Paediatric constipation and functional non-retentive faecal soiling
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Chapter 8

Functional Non-Retentive Faecal Soiling in children: 12 years of longitudinal follow-up

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Submitted
Abstract

Background
Functional non-retentive faecal soiling (FNRFS), encopresis in the absence of signs of faecal retention, is a frustrating phenomenon in children and difficult to treat. It is assumed that FNRFS will resolve spontaneously beyond puberty, however no data on long-term outcome are available.

Aim
To investigate the very long-term outcome of FNRFS patients after intensive medical treatment.

Methods
Between 1990 and 1999, 119 patients (96 boys) with FNRFS were enrolled in 2 prospective, randomised trials investigating the effect of biofeedback training and/or laxative treatment. Thereafter, follow-up (FU) was performed at 6 months, 1 year and thereafter annually until the end of data collection in September 2004. A standardised questionnaire was used, either during clinical visit or by telephone, to evaluate symptoms. Success was defined as having less than 1 encopresis episode in 2 weeks while not using medication for more than 1 month.

Results
Median age at entry (25th-75th percentiles) was 9.2 (7.9-11.6) years and the median duration of symptoms before intake (25th-75th percentiles) was 4.4 (3.0-6.7) years. A 90% follow-up was achieved at all stages of the study. After 2 years of intensive behavioural and medical therapy, 33 out of 112 (29.5%) patients were successfully treated. The cumulative success percentage after 7 years of FU was 80%. At the biological ages of 12 and 18 years, 49.4% (40/81) and 15.5% (9/58) of the patients still had encopresis, respectively. Age at intake younger than 6 years in combination with secondary encopresis was associated with a lower chance of achieving success (HR: 0.51 (95% CI: 0.27-0.98), P=0.04). Relapse occurred in 37% of patients (cumulative percentage after 7 years), and occurred most likely in the first two years after an initial success.

Conclusions
Only 29% of the patients with FNRFS are successfully treated after two years of
intensive treatment. Thereafter, a steady increase in success is observed. Nevertheless, at the age of 18 years, 15% still have encopresis.

**Introduction**

Encopresis, the involuntary loss of faeces in the underwear after the age of four is a frustrating symptom. Despite its high prevalence, 1-2% in otherwise healthy school children, a first visit to the paediatrician is frequently delayed because of shame and cultural taboos concerning encopresis (1). In more than 80% of the patients, encopresis is the result of constipation. Prolonged faecal stasis results in the involuntarily loss of faeces in the underwear several times per day and in severe constipation even during the night. These patients are best treated with oral and/or rectal laxatives.

In approximately 20% of the children with encopresis, the faecal loss exists without any sign of faecal retention and these patients are classified having functional non-retentive faecal soiling (FNRFS) (2-4). 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 encopresis, they have no faecal retention on abdominal and rectal examination. Furthermore, these children have normal colonic transit times and normal anorectal manometry results (3,5-7).

Recently, a prospective long-term follow up study in children with constipation showed that, in contrast to general belief, constipation continued beyond puberty in 30% of the constipated children (8). To date, no data exist concerning the long-term prognosis of patients with FNRFS. Since 1990 we yearly contacted our cohort of children with FNRFS who participated in two randomised controlled trials evaluating the effect of biofeedback training and / or laxatives. In the present study we aimed to investigate: 1) if FNRFS resolved during puberty, and 2) if there are clinical factors associated with the achievement of clinical success and with relapse after an initial successful period.
Methods

All FNRFS-patients enrolled in the present study participated in two randomised trials performed in our department between 1990 and 1999. The first study compared the effect of laxatives and additional biofeedback training (BF) to laxative treatment alone (5). During a 6-week treatment program all patients (N=71) received Lactitol (5 gram/10 kg bodyweight), whereas half of the children were randomly assigned to receive 5 additional sessions of BF.

The second study investigated the effect of biofeedback training with additional laxative treatment compared to BF alone (6). During a 7-week program all patients (N=48) underwent 5 BF sessions, whereas half of the children received additional Lactulose (5 gram/10 kg bodyweight). In addition to laxative therapy and/or BF training, therapy in both studies consisted of a protocol including education about encopresis, a high fibre diet and the fill in of a bowel diary. Motivation was enhanced by praise and small gifts.

Patients in both studies (N=119) were referred to our tertiary gastro-intestinal motility centre by general practitioners, paediatricians, psychiatrists, and school doctors. At intake all patients fulfilled the criteria for FNRFS, defined as: two or more encopresis episodes per week (for a period of at least one week in the preceding 12 weeks) with no signs of constipation: a defecation frequency of ≥3 per week, no periodic passage of very large amounts of stool at least once every 7–30 days and no palpable abdominal or rectal mass on physical examination (2). Encopresis was defined as the voluntary or involuntary loss of loose stool in the underwear after the age of 4 years (9). All children included in both treatment studies were older than 4 years of age in order to understand the BF procedure. Children with constipation and patients with organic causes of faecal incontinence such as Hirschsprung's disease, muscle disorders, prior recto-anal surgery, spina bifida (occulta), and mental retardation were excluded.

Data collection and Follow-up

After the end of the two randomised trials, all patients were enrolled in our long-term registry of follow-up (FU). In this long-term outcome registry we actively contacted our former patient cohort in order to monitor success and relapse. Children were contacted 6, 12 and 18 months after the end of the initial studies and thereafter annually until September 2004. A standardised questionnaire was
used for FU to evaluate symptoms, during an out-patient visit or by telephone when the patient had been discharged. Data about defecation- and encopresis-frequency, and the use of medication were collected. Children who (still) had encopresis were seen regularly at our outpatient clinic. Successful outcome ('clinical success') was defined as having less than 1 encopresis episode in 2 weeks while not using medication, such as loperamide for at least one month (8,9).

Statistical analysis
Baseline characteristics of the cohort were analysed in a descriptive way. The frequency and timing of first success and relapse was presented in a Kaplan Meier-curve without adjustment for the discrete nature of the FU. To gain insight into the clinical characteristics that are associated with clinical outcome during FU we set up two different analyses.

Prognostic factors for success and relapse after initial success
In the first model, prognostic factors for the occurrence of first success were analysed ('success model'). We used a complementary log-log regression model to examine the relation between predefined baseline characteristics and the probability of a good clinical outcome (10). This regression model assumes that a continuous time, proportional hazards model has generated the underlying observations, but because we observed only data grouped by 6 monthly or yearly intervals we used a discrete hazard model to estimate the contributions of the independent variables to the hazard. It has been shown that the discrete hazard model generates unbiased estimates of the coefficients of a continuous time proportional hazards model (10). A limited set of predefined baseline factors was entered. These factors were selected based upon previous research findings and own interest.

The following factors at intake were examined: gender, frequency of encopresis, family history, and earlier psychological treatment. If patients had primary encopresis (i.e. never toilet trained at intake) or secondary encopresis (i.e. the child had been toilet trained before intake but had regressed to incontinence) was sub-divided into three categories. No onset of complaints of encopresis before the age of 6 and secondary encopresis, onset of complaints before the age of 6 with secondary encopresis and onset of complaints before the age of 6 with primary encopresis. Frequency of encopresis at intake was dichotomised into: seven times
encopresis a week or less versus seven times per week or more.

Because of the low number of events, we analysed all relapses in the two years following initial success combined rather than in a discrete time model. A standard logistic regression model was used to examine predictive factors for relapse after an initial success. The same factors were examined as in the success model. Analyses were performed with SAS software version 8.2 and SPSS version 11.0.
Chapter 8

Results

Patient characteristics

A total of 114 patients were enrolled in this long-term follow up study of which 90 were boys (79%). Median age at intake was 9.2 years, whereas the median age of onset of defecation problems was 4 years. There were no significant differences between boys and girls with regards to patient characteristics at intake (table 1).

Table 1 patient characteristics at intake.

<table>
<thead>
<tr>
<th>Characteristic</th>
<th>Value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Number of patients in study</td>
<td>114 (90 male)</td>
</tr>
<tr>
<td>Median age at intake in years (25th-75th percentiles)</td>
<td>9.2 (7.9-11.6)</td>
</tr>
<tr>
<td>Median years of symptoms before intake (25th-75th percentiles)</td>
<td>4.4 (3.0-6.7)</td>
</tr>
<tr>
<td>Median encopresis frequency / week (25th-75th percentiles)</td>
<td>8.0 (5.0-14.0)</td>
</tr>
<tr>
<td>Median defecation frequency / week on toilet (25th-75th percentiles)</td>
<td>7.0 (7.0-10.0)</td>
</tr>
<tr>
<td>Primary encopresis (i.e. never toilet trained at intake)</td>
<td>32%</td>
</tr>
<tr>
<td>Daytime enuresis</td>
<td>28%</td>
</tr>
<tr>
<td>Night time enuresis</td>
<td>26%</td>
</tr>
<tr>
<td>Median months treatment before intake (25th-75th percentiles)</td>
<td>5.5 (2.0-24.0)</td>
</tr>
<tr>
<td>Psychological treatment before intake</td>
<td>28%</td>
</tr>
<tr>
<td>Positive family history</td>
<td>20%</td>
</tr>
</tbody>
</table>

Clinical outcome during follow-up

A total of 119 consecutive children with FNRFS took part in two previous randomised, large biofeedback studies (5). Five patients did not complete these treatment studies, consequently 114 children were enrolled in the present FU study. During the 12 year period of the current FU study, 10 of the 114 patients were lost to FU, in all cases due to a change to a new and unknown address. Despite all efforts, we were unable to trace the new addresses of these subjects. The overall FU percentage throughout the study was therefore 90%. Figure 1 shows the percentages of clinically successful patients at different years of FU. At one year of follow-up, only 28.6% of patients were successfully treated after the initial study protocol of 6-7 weeks. At two years, still only 29.5% of patients were successfully treated despite intensive treatment and frequent follow-up. Figure 1 shows a steady increase in success from three years of FU and further on. Thirteen patients reached twelve years of FU at the time of this report; they were all clinically successful.
Figure 1: Percentage of clinically successful patients at different years of follow-up. The number on top of each bar indicates the number of patients available for FU in that year.

Figure 2: Success percentages at different (biological) ages.

Figure 2 shows the FU results according to biological age. A steady increase in percentage of successfully treated patients can be observed from the age of 7. However, at the age of 12 years, still 49% of the patients was not successfully treated and had at least 1 encopresis episode per 2 weeks. At 18 years of age, 85% of the FNRFS patients were clinically successful. At this age, 81% of patients had no encopresis any more.
Frequency and prognostic factors for first success

**Success model**

Figure 3 shows the cumulative percentage of FNRFS patients achieving at least one successful period somewhere during the course of FU. Cumulative success percentages were not significantly different between boys and girls during all FU time points (p=0.67, log-rank test) (figure 3). Within one year after the initial treatment period of 6-7 weeks, clinical success was obtained at least once in 36% of patients. After 5 years of FU this percentage was 75%. After 7 years of FU the cumulative percentage of children who did experience at least one successful outcome was 80%. In other words, after 7 years of FU 20% of children had not once experienced successful treatment outcome.

![Figure 3: cumulative percentage of children who achieved successful treatment.](image)

The different variables associated with first success after treatment in the patient sample are depicted in table 2 (univariate- and multivariate analysis). We found age at onset of symptoms younger than 6 six years in combination with secondary enuresis to be associated with a lower chance on achieving success (HR: 0.51 (95% CI: 0.27-0.98), P=0.04). Other factors mentioned in table 2 were not associated with a lower chance on achieving a first success somewhere during FU. Nevertheless, a trend was observed towards a diminished number of clinical successful children who had received prior psychological treatment (HR: 0.60 (95% CI: 0.35-1.01), P=0.06).
Table 2 Factors predicting first treatment success (Hazard ratios (HR) and 95% confidence intervals (CI)).

<table>
<thead>
<tr>
<th>Factor</th>
<th>Hazard ratio (95%CI), univariate analysis</th>
<th>Hazard ratio (95% CI), multivariate analysis</th>
</tr>
</thead>
<tbody>
<tr>
<td>Male sex</td>
<td>1.06 (0.65-1.74)</td>
<td>1.24 (0.72-2.14)</td>
</tr>
<tr>
<td>Encopresis more than 7 /wk</td>
<td>0.65 (0.42-0.99)</td>
<td>0.77 (0.49-1.22)</td>
</tr>
<tr>
<td>Family history positive</td>
<td>0.93 (0.56-1.54)</td>
<td>0.91 (0.53-1.54)</td>
</tr>
<tr>
<td>Psychological treatment</td>
<td>0.62 (0.39-0.98)</td>
<td>0.60 (0.35-1.01)</td>
</tr>
<tr>
<td>&gt; 6 years and secondary encopresis</td>
<td>1.00 (reference)</td>
<td>1.00 (reference)</td>
</tr>
<tr>
<td>≤ 6 years and secondary encopresis</td>
<td>0.54 (0.29-1.01)</td>
<td>0.51 (0.27-0.98)</td>
</tr>
<tr>
<td>≤ 6 years and primary encopresis</td>
<td>0.55 (0.28-1.05)</td>
<td>0.62 (0.31-1.24)</td>
</tr>
</tbody>
</table>

Frequency and prognostic factors for relapse after initial successful treatment

Relapse

Figure 4 (KM curve) shows the cumulative percentage of relapses during FU after achievement of initial success. Cumulative relapse percentages were not significantly different between boys and girls during all FU time points (p=0.41, log-rank test). Within the first 2 years after initial success, relapses occurred most frequently: after 1 year 29%, and 2 years after their original successful treatment, 34% of patients experienced a relapse. After 7 years of FU the cumulative percentage of children who experienced a relapse at least once after initial success was 37%.

Multivariate analysis (table 3) showed that no single factor could be significantly related to relapse after an initial successful treatment outcome.
Figure 4: cumulative percentage of children who relapsed after initial successful treatment.

Table 3 Factors predicting relapse after an initial success (Odds ratios and 95% confidence intervals (CI)).

<table>
<thead>
<tr>
<th></th>
<th>Odds ratio (95%CI), univariate analysis</th>
<th>Multivariate Odds ratio (95% CI)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Male sex</td>
<td>1.54 (0.53-4.48)</td>
<td>1.78 (0.58-5.54)</td>
</tr>
<tr>
<td>Encopresis more than 7 / week</td>
<td>1.05 (0.44-2.48)</td>
<td>1.04 (0.41-2.62)</td>
</tr>
<tr>
<td>Family history positive</td>
<td>0.87 (0.31-2.44)</td>
<td>0.91 (0.31-2.69)</td>
</tr>
<tr>
<td>Psychological treatment</td>
<td>0.89 (0.34-2.30)</td>
<td>0.89 (0.31-2.60)</td>
</tr>
<tr>
<td>&gt; 6 years and secondary encopresis</td>
<td>1.00 (reference)</td>
<td>1.00 (reference)</td>
</tr>
<tr>
<td>≤ 6 years and secondary encopresis</td>
<td>0.60 (0.17-2.09)</td>
<td>0.50 (0.13-1.86)</td>
</tr>
<tr>
<td>≤ 6 years and primary encopresis</td>
<td>1.02 (0.28-3.77)</td>
<td>0.98 (0.23-4.10)</td>
</tr>
</tbody>
</table>
Discussion

This is the first study describing the long-term follow-up of patients with functional non-retentive faecal soiling (FNRFS). After two years of intensive medical and behavioural treatment only 29% of the children were successfully treated. At the age of 12, almost 50% of the children still suffered from encopresis. Although a steady increase in success was observed over the years and even during puberty, in 15% of the FNRFS patients, encopresis persisted into young adulthood. Clinical success was lower in patients with an onset of encopresis younger than six years in combination with secondary encopresis. Relapses were frequent (cumulative percentage 37%), occurring most likely within the first two years after an initial success.

Patients with functional non-retentive faecal soiling (FNRFS) differ substantially from constipated children with respect to patient characteristics and success percentage. FNRFS patients are mainly boys (79% in this sample) whereas in children with constipation the male: female ratio is 3:2. Compared to constipated children participating in our long-term FU study (8), FNRFS patients had less therapy before intake (5.5 months vs. 15 months), had more day- and night-time enuresis (28%/26% vs. 8%/18% respectively), and were more likely to have a positive family history (20% vs. 13%).

Another important difference was the delay in presentation; FNRFS patients presented to our outpatient clinic at the age of 9.2 years whereas constipated children were seen at the age of 6.5 years (11). We hypothesise, that parents of children with FNRFS are ashamed of failing to successfully toilet train their child and therefore postpone a doctors visit. In contrast, children with constipation complain of infrequent, painful defecation which is a good reason for the parents to visit a doctor earlier. While in FNRFS patients the absence of accompanying symptoms such as painful or infrequent defecation will often not alarm parents. Moreover, encopresis in the absence of constipation will often not be recognised as separate clinical entity by general physicians and paediatricians and might cause a delay in referral after unsuccessful therapy.

Clinical success, i.e. less than one encopresis episode in two weeks without the use of medication influencing motility, was achieved in only 29% of FNRFS patients at two years of FU. Thereafter, a steady increase in success was observed. Of all young adults (aged 18) 85% was free of encopresis. In contrast, we reported a
significant higher percentage of success after 1 year of treatment (59%) in children with chronic constipation (8). In these patients, from 9 years on until the age of 16, a steady but less pronounced increase in success percentage was observed (70% at 16 years). Thereafter this percentage of success remained stable into young adulthood (70% successful at age 18). We can only speculate about the difference in success between non-retentive and retentive encopretic children. Van Ginkel et al. clearly showed that laxatives were not helpful and even worsened the frequency of encopresis episodes in patients with FNRFS (6). We now know that behavioural therapy consisting of structured toilet training three times daily after meals and keeping a bowel diary together with education is the only effective therapy in FNRFS, while children with constipation need additional long lasting laxative treatment.

As stated, a strict toilet training programme is the corner stone of FNRFS treatment. Thus, motivation to adhere to this intensive programme is of paramount importance in children with FNRFS. During puberty, the increase in success percentage might be the result of 'peer-pressure'. During this critical period, influences of the social environment of the young adolescent could pursue him or her to sustain toilet-training three times daily.

We have no explanation for the observed negative relation between success and regression to incontinence in children younger than 6 years (i.e. secondary encopresis). In constipated children a young age at onset of symptomatology was a negative predictive factor for success as well (8,12). In these studies it was suggested that in young children underlying organic pathophysiological mechanisms are responsible for their symptoms in contrast to children who have an onset of constipation after many years of normal defecation.

The high percentage of relapse within the first two years after an initial success has consequences for the management of FNRFS patients. It underscores that these children should be closely monitored for at least two years after initial success. Based on our clinical experience we suggest continuing toilet training and keeping the bowel diary in combination with regular outpatient visits.

In conclusion, in children with FNRFS a cumulative success percentage of 80% is found after 12 years of follow-up. However, in those reaching the age of 18 years, still 15% have encopresis. Relapse after an initial success occurs frequently and most likely in the first two years after successful treatment. This high percentage of relapse stresses the importance of intensive monitoring and follow-up of FNRFS patients.
References


