Prevalence of Functional Defecation Disorders in Children: A Systematic Review and Meta-Analysis

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Objective To systematically review the literature regarding the epidemiology of functional constipation and functional nonretentive fecal incontinence (FNRFI) in children. Secondary objectives were to assess the geographical, age, and sex distribution of functional constipation and FNRFI and to evaluate associated factors.

Study design The Cochrane Library, PubMed, and Embase databases were searched from 2006 until September 2017. The following inclusion criteria were applied: (1) prospective studies of population-based samples; (2) reporting on the prevalence of functional constipation or FNRFI according to the Rome III/IV criteria; (3) in children aged 0-18 years; and (4) published in full manuscript form. A quality assessment of included studies was conducted. Random effect meta-analyses with meta-regression analyses of study characteristics were performed.

Results Thirty-seven studies were included, of which 35 reported on the prevalence of functional constipation and 15 of FNRFI. The reported prevalence of functional constipation ranged from 0.5% to 32.2%, with a pooled prevalence of 9.5% (95% CI 7.5-12.1). The prevalence of FRNFI ranged from 0.0% to 1.8%, with a pooled prevalence of 0.4% (95% CI 0.2-0.7). The prevalence of functional constipation was 8.6% in boys compared with 8.9% in girls (OR 0.99, 95% CI 0.9-1.4). Geographical location, dietary habits, and exposure to stressful life events were reported to be associated with the prevalence of functional constipation. Data on FNRFI were scarce and no associated factors were identified.

Conclusion Functional constipation is common in childhood and is associated with geographical location, lifestyle factors, and stressful life events. FNRFI is rare, and no associated factors were identified. *(J Pediatr 2018;■■:■■-■■).*

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The umbrella term functional defecation disorders is used to describe 2 distinct diagnoses: functional constipation and functional nonretentive fecal incontinence (FNRFI).[1] Functional defecation disorders are common in children and represent a frequent reason for healthcare consultations and increased healthcare expenditures.[2-5] Childhood functional constipation has a reported prevalence ranging from 0.7% to 29.6%.[6] The prevalence of FNRFI in the general pediatric population is estimated to be approximately 1%, although epidemiologic studies on FNRFI are scarce.[7] Functional defecation disorders are characterized by bothersome and embarrassing symptoms such as fecal incontinence and abdominal pain, which negatively impact quality of life of affected children and their families.[8-11]

Functional defecation disorders are diagnosed according to the symptom-based Rome criteria. In 2016, the Rome IV criteria were published, replacing the Rome III criteria, published in 2006.[1,12] For functional defecation disorders, the changes in Rome IV were minor compared with Rome III. According to the new Rome IV criteria for functional constipation, a distinction is made for young children between those who are toilet trained and those who are not. Furthermore, the time criterion has changed for both disorders; patients now need to fulfill the criteria for at least 1 month instead of 2 months.[1,12,13]

The most recent systematic review on the prevalence of functional constipation was published in 2011 and included studies with a variety of definitions for functional constipation, including self-reported diagnoses, stool consistency, stool frequency, Rome II, and Rome III criteria.[6] However, the use of different definitions of functional constipation significantly affects epidemiologic study outcomes, and the authors refrained from performing a meta-analysis on these studies with heterogeneous methodologies.[14] Currently, a systematic review on the

FNRFI	Functional nonretentive fecal incontinence
HRQoL	Health-Related Quality of Life

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prevalence of FNRFI is lacking. A systematic review on the epidemiology of functional constipation and FNRFI using uniform definitions, enabling a meta-analysis, could help to generate more insights into the worldwide epidemiology and potential risk factors. This could help to identify deficiencies in the current knowledge and to guide future research. Therefore, our aim was to perform a systematic review and meta-analysis on the epidemiology of functional defecation disorders in children according to the pediatric Rome III and Rome IV criteria. Our secondary aim was to evaluate geographic, age, and sex distribution for both disorders and to report on factors associated with both disorders, such as environmental, lifestyle, and psychosocial factors.

Methods

The Cochrane Library, PubMed, and Embase databases were searched from 2006 (the year the Rome III criteria were published) up to September 2017. The protocol including the full search strategy is provided in **Appendix 1** (available at www.jpeds.com). To identify additional studies, reference lists of review articles and included studies were searched manually. No language limits were used. This systematic review was not registered.

Articles were eligible for inclusion if they met the following inclusion criteria: (1) prospective and cross-sectional studies of population-based samples (eg, birth cohorts, school samples, or general population samples); (2) reporting the prevalence of functional constipation and/or FNRFI according to the pediatric Rome III or IV criteria; (3) in children aged 0-18 years; and (4) published in full manuscript form.

Abstracts were screened for eligibility by 2 authors independently through Covidence (Covidence systematic review software, Veritas Health Innovation, Melbourne, Australia; available at www.covidence.org). In case of disagreement, consensus was reached by discussion or by consulting a third author. All manuscripts that were considered eligible for inclusion were retrieved and read in full text to assess whether they fulfilled the inclusion criteria. One of the Rome III/IV criteria includes findings on physical examination. However, because a physical examination is often not performed in epidemiologic studies, we decided not to exclude studies for omitting this Rome criterion. Moreover, studies were included if only minor changes were made to a maximum of 1 of the Rome criteria based on the medical history, such as the criterion regarding duration of symptoms. Any changes made to the original Rome criteria are reported in the results and were taken into account during the quality assessment. Guidelines of the PRISMA statement (available at www.prisma-statement.org) were followed (**Appendix 2**; available at www.jpeds.com).

Quality Assessment

There is currently no gold-standard quality assessment tool for epidemiologic studies. For this study, we used an assessment tool that was developed previously by our research group, specifically designed for quality assessment of epidemiologic studies on functional abdominal pain disorders in children.[15] After adjusting this tool to address functional defecation disorders instead of functional abdominal pain disorders, we evaluated all manuscripts according to the following 6 questions: (1) Is the method of subject selection described and appropriate? (2) Are subject characteristics sufficiently described, ie, do they match the target population regarding to sex and age? (3) Are functional defecation disorders diagnosed according to the Rome III or Rome IV criteria? (4) Are the survey instruments reliable and valid? (5) Are the analytic methods described/justified and appropriate? (6) Were the results reported in sufficient detail? Each question was scored on a 3-point scale (no [0], partial [1], or yes [2]), with greater scores representing better methodologic quality. Quality assessment scores were not used to include or exclude studies from any of the analyses.

Data Extraction

The following data were extracted from each included study if possible: country where the study was performed, study design, type of population and sampling strategy, sample size, age range, sex distribution, method of data collection, definition of functional defecation disorders, overall prevalence of functional constipation, and FNRFI. If available, prevalence distributions according to sex, age, and geographic location were extracted from each study. Moreover, if factors associated with functional defecation disorders (eg, environmental, lifestyle, and psychosocial factors) were reported, we evaluated these study findings.

Statistical Analyses

Meta-analysis was performed with Comprehensive Meta-Analysis, version 3 (Biostat Inc, Englewood, New Jersey). First, heterogeneity of included studies was assessed with the Cochrane-Q-statistic and I^2 test. A P value <.10 and I^2>50% were considered as significant heterogeneity.[16] Pooled prevalence rates for functional constipation and FNRFI were calculated via a fixed-effect model in case of no heterogeneity; otherwise, the random-effect model was applied.

Subgroup analyses with univariate meta-regression were performed to assess the correlation between the prevalence and different study characteristics (eg, geographic location, age of patients, and study quality score). Pooled ORs were calculated to assess the sex distribution. Publication bias was evaluated by funnel plot and Egger tests; a P value of <.05 was considered to be statistically significant.[17]

Results

A total of 4146 search results were identified (**Figure**). After we screened titles and abstracts, 218 full-text articles were assessed for eligibility and finally 37 articles were included.[18-54] Reasons for exclusion are presented in the **Figure**. Of the 37 articles included in the qualitative analysis, 35 studies were included in the meta-analysis. One birth cohort study was excluded from the meta-analysis because of different reported prevalence rates and different sample sizes due to loss to follow-up at the last time point.[46] Another study was excluded because

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