Functional defecation disorders in children
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Chapter eight

TRANSANAL IRRIGATION IN THE TREATMENT OF CHILDREN WITH INTRACTABLE FUNCTIONAL CONSTIPATION

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ABSTRACT

Objectives
to assess the treatment efficacy of transanal irrigation and parental satisfaction in children with intractable functional constipation (FC) treated with Peristeen®.

Methods
Cross-sectional survey study among parents of children (age 0-18 years) treated with Peristeen® for FC (based on the Rome III criteria). Anonymous questionnaires were sent out to parents via mail, these consisted of 25 self-developed, multiple-choice questions regarding the use of Peristeen®, current gastrointestinal symptoms, adverse effects of Peristeen®, concomitant medication use and parental satisfaction.

Results
Out of 91 invited families, 67 (74%) returned the questionnaire. In total, 84% of patients suffered from fecal incontinence prior to treatment. Out of all children who still used Peristeen® at the time of survey (n=49), fecal incontinence had resolved completely in 41%, 12% experienced occasional episodes of fecal incontinence (<1 episode per week) and the remaining 47% still suffered from episodes of fecal incontinence regularly (≥1 time per week). A total of 28 children (42%) experienced pain during rectal irrigation, especially during insertion of the catheter, inflating the balloon or during irrigation. Overall, 86% of the parents were satisfied with the result of transanal irrigation and 67% reported that they would continue using transanal irrigation for the treatment of their child’s symptoms.

Conclusion
Transanal irrigation may be effective in the treatment of children with FC and renders a high parental satisfaction. Future prospective studies, preferably RCTs, are necessary to further evaluate this treatment option.
INTRODUCTION

Intractable constipation in children is a frustrating and difficult problem for affected children, their families and medical caregivers involved. Finding the best tailored therapeutic approach for these children can be challenging, especially after various conventional pharmacological options have been exhausted. Transanal irrigation with specifically designed equipment is a treatment modality that is not widely used yet, but that is potentially beneficial for a subset of these children. Unfortunately, published data on this treatment modality in children are scarce. In this survey study, the use of transanal irrigation is evaluated in children with intractable functional constipation (FC) in a single medical center.

Pediatric constipation is a commonly encountered problem in primary, secondary and tertiary pediatric healthcare and accounts for substantial healthcare costs (1,2). The vast majority of children with constipation suffers from FC as defined by the Rome III criteria (3,4). The prevalence of FC ranges from 0.7% to 29.6% (2). Organic causes of constipation are rare and include anorectal malformations, spina bifida and Hirschsprung’s disease (5).

FC symptoms such as fecal incontinence and painful defecation are bothersome, may impede children in their daily activities and have a significant impact on the health-related quality of life (6,7). Conventional treatment of FC in children comprises of education, a toilet training program, and pharmacological treatment with oral and/or rectal laxatives (8). Approximately 50% of children with FC treated with conventional therapy will recover and be taken off laxatives within 6-12 months (9). However, even after five years of intensive treatment, approximately 50% of children referred to a pediatric gastroenterologist still suffer from infrequent painful defecation and fecal incontinence (3). FC unresponsive to optimal conventional treatment for at least 3 months is referred to as intractable FC (3). In children with intractable FC, surgery is sometimes required. However, surgery always carries the risk of complications and does not provide a guarantee of success. Transanal irrigation is a non-surgical treatment modality and, if treatment is successful, transanal irrigation could prevent children from having to undergo surgery.

Peristeen® Anal Irrigation System is a relatively novel rectal irrigation system that can be used at home in children with constipation and fecal incontinence (figure 1). After inserting the rectal catheter, a balloon attached to the catheter is inflated with air to secure the position of the catheter in the rectum. Then, after infusion of the irrigation fluids into the colon, the balloon is deflated and both stools and irrigation water are evacuated from the rectum. By irrigating
Figure 1
Peristeen® Anal Irrigation System, figure provided by Coloplast. The system consists of a control unit with a pump, a water bag, and a rectal catheter through which water can be infused into the colon.

the colon, accumulation of large quantities of stools is prevented which consequently results in a decrease in the number of fecal incontinence episodes, thereby improving the quality of life in these children. Transanal irrigations are usually performed with a volume of 10-20 ml/kg of water and the frequency of irrigations depends on the patient’s response (10,11). Transanal irrigation has been shown to be effective in the management of fecal incontinence in children with spina bifida, Hirschsprung’s disease and defecation disorders due to anorectal malformations (11–16). However, data on the effectiveness of transanal irrigation in children with FC are scarce (10,17). Therefore, the aim of this survey study was to explore the treatment efficacy and parental satisfaction in children with FC who are treated with Peristeen®.

METHODS

Patients and materials
This cross-sectional survey study was performed in the Emma Children’s Hospital/Academic Medical Center (Amsterdam, the Netherlands), a tertiary referral center for pediatric defecation disorders. Parents of children who were treated with transanal irrigation for intractable FC (with or without fecal incontinence) at the time of the study were invited to participate in this cross-sectional survey study via mail between March 2014 and October 2014. Children with FC fulfilled the Rome III criteria, based on clinical assessment. In all
children, transanal irrigation was initiated when they were no older than 18 years of age. Patients with intractable FC had been referred to a specialized pediatric gastroenterology nurse (HV) for transanal irrigation by their pediatric gastroenterologist. Transanal irrigation was only considered as a treatment option if intensive conventional pharmacological treatment had failed. In addition, children with intractable FC are usually referred for psychological evaluation at our center. Transanal irrigation was performed with Peristeen® Anal Irrigation System (Coloplast A/S, Kokkedal, Denmark or Mallinckrodt, St. Louis, MO; FDA K103254).

Children and their parents received information about Peristeen® and were instructed on how to use this irrigation system conform the instructions of the manufacturing company. This included patient-tailored instructions on how to insert and inflate the balloon (e.g., maximum amount of air inflations based on age). During the first time of irrigation, the balloon was inflated until there was no more water leakage from the anus, this indicates that the balloon seals the anal canal. The patients (and their parents) were then supported during outpatient clinic visits until they were able to use Peristeen® at home, from then on follow-up by the pediatric gastroenterology nurse consisted mainly of telephone contacts.

Questionnaires
A questionnaire was sent out to the parents by mail, they were asked to fill out the questionnaire together with their children. The questionnaires were developed by the department of pediatric gastroenterology in collaboration with the department of psychology of the Emma Children’s Hospital/Academic Medical Center. The questionnaire involved 25 questions and consisted of multiple-choice questions regarding gastrointestinal symptoms, use of Peristeen®, concomitant medication use and parental satisfaction with treatment. Completion of the questionnaire took approximately 5-10 minutes. The questionnaires were returned anonymously by mail.

Data analyses
Data was analyzed using SPSS version 21 (Armonk, NY: IBM Corp.). Results are expressed in percentages, mean (with standard deviation) or median depending on whether the data was normally distributed.

Ethical considerations
This study was approved by the local IRB.
Table 1  Patient characteristics of children using Peristeen®

<table>
<thead>
<tr>
<th>Patient characteristics</th>
<th>N (% of total)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Frequency Peristeen use</td>
<td></td>
</tr>
<tr>
<td>- Daily</td>
<td>22 (33)</td>
</tr>
<tr>
<td>- Every other day</td>
<td>17 (25)</td>
</tr>
<tr>
<td>- Every 3 days</td>
<td>4 (6)</td>
</tr>
<tr>
<td>- 1 time/week</td>
<td>2 (3)</td>
</tr>
<tr>
<td>- &lt;1 time/week</td>
<td>2 (3)</td>
</tr>
<tr>
<td>- Other/varying frequency</td>
<td>2 (3)</td>
</tr>
<tr>
<td>- Stopped using Peristeen</td>
<td>18 (27)</td>
</tr>
<tr>
<td>Irrigation fluid</td>
<td></td>
</tr>
<tr>
<td>- Tap water</td>
<td>52 (78)</td>
</tr>
<tr>
<td>- Tap water + bisacodyl</td>
<td>9 (13)</td>
</tr>
<tr>
<td>- Tap water + PEG</td>
<td>2 (3)</td>
</tr>
<tr>
<td>- Tap water + glycerine</td>
<td>2 (3)</td>
</tr>
<tr>
<td>- Tap water + laxative enema</td>
<td>1 (1)</td>
</tr>
<tr>
<td>- Saline</td>
<td>1 (1)</td>
</tr>
<tr>
<td>- Missing</td>
<td>1 (1)</td>
</tr>
<tr>
<td>Concomitant medication</td>
<td></td>
</tr>
<tr>
<td>- No</td>
<td>31 (46)</td>
</tr>
<tr>
<td>- Yes</td>
<td>35 (52)</td>
</tr>
<tr>
<td>- Oral laxatives</td>
<td>29 (83)</td>
</tr>
<tr>
<td>- Enemas</td>
<td>23 (83)</td>
</tr>
<tr>
<td>- Bisacodyl suppositories</td>
<td>2 (6)</td>
</tr>
<tr>
<td>- Missing</td>
<td>1 (1)</td>
</tr>
<tr>
<td>Duration of irrigation</td>
<td></td>
</tr>
<tr>
<td>- ≤30 min</td>
<td>29 (43)</td>
</tr>
<tr>
<td>- 30 – 60 min</td>
<td>34 (51)</td>
</tr>
<tr>
<td>- &gt;60 min</td>
<td>4 (6)</td>
</tr>
</tbody>
</table>

RESULTS

In total, 91 families were invited to participate; 67 questionnaires (74%) were returned. At the time of survey the mean age of the children was 11.2 years (SD 3.8, range 4-19 years), of whom 55% was male. The median duration of symptoms was 7 years and children had started using Peristeen® at a mean age of 10.3 years (SD 3.7, range 3-18 years). At the time of survey, children had used Peristeen® for a median duration of 11 months (range 1 month-3 years). Patient characteristics are depicted in Table 1. A total of 22 children (33%) used Peristeen® daily and 15 children (22%) used it once every other day. Eighteen (27%) children had stopped using Peristeen®, mostly due to ineffectiveness (n=11) or
because they were in remission and did not need Peristeen® anymore ($n=4$). Tap water was used as irrigation fluid in the majority of patients, occasionally supplemented with added laxatives. Half of the children used concomitant oral and/or rectal laxatives in addition to Peristeen®. A total of 28 children ($42\%$) experienced pain during rectal irrigation, especially during insertion of the catheter, pumping up the balloon or during irrigation. More than half of the parents ($57\%$) reported that rectal irrigation was a feasible treatment modality for their child.

The majority of children ($n=56$, $84\%$) suffered from fecal incontinence before initiating treatment with Peristeen®. Out of the children still using Peristeen® at the time of survey ($n=49$), fecal incontinence had resolved in $41\%$, $12\%$ still suffered from infrequent episodes of fecal incontinence ($<1$ episode per week) and $47\%$ suffered regularly from episodes of fecal incontinence ($\geq1$ time per week).

When parents were asked to compare Peristeen® with previous treatment regimens, $72\%$ reported that transanal irrigation was an improvement in the management of their child’s symptoms, $24\%$ reported it was neither better nor worse. The majority of parents ($86\%$) reported that they were satisfied with Peristeen® as a treatment modality and $67\%$ reported that they would continue using Peristeen® in the treatment of their child’s symptoms.

**DISCUSSION**

This study shows that Peristeen® can be a feasible and effective bowel management system for children with intractable FC. In a subset of children, fecal incontinence was treated successfully. The most common adverse effect of treatment was pain. Overall, parental satisfaction was high and no severe complications or side effects were reported.

Intractable FC is a difficult and frustrating problem with a detrimental effect on quality of life. In the majority of children with constipation, conservative treatment including toilet training, dietary advices and oral laxatives is sufficient. In less than $10\%$ of our tertiary population however, invasive treatment such as rectal irrigation is necessary to treat these children and prevent them from undergoing surgery. To date, this study describes the largest population of children with FC using transanal irrigation. The results of our study are in accordance with previous studies demonstrating that transanal colonic irrigation with Peristeen® may be an effective treatment modality for children with constipation and/or fecal incontinence due to organic causes (11–15). Furthermore the results of this study are in accordance with two smaller studies assessing the use of Peristeen® in children with FC (10,17). In a small retrospective study
of 10 children (7 children with FC) using Peristeen®, an improvement of fecal incontinence was observed in all children (10). In this study, fecal incontinence symptoms were assessed using the fecal continence scoring system designed by Rintala et al. (18); this score ranges from 1 to 20, with higher scores reflecting better outcome. The authors describe a significant improvement in the mean fecal incontinence score from 9.7 prior to treatment to 14.8 after treatment. Especially scoring items regarding social problems related to fecal incontinence improved (10). In a more recent study using a prospectively maintained database, Ng et al. evaluated the use of transanal irrigation (Peristeen® or Qufora®) in a mixed population of 42 children with constipation (26 children with FC) (17). They also demonstrated a significant improvement in fecal continence score using the same scoring system and a significant increase in quality of life measured with the PedsQLTM 4.0 Generic Core Scales questionnaire. In accordance with these results, an improvement in quality of life has also been reported in studies evaluating children treated with Peristeen® for defecation disorders due to organic causes (12,19). At the time of our survey, fecal incontinence had resolved in only 41% of children who were still using Peristeen®. However, according to the majority of parents, Peristeen® had led to an improvement of symptoms compared to previous treatment regimens. The fact that these children, who would otherwise have undergone invasive surgery, are now treated with a non-surgical approach in the home setting with high parental satisfaction rates is of substantial clinical importance. Peristeen® can be regarded as a safe option for colonic irrigation in children with intractable constipation. Aside from pain, no severe adverse events have been reported by the parents participating in our study. This is in accordance with previous studies in children using Peristeen® (10–17). Pain and discomfort seem to be the most occurring adverse events and are reported throughout studies as reason for cessation of treatment (17). In addition, based on our experience, we would not recommend transanal irrigation for children with severe anxiety problems, since treatment via an anorectal route may enhance anxiety and refusal of further treatment (20). Furthermore, in patients with poor treatment adherence, transanal irrigation is likely to fail since it is time consuming, which requires commitment and effort from both patients and parents. We described that a proportion of patients added laxatives, especially bisacodyl, to the irrigation fluid, in an attempt to achieve better efficacy of transanal irrigation. The effect of stimulant laxatives on colonic motility has been described in colonic manometry studies, where it has been shown to induce high-amplitude propagated contractions (21–23). High amplitude propagated contractions are considered to enable the transfer of colonic contents over long
distances and they are known to be involved in the process of defecation. By that mechanism, inducing high amplitude propagated contractions during rectal irrigation may help to optimize the irrigation process. Similarly, stimulants have been successfully used in patients with antegrade continence enemas, without reported complications (24,25). Future studies should assess the safety and efficacy of using additives such as stimulant laxatives during transanal irrigation.

There are several limitations to this study. There is a risk of selection bias; some patients or their parents may have been reluctant to participate in this study because they had stopped using Peristeen® due to ineffectiveness of treatment or due to adverse effects. On the other hand, the response rate in our study was high (74%). Moreover, we also received questionnaires from children that had ceased using Peristeen® due to ineffectiveness. This type of survey study always carries the risk of social desirability bias, to minimize this risk the questionnaires in this study were anonymous. Children included in this study often used concomitant medication, which may have influenced our results. However, in all children, transanal irrigation had been initiated only after unsuccessful pharmacological treatment, indicating that pharmacological treatment alone is unlikely to account for these results in these patients. Furthermore, this study was based on a self-developed non-validated questionnaire, not taking into account validated measurements before and after treatment. To further assess the usefulness of Peristeen® in the management of children with constipation, prospective studies should be conducted, preferably with validated measurements (e.g. quality of life questionnaires) and standardized symptom-based outcome measures (e.g. bowel diary and fecal incontinence scores) so that patient and parent reported satisfaction can be matched with clinical information. A randomized controlled trial comparing the efficacy, side effects and patient/parental satisfaction of rectal enemas and Peristeen® would be of great interest.

In conclusion, transanal irrigation with Peristeen® is likely to be a valuable tool in the management of children with intractable FC and renders high parental satisfaction rates. It should therefore always be considered as a treatment modality for children with intractable FC before proceeding to more invasive, surgical management. Future prospective studies are necessary to further evaluate this treatment option.

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REFERENCES


20. Giorgio V, Borrelli O, Smith V V., et al. High-resolution colonic manometry accurately...


