Paediatric constipation and functional non-retentive faecal soiling

Voskuilj, W.P.

Citation for published version (APA):
Chapter 3

Use of Rome II criteria in childhood defecation disorders: applicability in clinical and research practice

Wieger P. Voskuijl, Jarom Heijmans, Hugo S. Heymans, Jan A. Taminiau, and Marc A. Benninga

Abstract

Aim
To evaluate the prevalence of paediatric functional gastrointestinal disorders with the use of the Rome II criteria and to compare these data with the 'classic' Iowa criteria.

Methods
Patients recorded defecation and encopresis frequency. A standard history was taken and a physical examination including a rectal examination was done. The prevalence of both criteria was assessed and compared.

Results
One hundred ninety-eight consecutive patients (age: 0.66 to 15.76 years; 131 male subjects) were included. According to the Rome II criteria, 64%, 18%, and 21% of patients fulfilled the criteria for functional constipation, functional faecal retention, and functional non-retentive faecal soiling, respectively. Using the classic criteria, 74% and 18% of patients fulfilled the criteria for paediatric constipation and solitary encopresis, respectively; 16% of the patients fulfilling the paediatric constipation criteria were not recognized by the Rome II constipation criteria. Fair agreement was found between functional non-retentive faecal soiling and solitary encopresis. Encopresis was present in 79% of constipated children.

Conclusions
The Rome criteria are restrictive and exclude several children with constipation. We recommend including encopresis and rectal digital examination and excluding arbitrary age limits and retentive behaviour in the revision of the Rome criteria.
**Introduction**

Constipation in childhood is characterized by a low defecation frequency in combination with either involuntary loss of stools (encopresis), passage of large amounts of stool, retentive posturing, or hard and often painful defecation (1). In a minority of patients, encopresis is present without other symptoms of constipation (2). These children have a normal defecation frequency and normal size and consistency of stool without faecal retention on physical examination. Furthermore, these children have normal colonic transit time measured by the radio-opaque marker technique (2). In more than 90% of patients with these symptom complexes, no organic or anatomic cause can be found, and therefore these patients are considered to have a functional defecation disorder (3). On the basis of this symptomatology, strict ‘classic’ criteria have been developed to define constipation and encopresis in children (Table 1) that were useful in evaluating end points of various treatment regimens (2,4).

Recently, a group of experts in the field of paediatric gastroenterology made an attempt to set criteria for functional gastrointestinal disorders in childhood, leading to the first paediatric Rome II criteria (5). These criteria have provided clinicians with a method for standardizing their definition of clinical disorders and have allowed researchers from various fields to study the (patho-)physiology and treatment of the same disorders from different points of view (5) (Table 1). However, little is known about the applicability in clinical practice and research of the paediatric Rome II criteria. Therefore, our aims were to study and compare the prevalence of functional constipation (FC), functional faecal retention (FFR), and functional non-retentive faecal soiling (FNRFS) according to the Rome II criteria and paediatric constipation (PC) and solitary encopresis (SE) according to the classic criteria, in children referred to a tertiary centre for evaluation of defecation disorders and to assess whether the Rome II criteria and classic criteria identify the same patients.
**Methods**

Between 2000 and 2002, a group of 198 otherwise healthy children with complaints of constipation and/or encopresis (duration >3 months) was referred by general practitioners, school doctors, and paediatricians to our outpatient clinic in a tertiary academic hospital. Children with organic causes for defecation disorders, including Hirschsprung’s disease, spina bifida occulta, or hypothyroidism, were excluded from the study. Two weeks before the initial visit, all patients were sent a diary in which they were asked to record defecation and encopresis frequency. Encopresis was defined as the loss of loose stool in the underwear. Soiling was defined as staining of the underwear. However, since parents are not able to accurately estimate the amount of faeces lost in the underwear and thus can not differentiate between soiling and encopresis, we will further only use the term encopresis, defined as the loss of any quantity of faeces in the underwear (6).

To objectively study symptoms, laxative treatment had to be discontinued during the 2-week record period. At intake, all patients were subjected to a standard history and physical examination, including digital rectal examination. Form and consistency of stool were assessed through the use of standardized drawings and clay models. Rectal examination was not performed in children considered too frightened to undergo this procedure.

Sixteen items were selected to identify whether patients met the diagnostic criteria for FC, FFR, FNRFS, PC, and SE (Table 1). All data were gathered, and computer algorithms were written to generate the prevalence of FC, FFR, FNRFS, PC, and SE.
**Table 1** Different diagnostic criteria

**Classic criteria**

A. Paediatric constipation (PC) (4) = at least two of the following criteria:
   - Defecation frequency <3 times per week
   - Two or more encopresis episodes per week
   - Periodic passage of very large amounts of stool once every 7 to 30 days
   - A palpable abdominal or rectal mass at physical examination
   (The criterion of a large amount of stool is satisfied if it is estimated to be twice the standard amount of stool, shown in a clay model, or if stools are so large that they clog the toilet)

B. Solitary encopresis (SE) (2) = in a child older than 4 years of age:
   - Two or more encopresis episodes per week
   - Defecation frequency ≥3 times per week
   - No passage of very large amounts of stool
   - No palpable abdominal or rectal mass at physical examination

**Rome II criteria** (5)

A. FC = in infants and preschool children (from 1 month to 6 years) at least 2 week of:
   - Scybalous, pebble like, hard stools for a majority of stools; or
   - Firm stools ≤2 times per week; and
   - No evidence of structural, endocrine, or metabolic disease.

B. FFR = from infancy to 16 years old, a history of at least 12 week of:
   - Passage of large-diameter stools at intervals <2 times per week; and
   - Retentive posturing, avoiding defecation by purposefully contracting the pelvic floor muscles; as these muscles fatigue, the child uses the gluteal muscles, squeezing the buttocks together

C. FNRFS = once per week or more for the preceding 12 week, in a child older than 4 years, a history of:
   - Defecation into places and at times inappropriate to the social context;
   - In the absence of structural or inflammatory disease; and
   - In the absence of signs of faecal retention
Results

In a 2-year period, a total of 198 consecutive patients (66% boys) referred for functional defecation disorders were included in this study. Of the 198 children originally included in this study, 68 children were excluded from further analysis: (1) 27 patients were too frightened to undergo rectal examination, (2) parents of 18 patients were not able to reliably answer if their child avoided defecation by contracting the pelvic floor muscles purposefully, and (3) in 23 patients, the parents could not report reliably about the form or consistency of stools of their child. Statistical sub-analysis of the initial sample of 198 patients and the final sample of 130 children showed no significant differences with respect to clinical features, treatment before intake, and final diagnoses, demonstrating no selection bias. All data are based on these 130 patients (Table 2). The median time of laxative treatment before intake was 8.5 months. Only 19% of all 130 patients were never treated for constipation and had never used laxatives. Twenty-eight children were taking laxatives 2 weeks before the initial visit to our outpatient clinic; 84% of all patients had encopresis. Of all children with a diagnosis of constipation according to the Rome II criteria (i.e., FC or FFR), 79% had encopresis. Primary encopresis, defined as never having been continent for faeces, was found in 45% of the children.

Table 2 Patient characteristics of the study population (n = 130)

<table>
<thead>
<tr>
<th>Characteristic</th>
<th>Value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Male, n (%)</td>
<td>88 (68)</td>
</tr>
<tr>
<td>Mean age at intake in years (min-max)</td>
<td>7.97 (0.66–15.76)</td>
</tr>
<tr>
<td>No. of patients with positive family history (%)</td>
<td>52 (40)</td>
</tr>
<tr>
<td>Mean months of laxative treatment before intake (min-max)</td>
<td>8.5 (0–180)</td>
</tr>
<tr>
<td>Symptoms (%)</td>
<td></td>
</tr>
<tr>
<td>Defecation frequency &lt;3/week</td>
<td>43</td>
</tr>
<tr>
<td>Encopresis</td>
<td>84</td>
</tr>
<tr>
<td>Faecal retention on physical examination</td>
<td>52</td>
</tr>
<tr>
<td>Large amounts of stool once weekly/monthly</td>
<td>53</td>
</tr>
<tr>
<td>Retentive posturing</td>
<td>54</td>
</tr>
<tr>
<td>Scybalous, pebble-like, hard stools</td>
<td>38</td>
</tr>
<tr>
<td>Successfully toilet trained for faeces (%)</td>
<td>55</td>
</tr>
<tr>
<td>Mean age in years (min-max)</td>
<td>2.8 (1–8.5)</td>
</tr>
</tbody>
</table>

Constipation

The prevalence of constipation as defined by the Rome II criteria (FC or FFR) and the classic criteria was 69% and 74%, respectively. A total of 45 children in the study population were younger than 6 years; 29 of these patients (64%) fulfilled the FC criteria (mean age, 4.3 years ± 1.2; range, 1.4–5.9). Furthermore, 32
additional children older than 6 years of age also fulfilled the FC criteria (mean age, 10.0 ± 2.1 years; range, 6.5-15.3). Large, firm stools were significantly more present in the group of FC children younger than 6 years of age compared with children older than 6 years of age with FC.

Of 130 patients, only 23 patients (18%) fulfilled the criteria for FFR. The mean age of these children was 6.2 ± 3.2 years (range, 2.2-14.3). As mentioned above, the question of retentive posturing could not adequately be answered by the parents in 18 children. Of the total patient group of 130 patients with defecation disorders, 13 patients (10%) fulfilled the criteria for FC and FFR simultaneously. These children had a defecation frequency of less than 2 per week and exhibited retentive posturing and/or passed large-diameter stools once per week to once per month. A total of 74% of the patient sample fulfilled the classic criteria for PC. These patients had a mean age of 7.9 ± 3.4 years (range, 1.4-15.8).

Non-retentive soiling
Twenty-four patients (21%) fit the Rome II criteria for FNRFS (67% boys). The mean age of these FNRFS patients was 9.1 ± 2.7 years (range, 4.2-15.8). Twenty patients (18%) fulfilled the criteria for SE (60% boys). The mean age in this group of children was 8.9 ± 2.2 years (range, 4.5-12.9).

Comparison of Rome II and Classic Criteria
The Figure shows that 16% of children fulfilling the classic PC criteria were not recognized by the Rome II criteria (FC or FFR). The diagnosis of PC was based on a low defecation frequency in combination with encopresis and/or faecal retention found upon physical examination. On the other hand, 11% of patients with positive Rome II criteria (FC or FFR) were not recognized by the classic criteria. These patients had Rome II–specific items such as retentive posturing and scybalous, pebble-like, hard stools, without other criteria as mentioned by the classic criteria.

All children matching the classic criteria for SE were recognized by the FNRFS criteria. In contrast, 4 children who met the Rome II criteria for FNRFS did not meet the criteria for SE. These 4 patients defecated fewer than 3 times per week.
Patients Not Fulfilling Any of the Rome II or Classic Criteria

Ten children (8%) did not fulfill any criteria. The bowel diary showed a normal defecation frequency without encopresis. Furthermore, standard history and physical examination were without any abnormalities.

**Figure** Relation between Rome II criteria and classic criteria in describing constipation prevalence in the cohort.
Discussion

This study prospectively compared the prevalence and applicability of the paediatric Rome II criteria and classic criteria in a group of children referred to a tertiary centre for defecation disorders. According to the paediatric Rome II criteria, 64%, 18%, and 21% of patients fulfilled the criteria for FC, FFR, and FNRFS, respectively, whereas 74% and 18% fulfilled the classic criteria (2,4) for PC and SE, respectively. Evaluation of the total prevalence of constipation according to both criteria showed acceptable similarity. Despite this, 16% of the patients fulfilling the PC criteria were not recognised as having FC or FFR. On the other hand, 11% of the children with a positive Rome II diagnosis were not recognized by the classic criteria.

Approximately 10% of the patients did not fulfil any of the Rome II or classic criteria. Their parents considered them constipated because they had no daily defecation. However, their bowel diary showed a stool frequency >2 per week, without any other criterion for constipation. More than 60% of the public believes that one bowel movement each day is necessary for good health (7). Moreover, different views and the absence of universally accepted definitions may give rise to misinterpretations between patients and doctors.

Using the classic and Rome II criteria, encopresis was the most frequent accompanying symptom (84%) of childhood constipation. This finding is in accordance with several other studies describing symptoms of childhood constipation (8-10). Encopresis is well-recognized and easy to score by the parents and is therefore useful as an objective marker for the severity of constipation and its effectiveness of treatment (6). Moreover, in large clinical studies, encopresis was one of the criteria for success (11-14). The current Rome II criteria lack encopresis as inclusion criterion for constipation and is only mentioned as an accompanying symptom. To better understand the pathophysiology of constipation, to enhance treatment monitoring, and to allow comparison of the outcome of different treatment regimens, it might be helpful to include encopresis in the next Rome criteria.

Functional constipation was the most common disorder in this study (64%), whereas the prevalence of functional faecal retention was low (18%). A similar outcome was reported by Loening-Baucke in children referred for functional encopresis (15). The low incidence of FFR might reflect the subjectivity of recognizing retentive posturing. Parents are often not able to reliably report whether their child exhibits stool-withholding behaviour. Moreover, an accurate history strongly depends on question
formulation and interpretation by the physician and parents. We agree, however, that retentive posturing is an important item of childhood constipation. It might be useful as an inclusion criterion in the next Rome criteria only if more attention is paid to precise questioning and scoring of this subjective symptom.

Faecal retention in children with constipation is mainly characterized by the accumulation of faeces in the rectum. This can be assessed either by abdominal radiograph or by rectal digital examination; the latter is often considered as too invasive, unethical, and negatively influences treatment compliance and outcome (16). For only a minority of children in this study (14%) did we decide not to perform a rectal examination because these children were considered to be too anxious. In 18 children (14%), faecal retention was only detected by rectal digital examination. These children had no other symptoms of constipation and no palpable faecal mass at abdominal examination. With the use of the Rome II criteria, 16 children (12%) would not have been recognized as constipated patients, since rectal examination is not mandatory. In our practice, however, we use this procedure as an important diagnostic tool. In children with a large rectal faecal mass, we usually start with a 3-day enema clean-out before starting oral laxatives (17). In children without rectal faecal retention, we only start with oral laxatives.

Of our total patient sample, approximately 50% fulfilled the criteria for functional constipation. Of this group, 32 patients were older than 6 years of age. According to the Rome II criteria, these patients should not be classified as FC, as the age limit was 6 years or younger. Surprisingly, only the occurrence of large amounts of stool varied between FC patients younger and older than 6 years of age. We have no clear explanation for this finding because no significant difference was found in defecation frequency between both groups. Because both the onset and the cause of childhood constipation are variable and are not fixed to certain age limits, we suggest excluding the age limit for the next Rome criteria.

A small subgroup of children fulfilled the FC and FFR criteria simultaneously. Both diagnoses contain “firm or large-diameter stool, ≤2/week,” whereas the diagnosis FC does not exclude “retentive posturing.” This overlap could be avoided by incorporating the exclusion of the symptom “retentive posturing” to the diagnosis FC.

In accordance with earlier studies, FNRFS was found in a minority of patients (21%) (9). These children defecate ≥3 times per week on the toilet and have no other symptoms of constipation. More importantly, and in contrast with children
with retentive soiling, they have no abnormalities on rectal examination. Therefore, it is essential to perform a rectal examination in these children to differentiate between retentive and non-retentive soiling. Children fulfilling the clinical criteria of FNRFS undergo radio-opaque marker studies to measure colonic transit time. Subsequently, if colonic transit is within the normal limit, a strict toilet training program is started, sometimes in combination with behaviour therapy. We suggest including a defecation frequency of $\geq 3$ times per week and no abnormalities on rectal examination as important criteria in the next Rome criteria for FNRFS.

In conclusion, although the current Rome II criteria mark an important first step in defining functional defecation disorders, they are difficult to use in clinical practice. Furthermore, these criteria do not supply a sufficient tool to make studies more comparable to select patients for clinical trials with respect to physiology and treatment outcome. These criteria are restrictive and exclude many children with constipation. Encopresis, a major feature of constipation, should be included in the next Rome criteria. Although rectal examination is an additional examination, it is usually well tolerated and important to differentiate between faecal retention and FNRFS. We therefore recommend including this important item in the Rome III criteria. International collaboration is necessary to develop and validate standard constipation questionnaires in which difficult items as large amounts of stool, consistency of stool, and retentive posturing are defined.
References


70