

Parental characteristics and functional constipation in children: a cross-sectional cohort study

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ABSTRACT

Objective To evaluate personality, psychological health, physical health and childrearing practices in mothers and fathers of children with functional constipation (FC) compared with mothers and fathers of healthy controls.

Design Cross-sectional cohort study.

Setting Outpatient paediatric gastroenterology clinic at a tertiary hospital in the Netherlands.

Patients Parents of children (4–16 years) presenting with FC were included between January 2010 and August 2012. Participating parents were asked to recruit parents of another child of the same age without FC as their own controls. Data of 116 mothers and 115 fathers of 127 children with FC, and 84 mothers and 73 fathers of 91 children without FC were collected.

Main outcome measures Parental characteristics were evaluated by using the NEO Five-Factor Inventory to assess personality, the Brief Symptom Inventory and Physical Symptom Checklist to assess psychological and physical health and the Ghent Parental Behavior Scale to assess childrearing practices.

Results Mothers of constipated children had significant higher scores on the neuroticism personality factor and reported higher rates of overall psychological distress and depression. Both mothers and fathers of children with FC reported significant more physical symptoms than parents of children without FC. Mothers of children with FC showed more positive childrearing practices compared with controls.

Conclusions Personality, psychological and physical health, and childrearing practices differ significantly between parents of children with FC and parents of control subjects. Parental factors should be taken into account when evaluating children with FC.

INTRODUCTION

Constipation is a common disorder in childhood with a worldwide prevalence rate up to 29.6%.¹ In more than 90% of children with symptoms of constipation, no organic cause can be identified and these children are considered to have functional constipation (FC).² The diagnosis of FC is based on the Rome criteria. Children may present with infrequent, painful, hard stools, faecal incontinence and stool withholding behaviour. In

What is already known on this topic?

- ▶ The association between functional constipation in children and parental childrearing attitudes has been acknowledged. However, research is limited.
- ▶ Personality dimensions have been shown to differ between mothers of children with constipation and control subjects. Data about fathers are lacking.

What this study hopes to add?

- ▶ Our data show that childrearing practices, physical and psychological health of mothers of children with functional constipation (FC) differ from mothers of healthy controls.
- ▶ Fathers of children with constipation report more physical complaints than fathers of healthy controls.
- ▶ Parental factors should be addressed when evaluating a child with FC. Family-based treatment strategies should be considered.

2016, the Rome Foundation published the new Rome IV criteria for FC, an updated version of the Rome III criteria in which only the duration of symptoms has been changed.^{3,4}

Multiple factors are thought to play a role in the aetiology and persistence of symptoms, including genetic and environmental factors and the co-occurrence of behavioural problems in constipated children.^{5–7} In most studies, constipated children have been the main subject of investigation and little to no attention has been paid to the parents. Using the Symptom Checklist-90 (SCL-90) revised version, Ozokutan *et al* found no significant differences in psychological characteristics between parents of 32 children with FC and parents of 30 controls.⁸ In 2009, an Iranian study showed that personality dimensions of mothers of 150 children with FC differed from mothers of 150 controls.⁹

Until now, research on parents of constipated children has been limited and most studies lack data with respect to fathers. Identification of personality patterns, levels of psychological distress, specific parental health problems and childrearing practices in mothers and fathers of constipated children might lead to an adaptation of the current treatment strategies for children with FC. Such information may provide insights into the complex pathophysiology of childhood FC, identify factors that perpetuate symptoms and uncover the psychosocial effects of FC on families. Thus, in the present study, we aimed to compare the abovementioned characteristics between mothers and fathers of children with FC and mothers and fathers of healthy controls.

METHODS

Study population

Between January 2010 and August 2012, we enrolled parents of paediatric patients with FC, aged 4–16 years, presenting at the outpatient paediatric gastroenterology clinic of a tertiary hospital in Amsterdam, the Netherlands. Children had to have a diagnosis of FC according to the internationally accepted Rome III criteria. Parents were invited to participate by their treating physician. Parents with insufficient Dutch language proficiency were excluded from participation.

Participating parents were asked to recruit parents of another child (healthy control) of the same age from their own environment without FC or other chronic disorders to serve as their own controls. In order to assess their eligibility for participation, potential control parents were contacted and inquired about signs of FC according to the Rome III criteria and the presence of other chronic disorders in their child.

Procedures

If parents were eligible for participation, both mothers and fathers from the two study groups were asked to fill out several questionnaires at home in a secured online testing environment. The following data were collected: demographics, the NEO Five-Factor Inventory (NEO-FFI),¹⁰ the Brief Symptom Inventory (BSI),¹¹ the Physical Symptom Checklist (PSC)¹² and the Ghent Parental Behavior Scale (GPBS).¹³ Personal codes and passwords were provided by email. The mean raw scores of the questionnaires were calculated. For interpretation and comparison purposes, the raw scores were converted into age-specific or sex-specific standardised scores and compared with the norm scores of the (Dutch) general population.^{12–15} The study was approved by the local Medical Ethics Committee of the Academic Medical Center.

Questionnaires

Demographics

To assess demographic characteristics, we used a questionnaire which consisted of questions regarding sex and

age, ethnicity, marital status and educational level of both children and parents.

Personality dimensions: NEO-FFI

Personality dimensions were evaluated by the Dutch version of the NEO-FFI, a validated and shortened questionnaire based on the NEO Personality Inventory. It provides a measure of the five basic personality factors: neuroticism, extraversion, openness to experience, agreeableness and conscientiousness by 60 items that are scored on a 5-point Likert scale.

Psychological distress and symptoms: BSI

The BSI is adapted from the longer SCL-90. It provides an overview of current (previous 7 days) psychological distress and symptoms on eight different subscales: somatisation, obsessive-compulsive, interpersonal sensitivity, depression, anxiety, hostility, phobic anxiety, paranoid ideation and psychoticism.¹¹ Higher scores indicate more severe symptoms. An overall score, which is an average rating of all 53 items, can be calculated as an overall indication of psychological functioning. Psychometric properties of the BSI have been demonstrated to be good.¹¹

Physical health: PSC

The PSC quantifies the number of reported physical symptoms in the preceding week.¹² The checklist consists of 51 non-gender-specific physical symptoms mentioned in the Diagnostic and Statistical Manual of Mental Disorders, Third Edition classification.¹⁶ Symptoms are rated on a severity scale ranging from 0 (never) to 3 (regularly), symptoms are considered to be bothersome when scored 2 or 3. The total score describes the number of bothersome symptoms present during the previous week. Items can be divided into different organ system categories.

Childrearing practices: GPBS

The GPBS was developed to provide insight into concrete parental behaviour.¹⁷ It includes nine parenting subscales: discipline, ignoring of unwanted behaviour, harsh punishment, positive parental behaviour, teaching rules, autonomy, monitoring, material rewarding and inconsistent discipline. In this study, parents were asked to rate their own parenting behaviour. Psychometric properties of the GPBS have reported to be sufficient to good.¹⁷

Outcome measures

The main outcome measures of this study were the differences in personality dimensions, psychological distress, physical health and childrearing practices between mothers and fathers of children with childhood constipation and mothers and fathers of control subjects.

Statistical analysis

The statistical program SPSS V.16.0 (SPSS Inc) was used for the statistical analyses. Normally distributed data are reported as mean with SD and compared between groups by independent t-tests. Skewed continuous

Table 1 Baseline characteristics of children and their participating parents

	Children with functional constipation (N=127)	Control children (N=91)	p-Value
Boys, n (%)	70 (55)	54 (56)	0.640
Mean age child in years (SD)	9.4 (±3.6)	9.1 (±3.3)	0.470
Mean duration of symptoms in years (SD)	5.1 (±3.4)	n/a	–
Presence of faecal incontinence, n (%)	99 (78)	n/a	–
Presence of withholding behaviour, n (%)	75 (59)	n/a	–
No of participating parents, n (%)			
Both mother and father	104 (82)	66 (73)	0.075
Only mother	12 (9)	18 (20)	0.038
Only father	11 (9)	7 (8)	0.780
Children with divorced/separated parents, n (%)	25 (20)	6 (7)	0.006*
Highest education parents, n (%)			
Primary school	0 (0)	1 (1)	0.417
Secondary school	17 (13)	11 (12)	0.839
Vocational education	81 (64)	46 (51)	0.053
University	23 (18)	33 (36)	0.003*
Other	6 (5)	1 (1)	0.243
Having at least one unemployed parent	23 (18)	12 (13)	0.356
	Parents of children with functional constipation (N=231)	Parents of control children (N=157)	p-Value
Mothers, n (%)	116 (50)	84 (53)	0.536
Fathers, n (%)	115 (50)	73 (47)	0.536
Caucasian, n (%)	220 (95)	150 (96)	1.000
Mean age parents in years (SD)	41.6 (±6.3)	40.7 (±6.0)	0.313

*Difference $p < 0.01$ between the constipation and control group.
n/a, not applicable.

data are reported as medians with IQR and compared between groups by Mann-Whitney U analyses. In order to compare proportions of parents with scores above certain cut-off points between groups, χ^2 analyses or Fisher's exact tests were performed. The significance level was set at $p < 0.01$ to correct for multiple testing between the study groups. To describe the internal consistency of the psychometric questionnaires in our study, Cronbach's α tests were performed. A Cronbach's $\alpha \geq 0.6$ was considered to be acceptable.

RESULTS

In total, 170 mothers and 170 fathers of 170 children with FC agreed to participate, of whom 116 mothers and 115 fathers of 127 children with FC completed the study (response rate 68%). In the control group, 104 mothers and 104 fathers of 104 children agreed to participate, and 84 mothers and 73 fathers of 91 children completed the study (response rate 75%). No parents were excluded based on the exclusion criteria. Baseline characteristics

of participating parents and their children are reported in (table 1).

Mothers of constipated children versus mothers of controls

Outcomes of mothers of children with FC and mothers of controls can be found in (table 2).

Personality

On the NEO-FFI, mothers of constipated children showed significantly higher raw scores, and sex-specific and age-specific norm scores for neuroticism compared with mothers of controls.

Psychological distress

Raw scores of the BSI overall score were found to be significant higher in mothers of children with FC compared with mothers of healthy children. Moreover, both raw and sex-specific norm scores for depression were significantly higher in mothers of constipated children.

Table 2 Differences between parents of constipated children and parents of controls

	Mothers		Fathers	
	Mothers of children with functional constipation, N=116	Mothers of healthy controls, N=84	Fathers of children with functional constipation, N=115	Fathers of healthy controls, N=73
Mean age (SD)	40.2 (±5.8)	40.0 (±5.8)	43.1 (±6.1)	41.5 (±6.1)
Caucasian, n (%)	111 (95.7)	77 (91.7)	109 (94.8)	73 (100)
Unemployed, n (%)	19 (16.3)	8 (9.5)	6 (5.2)	4 (5.5)
NEO-FFI				
Raw scores (mean (SD))				
Neuroticism	31.1 (8.1)	28.2 (6.3)	28.8 (8.1)	27.6 (5.9)
Extraversion	42.7 (5.8)	43.1 (5.4)	42.4 (6.5)	42.7 (4.8)
Openness to experience	36.0 (5.6)	37.0 (6.0)	35.2 (6.2)	36.3 (7.0)
Agreeableness	47.0 (4.6)	47.3 (3.9)	44.0 (5.3)	42.9 (5.5)
Conscientiousness	47.1 (5.1)	46.5 (4.6)	46.9 (6.2)	46.5 (5.2)
Sex dependent norm scores (mean (SD))				
Neuroticism	4.8 (2.0)	4.1 (1.6)	4.6 (2.0)	4.6 (1.5)
Extraversion	5.7 (1.8)	5.8 (1.7)	5.8 (2.1)	5.9 (1.5)
Openness to experience	4.9 (1.8)	5.1 (1.8)	5.0 (1.9)	5.3 (2.1)
Agreeableness	5.8 (1.8)	5.9 (1.5)	5.6 (1.9)	5.2 (2.1)
Conscientiousness	5.6 (1.9)	5.5 (1.8)	5.4 (2.1)	5.3 (1.8)
Age dependent norm scores (mean (SD))				
Neuroticism	5.0 (1.9)	4.3 (1.6)	4.3 (2.0)	4.2 (1.5)
Extraversion	5.5 (1.8)	5.5 (1.6)	5.5 (2.0)	5.5 (1.5)
Openness to experience	4.8 (1.8)	5.0 (1.7)	4.6 (2.0)	4.8 (2.2)
Agreeableness	5.9 (1.8)	5.9 (1.5)	4.8 (1.9)	4.4 (1.9)
Conscientiousness	5.5 (1.9)	5.3 (1.8)	5.4 (2.2)	5.3 (1.9)
Brief Symptom Inventory (BSI)				
Raw scores (median (IQR))				
Somatisation	0.14 (0–0.43)	0.0 (0–0.3)	0.0 (0–0.2)	0.0 (0–0.1)
Obsessive-compulsive	0.21 (0–0.5)	0.17 (0–0.5)	0.17 (0–0.5)	0.17 (0–0.5)
Interpersonal sensitivity	0.25 (0–0.75)	0.25 (0–0.5)	0.25 (0–0.5)	0.25 (0–0.5)
Depression	0.17 (0–0.5)	0.0 (0–0.3)	0.0 (0–0.5)	0.0 (0–0.3)
Anxiety	0.17 (0–0.5)	0.17 (0–0.3)	0.0 (0–0.3)	0.17 (0–0.3)

Continued

Table 2 Continued

	Mothers		Fathers		
	Mothers of children with functional constipation, N=116	Mothers of healthy controls, N=84	Fathers of children with functional constipation, N=115	Fathers of healthy controls, N=73	
			p-Value	p-Value	
Hostility	0.2 (0.2-0.4)	0.2 (0.2-0.4)	0.044	0.2 (0-0.4)	0.877
Phobic anxiety	0.0 (0-0.2)	0.0 (0-0.2)	0.103	0.0 (0-0.2)	0.082
Paranoid ideation	0.2 (0.2-0.6)	0.2 (0-0.4)	0.199	0.2 (0-0.4)	0.988
Psychoticism	0.0 (0-0.4)	0.0 (0-0.4)	0.065	0.0 (0-0.2)	0.465
Overall score	0.19 (0.1-0.4)	0.15 (0.1-0.3)	0.011*	0.17 (0.1-0.3)	0.155
Sex-specific norm scores (median (IQR))					
Somatisation	4 (3-5)	3 (3-4)	0.018	4 (4-6)	0.023
Obsessive-compulsive	4 (3-4)	3 (3-4)	0.177	3 (3-5)	0.194
Interpersonal sensitivity	4 (3-5)	4 (3-4)	0.084	4 (3-5)	0.232
Depression	3 (3-5)	3 (3-4)	0.006*	4 (4-5)	0.194
Anxiety	3 (3-5)	3 (3-4)	0.059	3 (3-5)	0.072
Hostility	4 (4-5)	4 (4-5)	0.052	4 (3-5)	0.710
Phobic anxiety	4 (4-5)	4 (4-5)	0.281	4 (4-5)	0.033
Paranoid ideation	4 (3-5)	4 (3-4)	0.154	4 (3-5)	0.623
Psychoticism	4 (4-5)	4 (4-5)	0.168	4 (4-4)	0.816
Overall score	3 (2-5)	3 (2-4)	0.035	3 (2-5)	0.371
Physical Symptom Checklist (PSC)					
Raw scores (median (IQR))					
Total PSC score†	1 (0-4)	0 (0-1)	0.000*	0.9 (0-2)	0.005*
% greater than sex-specific cut-off score for general population (>p75)†	34 (29%)	12 (14%)	0.017	31 (27%)	0.110
No of symptoms on subscale (median (IQR))					
Autonomic	0 (0-0)	0 (0-0)	0.004*	0 (0-0)	0.022
Neurological	0 (0-1.8)	0 (0-0)	0.002*	0 (0-0)	0.401
Musculoskeletal	0 (0-1)	0 (0-0)	0.022	0 (0-0)	0.972
Gastrointestinal	0 (0-1)	0 (0-0)	0.004*	0 (0-0)	0.027
Warm/cold/urogenital	0 (0-0)	0 (0-0)	0.010*	0 (0-0)	0.133

Continued

Table 2 Continued

	Mothers		Fathers	
	Mothers of children with functional constipation, N=116	Mothers of healthy controls, N=84	Fathers of children with functional constipation, N=115	Fathers of healthy controls, N=73
				p-Value
Ghent Parental Behaviour Scale (GPBS)				
Raw scores (mean (SD))				
Autonomy	11.4 (2.0)	11.6 (1.7)	11.2 (2.1)	0.469
Discipline	18.4 (5.3)	16.9 (3.5)	17.7 (4.5)	0.029
Positive parental behaviour	47.2 (7.5)	46.1 (5.5)	43.2 (7.1)	0.298
Harsh punishment	4.5 (1.9)	4.7 (1.8)	4.7 (1.7)	0.579
Monitoring	16.8 (5.2)	15.9 (4.7)	13.4 (4.4)	0.282
Teaching rules	26.0 (4.2)	25.8 (3.3)	24.9 (3.9)	0.727
Ignoring of unwanted behaviour	6.5 (2.2)	6.6 (2.5)	7.1 (2.7)	0.595
Material rewarding	7.6 (1.9)	7.4 (2.0)	8.1 (1.9)	0.474
Inconsistent discipline	7.7 (2.4)	7.9 (2.4)	8.2 (2.5)	0.720
Sex-specific norm scores (mean (SD))				
Autonomy	3.1 (0.9)	3.2 (0.8)	3.0 (0.9)	0.518
Discipline	3.2 (0.7)	3.0 (0.7)	3.3 (0.7)	0.054
Positive parental behaviour	3.4 (0.8)	3.1 (0.8)	3.3 (0.8)	0.005*
Harsh punishment	2.2 (0.6)	2.4 (0.8)	2.4 (0.8)	0.231
Monitoring	2.8 (1.1)	2.8 (1.0)	2.7 (0.8)	0.858
Teaching rules	2.6 (1.3)	2.3 (1.2)	2.7 (1.1)	0.101
Ignoring of unwanted behaviour	2.8 (0.5)	2.9 (0.6)	2.6 (1.0)	0.433
Material rewarding	3.0 (0.7)	2.9 (0.8)	3.2 (0.7)	0.233
Inconsistent discipline	2.7 (0.9)	2.8 (0.8)	2.8 (1.0)	0.453
				p-Value
				0.414
				0.279
				0.440
				0.222
				0.213
				0.626
				0.891
				0.058
				0.954
				0.116
				0.471
				0.875
				0.694
				0.413
				0.512
				0.675
				0.088
				0.970

*Difference $p < 0.01$ between the constipation and control group.†No of bothersome symptoms (often or regularly present during the previous week).
NEO-FFI, NEO-Five Factor Inventory.

Table 3 Internal consistency of the questionnaires

Questionnaire	Internal consistency (Cronbach's α)	Internal consistency of subscales (Cronbach's α)
NEO-FFI	Acceptable (0.7)	0.6–0.9
BSI	Excellent (>0.9)	0.6–0.8
PSC	Excellent (>0.9)	0.6–0.8
GPBS	Good (0.8)	0.6–0.9

BSI, Brief Symptom Inventory; GPBS, Ghent Parental Behaviour Scale; NEO-FFI, NEO-Five Factor Inventory, PSC, Physical Symptom Checklist.

Physical symptoms

Median total PSC scores were significantly higher in mothers of patients, indicating that bothersome physical complaints were more prevalent in mothers of constipated children compared with mothers of controls. On all organ subscales except the musculoskeletal subscale, the median number of symptoms of mothers of children with FC exceeded the scores of mothers of controls significantly.

Childrearing practices

For the GBPS, sex-specific scores were only significantly different between the two groups of mothers on the domain of positive parental behaviour.

Fathers of constipated children versus fathers of controls

There were no statistically significant differences between the two groups of fathers in personality, psychological distress and childrearing practices (table 2). The median total number of bothersome physical symptoms according to the PSC questionnaire was significantly higher in fathers of constipated children compared with fathers of controls.

Internal consistency of questionnaires

As shown in (table 3), the internal consistency of all questionnaires in this study was considered to be acceptable.

DISCUSSION

In this study, we explored characteristics of parents of children with FC and parents of controls. Mothers of constipated children had significantly higher scores on the neuroticism personality factor and reported higher rates of overall psychological distress and depression. Both mothers and fathers of children with FC reported significantly more bothersome physical symptoms than parents of children without FC. Mothers of children with FC showed more positive childrearing practices compared with controls. These results suggest that parental characteristics of children with FC differ from those of parents of healthy controls.

Our results are not entirely in line with the previous literature. Although Farnam *et al* also found differences

in personality characteristics, their study showed that mothers of constipated children scored lower on neuroticism compared with mothers of controls.⁹ Instead, mothers in our study demonstrated higher scores on the personality domains extraversion, conscientiousness and agreeableness. We were not able to find a good explanation for the differences between the study of Farnam *et al* and the present study.

Psychological health problems occurred more often in mothers of children with FC compared with controls. In contrast to these results, Ozukutan *et al* found no differences in psychological distress levels in parents of constipated children compared with parents of controls.⁸ However, this study included a smaller sample and excluded parents of children with faecal incontinence, which is known to be an important stressor in families of constipated children.¹⁸

Parents of constipated children reported significantly more physical symptoms. These results concur with previously published data from mothers of children with functional abdominal pain.¹⁹ Furthermore, anxiety and depression were more common among these mothers, which is in line with our results. Unfortunately, published data on this topic concerning fathers are lacking.

Our research group has previously shown that parental childrearing attitudes are associated with the frequency of symptoms in children with constipation.²⁰ High levels of frustration or irritability towards the child were shown to have a negative effect on faecal incontinence symptoms. Moreover, parental beliefs about faecal incontinence and constipation can change after consultation of a medical professional, resulting in decreased feelings of blame and punishment.²¹ Parents of children with constipation participating in our study had already consulted a medical professional, which could have resulted in adapted positive parenting behaviour. These results emphasise the need for physicians to provide parents of children with FC with appropriate education about the condition and the effect of certain childrearing practices. By avoiding blame and praising positive defecation behaviour, education about parental influence might enhance treatment.

The differences between the parents of children with FC and controls in our study are remarkable, but due to the cross-sectional study design the causality dilemma remains. One could hypothesise that the identified differences in parental characteristics may be a consequence of having a child with FC. Having a child suffering from constipation with faecal incontinence, abdominal pain and frequent school absenteeism, may lead to psychological distress in parents. FC often is a chronic condition and parents of chronically ill children have been shown to have an impaired health-related quality of life and report higher depression and anxiety scores compared with controls.^{22 23} On the other hand, FC symptoms in children may in some way be affected by parental characteristics. For example, it has been found that the presence of depressive symptoms in parents is associated with school absenteeism and increased healthcare usage in

their children.²⁴ Moreover, it has been shown that children of parents with somatic complaints report more health problems than children of healthy parents.^{25–27}

It is remarkable that most differences found in our study were found in mothers. It has been demonstrated that fathers are significantly less involved in childrearing than mothers.^{28–29} Although this practice is currently changing, mothers in general still tend to spend more time with their children as fathers are commonly the highest wage-earners. Especially mothers of chronically ill children tend to work fewer hours per week than mothers of healthy children.³⁰ Therefore, mothers are primarily facing the daily struggles connected to having a child with FC and this might explain the fact that most differences were demonstrated between the mothers of the study groups.

A major strength of our study is the use of a control group with parents of age-comparable, healthy children. However, several limitations should be taken in mind when interpreting the results. Our study may be at risk of selection bias towards more severe cases of constipated children since our study took place in a tertiary centre. However, nearly a third of all patients were directly referred by a primary care physician without consultation of a paediatrician. For this study, we asked parents to recruit their own controls. Although determinants of study participation are poorly investigated, it could be hypothesised that people are more likely to enrol in research if there is potential benefit for themselves or for people they know. By recruiting controls through their acquaintances, we attempted to enhance participant enrolment and to recruit controls from comparable social, economical and cultural backgrounds. However, by using this method of patient selection, there is a possibility of selection bias since the sampling was not performed at random. Furthermore, the proportion of parents that attended university was higher in the control group and parents of children with FC were more frequently separated than parents of controls. These differences may be of importance because studies have identified low socio-economic status and being a single parent as risk factors for impaired parental well-being and health-related quality of life.^{31–32} Yet, the control group appeared to be a reasonably representative for the general population as their scores were largely comparable with the norm scores of the questionnaires.

The demonstrated differences between parents, especially mothers, of constipated children and controls are important to keep in mind when evaluating and treating children with FC. In children with FC refractory to intensive medical treatment, parental factors which may affect treatment success should be evaluated and a more family-based multidisciplinary treatment strategy should be considered. Education about the possible influence of parental factors in the persistence of symptoms of FC might enhance success of treatment. Also, parents may benefit from the acknowledgement of the impact and stress caused by their child's FC on their own well-being.

Future research should focus on longitudinal prospective studies in order to answer the question of cause and effect in light of these observations. These studies should prospectively assess both psychological and physical health in young children with FC and their parents over time, evaluate the effect of therapeutic strategies and compare the results with a matched control group. Furthermore, randomised controlled trials are needed to investigate the effect of family-based intervention strategies.

Contributors BP, conceptualised the study, collected data, performed data analyses, drafted the initial manuscript and approved the final manuscript as submitted. MHV, interpreted data, critically reviewed and revised the manuscript and approved the final manuscript as submitted. IJNK, interpreted data, critically reviewed and revised the manuscript and approved the final manuscript as submitted. MvD, participated in the design of the study, critically reviewed and revised the manuscript and approved the final manuscript as submitted. MAG, participated in the design of the study, critically reviewed and revised the manuscript and approved the final manuscript as submitted. CDL, critically reviewed and revised the manuscript and approved the final manuscript as submitted. MAB, conceptualised the study, critically reviewed and revised the manuscript and approved the final manuscript as submitted.

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