Diagnostic dilemmas and results of treatment for chronic constipation

C W Keuzenkamp-Jansen, C J Fijnvandraat, C M F Kneepkens, A C Douwes

Abstract
Chronic functional constipation (CFC) may be difficult to recognise and information regarding its long term prognosis is scarce. The records of 244 children with CFC, aged 0-18 years, were analysed for symptoms at presentation and results of treatment, and long term outcome was evaluated by means of a telephone interview in 137 patients discharged for more than one year. The patients presented with a great variety of symptoms, only 22% having infrequent defecation of increased consistency, another 22% having an obviously normal defecation pattern. The mean duration of treatment was 13 months. At the time of discharge, 69% of the patients still used laxatives. At a median of four years after discharge, 66% of the children were free of symptoms and without medication, 39% having experienced a recurrence. It is concluded that CFC may be difficult to recognise and can be alleviated by an intensive laxative regimen. Recurrence of symptoms is common, but the long term prognosis is good in most patients.

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Keywords: chronic constipation, plain abdominal radiography, laxatives.

Although constipation in children is frequently encountered by general practitioners as well as paediatricians and psychologists, its diagnosis and treatment remain difficult and its prognosis uncertain.1-4 After exclusion of such relatively rare anatomical causes of constipation such as Hirschsprung's disease, anal atresia and spinal disease, and secondary causes such as being bedridden and hypothyroidism, in most cases chronic functional constipation (CFC) can be diagnosed. CFC is easily diagnosed, when decreased frequency of defecation and increased size and consistency of the faeces are the main symptoms. The diagnosis can be missed in many cases, however, if only these symptoms are relied on. The symptoms of CFC, including the altered defecation characteristics, are primarily caused by the delayed passage of faeces leading to colonic faecal retention; this implies that other symptoms, such as recurrent abdominal pain, may be prominent. For this reason we prefer to define CFC as a condition in which chronic faecal retention, not caused by an anatomical abnormality, induces abdominal and/or systemic symptoms. For these symptoms no obvious other explanation should be available, and they should subside after colonic evacuation.

The treatment of CFC requires at least several months to be successful. Evacuation of the colon followed by maintenance laxation, toilet training, and parental counselling and support are important aspects of the treatment. Even with optimal treatment, relapses frequently occur, both during treatment and afterwards. Until now, only one systematic survey of the presenting complaints of CFC has been reported, and little is known of the long term outcome.5 The purpose of this study was, therefore, to identify the presenting symptoms of CFC in childhood and to evaluate its long term outcome.

Patients and methods

PATIENTS
A total of 244 children with CFC were included out of a total of 409 children referred to our paediatric gastroenterological outpatient department, between 1976 and 1991, for complaints that were eventually shown to be caused by constipation. Roughly half of the patients were referred by family practitioners and half by paediatricians. Patients were included when they fulfilled the following criteria: signs and symptoms consistent with CFC; intake plain abdominal radiography available (in children older than 1 year); no other cause for constipation found as judged by selective screening; no history of anal surgery; and a treatment period at our outpatient department of at least six weeks. A total of 165 children were not eligible for the following reasons (numbers in parentheses): referred for diagnosis of Hirschsprung's disease (65), history of psychomotor retardation (7), history of anal dilatation (10), seen only for diagnostic purposes (30), less than three visits (25), no laxative treatment instituted (9), and records incomplete (19).

DIAGNOSIS AND TREATMENT REGIMEN
All children were initially evaluated and treated by two of the authors (ACD and CMFK). Medical histories were obtained and a complete physical examination was performed. In all children a plain abdominal radiograph was made and evaluated using the Barr score.6 In our experience, this score is useful when applied to children over 1 year of age.

Once CFC was diagnosed, the children were treated according to the standard three stage therapy schedule used at our centre. In the first stage, the evacuation phase, the colon is evacuated employing a stool softener (lactulose or lactitol) in combination with a mild irritant (bisacodyl or sodium picosulphate, depending

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on the age of the child). Mineral oil is added when the bulk of faecal retention is located in the distal colon and rectum and especially in the presence of scybala. Rectal medication (phosphate enemas) is used only in the first few days of treatment in case of huge faecal impaction in the rectosigmoid, and in case of difficult to treat recurrences, as an adjunct to oral medication. In the evacuation phase, the laxative doses are high enough to produce semiliquid stools, in order to obtain evacuation of the colon and to prevent active withholding of defecation. Parents and children receive oral and written information on the nature of the condition and the rationale for our therapeutic regimen. When old enough the children are told to visit the toilet twice daily and take enough time for defecation. No dietary advice is given at this stage apart from the introduction of wholemeal bread, if necessary. Initially, wholemeal bread is offered, either by means of outpatient visits or by telephone. The aim of the second stage is to prevent colonic reaccumulation of faeces. The laxative dose is decreased until the child produces soft stools daily, and tapered at intervals of two to three weeks until the lowest effective dose is reached. Finally, in the third stage, a fibre enriched diet is introduced and laxatives are gradually stopped. Insufficient progress or relapses prompt intensification of treatment. This may include a one to two week hospital admission in order to obtain evacuation in a clinical setting.

Dependent on the clinical judgment of the attending consultant and the willingness of patient and parents, a child psychologist may be consulted. Referral usually takes place when one of the following factors is present: constipation or symptoms refractory to medical treatment without obvious organic explanation; overt psychosocial pathology judged to interfere with medical treatment; or parents asking for help in the guidance of their constipated child.

DATA COLLECTION

The information used in this study was extracted from the patient records using a structured data collection form. When the child had been discharged for more than one year from the outpatient department by 1 June 1991, the parents were approached for a telephone interview. They were questioned about any relapses and any continuing use of laxatives. They were considered to be cured when they reported no actual complaints attributable to CFC and did not use any laxatives in the last six months.

STATISTICAL ANALYSIS

Frequency distributions were calculated for symptoms and treatment variables. The follow up data were analysed by constructing a Kaplan–Meier curve of the persistence of complaints for three age groups. Differences between these groups were tested with a log rank test (Mantel-Cox). Associations between outcome at follow up and variables about history or treatment variables were tested with the $\chi^2$ test.

Results

PATIENT CHARACTERISTICS AND SYMPTOMS AT PRESENTATION

The charts of 135 boys and 109 girls were reviewed (male to female ratio 1.23:1). The ages of the children at the onset of symptoms and the total delay between onset of symptoms and start of laxative treatment are depicted in figure 1. The long delays were principally caused by patient's delay, that is, the time between the onset of complaints and the first medical consultation. For 154 patients both the patient's delay and the doctor's delay were known. In this group medians and first and third quartiles for the patient's delay were 0.7, 2.4, and 9.0 months; for the doctor's delay, 0, 0.8, and 5.1 months; and for total delay, 1.6, 5.4, and 17.0 months, respectively. The longest patient's delay as reported by the parents was more than 16 years. There was no relation between age at start of symptoms and total delay.

The reasons for presentation at the outpatient department, as well as the symptoms mentioned during history taking, are shown in table 1. 'Constipation' as such (that is, hard, infrequent stools) was the presenting symptom in not more than 31% of the children. Encopresis, defined as involuntary loss of stools including soiling, was found in half of the children over 3.5 years of age, and in 40% of them was the principal reason for referral (table 1). As judged by a plain abdominal radiograph, constipation was present in all of them and they were treated not differently from the whole group. Interestingly, 15 children (6%) presented as an emergency because of acute abdominal pain. Most children (60%) experienced at least two other symptoms in addition to their main reason for referral (table 1). Information on defecation frequency and consistency, available on 210 children (86%) is given in table 2.

![Figure 1](http://adc.bmj.com/)

**Figure 1** (A) Age at start of treatment (n=231) and (B) total delay before laxative treatment started (n=226); median age was 3 years and median delay 10 months.
### Table 1 Reasons for presentation and symptoms of the 244 children, figures are number (%)

<table>
<thead>
<tr>
<th>Symptoms</th>
<th>Reasons for presentation</th>
<th>Symptoms at time of presentation*</th>
</tr>
</thead>
<tbody>
<tr>
<td>Abdominal pain</td>
<td>77 (31.6)</td>
<td>135 (55.3)</td>
</tr>
<tr>
<td>Constipation</td>
<td>75 (30.7)</td>
<td>106 (43.4)</td>
</tr>
<tr>
<td>Encopresis, soiling</td>
<td>34 (13.9)</td>
<td>87 (49.7)†</td>
</tr>
<tr>
<td>Anorexia</td>
<td>1 (0.4)</td>
<td>69 (28.3)</td>
</tr>
<tr>
<td>Malaise</td>
<td>4 (1.6)</td>
<td>38 (15.6)</td>
</tr>
<tr>
<td>Painful defecation</td>
<td>2 (0.8)</td>
<td>36 (14.8)</td>
</tr>
<tr>
<td>Diarrhoea</td>
<td>18 (7.4)</td>
<td>34 (15.9)</td>
</tr>
<tr>
<td>Enuresis</td>
<td>1 (0.4)</td>
<td>26 (10.7)</td>
</tr>
<tr>
<td>Vomiting</td>
<td>5 (2.0)</td>
<td>24 (9.8)</td>
</tr>
<tr>
<td>Rectal bleeding</td>
<td>2 (0.8)</td>
<td>18 (7.4)</td>
</tr>
<tr>
<td>Headache</td>
<td>0</td>
<td>17 (7.0)</td>
</tr>
<tr>
<td>Fear of defecation</td>
<td>1 (0.4)</td>
<td>15 (6.1)</td>
</tr>
<tr>
<td>Nausea</td>
<td>0</td>
<td>15 (6.1)</td>
</tr>
<tr>
<td>Low grade fever</td>
<td>0</td>
<td>14 (5.7)</td>
</tr>
<tr>
<td>Distended abdomen</td>
<td>0</td>
<td>11 (4.5)</td>
</tr>
<tr>
<td>Perianal problems</td>
<td>1 (0.4)</td>
<td>10 (4.1)</td>
</tr>
<tr>
<td>Flatulence</td>
<td>0</td>
<td>10 (4.1)</td>
</tr>
<tr>
<td>Sleeping problems</td>
<td>0</td>
<td>9 (3.7)</td>
</tr>
<tr>
<td>Urinary tract infections</td>
<td>1 (0.4)</td>
<td>9 (3.7)</td>
</tr>
<tr>
<td>Delayed faecal continence</td>
<td>10 (4.1)</td>
<td>0</td>
</tr>
<tr>
<td>Food allergy</td>
<td>6 (2.5)</td>
<td>0</td>
</tr>
<tr>
<td>Other</td>
<td>4 (1.6)</td>
<td>14 (5.7)</td>
</tr>
</tbody>
</table>

* One or two symptoms in 97 children (40%) and three or more in 147 (60%).
† Calculated as a percentage of the 175 children over 3.5 years of age, the age at which continence may be expected.

### Table 2 Defecation patterns of 210 children at time of presentation, figures are number (%)

<table>
<thead>
<tr>
<th>Consistency</th>
<th>Frequency</th>
<th>Consistency</th>
<th>Frequency</th>
</tr>
</thead>
<tbody>
<tr>
<td>Hard</td>
<td>46 (21.9)</td>
<td>Normal</td>
<td>6 (2.9)</td>
</tr>
<tr>
<td>Normal</td>
<td>20 (9.5)</td>
<td>&gt; 2 Daily</td>
<td>15 (7.1)</td>
</tr>
<tr>
<td>Semi-liquid</td>
<td>46 (21.9)</td>
<td>Variable</td>
<td>18 (8.6)</td>
</tr>
<tr>
<td>Variable</td>
<td>9 (4.3)</td>
<td>5 (2.4)</td>
<td>7 (3.3)</td>
</tr>
</tbody>
</table>

A positive family history for constipation (meaning that at least one first degree relative was known to have constipation) was present in 90 (50%) of the 180 patients in whom this information was available. Likewise, constipation had been present during infancy in 114 of 202 patients (56%). The parents of 82 out of 160 children (51%) recalled that their child had a fear of defecation as a toddler, resulting in withholding and straining.

**Defecation problems vs abdominal pain**

As most studies on the treatment and outcome of constipation focus on overt defecation problems, we compared patients presenting with abdominal pain as the sole or major problem (n = 77) with patients presenting with symptoms classically considered consistent with CFC ('constipation', encopresis, diarrhoea, delayed faecal continence, painful defecation, fear of defecation, and rectal bleeding; table 1; n = 142). The abdominal pain group tended to be older at the onset of symptoms (mean 85 ± 34 months; p < 0.001); they had a shorter patient's delay (mean 6 ± 15 months; p = 0.013) and a longer doctor's delay (mean 14 ± 5 months, p = 0.005), resulting in similar total delays (20 ± 19 months; NS).

**TREATMENT**

Dietary advice, that is, starting solid foods in infants or introducing fibre rich diets in older children, was the only treatment for 12 patients (5%). The other 232 children received lactulose during the evacuation phase at a mean (SD) dose of 0.88 (0.46) g/kg body weight/day (range 0.17-3.45 g/kg). In addition, 183 older children were treated with bisacodyl at a dose of 0.35 (0.16) mg/kg/day (range 0.10–1.07 mg/kg) and 39 infants and younger children with sodium picosulphate at a dose of 0.23 (0.11) mg/kg/day (range 0.06–0.58 mg/kg). Mineral oil was prescribed to 53 children at a dose of 1.4 (1.0) ml/kg/day (range 0.2–6.4 ml/kg). All dosages were taken close to eight weeks after the start of treatment; this is the time expected that the optimal evacuation phase dose was obtained. At some time during the course of treatment it was felt necessary to consult a child psychologist in 57 patients (23%), and 31 patients (13%) were hospitalised at least once for extensive clinical laxation. As a group, patients requiring psychological counselling were not worse than those in whom this was not judged necessary. Ninety-five children (39%) had a relapse of symptoms, requiring intensification of treatment, at some time before discharge.

At the time this study was performed 212 patients (87%) had been discharged from outpatient follow up. The average treatment period had been 13.3 months (median 9 months, range 6 weeks to 6.8 years). One hundred and sixty-four (69%) still used low doses of laxatives at discharge, but in 131 children (62%) symptoms had disappeared completely at the time of discharge. Improvement of symptoms was obtained in another 70 children (33%) who had been referred to their general practitioner or were judged able to manage their symptoms themselves. The remaining 11 children (5%) had unchanged symptoms when last seen at the outpatient department before they discontinued treatment. Thirty two patients were still under supervision. The mean treatment period in this group was longer than average at 23 months, with half of the patients being treated for at least 16 months. No association could be demonstrated between the duration of the treatment period and treatment delay.

**Defecation problems vs abdominal pain**

The doses of lactulose and bisacodyl given to patients with abdominal pain were lower than those given to patients with defecation problems: 0.69 (0.29) v 0.99 (0.51) g/kg/day (p < 0.001) and 0.30 (0.13) v 0.37 (0.18) mg/kg/day (p = 0.006), respectively. No significant differences were found for mineral oil and sodium picosulphate, the latter being given to only three children with abdominal pain. The percentages of children requiring psychological consultation or hospital admission were similar in both groups. The mean total duration of treatment in those children discharged from follow up was close to 14 months in both groups. None of the other parameters studied differed significantly between these groups, apart from the presence of encopresis at the time of discharge, which was less often present in the abdominal pain group (5/69) than in the defecation problems group (43/100).
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Discussion
Loening-Baucke evaluated 174 patients with constipation, all younger than 4 years of age, for symptoms at presentation. She found infrequent bowel movements in 58%, painful bowel movements in 77%, and stool withholding in 97%. In her study, CFC was diagnosed using clinical features only. In our experience, when using clinical features only the diagnosis of CFC may be easily missed. We therefore adopted the Barr score of plain abdominal radiographs as an objective measure of CFC. In our hands and others', this method, developed for schoolchildren, is a valuable tool for children over 1 year of age, although (re)validation is impossible for ethical reasons. In recent years new objective criteria have been developed for the assessment of constipation, including the segmental colonic transit time using markers and repeated radiography. While the use of such a technique would be useful in a prospective study, for clinical purposes the Barr score has the great advantage of enabling an instant diagnosis of constipation and the start of treatment after the first outpatient visit. Although there seems to be no direct relationship between Barr score and colonic transit time, the same holds for ease of rectal evacuation and colonic transit time in adult constipated patients. Nevertheless, there is little doubt that increased faecal mass on plain abdominal radiography represents faecal retention and therefore is a simple tool for the objective assessment of constipation.

In the present study, the records of 244 children, aged 0–18 years, with CFC as identified by plain abdominal radiography, were analyzed retrospectively. By using this technique we could demonstrate that the clinical picture of CFC is much more diverse than usually acknowledged. Only 46 children (22%) had the classical presentation of infrequent defecation of increased consistency. Many children had an apparently normal defecation pattern; this is a feature that is not generally appreciated as being consistent with CFC. The diagnosis may therefore easily be missed, as is illustrated by the long treatment delays, especially in children with abdominal pain as the presenting complaint; this also may explain the large number of patients with occult constipation in our population. Abdominal pain is the most frequently encountered single symptom, occurring in half of our patients. As is clear from the analysis of the abdominal pain group, they essentially differ only from the children presenting with defecation problems in that they are significantly older at the onset of symptoms. The lower doses of laxatives used to reach optimal treatment effect may be fully explained by the higher age at the start of treatment. It is our clinical impression that so-called recurrent abdominal pain in schoolchildren often coexists with CFC. We feel that CFC deserves a prominent place in the differential diagnosis of childhood recurrent abdominal pain. In this respect the shorter patient's delay and longer doctor's delay we found are very interesting; they may be interpreted as abdominal pain being a symp-
tom that provokes more concern in the parents while the doctor fails to make a timely diagnosis of CFC.

To our knowledge, headache and low fever have not been reported before as symptoms of CFC. We had several children presenting with temperatures up to 38°C, or even higher, as one of the main symptoms, CFC being the sole recognised cause. Once the constipation is treated adequately, the body temperature normalises. We are not aware of a report on this phenomenon, nor do we have a sound explanation for it. The importance of it lies in the fact that, apart from simple screening procedures, such as erythrocyte sedimentation rate and white cell count, extensive investigations may be postponed until the effect of laxative treatment is appreciated. In comparison with other studies we found a low prevalence of recurring urinary tract infections: 3.7% compared with 7–30%.5 15-18 This may be due to either the high number of cases with occult constipation or to the referral pattern of our patients, half of them being referred to us by general paediatricians. From the literature it is known that recurrent urinary tract infections often subside when constipation is adequately treated.19

The aetiology of CFC is still unknown. Some children demonstrate unconscious contractions of the external anal sphincter during defecation.20 This paradoxical reaction is probably based on fear of a painful defecation and may contribute to the pathogenesis of CFC because it limits colonic evacuation. Familial and constitutional factors seem to play a part in the aetiology of CFC. In half of our patients the parents reported that they themselves or one of the other children also had constipation, which is in agreement with other reports.18 21 22 Both the positive family history and the fact that 57% of our patients were reported to have suffered from constipation as an infant suggests a role for congenital factors in the aetiology of CFC.

Although in 23% of the families in our study the help of a child psychologist was sought, this was mostly the case only after our regular treatment regimen failed to reach any further improvement, or when overt psychosocial problems seemed to interfere. It was thought by us and by the team of psychologists that the behavioural problems were not the cause of the defecation abnormalities in any of the patients. These problems often seem to be a sequel of CFC and hence decrease after successful treatment.14 18 21-34

Contrary to our experience that encopresis is virtually always accompanied by CFC, Benninga et al recently suggested that encopresis could exist without CFC.23 In their study, constipation was primarily diagnosed on clinical grounds. Defecation dynamics as well as child behaviour scores were found abnormal in children with CFC with or without encopresis, and in children with encopresis without CFC, compared with controls.24 Rockney et al, using plain abdominal radiographs for diagnosis, found about one fifth of the encopreptic children to have no evidence of faecal retention.7 These children were less likely to have faeces palpable on rectal examination, and more likely to have experienced difficulties in toilet training.

Recently, Loening-Baucke published a study of CFC in children younger than 4 years of age.1 The treatment regimen in this study was similar to ours, although we do not routinely use enemas and never prescribe milk of magnesia. We conclude from that the laxative doses required to adequately treat constipation in children is much higher than the doses suggested by the manufacturer. The results of our treatment strategy are comparable with hers1 and to those of other studies.4 19 26 27

From our follow up data it may be concluded that for the majority of children, although symptoms from CFC may be tenacious, the long term prognosis is favourable. According to our results, children starting treatment before 3 months of age had a better prognosis than those starting treatment between 13 and 72 months of age. This may be due to difficulties associated with bowel habit training, as this is the age that continence must be accomplished. For those children that have persistent symptoms of constipation, biofeedback training might be a valuable alternative to conventional treatment.28-30

In summary, CFC, which may be difficult to recognise when classical symptoms of disordered defecation are absent, can effectively be controlled in the majority of children when using the paediatrician as the main agent. As a rule, long term prognosis is favourable, independent of the presenting symptoms.

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