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## Hirschsprung's disease: early diagnosis and long-term outcomes

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## CHAPTER 6

# Reproducibility, feasibility, and validity of the Groningen Defecation and Fecal Continence questionnaires

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# SUMMARY

## **Background**

Current questionnaires on defecation disorders are often brief and fail to include questions considering causative factors. Furthermore, adult and pediatric questionnaires differ, which makes it impossible to monitor defecation disorders during the transition from childhood to adulthood. With these points in mind we developed the Groningen Defecation and Fecal Continence (DeFeC) questionnaire and its pediatric equivalent, the P-DeFeC. The aim of this paper is to introduce the questionnaires and to assess their feasibility, reproducibility, and validity.

## **Methods**

Various Rome IV criteria and scoring tools for constipation and fecal incontinence were incorporated, resulting in nine categories. Feasibility and reproducibility were assessed by performing a test-retest survey in 100 adult participants. Concurrent validity was assessed in 27 patients and 18 healthy volunteers by comparing questionnaire-based diagnoses of constipation and fecal incontinence to final diagnoses based on anorectal function tests.

## **Results**

There were no remarks on the understandability of any questions. The Cohen's kappa coefficient of all main questions ranged from 0.26 to 1.00, with an average of 0.57. All but one category showed moderate agreement or higher. The sensitivity of the questionnaire-based diagnosis of constipation was 75%; specificity was 100%. The sensitivity of the questionnaire-based diagnosis of fecal incontinence was 77%; specificity was 94%.

## **Conclusions**

Overall reproducibility of the Groningen DeFeC questionnaire is acceptable and its validity is good. This makes it a feasible screening tool for defecation disorders and, equally important, with these questionnaires defecation disorders can now be monitored during the transition from childhood to adulthood.

# INTRODUCTION

Fecal disorders, such as constipation and fecal incontinence, are among the most common medical disorders. In the general population the prevalence of constipation is 16%<sup>1</sup> and for fecal incontinence it is 8%.<sup>2</sup> Despite their high rates of prevalence, constipation and fecal incontinence are not yet clearly defined, because the symptoms vary widely among patients and are often ambiguous. This explains why diagnosing constipation and fecal incontinence, and finding the causative factors, remains difficult.

Ambiguity regarding the definitions of defecation disorders led to the development of the Rome criteria<sup>3,4</sup> and different questionnaires, each with their own scoring systems.<sup>5-19</sup> To our knowledge, even though constipation and fecal incontinence are known to coexist, only one of these scoring systems incorporates both disorders.<sup>5</sup> Moreover, only a few questionnaires include questions relating to factors that may influence defecation disorders, such as diet and therapy,<sup>7-12</sup> and only one questionnaire includes questions relating to other causative factors for constipation and fecal incontinence, such as obstetric history, medicine use, or neurological illnesses.<sup>6</sup> Finally, and most importantly, all the questionnaires that are currently available are designed separately for children or adults. This makes it impossible to determine whether symptoms of constipation and fecal incontinence in pediatric patients change during the transition from childhood to adulthood, even though continuity of care during this transitional phase is an important issue.

Keeping all these points in mind we developed the Groningen Defecation and Fecal Continence (DeFeC) questionnaire, a comprehensive questionnaire that contains questions on anorectal functioning, associated disorders, and causative factors and its pediatric equivalent, the Pediatric DeFeC questionnaire (P-DeFeC). We based the questionnaires on the adult Rome IV criteria<sup>3,4</sup> for constipation and fecal incontinence, along with several validated constipation and fecal continence scores.<sup>10-14</sup> The aim of this paper is to introduce the two versions of the questionnaire and to explain their contents. In addition, we aimed to assess the reproducibility, feasibility, and validity of the adult version of the DeFeC questionnaire.

## METHODS

### **Design of the Groningen DeFeC questionnaire**

The Groningen DeFeC questionnaire was modeled on the Rome IV questionnaire,<sup>20</sup> the Rome IV criteria,<sup>3,4</sup> and various validated scoring tools.<sup>10-14</sup> We recruited patients, fellow physicians, and acquaintances to review the draft questionnaire and we obtained their

remarks on the understandability of the questions and of the language used in the questionnaire. The final version of the questionnaire consisted of 88 questions in nine different categories that covered various aspects of anorectal functioning, associated disorders, and causative factors (Table 1, and see the appendices for the complete version of the questionnaire). All the questions relate to a patient's bowel habits during the last six months. Below, we briefly explain the contents of the categories of the questionnaire:

0. Prior to the questions on bowel habits, we included several questions on demography. These questions were included primarily to collect reference data from the general population. They also allow future matching and comparisons between the patient data to the data of healthy peers. These questions do not have to be completed for the calculation of the various Rome IV criteria and scoring tools.
1. Defecation pattern was assessed by frequency and stool consistency. The options for defecation frequency ranged from less than once a month to more than five times a day. Stool consistency was defined in accordance with the stool types as described in the Bristol stool form scale.<sup>21</sup>
2. Constipation-related aspects were investigated more thoroughly than is done in current questionnaires. Patients were first asked whether they have difficulty emptying their bowels and, if so, how long had they suffered from these symptoms. The following questions allowed us to determine whether the patients fulfilled the Rome IV criteria for functional constipation. Dyssynergic defecation was assessed by inquiring about the most characteristic symptoms: excessive straining, duration of strain, anal blockage, incomplete defecation, failure to defecate, and anal pain. Because symptoms of constipation are often associated with abdominal bloating and pain, we added questions specifying abdominal discomfort.
3. A patient's daily diet is known to play a role in constipation.<sup>22</sup> Inquiring about diet can be troublesome and should preferably be addressed by using a dietary diary. Processing such a diary, however, is time-consuming – both for the patient and the physician. Instead, we decided to screen patients' dietary habits by asking four questions about intake of liquids and consumption of fruits, vegetables, and whole-wheat bread. Using various remedies to relieve constipation is common practice among patients with constipation problems. We therefore added detailed questions on the use of laxatives, as well as questions relating to other constipation remedies, such as rectal suppositories and rectal irrigation. Lastly, we added a question on help-seeking behavior for constipation symptoms.
4. Fecal incontinence was assessed by multiple questions relating to different types of incontinence, such as soiling, incontinence for solid stool, liquid stool, and

flatus. A positive answer to one of the aforementioned questions was followed by more detailed questions relating to severity and frequency of fecal incontinence. Other questions regarding incontinence related to the use of stool-hardeners, pads, rectal irrigation, and changes in daily activities. Finally, we asked patients about help-seeking behavior regarding their incontinence symptoms.

5. Both the ability to feel the urge to defecate and the ability to postpone the passage of stools play an important role in fecal continence. We asked patients whether they feel the urge to defecate before going to the toilet and whether they can differentiate between different kinds of stools and flatus. We also asked how long

**Table 1**

Contents of the Groningen DeFeC questionnaire

Categories	Questions	No. of questions
Demographic information	General information such as height, weight, gender, age, residence, and daily occupation	8
Defecation pattern	Defecation frequency, stool consistency	2
Constipation complaints	Difficulties passing stool, incomplete or failure to defecate, anal pain, bloating, and abdominal discomfort/pain	16
Constipation-related remedies	Use of diet, laxatives, and/or more invasive therapies	14
Fecal continence	Different types of incontinence ( <i>i.e.</i> soiling, solid, liquid, gas), time of incontinence, and incontinence related therapies	16
Anorectal sensation and voluntary contractions	Urge to defecate, ability to hold stool, ability to differentiate between various stool types.	4
Urinary continence	Urination frequency, straining during urination, urinary incontinence, time of incontinence, nocturnal urination, and urinary tract infections.	9
Obstetric and gynecologic history	Obstetric history and complications, gynecologic surgical history, and prolapse complaints	11
Pelvic floor-related medical history	Bowel surgery history, presence of blood or slime in stools, medical conditions affecting bowel movements, and overall medication use	8
Total:		88

they are able to postpone the passage of stools and how often they have to hurry to reach the toilet in time.

6. Fecal and urinary incontinence frequently coexist, especially in the elderly and nursing home residents.<sup>23</sup> For this reason we also included several questions on urinary continence and urinary tract infections.
7. For female respondents we included a category on obstetric and gynecologic medical history. We included detailed questions on the number of deliveries, types of delivery, and complications during delivery.
8. The last category of the Groningen DeFeC questionnaire inquired about the patient's medical history regarding bowel surgery, somatic bowel diseases, and other diseases that could influence anorectal physiology.

### **Pediatric version of DeFeC questionnaire**

The P-DeFeC questionnaire is the pediatric equivalent of the Groningen DeFeC questionnaire (see the supplementary files for the complete version). It contains almost the same questions and criteria as the adult version, but is more simply worded, thus making it understandable for a child of eight years or older. Questions on educational level, work, and obstetric history were removed as such questions would be irrelevant to most children. The pediatric version therefore consisted of 79 questions in eight different categories instead of the 88 questions in the adult version.

### **Completing the questionnaires**

Both versions of the questionnaire are self-administered and can be completed either online or on paper. The questionnaires were designed in such a way that patients would not be asked questions that were irrelevant to them. This is determined by positive or negative answers given at the beginning of certain categories. For instance, if patients reported never to experience abdominal pain, they could skip the subsequent questions on abdominal pain. In this way an adult male respondent who does not suffer from either constipation or fecal incontinence only needed to answer 55 out of the total set of 88 questions.

### **Assessing defecation disorders**

The Groningen DeFeC questionnaire includes questions that comply with several adult Rome IV criteria, as well as validated constipation and fecal incontinence scores. Firstly, we defined constipation in accordance with the Rome IV criteria for functional constipation<sup>3</sup> and we assessed its severity with the Constipation Scoring System as described by Agachan and colleagues.<sup>10</sup> In addition, we included a disease-specific

score for obstructed defecation syndrome by Renzi and colleagues.<sup>12</sup> We defined fecal incontinence in accordance with the Rome IV criterion for fecal incontinence, which is recurrent uncontrolled passage of fecal material.<sup>4</sup> We assessed the severity of fecal incontinence in terms of the Continence Grading Scale as described by Jorge and Wexner<sup>13</sup> and the Vaizey incontinence score.<sup>11</sup> Lastly, fecal incontinence following lower anterior resection surgery can be assessed with the lower anterior resection score developed by Emmertsen and Laurberg.<sup>14</sup>

The P-DeFeC questionnaire consisted of the same criteria and scores for constipation and fecal incontinence as the adult version. This enabled us to compare defecation disorders during the transition from childhood to adulthood. Additionally, in contrast to the adult Rome IV criteria for constipation, the pediatric Rome IV criteria required a psychological examination.<sup>24</sup> Because this questionnaire was designed as a screening tool, physical examination is not possible. For these reasons we decided to incorporate the same adult Rome IV criteria for constipation and fecal incontinence in the P-DeFeC questionnaire as in its adult equivalent.

### **Questionnaire translation**

Both versions of the DeFeC questionnaire were translated from Dutch to English and back to Dutch to ascertain that the contents of the questionnaires had not changed in the translation process. First, a bilingual translator, whose mother tongue was English and who had no knowledge of the subject, translated the questionnaire from Dutch to English. Subsequently, the questionnaires were translated back to Dutch by another bilingual translator whose mother tongue was Dutch and who also had no knowledge of the subject. The agreement of the contents between the Dutch original and the English-back-to-Dutch translation were analyzed. Any discrepancies were documented, discussed, resolved, and adjusted accordingly in the English version of the questionnaire. In this way we established that to all intents and purposes the contents of the English translation of both versions of the DeFeC questionnaire were the same as the Dutch original. A few items, such as the question on educational level and the four questions on dietary habits, were tailored to Dutch standards. A minimal cross-cultural adaptation is therefore required before the questionnaires can be used in other countries.

### **Reproducibility**

To test the reproducibility, that is the replicability of the instrument, we performed a test-retest survey by asking 100 adult participants to complete the Groningen DeFeC questionnaire twice, with a one-month interval in between. The participants were recruited from the general Dutch population by an external company specialized in



conducting surveys (Survey Sampling International, Rotterdam, the Netherlands). The test-retest survey was only performed in adults as we found it unethical to subject children to this survey twice.

### **Feasibility**

The initial draft was modified on the basis of the remarks made by the patients, physicians, and acquaintances in such a way that the questions and answers were understandable to someone with a basic level of education. At the end of the survey, in order to test feasibility, the adult participants of the test-retest survey were given the opportunity to remark on the contents of the questionnaire and their ability to complete the questionnaire. We also assessed the time required to fully complete the P-DeFeC and DeFeC questionnaires using data obtained from previously performed studies in the general population.<sup>25,26</sup>

### **Validity**

In order to test validity, that is the degree to which the questionnaire truly measures the construct it claims to measure,<sup>27</sup> we analyzed the sensitivity and specificity of questionnaire-based diagnoses using anorectal function tests as the gold standard. The anorectal functions tests were only performed in adults as we found it unethical to subject children to these tests for the purpose of validating our questionnaire.

The questionnaire-based diagnosis of constipation was based on the Rome IV criteria for functional constipation,<sup>3</sup> whereas the balloon evacuation test was used as the gold standard for constipation. After the balloon evacuation test, constipation was defined as the inability to pass a rectal balloon filled with a minimum of 50 mL within two minutes.<sup>28</sup> The questionnaire-based diagnosis of fecal incontinence was based on the Rome IV criteria for fecal incontinence,<sup>4</sup> whereas the rectal infusion test was used as the gold standard. After the rectal infusion test, fecal incontinence was defined by involuntary leakage of liquid following the infusion of 1000 mL of water at body temperature.<sup>29,30</sup>

We measured the ability to correctly identify positives by assessing the sensitivity in patients who were unable to expel the rectal balloon (that is who suffered from constipation) or who experienced leakage during the rectal infusion test (that is who suffered from fecal incontinence). We measured the ability to correctly identify negatives by assessing the specificity in healthy adult volunteers who expelled the rectal balloon effortlessly (that is who did not suffer from constipation) or who experienced no leakage (that is who did not suffer from fecal incontinence).

## Statistics

The data were analyzed using SPSS 23.0 for Windows (IBM SPSS Statistics, IBM Corporation, Armonk, NY). After the test-retest survey we analyzed the reproducibility of the questions of the questionnaire by calculating the weighted Cohen's kappa ( $\kappa$ ) coefficient for questions with multiple categorical response options and the unweighted  $\kappa$  coefficient for questions with a 'yes' or 'no' response option. We analyzed the reproducibility of criteria for functional constipation and fecal incontinence, based on the Rome IV diagnostic criteria, by using the unweighted  $\kappa$  coefficient. The reproducibility of continuous defecation scores, such as the Constipation Scoring System and the Continence Grading Scale, was analyzed by using the intraclass correlation coefficient (ICC).

The calculated  $\kappa$  coefficients and ICC values ranged between -1 and 1, where a negative value indicates poorer than chance agreement. Positive values below 0.2 indicate slight agreement, values between 0.21 to 0.40 indicate fair agreement, values between 0.41 to 0.60 indicate moderate agreement, values between 0.61 to 0.8 indicate substantial agreement, and values between 0.81 to 1.0 indicate almost perfect agreement.<sup>31</sup>

## RESULTS

### Reproducibility

One hundred adult individuals participated in the test-retest survey, consisting of 50% ( $n = 50$ ) female respondents and 50% ( $n = 50$ ) male respondents. Respondents' median age was 43 years, with a minimum of 19 years and a maximum of 65 years. Based on the

**Table 2**

Reproducibility of the adult version of Groningen DeFeC questionnaire

Category	$\kappa$ coefficient*	Interpretation**
Defecation pattern	0.48	Moderate
Constipation complaints	0.54	Moderate
Constipation-related remedies	0.73	Substantial
Fecal continence	0.39	Fair
Anorectal sensation and voluntary contractions	0.44	Moderate
Urinary continence	0.60	Moderate
Obstetric and gynecologic history	0.81	Almost perfect
Pelvic floor-related medical history	0.50	Moderate
Average:	0.57	Moderate

\* Weighted Cohen's kappa ( $\kappa$ ) coefficient

\*\* Interpretation of  $\kappa$  coefficients according to Landis and Koch<sup>31</sup>

primary survey, 20% (n = 20) of the respondents were eligible for the Rome IV criteria of functional constipation, while 7% (n = 7) were eligible for the Rome IV criteria of fecal incontinence.

When analyzing reproducibility of all the main questions of the adult version of the Groningen DeFeC questionnaire we found that the weighted  $\kappa$  coefficient ranged from 0.26 to 1.00, with an average of 0.57, indicating a moderate agreement for the entire questionnaire (Table 2). Additionally, the average weighted  $\kappa$  coefficients for each separate category of the Groningen DeFeC questionnaire were calculated and ranged from 0.39 to 0.81 (Table 2). All but one category showed moderate agreement or higher.

We also analyzed the reproducibility of scoring the Rome IV criteria for functional constipation and fecal incontinence (Table 3). The unweighted  $\kappa$  coefficient for functional constipation was 0.41 (95% CI, 0.18 to 0.64), indicating moderate agreement. The coefficient for fecal incontinence was 0.26 (95% CI, -0.08 to 0.60), indicating fair agreement. Finally, we analyzed the reproducibility of two validated scores for severity of constipation and fecal incontinence, that is for the scores obtained with the Constipation Scoring System and Continence Grading Scale. Both scores showed substantial agreement, 0.73 (95% CI, 0.63 to 0.81) for the Constipation Scoring System and 0.64 (95% CI, 0.51 to 0.74) for the Continence Grading Scale (Table 3).

### Feasibility

All 100 adult participants in our test-retest survey fully completed the questionnaire twice. There were no remarks on the understandability of any of the items. On average it took less than 11 minutes to complete the DeFeC and 8 minutes for P-DeFeC.

**Table 3**  
Reproducibility of criteria and scores

Rome IV criteria	Reproducibility		Interpretation**
	$\kappa$ coefficient*	95% CI	
Functional constipation	0.41	(0.18 to 0.64)	Moderate
Fecal incontinence	0.26	(-0.08 to 0.60)	Fair
Scores	ICC	95% CI	Interpretation**
Constipation Scoring System <sup>10</sup>	0.73	(0.63 to 0.81)	Substantial
Continence Grading Scale <sup>13</sup>	0.64	(0.51 to 0.74)	Substantial

\* Unweighted Cohen's kappa ( $\kappa$ ) coefficient

\*\* Interpretation of  $\kappa$  coefficients according to Landis and Koch<sup>31</sup>

## Validity

For the assessment of validity we included 27 referred adult patients, 74% (n = 20) of whom were women and 26% (n = 7) were men. The patients' median age was 55 years and ranged from 18 years to 75 years. We only included patients who suffered from constipation (n = 8), and/or from fecal incontinence (n = 22), based on anorectal function testing. Additionally, we included 18 healthy volunteers, 56% (n = 10) of whom were women and 44% (n = 8) were men. The healthy volunteers' median age was 22 years and ranged from 19 years to 26 years. We only selected healthy volunteers who did not suffer from constipation and fecal incontinence based on anorectal function testing.

The sensitivity of the questionnaire-based diagnosis of constipation was 75% and specificity was 100% (Table 4). Likewise, the sensitivity of the questionnaire-based diagnosis of fecal incontinence was 77% and specificity was 94% (Table 4).

## DISCUSSION

We developed the Groningen DeFeC questionnaires using existing criteria and validated scoring tools. It contained questions about anorectal functioning and associated disorders, such as constipation, fecal incontinence, and their combined forms. Because of their extensive range, which included urologic and gynecologic questions, the Groningen DeFeC questionnaires are a feasible tool to not only screen for defecation disorders, but to also determine causative factors. The questionnaires can be used to assess the adult Rome IV criteria for fecal incontinence and constipation,<sup>3,4</sup> as well as several fecal incontinence and constipation scores.<sup>10-14</sup> This makes the Groningen DeFeC questionnaire an excellent tool of choice in day-to-day clinical practice and for scientific studies. Importantly, this questionnaire has a pediatric version that consists of the same questions and scores. This provides the possibility to compare defecation disorders before and after the transition from childhood to adulthood.

To assess whether this questionnaire is understandable and easy to complete, whether it is reliable, and whether it measures what it claims to measure we analyzed

**Table 4**

Diagnostic accuracy of questionnaire-based diagnoses

Rome IV criteria	Sensitivity		Specificity	
	No.	%	No.	%
Functional constipation	6/8	75	18/18	100
Fecal incontinence	17/22	77	17/18	94

its feasibility, reproducibility, and validity in adult participants. For ethical reasons we did not perform the validation process in children. Nevertheless, we expect the outcomes to be comparable, because the questions are almost the same albeit that they were worded more simply than in the adult version of the Groningen DeFeC questionnaire.

Regarding feasibility, the questionnaire proved easy to understand and easy to complete. This was illustrated by our trial runs, such as this study and other studies in samples of the Dutch general population<sup>25,26</sup> that yielded only a small number of remarks from the participants, none of which concerned the understandability of the questionnaire. Although the questionnaire consisted of a wide range of questions, on average it only required 11 minutes to complete the adult version of the questionnaire and 8 minutes to complete the pediatric version. This also indicates that the questions were easy to understand and easy to answer. The relatively short time required to complete the questionnaires was partially the result of allowing patients to skip questions that are irrelevant to them, based on positive or negative answers given at the beginning of certain categories.

A test of reproducibility showed that the agreement between the two surveys was moderate, with all but one category in the questionnaire showing moderate agreement or higher. We point out that the  $\kappa$  coefficients are reduced due to the low frequency of positive responses in certain categories.<sup>32</sup> This was the case in the category on fecal incontinence especially, which only applied to seven of the 100 respondents. This low number of positive responses may have resulted in a lower  $\kappa$  value and agreement for this specific category. This was also reflected by a mere fair agreement of the Rome IV criteria of fecal incontinence. Moreover, the Rome IV criteria for functional constipation showed moderate agreement, while the scores for both severity of constipation and fecal incontinence showed substantial agreement.

The validity of the questionnaire was good as indicated by a relatively high specificity and sensitivity. This means that the questionnaire can be used to either exclude or confirm whether a patient is suffering from constipation or fecal incontinence-related symptoms. Due to the absence of a gold standard for constipation and fecal incontinence, we chose to use the balloon expulsion and rectal infusion tests for constipation and fecal incontinence, respectively, because these tests are performed routinely in our Anorectal Physiology Laboratory. These tests are also used by other medical centers to investigate these defecation problems.<sup>28-30</sup> Nevertheless, it remains difficult to establish a gold standard for constipation and fecal incontinence, because both disorders consist of a range of symptoms, often reported by the patient, and thus difficult to objectify by means of a test or by the physician. We are confident, however, that the balloon expulsion test can discriminate between severely constipated and healthy individuals, while the rectal

infusion test will easily discriminate between individuals with severely impaired fecal continence and those with perfect fecal continence.

### **Conclusions**

The Groningen DeFeC questionnaires are comprehensive questionnaires that contain questions on anorectal functioning, associated disorders, and causative factors. Importantly, these questionnaires provide the possibility to monitor symptoms and treatment efficacy of defecation disorders during the transition from childhood to adulthood. The adult version of the Groningen DeFeC questionnaire is well understood by the respondents, its reproducibility is acceptable, and its validity is good. Some cross-cultural adaptations will be necessary to extend the use of the questionnaire to other countries, and further use of the questionnaire is necessary to extend its validation in patient populations. Recently, we administered the questionnaire to a sample of the general Dutch population with the purpose of establishing norms to be used in future clinical studies.<sup>26</sup> We therefore encourage fellow researchers to also use the Groningen DeFeC questionnaire, or its pediatric equivalent, in clinical studies on fecal disorders.

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