Systematic Review and Meta-Analysis of Behavioral Interventions for Fecal Incontinence With Constipation

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Background Multiple treatments exist for fecal incontinence. However, the relative and additive influence of commonly used behavioral approaches remains unclear. Objective We conducted a systematic review of randomized controlled trials to synthesize the effects of behavioral treatment of fecal incontinence with constipation in children aged 4–18 years. Mixed treatment comparisons (MTCs) and random effects models were used to analyze outcomes. Risk of bias and quality of evidence were rated. Results Although 10 studies were identified for MTCs, results did not yield reliable or valid estimates. Four studies were retained for random effects pooled outcome analysis. Results indicated that behavioral intervention was more effective than control conditions for author-defined success and soiling frequency. Conclusion Although evidence supports behavioral treatments for fecal incontinence with constipation in children, available evidence is limited. More and higher-quality trials are needed to better understand the relative effects of different treatments, including behavioral strategies.

Key words encopresis; evidence-based practice; fecal incontinence; meta-analysis; systematic review.

Encopresis, or fecal incontinence1, marked by the evacuation of fecal stool at unwanted times and places, represents a common, distressing, and impairing condition for children and families. Estimates of prevalence of fecal incontinence in children have varied from 0.3 to 8% (Howe & Walker, 1992; Levine, 1975; Loening-Baucke, 2007; Van der Wal, Benninga, & Hirasing, 2005), with boys disproportionately affected. Historically, psychological impairments were emphasized in the etiology of fecal incontinence (Wright, Schaefer, & Solomons, 1979); however, numerous investigations have failed to support this perspective (Cox, Morris, Borowitz, & Sutphen, 2002; Friman, Mathews, Finney, Christophersen, & Leibowitz, 1988; Gabel, Hegedus, Wald, Chandra, & Chiponis, 1986; Loening-Baucke, Cruikshank, & Savage, 1987). Although fecal incontinence is not a product of psychopathology, several reports have characterized the potentially distressful nature of fecal soiling and associated negative psychosocial outcomes for affected children (Bongers, van Dijk, Benninga, & Grootenhuis, 2009; Cox et al., 2002; von Gontard, Baeyens, Van Hoecke, Warzak, & Bachmann, 2011) and their parents (Akça, Aysev, & Aycan, 2011; Demir et al., 2012). Further, successful treatment of fecal incontinence leads to behavioral improvement (Young, Brennen, Baker, & Baker, 1995). Thus, an interaction of physiological and behavioral processes is apparent, and successful treatment requires a good understanding of both. Improved understanding of specific treatment elements that optimize outcomes is important to informing clinical practice.

Processes of Defecation and Fecal Incontinence

To appreciate the need for a range of medical and behavioral treatments to address fecal incontinence and constipation, foundational understanding of the processes of...
defecation and fecal incontinence is useful. Normal defecation involves the integration and coordination of somatomotor functions of the colon, rectum, pelvic floor, and external anal sphincter (EAS; Palit, Lunniss, & Scott, 2012). Although most of these processes occur involuntarily, they can be strengthened or counteracted through volitional behavior.

Development and maintenance of fecal incontinence generally involves some dysfunction of the processes of defecation. The majority of cases of fecal incontinence occur in the context of constipation and bowel retention, with estimates of constipation prevalence in affected children ranging as high as 80–95% (Hatch, 1988; Partin, Hamill, Fischel, & Partin, 1992). Idiopathic constipation can affect the functioning of the colon and rectum in two important ways (Friman & Freeman, 2010; Lanfranchi, Bazzocchi, Brignola, Campieri, & Labo, 1984; Velio & Bassotti, 1996). First, large masses of fecal matter stretch the colon and thus reduce the ability of the muscles to engage in peristaltic movements. Second, nerve endings in the colon habituate to the sensation of fecal matter that typically signals the need for peristaltic movements, thereby decreasing the likelihood that an affected youth experiences the urge to defecate. In addition to disruption of colonic processes, a significant proportion of children with fecal incontinence evidence improper defecation dynamics (Loening-Baucke & Cruikshank, 1986; Loening-Baucke, Cruikshank, & Savage, 1987), particularly contraction of the EAS, which may develop as a learned avoidance response in reaction to the passage of large painful stools. In addition to direct aversive conditioning through pain, children who are incontinent are often admonished or punished for soiling accidents, which can result in a host of internal (e.g., sensation of full rectum) and external (e.g., toilets) stimuli associated with defecation becoming aversive. Attempted avoidance of these stimuli may lead to toileting resistance, stool withholding, or incomplete voiding (via paradoxical contraction), thus exacerbating constipation and maintaining a maladaptive pattern (Luxem, Christophersen, Purvis, & Baer, 1997; Taubman, Blum, & Nemeth, 2003). In this way, gastrointestinal, neuromuscular, behavioral, and socioemotional processes interact to establish and maintain fecal incontinence.

Available Treatments

To address the multiple processes involved in fecal incontinence, various treatments have been developed. The primary goal of treatment of fecal incontinence is to reduce soiling by establishing regular bowel movements in the toilet of sufficient size and appropriate consistency. Usually, the first step in achieving this goal is to cleanse the bowel completely of fecal matter, often through use of an enema, suppository, or oral laxative; this is often followed by a maintenance laxative regimen to avoid repeated constipation. These medical interventions are considered the frontline treatments for fecal incontinence associated with constipation; however, available evidence suggests a cure rate of only 40% with medical intervention alone (McGrath, Mellon, & Murphy, 2000).

Because of the limitations of medical intervention alone, behavioral methods are often used as an adjunct to laxative therapy. These methods typically are designed to ameliorate learned behavior that contributes to ongoing stool retention or toileting avoidance. A combined medical and behavioral approach may consist of (a) education about the physiological and behavioral processes associated with toileting, (b) medical intervention to ensure bowel evacuation using enemas or laxatives, (c) behavioral methods for skill-building and motivational enhancement (e.g., toilet scheduling, defecation skills training, differential reinforcement of bowel movements on the toilet), (d) dietary and fluid intake interventions and goal setting, and (e) maintenance of interventions (Friman & Freeman, 2010).

Given that a significant proportion of children with fecal incontinence and constipation evidence improper defecation dynamics (Loening-Baucke & Cruikshank, 1986; Loening-Baucke et al., 1987), another approach described in the treatment literature is biofeedback. This involves visual or auditory representation of the muscle tone of the EAS and engaging in specific exercises to improve either muscle tone or muscle coordination (Loening-Baucke, 1990).

Previous Reviews

With the availability of multiple treatments, understanding which treatment or combination of treatments is most effective for children with fecal incontinence is important. Two systematic reviews and one meta-analysis have been conducted to evaluate the relative benefit of available treatments. McGrath et al. (2000) reviewed 42 published articles, inclusive of 13 studies that used single-subject designs, evaluating behavioral and medical treatments for constipation and fecal incontinence using the “Chambless criteria” (Chambless & Hollon, 1998). At that time, comprehensive medical treatment alone did not meet efficacy criteria. Four treatments were identified as probably
Efficacious: medical intervention plus positive reinforcement with fiber recommendation, medical intervention plus positive reinforcement without fiber recommendation, comprehensive medical intervention plus biofeedback for paradoxical contraction (for children with constipation and abnormal defecation dynamics only), and one treatment with medical, behavioral, and biofeedback components. McGrath et al. noted a lack of systematic replication, such that it was difficult to identify consistency of treatment components across studies. This inconsistency, combined with heterogeneous reporting practices in terms of how interventions are classified and outcomes of interest, limited the strength of the conclusions.

Brooks and colleagues (2000) reviewed nine randomized controlled trials (RCTs) and three noncontrolled trials involving medical, behavioral, psychological, and biofeedback treatments for fecal incontinence published between 1973 and 1999. Behavioral treatment and combined medical-behavioral treatment were concluded to be equally efficacious when compliance with prescribed procedures was good. No evidence supported the additive value of biofeedback to comprehensive medical-behavior intervention, nor was improved paradoxical contraction found to be related to outcomes. Similar to McGrath et al. (2000), Brooks et al. (2000) noted a stark lack of quality randomized trials to support clinical practice, as well as inconsistency in reporting practices regarding treatments and outcomes across publications.

More recently, Brazzelli Griffiths, Cody, and Tappin (2011) completed a Cochrane Review of interventions for fecal incontinence. RCTs comparing behavior and/or cognitive interventions (with or without other interventions) with other behavioral/cognitive treatments, conventional treatment, or other methods of fecal incontinence treatment, or no treatment were included. Notably, samples of children with nonretentive encopresis, which represent a minority of those with fecal incontinence, were included. Twenty-four reports from 21 studies with 1371 participants were included in the review. Nineteen reports from 16 studies were included in the meta-analysis, although only results from 9 studies involving biofeedback were subjected to pooled outcome analysis; all other studies were analyzed by determining the odds ratio for treatment outcomes across treatment conditions. The authors concluded (a) biofeedback did not provide additional benefits beyond conventional treatment, and (b) weak evidence supported the benefit of behavioral methods adjunctive to laxative therapy. Like most meta-analyses, Brazzelli et al. (2011) limited statistical analyses to pairwise direct comparisons of competing treatments (i.e., Treatment A vs. Treatment B).

As noted by Brazzelli et al. (2011), the treatment literature for fecal incontinence lacks many of the necessary direct comparisons to conclusively answer important clinically relevant questions. For example, no trials comparing behavioral/cognitive intervention or other psychological intervention with no treatment, sham procedure, or another intervention were found. Additionally, each previous reviewer (Brazzelli et al., 2011; Brooks et al., 2000; McGrath et al., 2000) has described the literature as heterogeneous in terms of reporting quality, treatment components, treatment comparisons, outcome measures, and methodological rigor. As a result, characterizing the literature using direct comparison meta-analytic procedures have been difficult and conclusions have been limited.

**Current Review**

Different from McGrath et al. (2000) and Brooks et al. (2000), the current review and meta-analysis of behavioral treatments for pediatric fecal incontinence with associated constipation used rigorous methods for systematic reviews including risk of bias and quality of evidence assessment. The aim of the current review was to supplement and extend the findings of previous reviews by (a) focusing on RCTs of children with idiopathic constipation and incontinence, (b) using novel meta-analytic approaches to analyze information across heterogeneous studies, and (c) differentiating biofeedback from other behavioral approaches. It was hoped that the current systematic review and meta-analysis would increase understanding of contributions of behavioral interventions for fecal incontinence with associated constipation through the use of mixed treatment comparison (MTC) methods, which can provide useful information about the relative efficacy of interventions that have not been directly compared in clinical trials (Lumley, 2002; Song, Altman, Glenny, & Deeks, 2003). Unlike Brazzelli et al. (2011) who included studies of treatment for fecal incontinence with or without associated constipation, we restricted analysis to youth with idiopathic constipation and incontinence, the large majority of all children with fecal incontinence. This is an important distinction, as constipation is thought to play an important role in the development and maintenance of incontinence, and most therapies directly target alleviation of constipation as a primary component of treatment. Consistent with previous reviews (Brazzelli et al., 2011), in the current review, biofeedback was considered distinct from other behavioral interventions given evidence that biofeedback is likely effective only with a subset of children with fecal incontinence (Wald, 1983; Wald, Chandra, Gabel, &
Chiponis, 1987) and pooled analysis had been recently conducted (Brazzelli et al., 2011).

Because previous reviews have noted heterogeneity regarding reported treatments and outcomes, the feasibility of conducting MTC analyses was unclear at the outset of the current systematic review. Therefore, in the event MTC did not produce reliable outcomes, the review protocol was designed to conduct the usual meta-analyses of studies that used behavioral treatments as major components of treatment arms, and to compare findings from those conditions against laxative only/control conditions. Results of such analyses were not available in the literature and would provide a unique contribution.

Method
Criteria for Considering Studies for Review
This comparative effectiveness review focused on the effects of behavioral intervention to improve continence among youth aged 4–18 years with fecal incontinence and constipation.

Types of Studies
All RCTs in which at least one treatment arm was some combination of the following:

Behavioral intervention (single or combined)
Medical/laxative intervention (single or combined)
Biofeedback intervention

Specific inclusion criteria for studies were as follows:

- The studies were RCTs
- The samples included children with fecal incontinence and associated constipation
- Fecal incontinence was idiopathic [i.e., not due to a known physical condition (such as myelomeningocele) or anorectal malformation]
- There were at least 10 participants in each treatment arm
- The study was available in English

Types of Participants
Children between the ages of 4–18 years with a history of fecal incontinence with constipation were included. Fecal incontinence is defined here as the repeated passage of feces in any location other than the toilet. The term “constipation” may generally be defined as difficulty or delay in the passage of stool; for this review, study authors’ definitions of constipation were used.

Interventions of Interest
Given our primary interest was examining the relative effects of behavioral intervention, we included studies that compared behavioral interventions with other treatments. Using previous reviews and meta-analyses (Brazzelli et al., 2011; Brooks et al., 2000; McGrath et al., 2000) as a guide, behavioral interventions were defined as those that included one or a combination of the following components:

- Psychoeducation
- Rewards
- Response cost/punishment
- Defecation skills training
- Scheduled toileting sits
- Environmental modifications
- Prescription of diet modification

Medical/laxative interventions included one or any combination of the following:

- Oral laxatives
  - Osmotics
  - Stimulants
  - Stool softeners
- Anal laxatives
  - Glycerin suppository
  - Bisacodyl suppository
  - Enema

Biofeedback involves displaying muscle tone of the EAS through visual or auditory representation and engaging in specific activities to improve either muscle tone or muscle coordination. Although slight variations in biofeedback procedures exist in the literature, such distinction was not of interest for the current review because the effect of biofeedback was not of primary interest.

Treatment was defined as “Placebo” when described by authors as involving the use of an inert substance identical in physical properties so as to mimic a prescribed laxative.

Intervention was defined as “Treatment as Usual” (TAU) when the authors indicated either the typical intervention continued or that no specific intervention was described; given the virtually ubiquitous use of laxatives with children presenting with constipation, it is reasonable to presume that these children were receiving some form of laxative therapy.

Outcomes of Interest
Using Rome III criteria for fecal incontinence and constipation (Rasquin et al., 2006), and previous indications of
the variability in treatment outcomes included in this literature (Brazzelli et al., 2011), the following outcomes were included:

- Dichotomous outcomes
  - Number of children who had an 80% increase in accident-free days
  - Number of children with ≤1 soiling accident per month
  - Number of children with ≥50% increase in bowel movements in the toilet per week
  - Number of children defecating ≥3 times per week
  - Number of children who met “Author-Defined Success,” defined as the criteria used to determine treatment success in any individual study. This outcome was necessary given the inconsistency of reported outcome metrics used in the literature.

- Continuous outcomes (converted to weekly frequency as needed)
  - Frequency of soiling accidents
  - Frequency of bowel movements in the toilet

**Search Methods for Identification of Studies**

To identify published studies, we searched Ovid [MEDLINE, PsychINFO-search terms (fecal incontinence AND child) AND (psychotherapy OR drug therapy OR biofeedback)], EBSCOhost [CINAHL-search terms (fecal incontinence AND child) AND (psychotherapy OR cathartics OR biofeedback)], and PubMed-search terms [fecal (incontinence OR encopresis) AND child AND (psychotherapy OR laxatives OR biofeedback)]. The search included all studies published before April 1, 2013. In addition, reference lists for all previous systematic reviews and identified trials were searched to identify other potential studies.

**Data Collection and Analysis**

**Selection of Studies**

After removing duplicate studies identified via search procedures, titles and abstracts were reviewed by two investigators to determine whether they warranted further review based on inclusion and exclusion criteria. Disagreements regarding study inclusion were resolved by discussion; when the disagreement was due to a difference in interpretation, arbitration regarding the issue was facilitated by the first author. Discussion continued until consensus was reached.

**Data Extraction and Management**

Information on the characteristics of participants, interventions, and prespecified outcome variables was independently extracted from each study by two investigators (data extraction form available from the first author). Any disagreements in extracted information were resolved via re-review and discussion; a third author arbitrated discussion if needed. Discussion continued until consensus was reached.

**Assessment of Risk of Bias in Included Studies**

Information on random sequence generation (selection bias), allocation concealment (selection bias), blinding of participants and personnel (performance bias), blinding of outcome assessment (detection bias), incomplete outcome data (attrition bias), and selective reporting (reporting bias) was extracted from each trial using criteria recommended by the Cochrane Handbook for Systematic Reviews of Interventions (Higgins & Green, 2011). Each study was independently assessed for bias by two of the first three authors. Disagreements were resolved through conversation and reexamination of criteria.

**Quality of Evidence**

Extracted study data were used to determine the strength of evidence as per the Grading of Recommendations, Assessment, Development, and Evaluation (GRADE) system (Moher et al., 1999). This includes rating strength of evidence from very low to high based on several criteria (i.e., risk of bias, inconsistency, indirectness, and imprecision).

**Meta-Analytic Approach**

MTC analysis was attempted to estimate the relative effects of treatments that have not been directly compared (Lu & Ades, 2006). For example, if Treatment A and Treatment B have never been directly compared, but both have been compared with Treatment C, it is possible to indirectly estimate the relative effects of A and B if the included studies were comparable in patient characteristics, methods, and other features. The MTC analysis was conducted using WinBUGS 1.4.3 (Imperial College and MRC, U.K.).

In contrast to MTC analysis, a random effect model (DerSimonian & Laird, 1986) was used to determine the relative effect of treatments that have been directly compared in more than one study. For dichotomous outcomes, risk ratio was used as an effect measure; for continuous outcomes, mean difference (MD) was the effect measure. Statistical heterogeneity among studies was assessed using the \( \chi^2 \) test and \( I^2 \) statistics (Higgins & Thompson, 2002). The random effect analysis was conducted using RevMan 5.2.
Results

Description of Studies

Based on the search procedures, 545 articles were identified for initial review (Figure 1). Thirty-two studies were selected for full-text review, and 14 studies were included for the next level of review. For five of the studies (Loening-Baucke, Miele, & Staiano, 2004; Mellon, Houts, & Lazar, 1996; Ritterband et al., 2013; van Dijk et al., 2008; Wald et al., 1987), the methodology met inclusion criteria, but data for one or more outcomes of interest were presented in a manner that was incompatible for inclusion. The lead author of each of those articles was contacted via e-mail to request data be provided in a format useful for the current study. Only one was able to provide data suitable for inclusion (Ritterband et al., 2013). As a result, 10 studies were retained for inclusion in the MTC analysis (see Supplemental Material for detailed description of retained studies).

Summary of Articles Identified for Inclusion in Meta-Analysis

Participants

The 10 studies retained for inclusion in the meta-analysis including 562 youth. Consistent with previous findings that males are disproportionately affected by fecal incontinence, 74.3% of participants in these studies were male. Mean age was 7.9 years. Few studies reported participant race/ethnicity.

Location/Setting

Five studies were conducted in the United States, one in the Netherlands, two in Australia (note, Nolan, Debelle, Oberklaid, & Coffey, 1991, provided no information about location of study; however, as Nolan, Catto-Smith, Coffey, & Wells, 1998, was conducted in Australia by similar authors, it is presumed that the 1991 article was also conducted in that country), one in the U.K., and one in Mexico City, Mexico. Eight were conducted in ambulatory practices, often associated with pediatric gastroenterology. Two (Ritterband et al., 2003, 2013) were conducted via the internet.

Sample Size

Sample size of youth with fecal incontinence secondary to constipation ranged across studies from 24 to 169. The number of participants per treatment condition was generally small, often <20.

Intervention

When initially extracting information from selected trials, each specific intervention component included was indicated (e.g., “rewards,” “scheduled sits,” “osmotic laxative,” “placebo”). This revealed that most study arms used multiple intervention components and that treatment arms were rarely constrained to only one category of intervention (e.g., only laxatives; Online Supplemental Table of the Characteristics of Included Studies). Given the primary goal of this review to evaluate the relative effectiveness of including behavioral intervention in the treatment of fecal incontinence with constipation and to ensure outcome data were available for analysis, it was necessary to combine specific interventions into broader categories. Categories were based on what appeared to be the primary intervention components rather than attempting to investigate the relative influence of each intervention (e.g., rewards, oral laxative). Broad categories included “Behavioral,” “Laxative,” “Biofeedback,” “Placebo,” “TAU,” or “Fiber.” The first and second authors independently evaluated data extracted for each treatment component included in each arm of the RCTs and classified the major treatment components; disagreements would have been addressed via discussion but none occurred. This resulted in a determination of whether an intervention arm used a specific category of intervention (e.g., “Behavioral,” “Laxative”) or combination of interventions (e.g., “Behavioral + Laxative”). Based on this classification, only six trials had at least one arm for which behavioral intervention(s) were a primary component; the remaining four trials qualified for inclusion in the planned MTC analyses because they contained treatment arms that had been compared with behavioral interventions in other studies.

Outcomes

The only dichotomous variable available for analysis was “author-defined success” because no information regarding the other potential dichotomous outcomes was provided in a consistent manner across studies.

Five studies (Borowitz, Cox, Surphen, & Kovatchev, 2002; Cox, Surphen, Ling, Quillian, & Borowitz, 1996; Loening-Baucke, 1990; Nurko, Garcia-Aranda, Worona, & Zlochisht, 2000; Ritterband et al., 2003, 2013) provided information about frequency of soiling accidents. Data were converted to frequency per week for standardization across studies. Four studies (Borowitz et al., 2002; Cox et al., 1996; Loening-Baucke, 1990; Nurko et al., 2000; Ritterband et al., 2003) provided data regarding frequency of bowel movements; again, this outcome variable was converted to frequency per week for standardization across studies. However, only two of those studies (Borowitz et al., 2002; Cox et al., 1996) published data necessary for analysis. Data from a third study (Ritterband et al., 2003) were provided through direct communication with the authors.
Risk of Bias
Authors’ judgments of risk of bias are detailed for each study retained for quantitative synthesis is summarized in Figure 2 (for detailed description, see Online Supplemental Material Characteristics of Included Studies).

Random Sequence Generation
Randomization sequence generation was judged to be adequate for four trials (low risk of bias) and unclear for six trials (unclear risk). For the latter, authors simply did not state how they generated the randomization sequence.

Allocation Concealment
Randomization concealment was acceptable for four trials (low risk) and unclear for six trials (unclear risk). Again, risk was unclear because authors did not provide information that allowed determining how they concealed randomization.

Blinding of Participants and Personnel
Blinding of participants and personnel was either adequate (low risk for three trials) or high (seven trials). High risk trials used nonmedical interventions for which blinding of the treatment condition would be impossible.
Blinding of Outcome Assessment
Blinding of outcome assessment was either adequate (three trials) or high risk (seven trials). Again, those trials judged as having a high risk of bias for outcome assessment included nonmedical treatments for which it would have been impossible to disguise the treatment being provided to participants.

Incomplete Outcome Data
Five trials were judged to have low risk of attrition bias due to incomplete data. Three were judged to have unclear risk. Two were judged as high risk; those trials typically demonstrated reasonably high attrition, yet the authors described no method to account for this when conducting analyses.

Selective Reporting
Reporting of outcomes was judged as unclear (unclear risk) for all trials because specific protocols and convincing evidence that all expected outcomes, including those that were predetermined by the authors, were unavailable (Higgins & Green, 2011).

Meta-Analysis
Mixed Treatment Comparisons
To investigate the comparative effectiveness among different treatments, we attempted MTC analysis (Lu & Ades, 2006) for author-defined success and weekly frequency of fecal incontinence. However, because of the small number of included studies, small sample sizes, and heterogeneity across studies, analyses did not yield reliable or valid estimates for indirect comparisons. Attempting such comparisons for frequency of bowel movements in the toilet was not possible because only two studies provided the necessary data.

Random Effects Analysis
Because MTC did not produce reliable or valid results, four studies that compared behavioral intervention (i.e., “Behavioral + Laxative” or “Behavioral + Biofeedback + Laxative”) against laxative treatment/TAU (nonbehavioral control condition) (Borowitz et al., 2002; Cox et al., 1996; Ritterband et al., 2003, 2013) were pooled using the usual random effects model. These studies included 216 youth with a mean age of 8.6 years; 80.4% of participants in these studies were male.

For the dichotomous outcome variable “author-defined success,” all four studies reported data, and the pooled risk ratio indicated that participants were 1.78 [95% confidence interval (CI) = 1.23, 2.55; \( p = .002 \)] times more likely to meet criteria for success in conditions containing behavioral intervention (Figure 3).

For the continuous variable of soiling frequency, all four studies reported data suitable for the pooled analysis. Overall, based on 209 youth (\( M \) age = 8.75 years, 80.5% male), there was a significant effect at posttreatment, and inclusion of behavioral treatment resulted in an average of 2.81 fewer soiling episodes per week (95% CI = −5.04, −0.58; \( p = .01 \)) (Figure 4).

Three studies (Borowitz, et al., 2002, Cox et al., 1996; Ritterband et al., 2003) provided data useful for pooled analysis to examine the effects of behavioral treatment on the frequency of defecation in the toilet. This analysis included 148 youth (\( M \) age = 8.68 years; 80.2% were male) and did not show a significant effect (MD = −0.57, 95% CI = −2.90, 1.75, \( p = .63 \)) at posttreatment (Figure 5). Significant heterogeneity in treatment effects between studies was observed (\( I^2 = 72\% \); \( p = .03 \)). For two of the three studies (Borowitz et al.; Cox et al.), direction of findings favors laxative alone/TAU, whereas for the other (Ritterband et al., 2003) findings favored behavioral treatment.

GRADE Ratings
The quality of evidence for included studies was evaluated based on GRADE criteria. Overall, the quality of evidence in support of the included studies was rated as low (Table 1). Low quality suggests that further research is
very likely to have an important impact on the confidence in the estimate of effect and is also likely to change the estimate. Each outcome was independently evaluated and rated as low due to the following:

- **Author-defined success**: Multiple areas of risk of bias were not reported; thus, risk of bias was unclear. The total number of participants was <300 and variability existed in the definition of author-defined success.
- **Soiling frequency**: Multiple areas of risk of bias were not reported; thus, risk of bias was unclear. The total number of participants was <300.
- **Frequency of defecation in the toilet**: Multiple areas of risk of bias were not reported; thus, risk of bias was unclear. The total number of participants was <400, and the direction of treatment effects was not consistent across studies.

**Discussion**

**Summary and Interpretation of Findings**

For the current review, we attempted an MTC meta-analytic approach to investigate the relative effectiveness of behavioral interventions for fecal incontinence with associated constipation. Unfortunately, MTC results were not reliable because of variability in extant research, small sample sizes, and inconsistent outcome reporting. Random effects pooled analysis with four studies that included treatment arms with a behavioral component compared against laxative/TAU was completed for...
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<th>Number of studies</th>
<th>Design</th>
<th>Risk of bias</th>
<th>Inconsistency</th>
<th>Indirectness</th>
<th>Imprecision</th>
<th>Other considerations</th>
<th>Quality assessment</th>
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<th>Relative Effect (95% CI)</th>
<th>Absolute Effect</th>
<th>Quality</th>
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<td>4</td>
<td>Randomized trials</td>
<td>Serious&lt;sup&gt;a&lt;/sup&gt;</td>
<td>Serious&lt;sup&gt;b&lt;/sup&gt;</td>
<td>No serious indirectness</td>
<td>Serious&lt;sup&gt;c&lt;/sup&gt;</td>
<td>None</td>
<td>72/127 (56.7%)</td>
<td>24/89 (27%)</td>
<td>RR 1.78 (1.25–2.55)</td>
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<td>4</td>
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<td>Serious&lt;sup&gt;a&lt;/sup&gt;</td>
<td>No serious indirectness</td>
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<td>Serious&lt;sup&gt;d&lt;/sup&gt;</td>
<td>None</td>
<td>122</td>
<td>87</td>
<td>–</td>
<td>Mean difference (MD) 2.81 lower (5.04–0.58 lower)</td>
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<td>3</td>
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<td>MD 0.57 lower (2.9–1.75 lower)</td>
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Note. <sup>a</sup>Multiple domains of risk of bias rated as unclear.
<sup>b</sup>Difference in how success was defined across studies ("no soiling accidents" for a specified period of time vs. "statistically significant decrease in accidents").
<sup>c</sup>Total number of events is <300.
<sup>d</sup>Total number of events is <400.
<sup>e</sup>Heterogeneity in direction of treatment effects across studies.


**High quality**: Further research is very unlikely to change our confidence in the estimate of effect.

**Moderate quality**: Further research is likely to have an important impact on our confidence in the estimate of effect and may change the estimate.

**Low quality**: Further research is very likely to have an important impact on our confidence in the estimate of effect and is likely to change the estimate.

**Very low quality**: We are very uncertain about the estimate.
author-defined success, soiling frequency, and frequency of defeating in the toilet. Completing such an analysis is a unique addition to the literature beyond recent comprehensive reviews (Brazzelli et al., 2011).

The random effects pooled analysis suggests that inclusion of behavioral treatment results in a greater number of youth meeting author-defined success and less frequent soiling as compared with laxative therapy alone/TAU. However, the effect of behavioral treatment on the frequency of bowel movements in the toilet was not significant; inspection of studies that included this outcome revealed that two studies actually favor laxative therapy/TAU. The reason for differential influence of behavioral treatment across outcome variables is unclear as each of the studies included in the pooled analysis reported using similar behavioral interventions. However, it may be that the “TAU” control condition used by Ritterband et al. (2003) represents a markedly different comparison versus a known laxative comparison. However, this does not fully explain why laxative treatment alone would be favored in the Borowitz et al. (2002) and Cox et al. (1996) studies, as behavioral treatment conditions in those studies also included use of laxatives comparable with the control condition. Interestingly, participants actually defeated in the toilet less frequently overall in the study favoring behavioral treatments than in the other studies. Thus, it may be that the total number of bowel movements is less important than ensuring one complete bowel movement on the toilet per day on average. Additional research is needed to provide stable pooled analysis and to better understand whether behavioral treatments differentially affect various outcomes.

Limitations of Current Research

The inability to draw strong conclusions from the extant literature on behavioral treatment for fecal incontinence with idiopathic constipation is influenced by several factors. The number of high-quality RCTs that have investigated the influence of behavioral interventions remains small. Further, despite the use of advanced statistical techniques (MTC), reliable estimates of comparative treatment effects could not be obtained. Finally, only four studies remained available for the pooled analyses. Although pooled analysis results represent a unique contribution to the literature, estimates of treatment effects are considered unstable based on GRADE criteria. This reflects the continued need for a more coherent line of research examining the treatment of fecal incontinence.

Although there have been notable additions to the literature (Ritterband et al., 2013; van Dijk et al., 2008) since reviews by Brooks et al. (2000) and McGrath et al. (2000), conclusions and recommendations made over a decade ago remain germane. The treatment literature remains disjointed, disparate, and difficult to synthesize. A key issue is the use of imprecise treatment descriptors (e.g., “conventional medical treatment” or “behavior modifications”) which leads to equivocation of heterogeneous treatments. For example, Berg, Forsythe, Holt, & Watts (1983) compared a behavioral treatment that included scheduled sits on the toilet with the same treatment in combination with laxative therapy. Conversely, in comparing laxative therapy and behavioral intervention, Cox et al. (1996) included scheduled sits as a component of laxative therapy. These shared components across types of treatment confound and complicate comparisons between studies. Even when authors are careful to distinguish treatment approaches, methods tend to be subsumed under larger terminological umbrellas. As shown in the Characteristics of Included Studies (Online Supplemental Material), most intervention arms have not been purely “medical” or “behavioral” (e.g., many interventions designated as medical have also included use of behavioral components). Further, the use of sitting schedules and operant techniques has been common across behavioral and medical treatments. Whether these techniques are truly medical or behavioral in nature is unimportant, but inconsistency of nomenclature leads to inaccurate conclusions regarding the effectiveness of different treatments. Further, data rarely have been presented regarding dosing or treatment engagement (exceptions being frequency of engagement with the intervention Web site used by Ritterband et al., 2003 and 2013), so it is difficult to differentiate the effects of treatment from treatment dosing and compliance.

An additional weakness has been inconsistency in the reporting of outcomes. Of the 14 RCTs identified for potential inclusion in the quantitative synthesis, only 5 and 3 studies reported changes in the frequency of soiling and bowel movements in the toilet, respectively. Given these are the two primary outcomes of interest when treating fecal incontinence, this is a surprisingly low figure. Few studies report cure rates. Many have opted for a statistically significant reduction in soiling or “treatment success” as the primary outcome. Definitions of success have varied greatly, and the social validity of these outcomes is unknown.

The existing literature also lacks systematic study of sample characteristics both within and between studies. Samples may vary in several important ways including presence of constipation, inappropriate defecation dynamics, and previous treatment. While some studies document these characteristics well, others neglect to do so. Improved characterization of samples is important because...
available treatments may be differentially efficacious based on those characteristics. For example, although biofeedback appears to offer little benefit for fecal incontinence in general, for a subset of children with abnormal defecation dynamics, it may offer advantages beyond medical treatment (Wald, 1983; Wald et al., 1987). This type of linkage between individual characteristic and treatment outcomes has potential to greatly influence clinical practice, but potential moderation of treatment effects can only be assessed when samples have been properly characterized.

Implications for Future Research
The results of this and previous reviews clearly indicate the need for more and higher-quality trials of treatments for pediatric fecal incontinence. One factor in the quality of the literature is that most trials were carried out some time ago. Standards of methodological rigor, statistical analysis, and reporting have shifted considerably in recent years. More recent studies (Ritterband et al., 2013) are of higher quality but are few and far between. Reasons that more and better quality research has not occurred over time are unclear. Fecal incontinence is not rare; it accounts for up to 25% of all pediatric gastroenterology consultations (Partin et al., 1992). Further, fecal incontinence is clearly a distressing and impairing condition for patients and families. It may be that available treatments are perceived as more effective than research indicates; however, the near ubiquitous use of laxatives to address constipation boasts only modest empirical support (Gordon, Naidoo, Akobeng, & Thomas, 2012).

Despite the need for continued and higher-quality research, study of the treatment of fecal incontinence with constipation has several barriers. Although fecal incontinence is not rare, recruiting numbers sufficient to power large RCTs requires allocation of significant resources. Collaboration across multiple sites or use of technological approaches to deliver interventions as part of research may mitigate this obstacle to some degree. A recent RCT applying comprehensive intervention via a Web-based approach (Ritterband et al., 2013) has one of the larger samples sizes of research retained in this meta-analysis. This supports that future research endeavors may wish to continue exploration of how technology can be used to both deliver interventions but also to recruit and retain larger numbers of research participants.

Terminological and Methodological Coherence
Reasoned choices regarding populations studied, treatment components, and reported outcomes are paramount to increasing the coherence of this body of literature. A prerequisite to doing so is increasing the consistency of the terminology used. The term encopresis itself is used inconsistently (Benninga et al., 2005). The Paris Consensus on Childhood Constipation Terminology (Benninga et al., 2005) and Rome III criteria for functional gastrointestinal disorders (Rasquin et al., 2006) are efforts to alleviate this issue, and it is recommended researchers consider those guidelines when designing, conducting, and reporting trials. One application is in the reporting of participant characteristics. For instance, the proportion of participants with functional constipation and incontinence versus nonretentive fecal incontinence should be reported at baseline, and outcomes for those subsamples should be reported separately.

Collecting data on defecation and soiling can be challenging for several reasons: (1) it is reliant of self- and/or proxy-report, (2) defecation normally takes place in private, and (3) many children with fecal incontinence are secretive about their defecation habits. A well-constructed psychologically sound assessment tool designed to measure symptom severity and treatment progress might mitigate this obstacle, as well as encourage consistent reporting of outcomes across trials. The Virginia Encopresis-Constipation Apperception Test (Cox et al., 2003) is sensitive to change but is designed to measure behavioral aspects that may contribute to constipation and incontinence, rather than symptom severity. A valid repeatable measure would serve as a valuable adjunct to self- and/or proxy report.

Mechanisms of Action
A relative strength of the fecal incontinence literature is the identification of multiple treatments of varying intensities, each of which may be used in isolation or in combination. How to best apply these strategies for any individual patient remains unknown. Across RCTs, behavioral intervention “packages” are diverse in terms of specific components. As a result, even when pooled analysis is possible, determining which treatment components are responsible for clinical improvement is not possible. Methodologies that allow for component analysis are desirable. Recently, interest has grown in the use of “stepped-care” approaches to psychological and medical treatment, signified by the development of Sequential Multiple Assignment Randomized Trial (SMART) methodology (Lei, Nahum-Shani, Lynch, Oslin, & Murphy, 2012), which is designed to inform adaptive interventions that vary in type or dosage based on individualized patient characteristics. Given varied etiologies, complex interactions of medical and behavioral factors, and assortment of available treatments, SMART-type trials are well suited to answer important clinical
questions about the pediatric fecal incontinence population. The idiographic nature of single subject designs (SSD) also provides an opportunity for study of therapeutic mechanisms. Existing research using SSD helps inform how stimulus control and operant conditioning procedures can promote toileting habits and reduce soiling (Boles, Roberts, & Vernberg, 2008; Bornstein et al., 1983; Bornstein, Sturm, Retzlaff, Kirby, & Chong, 1981; Houts, Mellon, & Whelan, 1988; Houts, & Peterson, 1986; Luxem et al., 1997; Rolider & Van Houten, 1985; O’Brien, Ross, & Christophersen, 1986). Investigation using SSD remains appropriate given that they (a) require fewer numbers of participants, (b) are flexible and allow for testing treatment components at the individual level, and (c) have scientific rigor that allows for drawing conclusions about the impact of treatments on outcome variables. Further, modern methodological (Cushing, Walters, & Hoffman, 2014) and analytic (Cohen, Feinstein, Masuda, & Vowles, 2014) approaches to SSD may increase their value in propelling the research forward, including potentially elucidating whether subgroups of youth with fecal incontinence and constipation respond differentially to various treatments.

Regardless of methodology, researchers should take care to thoroughly describe any included aspects of treatment, as well as provide a clear rationale for why each component is included. For example, Cox et al. (1996) nicely associated each of laxatives, enemas, biofeedback, and a specific behavioral intervention package (i.e., Enhanced Toilet Training) with mechanisms hypothesized to maintain constipation and fecal incontinence to justify the inclusion of each component.

Beyond study design, use of modern data analytic approaches, such as analysis of moderated mediation or latent growth curve modeling, can be used as a means of studying the interaction of personal characteristics and therapeutic processes. Such analytic approaches are largely absent from the current literature and may prove useful in better understanding the relative influence of various interventions and how outcomes are affected by multiple variables that are potentially relevant in the study of fecal incontinence.

Social Validity
In addition to better quantification of outcomes, increased study of treatment acceptability and impact is needed. Available treatments offer benefits but are also associated with costs (e.g., physical discomfort, temporary increases in incontinence, response effort, unpleasant interactions), and little is known regarding parent and child perceptions of this offset. To date, only one study has investigated treatment acceptability (Bernard-Bonnin, Haley, Belanger, & Nadeau, 1993). Additional research is necessary to better understand treatment acceptability, especially because interventions have changed dramatically since the advent and widespread use of Polyethylene Glycol 3350. Further, inclusion of patient-/parent-defined success as an outcome variable, in addition to or instead of author-defined success, will likely improve the social validity of future findings.

Conclusions
In summary, drawing conclusions from published RCTs regarding the relative benefits of behavioral treatments for fecal incontinence and constipation is challenging given the current state of the literature. Additional and improved research is needed to offer more definitive evidence of the additive role of behavioral interventions above and beyond standard medical intervention, either in isolation or in combination.

The present findings provide support that behavioral interventions positively affect certain outcome variables. We submit that, when adding conclusions from the current pooled analyses to those drawn from previous summaries of research using SSD and within-subjects designs (McGrath et al., 2000), it is reasonable to conclude that using behavioral interventions is likely to improve desired toileting behavior, rates of success, and soiling frequency. Additionally, there is ample and robust literature that interventions based on behavioral principles (e.g., reinforcement) are effective at producing change in a variety of targeted behaviors. As such, it is appropriate for clinicians treating youth with fecal incontinence to draw on the broader evidence base regarding behavioral treatments when targeting critical outcomes and to consider doing so to be consistent with evidence-based practice.

Supplementary Data
Supplementary data can be found at: http://www.jpepsy.oxfordjournals.org/

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References

*Studies marked with an asterisk (*) were retained for inclusion in MTC. Those with two asterisks (**) were retained for both the MTC analysis and the pooled outcome analysis.


