



## RESEARCH ARTICLE

# Emotion regulation in 7-year-old children with familial high risk for schizophrenia or bipolar disorder compared to controls – The Danish High Risk and Resilience Study – VIA 7, a population-based cohort study

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## Abstract

**Objectives:** Emotion regulation is a predictor of overall life outcome. Problems of emotion regulation are associated with multiple psychiatric disorders and could be a potential treatment target for improving well-being and functioning. Children at familial high risk of severe mental illness have a markedly increased risk of various psychopathology and constitute a group at significant risk of emotion regulation problems. Investigations of emotion regulation in children at familial high risk of severe mental illness are sparse.

**Methods:** We applied an instrument for assessing emotion regulation, the Tangram Emotion Coding Manual (TEC-M), to a population-based cohort of 522 7-year-old children born to parents diagnosed with either schizophrenia or bipolar disorder and matched controls. The TEC-M is an ecologically valid, clinician-rated observational test measure of spontaneous emotion regulation. We aimed to compare emotion regulation between risk groups and to investigate

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associations between emotion regulation and psychopathology and daily life functioning, and between emotion regulation and an acknowledged questionnaire-based dysregulation profile.

**Results:** In this early developmental phase, we found no between group differences in emotion regulation. We found a significant but weak negative association between emotion regulation and both child psychopathology and the presence of a dysregulation profile on the Child Behavior Checklist and a weak positive association between emotion regulation and current level of functioning.

**Conclusions:** These findings contribute to the understanding of emotion regulation in familial high-risk children and further studies of emotion regulation in children at familial high risk of severe mental illness are warranted.

**KEYWORDS**

bipolar disorder, emotion regulation, familial high risk, offspring, schizophrenia spectrum psychosis

**Practitioner points**

- There are no significant differences in emotion regulation between young children with a familial high-risk of schizophrenia or bipolar disorder and community-based controls.
- Emotion regulation in young children is weakly and negatively associated with dimensions of their psychopathology.
- Emotion regulation in young children is positively associated with their current level of functioning.

**INTRODUCTION**

Emotions guide the individual in identifying what is potentially harmful and what is believed to be advantageous in relation to the individual's immediate well-being as well as general future functioning (Campos et al., 1994; Gross et al., 2019; Kimhy et al., 2016; Lazarus, 1991).

Adaptive and appropriate emotion regulation is a prerequisite for adequately adjusting to varying social contexts, and emotional dysregulation can have vast negative consequences for the individual as well as for the society as a whole (Moffitt et al., 2011; Philippot, 2004; Thompson, 2011). Emotion regulation in childhood and adolescence is an important predictor of overall outcomes throughout life, such as academic, occupational, and family functioning (Graziano et al., 2007; Moffitt et al., 2011).

Behaviours associated with inadequate or maladaptive emotion regulation are seen in multiple psychiatric disorders. Specifically, problems with emotion regulation are core symptoms of bipolar disorder and schizophrenia spectrum disorder and are often seen in, e.g., attention-deficit/hyperactivity disorder (ADHD), disruptive behaviour disorder, and autism spectrum disorders (Aldao et al., 2016; Fernandez et al., 2016; Kring & Werner, 2004). One study (Carlson et al., 2016) observed lower functioning using

Children's Global Assessment Scale (CGAS) (Shaffer et al., 1983) and worse aggression, based on a subscale from the Child Behavior Checklist (CBCL) (Achenbach & Rescorla, 2001), in 6-year-old community children who frequently lost their tempers and had severe temper tantrums compared to peers who also frequently lost their tempers but did not have real tantrums.

Studies in offspring of mothers with bipolar disorder suggest that problems with emotion regulation are present from infancy (Johnson et al., 2014). The capacity of emotion regulation is less studied in offspring of parents with schizophrenia, but studies imply that problems with emotion regulation are present from early childhood (4 and 7 years of age; 4–17 years of age) (Díaz-Caneja et al., 2018; Donatelli et al., 2010). Moreover, several studies have reported that children born to parents with severe mental illnesses such as schizophrenia and bipolar disorder are at a markedly increased risk of a wide array of psychiatric disorders (Ellersgaard et al., 2018; Rasic et al., 2014; Thorup et al., 2017). Thus, children of parents with a severe mental illness comprise a unique group of individuals, who potentially have emotion regulation difficulties due to their augmented risk of a broad spectrum of psychiatric disorders.

Most studies investigating emotion regulation in children are based on parent-, teacher-, or self-rated questionnaires. These questionnaires often focus on the occurrence of dysregulated behaviour and do not yield information about the context of said behaviour or the social mechanisms related to emotion regulation, and are known to be subject to bias as well as affected by the mood of the informant. Also, the need for ecologically valid assessments of spontaneous emotion regulation (meaning non-prompted and non-restricted emotion regulation, as opposed to settings/studies, in which the child has been instructed to use a specific emotion regulation strategy, e.g., cognitive reappraisal) has been highlighted in a meta-analytic review (Aldao et al., 2010). To this end, we employed the Tangram Emotion Coding Manual (TEC-M; Hagstrøm et al., 2019). The TEC-M provides an assessment of children's emotion regulation in an ecologically valid setting, in which the child participates in a frustrating puzzle task designed to prompt emotional reactions, in a social context, where only the child's primary caregiver is present during the task. The TEC-M allows for a standardized, lab-based, and clinician-rated assessment including observations of related mechanisms.

We aimed to investigate differences in emotion regulation across familial risk status in a population-based cohort of 522 7-year-old children consisting of children born to parents diagnosed with either schizophrenia or bipolar disorder and control children with neither parent diagnosed with schizophrenia nor bipolar disorder (Thorup et al., 2015). We hypothesized that children with a familial predisposition of severe mental illness would display more inadequate emotion regulation than children without this familial risk. Second, we aimed to investigate potential associations, both within each risk status group and across risk groups, between emotion regulation of the child and dimensional measures of child psychopathology, psychopathological symptom severity, daily life functioning, and a categorical emotional/behavioural dysregulation profile from The Child Behavior Checklist (CBCL-DP) (Achenbach & Rescorla, 2001; Althoff, 2010). We hypothesized that an increased dimensional psychopathological load and a positive dysregulation profile would be negatively associated with emotion regulation.

## MATERIALS AND METHODS

### Procedures

The Danish High Risk and Resilience Study – VIA 7 (The VIA 7 Study), a nationwide population-based cohort study, was approved by the Danish Data Protection Agency (J.nr.:2012-58-0004). The Danish National Committee on Health Research Ethics concluded that approval was not necessary due to the observational nature of the study. We obtained informed written consent from all participants and custody holders, including a separate written consent to contact the child's teachers. Trained psychologists, medical doctors, and nurses performed all assessments under the supervision of a senior specialist in child neuropsychology (JRM) and a specialist in child and adolescent psychiatry (AT). Child assessors were blinded to the risk status of the child.

## Participants

We composed a cohort of 522 children (aged 6.9–8.4 years) using the Danish Psychiatric Central Research Register (Mors et al., 2011) and the Danish Civil Registration System (Pedersen, 2011). The groups comprised children with familial high risk for bipolar disorder (FHR-BP) or schizophrenia spectrum psychosis (defined as schizophrenia, delusional disorder, or schizoaffective disorder) (FHR-SZ). Children with one parent with bipolar disorder and the other with schizophrenia were assigned to the FHR-SZ group. Children in the control group were likewise drawn from the registers and matched on sex, age, and municipality to the FHR-SZ children. Parents in the control group could have any psychiatric disorder other than schizophrenia spectrum disorder or bipolar disorder, thus creating a control group of children representative of the normal population. FHR-BP children comprised a non-matched sample but were comparable on age and sex to the children in the two other groups. All children and parents had to be born and living in Denmark. The VIA 7 study design has been described in detail elsewhere (Thorup et al., 2015). The age of seven was the desired age for the population-based cohort (VIA7) and was not related to or a specific age requirement for participation in the TEC-M.

A total of 465 children and their primary caregivers participated in the frustration task designed to measure emotion regulation (Figure 1). The participating adult was always the primary caregiver, defined as the adult who, at the time of the study, knew the child the best and cared for the child on a daily basis. This could be either one of the biological parents (independent of potential diagnosis of the parent), a step-parent, or a foster parent. TEC-M data of 57 children were not included in the study. Technical and administration problems accounted for 43.9% of missing data ( $N = 28$ ; lack of sound/picture, inappropriate film angle, etc.), but missing data were also due to, e.g., refusal to be videotaped or disparticipation. Missingness analyses revealed no significant differences between participants and non-participants neither on sex ( $p = .314$ ) nor age ( $p = .714$ ). Likewise, participation rates ( $p = .279$ ) and reason for missing ( $p = .438$ ) did not differ between risk groups.

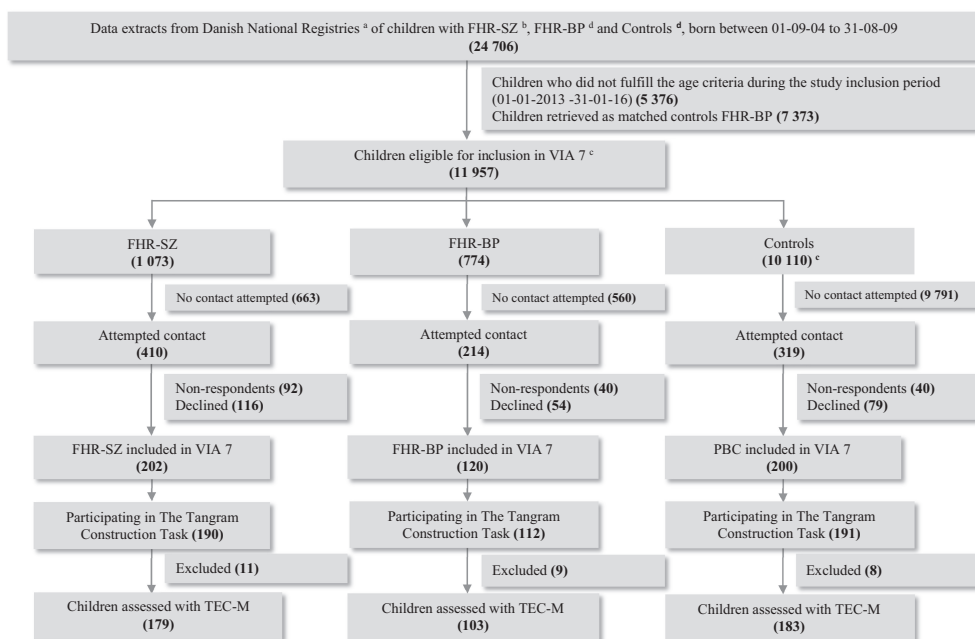
## Measures

### The tangram emotion coding manual (TEC-M)

The Tangram Emotion Coding Manual (TEC-M) (Hagstrøm et al., 2019) was developed to assess behaviours reflecting children's emotion regulation while performing a difficult puzzle (the Tangram Construction Task, [Hudson & Rapee, 2001]) in the context of the interaction between the primary caregiver and the child.

The TEC-M is a standardized manual for coding The Tangram Construction Task and is theoretically founded on the process model of emotion regulation, which is a well-recognized schematic model of the processes involved in emotion regulation (Gross, 1998, 2014, 2015; Hagstrøm et al., 2019). The process model describes five regulatory processes ('situation selection', 'situation modification', 'attention deployment', 'cognitive change', and 'response modulation'). The items on the TEC-M were designed to mirror these strategies, e.g., the use of verbal reappraisal under 'cognitive change'. TEC-M was developed as a tool to measure emotion regulation abilities across potential diagnoses as well as in typically developing children.

In the Tangram Construction Task, the child is instructed to solve as many puzzles as possible within 5 min. The child and the primary caregiver are told that most children can solve these puzzles, but that some find it a bit difficult. The primary caregiver is handed a booklet with the solutions to the puzzles. The primary caregiver is instructed to support the child, but it is emphasized that the primary caregiver should only help the child if it is genuinely needed. The task purpose is to frustrate the child and elicit emotional responses from both the child and the primary caregiver. Hence, the puzzle task is



**FIGURE 1** Flowchart of inclusion of children in The Danish High Risk and Resilience Study Via 7 and participation in The Tangram Construction Task<sup>1</sup> and assessed with the Tangram Emotion Coding Manual<sup>1</sup> (TEC-M) for emotion regulation ability. FHR-SZ: Children of parents with schizophrenia spectrum disorders. FHR-BP: Children of parents with bipolar disorder. Controls: Population-based control children of parents with no diagnoses of schizophrenia spectrum disorders or bipolar disorder. FHR: familial high risk; BP: bipolar disorder; PBC: population-based control; SZ: schizophrenia. <sup>a</sup> Danish Civil Registration System and Danish Psychiatric Central Research Register. <sup>b</sup> Parents with both diagnosis of schizophrenia and bipolar disorder were assigned to the schizophrenia high risk group as per the ICD-10 hierarchy. <sup>c</sup> Up to 10 controls were retrieved for each child in the schizophrenia spectrum disorder group and the bipolar disorder group. Controls were matched to cases on sex, municipality, and exact age<sup>1</sup>(Hagstrøm et al., 2019).

considerably more difficult than communicated to the participants. The session is videotaped with only the child and the primary caregiver in the room during the task. The video-recording is used for subsequent qualitative coding according to the manual, focusing on the child and primary caregiver. Eight items are coded for parental personal and interpersonal behaviour (intrusiveness, avoidance, control, verbal reappraisal, tension, positive expressions, negative expressions, and support sensitivity), and 11 items are coded for child behaviour (situation rejection, avoidance/resignation, control, narration, verbal reappraisal, reassurance-seeking, tension, incongruent positive affect, positive expressions, negative expression, and aggression). All child and parent items are scored on a four-point frequency scale (never, rarely, sometimes, often) and a three-point intensity scale (mild, moderate, marked). One item assessing the parent–child dyad (emotional warmth) is scored from 0 to 3.

The primary outcome measure of the TEC-M, the emotion regulation scale, EmReg, represents an assessment of the child's overall emotion regulation. The EmReg is a clinician-rated global assessment taking into account all factors of the evaluated test situation, such as the child's level of frustration, parental behaviour, and perceived level of difficulty in coherence with the coded items described above. The EmReg score is scored on a five-point scale, where a score of one represents very poor emotion regulation and a score of five represents excellent emotion regulation.

Two trained raters (KR and KSS) conducted the coding of all videos from The Danish High Risk and Resilience Study – VIA 7. Reliability and harmonizing ratings on TEC-M coding were carried out with regular intervals. Both raters coded a subset of 14 videos enabling the calculation of inter-rater reliability (IRR). IRR was found to be in the excellent range (Cicchetti, 1994) for the EmReg;

ICC = 0.79 (95% CI 0.34–0.93) when analysing the degree of rating consistency between raters, across subjects, assessed with intra-class correlation coefficient (ICC) based on raw data estimates by method of two-way mixed-effects model, average measures, and absolute agreement type (McGraw & Wong, 1996).

### The child behavior checklist (CBCL)

Primary caregivers filled out the CBCL (Achenbach & Rescorla, 2001). Beyond the CBCL-Total score, an emotional/behavioural dysregulation profile (CBCL-DP) can be computed from the CBCL. The CBCL-DP comprises three symptom scales from the CBCL (Aggressive Behavior, Anxious/Depressed, and Attention Problems) (Achenbach & Rescorla, 2001; Althoff, 2010). Two versions of the CBCL-DP are generally used, the stringent CBCL-DP70 (i.e., T-scores above or equal to 70 on all of the three individual symptom scales) or the broader CBCL-DP210 (i.e., a sum of the three symptom scale T-scores above or equal to 210) (Aitken et al., 2019). A CBCL-DP score above the cutoff suggestively predicts subsequent psychopathology and poor functioning (Biederman et al., 2012). We computed a normalized CBCL-DP T-score (mean = 50 and SD = 10) from our study sample's control group for boys and girls separately, with higher scores indicating more problem behaviour. In this study, we utilized both the stringent CBCL-DP70 and the broader CBCL-DP210.

### The attention-deficit/hyperactivity disorder-rating scale (ADHD-RS)

Primary caregivers and the child's teacher independently filled out the Danish version of the ADHD-RS (Barkley et al., 1999; DuPaul et al., 1998; Makransky & Bilenberg, 2014). Higher scores on ADHD-RS reflect worse symptom severity. ADHD-RS composite scores (ADHD-RS-comp) were calculated as the mean from the primary caregiver and teacher-rated ADHD-RS item 1–18, thus excluding conduct disorder item scores (Martel et al., 2015). A normalizing T-score was calculated by the same method as described for CBCL-DP.

### Children's global assessment scale (CGAS)

The child's current level of daily functioning was assessed with the CGAS (Shaffer et al., 1983). Higher CGAS scores reflect higher level of functioning.

## Statistical analysis

### Participant characteristics

Between-group differences of participant characteristics were analysed using one-way ANOVAs, chi-square tests, or the Mantel–Haenszel linear-by-linear test of associations, as appropriate (Table 1).

### Group differences in emotion regulation

The primary outcome measure, EmReg, was approximately normally distributed for both the total cohort and the three high-risk groups separately as evaluated from indicators of skewness, kurtosis, histograms, and boxplots. Assessing between-group differences of emotion regulation employing ANOVA with post hoc Tukey–Kramer was thus considered not to increase the risk of type I errors (Blanca et al., 2017).

TABLE 1 Characteristics of child and adult participants evaluated with The Tangram Emotion Coding Manual (TEC-M)

	Study group			p-value	p-value Pairwise comparison		
	FHR-SZ	FHR-BP	Controls		FHR-SZ vs. Controls	FHR-BP vs. Controls	FHR-BP vs. FHR-SZ
Children, N	179	103	183				
Female, N (%)	81 (45.3)	45 (43.7)	86 (47.0)	.859 <sup>a</sup>	-	-	-
Age at inclusion, mean (SD)	7.85 (0.21)	7.86 (0.21)	7.82 (0.20)	.200 <sup>b</sup>	-	-	-
CGAS, mean (SD)	67.7 (15.3)	73.6 (15.0)	77.7 (13.5)	.000 <sup>b</sup>	.000 <sup>b</sup>	.022 <sup>b</sup>	.001 <sup>b</sup>
Any Axis I diagnosis <sup>e</sup> , N (%)	65 (36.3)	39 (37.9)	28 (15.4)	.000 <sup>a</sup>	.000 <sup>a</sup>	.000 <sup>a</sup>	.795 <sup>a</sup>
Any ADHD diagnosis <sup>c</sup> , N (%)	36 (20.1)	9 (8.7)	13 (7.1)	.000 <sup>a</sup>	.000 <sup>a</sup>	.628 <sup>a</sup>	.012 <sup>a</sup>
Any DBD diagnosis <sup>c</sup> , N (%)	10 (5.6)	4 (3.9)	2 (1.1)	.063 <sup>a</sup>	.017 <sup>a</sup>	.116 <sup>a</sup>	.526 <sup>a</sup>
CBCL (N = 447), mean (SD) <sup>d</sup>	27.6 (21.2)	22.2 (18.9)	16.9 (14.6)	.000 <sup>b</sup>	.000 <sup>b</sup>	.021 <sup>b</sup>	.021 <sup>b</sup>
CBCL-DP210 (N = 446), N (%)	22 (12.7)	6 (6.3)	7 (4.0)	.008 <sup>a</sup>	.003 <sup>a</sup>	.395 <sup>a</sup>	.096 <sup>a</sup>
CBCL-DP70 (N = 446), N (%)	7 (4.0)	0 (0.0)	0 (0.0)	.004 <sup>a</sup>	.007 <sup>a</sup>	-	.046 <sup>a</sup>
ADHD-RS-Comp (N = 462), mean (SD) <sup>k</sup>	12.8 (9.9)	10.5 (9.2)	8.7 (7.4)	.000 <sup>b</sup>	.000 <sup>b</sup>	.022 <sup>b</sup>	.001 <sup>b</sup>
Primary caregivers <sup>c</sup> , N	179	103	183				
Female, N (%)	158 (88.3)	96 (93.2)	163 (89.1)	.398 <sup>a</sup>	-	-	-
Primary caregiver is index <sup>f</sup> , N (%)	81 (45.3)	61 (59.2)	-	-	-	-	.024 <sup>a</sup>
PSP <sup>f</sup> , mean (SD) <sup>g</sup>	72.6 (14.4)	74.8 (13.9)	84.3 (9.4)	.000 <sup>b</sup>	.000 <sup>b</sup>	.000 <sup>b</sup>	.153 <sup>b</sup>
Age at child's birth, mean (SD)	30.53 (6.68)	31.57 (5.54)	32.29 (4.25)	.011 <sup>b</sup>	.003 <sup>b</sup>	.302 <sup>b</sup>	.130 <sup>b</sup>
Employed or studying <sup>h</sup> , N (%) <sup>i</sup>	118 (66.3)	70 (68.0)	163 (90.6)	.000 <sup>a</sup>	.000 <sup>a</sup>	.000 <sup>a</sup>	.774 <sup>a</sup>

TABLE 1 (Continued)

	Study group		<i>p</i> -value Pairwise comparison		
	FHR-SZ	FHR-BP	Controls	<i>p</i> -value	FHR-SZ vs. FHR-BP vs. Controls
	FHR-SZ vs. Controls	FHR-BP vs. Controls	FHR-SZ vs. Controls	FHR-BP vs. Controls	FHR-SZ vs. FHR-BP vs. Controls
Education, N	176	102	181		
Primary/lower secondary, N (%)	31 (17.6)	5 (4.9)	5 (2.8)	.000 <sup>f</sup>	.248 <sup>g</sup>
Upper secondary, vocational, short-cycle tertiary, N (%)	76 (43.2)	33 (32.4)	77 (42.5)	.000 <sup>f</sup>	.000 <sup>h</sup>
Bachelor degree, equivalent or higher, N (%)	69 (39.2)	64 (62.7)	99 (54.7)		

*Note:* In case information is not available for all participants, an N indicates the number of informants.

In 8 cases, data for the same primary caregiver are counted twice because of siblings with shared primary caregiver. A total of 10 sibling pairs participated.

Abbreviations: ADHD, Attention-deficit/hyperactivity disorder; ADHD-RS-Comp, The modified version of the ADHD-Rating Scale parent-teacher composite score; BP, bipolar disorder; CBCL-DP, Child Behavior Checklist-Dysregulation Profile; CBCL-DP70, t-score above 70 on the three CBCL-DP subscales; CBCL-DP210, t-score above 210 on the combined CBCL-DP subscales; CGAS, Children's Global Assessment Scale; DBD, Disruptive behaviour disorder; FHR, familial high risk; PSP, The Personal and Social Performance Scale; SZ, schizophrenia spectrum disorder.

<sup>a</sup>Chi square test.

<sup>b</sup>ANOVA.

<sup>c</sup>Best estimate diagnosis made from diagnostics interviews (K-SADS-PI) with child and primary caregiver and all other available information on the child.

<sup>d</sup>173 FHR-SZ, 97 FHR-BP, 177 controls.

<sup>e</sup>Primary caregivers are defined as the parent or foster parent that knows the child best and spends most time with the child.

<sup>f</sup>Index is defined as the parent with a diagnosis of either schizophrenia spectrum disorder or bipolar disorder and their adult matched control.

<sup>g</sup>177 FHR-SZ, 103 FHR-BP, 181 control-group primary caregivers.

<sup>h</sup>Employed or studying is defined as being under employment (including temporary leave) or adhering to an acknowledged education for a minimum of 15 hours weekly.

<sup>i</sup>178 FHR-SZ, 103 FHR-BP, 180 control-group primary caregivers.

linear-by-linear association.

<sup>j</sup>179 FHR-SZ, 102 FHR-BP, 181 controls.



## Association between emotion regulation and dimensions of psychopathology and general functioning

Spearman's rank-order correlation analyses were performed as a monotonic relationship was evaluated between EmReg and the continuous dependent variables. For dichotomous dependent variables, Pearson's correlations were performed.

We used IBM SPSS Statistics, version 25.0, for all statistical analyses.

## RESULTS

### Participant characteristics

There was no difference between groups regarding age and sex. Compared to the control group, children from both FHR groups had significantly lower general levels of functioning (CGAS) and were rated higher on the dimensional measure of psychopathology (CBCL). Likewise, primary caregivers from both FHR groups had significantly lower levels of functioning than primary caregivers from the control group (Table 1). These results correspond to the results of the full VIA 7 cohort (Ellersgaard et al., 2018).

### Group differences in emotion regulation

We found no significant difference between the three groups on EmReg as determined by a one-way ANOVA ( $F [2462] = 1.155, p = .316$ ) (Table 2).

### Associations between emotion regulation and dimensions of psychopathology and general functioning

Analyses using Spearman's rank-order correlation on the total cohort revealed a statistically significant, weak, and inverse correlation between the EmReg score and ADHD-RS-comp ( $r_s[460] = -.170, p = < .001$ ) and CBCL-Total ( $r_s[445] = -.132, p = .005$ ). Pearson's correlations between EmReg and the broader CBCL-DP210 were significant, weak, and negative;  $r(444) = -.177, p = < .001$ . The stringent CBCL-DP70 was non-significantly correlated with the EmReg score (Table 3). CGAS was significantly, weak, and positively correlated with EmReg ( $r_s[462] = .150, p = .001$ ) (Table 3).

All within-group associations were likewise weak. Associations between EmReg and ADHD-RS were only significant in the FHR-SZ group ( $r_s[177] = -.273, p = < .001$ ). CBCL-DP210 was significantly associated with EmReg in both the FHR-SZ and the control group, and in the control group, associations between EmReg and the CBCL-Total and the CGAS were likewise significant. The only significant association in the FHR-BP group was between EmReg and CGAS (Table 3).

## DISCUSSION

We did not find any group differences between the two familial high-risk groups and the control group on the primary outcome measure of emotion regulation, EmReg, measured by the TEC-M. We did, however, find statistically significant, weak associations between emotion regulation (the EmReg score) and general functioning (CGAS), a dimensional measure of psychopathology (CBCL), ADHD symptom severity (ADHD-RS), and between emotion regulation and a dysregulation profile (CBCL-DP210) in the total cohort.

The stringently defined profile; CBCL-DP70, was only found in very few FHR-SZ children and none of the FHR-BP nor the control children. The insignificant results regarding the association between

TABLE 2 Emotion Regulation (EmReg) as measured with the Tangram Emotion Coding Manual (TEC-M)

	Study group			<i>p</i> -value	<i>p</i> -value Pairwise comparison		
	FHR-SZ	FHR-BP	Controls		FHR-SZ vs. Controls	FHR-BP vs. Controls	FHR-BP vs. FHR-SZ
Children, <i>N</i>	179	103	183				
EmReg, mean [95% CI]	3.21 [3.06–3.36]	3.39 [3.21–3.56]	3.25 [3.11–3.40]	.316 <sup>a</sup>	–	–	–

Abbreviations: BP: bipolar disorder; EmReg: Emotion regulation ability, higher scores represent better emotion regulation; FHR: familial high risk; SZ: schizophrenia spectrum disorder.

<sup>a</sup>ANOVA.

EmReg and CBCL-DP70 may thus be due to the small number of children found with a dysregulation profile (type II error). Associations between EmReg and the broader defined dysregulation profile, CBCL-DP210, were highly significant for the total cohort and in the FHR-SZ group, the group with the highest frequency of a CBCL-DP210 profile. The non-significant associations between EmReg and the CBCL-DB210 in the FHR-BP group and the control group may also be due to the small number of children identified with the dysregulation profile in these groups (type II error).

Better emotion regulation was positively related to higher levels of daily functioning; although we cannot infer causation, this could possibly suggest an underlying importance of apt emotion regulation for success in everyday activities like school and leisure time activities, and in initiating and maintaining social relations to peers. This is supported by findings of emotion regulation being associated with academic performance as early as age five and that academic performance is found relatively stable after 1 grade (Graziano et al., 2007). Furthermore, adaptive emotion regulation is recognized as a competence that can increase the individual's well-being and chances of overcoming adverse life events or minor obstacles in daily life (National Scientific Council on the Developing Child, 2015). Apt emotion regulation is thus likely a critical resilience factor in life.

On account of the higher prevalence of psychopathology in FHR-SZ and FHR-BP children in this FHR population as documented with diagnostic interviews (Ellersgaard et al., 2018) and the significantly worse psychopathological symptom severity measured dimensionally (Table 1), we expected to find poorer emotion regulation in the two high-risk groups compared to the control group. We did nevertheless not find any significant differences in emotion regulation between the three groups. It is, however, essential to consider that the participating children are at an age where their emotion regulation is still developing and maturing (Plessen & Kabicheva, 2010; Schweizer et al., 2020). Thus, later in the maturational development, an abnormal emotion regulation may emerge in the FHR groups (Dickson et al., 2018; Reichenberg et al., 2010). Poorer emotion regulation was modestly associated with worse ADHD symptom severity and more signs of psychopathology in the total cohort. These results could suggest that emotion regulation difficulties are not specific to offspring with a predisposition for schizophrenia or bipolar disorder but rather related to developmental disorders such as ADHD. Yet another consideration is that emotion regulation in all forms (as is true of all other types of regulation) requires resource capacity and that different forms of emotion regulation require different forms of cognitive capacity (Urry & Gross, 2010). This entails two different presumptions (1) regulation capacity as a depletable resource and (2) differences in cognitive capacity in different cognitive domains maybe the Tangram Construction Task was not demanding or lengthy enough to elucidate emotion regulation difficulties in average to high-capacity children and only children with an inherent low capacity to regulate would show emotion regulation difficulties. Following this hypothesis, the association between ADHD symptom load and poor emotion regulation could be explained by an inherent low capacity to regulate in children with ADHD. The second presumption of cognitive capacity again supports the association between poor emotion regulation and ADHD, as ADHD is defined on the basis of dysfunctional regulation of, e.g., attention (Petrovic & Castellanos, 2016).

TABLE 3 Correlation between emotion regulation (EmReg) and measures of psychopathology, symptom severity, and daily life functioning

	Total Cohort			FHR-SZ			FHR-BP			Controls		
	Correlation coefficient (df)	p-value	Correlation coefficient (df)	p-value	Correlation coefficient (df)	p-value	Correlation coefficient (df)	p-value	Correlation coefficient (df)	p-value	Correlation coefficient (df)	p-value
ADHD-RS-comp	-.170 (460) <sup>a</sup>	<.001	-.273 (177) <sup>a</sup>	<.001	-.107 (100) <sup>a</sup>	.284	-.130 (179) <sup>a</sup>	.082				
CBCL-Total	-.132 (445) <sup>a</sup>	.005	-.111 (171) <sup>a</sup>	.147	-.177 (95) <sup>a</sup>	.082	-.173 (175) <sup>a</sup>	.021				
CBCL-DP210	-.177 (444) <sup>b</sup>	<.001	-.199 (171) <sup>b</sup>	.009	-.194 (94) <sup>b</sup>	.058	-.156 (175) <sup>b</sup>	.039				
CBCL-DP70	-.087 (444) <sup>b</sup>	.065	-.140 (171) <sup>b</sup>	.068	-	-	-	-				
CGAS	.150 (462) <sup>a</sup>	.001	.137 (177) <sup>a</sup>	.068	.201 (101) <sup>a</sup>	.042	.173 (180) <sup>a</sup>	.020				

Note: ADHD-RS-Comp: The modified version of the ADHD-Rating Scale parent-teacher composite score.

EmReg: Overall Emotion Regulation Scale. Higher scores equal better emotion regulation.

CBCL: Child Behavior Checklist. Higher scores equal more symptomatology.

CBCL-DP: Child Behavior Checklist Dysregulation Profile. DP70 and DP210 are dichotomous outcome measure of whether the child fulfills criteria for the profile. CBCL-DP70: t-score above 70 on the three CBCL-DP subscales; CBCL-DP210: t-score above 210 on the combined CBCL-DP subscales.

CGAS: Children's Global Assessment Scale. Higher scores equal better performance.

<sup>a</sup>Spearman's rank-order correlation.

<sup>b</sup>Pearson's correlation.

We chose to analyse associations across the whole study cohort, regardless of risk status as well as within-group. This was done considering two different conceptualizations of emotion regulation in children with FHR for severe mental illness: First, emotion regulation and emotion regulation development is inherently affected by, and should be primarily understood in relation to, the specific psychiatric disorder of the parent. Second, emotion regulation is related to the child's own potential psychopathology. Hereby not neglecting the fact that both scenarios are intrinsically connected to genetic and environmental dispositions related to high-risk status. The fact that we find associations between emotion regulation and measures of psychopathology including ADHD symptom severity across groups and no differences in emotion regulation between groups could again indicate that emotion regulation is more related to the child's own psychopathology than FHR status. However, it is essential to note that when analysing associations by group, significant association was only found in the FHR-SZ group and most likely this carries the significant result of the total group association. However, it is likewise important to note that only a very few FHR-BP and control children fulfilled the criteria for an ADHD diagnosis, which could account for the nonsignificant results within these groups. Future studies should assess the potential association between categorical psychopathology and emotion regulation.

We know from a previous study of the same cohort that the FHR-SZ children as a group have poorer cognitive functioning than control children (Hemager et al., 2018), and we also found expected differences in parental and primary caregiver educational attainments, employment status, and daily life functioning, measured by the Personal and Social Performance Scale between FHR-groups and the control group (Ellersgaard et al., 2018) (Table 1). These factors could affect the child's capabilities of solving the puzzles and increase frustration levels. However, we did not control for these factors as this could potentially remove key effects of being an FHR child and introduce the risk of overcorrection and thus rendering the results unrepresentative. Likewise, we did not correct for sociodemographic factors as not to risk statistical overcorrection. Utilizing a population-based cohort, we wished to increase the knowledge of emotion regulation in children with a familial high risk of schizophrenia or bipolar disorder compared to controls by using a test that was easy to apply and ecologically valid. The finding of non-different emotion regulation between groups indicates that TEC-M may not be sensitive enough in this specific population at this age as previous findings indicate that problems with emotion regulation are present in children with familial high risk of severe mental disorders (Díaz-Caneja et al., 2018; Donatelli et al., 2010; Johnson et al., 2014). Even though the test is designed to resemble a naturalistic situation, it is a lab setting and thus does not entail all the demands of everyday life. Evaluation of real-life emotion regulation may potentially be more valid in this population at this age.

A considerable strength is the uniqueness of the sizeable register-based study sample of same-age children. Furthermore, most studies of emotion regulation rely on questionnaires (Compas et al., 2017; Wakschlag et al., 2007) that can be subject to recall biases. To overcome this methodological limitation, we investigated emotion regulation with an ecologically valid clinician-rated observational test measure, thus adding to previous knowledge of emotion regulation investigations employing other methods. A further strength contributing to the validity of the results of the study is the interrater reliability (IRR), which was found in the excellent range regarding the emotion regulation ability measure, EmReg. This indicates a high degree of agreement, suggesting that a negligible amount of measurement error was introduced by the two independent raters despite a rather small sample of 14 cases. The ICC confidence intervals of the IRR are somewhat wide, indicating a risk that the ICC value might not hold the standard of excellence. However, this is to be expected due to the low number of cases assessed for IRR. The high IRR corresponds to the results found in the TEC-M validation study, supporting the validity of the IRR in the present study (Hagstrøm et al., 2019). However, the present study also has some limitations. Emotion regulation partly develops through social interaction and in childhood, especially in close interaction with the primary caregiver (Plessen & Kabicheva, 2010). Severe mental illness can potentially affect the primary caregivers' ability to support their child's emotion regulation development (National Scientific Council on the Developing Child, 2015). As we used parental lifetime diagnoses in the inclusion process, the cohort includes children of parents with current illness as well as of parents that might have been ill shortly and in remission/well for many years and even before the birth of the child. Also,

the severity of illness of parents in the cohort may vary significantly. This limitation could account for why we, in the present study, did not find any significant group differences. Maybe a more uniform sample with more narrow inclusion criteria, e.g., diagnosis given no later than the year before the child's birth or, a requirement of active illness during the child's lifetime would have revealed a different result. A further limitation of the study is that we were not able to control for parental psychopathology in the control group, other than schizophrenia/bipolar disorder, which may partly explain the lack of significant differences between the FHR group and the control group, as parents in the control group may have other psychiatric disorders, which could constitute a factor in child emotion regulation. Conversely, allowing for some psychopathology in the control group increases the representativeness of the study.

Future studies should consider the potential association between the severity of parental psychopathology on the child's emotion regulation abilities, and not just a categorical diagnostic classification as in the current study.

## CONCLUSION

To our knowledge, this is the first study to compare emotion regulation between FHR-SZ and FHR-BP children applying an ecologically valid clinician-rated observational test. At age seven, emotion regulation in familial high-risk children did not differ from controls, although poorer emotion regulation was associated with a higher level of psychopathology and more ADHD symptoms. Also, emotion regulation efficacy correlated positively with daily life functioning of the child. These findings contribute to the understanding of emotion regulation in familial high-risk children and the difficulties related to poor emotion regulation. Identification of poor emotion regulation is important as maladaptive emotion regulation is associated with adverse outcomes, and conversely, adaptive emotion regulation is associated with a positive outcome and general well-being. Improving emotion regulation may be a relevant mean to improve the overall outcome and well-being of the individual, and further studies with observational measures of emotion regulation in familial high-risk children are warranted.

## AUTHOR CONTRIBUTIONS

**Julie Hagstrøm:** Conceptualization; formal analysis; investigation; methodology; writing – review and editing. **Ditte Ellersgaard:** Investigation; methodology; writing – review and editing. **Camilla Christiani:** Investigation; methodology; writing – review and editing. **Nicoline Hemager:** Investigation; methodology; writing – review and editing. **Birgitte Klee Burton:** Investigation; writing – review and editing. **Aja Neergaard Greve:** Investigation; writing – review and editing. **Kirsten Rohr:** Investigation; writing – review and editing. **Ditte Gantriis:** Investigation; writing – review and editing. **Signe Vangkilde:** Formal analysis; investigation; writing – review and editing. **Ole Mors:** Funding acquisition; resources; supervision; writing – review and editing. **Merete Nordentoft:** Funding acquisition; investigation; methodology; project administration; supervision; writing – review and editing. **Carsten Obel:** Methodology; supervision; writing – review and editing. **Kerstin Jessica Plessen:** Conceptualization; funding acquisition; investigation; project administration; supervision; writing – review and editing. **Jens Richardt Moellegaard Jepsen:** Conceptualization; methodology; formal analysis; supervision; writing – review and editing. **Anne A.E. Thorup:** Conceptualization; funding acquisition; investigation; methodology; project administration; supervision; writing – review and editing.

## ACKNOWLEDGEMENTS

The authors would like to express their gratitude to the dedicated families participating in the study, to M. Skjærbæk, A. Ranning, A. Søndergaard, M. Gregersen, H. Jensen, C. Gregersen, D-M. Henriksen, H. Stadsgaard, K. Zahle, and M. Melau for contributing to data collection; to C. Bøcker Pedersen and M. Gjørtz Pedersen for retrieving the register extract; to M. Chaine and J. Ohland for help with data management and Emma Louise Sillehoved for data entry; to P.B. Mortensen T. Werge, D. Hougaard, and A. Børglum for collaboration in iPSYCH; and to B. Hoff Esbjørn for

collaboration preceding the TEC-M development. This work was supported by TrygFonden, the Mental Health Services of the Capital Region of Denmark, The Lundbeck Foundation Initiative for Integrative Psychiatric Research (iPSYCH) (Grant Nos. R102-A9118 and R155-2014-1724), Aarhus University, and the Beatrice Surovell Haskell Fund for Child Mental Health Research of Copenhagen (Grant No. J.NR 11531).

### FUNDING INFORMATION

This work was supported by TrygFonden, the Mental Health Services of the Capital Region of Denmark, The Lundbeck Foundation Initiative for Integrative Psychiatric Research (iPSYCH) (Grant Nos. R102-A9118 and R155-2014-1724), Aarhus University, and the Beatrice Surovell Haskell Fund for Child Mental Health Research of Copenhagen (Grant No. J.NR 11531).

### CONFLICT OF INTEREST

The authors have no conflicts of interest to disclose.

### DECLARATIONS

The study was approved by the Danish Data Protection Agency (J.nr.:2012-58-0004) and follows all present laws concerning personal data. The Danish Ministry of Health granted the permission to draw data from Danish national Registers. The Danish National Committee on Health Research Ethics evaluated the study and concluded that further approval was not required due to the observational nature of the study. Verbal and written informed consent was obtained for all participants or custody holders, including a separate written consent to contact the child's teacher. The study was funded by TrygFonden, the Mental Health Services of the Capital Region of Denmark, The Lundbeck Foundation Initiative for Integrative Psychiatric Research (iPSYCH), Aarhus University, and the Beatrice Surovell Haskell Fund for Child Mental Health Research of Copenhagen.

### DATA AVAILABILITY STATEMENT

Data are available upon request.

### ETHICS APPROVAL

The VIA 7 study was approved by the Danish Data Protection Agency (J.nr.:2012-58-0004). The Danish National Committee on Health Research Ethics evaluated the study and concluded that further approval was not required due to the observational nature of the study.

### CONSENT TO PARTICIPATE

Verbal and written informed consent was obtained for all participants or custody holders, including a separate written consent to contact the child's teacher.

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**How to cite this article:** Spang, K. S., Hagstrøm, J., Ellersgaard, D., Christiani, C., Hemager, N., Burton, B. K., Greve, A. N., Rohr, K., Gantriis, D., Vangkilde, S., Mors, O., Nordentoft, M., Obel, C., Plessen, K. J., Jepsen, J. R. M., & Thorup, A. A. E. (2022). Emotion regulation in 7-year-old children with familial high risk for schizophrenia or bipolar disorder compared to controls – The Danish High Risk and Resilience Study – VIA 7, a population-based cohort study. *British Journal of Clinical Psychology*, *00*, 1–16. <https://doi.org/10.1111/bjc.12382>