Summary

The diagnosis functional constipation is based on a complex of symptoms in the absence of an organic cause. The main characteristics of constipation are infrequent, hard and painful defecation accompanied by the involuntary loss of feces in the underwear. Functional constipation represents the main complaint in 3% of pediatric outpatient’s visits. This percentage increases to 25–45% in specific pediatric-gastrointestinal motility clinics.

It is assumed that functional constipation is the result from the interplay between biological, psychological and social factors (the so-called biopsychosocial approach). Stool-withholding behavior because of fear for painful defecation is viewed as the major factor in the development and maintenance of functional constipation in children. Consequently, retained stools become harder, larger, and more painful to evacuate leading to even more fear and avoidance of defecation. Moreover, retained stools cause chronic distension of the rectum, which will subsequently lead to overflow incontinence, a terrible symptom for both parent and child. Together with toileting resistance, continued fecal incontinence can be a source of conflict between parents and children, which then contributes to the maintenance of functional constipation. Moreover, emotional and behavior problems are common in children with functional constipation. Successful treatment of functional constipation is often associated with improvement in emotional and behavioral problems suggesting that these problems are secondary to functional constipation. Nonetheless, it remains the question whether emotional and behavior problems are the result of functional constipation or vice versa.

Based on clinical experience, constipated children are traditionally treated by pediatricians combining laxative treatment with behavioral approaches like toilet training and education. However, long-term follow-up studies showed that despite intensive medical treatment, functional constipation persists into young adulthood in approximately one third of patients.

Previous studies showed that there is some evidence that behavioral interventions added to laxative therapy have advantage over laxative therapy alone for improving continence in children with fecal incontinence associated with functional constipation. Although many studies report on medical treatment and behavioral interventions, comparison of findings is hampered because descriptions of the interventions are unclear. Behavioral recommendations and education given by physicians have to be distinguished from behavioral therapy. Behavioral therapy is a form of psychotherapy that employs specific techniques derived from learning theory that constructively changes the patient’s behavior. The efficacy of a well-described behavioral therapy combined with laxatives compared with medical treatment has never been evaluated by randomized controlled trials.

Pediatric psychologists of the Psychosocial Department from the Emma Children’s Hospital/Academic Medical Center in Amsterdam developed a protocolized behavioral therapy for the treatment of functional constipation in childhood. The protocol is based on literature, clinical experience and learning theory (classic and operant conditioning learning principles). The primary aim of this thesis was to compare the efficacy of this behavioral therapy with laxatives, with conventional treatment in treating functional constipation in childhood in a randomized controlled trial. This resulted in the following research question:

1. Is behavioral therapy with laxative treatment compared with conventional treatment (laxative treatment combined with education and toilet training) more successful in treating functional constipation, stool-withholding behavior, and emotional and behavior problems?
The secondary aim of this thesis was to investigate health-related quality of life (HRQoL), emotional and behavior problems in children with functional constipation, and parental child-rearing attitudes. The following research questions were formulated:

2. How is the HRQoL of children with functional constipation, and are clinical characteristics of functional constipation associated with reported HRQoL?

3. What is the prevalence of emotional and behavior problems in children with functional constipation, and are clinical characteristics of functional constipation associated with reported emotional and behavior problems?

4. Are parental child-rearing attitudes associated with functional constipation?

Chapter 1 is the introduction to this thesis. A working model (the so-called SORC model), commonly used by behavioral therapists, to explain functional constipation is presented. Treatment techniques of the behavioral therapy follow from this SORC model. Furthermore, this SORC model was used as a biopsychosocial research model from which the research questions were derived.

Chapter 2 provides a review of the literature on functional constipation. Next, the behavioral therapy protocol is presented that was used in the randomized controlled trial to evaluate the efficacy of behavioral therapy compared with conventional treatment. The behavioral protocol has two age-related modules (4-8 years; ≥ 8 years) with similar content except for anxiety reduction procedures by means of play therapy, which is only used for the younger children. In the older children autonomy with regard to toileting is promoted.

In Chapter 3 the results are presented of a randomized controlled trial in which the efficacy of behavioral therapy with laxatives compared with conventional treatment is evaluated. One hundred thirty-four children aged 4-18 years with functional constipation were randomly assigned to 22 weeks (12 visits) of either behavioral therapy or conventional treatment. Outcomes were evaluated at the end of treatment and at 6-months follow-up. The results showed that behavioral therapy compared with conventional treatment was equally successful in treating functional constipation. However, conventional treatment resulted in a higher defecation frequency than behavioral therapy, and behavioral therapy was superior in addressing emotional and behavior problems.

Chapter 4 describes how children with constipation-associated fecal incontinence, aged 8 years and older, evaluate the emotional and social impact of having a defecation disorder. The sample included the children that participated in our randomized controlled trial, and children with treatment resistant constipation that participated in a separate study evaluating the efficacy of rectal enemas. The HRQoL in 114 children was measured with a disease-specific questionnaire, the Defecation Disorder List (DDL). Item description showed that the majority of children reported relatively more emotional concerns than social consequences. Frequent episodes of fecal incontinence in the child were associated with lower HRQoL regarding emotional and social functioning, but only explained a small amount of the variance in HRQoL.

Chapter 5 describes the results concerning emotional and behavior problems in children with functional constipation. This study made use of baseline data of 133 children who participated in the randomized controlled trial. Behavior problems were three to four-fold higher in children
with functional constipation compared with the Dutch general pediatric population. Clinical characteristics of functional constipation were found to be associated with behavior problems. Especially a long duration of treatment was strongly associated with behavior problems. Children with nighttime urinary incontinence were also more likely to have behavior problems. Fecal incontinence and the production of large stools, two prominent features of functional constipation, were exclusively related to externalizing behavior problems.

In Chapter 6 the results are presented of a study investigating the relation between parental child-rearing attitudes and functional constipation in childhood. This study also made use of the baseline data of 133 children who participated in the randomized controlled trial. The study showed that parental child-rearing attitudes are associated with prominent symptoms of functional constipation. Constipated children of parents who either mildly or strongly encourage autonomy had less bowel movements and more episodes of fecal incontinence than constipated children with parents showing no specific focus on this parental child-rearing style. Children with parents with a strong overprotective attitude or a strong self-pity attitude also showed more episodes of fecal incontinence. The associations between parental child-rearing attitudes and functional constipation were specifically found for older children (aged ≥ 6 years).

Finally, in the General Discussion in Chapter 7 the results of this thesis are summarized and discussed in light of methodological limitations. A methodological drawback of our randomized controlled trial was that the additive effect of behavioral therapy could not be satisfactorily evaluated. The pediatric psychologists were partly responsible for adjusting laxative dosages during the behavioral therapy which possibly resulted in differences in laxative treatment. Furthermore, generalization of the findings into general practice could be limited. The study population consisted of constipated children referred to a tertiary center for treatment, and the quality of the conventional treatment was unusually high. From this, the following main messages & recommendations were derived:

- Medical treatment should remain the first treatment choice in treating functional constipation in childhood. Children with functional constipation should not be routinely treated with behavioral therapy to cure constipation.

- Treatment of emotional and behavior problems is needed in children with functional constipation. A collaborative approach which integrates medical and psychological treatment is recommended for constipated children with concomitant behavior problems. Healthcare providers should refer constipated children with behavior problems for behavioral therapy, while the healthcare provider regulates laxative treatment.

- Therefore, a screening for emotional and behavior problems should be incorporated in the diagnostic workup of functional constipation.

- If screening instruments are not available healthcare providers should be aware that constipated children 1) with treatment resistant constipation, 2) with nighttime urinary incontinence, and 3) children aged ≥ 7 years are at risk for having emotional and behavior problems.
• Periodic evaluation and discussion of the constipated child’s disease-specific HRQoL should become an integral part of medical care. Psychosocial barriers can be early identified and tailored intervention can be provided.

• Without a standard screening for HRQoL issues, the emotional and social impact of fecal incontinence, especially being bullied, should be addressed by the health care provider during control visits.

• Because healthcare providers work with parents collaboratively in the management of functional constipation, parenting issues should be incorporated in the treatment. Healthcare providers should routinely discuss general parenting issues and toileting-specific problems. This encourages positive parenting practices, and may allow parents to develop better strategies for the conflicts that may easily develop regarding defecation and toileting. Moreover, discussing parenting issues will also result in early recognition of dysfunctional parenting. Referral to mental health services is needed when parenting issues are hindering treatment to be successful or when the parent-child relationship is at risk.