

# Role of daily anal stimulation for intractable functional constipation in infants

Tianqi Zhu, Xiaoyi Sun, Dan Zhu, Qian You, Xiaoting Wan, Xinyao Meng, Jiexiong Feng

**To cite:** Zhu T, Sun X, Zhu D, *et al.* Role of daily anal stimulation for intractable functional constipation in infants. *World Jnl Ped Surg* 2020;3:e000135. doi:10.1136/wjps-2020-000135

Received 16 April 2020

Revised 19 May 2020

Accepted 3 June 2020

## ABSTRACT

**Objective** Functional constipation is one of the most common problems in pediatric gastroenterology. The aim of the present study was to evaluate the effectiveness of daily anal stimulation in infants with intractable functional constipation (IFC). Our evaluation was based on clinical improvement and on changes in manometric parameters through time.

**Methods** Infants with IFC treated between January 2018 and December 2019 were included in this retrospective study. Treatment processing included daily anal stimulation for infants and psychological counseling for parents. All cases underwent a complete intervention program and were evaluated for improvement in symptoms and for changes in anorectal manometry within 1 year of follow-up.

**Results** A total of 161 patients were included in this study. Positive response was achieved in all patients. Frequency of defecation, change in stool form and decrease in the defecatory pain were significantly relieved in all infants after intervention. On anorectal manometry, no significant difference was found in the peristaltic frequency of distal rectum before and after treatment. There was a significant increase in the mean amplitude of peristalsis and improvement in the rhythm after intervention. In the 1 year of follow-up, three patients had recurrence of constipation requiring colectomy.

**Conclusion** In terms of the high clinical efficacy and limited side effects, daily anal stimulation may be included in the initial part of an intervention program for IFC in infants.

## Key messages

### What is already known about this subject?

- ▶ Infantile functional constipation is a very common disease that afflicts many patients and parents.
- ▶ There are various ways to treat functional constipation, but a number of infants with intractable constipation still lack of effective treatment.
- ▶ Toilet training through anal stimulation was widely used in clinical practice.

### What are the new findings?

- ▶ This is the first retrospective study with a large sample size on the use of toilet training for treatment of infantile functional constipation.
- ▶ Daily anal stimulation is a safe toilet training program, with rare side effects.
- ▶ Early toilet training is effective for infantile functional constipation; it triggers defecation regularly, and it effectively builds proper toilet habits.

### How might it impact on clinical practice in the foreseeable future?

- ▶ This study preliminarily proved the effectiveness of daily anal stimulation. Daily anal stimulation may be included in the initial part of an intervention program for intractable functional constipation in infants.



© Author(s) (or their employer(s)) 2020. Re-use permitted under CC BY-NC. No commercial re-use. See rights and permissions. Published by BMJ.

Department of Pediatric Surgery, Tongji Hospital of Tongji Medical College of Huazhong University of Science and Technology, Wuhan, China

## Correspondence to

Dr Jiexiong Feng; 31699788@qq.com

## INTRODUCTION

Childhood functional constipation (FC) is a common clinical problem, often seen in the first year of life.<sup>1</sup> Symptoms consist of infrequent, painful defecation and/or passage of hard stools,<sup>2</sup> and the Rome IV criteria represent the widely accepted method for diagnosis of FC in the pediatric age group.<sup>3</sup> The management of FC in infants continues to be challenging. The available treatments include oral probiotic and laxatives to assure painless defecation, which is an important part of the treatment.<sup>3</sup> However, some infants experience severe symptoms for long duration that respond poorly to the conventional dietary and pharmacological

management, and these infants are considered to have intractable FC (IFC).<sup>4</sup>

Many studies have described various alternative treatments, including toilet training, biofeedback, and other intervention methods, for FC in adolescents or adults, and the responses were almost significantly improved.<sup>5,6</sup> However, experience and reports of these treatments in infants are lacking. The primary aim of this study was to evaluate the application value of daily anal stimulation in infants with IFC.

## METHODS

### Study population

This study included consecutive infants with chronic constipation based on the Rome IV criteria<sup>3</sup> treated between January 2018 and

December 2019 at the children constipation outpatient clinic of our institution, a tertiary university hospital in China. The trial was registered at ClinicalTrials.gov (registration number: NCT01985646). All the procedures complied with the Declaration of Helsinki. Patients or the public were not involved in the design, conduct, reporting, or dissemination plans of our research.

Inclusion criteria were as follows: (1) age less than 1 year with the diagnosis of FC based on Rome IV criteria; according to these criteria, at least two of the following problems had to have been reported: two or fewer defecations per week, a history of excessive stool retention, painful bowel movements or hard bowel movements, and large-diameter stools, and the presence of a large fecal mass in the rectum<sup>3</sup>; (2) received or were receiving medication treatment (lactulose and probiotics and others) by their primary gastroenterologist, but were dependent on the medications or unresponsive to medication treatment with persistence of constipation for at least 1 month.

Exclusion criteria were as follows: (1) constipation due to organic causes diagnosed by histopathology or anorectal manometry (ARM) examination, such as Hirschsprung disease (HSCR) or internal anal sphincter achalasia (IASA), and so on; (2) personal history of anorectal malformation; (3) family history of inflammatory bowel disease, HIV infection, neurologic or psychiatric disorder, and so on.

### Treatment protocol

All the treatments were conducted on an outpatient basis. All the parents were educated about the study protocol after which written informed consent was collected from all participating parents. The intervention program included daily anal stimulation for infants and psychological counseling for parents.

### Anal stimulation

For IFC infants, anal stimulation with specific bougie was used to promote regular defecation. The bougies were made of plexiglass, which were approximately 21 cm in length and of various diameters ranging from 11 to 22 mm (F11–F22). Before therapy, the bougies were selected based on the anal size of the patient determined by digital rectal examination. In the beginning, the appropriate bougie was covered with paraffin and inserted into the anus gently for approximately 3–5 cm in depth. Usually obvious bowel movement patterns of infants could be observed, including passing of gas, groaning, crying, or other sounds related to distress, and an attempt to push out the bougies from the anus. Meanwhile, the caregiver gave vocal signals to promote his bowel movement, specifically a baby music sound, and a clockwise abdominal massage, which promoted intestinal peristalsis and assisted defecation smoothly.<sup>7</sup>

Patients underwent regular daily intervention for at least 1 month, up to a maximum of 3 months. Each intervention lasted about 5–10 minutes. At the same time, as a part of the training program and to enhance compliance,

all the participating patients were called for monthly clinical visits until 1 year of follow-up.

### Psychological counseling

Health education and the relevant materials were provided to the parents or caregivers, to help them fully understand the pathophysiology of FC. The entire intervention process was explained with the help of diagrams. The parents and caregivers were given opportunity to discuss any queries related to the nursing process. The importance of compliance was also explained to the parents.

### ARM procedure

ARM (Medical Measurement Systems, Netherlands) was performed before the treatment courses, with the patient in the lying position, using an 8-channel water perfusion catheter. The catheter was inserted in the anal canal, and manometric parameters, including rectoanal inhibitory reflex (RAIR) threshold, peristaltic frequency and amplitude of distal rectum, and anal canal pressure at rest, were measured.

ARM was performed in all children at the time of presentation and was compared with follow-up manometry results that were performed within 1 month after intervention. All procedures were performed by the same operator.

### Outcomes measured

The primary outcome was the change in the frequency of stool passage. The secondary outcomes included changes in the stool form, incidence of defecatory pain, and improvement of parameters in ARM.

### Statistical analysis

All results of pretreatment versus post-treatment comparisons were analyzed statistically. Continuous data were reported as the mean±SD.  $\chi^2$  test was used to analyze categorical variables (large-diameter stools or painful defecation). Differences in the ARM parameters between the groups were assessed by the Student's paired t-test for continuous data, such as resting pressure, RAIR threshold, and peristaltic frequency and amplitude of the distal rectum. A p value <0.05 was considered to be statistically significant. Data were analyzed using SPSS software (V.14.0 for Windows, IBM).

## RESULTS

A total of 161 infants with IFC were enrolled in the study. The baseline characteristics of the patients are included in [table 1](#).

### Reduction of symptoms

Of the 161 patients receiving toilet training intervention, all had positive response. Frequency of defecation before treatment was 2.6±1.4 (range: 1–4) per week and this increased to 9.3±2.3 (range: 7–12) per week. We were

**Table 1** Baseline characteristics of the patients

Variable	Value
Male, n (%)	83 (51.6)
Age at the beginning of toilet training intervention (months), mean (range)	5.2 (3–11)
Mean duration of toilet training intervention (months), mean (range)	1.4 (1–3)
Age at the end of follow-up (months), mean (range)	13.4 (11–15)

able to wean down or stop the laxatives in 116 (72.1%) infants after treatment.

Stool form became soft from dry and hard in 150 (93.2%) patients. The incidence of crying defecation decreased from 100 (62.1%) patients at the start of treatment to 21 (13.1%) patients at the end of treatment suggesting that most infants had relief of symptoms (table 2).

Patients were also evaluated for symptoms at each follow-up visit. During the first year of follow-up, most patients had a beneficial response, but three patients (0.02%) had recurrence of constipation. These patients underwent radical colectomy at the age of about 2 years. The histopathological examination of the resected colon revealed intestinal neuronal dysplasia (IND) in two patients and hypoganglionosis in one patient.

### Anorectal manometry

On examining the anal sphincter relaxation, we found all children to have RAIR. The RAIR was present at the mean threshold volume required to elicit RAIR which was similar before and after the treatment (13.38±7.38 mL vs 11.62±5.02 mL,  $p=0.15$ ). The mean anal resting pressure was 64.47±21.3 and 66.61±20.3 mm Hg before and after the treatment, respectively. No difference was found in the peristaltic frequency of distal rectum between before and after treatment (figure 1). However, there was significantly positive response in the peristaltic amplitude of distal rectum. The mean peristalsis amplitude increased

from 7.617±4.22 to 15.723±7.11 mm Hg, and the rhythm improved significantly after intervention (table 2).

### Adverse effects

One infant developed anal fissure at the first intervention which resolved after readjustment of bougie size. No patient developed soiling or bowel perforation (table 3).

### DISCUSSION

Currently, the key aspects of treatment for FC include the use of laxatives, dietary modifications, behavioral therapy, and so on.<sup>2</sup> In infants, oral and/or rectal laxatives and probiotics are the first-line treatments for fecal impaction.<sup>8</sup> However, the symptoms of patients with IFC often persist, and relapses are common.<sup>9</sup> The quality of life is adversely affected in various aspects including social interactions, school achievements, and self-esteem. And finally, surgery decisions were made in the management of children with IFC in many centers.<sup>4 10</sup>

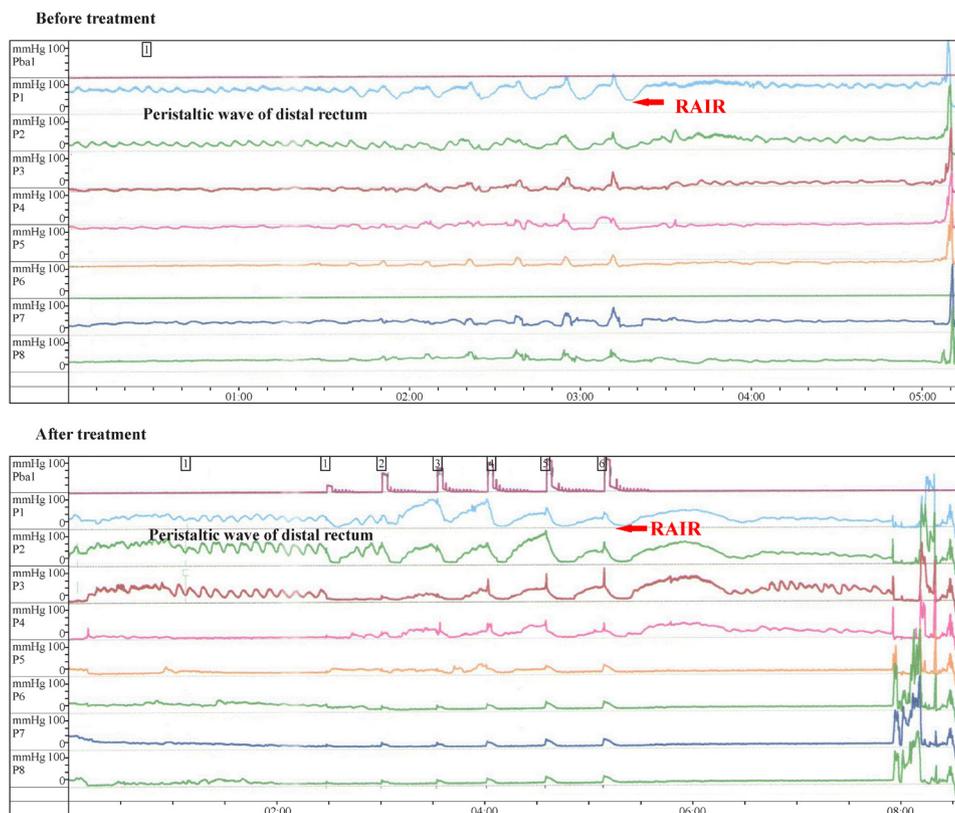
Nowadays, the optimal time of toilet training for infants is still controversial between eastern and western countries.<sup>11</sup> In Chinese culture, early toilet training is practiced as it helps the parents to make the infants aware about the sensory of defecation, gives sufficient time for the infant to get adapted to toilet training, and makes the transition to independent toileting easier for the infants.<sup>12</sup> In the past, the ‘infant toilet training’ approach received a lot of media attention, but it was infrequently used in routine clinical practice and was not discussed in the published guidelines.<sup>13</sup>

In the current study, the toilet training program was started as early as 3 months of age. During the follow-up period we found that most infants benefited from promoting intestinal peristalsis and stimulating defecation, and they were trained to control stool activities spontaneously, including the muscles involved in defecation, and to establish a defecation reflex.<sup>14 15</sup> Clinically, the ability to control the defecation and to achieve coordinated rectal muscle contraction with sphincter relaxation

**Table 2** The comparison of severity of symptoms and manometric results preintervention and postintervention

Variable	Toilet training intervention		P value
	Pretreatment	Post-treatment	
Frequency of defecation per week, mean±SD	2.6±1.4	9.3±2.3	0.023
Large-diameter stools, n (%)	125 (77.6)	11 (6.8)	0.005
Painful defecation, n (%)	100 (62.1)	21 (13.1)	0.012
Manometry parameters, mean±SD			
Resting pressure (mm Hg)	64.47±21.3	66.61±20.3	0.472
RAIR threshold (mm Hg)	13.38±7.38	11.62±5.02	0.150
Peristaltic frequency of distal rectum (/min)	9.925±2.22	10.547±2.10	1.102
Peristalsis amplitude of distal rectum (mm Hg)	7.617±4.22	15.723±7.11	0.001

RAIR, rectoanal inhibitory reflex; SD, standard deviation.



**Figure 1** Changes of the anorectal manometry before and after toilet training intervention. RAIR, rectoanal inhibitory reflex.

matures over time.<sup>16</sup> Undoubtedly, toilet training accelerated this process.

Furthermore, the treatment method of toilet training program was also varied. For a long time, anal stimulation is considered to be less invasive and commonly employed in China, and use of mechanical stimulants, such as soap or cooking oil, or other types of anal stimulation, was popular. By using daily anal stimulation as a part of toilet training process, the effect is generally believed to present recognizable motility pattern and to stimulate colonic propagated contraction ('colo-anal reflex').<sup>17</sup> Finally, the aim is to promote defecation regularly and to build proper toilet habits. To the best of our knowledge, this is the first study to evaluate the effects of daily anal stimulation in infants with IFC.

At the same time, all the parents were provided with health education and psychological counseling. The anxiety and depression of parents were relieved, and trust was created between doctors and patients, so as to obtain their full cooperation and compliance. Therefore, the process of intervention could be performed in a pleasant and encouraging environment. Furthermore,

besides mechanical anal stimulation, close touch from caregivers, such as specific whisper nursery rhymes and soft abdominal massage, could also arouse defecation awareness and could play important roles in the onset of toilet training with more sensitive approaches.<sup>12</sup>

ARM is an effective tool to assess anorectal sensations, pressure changes, and rectal compliance.<sup>18</sup> In this study, we evaluated the manometric parameters in infants to detect possible changes in the defecation dynamics and anorectal neuromuscular functions before and after intervention. All children had intact RAIR, which is important to exclude HSCR and IASA. The frequency of peristalsis in the distal rectum was similar before and after intervention. However, the peristaltic wave of distal rectum was disorganized, with slow, irregular waveform and low amplitude before intervention. After intervention, the amplitude of peristalsis became significantly greater and the rhythm also significantly improved (table 2). These findings suggested that with intervention the intestinal peristalsis improved and the coordination function of defecation was obviously strengthened. The mean resting pressure measured in a quiet state mainly reflects the tension of the internal sphincter. A normal sphincter apparatus helps patients to achieve control over the muscles of the defecation.<sup>19</sup> Although normal values for ARM have been published in adults, there have been few studies reporting the manometric data of normal and constipated infants. In this study, there was no significant difference in the anal resting pressure before and after intervention. This means that the intervention, including

**Table 3** Side effects of toilet training intervention

Variable	Value
Anal fissure, n (%)	1 (0.6)
Bowel perforation, n (%)	0
Soiling, n (%)	0

anal stimulation, did not adversely impact the structure and function of the internal sphincter.<sup>20</sup>

In order to support the management program and to enhance compliance, all patients underwent monthly clinical visits during the first year of follow-up. We found that most patients had improvement in defecation lasting at least 1 year suggesting that the toilet training had a beneficial response. Three patients had recurrence of symptoms over 6 months and required colectomy. Among these patients, two had IND and one had hypoganglionosis on histopathology of the resected colon. These are rare entities of congenital neuronal intestinal malformation and are classified as variants of HSCR.<sup>21</sup>

This study found that daily anal stimulation was one treatment of the early toilet training program (as early as 3 months of age) and might be an effective strategy for the management of infants with IFC. It triggers defecation regularly and effectively builds proper toilet habits.

There were some limitations in the present study. First, retrospective single-center research was the main limitation; then, uncontrolled study and short-term follow-up also limited our results and conclusion. Therefore, future prospective randomized controlled trial studies are required to validate our findings and to further improve the outcomes.

**Contributors** TZ designed the study and drafted the manuscript. XS contributed to the interpretation of the critical revision of the manuscript for important intellectual content and approved the final version of the manuscript. DZ, QY, and XW were involved in practice of training program and nursing care. XM drafted the manuscript. JF designed the study and contributed to the interpretation of the critical revision of the manuscript for important intellectual content and approved the final version of the manuscript.

**Funding** This study was supported by grants from the National Natural Science Foundation of China (No 81270441).

**Competing interests** None declared.

**Patient consent for publication** Parental/guardian consent obtained.

**Ethics approval** The retrospective study protocol was approved by the institutional review board of our institution (No 2019CR104). All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki Declaration and its later amendments or comparable ethical standards.

**Provenance and peer review** Not commissioned; externally peer reviewed.

**Data availability statement** Data sharing is not applicable as no data sets were generated and/or analyzed for this study.

**Open access** This is an open access article distributed in accordance with the Creative Commons Attribution Non Commercial (CC BY-NC 4.0) license, which permits others to distribute, remix, adapt, build upon this work non-commercially, and license their derivative works on different terms, provided the original work is properly cited, appropriate credit is given, any changes made indicated, and the use is non-commercial. See: <http://creativecommons.org/licenses/by-nc/4.0/>.

## REFERENCES

- Rajindrajith S, Devanarayana NM. Constipation in children: novel insight into epidemiology, pathophysiology and management. *J Neurogastroenterol Motil* 2011;17:35–47.
- Tabbers MM, Di Lorenzo C, Berger MY, et al. Evaluation and treatment of functional constipation in infants and children: evidence-based recommendations from ESPGHAN and NASPGHAN. *J Pediatr Gastroenterol Nutr* 2014;58:258–74.
- Benninga MA, Faure C, Hyman PE, et al. Childhood functional gastrointestinal disorders: Neonate/Toddler. *Gastroenterology* 2016;150:1443–55.
- Kuizenga-Wessel S, Koppen IJN, Zwager LW, et al. Surgical management of children with intractable functional constipation; experience of a single tertiary children's Hospital. *J Neurogastroenterol Motil* 2017;29:e13005.
- Gordon M, Naidoo K, Akobeng AK, et al. Cochrane review: osmotic and stimulant laxatives for the management of childhood constipation (review). *Evid Based Child Health* 2013;8:57–109.
- Koppen IJN, Di Lorenzo C, Saps M, et al. Childhood constipation: finally something is moving! *Expert Rev Gastroenterol Hepatol* 2016;10:141–55.
- Turan N, Ast TA. The effect of abdominal massage on constipation and quality of life. *Gastroenterol Nurs* 2016;39:48–59.
- Miller MK, Dowd MD, Friesen CA, et al. A randomized trial of enema versus polyethylene glycol 3350 for fecal disimpaction in children presenting to an emergency department. *Pediatr Emerg Care* 2012;28:1–9.
- Zar-Kessler C, Kuo B, Belkind-Gerson J. Botulinum toxin injection for childhood constipation is safe and can be effective regardless of anal sphincter dynamics. *J Pediatr Surg* 2018;53:693–7.
- Bharucha AE, Rao SSC, Shin AS, et al. Surgical interventions and the use of Device-Aided therapy for the treatment of fecal incontinence and Defecatory disorders. *Clin Gastroenterol Hepatol* 2017;15:1844–54.
- Schum TR, Kolb TM, McAuliffe TL, et al. Sequential acquisition of toilet-training skills: a descriptive study of gender and age differences in normal children. *Pediatrics* 2002;109:E48.
- Sun M, Rugolotto S. Assisted infant toilet training in a Western family setting. *J Dev Behav Pediatr* 2004;25:99–101.
- Zickler CF, Richardson V. Achieving continence in children with neurogenic bowel and bladder. *J Pediatr Health Care* 2004;18:276–83.
- Jarzebicka D, Sieczkowska J, Dadalski M, et al. Evaluation of the effectiveness of biofeedback therapy for functional constipation in children. *Turk J Gastroenterol* 2016;27:433–8.
- Rodriguez L, Siddiqui A, Nurko S. Internal anal sphincter relaxation associated with bisacodyl-induced colonic high amplitude propagating contractions in children with constipation: a colo-anal reflex? *J Neurogastroenterol Motil* 2012;24:1023–545.
- Robin SG, Keller C, Zwiener R, et al. Prevalence of pediatric functional gastrointestinal disorders utilizing the Rome IV criteria. *J Pediatr* 2018;195:134–9.
- Rodriguez L, Sood M, Di Lorenzo C, et al. An ANMS-NASPGHAN consensus document on anorectal and colonic manometry in children. *J Neurogastroenterol Motil* 2017;29:e12944.
- Lee TH, Bharucha AE. How to perform and interpret a high-resolution anorectal manometry test. *J Neurogastroenterol Motil* 2016;22:46–59.
- Levitt MA, Dickie B, Peña A. The Hirschsprungs patient who is soiling after what was considered a "successful" pull-through. *Semin Pediatr Surg* 2012;21:344–53.
- El-Shabrawi M, Hanafi HM, Abdelgawad MMAH, et al. High-Resolution anorectal manometry in children with functional constipation: a single-centre experience before and after treatment. *Prz Gastroenterol* 2018;13:305–12.
- Friedmacher F, Puri P. Classification and diagnostic criteria of variants of Hirschsprung's disease. *Pediatr Surg Int* 2013;29:855–72.