

## ORIGINAL ARTICLE

## Gastroenterology

# Transanal irrigation in children with functional constipation: A systematic review and meta-analysis

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## Abstract

**Objectives:** Refractory functional constipation is a challenging condition to manage in children. The use of transanal irrigation (TAI) is well reported in children with neurological disorders as well as anorectal malformations but less so in children with functional disorders of defecation. The objective of our study was to evaluate the effectiveness, safety and outcomes of TAI in children with functional constipation.

**Methods:** PubMed, Scopus and Google Scholar were searched for publications related to the use of TAI in functional constipation. Data regarding the study design, sample size, patient characteristics, investigator-reported response to TAI and adverse effects were extracted from studies that met the selection criteria. The inverse variance heterogeneity model was used for ascertaining the summary effect in this meta-analysis.

**Results:** The search strategy yielded 279 articles of which five studies were included in the final review. The studies were from the United Kingdom ( $n = 2$ ), Netherlands ( $n = 2$ ) and Denmark ( $n = 1$ ). These studies included 192 children with a median age ranging from 7 to 12.2 years old. The TAI systems used in these studies were: Peristeen ( $n = 2$ ), Peristeen or Qufora ( $n = 1$ ), Alterna ( $n = 1$ ) and Navina ( $n = 1$ ). The follow-up duration ranged from 5.5 months to 3 years. Eleven (5.7%) children did not tolerate TAI and withdrew from treatment soon after initiation. The pooled investigator-reported success of TAI was 62% (95% CI: 52%–71%). The most common adverse event was pain which was experienced by 21.7% of children. A total of 27 (14%) were successfully weaned off TAI at the last follow-up.

**Conclusions:** TAI is reported to be successful in 62% of children with refractory functional constipation. There is a need for well-designed prospective trials to evaluate this treatment option in children with refractory functional constipation.

## KEYWORDS

faecal incontinence, refractory constipation

## 1 | INTRODUCTION

Functional constipation (FC) is a common problem in childhood with a reported global pooled prevalence of 9.5%.<sup>1</sup> The conventional treatment of FC includes laxatives (including softeners and stimulants), education and demystification, toilet training, cognitive behavioural

therapy and biofeedback. While a large proportion respond to these measures, nearly one-fourth continue to experience symptoms despite treatment.<sup>2</sup>

In recent times, transanal irrigation (TAI) has emerged as a treatment for children with refractory constipation and faecal incontinence who do not respond to conservative medical treatments.<sup>3</sup> It involves instilling water through the

anus using either a balloon catheter or cone delivery system. This not only cleans out the rectum but the instilled water also appears to stimulate colonic movement. Given it is less invasive than surgical options such as an antegrade colonic enema, some authors recommend that TAI should be considered before any surgical treatment in children with nonresponsive constipation.<sup>3</sup>

TAI was first described in a subset of children with spina bifida and faecal incontinence and has since then become a well-established treatment modality in this subgroup with some studies reporting a success rate as high as 100%.<sup>4,5</sup> It not only improves clinical outcomes but also improves the quality of life. However, the role of TAI in paediatric functional constipation is not clear. We carried out this systematic review to evaluate the role of TAI in children with FC.

## 2 | METHODS

This systematic review and meta-analysis has been performed using the Preferred Reporting Items for Systematic Reviews and Meta-analysis (PRISMA).

### 2.1 | Criteria for considering the studies for review

Inclusion and exclusion of studies were planned using the PECO format—participants, exposure group, control group and outcomes. Participants were children (<19 years) with constipation undergoing TAI. Studies in which adult patients were included and results from children were not reported separately were excluded.

Exposure groups were children with functional constipation. Only the studies in which the data for TAI in FC was given separately were included. Studies in which the outcome of TAI (success or failure) was not clearly defined were excluded.

The outcome studied was the success rate of TAI in the participants. If a study gave the success rate at different time-frames after the initiation of TAI then the success rate at the last follow-up was included in the analysis. If a patient withdrew from treatment because of inability to tolerate TAI then they were deemed as a treatment failure. Only observational studies were considered for this review.

### 2.2 | Search strategy for identification of studies

A systematic literature search was carried out with no time or language restrictions using PubMed, Scopus and Google Scholar. The following strategy was used: ('Transanal Irrigation' OR 'Trans-anal irrigation' OR 'Rectal Irrigation' OR 'Therapeutic Irrigation' [Mesh] OR 'Irrigation')

#### What is Known

- Refractory functional constipation is a challenging condition to manage in children.
- The use of transanal irrigation (TAI) is well reported in children with spine and gut-related neurological disorders as well as anorectal malformations but less so in children with functional disorders of defecation.

#### What is New

- TAI is reported to be successful in 62% of children with refractory functional constipation.

AND ('Constipation' [Mesh] OR constipate\* OR 'defecation' OR 'defaecation' OR 'bowel dysfunction') AND ('Infant, Newborn' [Mesh] OR 'Infant' [Mesh] OR 'Child, Preschool' [Mesh] OR 'Child' [Mesh] OR 'Adolescent' [Mesh] OR child\* OR Pediatric OR Paediatric).

The reference lists of the selected studies and review articles were also reviewed manually to ensure the inclusion of all pertinent articles. The last search for articles was performed in December 2022.

### 2.3 | Study selection and data collection process

Studies selected were imported into the Rayyan QCRI Software for screening by two reviewers independently. After removing duplicate articles, the title and abstract of all other articles were reviewed. Articles thus filtered were assessed for eligibility by reading their full-texts by two reviewers independently. Authors of any study reporting incomplete data or having scope of additional data were contacted for sharing full details for the review via email.

### 2.4 | Data extraction process

Two reviewers independently extracted data using a predetermined data extraction criteria. This included year of publication, authors' names, number of included patients, gender distribution, age, transanal irrigation system used, duration of use, outcomes, that is, treatment success and adverse effects (if any). Data were extracted into a standardised format excel spreadsheet

### 2.5 | Risk of bias in individual studies

Appraisal tool for cross-sectional studies (AXIS) was used for the quality assessment of the individual

studies. The AXIS tool contains a 20-point questionnaire with 'yes', 'no', and 'don't know' answers that address study quality and reporting. The critical areas in the AXIS tool included are study design, sample size justification, target population, sampling frame, sample selection, measurement validity and reliability, overall methods, and conflict of interest and ethical issues.

## 2.6 | Statistical methods

The inverse variance heterogeneity (IVHet) model was used for the meta-analysis to account for heterogeneity. A  $p$  value of less than 0.05 was considered indicative of statistically significant heterogeneity. We performed sensitivity analyses in which we excluded each study individually to determine the effect on the test of heterogeneity and the overall pooled estimates. Poor quality studies were considered for exclusion in sensitivity analysis.

Small study effects which may be due to a publication bias were assessed using Luis Furuya Kanamori (LFK) index. A value of LFK index  $<1$  is indicative of no symmetry, between 1 and 2 indicates minor symmetry and more than 2 is indicative of major asymmetry.

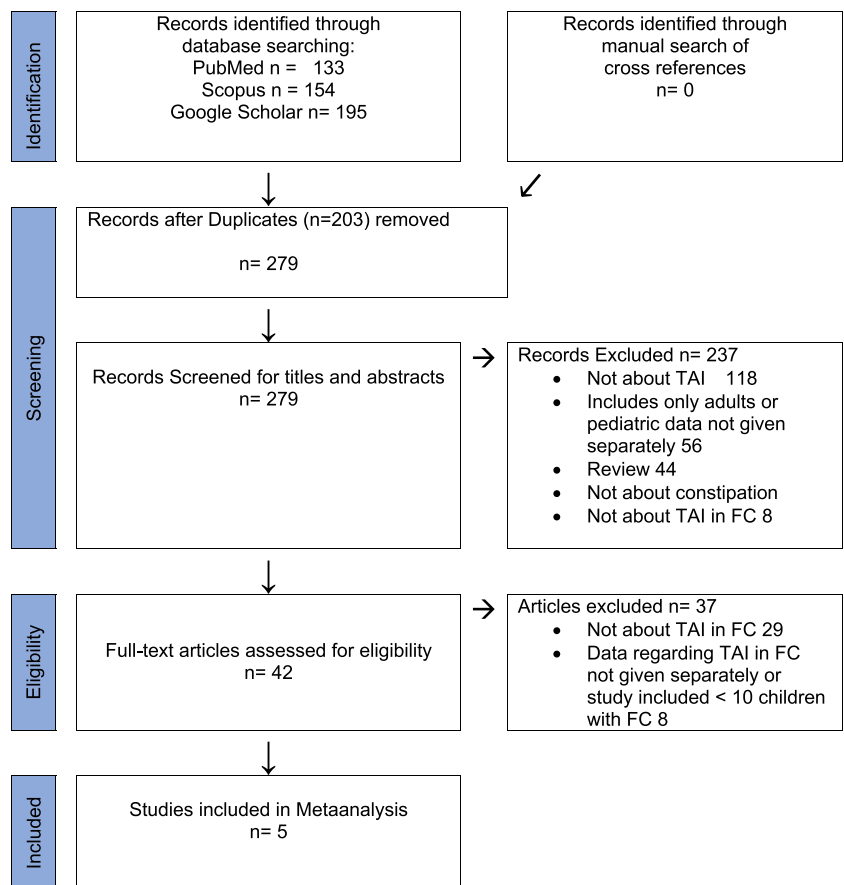
Meta-analysis was performed using MetaXL v5.3 software (EpiGear International).

## 3 | RESULTS

A total of 482 articles were identified on the systematic search (133 PubMed, 154 Scopus and 195 Google Scholar). After the removal of 203 duplicates, the title and abstract of 279 articles were screened (Figure 1). After screening, 274 articles were removed, and the remaining five were included in the final analysis.<sup>6–10</sup>

Five studies ( $n=192$ ) detailing the utility of transanal irrigation in FC were included. The studies were from the United Kingdom ( $n=2$ ), the Netherlands ( $n=2$ ) and Denmark ( $n=1$ ). Two studies were conducted prospectively, while the remaining were either retrospective reviews or cross-sectional surveys.

These studies included 192 children with a median/mean age ranging from 7 to 12.2 years old. The TAI systems used in these studies were as follows: Peristeen ( $n=2$ ), Peristeen or Qufora ( $n=1$ ), Alterna ( $n=1$ ) and Navina ( $n=1$ ). All these TAI devices are approved for use in children with the minimum age dictated by the regulatory authorities in each region. As



**FIGURE 1** PRISMA flow diagram depicting the flow of information through different phases of the systematic review. FC, functional constipation; PRISMA, Preferred Reporting Items for Systematic Reviews and Meta-analysis; TAI, transanal irrigation.

a guide, this is usually 3 years of age in Europe and 2 years in the United States.

The follow-up duration ranged from 5.5 months to 3 years.

Eleven (5.7%) children did not tolerate TAI and withdrew from treatment soon after initiation. Nine of these patients were from a single study (Ng et al.).<sup>8</sup> In this study, 9/26 children stopped TAI within a month of commencement.

The pooled investigator-reported success of TAI was 62% (95% confidence interval [CI]: 52%–71%,  $I^2 = 31%$ ) (Figure 2). There were considerable differences in the definition of treatment success used by the individual studies (Table 1).

On sensitivity analysis, the exclusion of the study by Koppen et al. reduced the heterogeneity to 16% (Supporting Information S1: Table 1). The resulting pooled success rate was 65.5% (95% CI: 55.4%–74.9%).

There was no asymmetry for small study effects (LFK index = 0.81, Figure 3).

Details of concomitant oral laxatives were reported in three studies. A large majority 107/132 (81%) were on concomitant oral laxatives (Table 2). One study (Koppen et al.) also reported the addition of laxatives (mainly bisacodyl) to the irrigation fluid.<sup>10</sup>

The most common adverse event was pain which was experienced by 36/166 (21.7%) children. One study did not report the adverse events for children with functional constipation separately. A total of 27 (14%) were successfully weaned off TAI at the last follow-up (Table 2).

### 3.1 | Risk of bias assessment

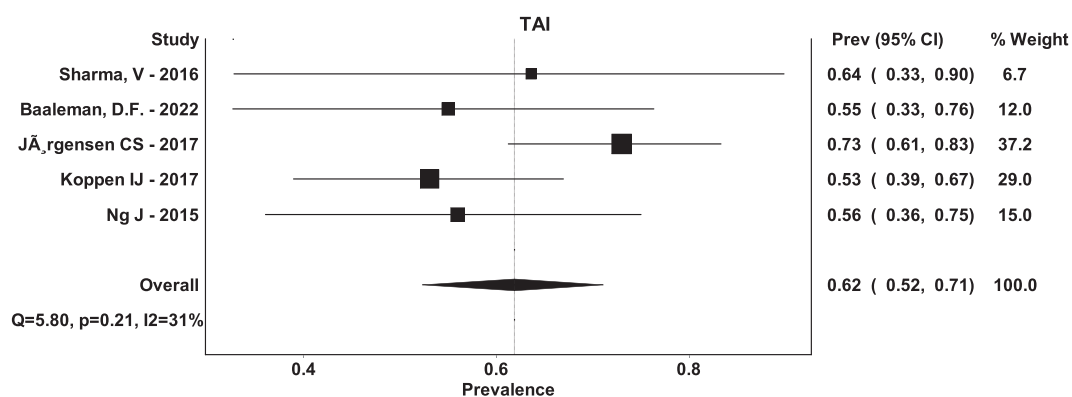
The risk of bias assessment of the studies included in the meta-analysis has been summarised in Supporting Information S1: Table 2. No studies were excluded because of poor quality.

## 4 | DISCUSSION

This review summarises the findings of five studies that evaluate the utility of TAI in children with refractory functional constipation. In these studies, TAI was reported to be successful in nearly 62% of children which makes it a promising modality in this subgroup of patients. TAI is simple and reversible and was well-accepted by the majority of patients in which it is instituted.

It is important to choose the right patients for TAI. Age is likely an important determinant for the acceptance of TAI. Younger children may not fully understand the rationale of the treatment and be understandably anxious about a rectal approach. In the study by Ng et al. in which the median age of the included children was 7 years (compared to 9.2–12.2 years in the others) a large proportion stopped treatment soon after commencement.<sup>8</sup> In that study a younger age was found to be a predictive factor for treatment withdrawal. This should be a consideration when offering TAI as an option for constipation management. In selected cases, the input of a psychologist or occupational therapist who is familiar with dealing with younger children may help in allaying any fears or anxiety around the proposed treatment and increase acceptance and adherence.

A number of different commercial TAI devices are available. Even though their basic principle is the same, they differ in their type of rectal catheter (balloon or cone tip), method of water instillation (by gravity or by manual or electronic pumps) and volume of irrigant used (low or high volume). Four different devices were used in the five studies included in this review. We did not attempt to compare the success rate between different devices because of the small number of patients using each device and differences in the definition of a successful outcome used in each study. In all probability, all devices have a similar efficacy and



**FIGURE 2** Investigator reported response to TAI in children with functional constipation. CI, confidence interval; TAI, transanal irrigation.

TABLE 1 Characteristics of studies included in this meta-analysis.

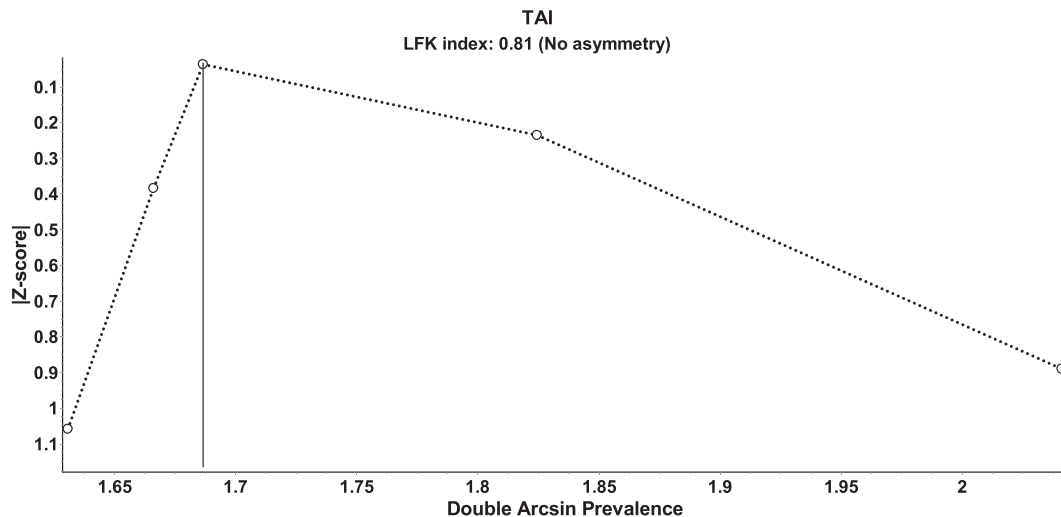
| Reference                     | Study design | Study years | n   | Symptoms   | Age range (median or mean)        | System used         | Definition of success used   | Follow-up method (if available) and duration  |
|-------------------------------|--------------|-------------|---|--|-----------------------------------|---------------------|--|---|
| Sharma et al. <sup>6</sup>    | P            | 2014        | 11<br>(two did not tolerate and withdrew)               | All refractory constipation and FI   | NA (12.2) years                   | Peristeen           | Improvement in soiling   | —   |
| Baaleman et al. <sup>7</sup>  | R            | —           | 26  | FI 68%, large stools 60%, hard stools 53%, withholding behaviour 44%, abdominal pain 53% | 8.3–12.8 (11) years               | Navina              | Rome IV criteria   | Median: 3 years; 20 children continued to be on TAI at the last follow-up   |
| Ng et al. <sup>8</sup>        | P            | 2008–2014   | 25 <sup>a</sup><br>(nine did not tolerate and withdrew) | All refractory constipation (with or without FI)   | 3–16 years (7) years <sup>b</sup> | Peristeen<br>Qufora | total continence or occasional soiling                                   | Median: 14 (3–78) months <sup>c</sup>   |
| Jargensen et al. <sup>9</sup> | R            | 2010–2016   | 63  | All refractory constipation (with or without FI)   | NA (9.2 ± 2.3) years              | Alterna             | Rome III criteria/<br>International Children's Continence Society (ICCS) | Telephone by a specialist nurse starting 3–4 weeks.<br>Mean 5.5 ± 8.8 months  |
| Koppen et al. <sup>10</sup>   | R            | 2014        | 67  | All refractory constipation (with or without FI)<br>FI—84%                               | 3–18 (10.3) years                 | Peristeen           | FI resolved or <1 episode per week                                       | Outpatient clinic visits until able to use device, then follow-up by nurse by telephone contact.<br>Median 11 months (range 1 month to 3 years) |

Abbreviations: FC, functional constipation; FI, faecal incontinence; NA, not applicable; P, prospective; R, retrospective; TAI, transanal irrigation.

<sup>a</sup>Twenty-six children were initially included but one was lost to follow-up and treatment success could not be evaluated.

<sup>b</sup>Age range (median) for all children included in the study. Includes children who received TAI for indications apart from FC.

<sup>c</sup>Includes follow-up duration of children who received TAI for indications apart from FC.



**FIGURE 3** DOI plots to estimate small study effects. LFK, Luis Furuya Kanamori; TAI, transanal irrigation.

patients/carers can choose a device of their choice based on their preference, availability, portability and cost.

Tap water is the most common irrigant used for TAI.<sup>3</sup> As most of these children have dilated colons and often need a large volume to be instilled there is a theoretical risk of iatrogenic hyponatremia. However, none of the included studies reported such a side effect indicating that using tap water is safe. In the study by Koppen et al. additives (bisacodyl, polyethylene glycol, glycerine and laxative enemas) were added to the irrigating fluid in 20% of children.<sup>10</sup> There is limited literature on the use of additives with TAI but should likely be a consideration in children with a suboptimal response to tap water TAI, since a similar strategy has also been successfully used in those undergoing antegrade colonic enemas.<sup>11,12</sup>

One of the goals of TAI is to wean off the oral laxatives. As most of the patients in whom TAI is started would have been on oral laxatives for a long time, most centres continue oral laxatives till TAI is successfully established and then attempt gradual withdrawal. It was interesting to note in the studies included in this review that a large proportion (~80%) of children continued on oral laxatives long after the initiation of TAI. Unfortunately, these studies have not discussed dosages and it would not be unreasonable to assume that in a large proportion, the dosages were being weaned down and were much lower than the dose pre-TAI.

Most centres initiate TAI on a daily basis and then gradually reduce the frequency once a successful result is achieved with the ultimate aim to do away with it completely in due course of time. In the studies included in this review, 14% were weaned off TAI (and all other treatments) completely at the last follow-up.

This suggests that TAI can not only optimise bowel management but can also provide a complete cure in those with refractory FC.

TAI was found to be safe. No serious adverse effects were reported in any study. Pain during irrigation was the most commonly reported side effect and was experienced by nearly one-fifth of all patients. However, it was generally mild and did not lead to discontinuation of TAI in most instances.

One of the drawbacks of TAI is the need for ongoing assistance with the procedure. In the study by Baaleman et al. that discussed independent usage in FC only two children (out of 26) were able to perform TAI independently.<sup>7</sup> It is important that this aspect is discussed with families before starting TAI.

The success of TAI depends on using a tailored approach to an individual patient, proper training and careful follow-up. Optimising the volume of the irrigant, number of puffs of balloon (in those with a balloon catheter) and ongoing supervision can improve treatment adherence and success.<sup>13,14</sup> Caruso et al. evaluated a cohort of 70 children on TAI and found that nearly 63% of children needed adjustment to their initial TAI regime to improve its effectiveness.<sup>13</sup> The importance of close follow-up was exemplified in the study by Jorgensen et al. in which regular follow-up by a specialist nurse (on phone and in-person) helped achieve success in nearly three-fourth of children.<sup>9</sup>

The strength of this meta-analysis is the first such study that focuses only on children with functional constipation. FC is by far the commonest cause of constipation in children and our review suggests that TAI should be considered in those who do not respond to conventional measures.



**TABLE 2** Details of TAI in the individual studies.

| Reference                     | Volume and duration of irrigation                                  | Frequency   | Additional treatment  | Success evaluation and rate   | GoL/treatment satisfaction assessment   | Adverse effects   | TAI successfully stopped |
|-------------------------------|--|---|---|---|---|---|--------------------------|
| Sharma et al. <sup>6</sup>    | —  | —   | —   | 64%   | —   | None  | 1                        |
| Baaleman et al. <sup>7</sup>  | —  | Median (IQR): 7 (1.5–7) per week.   | Oral laxatives, 72%; pharmacological enemas, 11%                        | Questionnaire electronically, 55%   | HRQoL (PedsQL and PedsQL-GI symptom scales) scores were high compared with other studies in similar patient populations | Abdominal pain ( <i>n</i> = 6), pain during catheter insertion ( <i>n</i> = 2) <sup>a</sup> | 4                        |
| Ng et al. <sup>8</sup>        | —  | —   | —   | Prospectively maintained database questionnaire for family, 73%   | Significant improvement in both Rintala score and QoL (PedsQL)  | Not given <sup>b</sup>  | None                     |
| Jargensen et al. <sup>9</sup> | 20 mL/kg, tap water. Procedure time was approximately 35–45 min.   | Initially three times weekly, subsequently titrated. 47 (65%) used TAI 3–4 times weekly, 25 (35%) daily irrigation session. Mean: 4.2 ± 1.9 per week. | Oral laxatives, 79%   | Reviewing patient files and based on the children and their parents' reporting during the outpatient consultations, 53% | —   | None  | 18                       |
| Koppen et al. <sup>10</sup>   | 10–20 mL/kg. Duration of irrigation: <30 min, 43%; 30–60 min, 52%. | 22 (33%) daily, 15 (25%) alternate day. Every third day, 6%. Rest other/varying frequency.  | Additional medications (oral laxatives and/or enema/suppositories), 52% | Cross-sectional questionnaire (25 questions) sent out to the parents by mail, 56%                                       | 72% of parents reported TAI improved management compared to previous treatment  | Pain during irrigation ( <i>n</i> = 28)   | 4                        |

Abbreviations: FC, functional constipation; GI, gastrointestinal; HRQoL, health-related quality of life; IQR, interquartile range; PedsQL, Paediatric Quality of Life Inventory; QoL, quality of life; TAI, transanal irrigation.

<sup>a</sup>Events that occurred often or always. Other adverse events were—fluid leak during infusion (*n* = 2), catheter did not remain in place (*n* = 1) and bursting of balloon (*n* = 1).

<sup>b</sup>In this study, the adverse events for FC children were not given separately. Overall, 50% of respondents (functional + organic) experienced abdominal pain, bloating, nausea and pain on insertion.

## 5 | CONCLUSION

This study, however, has a number of limitations. Overall, only a small number of studies could be included in this analysis. The studies were not of robust methodological quality and used different definitions of treatment success. The use of different TAI systems in these studies also makes comparability difficult. Some data could not be obtained despite attempts to contact the authors. These points underline the fact that the evidence for the use of TAI in children with FC is weak. There is a need for well-designed prospective trials to evaluate this treatment option in children with refractory functional constipation.

### ACKNOWLEDGEMENTS

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### CONFLICT OF INTEREST STATEMENT

The authors declare no conflict of interest.

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### SUPPORTING INFORMATION

Additional supporting information can be found online in the Supporting Information section at the end of this article.

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