## 1. Introduction

Fecal incontinence (FI) is one of the most disturbing and psychologically distressing problems affecting children.<sup>[1]</sup> Several psychological problems are associated with unsuccessful bowel programs in the pediatric surgery field. It is a problem that affects more children than previously thought, affecting those born with anorectal malformation (ARM), Hirschsprung disease (HD), presacral tumor, or spinal cord problems.

Fecal continence depends on 3 main factors: voluntary sphincter muscles, anal canal sensation, and colonic motility.<sup>[2]</sup> Patients with true FI are those who lack voluntary bowel movement ability, either because they were born with a congenital malformation with a poor functional prognosis or because of hypothyroidism, hypocalcemia, allergy, and other pathogenesis; in this study, we focused on the former cases in the pediatric surgery field. We have learned how to evaluate these patients to recognize the exact pathogenesis of FI. Of the patients with ARM, 25% experience FI due to functional defecation disorder, weak sphincter muscles, iatrogenic injury, and abnormal innervation.<sup>[2]</sup> Yet, a small significant number of patients with HD (< 5%) experience FI<sup>[3,4]</sup> owing to their destroyed dentate line and bowel dismotility.<sup>[5]</sup> Patients with spinal problems, such as tethered cord, presacral tumor, or injuries, can lack voluntary bowel movement ability.<sup>[6]</sup>

The Bowel Management Program (BMP), developed by Drs Alberto Peña and Marc Levitt for over 30 years, has a low cost, can be implemented at home, and ensures that the patients remain clean for 24 hours.<sup>[5,7,8]</sup> It is an artificial tailored enema program to keep patients clean on a 24-hour basis. It is very popular in the United States<sup>[5,8,9]</sup> and some South American and Asian countries as well.<sup>[10,11]</sup> However, there have been no reports related to BMP in China. Since the implementation of the BMP in 2013, we have treated 48 cases to date. The objective of this study was to evaluate the status of BMP implementation in a children's medical center. We summarized the effectiveness of the BMP in improving the quality of life of children with FI. The patients' bowel function, satisfaction with the program, treatment cost, and family acceptance were also assessed.

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## 2. Methods

This cross-sectional study was conducted in a children's medical center in South China. Children above 3 years of age with FI postoperations who underwent the BMP from February 2013 to December 2015 were considered for this study. The children who did not participate in all the interviews pertaining to data collection or who had FI not related to their primary diagnosis of ARM, HD, presacral tumor, or spinal cord problems or who were treated successfully with diet and drug were excluded from the study. This study was approved by the ethics committee of Guangzhou Women and Children's Medical Center. In total, 48 patients fulfilled the inclusion criteria of the study.

The BMP includes administration of daily enemas with saline solution alone or with glycerin, with the volume and concentration determined via trial and error over a period of 1 week based on the clinical response and the amount and distribution of stool in the colon (as revealed by a plain abdominal X-ray).<sup>[5,7,8]</sup>

We collected data related to the FI cases treated with the BMP via a face-to-face interview or phone. The basic data collected from the patients included sex, age, time required for the treatment process, treatment cost, and BMP time. We observed the clinical efficacy of the BMP in terms of the improvement in the defecation symptoms.

The Cleveland score was used to evaluate the efficacy of the program for FI; the score ranges from 0 to 20, and the test evaluates the type (solid, fluid, or gas) and frequency of stools (0=perfect control, 20=severe incontinence). In 2013, all the parents who answered the first questionnaire were asked to complete a second questionnaire in 2015 via a phone call or a face-to-face interview.

The PedsQL 4.0 questionnaire was filled out by the parents and was used to assess the quality of life before and after the program.<sup>[10,11]</sup> The questionnaire had 4 modules with 23 items encompassing the physical (8 items), emotional (5 items), social (5 items), and school (5 items) domains. The items were reverse-scored and linearly transformed to a scale from 0 to 100 (0=100, 1=75, 2=50, 3=25, 4=0), with high scores indicating a better quality of life.

### 2.1. Statistical analysis

Continuous variables were expressed as medians and interquartile ranges (IQR). The paired two-tailed Student *t* test was used to compare the Cleveland FI and PedsQL scores before and after the program when the data conformed to a normal distribution. Otherwise, the Wilcoxon-signed rank test was used. Correlations between the Cleveland FI score and PedsQL score as well as its dimension scores were assessed using the Spearman correlation analysis. Results with  $P < .05$  were considered statistically significant. All the analyses were performed using the R version 3.3.2.

## 3. Results

### 3.1. Characteristics of the patients

The patient characteristics are listed in Table 1. Of the 48 patients who met the inclusion criteria, 26 were born with ARMs, 12 had Hirschsprung disease, and the remaining had meningocele, tethered cord, and sacral teratoma. Thirty-eight of the patients were boys. The median treatment fee was 660.1 (IQR: 471.0–821.8) dollars. The median age and follow-up period were 7 years (IQR: 6–8 years) and 1.75 years (IQR: 1.0–2.0 years), respectively. Only 2 patients agreed to undergo the Malone procedure. Complications

**Table 1**

### Characteristics of children with fecal incontinence.

Variables	n=48
Sex	
Boys	38/48
Age, y	7.0 (5.3–8.0)
Diagnosis	
ARM	26/48
HD	12/48
M+TT	8/48
ST	2/48
Treatment fee, dollar	660.1 (471.2–821.8)
Hospital stay, d	7 (6–8)
BMP time, y	1.75 (1.0–2.0)
Malone procedure	2/48

Data are presented as median (interquartile range).

ARM=anorectal malformation, HD=Hirschsprung disease, M=meningocele, ST=sacral teratoma, TT=tethered cord.

included abdominal pain (4 patients, 8%), occasional vomiting (2 patients, 4%), and hypoglycemia (1 patient, 2%).

### 3.2. Treatment effects before and after the program

Table 2 shows that the Cleveland FI score significantly decreased after the program (from 10.42 to 2.60). Compared with the scores before the program, there were statistically significant increases in the PedsQL (from 59.14 to 73.09), emotional function (from 36.67 to 63.65), social function (from 35.42 to 57.81), and school function (from 40.00 to 54.79) scores at the end of the study,<sup>[8,9]</sup> other than the physical function score.

### 3.3. Correlations between the Cleveland FI score and PedsQL score

To assess the correlation between the Cleveland FI score and PedsQL score, the Spearman correlation analysis was used. As shown in Table 3, changes in the Cleveland FI score were significantly negatively correlated with the changes in the total PedsQL score ( $r = -0.32$ ,  $P < .05$ ) and the emotional function score ( $r = -0.42$ ,  $P < .01$ ).

### 3.4. Trends in the program and improvement in the Cleveland FI score and PedsQL score

As shown in Fig. 1, the total PedsQL, emotional function, social function, and school function scores increased with the program.

**Table 2**

### Comparisons of the Cleveland FI score and PedsQL score before and after treatment.

	Score		P
	Pre	Post	
Cleveland FI score	10.42 ± 4.12	2.60 ± 1.83	<.001*
PedsQL total score	59.14 ± 7.59	73.09 ± 7.04	<.001†
Physical function	100	100	—
Emotional function	36.67 ± 16.89	63.65 ± 17.56	<.001*
Social function	35.42 ± 18.27	57.81 ± 16.01	<.001†
School function	40.00 ± 11.81	54.79 ± 10.67	<.001†

FI=fecal incontinence, PedsQL=Pediatric Quality of Life Inventory.

\* *P* values are shown for comparisons of absolute scores using Wilcoxon signed rank test.

† *P* values are shown for comparisons of absolute scores using paired Student *t* test.

**Table 3**  
**Spearman correlations between the Cleveland fecal incontinence and pediatric quality of life inventory scores.**

Variables	PedsQL total score	Emotional function score	Social function score	School function score
Cleveland FI score before treatment	-0.35 <sup>†</sup>	-0.15	-0.38 <sup>‡</sup>	-0.25
Cleveland FI score after treatment	0.04	-0.24	0.25	-0.01
Change of Cleveland FI score <sup>*</sup>	-0.32 <sup>†</sup>	-0.42 <sup>‡</sup>	-0.14	-0.09

FI=fecal incontinence, PedsQL=Pediatric Quality of Life Inventory.  
<sup>\*</sup>Change of Cleveland FI score=Cleveland FI score before treatment – Cleveland FI score after treatment.  
<sup>†</sup>P<.05.  
<sup>‡</sup>P<.01.

Conversely, the Cleveland FI score decreased, indicating that both anorectal function and quality of life improved following the BMP.

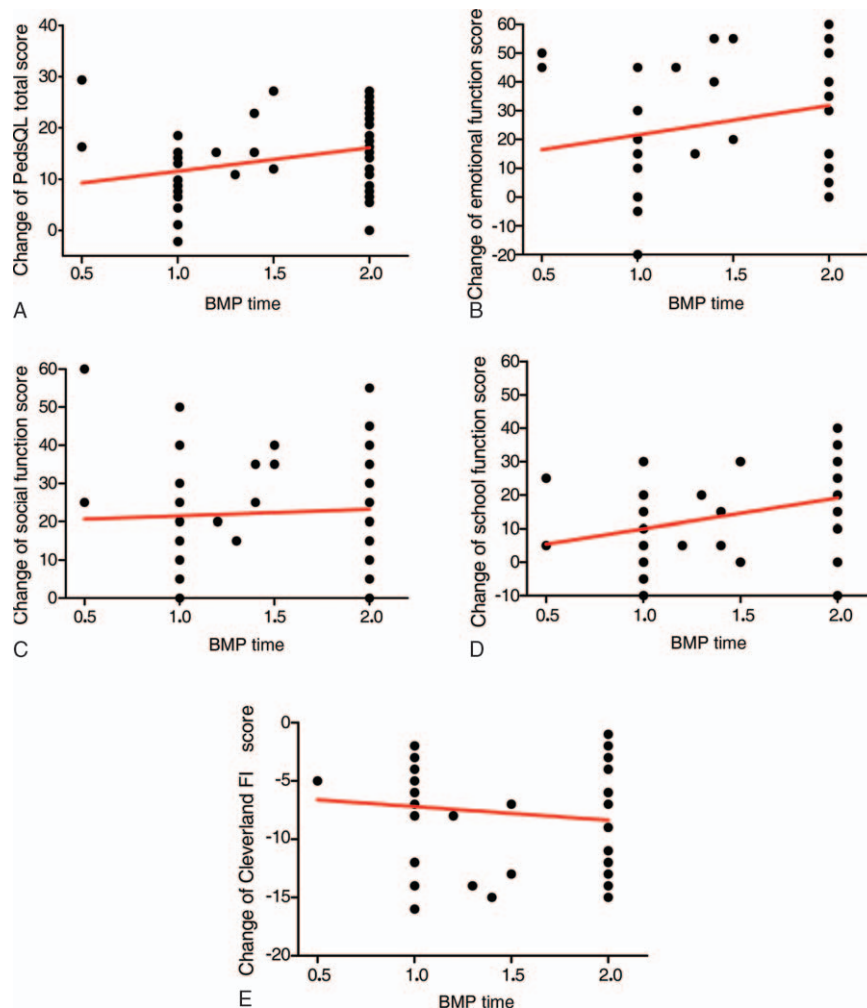
1 patient abandoned the program because of the high treatment fee (Fig. 2).

**3.5. Reasons for abandonment of the program**

Twenty-four patients discontinued their BMP; the parents of 9 patients believed there should be better methods, such as operations and defecation training, that could allow their children to gain bowel control. About 4 (17%) and 4 (17%) of the parents commented that they are too busy and that the implementation of the program is complicated, respectively. Only

**4. Discussion**

This is the first study that illustrates the practice and outcomes of bowel care for pediatric FI in China. The most interesting result from our study is that even though the patients showed improvements in the quality of life and bowel care, ~50% discontinued the BMP, and most of them refused to undergo the Malone procedure to follow the program.



**Figure 1.** Trends of the BMP time and effects (changes in the total PedsQL score and its dimensions and Cleveland FI score). BMP = Bowel Management Program, FI=fecal incontinence.

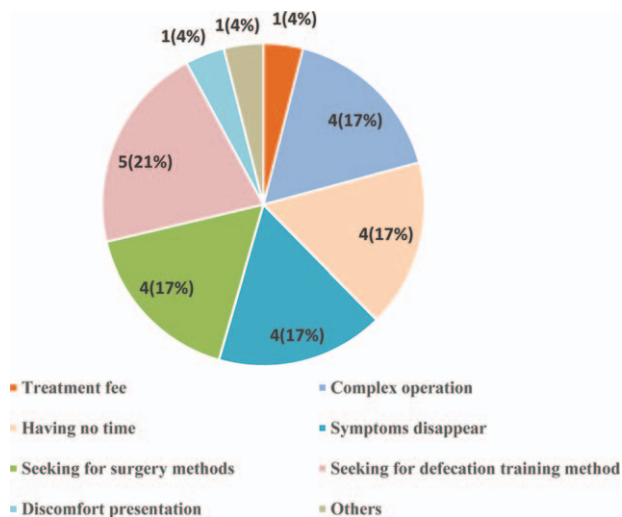


Figure 2. Reasons for abandonment.

There are several therapeutic approaches to treat FI in China, including biofeedback, antegrade enema, sphincteroplasty, sphincter replacement, or colostomy, when all other treatment methods fail. Biofeedback is a noninvasive approach; however, its functional results are dubious in children.<sup>[12,13]</sup> Surgical interventions, such as rebuilding or replacement of the anal sphincter, showed “good” results in 20% to 60% of the cases; however, the complication rates were high at 50% to 60%.<sup>[14]</sup> With this, the Malone procedure may be considered one of the options for the treatment of FI. However, antegrade enema should be recommended only after proving that the bowel management has been successful. This enema requires only a different route; the key to its success depends on the enema formula and not the route.<sup>[8]</sup>

The Cleveland score based on a questionnaire was lower in patients with better social continence. We decided to use the Cleveland score because it is easy to use and complete and can be useful in prospective studies.<sup>[15]</sup> The adoption of the BMP in our study enabled a dramatic reduction in the FI after the BMP. The incontinence status of some patients who discontinued their BMP for various reasons was better than that of those who continued their BMP, possibly owing to an improvement in the bowel function using enema or bowel control with age;<sup>[16–20]</sup> further, the parents who participated in the program may have started to notice the importance of eating habits, including not eating very cold or spicy food, not eating too many snacks, and maintaining a 3-meal-a-day diet plan for their children.

Another important method to measure the severity of FI is to determine its impact on the quality of life of the patients. The PedsQL 4.0 survey has been widely used to assess the quality of life of children in general.<sup>[21]</sup> The PedsQL is a modular instrument designed to measure the health-related quality of life of healthy and acutely/chronically ill children and adolescents (aged 0–18 years). We used the PedsQL 4.0 for our cases. Miner reported that the physical function of children with FI is poorer than that of normal children, probably because of the dampness related to the diaper.<sup>[22]</sup> Parents in China want their children to live the same lifestyle as normal children; thus, they prefer using pants over using diaper wraps. As such, patients have greater freedom to walk, run, and play sports; moreover, they can have a normal physical presentation. In our cases, the poor quality of life

was mainly related to the emotional and learning function domains. The children with FI were more sensitive and shy in communicating and always had more difficulties in concentrating than normal children. Our data suggest that compared with the children who discontinued their BMP a few months later, those who continued their BMP showed significant improvements in their quality of life, especially in the emotional and school function domains; their bowel function was also observed to improve.

The other finding of this study is that the FI score was highly correlated with the quality-of-life measures. The changes in the anal function were closely related to the quality of life before and after the program, especially in the emotional function. The relationship between FI and quality of life has been repeatedly observed, which indicates that children with FI have a lower quality of life than healthy children. Grano et al<sup>[23]</sup> investigated how FI could influence the different aspects of the quality of life of children and adolescents with ARM. Further, Bai et al<sup>[24]</sup> reported that the incidence of behavioral problems was significantly higher in children with poor fecal continence than in those with good fecal continence.

Although there were great improvements in the quality of life and the FI score in our patients with FI, the 2-year follow-up call revealed that 50% of the families discontinued the program. The analysis of the reasons for abandonment showed that some parents were more inclined to seek operations to achieve optimal bowel control, as they cannot accept the truth that their children will experience true incontinence for their entire life. This can explain why many parents did not select the Malone procedure (only 2 cases agreed to undergo the procedure); they believe that performing the Malone procedure would mean giving up the opportunity of achieving optimal bowel control by other means. Some parents attempt to train their children at home to defecate in their own way. They order their children to practice defecation regularly (twice or thrice a day) or use a dilator to stimulate their children’s anus to make them “feel” the sensation corresponding to defecation; most parents also report that it is, indeed, effective. As for the reason of having no time and/or finding the BMP implementation complicated, we believe that the key reason for discontinuing the BMP is their lack of confidence in the BMP itself and the fact that they want to cure their children “completely” using other approaches.

We report some complications in our study, including abdominal pain, occasional vomiting, and hypoglycemia ( $n=1$ ) after the enema administration. We believe that this may be due to the hyperosmosis of glycerin. Thus, we have guidelines for the concentration of the enema, stating that not more than 30 mL of glycerin (in 500 mL saline) must be used.

The limitations of this study include its retrospective design and small sample size. As it is a questionnaire-based study, it relied on the respondents’ memories of their bowel care, which may have introduced a recall bias. In addition, it is a single-center retrospective study, we missed the specialized tests to rule out conditions such as hypothyroidism, hypocalcemia, lead poisoning, celiac, and allergy in clinical, but now we have started these protocols to complete our diagnosis for appropriate treatments. Considering that it was conducted in a country with a population of more than 1 billion, the sample population of the study does not represent the general population. Besides, in future studies, age-matched healthy samples drawn from local populations would be optimal to allow for a more direct comparison between the population with FI and healthy control children.



## 5. Conclusions

In conclusion, this study demonstrates that the BMP can be successfully implemented in China, despite its limitations. It can significantly improve the quality life of children with FI. However, great efforts are required to make the program acceptable to parents in China. As surgeons, we should explain the pathophysiological issues of the BMP patiently and in considerable detail to the families; further, the government should consider establishing an FI treatment center. Early interventions for FI can greatly improve the patient's quality of life and reduce the psychological impact on children with FI.

## References

- [1] Yusuf SA, Jorge JM, Habr-Gama A, et al. [Evaluation of quality of life in anal incontinence: validation of the questionnaire FIQL (Fecal Incontinence Quality of Life)]. *Arq Gastroenterol* 2004;41:202–8.
- [2] Pena A, Levitt MA. Colonic inertia disorders in pediatrics. *Curr Probl Surg* 2002;39:666–730.
- [3] Bax KN. Duhamel Lecture: the incurability of Hirschsprung's disease. *Eur J Pediatr Surg* 2006;16:380–4.
- [4] Teitelbaum DH, Drongowski RA, Chamberlain JN, et al. Long-term stooling patterns in infants undergoing primary endorectal pull-through for Hirschsprung's disease. *J Pediatr Surg* 1997;32:1049–52.
- [5] Levitt M, Peña A. Update on paediatric faecal incontinence. *Eur J Pediatr Surg* 2009;19:1–9.
- [6] Vande Velde S, Van Biervliet S, Van Renterghem K, et al. Achieving fecal continence in patients with spina bifida: a descriptive cohort study. *J Urol* 2007;178:2640–4.
- [7] Pena A, Guardino K, Tovilla JM, et al. Bowel management for fecal incontinence in patients with anorectal malformations. *J Pediatr Surg* 1998;33:133–7.
- [8] Bischoff A, Levitt MA, Bauer C, et al. Treatment of fecal incontinence with a comprehensive bowel management program. *J Pediatr Surg* 2009;44:1278–83.
- [9] Colares JH, Purcaru M, da Silva GP, et al. Impact of the Bowel Management Program on the quality of life in children with fecal incontinence. *Pediatric surgery international* 2016;32:471–6.
- [10] Colares JH, Purcaru M, da Silva GP, et al. Impact of the Bowel Management Program on the quality of life in children with fecal incontinence. *Pediatr Surg Int* 2016;32:471–6.
- [11] Engkasan JP, Sudin SS. Neurogenic bowel management after spinal cord injury: Malaysian experience. *J Rehabil Med* 2013;45:141–4.
- [12] Norton C, Chelvanayagam S, Wilson-Barnett J, et al. Randomized controlled trial of biofeedback for fecal incontinence. *Gastroenterology* 2003;125:1320–9.
- [13] Wang X, Li H. [Current status of the treatments of fecal incontinence]. *Zhonghua Wei Chang Wai Ke Za Zhi* 2014;17:1244–50.
- [14] Tan JJ, Chan M, Tjandra JJ. Evolving therapy for fecal incontinence. *Dis Colon Rectum* 2007;50:1950–67.
- [15] Rockwood TH, Church JM, Fleshman JW, et al. Fecal Incontinence Quality of Life Scale: quality of life instrument for patients with fecal incontinence. *Dis Colon Rectum* 2000;43:9–16.
- [16] Conway SJ, Craigie RJ, Cooper LH, et al. Early adult outcome of the Duhamel procedure for left-sided Hirschsprung disease—a prospective serial assessment study. *J Pediatr Surg* 2007;42:1429–32.
- [17] Menezes M, Corbally M, Puri P. Long-term results of bowel function after treatment for Hirschsprung's disease: a 29-year review. *Pediatr Surg Int* 2006;22:987–90.
- [18] Moore S, Albertyn R, Cywes S. Clinical outcome and long-term quality of life after surgical correction of Hirschsprung's disease. *J Pediatr Surg* 1996;31:1496–502.
- [19] Yanchar NL, Soucy P. Long-term outcome after Hirschsprung's disease: patients' perspectives. *J Pediatr Surg* 1999;34:1152–60.
- [20] Hartman EE, Oort FJ, Aronson DC, et al. Explaining change in quality of life of children and adolescents with anorectal malformations or Hirschsprung disease. *Pediatrics* 2007;119:e374–83.
- [21] Varni JW, Seid M, Kurtin PS. PedsQL™ 4.0: Reliability and validity of the Pediatric Quality of Life Inventory™ Version 4.0 Generic Core Scales in healthy and patient populations. *Med Care* 2001;39:800–12.
- [22] Miner PBJr. Economic and personal impact of fecal and urinary incontinence. *Gastroenterology* 2004;126(1 suppl 1):S8–13.
- [23] Grano C, Aminoff D, Lucidi F, et al. Long-term disease-specific quality of life in children and adolescent patients with ARM. *J Pediatr Surg* 2012;47:1317–22.
- [24] Bai Y, Yuan Z, Wang W, et al. Quality of life for children with fecal incontinence after surgically corrected anorectal malformation. *J Pediatr Surg* 2000;35:462–4.