

# Prevalence of suspected child abuse in children with constipation: a case-control study

Mana H Vriesman <sup>1</sup>, Thekla F Vrolijk- Bosschaart,<sup>2</sup> Ramón J L Lindauer,<sup>3,4</sup> Johanna H van der Lee,<sup>5,6</sup> Sonja Brilleslijper-Kater,<sup>2</sup> Arianne H Teeuw,<sup>2</sup> Marc A Benninga<sup>1</sup>

**To cite:** Vriesman MH, Vrolijk- Bosschaart TF, Lindauer RJL, *et al*. Prevalence of suspected child abuse in children with constipation: a case-control study. *BMJ Paediatrics Open* 2022;**6**:e001338. doi:10.1136/bmjpo-2021-001338

► Additional supplemental material is published online only. To view, please visit the journal online (<http://dx.doi.org/10.1136/bmjpo-2021-001338>).

Received 2 November 2021  
Accepted 28 December 2021



© Author(s) (or their employer(s)) 2022. Re-use permitted under CC BY-NC. No commercial re-use. See rights and permissions. Published by BMJ.

For numbered affiliations see end of article.

## Correspondence to

Dr Mana H Vriesman; m.h.vriesman@amsterdamumc.nl

## ABSTRACT

**Background** A possible association between child abuse and neglect (CAN) and functional constipation (FC) has been described in adults, however, limited data are available in children. Our objective was to determine the prevalence of suspected CAN in children with FC as compared with their healthy peers.

**Methods** A case-control study was carried out in children aged 3–10 years. Children with FC were recruited at a tertiary outpatient clinic, and healthy controls were recruited at schools. Parents were asked to fill out questionnaires about the history and behaviour of their child, children were inquired using a semistructured interview about experienced traumatic events and sexual knowledge. The interview was scored by two independent observers. The prevalence of suspected CAN was determined according to the questionnaires and interview.

**Results** In total, 228 children with FC and 153 healthy controls were included. Both groups were age and gender comparable (50% females, median age 6 years (not significant)). No significant difference in the prevalence of suspected CAN was found between children with FC and healthy controls (23.3% vs 30.1%, 95% CI 0.44 to 1.12,  $p=0.14$ ), including a suspicion of sexual, emotional and physical abuse.

**Conclusion** Suspected CAN was detected in both children with FC as in healthy controls. The possible association between CAN and FC in children could not be confirmed.

## INTRODUCTION

Functional constipation (FC) is a common and distressing health condition, affecting around 10% of children worldwide.<sup>1</sup> Multiple studies have shown that stressful life events, such as bullying, separation of parents or illness of a family member are associated with functional defecation problems such as FC and faecal incontinence in children.<sup>2,3</sup>

In adults suffering from functional defecation disorders, such as slow-transit constipation and pelvic floor dyssynergia, a possible association with child abuse has been described.<sup>4–10</sup> In addition, abused

## What is known about the subject?

- A possible association between child abuse and neglect and functional constipation has been described in adults, however, limited data are available in children.
- Diagnosing child abuse in young children is extremely challenging, it is often not recognised.

## What this study adds?

- We found no significant difference regarding the prevalence of suspected child abuse and neglect in young children with functional constipation as compared with healthy controls.

adult patients reported more severe symptoms of constipation,<sup>7</sup> decreased quality of life<sup>7</sup> and more frequent use of surgical strategies<sup>8</sup> as compared with patients with no history of child abuse. Early detection of abuse is therefore important in the workup of both paediatric and adult patients with FC. This is supported by the current paediatric and adult guidelines, advising to ask for a possible history of abuse in all patients with FC.<sup>11,12</sup>

Child abuse and neglect (CAN) as defined by WHO includes ‘all types of physical and/or emotional ill treatment, sexual abuse, neglect, negligence and commercial or other exploitation, which results in actual or potential harm to the child’s health, survival, development or dignity in the context of a relationship of responsibility, trust or power’.<sup>13</sup> Different subtypes of CAN have been recognised and include sexual abuse, physical and emotional abuse, and physical and emotional neglect.

The diagnosis of CAN remains challenging, since there is no gold standard and feelings of shame, guilt and blame

play an important role in the disclosure of abuse.<sup>14</sup> In children, the diagnosis of abuse is even more difficult because of verbal limitations<sup>15</sup> and the often absence or non-specificity of physical and psychological symptoms related to CAN.<sup>16 17</sup> Several screening tools have therefore been proposed for the paediatric assessment of CAN, using specific signs such as abnormal (sexual) behaviour,<sup>18</sup> age-inappropriate (sexual) knowledge<sup>15</sup> and abnormalities found on physical examination to detect patients with suspected CAN.

Several pathophysiological mechanisms in adults have been proposed to underlie the association between CAN and FC. Exposure to abuse may result in emotional distress, which in turn, through the brain-gut axis, may lead to visceral hypersensitivity and somatisation generating gastrointestinal symptoms.<sup>10 19</sup> In addition, trauma to the anorectal area or abdomen related to abuse may result in withholding behaviour and pelvic floor dysfunction.<sup>10</sup> However, the current evidence in adults is also criticised due to methodological shortcomings such as small study populations, the lack of control groups and recall bias.<sup>20</sup>

Only a few small cohort studies described a possible association between CAN and gastrointestinal symptoms in children, such as FC,<sup>21</sup> faecal incontinence<sup>22 23</sup> and abdominal pain.<sup>24–26</sup> Unfortunately, to date, there is a lack of well-designed studies investigating the prevalence of CAN in children presenting with functional defecation disorders.

Therefore, our objective was to determine the prevalence of suspected CAN in a large cohort of children with FC as compared with their healthy peers using specific signs of CAN.

## METHODS

A case–control study was carried out between April 2014 and March 2019 including children with FC (index group) and healthy children without gastrointestinal complaints (control group).

### Study population

Children aged 3–10 years presenting at our specialised outpatient clinic with FC according to the Rome III criteria<sup>27</sup> were included in the index group. Children with signs of organic or surgical causes of their symptoms were excluded.

For the healthy control group, children aged 3–10 years without gastrointestinal symptoms or any other functional or chronic disease were eligible for inclusion and recruited at randomly selected elementary schools throughout the Netherlands. Parents of children were asked about the medical history, current gastrointestinal symptoms and medication use of their child, and children in the control group were excluded if they had any sign of an organic or functional (somatic) disorder.

In both the index and control group, children were excluded if they or their parents had too little knowledge

of the Dutch language or if they suffered from a known intellectual disability.

### Recruitment and study procedure

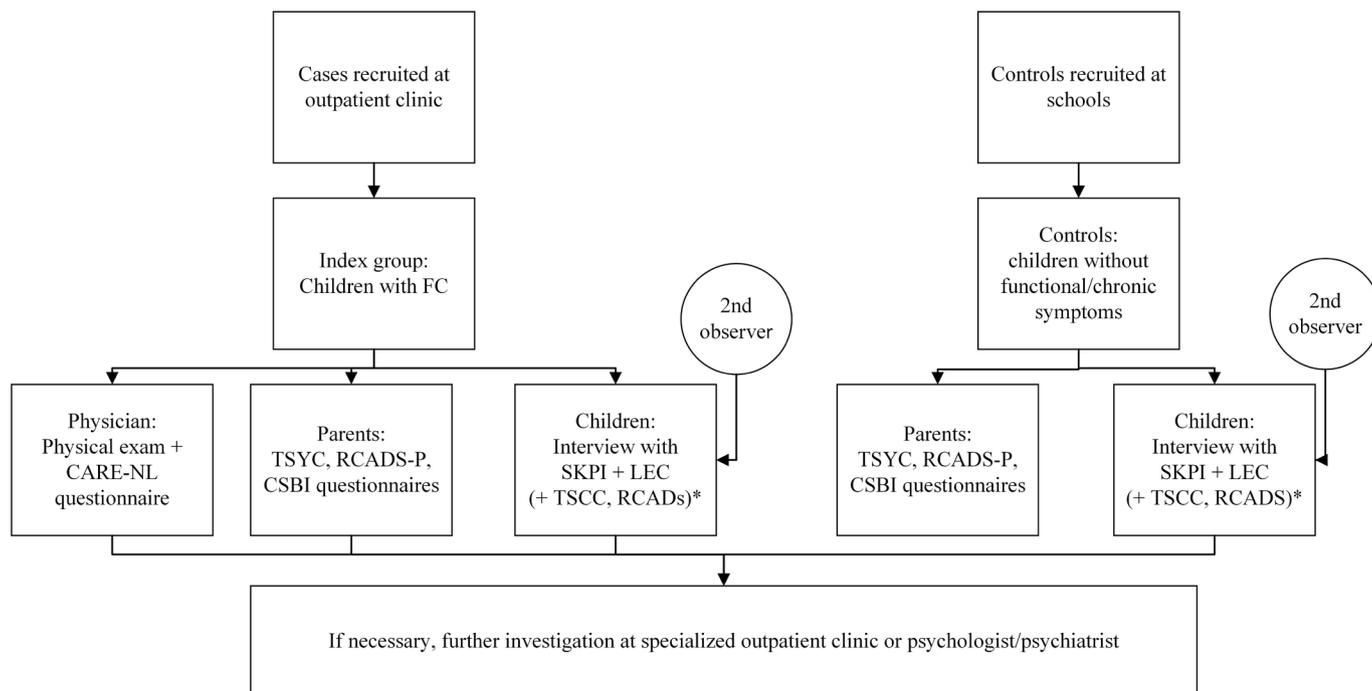
An overview of the study recruitment and procedure is depicted in figure 1. For the index group, all parents of newly referred children as patients with FC received an information letter about the study. After their first appointment at the outpatient clinic, parents were verbally informed by the local investigator. After their consent, the physician filled out a questionnaire estimating the risk for CAN using the Child Abuse Risk Evaluation—Dutch version (CARE-NL, and recorded the findings during the physical examination of the child. Next, after the clinic visit, a semi-structured interview (described below) with the child took place at the outpatient clinic by a trained member of the research team. During this interview, the parents were not present in the room. The interview was recorded on video and scored by the interviewer. At a later time point, the videorecorded interview was scored a second time by an independent member of the research team, who was not involved in the preparation of the manuscript. Parents were asked to fill out questionnaires about their child in a separate room.

Parents of healthy controls received an information letter about the study in the school newsletter or at school meetings. When interested in participating, parents were asked to send an email to the local investigator. The child was interviewed at school in the same way as the patients were interviewed in the index groups. However, no physical examination was performed as this was considered as too much of a burden in an otherwise healthy child. Parents were asked to fill out the same questionnaires at home.

### Questionnaires and interview

The parental questionnaires and interview with children was used to assess the prevalence of suspected CAN. Parents filled out a demographic questionnaire on a possible history of trauma of their child, and the Child Sexual Behaviour Inventory (CSBI-3),<sup>28</sup> measuring children's inappropriate sexual behaviour as a marker for sexual abuse.<sup>18</sup> Children were interviewed using the Life Events Checklist (LEC),<sup>29</sup> on traumatic events, and the Sexual Knowledge Picture Instrument (SKPI),<sup>30</sup> a child-friendly picture book to assess young children's sexual knowledge. Abnormal reaction to the SKPI included an abnormal non-verbal reaction to the images or age-inappropriate sexual knowledge, which can be indicators of sexual abuse.<sup>15</sup>

The Revised Child Anxiety and Depression Scale-Parent Version (RCADS/RCADS-P)<sup>31</sup> and Trauma Symptom Checklist for Young Children (TSCYC/Trauma Symptom Checklist for Children (TSCC))<sup>32</sup> were used to determine psychological symptoms in children with suspected CAN. A demographic questionnaire was filled out by parents to obtain information on social factors and



**Figure 1** Overview of study design. \*TSCC and RCADS if  $\geq 8$  years old. CARE-NL, Child Abuse Risk Evaluation—Dutch version; CSBI, Child Sexual Abuse Inventory; FAP, functional abdominal pain; FC, functional constipation; LEC, Life Events Checklist; RCADS(-P), Revised Child Anxiety and Depression Scale-Parent Version; SKPI, Sexual Knowledge Picture Instrument; TSCC, Trauma Symptom Checklist for Children; TSYC, Trauma Symptom Checklist for Young Children.

parental characteristics of the child. Detailed description of the questionnaires is available in online supplemental appendix 1.

### Prevalence of suspected can

The prevalence of suspected sexual abuse, physical abuse, neglect and total prevalence of suspected CAN (all subtypes combined) was calculated. Suspected sexual abuse was determined according to one of the following abnormal results; a parent reported child's history of sexual abuse, a clinical CSBI, an abnormal reaction to the SKPI as scored by two independent observers and/or a child report of sexual abuse on the LEC questionnaire. The prevalence of suspected physical abuse and emotional abuse or neglect was determined according to the following abnormal results; a parent reported child's history of abuse and/or child report of physical abuse, emotional abuse or neglect on the LEC questionnaire.

Abnormal findings were discussed with a blinded member of our specialised CAN team (SB-K). In case of a strong presumption of CAN and concerns for safety, findings were discussed with the parents and children were referred to our specialised multidisciplinary outpatient clinic (including a specialised paediatrician, child psychologist and social worker) according to the protocol of our hospital.

### Study outcomes

Primary outcome was the prevalence of suspected CAN in children with FC as compared with healthy controls. In addition, we aimed to determine clinical characteristics

of children with suspected CAN and to determine psychological symptoms in children with suspected CAN.

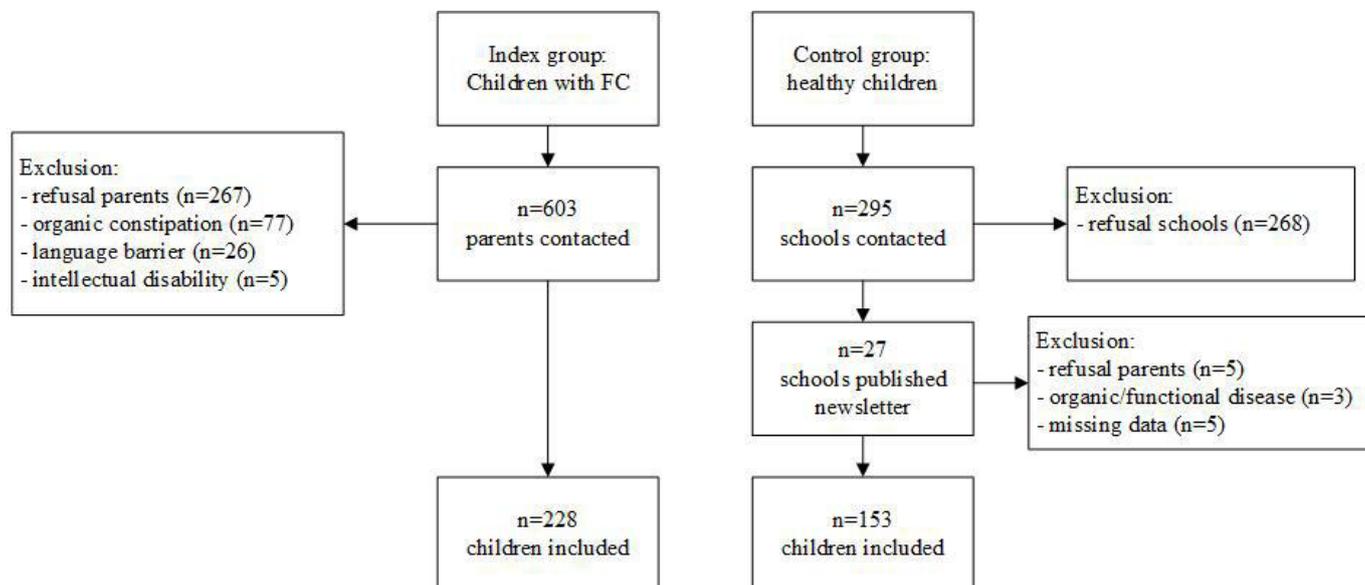
### Statistical analysis

Due to the explorative character of this study and lack of well-designed comparable studies in children, it was not possible to predict the frequency of suspected CAN. Therefore, we were unable to make exact power calculations. The exact prevalence of CAN in young children is unknown. The sample size was therefore calculated based on interim analysis (calculating a prevalence of CAN of 15.7% in children with FC) using nQuery Advisor (Statistical Solutions, Cork, Ireland) as we hypothesised that the prevalence of symptoms of CAN in the index group was twice as high as compared with healthy children. For 80% power to detect an OR of 2.17, 300 children were needed in each group.

Data were stored anonymously in SPSS V.25 (IBM). Statistical significance was accepted at  $p < 0.05$ . The study groups were compared by using one-way analysis of variance and independent t-test for continuous variables. Analysis of dichotomous variables were performed using  $\chi^2$  tests or Fisher's exact tests. Previously reported child and parental risk factors for CAN<sup>9</sup> (including age and gender of children, parental education level and mental health problems, parental history of abuse and parental substance abuse) were included.

### Patient and public involvement

Patients were involved in the design and conduct of this research. Choice of outcome measures and methods of



**Figure 2** Inclusion of index and control group. FC, functional constipation.

recruitment were informed by discussions with parents of patients based on previous studies of our research group.<sup>15–17</sup> Once the trial has been published, participants will be informed of the results through a newsletter.

## RESULTS

### Study population

In total, 228 children were diagnosed with FC and included in the index group (figure 2).

Reasons to decline participation were mostly practical (eg, no time) or concerns about the implications for the child. For the healthy control group, we randomly selected 295 elementary schools throughout different regions of the Netherlands, out of which 27 schools agreed to publish the study in their newsletter. The majority of schools reported to be already enrolled in other research projects or had hesitations concerning the research topic. In total, 153 parents of healthy children responded and children were included in the control group.

Of the included children, 50.1% were girls and the median age was 6 years (table 1). According to the CARE-NL questionnaire, significant differences with respect to education level and social history between parents of children with FC and parents of healthy controls were found (table 1).

### Prevalence of suspected CAN

We calculated the prevalence of suspected different subtypes of CAN (table 2). When combining all subtypes of CAN, no significant difference was found in the prevalence of suspected CAN between children with FC and healthy controls (23.3% vs 30.1%, 95% CI 0.44 to 1.12). Out of the 99 children with a suspicion of CAN, 17 children with FC and 12 healthy controls were referred to our specialised multidisciplinary outpatient clinic for further follow-up.

Next, our secondary objective was to determine clinical characteristics and psychological symptoms of children with suspected CAN. Clinical characteristics of children with suspected CAN are described in table 3. For this analysis, all children with suspected CAN were grouped together (n=99) and were compared with children without a suspicion of CAN (n=282). In children with FC, no significant difference with respect to the presence of faecal incontinence symptoms was found between children with and without suspected CAN (71.6% vs 74.3%, OR 0.9 95% CI 0.4 to 1.8, p=0.67).

According to the RCADS/RCADS-P and TSCYC/TSCC questionnaires, no significant differences were found in the prevalence of anxiety and depression (3 (3.0%) vs 3 (1.1%), p=0.18) and PTSD symptoms (7 (7.2%) vs 12 (4.7%), p=0.35) between children with (n=99) and without a suspicion of CAN (n=282).

## DISCUSSION

In this large case-control study including 381 children, we found a high number (26%) of children with a suspicion of CAN. No significant difference was found in the prevalence of suspected CAN in children with FC as compared with their healthy peers.

Unfortunately, paediatric data on this topic are scarce and the limited published studies mostly report on symptoms of faecal incontinence in children with a confirmed diagnosis of CAN.<sup>22 23</sup> With this current study, we tried to determine the prevalence of suspected CAN in children with a confirmed diagnosis of FC. To our knowledge, only two other community based studies conducted in Sri Lanka and Hong Kong assessed this association.<sup>21 33</sup> Rajindrajith *et al* found a significantly higher prevalence of sexual (5.8% vs 2.6%), emotional (40.9% vs 20.8%) and physical abuse (41.6% vs 23.2%) according to self-reports of adolescents

**Table 1** Demographic characteristics

	FC (n=228)	Controls (n=153)	P value
<b>Child characteristics</b>			
Age in years, median (IQR)*	6 (5–8.8)	6 (5–8)	0.34
Girls, n (%)†	112 (49.1)	79 (51.6)	0.63
Faecal incontinence n (%)	177 (77.6)	–	–
Duration symptoms in years, median (IQR)	4 (2–6)	–	–
Behavioural problems, n (%)	17 (7.6)	–	–
Abnormalities during physical examination suspect for CAN, n (%)‡	64 (28.1)	–	–
Anxiety/depression symptoms, n (%)§¶	6 (2.6)	0 (0.0)	0.09
PTSD symptoms, n (%)**†	15 (7.0)	4 (2.9)	0.09
<b>Parental characteristics</b>			
Parent(s) Dutch origin, n (%)†	177 (83.1)	126 (86.3)	0.46
Parent(s) low education††, n (%)†	149 (66.2)	48 (32.7)	(is less than sign) 0.001
Parent(s) history abuse, n (%)†	28 (13.7)	9 (6.4)	0.03
Parent(s) history domestic violence, n (%)†	21 (10.3)	6 (4.2)	0.04
Parent(s) daily alcohol use, n (%)†	23 (18.0)	18 (15.7)	0.63
Parent(s) drug abuse, n (%)†	11 (5.2)	4 (2.8)	0.27
Parent(s) mental health problems, n (%)†	75 (35.4)	28 (19.6)	0.001

\*Mann-Whitney U test.

†Pearson  $\chi^2$  test

‡According to physician, includes abnormal behaviour during examination (n=34), refusal of examination (n=17), haematomas (n=10) anal fissures (n=8), abnormal interaction between parent and child (n=5).

§According to the RCADS/RCADS-P questionnaires.

¶Fisher's exact test.

\*\*According to the TSCYC/TSCC questionnaires.

††<college or university degree.

–, not applicable; ADHD, autism and anxiety disorder; CAN, child abuse and neglect; FC, functional constipation; PTSD, post-traumatic stress disorder; RCADS-P, Revised Child Anxiety and Depression Scale-Parent Version; TSCC, Trauma Symptom Checklist for Children; TSCYC, Trauma Symptom Checklist for Young Children.

with FC as compared with healthy controls.<sup>21</sup> In contrast, Tam *et al* could not confirm this association.<sup>33</sup> Since the prevalence of CAN is highly dependent on community and societal factors, and differs between low-income and high-income countries, it is hard to compare our findings with these different widespread geographical locations.<sup>13</sup> Moreover, comparison between study results is challenging owing to the difference in instruments used to diagnose the prevalence of CAN.

We found a high prevalence of suspected CAN in our cohort; 23% and 30% of children with FC and healthy controls respectively. These numbers are conflicting with the previous Dutch NPM-2017 study, reporting a prevalence of confirmed cases of CAN of less than 3% in children.<sup>34</sup> However, it is important to keep in mind that we could only identify children with a suspicion of CAN, and a final diagnosis by a multidisciplinary team was not made in the time span of our study. This in contrast to the NPM study, which reported on the number of confirmed cases of CAN using data from 'Safe at Home' organisations and professionals. This major difference in methods could justify the conflicting numbers. Nonetheless, our results are in line

with the previously self-reported prevalence of CAN in Dutch school children of 26.7%.<sup>35 36</sup>

Research on the exact prevalence of CAN is challenging. Consent of parents is always required, while parents could be the perpetrators of the abuse. Furthermore, the diagnosis of CAN in young children is extremely difficult to confirm, while these young children are most at risk for CAN.<sup>13</sup> Not only important emotional barriers in disclosing CAN are involved, especially in a research setting where subjects have not met the interviewer before. In contrast to adults, (young) children often lack the proper words and knowledge to report abuse.<sup>15</sup> Consequently, CAN in children is often not recognised by healthcare professionals, and up to a 10-fold gap between studies based on self-report and child protection agencies is reported.<sup>37</sup> Therefore, for this current study, we chose to use both an interview with children and parental questionnaires to diagnose a suspicion of CAN. However, a reference standard for the diagnosis of CAN in children is lacking and instruments differ widely between studies and centres. We, therefore, decided to use several instruments, suitable for a young population, in order to not miss any children with a risk for CAN. Although

**Table 2** Prevalence of suspected CAN

N (%)	FC (n=228)	Controls (n=153)	Unadjusted OR (95% CI)	P value
Total suspected CAN	53 (23.2%)	46 (30.1%)	0.7 (0.4 to 1.1)	0.14
Suspected sexual abuse*	27 (11.8%)	25 (16.3%)	0.7 (0.4 to 1.2)	0.21
History sexual abuse	2 (0.9%)	4 (2.7%)		
Clinical CSBI	15 (6.7%)	18 (12.0%)		
Abnormal LEC	0 (0.0%)	3 (2.1%)		
Abnormal SKPI	13 (5.8%)	2 (1.4%)		
Suspected emotional abuse and neglect†	17 (7.5%)	11 (7.2%)	1.1 (0.5 to 2.3)	0.90
History neglect	1 (0.5%)	1 (0.7%)		
Abnormal LEC	16 (7.3%)	11 (7.8%)		
Suspected physical abuse‡	20 (8.8%)	18 (11.8%)	0.7 (0.4 to 1.4)	0.34
History physical abuse	2 (0.9%)	1 (0.7%)		
Abnormal LEC	18 (8.3%)	17 (12.0%)		

\*According to a parent reported child's history of sexual abuse, a clinical CSBI, abnormal SKPI and/or a child report of sexual abuse on the LEC questionnaire.

†According to a parent reported child's history of neglect and/or child report of emotional abuse and neglect on the LEC questionnaire.

‡According to a parent reported child's history of physical abuse and/or child report of physical on the LEC questionnaire.

CAN, child abuse and neglect; CSBI, Child Sexual Behaviour Inventory; FC, functional constipation; LEC, Life Events Checklist; SKPI, Sexual Knowledge Picture Instrument.

these instruments are already in clinical use by our specialised CAN team, the validation of the SKPI interview is still in progress.<sup>38</sup> Moreover, a recent systematic review showed low specificity of the CSBI for diagnosing sexual abuse.<sup>39</sup> It is therefore possible that some children were misclassified as suspected victims of CAN.

Another challenging aspect of diagnosing CAN in children with FC is that symptoms might overlap. CAN may cause behavioural and psychological problems, but these disorders are also common in children with FC.<sup>40</sup> Physical examination of children with FC, including inspection of the anorectal area and digital rectal examination, may lead to abnormal behaviour that is also seen in children after CAN. Because the interview was conducted right after the clinic visit, this could also have contributed to the high prevalence of abnormal reactions to the SKPI pictures in this group. Other important markers of sexual abuse during physical examination, such as anal fissures, are also common problems in children with constipation.<sup>41 42</sup> Therefore, our findings on physical examination and psychological symptoms in children with FC should be interpreted with care. The use of the SKPI in children with FC should be further investigated.

In addition, the inclusion of a representative healthy control group was not easy. For ethical reasons, recruitment of the index and control group occurred in a different way. While all new patients with FC visiting the outpatient clinic were personally informed about the study, we were only allowed to recruit healthy controls via a newsletter and parents had to actively contact us in order to participate.

Our results show that the topic of the study played an important role in the non-participation of many of the contacted schools and parents of both the index and control group. The recruitment is therefore at risk for selection bias, which has important consequences for our study results. Due to the low number of participating parents of healthy controls, it could be that these parents had specific reasons to respond to the research. These parents might have had existing concerns about possible psychological problems or abuse of their child, thereby contributing to the high prevalence of suspected CAN in this group. Moreover, important differences in demographic characteristics between the two study groups in terms of socioeconomic status were found. These differences could have affected our results, given that social factors including socioeconomic status are correlated with risk of CAN.<sup>13</sup> Owing to these differences in baseline characteristics and potential selection bias of parents, we cannot draw firm conclusions on our results on parental factors (ie, domestic violence, drug and alcohol abuse) in children with suspected CAN. However, given the current ethical obligations, these challenges are unavoidable and previously described in similar research projects.<sup>43</sup>

General limitations of the study should be considered when interpreting our results. Unfortunately, we did not meet our power calculations due to slow inclusion rates. It could, therefore, be hypothesised that the study groups were too small to draw firm conclusions. However, the exact prevalence of CAN is unknown, the power calculations were based on an interim analysis, and the important differences in baseline characteristics between study groups, made us

**Table 3** Clinical characteristics of children with suspected CAN

	Children with suspicion of CAN (n=99)	Children without suspicion of CAN (n=282)	Unadjusted OR (95% CI)	P value
<b>Child characteristics</b>				
Age in years, median (IQR)	7 (6–9)	5 (5–8)	1.1 (1.0 to 1.3)	0.02
Girls, n (%)	50 (50.0)	141 (50.0)	1.0 (0.7 to 1.6)	0.93
Faecal incontinence, n (%)†	38/53 (71.6)	130/175 (74.3)	0.9 (0.4 to 1.8)	0.67
Behavioural problems, n (%)	7 (7.1)	10 (3.5)	2.5 (0.9 to 6.8)	0.16
<b>Parental characteristics</b>				
Parent(s) Dutch origin, n (%)	14 (15.4)	42 (15.7)	0.9 (0.5 to 1.9)	0.95
Parent(s) low education*, n (%)	52 (54.2)	145 (52.5)	1.1 (0.7 to 1.7)	0.78
Parent(s) history abuse, n (%)	13 (14.9)	24 (9.3)	1.7 (0.8 to 3.5)	0.14
Parent(s) history domestic violence, n (%)	14 (15.6)	13 (5.1)	3.4 (1.6 to 7.7)	0.002
Parent(s) daily alcohol consumption, n (%)	19 (30.6)	22 (12.2)	3.2 (1.6 to 6.4)	0.001
Parent(s) drug abuse, n (%)	8 (8.8)	7 (2.7)	3.6 (1.3 to 10.2)	0.03
Parent(s) mental health problems, n (%)	32 (34.0)	71 (27.2)	1.4 (0.8 to 2.3)	0.21

\* <college or university degree.

† only in children with FC

-, not applicable; CAN, child abuse and neglect.

decide to preliminary terminate the study. Another limitation is that the interview and questionnaires of cases and controls were not blinded, possibly leading to bias in the interpretation of results. However, to minimise this bias, an independent second observer was appointed to independently score the interviews and only abnormal results of both the first and second observer were used to calculate the prevalence of suspected CAN.

In contrast to the adult literature, we could not confirm the association between CAN and FC. Pathophysiologic differences between adults and children with FC could also play a role. As the association between CAN and FC is mostly described in adults with slow-transit constipation and pelvic floor dyssynergia,<sup>4–10</sup> these types of constipation are less commonly acknowledged in young children. It could also be hypothesised that constipation, as a symptom of CAN, will develop over time and the association in young children is, therefore, not present yet. Well-designed longitudinal paediatric studies, with longer follow-up, are therefore needed to confirm this hypothesis.

In conclusion, in this study, we found a high prevalence of suspected CAN, but could not confirm an association between suspected CAN and a diagnosis of FC. Our study demonstrates the many challenges, both ethical and methodological, related to research on the association between CAN and FC. Future research is needed to further unravel

the possible gastrointestinal consequences after CAN in order to establish an early detection and prevention of reabuse in children. These observational studies should use a comparable recruitment procedure to include a representative patient and control group, and blinded multidisciplinary assessment to determine the prevalence of CAN.

#### Author affiliations

<sup>1</sup>Department of Pediatric Gastroenterology and Nutrition, Emma Childrens' Hospital UMC, Amsterdam, The Netherlands

<sup>2</sup>Department of Social Pediatrics, Emma Childrens' Hospital UMC, Amsterdam, The Netherlands

<sup>3</sup>Department of Child and Adolescent Psychiatry, Amsterdam UMC Locatie AMC, Amsterdam, The Netherlands

<sup>4</sup>De Bascule, Academic Center for Child and Adolescent Psychiatry, Amsterdam UMC, University of Amsterdam, Amsterdam, Netherlands

<sup>5</sup>Pediatric Clinical Research Office, Emma Childrens' Hospital UMC, Amsterdam, The Netherlands

<sup>6</sup>Knowledge Institute of the Dutch Association of Medical Specialists, Utrecht, Netherlands

**Contributors** MHV: conceptualised the study, collected data, drafted the initial manuscript and performed initial data analyses. TFV-B: conceptualised the study, collected data and reviewed and revised the manuscript. SB-K: participated in the design of the study, coordinated and supervised data collection, and critically reviewed and revised the manuscript. JHvdL: coordinated and supervised data analyses, and critically reviewed and revised the manuscript. AHT, RJLL and MAB: participated in the design of the study, supervised drafting of the manuscript and critically reviewed the manuscript for important intellectual content. MAB:

responsible for the overall content as the guarantor. All authors approved the final manuscript as submitted and agree to be accountable for all aspects of the work.

**Funding** The authors have not declared a specific grant for this research from any funding agency in the public, commercial or not-for-profit sectors.

**Competing interests** None declared.

**Patient consent for publication** Consent obtained from parent(s)/guardian(s)

**Ethics approval** The study was approved by the local Medical Ethics Committee (file no. METC 2011\_059, NL35893.018.11). All parents were asked for written informed consent.

**Provenance and peer review** Not commissioned; externally peer reviewed.

**Data availability statement** All data relevant to the study are included in the article or uploaded as online supplemental information. Not applicable.

**Supplemental material** This content has been supplied by the author(s). It has not been vetted by BMJ Publishing Group Limited (BMJ) and may not have been peer-reviewed. Any opinions or recommendations discussed are solely those of the author(s) and are not endorsed by BMJ. BMJ disclaims all liability and responsibility arising from any reliance placed on the content. Where the content includes any translated material, BMJ does not warrant the accuracy and reliability of the translations (including but not limited to local regulations, clinical guidelines, terminology, drug names and drug dosages), and is not responsible for any error and/or omissions arising from translation and adaptation or otherwise.

**Open access** This is an open access article distributed in accordance with the Creative Commons Attribution Non Commercial (CC BY-NC 4.0) license, which permits others to distribute, remix, adapt, build upon this work non-commercially, and license their derivative works on different terms, provided the original work is properly cited, appropriate credit is given, any changes made indicated, and the use is non-commercial. See: <http://creativecommons.org/licenses/by-nc/4.0/>.

#### ORCID iD

Mana H Vriesman <http://orcid.org/0000-0002-4759-8447>

#### REFERENCES

- Koppen IJN, Vriesman MH, Saps M, *et al.* Prevalence of functional defecation disorders in children: a systematic review and meta-analysis. *J Pediatr* 2018;198:121–30.
- Philips EM, Peeters B, Teeuw AH, *et al.* Stressful life events in children with functional defecation disorders. *J Pediatr Gastroenterol Nutr* 2015;61:384–92.
- Joinson C, Grzeda MT, von Gontard A, *et al.* Psychosocial risks for constipation and soiling in primary school children. *Eur Child Adolesc Psychiatry* 2019;28:203–10.
- Leroi AM, Bernier C, Watier A, *et al.* Prevalence of sexual abuse among patients with functional disorders of the lower gastrointestinal tract. *Int J Colorectal Dis* 1995;10:200–6.
- Drossman DA, Leserman J, Nachman G, *et al.* Sexual and physical abuse in women with functional or organic gastrointestinal disorders. *Ann Intern Med* 1990;113:828–33.
- Solé LI, Bolino MC, Lueso M, *et al.* Prevalence of sexual and physical abuse in patients with obstructed defecation: impact on biofeedback treatment. *Rev Esp Enferm Dig* 2009;101:464–7.
- Imhoff LR, Liwanag L, Varma M. Exacerbation of symptom severity of pelvic floor disorders in women who report a history of sexual abuse. *Arch Surg* 2012;147:1123–9.
- O'Brien S, Hyman N, Osler T, *et al.* Sexual abuse: a strong predictor of outcomes after colectomy for slow-transit constipation. *Dis Colon Rectum* 2009;52:1844–7.
- Beck JJH, Elzevier HW, Pelger RCM, *et al.* Multiple pelvic floor complaints are correlated with sexual abuse history. *J Sex Med* 2009;6:193–8.
- Drossman DA, Talley NJ, Leserman J, *et al.* Sexual and physical abuse and gastrointestinal illness. review and recommendations. *Ann Intern Med* 1995;123:782–94.
- Tabbers MM, DiLorenzo C, Berger MY, *et al.* Evaluation and treatment of functional constipation in infants and children: evidence-based recommendations from ESPGHAN and NASPGHAN. *J Pediatr Gastroenterol Nutr* 2014;58:258–74.
- Bharucha AE, Pemberton JH, Locke GR. American gastroenterological association technical review on constipation. *Gastroenterology* 2013;144:218–38.
- World Health Organisation. Child maltreatment fact sheet no 150. Available: <https://www.who.int/news-room/fact-sheets/detail/child-maltreatment> [Accessed 20 Sep 2019].
- Lemaigre C, Taylor EP, Gittoes C. Barriers and facilitators to disclosing sexual abuse in childhood and adolescence: a systematic review. *Child Abuse Negl* 2017;70:39–52.
- Brilleslijper-Kater SN, Friedrich WN, Corwin DL. Sexual knowledge and emotional reaction as indicators of sexual abuse in young children: theory and research challenges. *Child Abuse Negl* 2004;28:1007–17.
- Vrolijk-Bosschaart TF, Brilleslijper-Kater SN, Widdershoven GA, *et al.* Physical symptoms in very young children assessed for sexual abuse: a mixed method analysis from the ASAC study. *Eur J Pediatr* 2017;176:1365–74.
- Vrolijk-Bosschaart TF, Brilleslijper-Kater SN, Widdershoven GAM, *et al.* Psychosocial symptoms in very young children assessed for sexual abuse: a qualitative analysis from the ASAC study. *Child Abuse Negl* 2017;73:8–23.
- Drach KM, Wientzen J, Ricci LR. The diagnostic utility of sexual behavior problems in diagnosing sexual abuse in a forensic child abuse evaluation clinic. *Child Abuse Negl* 2001;25:489–503.
- Salmon P, Skaife K, Rhodes J. Abuse, dissociation, and somatization in irritable bowel syndrome: towards an explanatory model. *J Behav Med* 2003;26:1–18.
- Hobbis ICA, Turpin G, Read NW. A re-examination of the relationship between abuse experience and functional bowel disorders. *Scand J Gastroenterol* 2002;37:423–30.
- Rajindrajith S, Devanarayana NM, Lakmini C, *et al.* Association between child maltreatment and constipation: a school-based survey using Rome III criteria. *J Pediatr Gastroenterol Nutr* 2014;58:486–90.
- Rajindrajith S, Devanarayana NM, Benninga MA. Fecal incontinence in adolescents is associated with child abuse, somatization, and poor health-related quality of life. *J Pediatr Gastroenterol Nutr* 2016;62:698–703.
- Mellon MW, Whiteside SP, Friedrich WN. The relevance of fecal soiling as an indicator of child sexual abuse: a preliminary analysis. *J Dev Behav Pediatr* 2006;27:25–32.
- Sonneveld LP, Brilleslijper-Kater SN, Benninga MA, *et al.* Prevalence of child sexual abuse in pediatric patients with chronic abdominal pain. *J Pediatr Gastroenterol Nutr* 2013;56:475–80.
- van Tilburg MAL, Runyan DK, Zolotor AJ, *et al.* Unexplained gastrointestinal symptoms after abuse in a prospective study of children at risk for abuse and neglect. *Ann Fam Med* 2010;8:134–40.
- Devanarayana NM, Rajindrajith S, Perera MS, *et al.* Association between functional gastrointestinal diseases and exposure to abuse in teenagers. *J Trop Pediatr* 2014;60:386–92.
- Rasquin A, Di Lorenzo C, Forbes D, *et al.* Childhood functional gastrointestinal disorders: child/adolescent. *Gastroenterology* 2006;130:1527–37.
- Friedrich WN, Fisher JL, Dittner CA, *et al.* Child sexual behavior inventory: normative, psychiatric, and sexual abuse comparisons. *Child Maltreat* 2001;6:37–49.
- Gray MJ, Litz BT, Hsu JL, *et al.* Psychometric properties of the life events checklist. *Assessment* 2004;11:330–41.
- Brilleslijper-Kater SN. *Beyond Words: Between-Group Differences in the Ways Sexually Abused and Nonabused Preschool Children Reveal Sexual Knowledge*. Enschede: Febedruk, 2005.
- Chorpita BF, Moffitt CE, Gray J. Psychometric properties of the revised child anxiety and depression scale in a clinical sample. *Behav Res Ther* 2005;43:309–22.
- Odessa JB, Resources FPA. Trauma symptom checklist for young children (TSCYC), 2005. Available: <http://v-psyche.com/doc/ClinicalTest/Trauma> [Accessed 23 Apr 2019].
- Tam YH, Li AM, So HK, *et al.* Socioenvironmental factors associated with constipation in Hong Kong children and Rome III criteria. *J Pediatr Gastroenterol Nutr* 2012;55:56–61.
- van Berkel SR, Prevoo MJL, Linting M, *et al.* Prevalence of child maltreatment in the Netherlands: an update and cross-time comparison. *Child Abuse Negl* 2020;103:104439.
- Vink R, van der Pal S, Eekhout I. *Bij Leerlingen in Groep 7/8 van Het Regulier Basisonderwijs*, 2016. file:///H:/Downloads/TNO-2016-R11157.pdf
- Alink L, Prevoo M, van Berkel S. *NPM-2017: Nationale Prevalentiestudie Mishandeling van*. Leiden: Kinderen En Jeugdigen, 2018. <https://www.wodc.nl/onderzoeksdatabase/2668g-informantenstudie-kindermishandeling.aspx>
- Gilbert R, Widom CS, Browne K, *et al.* Burden and consequences of child maltreatment in high-income countries. *Lancet* 2009;373:68–81.
- van Ham K, Brilleslijper-Kater S, van der Lee H, *et al.* Validation of the sexual knowledge picture instrument as a diagnostic instrument for child sexual abuse: study protocol. *BMJ Paediatr Open* 2020;4:e000799.

- 39 Vrolijk-Bosschaart TF, Verlinden E, Langendam MW, *et al.* The diagnostic utility of the child sexual behavior inventory for sexual abuse: a systematic review. *J Child Sex Abus* 2018;27:729–51.
- 40 van Dijk M, Benninga MA, Grootenhuis MA, *et al.* Prevalence and associated clinical characteristics of behavior problems in constipated children. *Pediatrics* 2010;125:e309–17.
- 41 Rougé-Maillart C, Houdu S, Darviot E, *et al.* Anal lesions presenting in a cohort of child gastroenterological examinations. Implications for sexual traumatic injuries. *J Forensic Leg Med* 2015;32:25–9.
- 42 Anderson B, Thimmesch I, Aardsma N, *et al.* The prevalence of abnormal genital findings, vulvovaginitis, enuresis and encopresis in children who present with allegations of sexual abuse. *J Pediatr Urol* 2014;10:1216–21.
- 43 Brilleslijper-Kater SN, Baartman HEM. What do young children know about sex? research on the sexual knowledge of children between the ages of 2 and 6 years. *Child Abuse Rev.* 2000;9:166–82.

## APPENDIX 1. Interview and questionnaires

The following questionnaires were used for the diagnosis of suspected CAN:

### *Physician's questionnaire- Child Abuse Risk Evaluation – Dutch version (CARE-NL)*

The physician filled out the CARE-NL after the consultation of children with FC. The CARE-NL is a structured questionnaire for healthcare professionals to estimate the risk of CAN in children. This validated questionnaire consists of 18 risk factors divided on 4 subscales: 1) parent factors, 2) parent-child factors, 3) child factor and 4) family factors. All risk factors will be scored on a 3-point scale (yes, no or lack of information). The inter-rater reliability was described to be high (ICC 0.6-0.75) and predictive validity for court custody to be good (AUC = 0.77).<sup>1,2</sup>

### *Parental questionnaire – Child Sexual Behavior Inventory (CSBI-3)*

Parents filled out the validated Dutch version of the CSBI, a parent reported measure of sexual behavior in children aged 2-12 years.<sup>3,4</sup> The CSBI was developed based on the fact that age-inappropriate sexual behavior can be an indication for sexual abuse.<sup>5-7</sup> For this study, the CSBI-Total scale was used to identify children with a suspicion of sexual abuse. The CSBI total scale indicates the overall level of sexual behavior the child exhibits and provides clinical cutoff scores depending on the age and gender of the child. Psychometric properties such as internal consistency (0.47 to 0.86 in normative samples) and inter-rater reliability between primary caregivers ( $r = 0.79$ ,  $p < 0.01$ ) are described to be good.<sup>4,6,8</sup>

### *Child's interview – Sexual Knowledge Picture Instrument (SKPI)*

Children were interviewed using the SKPI. The SKPI is developed by one of the authors (SB) and consists of fourteen child friendly pictures to assess young children's sexual knowledge.<sup>9</sup>

Research to validate the SKPI is now in progress.<sup>10</sup> Pictures are used to assess knowledge of genital differences, gender identity and body parts and functions, by showing four pictures of clothed and naked adults and children. Other pictures are used to assess knowledge of adult sexual behavior and knowledge of differences between physical intimacy and sexual interactions.<sup>9</sup> Children were asked to describe the pictures and the verbal and non-verbal reactions were evaluated by a trained member of the study team. Abnormal reaction to the SKPI included an abnormal non-verbal reaction to the images or age-inappropriate sexual knowledge, that can be indicators of sexual abuse.<sup>11</sup> A previous study showed that non-abused children's reactions to the SKPI were mostly open-minded and relaxed when interviewed, whereas abused children seemed remarkably more stressed, revealed less basic sexuality knowledge and showed significantly more avoidant non-verbal/emotional reactions during the interview.<sup>9</sup> A second blinded independent observer was asked to assess the video-recorded interview. Discrepancies were discussed with a member of our specialized CAN team (SB).

#### *Child's interview– Life Events Checklist (LEC)*

The LEC is a validated self-report measure to assess potentially traumatic events in children. The LEC consists of 25 questions about (life-time) traumatic life events including the subtypes of CAN and participants rate their experience with that event on a 5-point nominal scale (1 = happened to me, 2= witnessed it, 3= learned about it, 4= not sure, and 5 = does not apply). The reliability of the LEC as a measure of direct trauma exposure was found to be good (kappa coefficients of physical and sexual assault were 0.59 and 0.76 respectively).<sup>12</sup>

Moreover, we determined possible psychological consequences of CAN using the:

#### *Revised Child Anxiety and Depression Scale (RCADS & RCADS-P)*

The RCADS-P is a validated parent-report questionnaire assessing signs of anxiety and depression, the RCADS is the self-report version of the RCADS-P for children  $\geq 8$  years of age.<sup>13,14</sup> For this study, the subscales major depressive disorder (MDD), total anxiety and total anxiety and depression were used to diagnose anxiety and depression corresponding to the Diagnostic and Statistical Manual of mental disorders (DSM-IV). The questionnaire consists of 47 items and parents or children rate how often each item applies to them on a 4-point Likert scale, ranging from 0 (never) to 3 (always). Raw scores were converted into gender-normed T-scores, where a T-score of  $\geq 70$  was defined as the clinical threshold. The RCADS(-P) shows good psychometric properties<sup>15,16</sup> and the Dutch version is validated.<sup>14</sup>

*Trauma Symptom Checklist for Young Children (TSCYC) & Trauma Symptom Checklist for Children (TSCC)*

The TSCYC is a validated caretaker-report instrument developed for the assessment of trauma-related symptoms in children aged 3-10 years old.<sup>17,18</sup> The TSCC is the self-report version of children  $\geq 8$  years of age.<sup>18</sup> The TSCYC consists of 90-items, rating trauma-related symptoms on a 4-point scale according to how often the symptom has occurred in the previous month (ranging from 1= not at all to 4= very often). For this study, we used the summary posttraumatic stress scale to determine symptoms of posttraumatic stress disorder (PTSD). A T-value  $\geq 70$  was considered clinical.<sup>19</sup> The TSCYC was found to have reasonable psychometric properties and correlated with trauma exposure.<sup>52</sup>

**REFERENCES**

1. de Ruiter C, Hildebrand M, van der Hoorn S. Gestructureerde risicotaxatie bij kindermishandeling: De Child Abuse Risk Evaluation - Nederlandse versie (CARE-NL). *GZ-Psychologie*. 2012;4(3):10-17.
2. de Ruiter C, de Jong EM. *CARE-NL Richtlijn Voor Gestructureerde Beoordeling van Het Risico van Kindermishandeling*. Utrecht: Corine de Ruiter; 2005. <http://www.corinederuiter.eu/>. Accessed April 23, 2019.
3. Friedrich WN. *Child Sexual Behavior Inventory: Professional Manual*. Odessa, FL: Psychological Assessment Resources. ; 1997. <http://www4.parinc.com/>. Accessed April 23, 2019.
4. E S, D D, W F. Child Sexual Behavior Inventory: A Dutch-speaking Normative Sample. *Pediatrics*. 1999;104(4 Pt 1). doi:10.1542/PEDS.104.4.885
5. Drach KM, Wientzen J, Ricci LR. The diagnostic utility of sexual behavior problems in diagnosing sexual abuse in a forensic child abuse evaluation clinic. *Child Abuse Negl*. 2001;25(4):489-503. <http://www.ncbi.nlm.nih.gov/pubmed/11370722>. Accessed September 3, 2019.
6. Friedrich WN, Fisher JL, Dittner CA, et al. Child Sexual Behavior Inventory: Normative, Psychiatric, and Sexual Abuse Comparisons. *Child Maltreat*. 2001;6(1):37-49. doi:10.1177/1077559501006001004
7. Baker AJL, Gries L, Schneiderman M, Parker R, Archer M, Friedrich B. Children with problematic sexualized behaviors in the child welfare system. *Child Welfare*. 2008;87(1):5-27. <http://www.ncbi.nlm.nih.gov/pubmed/18575256>. Accessed April 23, 2019.
8. Friedrich WN, Fisher J, Broughton D, Houston M, Shafran CR. Normative sexual behavior in children: a contemporary sample. *Pediatrics*. 1998;101(4):E9. <http://www.ncbi.nlm.nih.gov/pubmed/9521975>. Accessed April 23, 2019.
9. Brilleslijper-Kater SN. *Beyond Words : Between-Group Differences in the Ways Sexually Abused and Nonabused Preschool Children Reveal Sexual Knowledge*. Enschede: Febodruk; 2005.
10. van Ham K, Brilleslijper-Kater S, van der Lee H, et al. Validation of the Sexual Knowledge Picture Instrument as a diagnostic instrument for child sexual abuse: study protocol. *BMJ Paediatrics Open* 2020;4:e000799. doi:10.1136/bmjpo-2020-00799.
11. Brilleslijper-Kater SN, Friedrich WN, Corwin DL. Sexual knowledge and emotional reaction as indicators of sexual abuse in young children: theory and research challenges. *Child Abuse Negl*. 2004;28(10):1007-1017. doi:10.1016/j.chiabu.2004.06.005
12. Gray MJ, Litz BT, Hsu JL, Lombardo TW. Psychometric Properties of the Life Events Checklist. *Assessment*. 2004;11(4):330-341. doi:10.1177/1073191104269954
13. Chorpita BF, Yim L, Moffitt C, Umemoto LA, Francis SE. Assessment of symptoms of DSM-IV anxiety and depression in children: a revised child anxiety and depression scale. *Behav Res Ther*. 2000;38(8):835-855. doi:10.1016/s0005-7967(99)00130-8
14. Kösters MP, Chinapaw MJM, Zwaanswijk M, van der Wal MF, Koot HM. Structure, reliability, and validity of the revised child anxiety and depression scale (RCADS) in a multi-ethnic urban sample of Dutch children. *BMC Psychiatry*. 2015;15(1):132. doi:10.1186/s12888-015-0509-7
15. Chorpita BF, Moffitt CE, Gray J. Psychometric properties of the Revised Child Anxiety and Depression Scale in a clinical sample. *Behav Res Ther*. 2005;43(3):309-322. doi:10.1016/j.brat.2004.02.004
16. Ebesutani C, Chorpita BF, Higa-McMillan CK, Nakamura BJ, Regan J, Lynch RE. A Psychometric Analysis of the Revised Child Anxiety and Depression Scales—Parent

- Version in a School Sample. *J Abnorm Child Psychol.* 2011;39(2):173-185.  
doi:10.1007/s10802-010-9460-8
17. Lamers-Winkelmann F. *Experimentele Vertaling Trauma Symptom Checklist for Children.*; 1998.
  18. Briere J, Johnson K, Bissada A, et al. The Trauma Symptom Checklist for Young Children (TSCYC): reliability and association with abuse exposure in a multi-site study. *Child Abuse Negl.* 2001;25(8):1001-1014.  
<http://www.ncbi.nlm.nih.gov/pubmed/11601594>. Accessed September 3, 2019.
  19. Odessa JB-, Resources FPA, 2005 undefined. Trauma symptom checklist for young children (TSCYC). *v-psyche.com*. [http://v-psyche.com/doc/Clinical Test/Trauma Symptom Checklist for Young Children\(TSCYC\).doc](http://v-psyche.com/doc/Clinical Test/Trauma Symptom Checklist for Young Children(TSCYC).doc). Accessed April 23, 2019.