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Gastroenterology

Prevalence of functional defecation disorders in European children: A systematic review and meta-analysis

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Abstract

Objectives: Functional defecation disorders (FDDs) are common among children worldwide. The prevalence of these disorders has not been clearly described in Europe. This study performed a systematic review and meta-analysis on the prevalence of FDD in European children and assessed geographical, age, and sex distribution and associated factors.

Methods: PubMed, Embase, Psycinfo, Cochrane Library, and Cinahl were searched from 1999 to July 2023. Included studies were (1) prospective or cross-sectional studies of European population-based samples; (2) reporting the prevalence of infant dyschezia (ID) according to Rome II, III, or IV criteria or functional constipation (FC) or functional non-retentive fecal incontinence (FNRFI) according to Rome III or IV criteria; (3) aged 0–18 years; and (4) published in English, Dutch or Spanish. PRISMA guidelines for extracting data and assessing data quality were followed.

Results: Twenty-eight studies were included. Pooled prevalence was 6.9% (95% confidence interval [CI]: 3.1%–11.9%) for ID in infants 0–12 months (9 studies, $n = 5611$), 8.17% (95% CI: 6.33%–10.22%) for FC in children <4 years (25 studies, $n = 35,189$), 11.39% (95% CI: 9.34%–14.11%) for FC in children 4–18 years, and 0.24% (95% CI: 0.07%–0.49%) for FNRFI in children 4–18 years (7 studies, n = 16,873). No sex predominance was found for FC. FC prevalence did not differ significantly when diagnosed according to Rome III versus IV. FC prevalence differed between countries, with greatest rates in Italy, Germany, and Spain. No meta-analysis could be performed on other factors associated with FDD.

Conclusions: FDD is common in European children. Future longitudinal studies are needed to provide better insight into associated factors in pathogenesis.

Abbreviations: ESPGHAN, European Society for Pediatric Gastroenterology, Hepatology, and Nutrition; FC, functional constipation; FDD, functional defecation disorder; FNRFI, functional non-retentive fecal incontinence; ID, infant dyschezia.

Michelle N. Bloem and Desiree F. Baaleman contributed equally as co-first authors.

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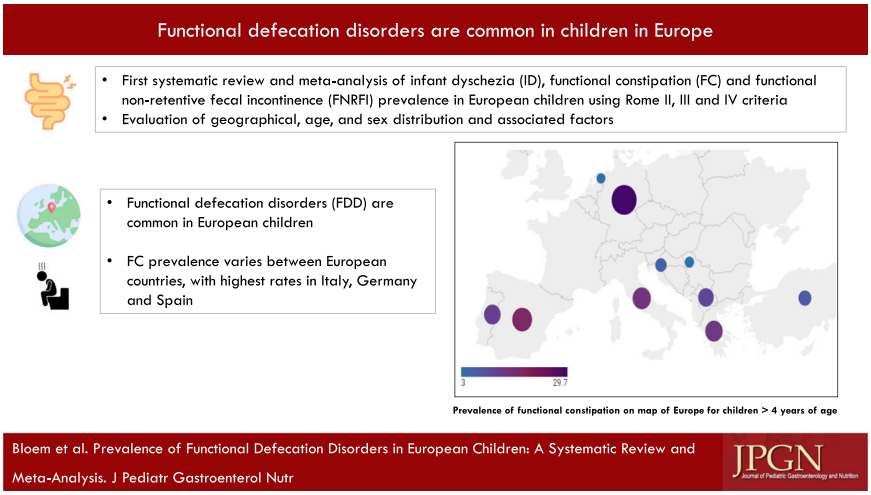

KEYWORDS

child, constipation, dyschezia, Europe, incontinence

1 | INTRODUCTION

Functional defecation disorders (FDDs) are common in childhood worldwide.[1] Symptoms of these disorders vary from mild, such as crying before the passage of soft stools, to severe with fecal impaction and daily fecal incontinence, which negatively impact the quality of life.[2] These disorders are diagnosed according to the symptom-based Rome criteria and include infant dyschezia (ID), functional constipation (FC), and functional non-retentive fecal incontinence (FNRFI).[3,4] The Rome IV criteria replaced the Rome III criteria in 2016 with minimal changes to the criteria of these disorders.[5] Supporting Information S1: eTable 1 outlines the Rome IV diagnostic criteria for each disorder, including the most relevant changes compared to previous Rome (II and III) criteria.[6,7] Previous studies have shown that the pediatric Rome II criteria were significantly more restrictive than the Rome III criteria for FC diagnosis.[8,9]

The most recent systematic review (2017) on the worldwide prevalence of FC and FNRFI, only included studies with FC or FNRFI diagnosed according to the Rome III criteria given no study using the Rome IV criteria had yet been published.[1] The systematic review reported that geographic location was significantly associated with the prevalence of FC, with a higher prevalence in the Americas and Europe (combined) compared to South and Central America (combined), and Asia. However, of the 33 included studies, only 3 originated from Europe. In addition, the prevalence of ID was not investigated.

A collaboration of members of the European Society for Pediatric Gastroenterology, Hepatology, and Nutrition and an epidemiological expert team proceeded to systematically review the prevalence of all FDD in European children. Insight into differences in FDD prevalence among European countries has the potential to identify associated factors and explain the previously found higher prevalence of FC in Europe. Therefore, our aim was to perform a systematic review and meta-analysis on the prevalence of ID, FC, and FNRFI in European children according to the pediatric Rome III and IV criteria. Secondary aims included identifying associated factors (i.e., age, sex, and geographical location) and evaluating for differences between reported prevalence according to Rome III and IV criteria.

What is Known

- Functional defecation disorders (FDDs) are common in children worldwide.
- The prevalence of FDD in Europe has not been described.

What is New

- First systematic review and meta-analysis of infant dyschezia, functional constipation (FC), and functional non-retentive fecal incontinence prevalence in European children using Rome II, III, and IV criteria.
- FDD is common in European children.
- No sex predominance found in FC. No meta-analysis could be performed for other factors associated with FDD.
- FC prevalence varies between European countries, with highest rates in Italy, Germany, and Spain.

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2 | METHODS

This systematic review was registered at PROSPERO with registration number CRD42022299526 and follows the PRISMA statement.[10]

2.1 | Search strategy and study selection

PubMed, Embase (Ovid), Psycinfo (Ovid), Cochrane Library, and Cinahl (Ebsco) were searched from 1999 (the year the Rome II criteria were published) to July 4, 2023, with the help of a clinical librarian. The full search strategy is provided in Supporting Information S1: eFile 1. Reference lists of included studies and (systematic) review articles were searched manually. Studies were eligible for inclusion if they met the following criteria: (1) prospective and cross-sectional studies of population-based samples; (2) reporting the prevalence of FC or FNRFI according to the Rome III or IV criteria, or ID according to the Rome II, III, or IV criteria; (3) in children aged 0–18 years; (4) carried out in Europe; and (5) published in English, Dutch, or Spanish. Since only minimal changes were made to ID diagnosis since Rome II, we also included studies using Rome II criteria for ID. Two reviewers (DFB and MNB) independently screened abstracts for eligibility with the use of Rayyan, a web application for SRs,[11] followed by full-text evaluation.

2.2 | Data extraction

Two authors (DFB and MNB) extracted data, including study information (author, year, country), design, diagnostic criteria, data collection method, response rate (if applicable), population information (sample size, age, and sex distribution), and results of possible associating factors. Disputes were resolved by a third author (IJK). In case reported data were unclear or incomplete (e.g., prevalence rates of one of the diagnoses were missing), the corresponding author of the respective study was contacted for clarification. If data were only reported graphically and corresponding authors were unable to provide the data, we used WebPlotDigitizer, a web-based plot digitizing tool, to extract data from figures with an accuracy of 1 decimal point.[12,13]

2.3 | Quality assessment

The quality of studies was critically appraised independently by two authors (DFB and MNB) using a modified version of the tool used in the previous systematic review.[1] Manuscripts were evaluated according to seven questions, scoring each question on a 3-point scale (no [0], partial/maybe [1], or yes [2]), with greater scores representing better methodologic quality (Supporting Information S1: eFile 2). Quality assessment scores were not used to include or exclude studies from any of the analyses.

2.4 | Data synthesis and statistical analysis

Analyses were performed in R version 4.3.2, using the meta package to produce the pooled estimates and forest plots and the metafor package to conduct meta-regression using restricted maximum likelihood estimation. The heterogeneity of included studies was assessed with Cochrane-Q-statistic and I^2 test, with $p < 0.10$ and $I^2 > 50\%$ considered as significant heterogeneity. Pooled prevalence rates were calculated via a fixed or random effects model depending on heterogeneity.

Subgroup analyses with univariate meta-regression assessed correlations between FC prevalence and different characteristics (e.g., geographic location and age). Pooled odds ratios (ORs) were calculated for sex distribution. Pairwise meta-regression compared ID prevalence between age subgroups. If cohort studies provided data on multiple time points, the mean prevalence was taken into consideration for the overall prevalence. Cohort studies only reporting a cumulative prevalence rate were excluded from meta-analyses. Significance was set at $p < 0.05$.

3 | RESULTS

We identified 5725 studies, of which 28 were eligible for inclusion (Supporting Information S1: eFigure 1). Twenty studies had a cross-sectional study design,[14–32] 7 studies were prospective cohort studies,[33–39] and 1 study was designed as a reliability study.[40]

3.1 | Quality assessment

A detailed overview of the quality assessment is available in Supporting Information S1: eFile 2. Eighteen studies (64%) scored 1 out of 2 points for their method of subject selection due to unclear exclusion of organic causes of constipation or fecal incontinence, or because children were recruited from outpatient clinics. Eighteen studies (64%) used a validated questionnaire or physician assessment to diagnose an FDD; scoring 2 out of 2 points for the diagnostic instrument used (item 4 on the quality assessment tool). Most studies lacked clear reporting on fecal impaction assessment. Five studies possibly assessed fecal impaction during physical examination,[15,20,23,27,37] one study reported to have assessed fecal impaction in a subset of

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children,[17] and one study reported the number of children in their cohort with fecal impaction.[19] Two studies received maximum scores on all qualitative criteria.[19,38]

3.2 | Infant dyschezia

Nine of the included studies reported on the prevalence of ID including 5611 children aged 0–9 months (Table 1).[15,18,29,34,35,37–39,41] One study did not specify how many infants were included, and the study was therefore excluded from meta-analysis.[18] ID diagnosis was based on Rome II criteria ($n = 1$),[41] Rome III criteria ($n = 6$),[15,18,34,35,37,38] or Rome IV criteria ($n = 2$).[29,39] Due to significant heterogeneity, data were pooled according to a random effects model, revealing a pooled prevalence of 6.89% (95% confidence interval [CI]: 3.1%–11.9%; Figure 1) for ID.

Sub-analyses were performed to calculate ID prevalence at 1, 3, and 6 months, revealing prevalence rates of 16.5% (95% CI: 3.9%–35.1%), 7.4 (91% CI: 0.0%–27.0%), and 5.0 (95% CI: 3.3%–7.0%), respectively.

Several factors were examined for their potential association with ID prevalence. One study reported an association between ID and younger age.[15] Two studies reported that the presence of ID was not associated with the subsequent development of FC.[35,38] Conflicting data were found concerning an association between ID and C-section birth.[15,34] No associations were found between ID and sex,[15] birth weight,[15] prematurity,[34] newborn hospital stay (>4 days),[34] neonatal antibiotic use,[34] time of commencing breastfeeding,[15] type of feeding,[15,34] bottle/pacifier usage,[15] or type of first consumed food.[15] Concerning maternal factors, no associations were found between ID and number of miscarriages,[15] parity,[15] education level or family type,[15] symptoms of postpartum depression,[37] "maternity blues,"[37] psychiatric symptoms[37] or adult attachment style.[37]

3.3 | Functional constipation

Twenty-five included studies reported FC prevalence including 35,189 children aged 0–18 years old (Table 1).[14–31,33–37,39] The FC prevalence reported by Kramer et al. was not taken into account as the described criteria for the diagnosis of FC did not correspond with either the Rome III or Rome IV criteria.[38] Most studies ($n = 13$; 52%) used the Rome III criteria to diagnose FC, followed by the Rome IV criteria ($n = 9$; 36%), or a modified version of the Rome III criteria ($n = 3$; 12%). FC was usually diagnosed via questionnaire ($n = 19$; 76%), often a validated questionnaire such as the QGPS-III or R4PDQ ($n = 11/19$; 58%). Only two studies reported on presence of fecal impaction, suggesting this was assessed in their population.[17,19]

Data were pooled for children <4 years ($n = 12$ studies) and >4 years ($n = 14$ studies) to evaluate for differences in FC prevalence between infants/toddlers and children/adolescents as per the Rome criteria. There was no significant difference in prevalence between these groups ($p > 0.05$). For all children, the pooled prevalence of FC was 8.76% (95% CI: 5.83–12.20).

Because of high levels of heterogeneity, the random-effects model was used to calculate pooled prevalence rates. The pooled prevalence of FC was 8.17% (95% CI: 6.33%–10.22%) for children from 0 to 4 years old (Figure 2) and 11.39% (95% CI: 9.34%–14.11%) for children 4–18 years old (Figure 3).

Rome III versus IV criteria yielded no significant FC prevalence disparities in children in both age groups. For children until 4 years old, the FC prevalence was 9.29% (95% CI: 6.11%–12.48%) according to Rome III criteria and 8.54% (95% CI: 5.35%–11.73%) according to Rome IV criteria. For children from 4 years old, the prevalence of FC was 10.54% (95% CI: 10.10%–10.99%) according to Rome III criteria and 14.23% (95% CI: 13.37%–15.13%) according to Rome IV criteria.

Sex distribution of FC was reported in 10 studies.[15,16,19,23–26,28,32,35] Six of these studies provided prevalence data of both sexes and could be included in our meta-analysis,[19,23,24,32,35] revealing a similar percentage of female cases for subjects aged 0–4 years old (50.7% [95% CI: 41.5%–59.8%]) and for 4–18 years old (53.6% [95% CI: 47.6%–59.6%]). The studies which did not provide prevalence data for either sex reported an equal sex distribution in both sexes.[15,16,25,28]

Geographic location was reported by all studies (Supporting Infomation S1: eFigures 2 and 3, eTable 2). There was a significant difference in FC prevalence among countries ($p < 0.001$). The highest prevalence of FC was 29.8% reported in 706 six- to eleven-year-old German children.[25] The lowest prevalence was 1.3% reported in 80 eleven- to twelve-month-old French children.[20]

Data of other studied associated factors with FC are summarized in Supporting Information S1: eFile 3, including parental factors, factors during infancy, food and lifestyle factors, urinary tract comorbidity, and social factors.

3.4 | Functional non-retentive fecal incontinence

Seven of the included studies reported FNRFI prevalence including 16,873 children aged 4–18 years old (see Table 1).[16,22,26,28,30,40] One study was not taken into account as they included patients only from outpatient visits and their reported prevalence of FNRFI

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TABLE 1 Study characteristics.

ID									
Authors and reference	**Ref.**	**County, region, study period**	**Study setting/design**	**In- and exclusion criteria**	**Rome criteria used**	**Method of data collection**	**Age group**	**Sample size (% female)**	**Prevalence ID, *n* (%)**
Baldassarre et al., 2021	37	Italy, Bari, 2015–2019	Birth cohort study	Mothers with pregnancy- and delivery-related medical problems were excluded from the study	Rome III	Questionnaire by physician	1 m 3 m	113 (45%)	1-mo: *n* = 17 (15%) 3-mo: *n* = 4 (3.5%)
Bekem et al., 2021	15	Turkey, İzmir, 2013	Cross-sectional outpatient clinics	Exclusion criteria were severe conditions that require urgent intervention, chronic illnesses and babies brought to the hospital without their mother. Organic diseases presenting like FGID were excluded via history, physical examination, and absence of alarm signs.	Rome III	NI	4–6 w 3–4 m 6–7 m	61 86 66 Whole group: 213 (49.3%)	*N* = 17 (27.9%) *N* = 7 (8.1%) *N* = 1 (1.5%) Whole population: *N* = 25 (11.7%)
Chouraqui et al., 2010	18	France, NI, NI	Private pediatrician visit for regular follow-up	None described	Rome III	Parent questionnaire	0–6 m	Unclear	5.6%
Kramer E et al., 2015	38	The Netherlands, National sample, 2003–2004	Well baby clinic study	Infants were included when fulfilling the following criteria: (1) duration of pregnancy ≥37 weeks; (2) weight at birth ≥2500 g; (3) postnatal hospitalization ≤2 days and (4) no congenital defect, metabolic disturbance, disorder of the gastrointestinal tract or indication of allergy to cow's milk.	Rome III	Parent questionnaire	1 m 3 m 9 m	1292	*N* = 51 (3.9%) *N* = 11 (0.9%) *N* = 11 (0.9%)
Miele E et al., 2004	41	Italy, Campania region	Primary care pediatric clinic survey	None described	Rome II	Pediatrician questionnaire	0–6 m	1020	*N* = 2 (0.20%)
Salvatore S et al., 2019	34	Italy, Varese, Bari, Catanzaro, Saronno, Milan, 2014–2016	Birth cohort study, prospective study at 1, 3, 6, and 12 months of age.	Exclusion criteria were severe acute infection or neonatal complications; known genetic syndromes, congenital and/or malformation disorders; surgery; major neurologic, immune, metabolic, cardiac, or renal diseases; absence of parental consent; or language difficulty.	Rome III	Clinical evaluation	0–6 m	934 (52.9%)	31.8% (28.8–34.9)
Steutel NF et al., 2020	29	Belgium, Brussels, 2015–2016	Well-baby visit at general pediatric clinic study	NI	Rome IV	Modified QPGS-III	0–9 m	25	*N* = 4[a] (16.0% [4.54–36.1])

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TABLE 1 (Continued)

ID

Authors and reference	Ref.	County, region, study period	Study setting/design	In- and exclusion criteria	Rome criteria used	Method of data collection	Age group	Sample size (% female)	Prevalence ID, *n* (%)
Steutel NF et al., 2020	29	Italy, Naples, 2015–2016	Well-baby visit at general pediatric clinic study	NI	Rome IV	Modified QPGS-III	0–9 m	427	*N* = 21[a] (4.92% [3.07–7.42])
Steutel NF et al., 2020	29	The Netherlands, Amsterdam, 2015–2016	Well-baby clinic study	NI	Rome IV	Modified QPGS-III	0–9 m	948	*N* = 42[a] (4.43% [3.21–5.94])
Turco R et al., 2014	35	Italy, Six regions, 2009	Birth cohort study	The infants were excluded if they had symptoms or findings suggestive of illnesses that could cause GI symptoms, a history of major abdominal surgery, acute or chronic physical disease, a developmental disability or diagnosis of organic causes of FC, or adverse effects of drugs.	Rome III	Parent questionnaire	3 m 6 m	465 (54.2%) 465 (54.2%)	*N* = 29 infants <6 months (6%)
Vlad et al., 2019	39	Romania, Ilfov County, 2018	Prospective at family medicine clinics	None described	Rome IV	R4PDQ parent report	0–9 m	174 (NI)	*N* = 12[a] 6.9%

FC and functional nonretentive fecal incontinence

Author, year	Ref.	County, region, study period	Study setting/ design	In- and exclusion criteria	Rome criteria used	Method of data collection	Fecal impaction assessment	Age group	Sample size (% female)	Prevalence FC, *N* (%)	Prevalence FNRFI, *N* (%)
Agakidis et al., 2019	36	Greece, Thessaloniki, 2017	School survey	None described	Rome III	Parent and child questionnaire	No	6–12 y 13–18 y	387 (NI) 448 (NI)	*N* = 54 (14.0%) *N* = 68 (15.1%)	-
Argibay et al., 2019	14	Spain, Gallega, NI	School survey	None described	Modified Rome III (not specified)	Parent questionnaire	No	3–9 y	1069 (48.2%)	*N* = 214 (20.0%)	-
Baldassarre et al., 2021	37	Italy, Bari, 2015–2019	Birth cohort study	Mothers with pregnancy- and delivery-related medical problems were excluded from the study	Rome III	Questionnaire by physician	NI (physician evaluation is performed)	1 m 3 m	113 (45%)	*N* = 9 (8%) *N* = 11 (9.7%)	-
Bekem et al., 2021	15	Turkey, İzmir, 2013	Cross-sectional outpatient clinics (pediatric	Exclusion criteria were severe conditions that	Rome III	NI	NI (physical examination is performed)	4–6 w 3–4 m 6–7 m	61 86 66	*N* = 0 (0%) *N* = 2 (2.3%) *N* = 4 (6.1%)	-

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TABLE 1 (Continued)

FC and functional nonretentive fecal incontinence

Author, year	Ref.	County, region, study period	Study setting/ design	In- and exclusion criteria	Rome criteria used	Method of data collection	Fecal impaction assessment	Age group	Sample size (% female)	Prevalence FC, *N* (%)	Prevalence FNRFI, *N* (%)
			gastroenterology, social pediatrics, general pediatrics)	require urgent intervention, chronic illnesses, and babies brought to the hospital without their mother. Organic diseases presenting like FGID were excluded via history, physical examination, and absence of alarm signs.					Whole group: 49.3% female Total N = 213	Whole population: *N* = 6 (2.8%)	
Bouzios et al., 2017	16	Greece, Athens, and seven other regions, 2014	School, gymnasium, and lyceum surveys	None described	Rome III	QPGS-RIII	No	6–17 y	1658 (51.8%)	*N* = 231 (13.9%)	*N* = 8 (0.48%)
Campeotto et al., 2020	17	France, national sample 2016–2017	Pediatrician and GP survey	Infants participating in a clinical trial were excluded	Rome IV	Pediatrician and GP records	In some children	0–12 m	1570	*N* = 141 9.0% (7.6–10.4)	-
Chouraqui et al., 2010	18	France, NI, NI	Private pediatrician visit for regular follow-up	None described	Rome III	Parent questionnaire	No	0–4 y	1032 (NI)	*N* = 69 (calculated) 6.7%	-
Chouraqui et al., 2019	20	France, National online survey, 2016	Pediatrician and GP survey	Healthy infant	Rome III	Physician diagnosis	NI (physician diagnosis)	1–2 m 3–4 m 5–6 m 8–9 m 11–12 m	151 132 145 90 80 Total 598 (M/F ratio 1.19)	*N* = 9[a] 6% *N* = 8[a] 6.1% *N* = 8[a] 5.5% *N* = 7[a] 7.8% *N* = 1[a] 1.3%	-
Cinquetti et al., 2021	21	Italy, Verona, 2019	School survey	None described	Rome IV	R4PDQ self-report	No	11–14 y	1594 (49.1%)	*N* = 280 (17.6%)	-

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TABLE 1 (Continued)

FC and functional nonretentive fecal incontinence

Author, year	Ref.	County, region, study period	Study setting/ design	In- and exclusion criteria	Rome criteria used	Method of data collection	Fecal impaction assessment	Age group	Sample size (% female)	Prevalence FC, *N* (%)	Prevalence FNRFI, *N* (%)
Farello et al., 2021	22	Italy, NI, NI	School survey	Incomplete questionnaires	Rome III	QPGS-RIII self-report	No	10–17 y	407 (54.1%)	*N* = 54 (13.6%)	*N* = 0 (0%)
Gulsen et al., 2020	23	Turkey, Istanbul, 2019	Pediatric health and diseases outpatient clinic or emergency clinic	We excluded children with any chronic diseases, infants that were given nutritional supplements other than vitamin D and iron supplements, and patients with chronic recurrent episodes of diarrhea or constipation and warning signs.	Rome IV	Clinical evaluation	NI (physician diagnosis)	4–24 m	400 (48.8%)	*N* = 23 (5.8%)	-
Harris et al., 2021	33	Netherlands, Rotterdam, 2012–2016	Birth cohort study	Children with IBS were excluded	m-Rome III (fecal retention missing)	Parent questionnaire	No	10 y	2818 (50.4%)	*N* = 85; 3.0%	-
Inan et al., 2007	24	Turkey, Erdine city region, 2005	Primary school survey	Incomplete questionnaires	NASPGHAN (2005) m-Rome III: (hard defecation and fecal retention missing)	Parent questionnaire	No	7–12 y	1689 (51.2%)	*N* = 122; 7.22% (6.50–7.54)	-
Laffoilie et al., 2020	25	Germany, Giessen country, 2016–2017	School survey	None described	Rome IV	Parent questionnaire	No	6–11 y	706	*N* = 210[a] 29.8% (26.4–33.3)	-
Rodríguez-Ruiz et al., 2021	26	Spain, Galicia, NI	School survey	None described	Rome III	Parent questionnaire	No	5–9 y	772 (47.2%)	*N* = 154 (20.0%)	*N* = 6 (0.78%)

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TABLE 1 (Continued)

FC and functional nonretentive fecal incontinence											
Author, year	**Ref.**	**County, region, study period**	**Study setting/ design**	**In- and exclusion criteria**	**Rome criteria used**	**Method of data collection**	**Fecal impaction assessment**	**Age group**	**Sample size (% female)**	**Prevalence FC, *N* (%)**	**Prevalence FNRFI, *N* (%)**
Russo et al., 2019	27	Italy, Naples, NI	General pediatrician visit for growth monitoring	Patients with a clinical history of organic gastrointestinal disease were excluded.	Rome IV	QPGS-RIV (including physical examination)	NI	0–3 y 4–17 y	81 (44.4%) 133 (56.4%)	*N* = 11 (13.6%) *N* = 28 (21.0%)	-
Salvatore et al., 2019	34	Italy, Varese, Bari, Catanzaro, Saronno, Milan, 2014–2016	Birth cohort study, prospective study at 1, 3, 6, and 12 months of age.	Exclusion criteria were severe acute infection or neonatal complications; known genetic syndromes, congenital and/or malformation disorders; surgery; major neurologic, immune, metabolic, cardiac, or renal diseases; or language difficulty.	Rome III	Standardized interview	No	0–12 m	934 (52.9%)	26.6% (23.7–29.5)	-
Scarpato et al., 2018	28	Croatia, National sample, 2016	School survey	Absence of alarm symptoms suggestive of an organic disease.	Rome III	QPGS-RIII parent report self-report	No	4–10 y 11–18 y	809 (51.1%) 907 (69.3%)	*N* = 37[a] (4.6%) *N* = 63[a] (6.9%)	*N* = 6[a] (0.7%) *N* = 0[a] (0%)
Scarpato et al., 2018	28	Greece, National sample, 2016	School survey	Absence of alarm symptoms suggestive of an organic disease.	Rome III	QPGS-RIII parent report self-report	No	4–10 y 11–18 y	727 (41.7%) 589 (45.1%)	*N* = 97[a] (13.4%) *N* = 93[a] (15.8%)	*N* = 3[a] (0.4%) *N* = 0[a] (0%)
Scarpato et al., 2018	28	Italy, National sample, 2016	School survey	Absence of alarm symptoms suggestive of an organic disease.	Rome III	QPGS-RIII parent report self-report	No	4–10 y 11–18 y	1070 (49.8%) 1048 (49.8%)	*N* = 199[a] (18.6%) *N* = 147[a] (14.0%)	*N* = 1[a] (0.1%) *N* = 1[a] (0.1%)
Scarpato et al., 2018	28	Macedonia, National sample, 2016	School survey	Absence of alarm symptoms suggestive of an organic disease.	Rome III	QPGS-RIII parent report self-report	No	4–10 y 11–18 y	711 (51.2%) 844 (49.7%)	*N* = 54[a] (7.6%) *N* = 188[a] (22.3%)	*N* = 1[a] (0.1%) *N* = 8[a] (0.9%)

(Continues)

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TABLE 1 (Continued)

FC and functional nonretentive fecal incontinence

Author, year	Ref.	County, region, study period	Study setting/ design	In- and exclusion criteria	Rome criteria used	Method of data collection	Fecal impaction assessment	Age group	Sample size (% female)	Prevalence FC, *N* (%)	Prevalence FNRFI, *N* (%)
Scarpato et al., 2018	28	Serbia, National sample, 2016	School survey	Absence of alarm symptoms suggestive of an organic disease.	Rome III	QPGS-RIII parent report self-report	No	4–10 y 11–18 y	828 (62.6%) 829 (46.6%)	*N* = 36[a] (4.4%) *N* = 21[a] (2.5%)	*N* = 0[a] (0%) *N* = 7[a] (0.8%)
Scarpato et al., 2018	28	Spain, National sample, 2016	School survey	Absence of alarm symptoms suggestive of an organic disease.	Rome III	QPGS-RIII parent report self-report	No	4–10 y 11–18 y	699 (51.4%) 866 (50.8%)	*N* = 125[a] (17.9%) *N* = 76[a] (8.8%)	*N* = 12[a] (1.7%) *N* = 5[a] (0.6%)
Steutel et al., 2020	29	Belgium, Brussels, 2015–2016	Well-baby visit at general pediatric clinic study	None described	Rome IV	Modified QPGS-III	No	0–12 m 13–48 m	38 77	*N* = 2 (5.26% [0.64–17.8]) *N* = 9 (11.7% [5.49–21.0])	-
Steutel et al., 2020	29	Italy, Naples, 2015–2016	Well-baby visit at general pediatric clinic study	None described	Rome IV	modified QPGS-III	No	0–12 m 13–48 m	499 253	*N* = 17 (3.41% [2.00–5.40]) *N* = 41 (16.2% [11.9–21.3])	-
Steutel et al., 2020	29	The Netherlands, Amsterdam, 2015–2016	Well-baby clinic study	None described	Rome IV	modified QPGS-III	No	0–12 m 13–48 m	1156 706	*N* = 31 (2.68% [1.83–3.78]) *N* = 51 (7.22% [5.43–9.39])	-
Strisciuglio et al., 2022	30	Croatia, national sample, 2019–2020	School survey	Children were included in the absence of alarm symptoms suggestive of an organic disease.	Rome IV	R4PDQ parent report; self-report	No	4–10 y 11–18 y	378 358	*N* = 17 (4.50% [2.64–7.11]) *N* = 23 (6.55% [4.46–9.22])	*N* = 0 (0%) *N* = 0 (0%)
Strisciuglio et al., 2022	30	Greece, national sample, 2019–2020	School survey	Children were included in the absence of alarm symptoms suggestive of an organic disease.	Rome IV	R4PDQ parent report; self-report	No	4–10 y 11–18 y	310 354	*N* = 42 (13.6% [9.98–17.9]) *N* = 39 (11.0% [7.94–14.7])	*N* = 0 (0%) *N* = 0 (0%)
Strisciuglio et al., 2022	30	Italy, national sample, 2019–2020	School survey	Children were included in the absence of alarm symptoms	Rome IV	R4PDQ parent report; self-report	No	4–10 y 11–18 y	338 368	*N* = 47 (13.9% [10.5–17.9]) *N* = 38 (10.3% [7.39–13.9])	*N* = 0 (0%) *N* = 0 (0%)

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TABLE 1 (Continued)

FC and functional nonretentive fecal incontinence											
Author, year	**Ref.**	**County, region, study period**	**Study setting/ design**	**In- and exclusion criteria**	**Rome criteria used**	**Method of data collection**	**Fecal impaction assessment**	**Age group**	**Sample size (% female)**	**Prevalence FC, *N* (%)**	**Prevalence FNRFI, *N* (%)**
				suggestive of an organic disease.							
Strisciuglio et al., 2022	30	Macedonia, national sample, 2019–2020	School survey	Children were included in the absence of alarm symptoms suggestive of an organic disease.	Rome IV	R4PDQ parent report; self-report	No	4–10 y 11–18 y	370 420	*N* = 22 (6.0% [3.77–8.86]) *N* = 73 (17.4% [13.9–21.3])	*N* = 0 (0%) *N* = 0 (0%)
Strisciuglio et al., 2022	30	Serbia, national sample, 2019–2020	School survey	Children were included in the absence of alarm symptoms suggestive of an organic disease.	Rome IV	R4PDQ parent report; self-report	No	4–10 y 11–18 y	398 450	*N* = 8 (2.0% [0.84–3.97]) *N* = 51 (11.3% [8.52–14.6])	*N* = 0 (0%) *N* = 0 (0%)
Sütcü et al., 2019	19	Turkey, Karapnar (rural area), 2016	Cross-sectional at Family Health Center	No chronic disease, no congenital aganglionic megacolon disease, no 1-month hospital stays, and no neurological disease.	Rome III	face-to-face interview	Yes	0–12 m	203 (47%)	*N* = 36[a] (17.7%)	-
Teixeira et al., 2018	31	Portugal, Braga, NI	School survey	The exclusion criteria were history of organic medical conditions or learning/ language problems.	Rome III	QPGS-RIII	No	4–18 y	400 (NI)	*N* = 50[a] (12.5%)	-
Turco et al., 2014	35	Italy, six regions, 2009	Birth cohort study	The infants were excluded if they had symptoms or findings suggestive of illnesses that could cause GI	Rome III	Parent questionnaire	No	3 m 6 m 12 m	465 (54.2%) 465 (54.2%) 402 (45.8%)	*N* = 54 (11.6% [8.85–14.9]) *N* = 64 (13.7% [10.6–17.0]) *N* = 43 (10.7% [7.85–14.1])	-

(Continues)

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TABLE 1 (Continued)

FC and functional nonretentive fecal incontinence											
Author, year	**Ref.**	**County, region, study period**	**Study setting/ design**	**In- and exclusion criteria**	**Rome criteria used**	**Method of data collection**	**Fecal impaction assessment**	**Age group**	**Sample size (% female)**	**Prevalence FC, *N* (%)**	**Prevalence FNRFI, *N* (%)**
				symptoms, a history of major abdominal surgery, acute or chronic physical disease, a developmental disability or diagnosis of organic causes of FC, and adverse effects of drugs.							
Verkuijl et al., 2023	32	Netherlands, national sample, 2021	Cross-sectional, country-wide population-based survey	Exclusion based on physically impossible combination of age, weight, and height according to the Dutch child growth charts, and/or invalid answers to open questions	Rome IV	EP-DeFeC validated parent questionnaire	No	1 m to 7 y	791 (45%)	*N* = 108 (13.7%) Boys: *N* = 58 (13.4%) Girls: N = 50 (13.9%)	*N* = 31/431 (calc) 7%
Vlad et al., 2019	39	Romania, Ilfov County, 2018	Prospective at family Medicine clinics	None described	Rome IV	R4PDQ parent report	No	0–12 mo 1–3 y	174 (NI) 134 (NI)	*N* = 11–calculated (6.3%) *N* = 12–calculated (8.9%)	-

Abbreviations: EP-DeFeC, Early Pediatric Groningen Defecation and Fecal Continence; FC, functional constipation; FRNFI, functional non-retentive fecal incontinence; GI, gastrointestinal; ID, infant dyschezia; m, months old; NI, no information; QPGS-RIII/IV, questionnaires on Pediatric Gastrointestinal Symptoms (QPGS) based on the Rome III and Rome IV criteria (QPGS-RIII and QPGS-RIV); R4PDQ, Rome IV Diagnostic Questionnaire on Pediatric Functional Gastrointestinal Disorders; w, weeks old; y, years old.

[a]Denotes calculated number of cases based on reported percentage and original sample size.

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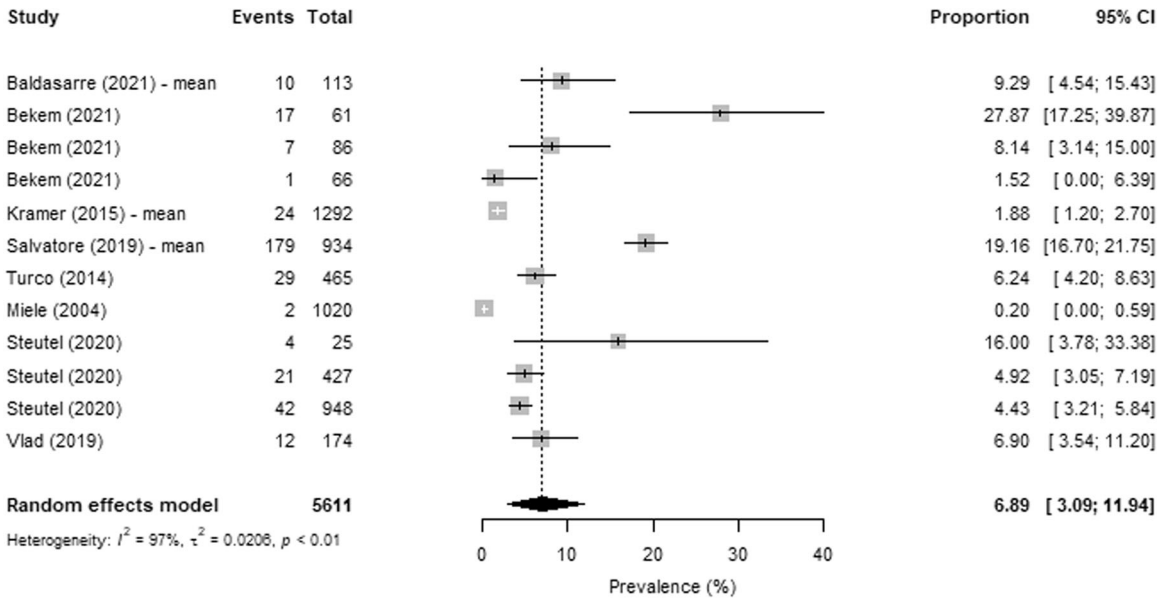

FIGURE 1 Forest plot of the prevalence of infant dyschezia reported across Europe with meta-analysis. Where studies provide more than one prevalence rate on the same group of subjects at different points in time, the average prevalence is included in the meta-analysis calculation. CI, confidence interval.

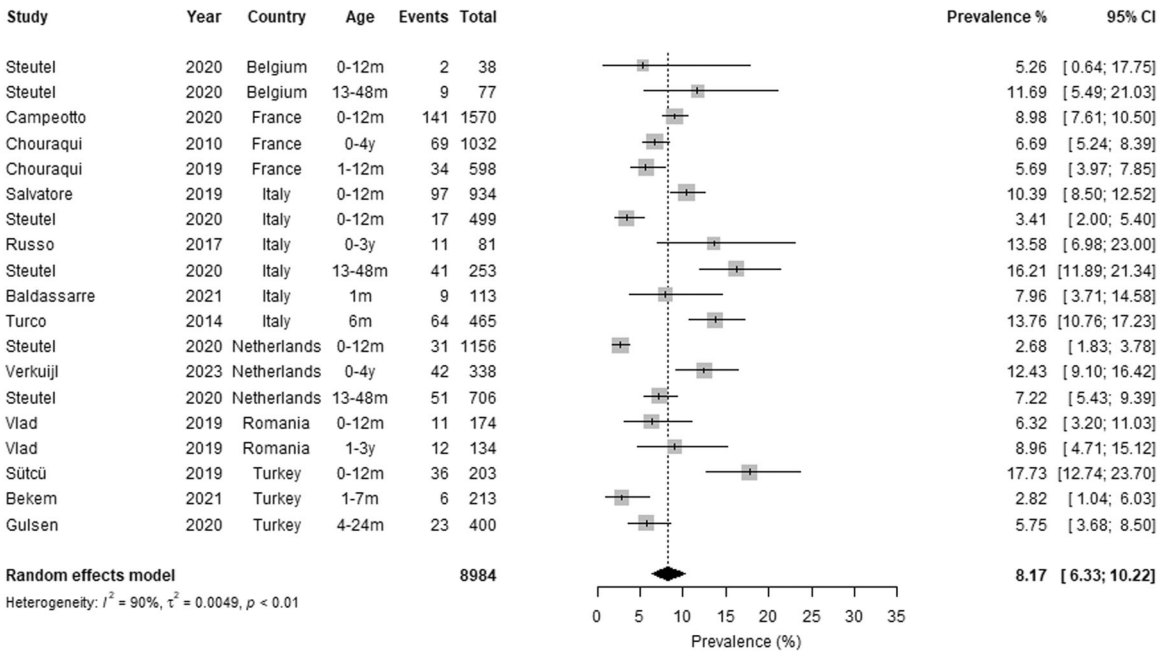

FIGURE 2 Forest plot of the prevalence of functional constipation reported. across Europe among children aged 0–4 years old with meta-analysis. Where studies provide more than one prevalence rate on the same group of subjects at different points in time, the average prevalence is included in the meta-analysis calculation. CI, confidence interval.

was excessively high.[40] All but one study used validated questionnaires to diagnose FNRFI.[26] None of the studies reported assessing for fecal impaction. According to the random-effects model, the prevalence of FRNFI ranged from 0.0% to 7.2% (Supporting Information S1: eFigure S1), and the pooled prevalence was 0.24% (95% CI: 0.07%–0.49%).

One multicountry study, including 9927 children, found that FNRFI was more prevalent in boys (0.8% vs. 0.3% in girls; $p = 0.036$)[28] reporting a significantly higher FNRFI prevalence in Spain (1.1%) compared to Italy (0.09%), Serbia (0.4%), and Macedonia (0.6%). No other factors were specifically assessed to be associated with FNRFI prevalence.

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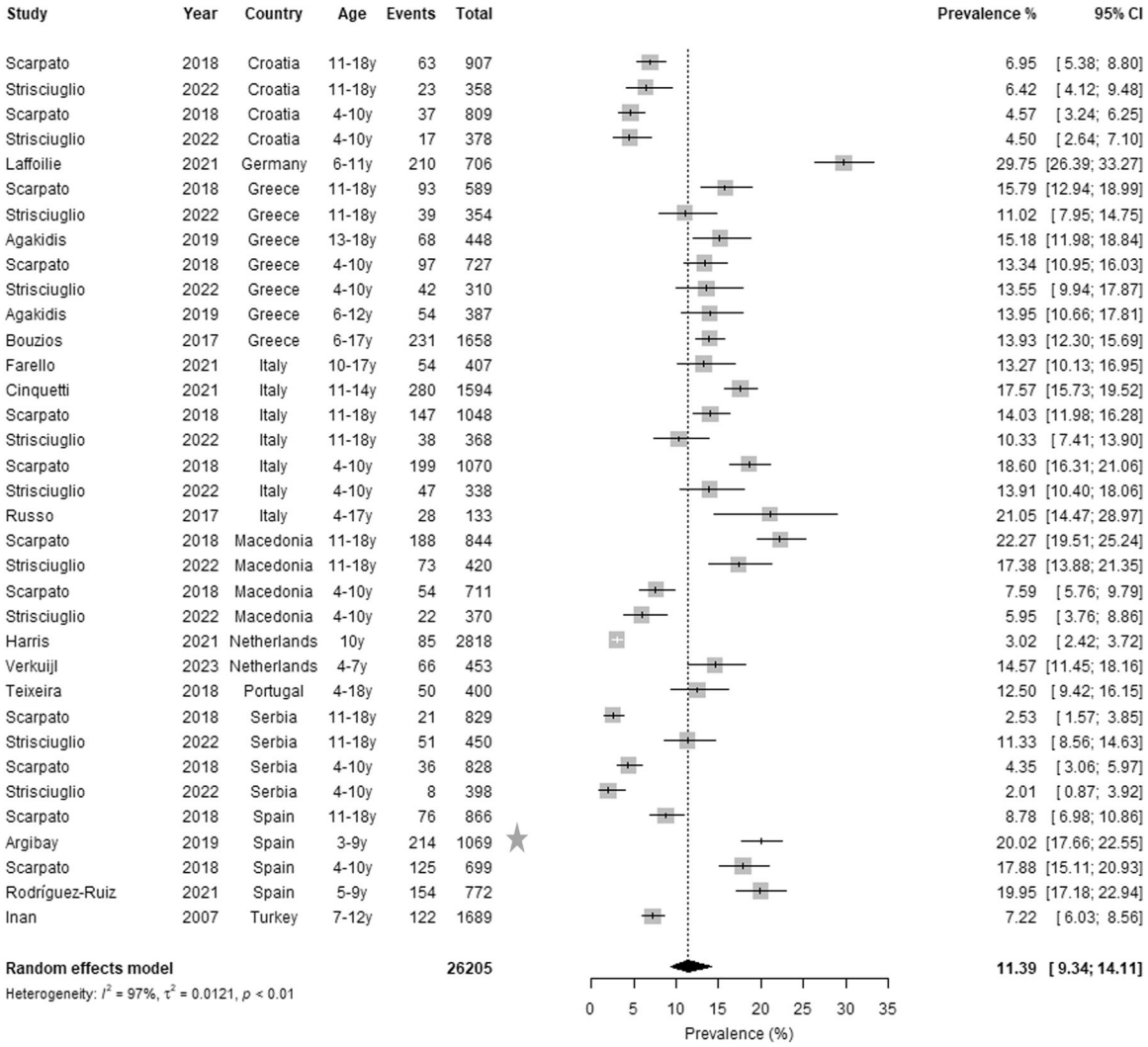

FIGURE 3 Forest plot of the prevalence of pediatric functional constipation reported. across Europe among children aged 4–18 years old, with meta-analysis. ★ denotes that the study is excluded from the meta-analysis, since the study age group overlaps the 4-year age threshold. CI, confidence interval.

4 | DISCUSSION

This systematic review and meta-analysis aimed to provide the prevalence of FDD among European children. The meta-analysis revealed a pooled prevalence of 6.9% for ID. FC was reported in 8.17% of children 0–4 years old and 11.6% of those aged 4–18 years old. The pooled prevalence of FNRFI was 0.24% in European children. Sex distribution of FC revealed no sex predominance in either age group. FC prevalence was not significantly different when diagnosed according to Rome III versus IV criteria. FC prevalence significantly differed between countries, with the greatest prevalence reported in Italy, Germany, and Spain. No meta-analysis could be performed on other associated factors.

To our knowledge, this is the first systematic review and meta-analysis providing the prevalence of FDD according to Rome criteria amongst European children. This review reveals a decrease in the prevalence of ID with increasing age, which is in line with the current theory on the etiology of ID, related to the inability to coordinate increased intra-abdominal pressure with pelvic floor muscle relaxation.[42] Infants are thought to learn how to coordinate this over time, with eventual spontaneous resolution of the behavior.[42] This is supported by the fact that ID is not associated with the development of FC.[42]

No significant differences were observed among European countries, but environmental or cultural factors may influence the etiology of ID. Although this review found no link between dyschezia and prematurity, conflicting evidence from studies in Asia[43] and South America[44] suggests regional variations. These discrepancies warrant further investigation into genetic, healthcare, and dietary factors potentially

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impacting the relationship between prematurity and dyschezia across diverse populations. Regarding FC, our results are in line with the previous systematic review on the worldwide prevalence of FC in children which included 35 studies, 3 of which are also included in our systematic review.[1] In the worldwide systematic review, the prevalence of FC in children, according to Rome III criteria, ranged from 0.5% to 32.2%, with a pooled prevalence of 9.5%. For Europe and North America, four studies were included resulting in a pooled prevalence of FC in these parts of the world of 12.4% (95% CI: 8.7–17.3). In contrast to the findings in the current review, no association was found between age and the worldwide prevalence of constipation. The previous review only included two studies that reported FC prevalence in children under 4 years old, whereas the current review found a higher FC prevalence among children aged 4–18 years, contrasting the previously reported peak incidence during potty-training age, around a median age of onset of 2.3 years old.[41,45] The lower prevalence in our current review of FC amongst European children 0–4 years old is likely due to all but two studies included in this category reported FC prevalence in very young children, specifically 0–24 months old, and therefore before the onset of potty-training for most children. In comparison to Asia, a recent systematic review of constipation prevalence in 311,660 children reports a pooled prevalence of 12.0% (95% CI: 9.3%–14.6%, $I^2 = 99.8\%$), which is in line with our results.[46] Similarly to our current review, a lower prevalence of constipation was reported in infants (<1 year old; 6.2%) compared to children (1–9 years old; 13.4%) and adolescents (10–18 years old; 14.0%) in Asia.[46]

In accordance with earlier data, our meta-analysis did not reveal a sex-related predominance.[1,46] No significant difference in FC prevalence was found depending on whether the Rome III or IV diagnostic criteria were applied, but an opposite trend was observed in both age groups. A higher prevalence was found in 4- to 18-year-old children diagnosed with the Rome IV compared to the Rome III criteria, which is in line with previous literature, although this trend has previously equally not been found to be statistically significant.[47] Rome IV, introduced a differentiation in toilet-trained and non-toilet-trained children and shortened the duration of symptoms from 2 months in Rome III, to 1 month in Rome IV. These changes likely prompt earlier diagnosis, specifically in children older than 4 years of age.[48]

The highest prevalence of FC for children 0–4 years old was reported in Italy (10.0%) and the lowest in the Netherlands (6.3%). In children 4–18 years old, the highest prevalence of FC was reported in Germany (29.8%), Spain (16.1%), and Italy (15.3%), and the lowest prevalence was in Serbia (4.2%), Croatia (5.7%), and the Netherlands (6.8%). These differences are surprising and possibly a result of reporting bias. Indeed, reporting bias could have led to either an over- or underestimation of FC prevalence in certain regions. In addition, not all European countries were included in this analysis due to the absence of published data. Paradoxically, some countries with a diet typically high in fiber, such as Spain and Italy, report high prevalences of FC, although a Mediterranean diet has been reported to be negatively associated with FC.[16,36] Additional factors such as socioeconomic factors, parental factors, and dietary and lifestyle habits and cultural norms could influence regional differences and warrant further investigation. For instance, discussing bowel habits may be taboo, leading to underreporting in some countries. Of note, only 13 countries were represented amongst all 44 European countries, challenging the accuracy of the current prevalence distribution.

Many studies have investigated factors potentially associated with FC, though causality cannot be established due to the cross-sectional design of most studies. Being bullied, physical or psychological trauma, and personal health problems were positively associated with FC.[16,24] This aligns with previous findings associating negative psychosocial factors with a greater prevalence of FDD.[1] Regarding lifestyle factors, FC was positively associated with increased TV exposure and negatively associated with an increased amount of physical activity.[16,24] Adherence to a Mediterranean diet and higher fiber consumption were negatively associated with FC,[16,36] supporting a diet higher in fiber and regular physical activity contribute to healthy stool passage.

Our reported pooled prevalence of FNRFI (0.24%; 95% CI: 0.1%–0.5%) in European children is in line with the worldwide pooled prevalence of FNRFI (0.4%; 95% CI: 0.2–0.6).[1] A recent Dutch study (2023) however reported a pooled prevalence of FNRFI of 7.2% among children aged 4 and 7 years.[32] This higher prevalence may be due to the younger age group studied and the use of the Rome IV criteria, which have a shorter diagnostic window (1 month) compared to the Rome III criteria (2 months).[1,32] Although not significant, a trend towards lower FNRFI prevalence with increasing age was observed. This is in line with a Dutch long-term follow-up study showing that more than 85% of FNRFI patients recover beyond puberty.[49] Male-predominance of FNRFI has been described in the literature with male:female ratios ranging from 3:1 to 6:1.[50] Our systematic review includes one large study confirming a significant male-predominance of FNRFI, whereas other studies found no significant sex-related predominance.[51,52] With such an uncommon disorder, it may be difficult to identify these sex-related differences in epidemiological studies.

This is the first systematic review on the prevalence of all FDD in European children, including ID, encompassing the most recent Rome IV criteria. The large sample size provides insight into differences in

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prevalence between European countries and factors associated with a higher prevalence of these disorders, as well as differences in prevalence for FC when considering the Rome III and IV criteria.

The main limitations of this systematic review arise from the nature of the included studies. The majority did not exclude organic causes of FDD beforehand, nor assessed presence of alarm symptoms or signs of organic causes during diagnostic assessment, potentially overestimating the prevalence of FDD. Given the rarity of organic etiologies, the impact on estimates is likely minimal. Since most studies used questionnaires to measure the prevalence of FDD, limitations with the use of these questionnaires to measure prevalence have to be taken into account.[53] Additionally, quality assessment was performed using a self-developed instrument. The lack of fecal impaction assessment in most studies may have resulted in underestimation of FC prevalence, and overestimation of FNRFI prevalence. Moreover, even though questionnaires were completed anonymously, children may feel ashamed about their symptoms, especially when completing the questionnaire in school, which may have resulted in an underestimation of the prevalence of FC and FRNFI.

This systematic review shows that FDDs are common among European children. However, results were constrained by inadequate reporting and limitations of self-reported questionnaires. Future longitudinal studies are needed to provide better insight into associated factors in the pathogenesis of FDDs.

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CONFLICT OF INTEREST STATEMENT

Marc A. Benninga is a consultant for Mallinckrodt, Shire, Danone, FrieslandCampina, HIPP, Norgine, Coloplast, United Pharmaceuticals, and Wellspect. All honoraria were paid to the hospital (Amsterdam UMC). Ilan J. N. Koppen received research funding from Wellspect Healthcare and The EuroQol Research Foundation. N. Thapar has participated as a speaker and/or consultant for Danone/Nutricia, Reckitt Benckiser, Biogaia, Abbott Nutrition and Nestle. The remaining authors declare no conflicts of interest.

DATA AVAILABILITY STATEMENT

All our data and metadata will be made available upon request.

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