Functional defecation disorders in children
Koppen, I.J.N.

Citation for published version (APA):

General rights
It is not permitted to download or to forward/distribute the text or part of it without the consent of the author(s) and/or copyright holder(s), other than for strictly personal, individual use, unless the work is under an open content license (like Creative Commons).

Disclaimer/Complaints regulations
If you believe that digital publication of certain material infringes any of your rights or (privacy) interests, please let the Library know, stating your reasons. In case of a legitimate complaint, the Library will make the material inaccessible and/or remove it from the website. Please Ask the Library: http://uba.uva.nl/en/contact, or a letter to: Library of the University of Amsterdam, Secretariat, Singel 425, 1012 WP Amsterdam, The Netherlands. You will be contacted as soon as possible.
USING THE BRISTOL STOOL SCALE AND PARENTAL REPORT OF STOOL CONSISTENCY AS PART OF THE ROME III CRITERIA FOR FUNCTIONAL CONSTIPATION IN INFANTS AND TODDLERS

Ilan J.N. Koppen, Carlos A. Velasco-Benítez, Marc A. Benninga, Carlo Di Lorenzo, Miguel Saps

Journal of Pediatrics 2016;177:44-48.e1
ABSTRACT

Objectives: To evaluate among parents of infants and toddlers the agreement between parental report and the Bristol Stool Scale (BSS) in assessing stool consistency and the effect of both methods on determining the prevalence of functional constipation (FC) according to the Rome III criteria.

Study design: Parents of children ≤48 months of age who were seen for a well-child visit completed a questionnaire about their child’s bowel habits during the previous month. Cohen’s kappa coefficient (κ) was used to measure intrarater agreement between parental report of stool consistency (“hard,” “normal,” “soft/mucous/liquid”) and the BSS (types 1-2, hard; types 3-5, normal; types 6-7, loose/liquid). The prevalence of FC was assessed based on the questionnaire according to the Rome III criteria, comparing both methods of stool consistency assessment.

Results: Parents of 1,095 children (median age 15 months; range, 1-48) were included. Only fair agreement existed between the two methods of stool consistency assessment (κ = 0.335; P < .001). According to the Rome III criteria, using parental report the prevalence of FC was 20.5% and using the BSS the prevalence was 20.9% (P = .87). The agreement between these two methods for assessing the prevalence of FC was excellent (κ = 0.95; P < .001).

Conclusions: Only fair agreement exists between the BSS and parental report of stool consistency among parents of infants and toddlers. Different methods of stool consistency assessment did not result in a difference in the prevalence of FC.
INTRODUCTION

Functional constipation (FC) is a common defecation disorder in children that is characterized by difficult, painful, and infrequent evacuation of hard stools.¹ The prevalence of FC in the pediatric population ranges between 0.7%-29.6%.² Symptoms often occur early in life; a recent study from the United States has shown that the median age of onset of FC is 2.3 years.³

Constipation symptoms are known to have a significant impact on the quality of life of children and on health care costs.¹ Because FC is such a major pediatric health care problem occurring at a young age, it is of great importance that it be evaluated and diagnosed accurately. A correct diagnosis allows early therapeutic intervention, which is of key importance in the management of childhood FC; a delay in presentation is negatively related to recovery.⁴ Currently, the diagnosis of FC is based on the Rome III criteria, which include measures of defecation frequency, stool consistency, and other symptoms of FC (Table 1).⁵ In clinical trials of pediatric FC, outcome measures to evaluate treatment efficacy are also often based on the Rome III criteria.⁶ Although outcome measures significantly differ among clinical trials, improvement in defecation frequency (i.e., more frequent stools) or stool consistency (i.e., softer stools) are frequently used to assess treatment success.⁶,⁷ There is currently no gold standard for assessing stool consistency in young children, and various methods are used throughout the literature and in clinical practice, such as the Bristol Stool Scale (BSS), the modified BSS, the Amsterdam Infant Stool Scale, and parental or patient report of stool consistency.⁸–¹⁰ The BSS is the most commonly used standardized instrument to rate stool consistency in children, both in clinical care and in research. According to the BSS, which classifies stools into seven types, types 1 and 2 are hard and suggestive of constipation, types 3-5 are considered to be within normal range (type 4 being the most normal), and loose and liquid stools (types 6 and 7) are associated with diarrhea (Figure 1). In infants and toddlers, assessment of stool consistency is usually based on parental recall of their child’s stools, but when children get older, assessment of stool consistency is usually based on self-report. For infants and toddlers, the agreement between the BSS and report of stool consistency as communicated by the parents is unknown. The primary aim of this study was to assess the agreement between parental report of stool consistency and the BSS in children ≤48 months of age. A secondary aim was to assess the prevalence of FC in children ≤48 months of age using both methods of stool consistency assessment as part of the Rome III criteria and to evaluate the agreement between the two methods for diagnosing FC.
## TABLE 1. Rome III criteria functional constipation in children <4 years of age

<table>
<thead>
<tr>
<th>Functional constipation</th>
</tr>
</thead>
<tbody>
<tr>
<td>1. &lt;3 defecations per week</td>
</tr>
<tr>
<td>2. ≥1 episode of fecal incontinence per week after the acquisition of toileting skills</td>
</tr>
<tr>
<td>3. History of excessive stool retention</td>
</tr>
<tr>
<td>4. History of painful or hard bowel movements</td>
</tr>
<tr>
<td>5. Presence of a large fecal mass in the rectum</td>
</tr>
<tr>
<td>6. History of large diameter stools which may obstruct the toilet</td>
</tr>
</tbody>
</table>

Must fulfill ≥2 criteria for ≥1 month prior to diagnosis

---

#### FIGURE 1. Bristol Stool Scale.

METHODS

Between April and December 2015, we asked parents of children ≤48 months of age who were seen for a well-child visit in pediatric health care clinics in four different cities and/or municipalities across Colombia (Cali, Florencia, San Andrés de Sotavento, and Bogotá) to complete a survey. The survey was completed after the well-child visit and was not part of the clinical visit. By using a questionnaire in Spanish, parents were asked to answer questions about their child’s bowel habits. This survey included questions on defecation frequency, withholding behavior, painful defecation, stool consistency, and the presence of large diameter stools. Parents were asked to describe the consistency of their child’s stools during the previous month, choosing between the terms “hard or very hard,” “not too hard, not too soft (normal),” “soft or very soft,” “mucous, with undigested food,” and “liquid,” they also had the opportunity to answer that the stools were “variable” in consistency. In addition, parents were asked to choose the BSS stool type that best represented their children’s stools during the previous month on a picture chart. The BSS picture chart was accompanied by descriptors, which had been translated into Spanish. The translation of the descriptors was performed by members of the research team who represented all Colombian regions involved in this study, because dialects and minor language nuances may differ between regions. Eventually, the final translation was considered to be adequate for all regions. The questionnaire also included general questions on the medical history of the children. All children with organic conditions known to cause defecation disorders or other gastrointestinal disorders were excluded from the study. This study was approved by the Ethics Committee of Clinical Investigation at the University del Valle (Cali, Colombia).

A diagnosis of FC was based on the questionnaires according to the Rome III criteria, using either one of the two methods of stool consistency assessment (Table 1). In young infants, FC can easily be confused with infant dyschezia, a benign condition in children <6 months of age, who are otherwise healthy and do not suffer from FC, but who strain or cry ≥10 minutes before successful passage of soft stools. Therefore, we also performed a sub-analysis for children <6 months of age to assess the prevalence of infant dyschezia and the association between hard stool consistency and painful defecation in this group. Information obtained during the well-child visit, including the results from physical examinations, was not collected as part of our study, and was not used for assessing the Rome III criteria.

Statistical analyses

Data were analyzed using IBM SPSS Statistics for Windows v 22.0 (IBM Corporation, Armonk, New York). Results are shown as total numbers and proportions. Comparisons of proportions were performed using Fisher exact test or the χ² test. P < .05 was considered statistically significant. Cohen’s kappa coefficient (κ) was used to measure intrarater agreement between
parental report of stool consistency (3 categories: “hard,” “normal,” “soft/mucous/liquid”) and the BSS (three categories: type 1-2: hard; type 3-5: normal; type 6-7: loose/liquid). Parents who had answered that their child’s stools were “variable” were excluded from all analyses comparing the BSS with parental report (including the FC prevalence assessments), because they could not be assigned to any of the predefined consistency groups. Subanalyses were performed to investigate if agreement was different for different age categories. The level of agreement was determined based on the κ coefficient: 0.00 = no agreement; 0.01-0.20 = slight agreement; 0.21-0.40 = fair agreement; 0.41-0.60 = moderate agreement; 0.61-0.80 = good agreement; 0.81-0.99 = excellent agreement; and 1.00 = perfect agreement.

RESULTS

Of the 1,530 invited parents, 1,207 (78.9%) agreed to participate in this study. Eighty-eight parents were excluded because they did not complete the BSS question and another 24 children were excluded because they suffered from organic diseases known to cause defecation disorders or other gastrointestinal disorders. In total, the questionnaires from 1,095 parents (71.6%) were included. The median age of the children was 15 months (range, 1-48) and they were balanced in terms of sex (50.8% boys). Families lived in or around Cali (50.6%), Florencia (27.8%), San Andrés de Sotavento (10.9%), and Bogotá (10.8%). The results of the questionnaire on bowel habits are displayed in Table 2.

Bowel habits

Eighty-eight parents reported that their child had hard stools; these same parents reported that the BSS that best represented their child’s stools over the past month was BSS type 1-2 (hard stools, n = 36, 40.9%), BSS type 3-5 (normal stools, n = 45, 51.1%), and BSS type 6-7 (loose/liquid stools, n = 7, 8.0%; Figure 2, A). Pain during defecation was reported by 41 of 88 parents (46.6%) and 24 of 88 (27.3%) reported a defecation frequency <3 times/week.

Among the 114 parents who chose BSS type 1-2 (hard stools) as the most appropriate representation of their child’s stools over the previous month, parents reported that their child had hard stools (n = 36, 31.6%), normal stools (n = 42, 36.8%), soft/mucous/liquid stools (n = 28, 24.6%), or variable stools (n = 8, 7.0%; Figure 2, B). In this group (n = 114), pain during defecation was reported by 51 (44.7%) and 39 (31.6%) reported a defecation frequency <3 times/week.


**TABLE 2.** Results from the questionnaire on bowel habits

<table>
<thead>
<tr>
<th></th>
<th>Total</th>
<th>&lt; 2 years</th>
<th>≥2 years</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>(n=1,095), n (%)</td>
<td>(n=674), n (%)</td>
<td>(n=421), n (%)</td>
</tr>
</tbody>
</table>

**Defecation frequency**
- <3x/week: 235 (21.5)
- 3-6x/week: 79 (7.2)
- Daily: 348 (31.8)
- 2-3x/day: 377 (34.4)
- >3x/day: 56 (5.1)

**Stool consistency (parental report)**
- hard: 88 (8.0)
- normal: 484 (44.2)
- soft: 250 (22.8)
- mucous: 169 (15.4)
- liquid: 58 (5.3)
- variable: 46 (4.2)

**Stool consistency (Bristol Stool Scale)**
- BSS 1-2 (hard): 114 (10.4)
- BSS 3-5 (normal): 619 (56.5)
- BSS 6-7 (loose/liquid): 362 (33.1)

**Painful defecation**
- yes: 267 (24.4)
- no: 828 (75.6)

**Large diameter stools**
- yes: 69 (6.3)
- no: 1026 (93.7)

**Withholding behaviour**
- never: 977 (89.2)
- occasionally: 104 (9.5)
- frequently (≥1 time/week): 14 (1.3)

---

**A) hard stools (parental report)**

![Diagram A]

**B) Hard stools (Bristol Stool Scale 1-2)**

![Diagram B]

**FIGURE 2.** Differences between 2 methods of stool consistency assessment. Distribution of stool consistency assessment according to **A**, the BSS in 88 parents who reported that their child had hard stools (parental report) and **B**, parental report in 114 parents who reported that their child had stools resembling BSS 1-2 (hard stools).
A total of 260 children were reported to suffer from painful defecation, and according to parental report these children had hard (15.4%), normal (18.0%), soft/mucous/liquid (64.0%), or variable stools (2.6%). The same parents reported that their children had BSS type 1-2 (hard stools, 19.1%), BSS type 3-5 (normal stools, 56.6%), or BSS 6-7 type (loose/liquid stools, 24.3%).

A total of 290 children were <6 months of age, and 132 of these infants (45.5%) reportedly struggled, grunted, or cried before having a bowel movement. In only 9 infants (3.1%) this lasted ≥10 minutes according to parental recall (10-20 minutes in five infants and >20 minutes in four infants). To compare the effect of the BSS and parental report of stool consistency on diagnosing infant dyschezia, two children had to be excluded because parents had reported that their child’s stools were variable in consistency. Of the seven remaining infants, three of the seven fulfilled the criteria for FC according to both methods (no difference between parental report and BSS; κ = 1.00). The remaining children (1.4%) met the criteria for infant dyschezia.

A total of 60 infants <6 months of age were reported to suffer from painful defecation. According to parental report these children had hard (8.3%), normal (18.3%), soft/mucous/liquid (68.3%), or variable stools (5.0%). The same parents reported that their children had BSS type 1-2 (hard stools, 6.7%), BSS type 3-5 (normal stools, 33.3%), and BSS 6-7 type (loose/liquid stools, 60.0%).

**Agreement between parental report and the BSS**

Forty-six parents were excluded from the analyses on the agreement between the BSS and parental report of stool consistency because parents had reported that their child’s stools were of variable consistency. The remaining 1,049 parental questionnaires were analyzed and the results for both methods of stool consistency assessment were grouped together into three categories as previously described; “hard,” “normal,” and “soft/mucous/liquid” for parental report and BSS type 1-2 (hard), BSS type 3-5 (normal), and BSS type 6-7 (loose/liquid). Overall, the κ statistic was 0.335 (P < .001), indicating that only fair agreement existed between the BSS and parental report of stool consistency. Subanalysis of the different age categories showed similar results (slight to fair agreement) for all age categories: 0-12 months of age (κ = 0.292, P < .001), 13-24 months of age (κ = 0.165, P = .001), 14-36 months (κ = 0.227, P < .001), and 37-48 months of age (κ = 0.296, P < .001). Finally, when only the categories of hard stools were compared between both methods (i.e., children who fulfilled the Rome III criterion of having hard stools according to both methods), results were similar (κ = 0.31, P < .001).
Functional constipation

The prevalence of FC was assessed based on the questions regarding defecation frequency (<3 times per week), stool consistency (parental report versus BSS), painful defecation, stool withholding behavior, and large diameter stools. All parents who had reported that their child’s stools were variable were excluded from this analysis; therefore, the total number of parents was 1,049.

Applying the Rome III criteria with parental report as the method of assessing stool consistency, the prevalence of FC was 20.5% (215/1,049). Using the BSS, it was 20.9% (219/1,049); this difference was not statistically significant ($P = .87$). The agreement between these two methods of determining FC prevalence was excellent ($\kappa = 0.95$, $P < .001$). Eleven children were found to have FC according to the BSS, but not based on parental report. In contrast, seven children were found to have FC based on parental report, but not according to the BSS.

Because the Rome III criteria combine hard stools and painful defecation into one criterion, we evaluated how often stool consistency assessment played a pivotal role in diagnosing FC. Using parental report, 167 children fulfilled two of the Rome III criteria and in 26 children (15.6%) hard stools were reported; in 10 of the 26 children, defecation was reported not painful and the diagnosis was therefore dependent on the stool consistency criterion. Using the BSS, 172 children fulfilled two Rome III criteria; 36 children (20.9%) had BSS 1-2 (hard stools) and in 15 of those 36, no pain during defecation was reported; in these children, the diagnosis was dependent on the BSS.

DISCUSSION

This survey demonstrates that there is only fair agreement between the BSS and parental report of stool consistency as rated by parents of infants and toddlers. Although there is only fair agreement between the two methods, this did not affect the assessment of the prevalence of FC according to a questionnaire based on the Rome III criteria. This is most likely because the Rome III criteria encompass more criteria than stool consistency alone and because the Rome III criterion for hard and/or painful stools is a combined criterion, meaning that children fulfill this criterion if they have either hard stools or painful defecation. This decreases the impact of stool consistency alone in diagnosing FC.

It is generally assumed that hard stools lead to painful defecation. Interestingly, in this study passing hard stools did not always coincide with painful defecation according to parents; less than one-half of all parents who reported that their child had hard stools, using either
of the two methods for stool consistency assessment, reported that their child suffered from painful defecation. Painful defecation was also reported commonly in children with normal or soft to liquid stools, and this was especially true for infants <6 months of age. Hard stools and painful defecation, as reported by parents, are therefore not as strongly correlated as one might expect. From a pathophysiologic perspective, pain during defecation is traditionally seen as an important etiologic and perpetuating factor in the pathophysiology of FC, especially in young children. Painful defecation may induce stool withholding behavior.2 Stool withholding behavior in turn may lead to fecal impaction with the presence of a large fecal mass in the rectum that is difficult to evacuate.1 Therefore, painful defecation plays an important causative role in the pathogenesis of FC and, if stool consistency assessment with currently available tools does not correlate well with painful defecation, as may have been assumed previously, this should warrant careful use of stool consistency as an outcome measure for clinical trials. A recent systematic review has shown that stool consistency is frequently used as an outcome measure in children aged 0-4 years.6 However, if different methods of assessment of stool consistency do not show acceptable agreement, the value of stool consistency as an outcome measure might be questioned, especially because different studies assess this variable with different methods. Pain during defecation may represent a more clinically relevant outcome measure, especially in young children, in whom withholding behavior plays an important role in the pathophysiology of FC.

It has been suggested previously that stool consistency assessment in infants and toddlers is difficult (e.g., owing to deformation of the stools in the diapers) and that the BSS, which was developed for adults, may not be applicable in young children, especially before they have acquired toileting skills. This was the reason for the development of the Amsterdam Infant Stool Scale, with 4 categories of stool consistency based on images: hard, formed, soft, and watery.10 In a study using both the BSS and the Amsterdam Infant Stool Scale, the latter has been suggested as more appropriate for young children, because it also takes into account volume and color and is designed for evaluating children who have not yet been toilet trained (assessment is based on pictures of diapers).12 There have been no studies on the agreement between the BSS and the Amsterdam Infant Stool Scale. For the present study, we have chosen to assess the BSS and not the Amsterdam Infant Stool Scale because we assessed children <4 years of age (based on the Rome III criteria). The Amsterdam Infant Stool Scale is only validated in children <1 year of age and our study population included mostly children ≥1 year of age. Furthermore, the BSS is the most commonly used tool for stool consistency assessment worldwide and we specifically aimed to assess its clinical applicability in young children. Unlike previous studies evaluating methods of stool consistency assessment,6,10,12,13 we did not use pictures of stools to assess stool consistency for comparison between the two stool consistency evaluation methods, and our ratings
were not performed by trained professionals. Instead, we assessed intrarater agreement of two methods of stool consistency assessment among parents, based on the recall of their child’s stools. This is highly relevant for clinical practice because, in reality, parents are most often the ones providing information about their child’s stool consistency, both in clinical practice and for the purpose of clinical trials.

We hypothesized that agreement between the two methods might have been better in parents of infants compared with older children, because of the more frequent diaper care. However, a subanalysis of the different age categories showed that this was not the case. This might be owing to the lack of applicability of the BSS in infants. Therefore, it would have been interesting to see how parents would have rated their child’s stools on the Amsterdam Infant Stool Scale.

There are limitations to this study. To allow comparison between the BSS (7 items, 3 categories) and parental report (5 categories + variable), we needed to group certain answers together to enable comparison of categories. This only applied to the stools of soft to liquid consistency, which needed to be grouped together for both the BSS and parental report. We believe that because this was done for both methods, the risk of potential bias of our results is acceptable. To support this, a subanalysis of only children who fulfilled the criterion of having hard stools (according to both methods of stool consistency assessment) revealed similar results regarding the level of agreement. Furthermore, the grouping of the stool ratings could not have affected our results on the diagnosis of FC using both methods, because these analyses only involved the report of hard stools, which were not grouped differently than in their original form. Another limitation inherent to our study design was the fact that parents had the option of reporting their child’s stools as “variable.” Although this likely reflects a common occurrence in real life, we were unable to group these parents in the predefined categories and therefore they needed to be excluded. Furthermore, we “diagnosed” FC based on a questionnaire and did not include a physical examination (with a digital rectal examination), which could have resulted in more children fulfilling the Rome III criteria for FC. It is obvious that performing a physical examination with a digital rectal examination in all children without a medical indication and for research purposes only would not have been ethically acceptable in our study design. It is still important to recognize that a questionnaire is only a screening tool, and an evaluation by a physician is always necessary to actually diagnose functional defecation disorders. Furthermore, our questionnaire did not include questions about the age of toilet training and the occurrence of fecal incontinence after the acquisition of toileting skills; together with the lack of a physical examination, this may have resulted in an underrepresentation of the actual prevalence of FC, especially in the older children in our sample.
Only fair agreement exists between the BSS and parental report of stool consistency among parents of infants and toddlers. In our sample, the different methods of stool consistency assessment did not result in a difference in the prevalence of FC. Stool consistency is often used as an important outcome measure in clinical research; however, the results of this study suggest that attention needs to be exercised in attributing much importance to stool consistency. Future studies are warranted to further evaluate reasons for disagreement and to optimize stool consistency assessment for clinical and research purposes. These studies could potentially include different methods of stool consistency assessment to evaluate what the most appropriate tool for stool consistency assessment is in their specific study population. Furthermore, it is important to consider whether stool consistency is indeed the appropriate outcome measure to assess treatment success, or if other outcome measures are more relevant and more reliable. This may depend on the study population, the tested treatment modality and the study design.
REFERENCES