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ORIGINAL ARTICLE

Transanal irrigation is effective in functional fecal incontinence

Cecilie Siggaard Jørgensen[1] · **Konstantinos Kamperis**[1] · **Line Modin**[2] · **Charlotte Siggaard Rittig**[1] · **Søren Rittig**[1]

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Abstract Functional fecal incontinence (FFI) is divided into cases related to functional constipation (FC) and cases without concomitant constipation termed functional non-retentive fecal incontinence (FNRFI). Transanal irrigation (TAI) is widely used in children with neurogenic fecal incontinence but is less studied in children with functional defecation disorders. The aim was to evaluate the feasibility and efficacy of TAI in the treatment of FFI. A retrospective study in 72 children (mean age 9.2 ± 2.2 years, 47 males) with treatment-resistant FFI was performed. All children accepted treatment and 35% (n = 25) were titrated to daily sessions. Of the 63 children who fulfilled the Rome III criteria of constipation, 46 (73%) showed full response with complete remission of incontinence episodes. Eleven (17%) showed partial response (≥50% reduction). Of nine children with FNRFI, four (44%) showed full response whereas two (22%) showed partial response. We found no significant difference in the reduction of incontinence episodes between the children with FC (87%) and children with FNRFI (68%) (p = 0.11).

Conclusion: TAI is an effective, well-tolerated, and safe choice in children with FC. No clinical parameters seemed to predict response to treatment. The number of children with FNRFI was low, but TAI seemed effective in this group of children.

What is Known:
- *Functional fecal incontinence (FFI) is a frequent, chronic condition with significant impact on children's quality of life.*
- *Transanal irrigation (TAI) is used in children with neurogenic bowel dysfunction but less studied in children with functional defecation disorders.*

What is New:
- *TAI seems an effective, well-tolerated, and safe choice in children with FFI due to functional constipation.*
- *Albeit the number of children with functional non-retentive fecal incontinence was low in our study, TAI seems effective also in this group of children.*

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✉ Cecilie Siggaard Jørgensen
cecilie.siggaard@clin.au.dk

Konstantinos Kamperis
kostas.kamperis@clin.au.dk

Line Modin
line.modin@rsyd.dk

Charlotte Siggaard Rittig
charritt@rm.dk

Søren Rittig
rittig@clin.au.dk

[1] Center for Child Incontinence, Department of Paediatrics, Aarhus University Hospital, Palle Juul-Jensens Boulevard 99, 8200 Aarhus N, Denmark

[2] HC Andersen Children Hospital, Odense University Hospital, Odense, Denmark

Keywords Functional fecal incontinence · Functional constipation · Functional non-retentive fecal incontinence · Transanal irrigation · Children

Abbreviations

ACE	Antegrade continence enema
ICCS	The International Children's Continence Society
FC	Functional constipation
FNRFI	Functional non-retentive fecal incontinence
FFI	Functional fecal incontinence
TAI	Transanal irrigation

Introduction

Functional fecal incontinence (FFI) is traditionally divided into cases related to functional constipation (FC) due to fecal retention with subsequent stool overflow and cases without concomitant constipation called functional non-retentive fecal incontinence (FNRFI). FFI is a more common among boys than girls [2]. The estimated prevalence of FC in the pediatric population is 0.7–29.6% [9], out of whom 75–90% are reported to present with FFI [18]. However, of all children with FFI, only 10–30% are reported to have FNRFI [16]. FFI has a significant negative impact on the child's quality of life regarding both physical and psychological aspects [9]. Standard treatment is primarily conservative including education, regular toilet visits, correct seating posture during defecation, adequate fluid intake, and secondly oral and/or rectal laxatives if needed [19]. Approximately 50 % of all children with FC respond to standard treatment within the first 6–12 months leaving a significant proportion of non-responders [14]. Some treatment refractory children require surgical procedures such as antegrade continence enema (ACE) stoma formation, which is effective in children with FC [6]. However, this modality is associated with complications in more than 50% including leakage, infection, and development of strictures [13].

Transanal irrigation (TAI) was introduced by Shandling and Gilmour in 1987 to treat FI in children with neurogenic bowel disease and is today widely used in this group of children [17]. The treatment is a non-surgical alternative which empties the bowel from approximately the rectal ampulla to the splenic flexure [4], thereby preventing fecal leakage between treatments. The idea is to keep the colon empty for longer periods of time to regain its propulsive ability and re-establishing control over defecation. A recent review evaluating the use of TAI in children recommends considering the use of TAI in all children before surgical intervention [8]. We have prospectively offered TAI as an ambulatory and non-surgical alternative in children with treatment refractory FFI (FC and FNRFI) since August 2010. The aim of this study was to evaluate the feasibility and efficacy of TAI in the treatment of children with FFI.

Materials and methods

Patients and study design

A retrospective follow-up study was performed by reviewing patient files for all consecutive children with FFI who were treated with TAI at the Center for Child Incontinence, Aarhus University Hospital, Denmark, a tertiary referral center for childhood incontinence, between August 2010 and January 2016. Exclusion criteria were confirmed neurogenic bowel disease, Hirschsprung's disease, anorectal malformations, and use of medication known to cause constipation. All children were characterized as having FC or FNRFI according to the Rome III criteria of FC [15]. Transverse rectal diameter was measured with transabdominal ultrasound for all children at the first outpatient visit [5]. The project was preformed according to the regulations of The Central Denmark Region Committees on Health Research Ethics.

Colonic irrigation system

The irrigation system consisted of a cone-shaped colostomy tip, which runs into the rectum (Alterna®, Coloplast A/S, Humlebaek, Denmark) and a bag system. The initial regimen consisted of using the irrigation system three times weekly, and the frequency was subsequently titrated according to response. Our specialist nurses instructed the patient and their parents. The amount of water used was 20 ml/kg b.w. of tap water with a temperature of approximately 37 °C. The water drained into the intestine over 5–10 min, after which the tip was removed. The child was asked to sit at the toilet for 15–30 min to open the bowel for water and stools. The total procedure time was approximately 35–45 min.

As follow-up, all parents were contacted by telephone by a specialist nurse 3 to 4 weeks after start of TAI. If the child or the family experienced difficulties, they were offered additional visits for training and supervision. If the child remained fecal continent over a period of at least 6 months, the family was instructed to reduce the frequency of treatments until no treatment was further needed. TAI was combined with conventional treatment including oral laxatives if necessary.

Evaluation of efficacy

The primary outcome variable was number of fecal incontinence episodes per week. This was evaluated by reviewing patient files and based on the children and their parents' reporting during the outpatient consultations. Children were subsequently characterized according to The International Children's Continence Society (ICCS). Full response was defined as complete remission of incontinence episodes with or without the use of TAI and partial response as at least a 50% reduction of incontinence episodes. [1].

Statistics

Data is presented as mean ± SD or proportions unless otherwise stated. Student's t test (based on the actual distribution of the data) and chi-squared tests were used to evaluate differences between children with FC and FNRFI and response to treatment. Correlation was tested with the Pearson's correlation coefficient. p values of <0.05 were considered significant. Data analysis was performed with SPSS version 23 (IBM Corp).

Results

Patient characteristics

Overall, 72 children with treatment-resistant FFI (defined as no response to standard treatment) who underwent TAI during the study period were identified. Forty-seven children (65%) were males, and mean age at commence of treatment was 9.2 ± 2.2 years. The median of baseline fecal incontinence episodes before the treatment start was 12.7 ± 12.3 per week. All children had previously been unsuccessfully treated with Macrogol 3350, 60 children (83%) had been treated with bisacodyl or docusate/glycerol, six children (8%) with lactulose, 28 children (39%) with sodium picosulfate, and 21 children (29%) had been subjected to oil enemas. We identified 19 children (26%) with psychological comorbidities (ADD, ADHD, autism, externalizing behavioral disorder, ticks, and depression). Two children (2.8%) were previously diagnosed with celiac disease and one child (1.4%) with lactose intolerance.

Compliance and adherence to the treatment

No children stopped treatment due to discomfort. Forty-seven children (65%) used TAI three to four times weekly, while 25 children (35%) needed daily irrigation session. Fifty-seven children (79%) combined TAI with oral laxatives use. Mean follow-up time while on treatment was 14.3 ± 11.8 months.

Response to treatment in children with FC

Sixty-three children (88%) fulfilled the Rome III criteria for constipation. In total, 46 children (73%) showed full response with complete remission of incontinence episodes, whereas 11 children (17%) showed partial response. Only six children (10%) experienced no improvement (<50% reduction of FFI episodes) to TAI. Mean treatment duration before obtaining full response was 5.5 ± 8.8 months. Figure 2 show the time to achieve continence for the responders. Of the children with full response, 18 (39%) were able to stop treatment and experienced no relapse until the last follow-up visit. No correlation was found between the number of Rome III criteria and response (reduction of incontinence episodes). We found no difference in age, gender, presence of psychological comorbidity, number of incontinence episodes per week before treatment during follow-up, or number of treatments per week between responders and non-responders to TAI (Table 1).

Children with FC vs. FNRFI

Nine children only met one criterion of the Rome III criteria for constipation (fecal incontinence) and were characterized as having FNRFI. Transverse rectal diameter was larger in children with FC than in children with FNRFI (4.0 ± 1.1 vs. 2.8 ± 0.5 cm, $p < 0.01$). We found no significant differences between children with FC and FNRFI regarding age, sex, or pretreatment fecal incontinence episodes per week (Table 2). However, 5 (56%) of children with FNRFI were diagnosed with psychological comorbidities (ADD and externalizing behavioral disorder), which were significantly higher than in the group of children with FC (14 (22%), $p = 0.03$). Furthermore, we found no difference in follow-up time, treatments per week or time to response between the groups (Table 2). We found no significant difference in reduction of incontinence episodes between the children with FC (87%) and children with FNRFI (68%) ($p = 0.11$). However, we found a higher response rate in children with FC with 57 children (90%)

Table 1 Patient characteristics and treatment parameters for children with functional constipation (FC): full and partial responders vs. non-responders

	Responder ($n = 57$)	Non-responders ($n = 6$)	p value
Age when starting irrigation (years)	9.2 ± 2.4	9.0 ± 2.1	0.83
Gender (male, *n*)	37 (65%)	3 (50%)	0.47
Psychological comorbidity (*n*)	13 (23%)	1 (17%)	0.73
Fecal incontinence episodes per week before treatment	13.4 ± 13.3	8.2 ± 2.9	0.34
Follow-up (months)	14.1 ± 11.6	10.2 ± 7.3	0.46
Treatments per week	4.3 ± 1.9	4.1 ± 1.6	0.83

Data are presented as the mean ± SD or percentage

n number of patients

*$p < 0.05$

Table 2 Patient characteristics and treatment parameters: children with functional constipation (FC) vs. children with functional non-retentive fecal incontinence (FNRFI)

	FC ($n = 63$)	FNRFI ($n = 9$)	p value
Age when starting irrigation (years)	9.2 ± 2.3	9.1 ± 1.8	0.92
Gender (male, *n*)	40 (63%)	7 (78%)	0.40
Psychological comorbidity (*n*)	14 (22%)	5 (56%)	0.03*
Fecal incontinence episodes per week before treatment	12.9 ± 12.8	11.3 ± 8.5	0.72
Rectal diameter (cm)	4.0 ± 1.1	2.8 ± 0.5	0.006**
Follow-up (months)	13.8 ± 11.3	17.6 ± 15.2	0.38
Treatments per week	4.2 ± 1.9	5.3 ± 2.0	0.11
Time to response (months)	5.5 ± 8.8	6.5 ± 6.2	0.83
Fecal incontinence episodes per week after treatment	1.4 ± 3.2	3.6 ± 5.2	0.09
Responders (*n*)	57 (90%)	6 (67%)	0.04*
(Full)	46 (73%)	4 (44%)	

Data presented as mean ± SD or percentages; n, number of patients

*$p < 0.05$; **$p < 0.01$

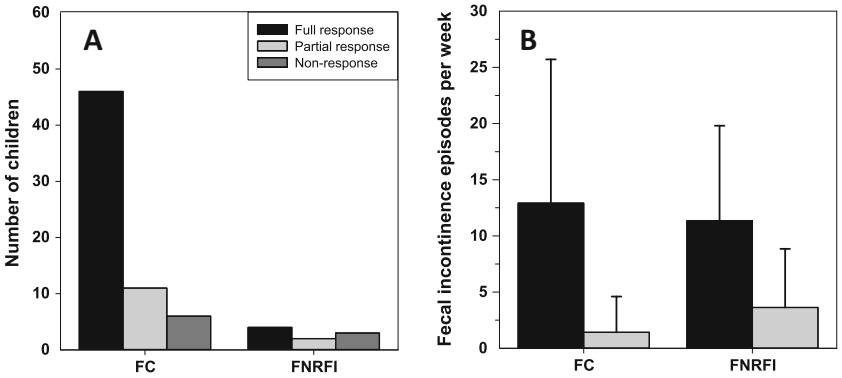

Fig. 1 Response to treatment when comparing children with FFI due to functional constipation (FC) and functional non-retentive fecal incontinence (FNRFI) regarding (a) number of responders (full response was defined as completely remission of incontinence episodes and partial response as a 50–99% reduction), $p = 0.04$, and (b) fecal incontinence episodes per week before (*black*) and after (*gray*) use of irrigation, NS

showing full or partial response compared with 6 children (67%) with FNRFI ($p = 0.04$) (Fig. 1).

Discussion

The present study showed a high response rate of TAI in children with FC (90%) and FNRFI (67%) who failed conventional first-line treatment. Furthermore, TAI showed a prompt effect, since approximately 50% of the children with FC showed response within the first 2 months (Fig. 2). This is, to our knowledge, the largest clinical study of the effect of TAI in children with FFI, and it is the first study evaluating the efficacy in children with FNRFI.

TAI is an established and accepted treatment for children with neurogenic bowel dysfunction. A recent meta-analysis of 16 studies on long-term management of neurogenic FI supported that TAI is an effective treatment option in children and reported that TAI improved quality of life in both children and their families [3]. Only few studies have been conducted to evaluate TAI in FFI. Nasher et al. conducted a small retrospective study of ten children (seven with chronic idiopathic constipation) treated with TAI [11] and found the treatment to be very successful since all children had an improvement in fecal continence score, episodes of soiling, and quality of life score. Ng et al. performed a prospectively study with 42 children (26 children with idiopathic constipation) using TAI [12]. Of the children who succeeded treatment, 84% responded and a significant improvement in quality of life was evident. A recent study performed by Koppen et al. assessed treatment efficacy of TAI in children with intractable FC ($n = 49$) by evaluating questionnaires. The study reported full response or less than one fecal incontinence episode per week in 53%, and most parents (86%) reported satisfaction with the result of the treatment [7]. Koppen et al. added laxatives (mainly bisacodyl) to the irrigation fluid in some children to improve response to treatment [7]. It could be indicated to perform further studies evaluating a possible synergistic effect of laxatives in the fluid, since this has also been reported in patients using ACE [10]. However, some concerns about electrolyte disturbances when adding laxatives to the irrigation water have been raised [8] and should be addressed before using it as standard procedure.

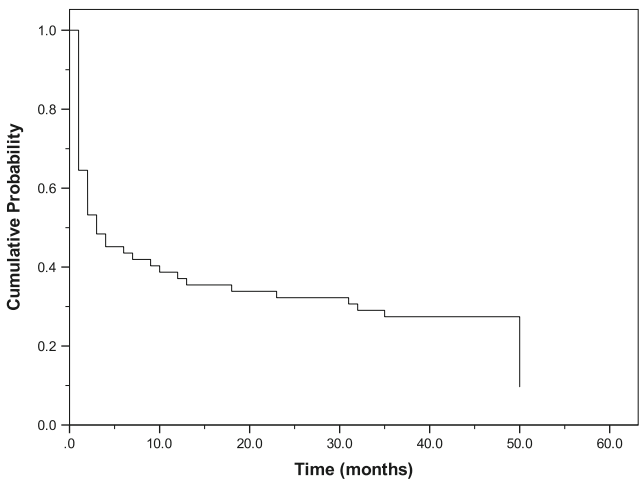

Fig. 2 Kaplan-Meier plot of response over time (full response) for children with functional constipation. Approximately 50% of the population shows response within the first 2 months. Responses rates continue to improve during several years after commence of irrigation

Today, two different systems are available for TAI. All three earlier studies in FFI have evaluated systems where a rectal catheter with a balloon attached is inserted and inflated in the rectum to secure the position of the catheter, after which irrigation fluid is infused. Our treatment consisted of a cone-shaped colostomy tip, where no balloon is attached to the catheter. The earlier studies investigating TAI in FFI all reported that some children experienced pain during treatment, a complaint also reported by children with neurogenic bowel dysfunction using TAI [8]. Koppen et al. found that 42% suffered pain during the procedure [7]. Although not specifically asked about pain, all children in our study tolerated the treatment. One could speculate if there is difference in tolerability

in the two systems. No serious complications were reported during follow-up, which is in accordance with previous studies [7, 11, 12].

Nasher et al. and Ng et al. both reported that higher age was associated with better acceptance of treatment and improved response [11, 12]. We found the system to be well tolerated by all ages maybe due to an irrigation system without an inflated balloon causing discomfort. However, it remains to be clarified whether TAI can or should be introduced in younger age groups.

Although not systematically examined, psychological comorbidity was diagnosed in 26% of all children and 56% in the group of children with FNRFI. In a recent study investigating 312 children with FFI, the frequency of behavioral comorbidity in children with FC was higher (49%) than the present study but comparable (59%) among children with FNRFI [20]. We found a significantly higher rate of children with psychological comorbidities among children with FNRFI than children with FC (p = 0.03), which, despite the small group of children, could indicate psychological comorbidity as a predisposing factor for FNRFI. There was no correlation between psychological comorbidity and response to treatment showing that children with psychological comorbidity were also well treated with TAI.

The limitation of the present study is the retrospective and un-controlled design as well as the fact that 79% of the children used combination of TAI and oral laxatives, which could have influenced our results. However, all children were refractory to laxatives before TAI started. In addition, the response to treatment was evaluated though reviewing patient files, and based on self-reporting, and the follow-up period was relatively short. A randomized, controlled trial with a longer follow-up period comparing TAI with other non-surgical procedures where the response is evaluated with a bowel diary and fecal incontinence scoring system should be performed. Also, a larger cohort of children with FNRFI should be included to evaluate the potential use for this group of children.

In conclusion, TAI seems an effective, well-tolerated, and safe choice in children with FFI due to FC who are refractory to first-line therapy. No clinical parameters seem to predict response to treatment. Albeit the number of children with FNRFI was low in our study, TAI seems effective also in this group of children.

Authors' Contributions Cecilie Siggaard Jørgensen—contribution: study design, data collection, data analysis, and writing the first draft. Konstantinos Kamperis—contribution: study design, data analysis, and critical revision of manuscript. Line Modin—contribution: study design, and critical revision of manuscript. Charlotte Siggaard Rittig—contribution: study design, and critical revision of manuscript. Søren Rittig—contribution: study design, data analysis, and critical revision of manuscript.

Compliance with ethical standards The project was preformed according to the regulations of The Central Denmark Region Committees on Health Research Ethics. For this type of study, formal consent is not required.

Conflict of interest The authors declare that they have no conflict of interest.

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