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Emotion Regulation in Children with Tourette Syndrome

PhD Thesis

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Preface

This thesis was submitted in February 2019 to obtain the PhD degree in Psychiatry at the Graduate School of Health and Medical Sciences, University of Copenhagen. The work on which this thesis was based was carried out at the Child and Adolescent Mental Health Center, Mental Health Services in the Capital Region of Denmark in the period from 2013-2018.

When I joined the research unit after completing my master's education in psychology in 2012, I became involved in research projects centered around children with Tourette syndrome (TS). I was immediately intrigued by this group of children at the intersection between neurology and psychiatry and the resulting variations of clinical presentations. When meeting the families affected by TS, it was interesting to see that the behavior causing distress was rarely related solely to the tics per se. Quite often, the focus was on dysregulated emotional behavior and rage attacks of varying severity. Arriving with an initial interest in emotion regulation (ER), it was clear that the connection between ER and TS would be interesting to explore. A literature search at the time on studies of TS and ER did not produce a single published paper addressing the specific association. Whereas TS had been studied in relation to the prevalence of explosive behavior and rage attacks, no one had investigated the direct link with ER. This marked the beginning of my PhD project. Five educational years later (due to two one-year maternity leaves), this work has now come to an end, though my interest in research in child and adolescent mental health most certainly has not.

Julie Hagstrøm
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List of papers

This thesis is based on the following papers.

I. **The Puzzle of Emotion Regulation – Development and Evaluation of the Tangram Emotion Coding Manual for Children**

   Julie Hagstrøm, Katrine S. Spang, Bianca Munkebo Christiansen, Katrine Maigaard, Signe Vangkilde, Barbara Hoff Esbjørn, Jens Richardt Møllegaard Jepsen, Kerstin Jessica Plessen. *Manuscript in review.*

II. **Emotion Regulation in Children with Tourette Syndrome – Associations with ADHD and Tic Severity**

   Julie Hagstrøm, Katrine S. Spang, Jens Richardt Møllegaard Jepsen, Signe Vangkilde, Katrine Maigaard, Liselotte Skov, Anne Katrine Pagsberg, Kerstin Jessica Plessen. *Manuscript in review.*

III. **Reappraisal is an Effective Emotion Regulation Strategy in Children with Tourette Syndrome and ADHD**

   Julie Hagstrøm, Katrine Maigaard, Anne Katrine Pagsberg, Liselotte Skov, Kerstin Jessica Plessen, Signe Vangkilde.*Manuscript in review.*

List of abbreviations

The following abbreviations are used throughout this thesis.

ACC	Anterior cingulate cortex
ADHD	Attention-deficit/hyperactivity disorder
ADHD-RS	ADHD Rating Scale
ANOVA	Analysis of variance
ANCOVA	Analysis of covariance
BRIEF	Behavior Rating Inventory of Executive Function
CAMCH	Child and Adolescent Mental Health Center, Mental Health Services, Capital Region of Denmark
CBCL	Child Behavior Checklist
CD	Conduct disorder
CI	Confidence interval
CSTC	Cortico-striato-thalamo-cortical
CVR	Content validity ratio
DESR	Deficient Emotional Self-Regulation profile
DSM-IV	Diagnostic and Statistical Manual of Mental Disorders, Fourth Edition
DSM-5	Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition
EC	Emotional Control Scale
ECG	Electrocardiogram
EFA	Exploratory factor analysis
EmReg	Overall emotion regulation scale from the TEC-M

ER	Emotion regulation
fMRI	Functional magnetic resonance imaging
HRV	Heart rate variability
ICC	Intra-class correlation coefficient
ICD-10	International Statistical Classification of Diseases and Related Health Problems, 10th Revision
K-SADS-PL	Kiddie-Schedule for Affective Disorders and Schizophrenia, Present and Lifetime Version
MD	Mean difference
Mdn	Median
MSCEIT	The Mayer-Salovey-Caruso Emotional Intelligence Test
OCD	Obsessive-compulsive disorder
ODD	Oppositional defiant disorder
PANDAS	Pediatric Autoimmune Neuropsychiatric Disorders Associated with Streptococcal infections
PFC	Prefrontal cortex
PRI	Perceptual Reasoning Index
PRT	Picture Reappraisal Task
PU	Premonitory urge
PUTS	Premonitory Urge for Tics Scale
SPSS	Statistical Package for the Social Sciences
VCI	Verbal Comprehension Index
WISC-IV	Wechsler Intelligence Scale for Children – fourth edition
YGTSS	Yale Global Tic Severity Scale
SES	Socioeconomic status
TEC-M	Tangram Emotion Coding Manual
TS	Tourette syndrome

Thesis summary

Children's abilities to regulate their emotions undergo substantial developmental changes during childhood and are pivotal to engaging adaptively in differing social contexts. Fluctuations in these abilities are a natural part of development, but in some children with mental illness, emotionally dysregulated behavior can cause severe impairment and reduce quality of life. Researchers, clinicians, and parents of children with Tourette syndrome (TS) have often perceived rage attacks as far more detrimental to daily functioning than the child's tics, and explosive outbursts have frequently been considered integral to TS. Studies of the prevalence of disruptive behavior in individuals with TS, however, indicate that emotional dysregulation is associated with comorbid attention-deficit/hyperactivity disorder (ADHD)a. This association has primarily been examined with questionnaires and has not been investigated using experimental measures – possible due to a shortage of experimental, observational tests of emotion regulation (ER) ability.

The aim of the present study was to develop and validate a novel manual for coding children's ER abilities, and to examine these abilities in children with TS. Approximately 90% of individuals with TS have one or more comorbid disorders, resulting in a wide array of clinical presentations. We thus included groups of children with ADHD, TS + ADHD, and typically developing controls, respectively, to enable a clarification of ER difficulties as either a general factor in childhood psychopathology, or specific to separate diagnoses. We examined a total of 169 medication-naïve children, aged 8-12 years, with 60 children in the TS group, 27 children in the ADHD group, 22 children in the TS + ADHD group, and 60 controls matched on age and sex to the TS group. Children participated in two experimental tests of ER. The first was a frustration task, allowing for an assessment of children's spontaneously occurring behavioral responses to frustration with the newly developed manual. The second test evaluated children's ability to employ a specific ER strategy (cognitive reappraisal).

Children from the TS group and the control group did not differ on either measure. Children with ADHD and TS + ADHD, however, demonstrated significantly poorer abilities of spontaneous ER, but performed equally to children from the TS group and controls on the task assessing their effective use of cognitive reappraisal. The present results indicate that ER difficulties are not directly related to TS, when no comorbid disorders are present. Conversely, children with ADHD demonstrate difficulties in spontaneous regulation; however, they are capable of learning and employing an effective strategy, even when this strategy is cognitively demanding. These findings are highly relevant to the clinical approach to dysregulated emotional behavior. Although emotional impairments do not appear to be central to individuals with TS without comorbidity, high rates of comorbid disorders are the rule rather than the exception, emphasizing the need for thorough assessment of comorbidity and any associated disruptive behavior. Hopefully, an increased understanding of the underlying features of disruptive behavior, as well as a novel measure to assess ER abilities, will contribute to optimizing options for treatment.

Resumé (Danish summary)

Børns evner til emotionsregulering (ER) udvikles mærkbart igennem barndommen og er afgørende for at kunne indgå hensigtsmæssigt i relationer med andre. Udsving i disse evner er en naturlig del af udviklingen hos alle børn. Ved en række børne- og ungdomspsykiatriske lidelser ses der dog problemer med uhensigtsmæssig ER i en sådan grad, at det nedsætter barnets funktionsevne og livskvalitet. Mange forskere, klinikere og forældre til børn med Tourettes syndrom (TS) oplever raserianfald og eksplosiv adfærd som langt mere ødelæggende end barnets tics, og disse problemer med ER er blevet opfattet som en indgroet del af TS. Studier af prævalensen af raserianfald hos personer med TS har imidlertid peget på, at den eksplosive adfærd primært forekommer, når disse personer har attention-deficit/hyperactivity disorder (ADHD) i tillæg til deres tics. Denne sammenhæng er blevet studeret ved hjælp af spørgeskemaer, men er ikke blevet undersøgt eksperimentelt – potentielt på grund af en mangel på eksperimentelle, observationsbaserede tests af evner inden for ER.

Formålet med dette studie var således dels at udvikle og validere en ny manual til kodning af børns emotionsreguleringsevner og dels at undersøge ER hos børn med TS. Omkring 90% af personer med TS har en eller flere lidelser i tillæg, hvilket medfører vidt forskellige kliniske præsentationer. Vi inkluderede således grupper af børn med henholdsvis ADHD, TS + ADHD og en rask kontrolgruppe for at kunne specificere vores fund som enten en del af generel psykopatologi eller som gældende for specifikke diagnoser. I alt undersøgte vi 169 børn i alderen 8-12 år fordelt på 60 børn med TS, 27 børn med ADHD, 22 børn med TS + ADHD og 60 kontroller, som var matchet på alder og køn til TS-gruppen. Børnene deltog i to eksperimentelle tests af ER. Den første test var en frustrationsopgave, der muliggjorde en kodning med den nyudviklede manual af børnenes adfærd i frustrerende situationer, når de skulle regulere deres følelser spontant. Den anden test evaluerede børnenes evner til at anvende en specifik strategi til regulering (kognitiv revurdering).

Der var ingen forskel på, hvordan børn fra TS-gruppen og børn fra kontrolgruppen klarede sig på testene. Børn med ADHD og med TS + ADHD derimod klarede sig signifikant dårligere på opgaven, hvor de skulle regulere spontant, men klarede sig på linje med børn med TS og børn fra kontrolgruppen i forhold til brug af kognitiv revurdering. Resultaterne indikerer, at vanskeligheder med ER ikke er en del af TS, når disse børn ikke har andre komorbide lidelser. Samtidig vidner fundene om, at børn med ADHD har vanskeligt ved spontant at regulere deres emotioner, mens de er i stand til at lære og anvende en effektiv strategi, som ellers er forbundet med store krav til kognitiv kapacitet. Dette fund har stor relevans for den kliniske tilgang til dysreguleret emotionel adfærd. Fundene vedrørende børn med TS styrker den diagnostiske forståelse og understreger samtidig, at man skal være opmærksom på problemer med ER, når der er komorbiditet (særligt ADHD) til stede. Det er håbet, at en øget forståelse af evner inden for ER, som potentielt ligger til grund for eksplosiv adfærd, samt en ny metode til at undersøge disse evner, på sigt vil kunne bidrage til udviklingen af optimerede behandlingsstrategier.

Contents

1. Introduction

The link between Tourette syndrome (TS) and emotion regulation (ER) presents a research area of great importance to the understanding of childhood disruptive behavior and social and emotional problems persisting through adulthood in individuals with TS. From the clinical work with children with TS it was evident that rage attacks were frequent causes of concern in the family, and that parents of children with TS often tended to place widely differing types of behavior under the umbrella of TS. This prompted an investigation of the nature of dysregulated behavior in TS. Previous studies had demonstrated an impaired level of functioning in young adults with TS with this impairment being partly attributable to a comorbid diagnosis of attention-deficit/hyperactivity disorder (ADHD; Gorman et al., 2010). Additionally, a longitudinal study of children with tics demonstrated an increased occurrence of emotional and peer problems over time (Hoekstra, Lundervold, Lie, Gillberg, & Plessen, 2012). Despite reports of emotional problems accompanying the diagnosis of TS, our knowledge is limited in terms of the direct association between ER and TS and the potentially mediating effect of ADHD. The first part of this introduction will provide an overview of previous studies assessing any aspect of emotional processes in TS. Characterizations of TS and ADHD will follow, before operationalizing the concept of ER as applied in this thesis. For the final part of the introduction, an attempt will be made to tie things together by addressing the role of ER in psychopathology.

1.1 Existing literature on emotional processes in TS

To obtain an overview of previous research performed in the domain of emotional processes within the context of TS, we systematically searched the literature prior to commencing the study and replicated this search in January 2019. Using the terms "tourette*" and "emotion*", one investigator (JH) searched PubMed and PsycINFO following up with a manual search of relevant reference lists. The aim was to identify all studies using an experimental measure to investigate emotional processes in individuals with TS, be it in the domain of social cognition or emotion regulation, to achieve an understanding of any potential deviant profiles in the way these individuals perceive, experience, and respond to emotional stimuli. All abstracts were initially screened. We included all experimental case-control studies of an emotional process while also accepting questionnaire-based measures if they specifically focused on an emotional process as

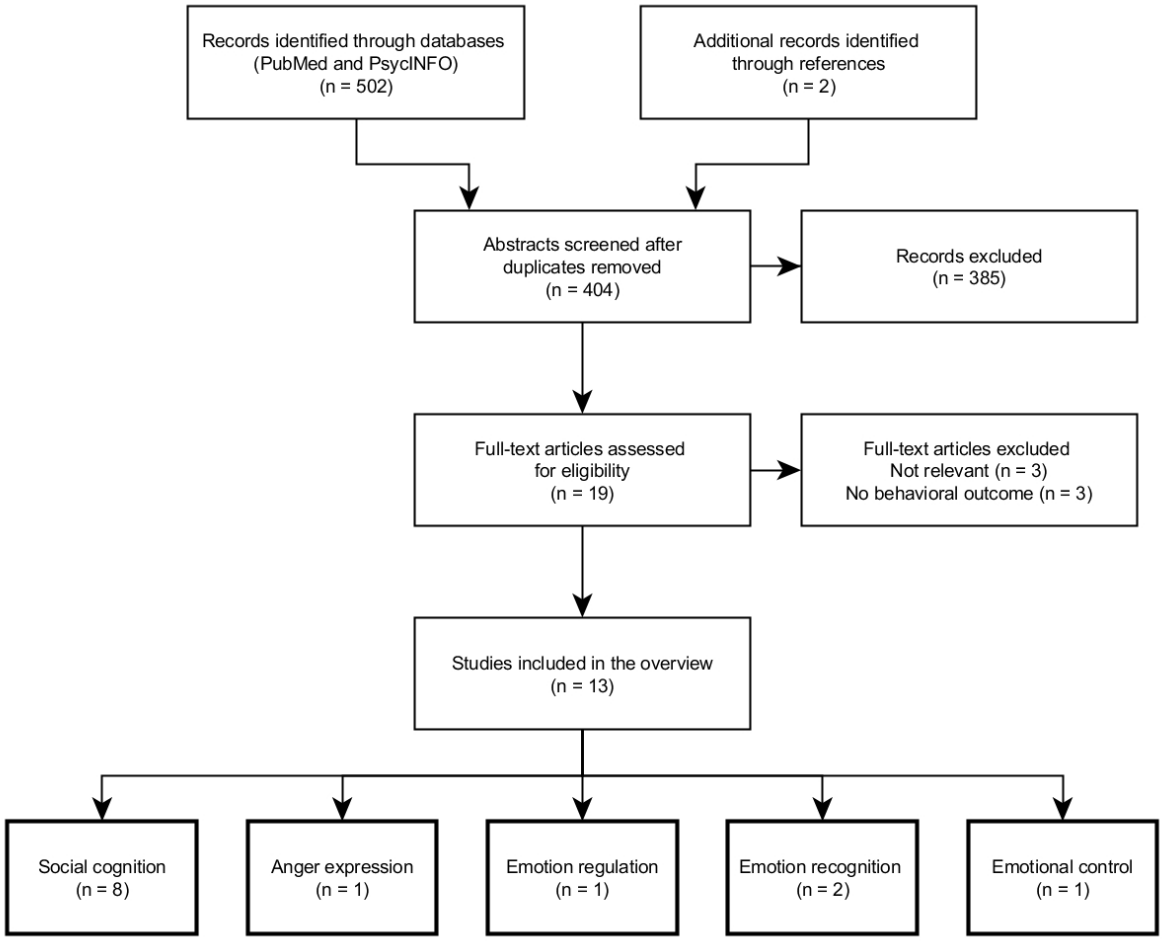

Figure 1. Flowchart of the systematic literature search. Studies presented in Table 1.

the main objective. We excluded reviews, theoretical papers, and studies addressing emotional problems, such as anxiety and depression, typically assessed with standard questionnaires on childhood problem behaviors.

1.1.1 Overview of findings

A total of 13 papers were reviewed for relevant findings (Table 1). For studies with neurophysiological measures, relevant findings were restricted to behavioral data. The resulting studies were divided into five categories depending on the domain under investigation (Figure 1). Eight studies addressed aspects of social cognition, making this field the most studied emotional process in TS. Social cognition broadly refers to cognitive processes involving other people, among those the ability to attribute thoughts and feelings to others (theory of mind) and to perceive and correctly interpret social stimuli. Social cognition may play a role in adaptive emotion regulation (Tamir & Mauss, 2011) and is thus relevant to the understanding of emotional processes in individuals with TS. The studies of social cognition mainly included adults with TS or TS-alone

(TS without any comorbidities) and produced mixed results, with some studies suggesting mild impairments in the TS samples, while others did not find any significant differences between controls and individuals with TS (Baron-Cohen, Jolliffe, Mortimore, & Robertson, 1997; Channon, Drury, Gafson, Stern, & Robertson, 2012; Channon, Sinclair, Waller, Healey, & Robertson, 2004; Drury, Shah, Stern, Crawford, & Channon, 2018; Eddy, Cavanna, & Hansen, 2017; Eddy, Mitchell, Beck, Cavanna, & Rickards, 2011, 2010b, 2010a). One study in adolescents and young adults with TS found no significant difference in the expression of anger compared with controls on a self-report questionnaire (Cavanna et al., 2015). Two studies of emotion recognition similarly suggested mild impairments in children and adults with TS in the recognition of certain feelings (Drury et al., 2012; Mermillod et al., 2013). One study on emotional control in children with TS assessed with a parent-reported questionnaire suggested more emotional problems in children with TS than in controls (Hovik et al., 2015). Of particular relevance to the work presented in this thesis is that only a single study addressed ER in individuals with TS (Drury, Wilkinson, Robertson, & Channon, 2016), and this study was carried out in a small sample of adults with mixed medication status using a self-report questionnaire.

It is noteworthy that the majority of studies included adult participants, as dysregulated emotional behavior and emotional problems are often childhood associated, and as adults may represent a selected group in terms of symptom severity and impairment (Bloch, 2013). Further, studies were generally carried out in small samples with wide age ranges. Remarkably, all studies accepted various types of medication, although the effects of psychotropic medication on emotional and cognitive processes have not been fully understood (Moritz, Andreou, Klingberg, Thoering, & Peters, 2013). The presence of comorbidity markedly affects behavior and level of functioning in children with TS (O'Hare, Helmes, Reece, Eapen, & McBain, 2016), and comorbid ADHD is known to cause impairment (Gorman et al., 2010; Tabori Kraft et al., 2012). Still, approximately half the studies comprised TS samples with varying comorbidity, thus obscuring findings from such groups in regard to TS specific impairment.

1.2 Tourette syndrome

The physician Georges Gilles de la Tourette became renowned for his characterization of TS as a distinct disorder following his 1885 publication describing nine individuals with TS (Ganos, Roessner, & Münchau, 2013). Although Gilles de la Tourette is often credited as the first to describe TS due to the eponymously named disorder, the first known record of TS dates back to 1825, when Jean Marc Itard from the Salpêtrière Hospital in Paris documented the syndrome in his description of motor and vocal tics in a French noblewoman, who would later become one of the nine individuals described by Gilles de la Tourette (Robertson, 2015b).

1.2.1 Clinical characteristics, prevalence, and course of the disorder

TS is a neurodevelopmental disorder characterized by multiple motor and at least one vocal tic presenting before the age of 18 with a duration of more than one year. The classifications of TS are identical for the International Statistical Classification of Diseases and Related Health Problems, 10th Revision (ICD-10; World Health Organization, 2016) and the Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition (DSM-5, American Psychiatric Associa-

Table 1. Identified studies with a main objective of examining an emotional process in children, adolescents, and adults with Tourette syndrome.

Reference	Sample	Age [years]	Domain	Measure	Comorbidity/medication	Relevant findings
Baron-Cohen et al., 1997	High functioning autism or Asperger's syndrome (n = 16) TS (n = 10) Controls (n = 50)	18-48	Social cognition	The Eyes Task (experimental)	Unknown	No difference between patients with TS and controls.
Cavanna et al., 2015	TS (n = 25) Controls (n = 41)	12-22	Anger expression	State-Trait Anger Expression Inventory (STAXI) (self-report)	Varying comorbidity in the TS sample, mixed medication status	TS and controls did not differ on any measure of the STAXI.
Channon et al., 2004	TS-alone (n = 15) Controls (n = 23)	18-65	Social cognition	Advanced Theory of Mind, Story Comprehension, Interpersonal Reactivity Index (experimental and self-report)	No comorbidity, unknown medication status	Patients with TS perform similarly to controls on tasks of social cognition.
Channon et al., 2012	TS-alone (n = 20) Controls (n = 20)	18-65	Social cognition	Emotional self-disclosure task with the Perceived Appropriateness of Disclosures Scale and the Faux Pas Task (experimental)	No comorbidity, mixed medication status	No group differences in self-disclosure. TS group was less accurate than controls on the Faux Pas Task.
Drury et al., 2012	Study 1: TS-alone (n = 16) TS+ADHD (n = 15) Controls (n = 27) Study 2: TS-alone (n = 23) Controls (n = 21)	Study 1: 10-18 Study 2: 18-65	Emotion recognition	Emotion perception in faces and voices (experimental)	No comorbidity in the TS sample, mixed medication status	No difference between children and adults with TS-alone and controls, except for subtle impairments for adults on conflicting emotional prosody. TS+ADHD performed poorer than controls on conflicting emotional prosody and voice-emotion naming.
Drury et al., 2016	TS-alone (n = 20) Controls (n = 20)	18-60	Emotion regulation	Berkeley Expressivity Questionnaire (BEQ), Emotion Regulation Questionnaire (ERQ), Cognitive Emotion Regulation Questionnaire (CERQ), Courtauld Emotional Control Scale (CECS) (self-report)	No comorbidity, mixed medication status	Higher scores of suppression for the TS group on the ERQ and the CECS. No difference between groups on reactivity or reappraisal.
Drury et al., 2018	TS-alone (n = 15) TS+ADHD (n = 13) Controls (n = 25)	10-17	Social cognition	The Sarcasm Comprehension Task (experimental)	No comorbidity in the TS sample, mixed medication status	TS-alone and TS+ADHD group showed mild impairments in comprehension of indirect sarcasm, but performed similarly to controls on direct sarcasm.

Table 1. (Continued)

Reference	Sample	Age [years]	Domain	Measure	Comorbidity/medication	Relevant findings
Drury et al., 2018	TS-alone (n = 15) TS+ADHD (n = 13) Controls (n = 25)	10-17	Social cognition	The Sarcasm Comprehension Task (experimental)	No comorbidity in the TS sample, mixed medication status	TS-alone and TS+ADHD groups showed mild impairments in comprehension of indirect sarcasm, but performed similarly to controls on direct sarcasm.
Eddy et al., 2010	TS (n = 16) Controls (n = 8)	TS M = 32 Controls M = 34	Social cognition	Theory of Mind Vignettes, Faux Pas Task (experimental)	Varying comorbidity in the TS sample, mixed medication status	No significant difference between groups on theory of mind. Patients with TS performed poorer than controls in recognizing faux pas.
Eddy et al., 2010	TS (n = 18) Controls (n = 10)	17-54	Social cognition	Pragmatic Story Comprehension Task, The Hinting Task, Faux Pas Task (experimental)	Varying comorbidity in the TS sample, mixed medication status	Patients with TS were impaired in detection of sarcasm and performance on the Faux Pas Task.
Eddy et al., 2011	TS (n = 18) Controls (n = 20)	16-47	Social cognition	Socially competitive emotions task (SCET), Ultimatum Game, Humorous cartoons, Reading the Mind in the Eyes (experimental)	Comorbid OCD, mixed medication status	Patients with TS were impaired on all tasks of theory of mind.
Eddy et al., 2017	TS-alone (n = 25) Controls (n = 25)	17-59	Social cognition	The Eyes Task (experimental)	No comorbidity, mixed medication status	No difference between groups.
Hovik et al., 2015	TS (n = 19) ADHD (n = 23) Controls (n = 50)	M= 12 (for all groups)	Emotional control	Emotional Control Scale from the Behavior Rating Inventory of Executive Function (BRIEF) (parent-report)	Varying comorbidity in the TS sample, mixed medication status	Children with TS and children with ADHD displayed significantly higher scores on problems with emotional control than controls.
Mermillod et al., 2013	TS (n = 18) Controls (n = 18)	16-70	Emotion recognition	Emotional Facial Expressions (experimental)	All patients with TS were medicated, varying comorbidity	Patients with TS were impaired in recognition of anger, sadness, and surprise compared to controls. No difference between groups on disgust, fear, happy, and neutral.

Note. TS = Tourette syndrome. TS-alone = Tourette syndrome without comorbidity. ADHD = Attention-deficit/hyperactivity disorder. Age range was reported, where this was evident from the text. Alternatively, mean age (M) was stated.

tion, 2013). Tics are sudden, rapid, recurring, nonrhythmic movements or sounds (motor tics and vocal tics) that may change over time and wax and wane in frequency. Tics are classified as simple or complex with the former involving only one muscle group and the latter involving multiple muscle groups. Motor and simple tics often emerge before vocal and complex tics with an average onset of TS of around six years of age (Freeman et al., 2000; Robertson et al., 2015) and a peak in severity around the ages of 10-12 years (McNaught & Mink, 2011). A small group of individuals with TS present with coprophenomena which manifest as either verbal (coprolalia) or motor (copropraxia) socially inappropriate sounds, words, or gestures. Lifetime coprolalia has been reported in around 15% of individuals with TS, and copropraxia in around 5%; both typically occurring several years after first tic onset (Freeman et al., 2009). Coprophenomena are well-described in the literature and are not to be confused with explosive emotional outbursts.

Prevalence estimates of TS vary, but are believed to lie around 0.3-0.9% in the school-age population (Scharf et al., 2015) with a higher prevalence among boys than girls of at least 3:1 (Cavanna, 2018; Robertson, 2015b). It is well-established that TS is a genetically heterogenous disorder with a large heritable component (Robertson, Cavanna, & Eapen, 2015), although the etiological factors have not been fully understood. The evidence regarding the so-called PANDAS (Pediatric Autoimmune Neuropsychiatric Disorders Associated with Streptococcal infections) hypothesis of tic onset due to a group A β-hemolytic streptococcal (GAS) infection (Swedo et al., 1998) is inconclusive. Although children with TS appear to be more prone to GAS infections, these infections did not predict tic exacerbations prospectively (Martino et al., 2011). Others have implicated immune mechanisms more generally as potentially affecting the neural development (Martino, Zis, & Buttiglione, 2015). Finally, further environmental factors, such as maternal smoking and low birth weight, have been suggested as risk factors of TS (Hoekstra, Dietrich, Edwards, Elamin, & Martino, 2013).

Previous studies on cognitive abilities in individuals with TS have produced mixed findings, presumably due to a lack of control for comorbidity, but generally point to intact cognitive abilities in TS (Eddy, Rizzo, & Cavanna, 2009) or only slight impairments (Morand-Beaulieu et al., 2017), and a main effect of comorbid ADHD on potential cognitive impairment (Roessner, Becker, Banaschewski, & Rothenberger, 2007).

Traditionally, the view on the course of the disorder has been quite optimistic with reports of substantial improvements if not complete recovery from tics in as many as one third of individuals with TS (Bloch & Leckman, 2009). The idea of frequent remission, however, has not been confirmed in longitudinal studies, although the severity of tics appears to decrease significantly from young adulthood, independent of medical treatment (Pappert, Goetz, Louis, Blasucci, & Leurgans, 2003). One study examined videotaped tics in a sample of 31 individuals with TS when they were children (8-14 years) and once more when they were adults ($>$ 20 years) and found that 90% still had tics as adults, although some of these individuals reported to be tic free (Pappert et al., 2003). This discrepancy between self-report and objective measures may explain why other studies have reported tic remission with higher frequency. Interestingly, not only tic severity, but also patterns of co-occurring diagnosis appear to change over time. In a study of 100 individuals with TS-alone, TS + ADHD, and TS + ADHD + OCD (obsessive-compulsive disorder), the majority changed phenotype over ten years (Rizzo, Gulisano, Calì,

& Curatolo, 2012). Approximately half of the individuals with TS-alone continued with this diagnosis, whereas the other half developed a comorbid OCD. Remarkably, all participants with TS + ADHD except for one were ADHD free after ten years with their phenotype having changed to either TS-alone for the majority or TS + OCD. A similar result was found for the group with three diagnoses. Tics remained present for all 100 participants, although they reported to be in better control of their tics (Rizzo et al., 2012). Despite the natural reduction in severity of tics following adolescence, a general finding of studies of impairment in individuals with TS illustrate a marked reduction in quality of life and psychosocial outcome (Robertson, 2015a).

1.2.2 Premonitory urges

The degree of voluntariness in tics has been debated since tics were first described as forms of hysteria with poor will-power and lack of self-control in the days of Gilles de la Tourette (Ganos et al., 2013). Whereas tics were earlier perceived as involuntary movements or sounds, they are now widely accepted as partly voluntary and suppressible at least for shorter periods of time (Leckman, Bloch, Scahill, & King, 2006). The study of premonitory urges (PU's) is relatively new in the field of TS, and is helpful to the understanding of the semi-voluntary nature of tics. A PU is an unpleasant sensation preceding a tic (Woods, Piacentini, Himle, & Chang, 2005), which can be more unpleasant than the tics per se, and which constitute an interesting dissociation from other motor disorders such as stereotypies, dystonia, or tremor, in which PU's are not present (Robertson et al., 2015). PU's are present in about 90% of individuals with tics (Robertson et al., 2015) with examples being sensations of itching, tension, pressure, or "just right" phenomena (Woods et al., 2005), referring to the well-known feeling to many individuals with TS of having to achieve a certain sensory state that feels "just right" to them through motor or vocal activity. The occurrence of PU's appears to increase with age following the general cognitive development (Banaschewski, Woerner, & Rothenberger, 2003), with individuals becoming aware of these sensations around the age of 10 (Leckman et al., 2006; Robertson et al., 2015). In one of the first studies to examine PU's in 135 individuals with TS aged 8-71, 93% reported to feel PU's and 92% of patients characterized their tics as under complete or partial voluntary control. Eighty-four percent of participants reported a sense of relief following execution of their tic(s) (Leckman, Walker, & Cohen, 1993). In sum, PU's aid in the understanding of tics as a voluntary capitulation to an involuntary urge (Cavanna & Nani, 2013).

1.2.3 The neural basis of TS

Imbalances in cortico-striato-thalamo-cortical (CSTC) circuits connecting the cerebral cortex with subcortical regions have constituted the framework for explaining the neurobiological mechanisms of TS and is still the leading hypothesis for tics (Conceição, Dias, Farinha, & Maia, 2017; Felling & Singer, 2011; Greene, Black, & Schlaggar, 2013). In brief, the basal ganglia has a central role in models of movement disorders and tic generation (Ganos et al., 2013) with some variation in the specific mechanisms. One widely accepted neuroanatomical model of TS has proposed an imbalance of the direct versus indirect pathway within these CSTC's, which normally cooperate in the relative selection and inhibition of appropriate responses (Greene et al., 2013). In the direct pathway, an inhibitory signal is sent from the striatum to the internal part of the globus pallidus, resulting in a disinhibition of the thalamus and ultimately an excitatory

effect on the prefrontal cortex (PFC) leading to movements (tics). In the indirect pathway, the signal is sent from the striatum to the external part of the globus pallidus and the subthalamic nucleus, which conversely increases inhibition of the thalamus and thus decreases excitation of the PFC (Felling & Singer, 2011; Leckman & Bloch, 2015). Structurally, the basal ganglia have consistently been implicated in TS, with reductions in volume of the caudate nucleus in both children and adults (Greene et al., 2013; Peterson et al., 2003). Other structures, such as the prefrontal lobes, the amygdala, and the hippocampus, most likely play a part in TS as well (Ganos et al., 2013). Despite numerous studies on the structural and functional neurobiology of tics and TS, differing results have not produced one accepted and reproducible model of TS, and regardless of the relative popularity of the CSTC model, TS most likely results from a complicated interplay of minor impairments at many different levels (Ganos et al., 2013). Biochemically, TS is believed to be associated with imbalances in primarily the dopaminergic system, with findings of an increased number of dopamine receptors in striatal and cortical regions in TS creating a hyper-dopaminergic state (Bloch, State, & Pittenger, 2011), but most likely deviances appear in a wide range of systems (Roessner et al., 2011).

1.2.4 Treatment

The primary therapeutic approach for TS is psychoeducation (Robertson, 2015a), which entails information regarding tics, course of the disorder, possible comorbidity, and treatment, in the hope that information about the disorder can ameliorate family stress. A vast number of individuals with TS do not need further treatment (Roessner et al., 2011), but for those who experience functional impairment in relation to their tics, the next step is behavioral intervention (Robertson, 2015a; Roessner et al., 2011; Verdellen, van de Griendt, Hartmann, Murphy, & the ESSTS Guidelines Group, 2011). A number of behavioral interventions exist that have shown some promise, specifically habit reversal training and exposure and response prevention (Verdellen et al., 2011). These therapeutic approaches have proved effective in the reduction of tics and can be used interchangeably if one type does not have an effect. They do, however, require a substantial work effort from the child and an awareness of PU's, and are thus not always suitable for younger children (Verdellen et al., 2011). As a final option when tics continue to cause subjective discomfort in the shape of pain, injury, social, emotional, or functional problems, a pharmacological approach can be beneficial (Roessner et al., 2011). Due to the hypothesis of an imbalance in the dopaminergic system, the pharmacological approach has primarily targeted the dopaminergic neurotransmission using antipsychotic medicine (Quezada & Coffman, 2018; Roessner et al., 2011). Other types of medication, particularly the α_2 agonist, clonidine, is increasingly being used to avoid the side effects of dopamine blockade (Quezada & Coffman, 2018). However, actual evidence-based guidelines are difficult to formulate due to the natural of course of tics waxing and waning and changing in type, which complicates the evaluation of treatment, and due to a lack of randomized controlled trials (Roessner et al., 2011).

1.2.5 Clinical heterogeneity

A challenge for treatment is the extensive comorbidity associated with TS. Only 10% of individuals with TS have so called pure TS or TS-alone without any co-existing conditions (Robertson et al., 2015). The vast majority thus presents with comorbid psychiatric disorders with ADHD being the most common, followed by OCD. Anxiety, mood disorders, and disruptive behaviors

are seen frequently as well (Hirschtritt et al., 2015). Approximately 70% of individuals with TS are estimated to fulfill criteria for either ADHD, OCD, or both, and there is a tendency for females to present more frequently with comorbid OCD and males with ADHD (Hirschtritt et al., 2015). The co-occurrence of psychopathology other than TS influences practices for symptom management, as symptoms other than tics frequently dominate the clinical presentation (Robertson et al., 2015). Specifically, comorbid ADHD has been associated with more psychosocial problems and maladaptive behaviors than in individuals with TS-alone (Hirschtritt et al., 2015).

1.3 Attention-deficit/hyperactivity disorder

The fact that ADHD is the most frequent comorbid disorder in individuals with TS, and is also associated with emotional dysregulation (Graziano & Garcia, 2016), made this group a particularly interesting subject for comparison. The diagnostic criteria for ADHD are largely the same for DSM-5 and ICD-10, describing a persistent pattern of inattention and/or hyperactivity/impulsivity that interferes with functioning and lasts for more than six months, with significant impairment in more than one context, e.g., in school and at home (American Psychiatric Association, 2013; World Health Organization, 2016). Previously, the two classification systems had seven years as the required age of onset, but following the update from DSM-IV to DSM-5, this criterion has been changed to age of onset before age 12. Finally, the ICD-10 labels the disorder as a hyperkinetic disturbance. The ICD-10 mentions that symptoms are frequently isolated to either inattention or hyperactivity/impulsivity, whereas the DSM-5 has specific subgroupings of predominantly inattentive, predominantly hyperactive/impulsive, or combined subtype. Prevalence estimates of ADHD vary greatly, but are believed to lie around 5% in children (Leckman & Bloch, 2015; Sayal, Prasad, Daley, Ford, & Coghill, 2018) and 2.5% in adults (Simon, Czobor, Bálint, Mészáros, & Bitter, 2009). More boys than girls have ADHD with a ratio of 2-3:1 (Sayal et al., 2018).

ADHD is a highly heritable disorder with symptoms emerging in early childhood in a interplay between genes and environment (Tarver, Daley, & Sayal, 2014). However, no specific genes have been identified as either necessary or sufficient to the development of ADHD. A number of structural changes in the brain have been associated with ADHD, particularly an overall reduction in brain size and reduced thickness of the cortex (Leckman & Bloch, 2015), which are likely to be genetically based (Tarver et al., 2014). Treatment of ADHD consists of either behavioral treatment, dopamine agonists, or a combination of the two (Sharma & Couture, 2014), but also psychoeducation can have a positive effect on coping with the disorder (Leckman & Bloch, 2015).

1.4 Emotion regulation

The scientific interest in ER increased drastically in the mid-nineties and has evolved from theoretical studies of the concept to focusing on the specific role of ER in models of psychopathology (Aldao, Nolen-Hoeksema, & Schweizer, 2010). The following sections will provide an overview of the conceptualization of ER that is employed in the present thesis, the developmental trajectory and neural basis of ER, and the link between ER and disruptive behavior.

1.4.1 A model of emotion regulation

ER can occur consciously or unconsciously on a continuum and represents the process of minimizing, intensifying, or maintaining an emotion in the context of subjective goals (Gross, 2014). Conversely, emotion dysregulation represents a deficit in the ability to adaptively modify these responses (Thompson, 1994). Various models of ER exist, but for the present thesis we will use the widely recognized process model of emotion regulation (Gross, 2014) as the theoretical framework for understanding ER. In order to discuss the regulation of emotions, it is central to first conceptualize the target of regulation. Emotions arise when an individual appraises a situation as relevant to a conscious or unconscious goal, emotions influence the subjective experience of a situation as well as behavior and physiology, and they may lead to action (Gross, 2014). Importantly, an emotion should not always be subject to regulation. The experience of strong emotions is not maladaptive in itself and often emotions serve as indicators of action. At other times, however, emotions lead to maladaptive behaviors or subjective experiences that are in need of regulation.

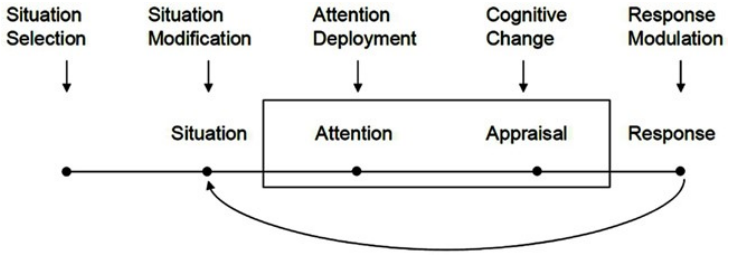

Figure 2. The process model of emotion regulation, (Gross, 2014, p. 7).

The process model of emotion regulation (Figure 2) builds on a previous model (the modal modal of emotion) by illustrating five groups of processes related to ER, and how they target emotion-generative processes (Gross, 2014). The first four groups of processes are antecendent-focused, meaning that they center around the input of the emotional system, whereas the final group is response-focused, referring to a role in the output of the emotional system (Gross, 1998). The first type of ER process is situation selection, which occurs before a stimulus can affect emotion generation. Individuals may select certain situations that are likely to generate specific emotional outcomes, such as choosing to attend a party to have a good time or to avoid that same party as part of social anxiety. In the next stage, actions can be taken to modify one's situation to consequently modify associated emotions. The third antecedent-focused process refers to the impact of attention on emotional experiences. Distraction is a classic example of redirecting attention away from something unpleasant, but it may also go in the opposite direction as in rumination central to depression, in which the attention on worrisome stimuli is excessive. The fourth group of ER processes is cognitive change, which has to do with the subjective evaluation of an emotional stimulus which determines the experience of the emotion (Gross, 2014). Cognitive reappraisal belongs to this category. Reappraisal refers to reinterpreting the meaning of emotional stimuli to change their impact on the subjective experience (Ochsner & Gross, 2008). This ability has been shown to be age-dependent (Silvers et al., 2012) and related to brain areas associated with cognitive control, thus reliant on the maturation of the PFC (McRae, 2016). The connection between reappraisal and cognitive abilities is further

strengthened by findings showing that the ability to reappraise is reduced in the context of competing cognitive demands (McRae, Ciesielski, & Gross, 2012). Interestingly, distraction can prove more effective than reappraisal if the stimulus is very intense or if there is not enough time to reappraise adequately. Distraction has also been shown to reduce activity in amygdala more than reappraisal; however, this effect is not as long-lasting as that of reappraisal (McRae, 2016). Response modulation is the final process of the process model and the only one to be response-focused. Response modulation encompasses experiential, behavioral, and physiological responses to an emotion which in turn will restart the process of regulation by influencing the situation (Gross & Jazaieri, 2014). Suppression is one type of strategy associated with this process. In suppression, the individual makes an effort to eliminate all bodily expressions of an emotion (expressive suppression; McRae, 2016) or simply to suppress the emotion by ignoring its existence. This strategy does not reduce negative affect and appears to increase activity in the amygdala (Gross, 2014). The process model of emotion regulation constitutes the current conceptualization of ER and will be referred to continuously throughout the thesis and the three papers.

1.4.2 The developmental trajectory of emotion regulation

The developmental trajectory of ER is under the simultaneous influence of trajectories of biological, behavioral, social, cognitive, and affective processes throughout life (Diamond & Aspinwall, 2003). The first type of ER to develop is the dyadic contact with the primary caregiver (Stringaris, 2015). Infants and small children mostly rely on interactions with their primary caregiver(s) for ER, and as the child grows older, these processes of regulation are internalized (Diamond & Aspinwall, 2003). Other relations such as that with peers will gain precedence during later childhood and adolescence; periods characterized by impulsivity and risk behaviors which are possibly the result of an imbalance in the maturation of subcortical versus prefrontal structures of the brain, with the former maturing earlier than the latter (Stringaris, 2015). Children down to the age of three are able to employ the strategy of cognitive reappraisal (Hua, Han, & Zhou, 2015; Mischel & Baker, 1975), and the frequency and efficacy of spontaneous use of regulatory strategies increase with age (Silvers et al., 2012). The development of ER does not stop at a fixed age, but is continuously advanced through experiences and interactions throughout life as a constant subject for optimization (Diamond & Aspinwall, 2003).

1.4.3 The neural basis of emotion regulation and dysregulation

The neural circuitry underlying emotions is constituted by dorsolateral and ventral regions of the PFC (including the anterior cingulate cortex (ACC)), amygdala, hippocampus, and insula (Davidson, Fox, & Kalin, 2001), with the PFC and the amygdala most consistently implicated in emotion regulation. The PFC is involved in cognitive control, but also plays a part in affective processes. Specifically, the PFC monitors competing stimuli, focusing on a specific goal, by inhibiting or activating other brain areas accordingly. Additionally, the PFC manages the balance between internal stimulus-independent thought and external stimulus-dependent input, which is part of ER (Davidson et al., 2001). Interestingly, the specific area keeping track of internal and external stimuli, the most rostral part of the PFC which is believed to play a part in ER, is the area of the PFC which shows the greatest relative increase in size compared to the rest of the brain across primate species with the largest relative size in humans

(Davidson et al., 2001). These parts of the PFC along with the medial PFC and the lateral orbitofrontal cortex are reciprocally connected with another relevant structure, the amygdala, which is involved in emotion generation and expression (Stringaris, 2015). Amygdala further directs attention towards affectively salient stimuli relevant to the individual, by recruiting cortical arousal and attention (Davidson et al., 2001). Neural activation is dictated by the chosen ER strategy. Areas of the PFC are activated during reappraisal, in which prefrontal and cingulate regions exert inhibitory control over the insula and the amygdala (Stringaris, 2015). As an interesting dissociation, the neural activity associated with suppression leads to increased activity in the amygdala (Ochsner & Gross, 2008). In a balance between cognitive and behavioral processes, it is sometimes adaptive for emotions to interrupt behavior and at other times emotions need to be managed properly. Possibly, impaired ER is associated with a dysfunction in these neural circuits, but it is unclear whether specific patterns of dysfunction contribute to specific maladaptive ER (Johnstone & Walter, 2015).

1.4.4 The link between emotion regulation and disruptive behavior

The focus of the present thesis is on the experimental expression of ER as a measurable component, potentially underlying disruptive behavior. Although it is highly relevant to establish the prevalence of rage attacks or explosive behavior on a clinical, behavioral level by categorically investigating the number of children fulfilling criteria for a disruptive behavior disorder or quantifying the number of explosive outbursts, our focus was on the dimensional capacity of ER on an experimental level, under the premise that general ER abilities are associated with the actual clinical presentation. The link between ER and disruptive behavior has been addressed in previous research. On a neural level, disturbances in the circuits described in the previous section are associated with both internalizing and externalizing psychopathology in adults, and on a clinical level, aggression can be conceptualized as a disturbance of ER (Lewis, Granic, & Lamm, 2006). A distinction is often made between reactive and proactive aggression. Reactive aggression is triggered by an external provocation and has shown associations with ER. Proactive aggression reflects willful, instrumental aggression and has been linked with more stable personality traits such as calleous-unemotional traits in childhood and adolescence, and psychopathic traits and adulthood (Ahmed, Bittencourt-Hewitt, & Sebastian, 2015). The concept of disruptive behavior encompasses several differing types of behavior. The merging of oppositional-defiant disorder (ODD) and conduct disorder (CD) as disruptive behavior disorders is gaining ground, but disruptive behavior can also be expressed dimensionally as anger, aggression, and general noncompliance in the absence of any psychiatric diagnosis. Explosive outbursts can be equated with rage attacks as further reflections of disruptive behavior. While explosive outbursts represent a normal part of development, these outbursts usually come to an end as the child matures (Piasecka, Bertschinger, Tudor, & Sukhodolsky, 2018). Sometimes, however, deficits in the ability to regulate emotions manifest as maladaptive aggression, which may be associated with long-term poor psychosocial functioning and substance abuse (Lewis et al., 2006).

1.5 Emotion regulation in psychopathology

Adaptive ER is pivotal to emotional health and mental well-being, and inappropriate emotional responses have for some time been implicated in various psychiatric disorders (Tamir & Mauss, 2011). People respond differently to emotional challenges with great variation in individual ER styles (McRae, Misra, Prasad, Pereira, & Gross, 2012). Some of these differences are hypothesized to be visible across groups, such as a general reliance on maladaptive strategies like rumination and suppression in depression (Aldao et al., 2010) or difficulties managing anger in ADHD (Bunford, Evans, & Wymbs, 2015). Importantly, maladaptive regulation depends on the individual's goal of ER. Two persons with anxiety may have opposite goals of avoiding the fear-inducing stimulus or exposing oneself to the stimulus to increase tolerance, respectively, which will result in widely differing responses (Fernandez, Jazaieri, & Gross, 2016). Thus, ER is not merely a process of down-regulating negative emotions, but must always be viewed in light of subjective goals, which determine the success of the process. The following sections will look specifically into the role or ER in TS and ADHD. Due to the relatively well-established relationship between ADHD and dysregulated behavior, we first address this connection before turning to the association between ER and TS.

1.5.1 Emotion regulation in ADHD

Reviews of the literature on emotion dysregulation in ADHD have established that impaired ER is a central part of ADHD that causes significant impairment in these children (Graziano & Garcia, 2016; Shaw, Stringaris, Nigg, & Leibenluft, 2014). Estimates of the prevalence of dysregulated emotional behavior in ADHD lie around 25-40% of children and 30-70% of adults (Shaw et al., 2014). Emotional dysregulation is a heterogeneous feature in ADHD (Tarver et al., 2014), which may be affected by cognitive deficits, but is not completely explained by executive and cognitive impairment (Shaw et al., 2014). One study suggested that the respective presence or absence of the impairments associated with emotional dysregulation may differentially predict treatment response (Bunford et al., 2015) with the same group noting that current treatment options do not sufficiently ameliorate problems with social functioning (Bunford, Evans, & Langberg, 2018).

1.5.2 Emotion regulation in TS

A broad estimate suggests that disruptive behavior in the form of explosive outbursts is present in 25-70% of children with TS (Chen et al., 2013) and represents the greatest source of impairment (Leclerc, O'Connor, Forget, & Lavoie, 2011). Longitudinally, emotional problems have been shown to persist into adulthood even in the absence of tics (Gorman et al., 2010; Hoekstra et al., 2012) pointing to a need for a further understanding of the emotional component in TS.

Many parents of children with TS believe that their children's rage attacks are part of their diagnosis of TS and are often less concerned with reducing tics than with their child's disruptive behavior. As illustrated in the systematic search described in the beginning of this thesis, some studies indicate mild impairments within aspects of social cognition, but a general impairment in ER has not been studied experimentally and has been the subject of debate regarding its diagnostic affiliation. In Gilles de la Tourette's original records, behavior mimicking what we

conceptualize as explosive outbursts today was described for some of his cases, referring for example to "ferocious fits of rage" (Cath & Ludolph, 2013, p. 92). Qualitatively, explosive outbursts and rage attacks can be used interchangeably in TS and manifest as sudden episodes of intense aggression (verbal or physical) which is disproportionate to the triggering stimulus. The child often reports a sense of loss of control, and the behavior is uncharacteristic to the child (Piasecka et al., 2018). Recalling the distinction between reactive and proactive aggression, it is the reactive aggression that has been associated with TS. Phenomenologically, reactive rage attacks can resemble tics in regard to premonitory urges, a sense of relief, and a lack of complete control (Budman, Bruun, Park, Lesser, & Olson, 2000). The apparent phenomenological similarities have led some to classify rage attacks as a subtype of tics (Leclerc et al., 2011). Simultaneously, a number of factor analytic studies of large cohorts of individuals with TS have indicated an aggressive phenotype in TS (Robertson et al., 2015). A number of studies controlling for the presence of ADHD, however, do not show any differences between individuals with TS and control groups regarding presence of explosive outbursts, and find the prevalence of these to be dependent on comorbidity with ADHD (Budman et al., 2000; Chen et al., 2013; Robertson et al., 2015; Stephens & Sandor, 1999). These consistent findings suggest that, on a strictly behavioral level, children with TS-alone are not more prone to rage attacks than children without a psychiatric disorder and indicate once again that ADHD appears to be responsible for the vulnerability to emotion dysregulation.

Hence, current views fairly clearly attribute a co-existing disorder of ADHD as decisive to the presence or absence of disruptive behavior in TS. The pronounced rates of comorbidity in TS other than ADHD, however, pose a challenge to the investigation of disorder specific impairments and indicate that disruptive behavior in TS is caused by a multifactorial etiology.

1.6 How to measure emotion regulation?

Several researchers have highlighted the need for new experimental paradigms of ER to increase options for investigating the concept and aid in the translation from clinical observations to empirical studies (Gross, 2014; Stringaris, 2015; Tamir & Mauss, 2011). An inherent challenge in the measurement of ER, however, lies in the close coupling with other emotional processes, such as emotion generation and emotional reactivity; the latter referring to the required threshold for a stimulus to create a response in an individual (Carthy, Horesh, Apter, Edge, & Gross, 2010). The fluid boundaries between concepts make it difficult to establish whether we are measuring behavior caused by an intense emotion or a lack of ability to regulate that emotion.

In March 2013, we searched the literature for existing tests of emotion regulation across diagnostic groups and outside of child psychiatric settings and identified several questionnaires and a few experimental tests. Broadly speaking, there are four approaches to examining ER: 1) The clinician or researcher asks an individual to answer a number of questions about their ER ability (self-report), 2) the clinician/researcher asks the parents of a child to evaluate the child's ER abilities (parent-report), 3) the clinician/researcher makes his or her own observations, preferably based on standardized, experimental measures (clinician-rated), and 4) the clinician/researcher employs objective measures of physiological responses such as heart rate variability (HRV) or galvanic skin response (physiological measures). We will briefly touch upon

these different types of measurement in the following.

1.6.1 Questionnaires

Following the interest in the study of ER, several questionnaires for children and adults have been developed that target various aspects of the ER continuum through self- or parent-report (Aldao et al., 2010). Questionnaires are widely used in the literature and are easy to administer. However, there are disadvantages associated with using this approach. Children are not always optimal self-reporters and thus studies of ER in children using questionnaire-based measures often rely on parent accounts of child behavior. While there is plenty of information to gain from parent reports, these typically cover the past six months, potentially missing nuances of behavior. Furthermore, parents' own possible psychopathology and interpretation of their child's behavior are also likely to affect their answers. Further, it may be in the interest of some parents to either exaggerate or understate symptoms or impairment to obtain help and support or to give the impression that there are no problems. In cases in which the child is old enough to self-report, additional challenges arise. The ability to accurately report on one's own emotional states requires proficient metacognitive abilities and is highly likely to be influenced by current moods (Aldao et al., 2010). In general, any self-reported information on ER strategies should be interpreted with caution (Robinson & Clore, 2002) and would benefit from the inclusion of additional measures.

1.6.2 Behavioural measures

Behavioral tasks are ideal to overcome potential bias related to self- or parent-report, although these measures are far from perfect themselves. One problem associated with the use of experimental, observational tasks is the heterogeneity in design and outcome variables, which complicates cross-study comparisons (Aldao et al., 2010). Another problem is the lack of ecological validity of tests, which hinders the transfer of behavioral paradigms to everyday situations (Fogleman, Leaberry, Rosen, Walerius, & Slaughter, 2018) and calls for methods of assessment that focus on spontaneous ER in ecologically valid settings (Aldao et al., 2010). Rage attacks have been documented to occur with the highest frequency in the child's home, which emphasizes the need for tests that capture the interpersonal component of ER (Budman et al., 2000).

The basic premise of most experimental tests of ER is artificially induced frustration with the hope that any elicited behavior will resemble the behavior displayed in real-life situations. These types of tests have primarily been used with younger children, such as having an attractive toy in a transparent box outside the child's reach or inducing disappointment with undesirable presents (Jahromi, Meek, & Ober-Reynolds, 2012; Stifter, Dollar, & Cipriano, 2010). The most frequently used frustration tests are variants of unsolvable puzzle tasks. The idea of these types of tasks is to have the child try to solve a puzzle with one or more pieces missing or switched with similarly looking pieces and subsequently code behavior. Different versions of the task have used regular puzzles with cartoon images with pieces missing (Day & Burns, 2011; Farmer & Garfield, 1971; Jahromi, Gulsrud, & Kasari, 2008; Jahromi et al., 2012; Smiley & Dweck, n.d.), solving a puzzle blind-folded (Scime & Norvilitis, 2006), Lego bricks or blocks with pieces missing (Gentzler, Wheat, Palmer, & Burwell, 2013; Melnick & Hinshaw, 2000), or the Tangram puzzle (Hudson & Rapee, 2001, 2002). The latter is difficult, but not unsolvable, and has also

been used in the work presented in this thesis, but with a novel method of coding. Similar to the design of these tests, outcome measures vary accordingly. The majority of participants taking these tests have been very young children, and in cases in which older children have been included, these have for the most part had developmental challenges such as learning disabilities, Down syndrome, mental retardation, or autism.

A few experimental tasks exist in addition to frustration tasks. The Mayer-Salovey-Caruso Emotional Intelligence Test (MSCEIT) is widely used as a test of emotional intelligence, but the required age is 17 years (Mayer, Salovey, & Caruso, 2002). Other methods have focused on specific strategies within the context of ER such as reappraisal, suppression, or rumination and how these strategies influence emotions (Aldao et al., 2010), and have employed a combination of self-report and clinician rating in their measures. Such studies are important to the understanding of these specific strategies, but do not elucidate participants' natural tendency to apply these strategies and have not previously been related to psychopathology (Aldao et al., 2010).

1.6.3 Physiological measures

Although we did not include physiological measures in the present work, it is relevant to briefly comment on these methods. Various measures have been included in a great number of studies of physiological measures of ER (Fernandez et al., 2016). These techniques serve as objective components that are not reliant on human evaluation. Multiple possible methods exist, covering among others functional magnetic resonance imaging (fMRI), HRV, and cortisol levels. An advantage associated with these measures is measurement precision (Bunford et al., 2015), but similarly to experimental measures, the physiological ones do not clarify what is being measured, i.e., whether a physiological response is caused by a powerful emotion or a lack of regulation.

2. Objectives and hypotheses

The overarching aim of the work presented in this thesis was to shed light on the phenomenology of ER in children with TS to build a bridge between clinical observations and experimental studies to ultimately benefit assessment and intervention. Specifically, we examined whether emotional dysregulation is an inherent part of TS, or whether it is associated with comorbid psychopathology. While an extensive body of literature has investigated the cognitive and executive functioning in children with TS, their ability to regulate emotions has not been tested experimentally and the existing knowledge is thus built on data obtained through self- or parent-report. Due to the lack of suitable tests of ER, the initial objective became the development of an experimental task for testing the capacity of ER in children (paper 1). Second in chronology, but not relevance, was the objective of assessing children with TS with and without comorbid ADHD on their ability to adaptively regulate their emotions by applying the newly developed manual for coding ER (paper 2). Finally, we wished to assess ER through a multi-method approach by using a second experimental test of a specific type of ER, cognitive reappraisal (paper 3). This latter type of ER differs from the unguided ER examined in paper 2, thus allowing for a nuanced characterization. As this work represents a first step towards better understanding the nature of emotion regulation in TS, the direct implications for treatment lie further ahead. We believe it to be an important contribution, however, to increase our comprehension of the diagnosis-specific and non-specific emotional processes in child psychopathology to ultimately benefit options for treatment. Aims and hypothesis for the individual papers are specified below.

2.1 Paper 1: The Puzzle of Emotion Regulation - Development and Evaluation of the Tangram Emotion Coding Manual for Children

Having searched the field across diagnoses for objective measures of ER prior to beginning the study, we discovered a significant dearth in experimental tests. To overcome this shortage in methods of assessing ER that were not reliant on self- or parent-report, we developed an instrument offering a novel approach to examining ER through clinician-rated evaluation of child behavior. In this work, we wanted to take into account recent requests for more ecologically valid tests incorporating the parent-child dyad, which often constitutes the frame for dysregulated

behavior. Thus, the aim of the first paper was to describe the development of the manual and to examine its psychometric properties, comprising interrater and intrarater reliability, convergent validity, discriminant validity, content validity, and the instrument's ability to delineate a group of children with ADHD from a group of typically developing controls. We hypothesized that the primary outcome of the test, an overall measure of ER, would show convergent validity in the medium range with two parent-report questionnaires on child emotional control and child deficient emotional self-regulation, respectively, and a small to medium correlation with IQ as a representation of discriminant validity. We hypothesized that children with ADHD would present with poorer ER ability than the control group.

2.2 Paper 2: Emotion Regulation in Children with Tourette Syndrome – Associations with ADHD and Tic Severity

As ER has not been studied experimentally in children with TS before, the aim of the second paper was to investigate ER profiles in groups of children with TS and ADHD compared with typically developing controls to explore the specific associations between dysregulated behavior and psycho¬pathology typical for TS and ADHD. In continuation of our previous validation of the instrument for assessing ER, we conducted an exploratory factor analysis to investigate the latent structure of items. Finally, we explored associations between ER ability and symptoms related to both TS (tic severity and premonitory urges) and ADHD (severity of inattention and hyperactivity/impulsivity). We hypothesized a staircase distribution of ER ability with the control group presenting with the highest scores of ER ability, followed closely by the TS group, and ultimately the groups including children with ADHD.

2.3 Paper 3: Reappraisal is an Effective Emotion Regulation Strategy in Children with Tourette Syndrome and ADHD

The aim of the third paper was to investigate group differences in a specific ER strategy (cognitive reappraisal) when children were explicitly trained in this strategy. We additionally examined emotional reactivity, and finally, in an explorative manner, the efficacy of specific strategies within the frame of reappraisal. We hypothesized an effect of employing the strategy of cognitive reappraisal for all groups, with the highest regulation success for the control group and the TS group, partly due to the cognitive component in the task. We expected children with ADHD to achieve the lowest scores, and the reverse pattern for reactivity, with children with ADHD presenting with the highest reactivity scores.

3. Methods

In the present cross-sectional case-control study we investigated ER in children with TS. Due to the seemingly impairing effect of comorbid ADHD on individuals with TS, we included clinical contrast groups of children with ADHD and children with TS + ADHD, respectively, to be able to systematically investigate whether difficulties in ER, as measured with two experimental tasks, were specific to one diagnostic group or a general factor in developmental neuropsychiatry (Aldao, 2016). Recruitment procedures, in- and exclusion criteria, characteristics of the total participant sample, and descriptions of the applied measures follow below.

3.1 Recruitment

Children were recruited in the period from 2013 to 2016 (Figure 3) with different recruitment procedures for children with TS, ADHD, and controls, respectively, as described below. For the comparison between the primary group (children with TS) and the control group (typically developing children), we aimed to be able to detect at least medium sized possible differences. As no prior studies could guide the selection of an appropriate sample size, we set an a priori alpha value of .05 and assumed power of .80, which required a sample size of minimum 51 in each of these two groups as calculated with the statistical power analysis program, G*Power (Faul, Erdfelder, Lang, & Buchner, 2007).

3.1.1 Children with TS

We recruited participants with TS in collaboration with a pediatric clinic at Herlev University Hospital in the Capital Region of Denmark with nationwide responsibility for the care and management of children with TS. Parents of children eligible for participation were provided with oral and written information material about the study at the clinic and were afterwards contacted via the phone. In case a family wished to participate, they would be invited to two days of assessment at the Child and Adolescent Mental Health Center (CAMCH), Mental Health Services, also in the Capital Region of Denmark.

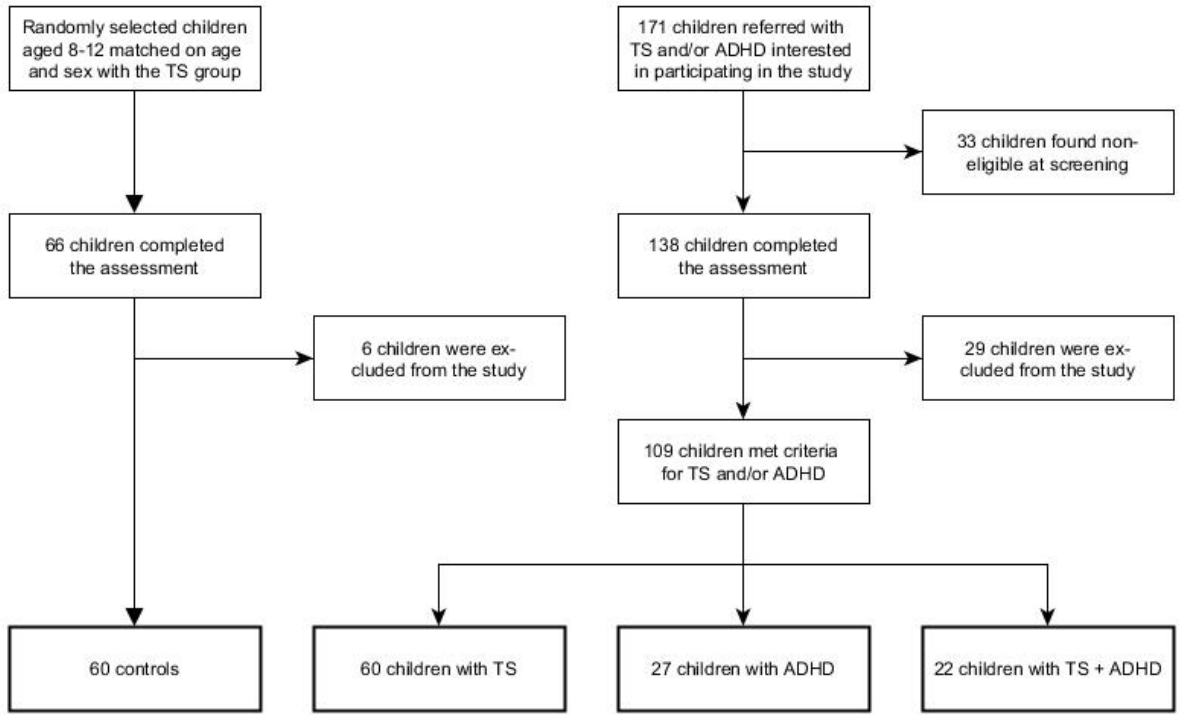

Figure 3. Flowchart of the recruitment of participants. TS = Tourette syndrome. ADHD = Attention-deficit/hyperactivity disorder. Six potential control participants were excluded due to meeting criteria for anorexia nervosa (n = 1) or TS (n = 2), presence of tics (n = 2), or age (n = 1). Thirty-three potential TS/ADHD participants were deemed not eligible at the initial screening, primarily due to current or prior use of psychotropic medication or an established diagnosis of autism spectrum disorder. Twenty-nine children were excluded from the participant sample following complete assessment due to age > 12 (n = 3), IQ below 80 (n = 5), severe comorbidity (n = 7), premature birth (n = 1), tics not fulfilling criteria for TS (n = 10), neurological conditions (n = 2), and one participant meeting only criteria for ODD.

3.1.2 Children with ADHD

Participants with ADHD were recruited from the local child and adolescent psychiatric outpatient clinic at CAMCH. When a child was referred for diagnostic assessment with a provisional diagnosis of ADHD and before beginning treatment, a member of the research team would participate in the initial consultation to evaluate eligibility, provide oral information about the study, and hand out information material. After minimum one week, the potential participant would be contacted via the phone. If a family did not wish to participate or met one or more exclusion criteria, the child would follow standard assessment procedures. If a family wished to participate, the child would be allocated to the research team with procedures comprising the standard assessment along with several other measures.

3.1.3 Children from the control group

We recruited children for the control group via the Danish Civil Registration System, which contains information on all individuals with a permanent residence in Denmark (Pedersen, 2011). An extract was randomly generated with children from the Capital Region of Denmark aged 8-12 with approximately 10 potential participants for each child with TS and a male-female ratio of 3.5:1. The control group was matched individually on age and sex with the TS group with up to four months of difference in age being accepted. Parents of potential participants received information material through hard copy letters and were subsequently contacted via the phone after two weeks, allowing for clarification and/or questions. Participants from the control group were recruited continuously, mirroring the recruitment of participants with TS.

3.2 Inclusion and exclusion criteria

Inclusion and exclusion criteria were similar for children in the clinical groups and the control groups with few exceptions regarding diagnostic status. Inclusion criteria for all participants were age between eight and 12 years (both included) and informed consent from the legal caregiver(s). Participants in the clinical groups additionally had to be referred for psychiatric assessment with a working diagnosis of either TS or ADHD according to the ICD-10 (World Health Organization, 2016). Exclusion criteria for all participants were birth at gestational age < 37 weeks, presence of any lifetime neurological condition, IQ below 80, and current or prior use of psychotropic medication, as any effects on cognitive and emotional processes stemming from the use of medication may prove difficult to disentangle from specific characteristics of the diagnostic entities. For children in the clinical groups, additional exclusion criteria were severe lifetime psychiatric comorbidity (autism spectrum disorder and psychotic disorders) and presence of tics not qualifying for a diagnosis of TS. As the shared genetic background and pathophysiology for different types of tic disorders have not been fully established, we preferred to be consistent in only including children meeting with diagnostic criteria for TS. For children in the control group, the presence of any psychiatric disorder according to ICD-10 criteria would lead to exclusion, with the exception of enuresis and encopresis when not associated with any other type of psychopathology.

3.3 Participants

The final participant sample consisted of 169 medication-naïve children living in the Capital Region of Denmark, aged 8-12 years. The TS group comprised 60 children, the ADHD group 27 children, the TS + ADHD group 22 children, and finally the control group comprised 60 children matched on age and sex with the TS group (Table 2). One child was recruited for participation in the control group, but was found to meet criteria for TS and was thus included in the TS group. The work described in all three papers was carried out with this participant sample with some exceptions. For paper 2, the employed task was implemented later than other existing measures, and we thus lack data from the first approximately 35 participants. For paper 3, we were missing data files or video material for nine participants. Finally, we included an additional 15 seven-year-old participants from a Danish high risk study (Thorup et al., 2015) in papers 1 and 2. Of these children, 13 were part of the control group, and two children were included in

Table 2. Participant characteristics for the total sample.

Characteristic	TS (n = 60)	ADHD (n = 27)	TS + ADHD (n = 22)	Controls (n = 60)	*p*
Mean age, years (SD)	9.97 (1.3)	9.60 (1.1)	9.50 (1.3)	9.90 (1.3)	0.366
Male, n (%)	49 (81.7)	19 (70.4)	17 (77.3)	49 (79.0)	0.620
Mean IQ (SD)	102.6 (9.9)	94.4 (8.5)	98.8 (10.9)	103.9 (11.2)	0.001
Mean SES (SD)	5.8 (1.4)	4.3 (1.7)	4.8 (1.7)	5.9 (1.4)	< 0.001
Comorbidity, n					
CD (%)	0 (0)	1 (3.7)	1 (4.5)	0 (0)	0.200
ODD (%)	4 (6.7)	6 (22.2)	5 (22.7)	0 (0)	0.050
OCD (%)	3 (5.0)	0 (0)	3 (13.6)	0 (0)	0.136
GAD (%)	2 (3.3)	0 (0)	0 (0)	0 (0)	1.000
SAD (%)	2 (3.3)	1 (3.7)	0 (0)	0 (0)	1.000
Phobia (%)	4 (6.7)	0 (0)	0 (0)	0 (0)	0.398
Tics NOS (%)	0 (0)	1 (3.7)	0 (0)	0 (0)	0.450
ADHD subtype, n					
Combined (%)	NA	20 (74.1)	12 (54.5)	NA	0.237
Inattentive (%)	NA	7 (25.9)	7 (31.8)	NA	0.755
Hyperactive/impulsive	NA	0 (0)	3 (13.6)	NA	0.084

Note. TS = Tourette syndrome. ADHD = Attention-deficit/hyperactivity disorder. SES = Socioeconomic status of participants' parents classified using the International Standard Classification of Education (Unesco, 2012). CD = Conduct disorder. ODD = Oppositional defiant disorder. OCD = Obsessive-compulsive disorder. GAD = Generalized anxiety disorder. SAD = Separation anxiety disorder. NOS = Not otherwise specified.

the ADHD group. The manual for coding ER in children was created in collaboration with a PhD student from this high risk study, and for the validation of the manual we thus chose to expand our participant sample to increase the sample size as well as the age span. Consequently, these children were also part of the participant sample in paper 2. Participant characteristics for the slightly varying samples across papers will be evident from the respective paper.

3.4 Clinical assessment

All clinical assessments were conducted by a medical doctor or a psychologist who reached a diagnosis according to ICD-10 criteria (World Health Organization, 2016) in consultation with a specialist in child and adolescent psychiatry. We adopted a multi-informant approach in the assessment, comprising a clinician-administered diagnostic interview, interview measures of tic related symptoms, parent- and teacher-reported questionnaires on problem behaviors, executive functions, and symptoms of ADHD, and clinician-assessed IQ. All participants were administered the measures listed below with the exception of the Yale Global Tic Severity Scale (YGTSS) and the Premonitory Urge for Tics Scale (PUTS), which were only administered if a child screened positive for TS in the diagnostic interview.

3.4.1 The Kiddie-Schedule for Affective Disorders and Schizophrenia, Present and Lifetime Version (K-SADS-PL)

We ascertained children's diagnostic status employing the Danish version of the Kiddie-Schedule for Affective Disorders and Schizophrenia, Present and Lifetime Version (K-SADS-PL; Kaufman

et al., 1997). The K-SADS-PL is a semi-structured diagnostic interview which assesses any present or lifetime occurrence of psychopathology based on DSM-IV (American Psychiatric Association, 2000) criteria. Based on an initial screening interview, the clinician can employ supplementary interviews depending on the potential diagnoses. The interview is designed as a multi-informant tool where the child and caregiver(s) are interviewed separately, and in that order in accordance with the manual, ultimately allowing the clinician to make the final assessment factoring in both child and parent reports, questionnaires, and the overall clinical impression.

3.4.2 The Yale Global Tic Severity Scale (YGTSS)

All children screened positive for tics during the K-SADS-PL were assessed with the Norwegian version of the Yale Global Tic Severity Scale (YGTSS; Leckman et al., 1989), which for nearly two decades has been the gold standard for measuring tic severity (Robertson, 2015a). The YGTSS is a diagnostic interview that evaluates the number, frequency, intensity, complexity, and interference of motor and vocal tics separately with scores from 0-5 along with tic-related impairment rated from 0-50. In the present study, we used the YGTSS with and without the impairment score in our analyses.

3.4.3 The Premonitory Urge for Tics Scale (PUTS)

In the further assessment of children with tics, we used the Premonitory Urge for Tics Scale (PUTS), which is a clinician-administered questionnaire assessing type, frequency, and control of premonitory urges on a four-point scale from "Never" to "Always" (Woods et al., 2005). As recently suggested by Dr. Andrea Cavanna (personal communication, October 29, 2015), we did not limit our analyses to look at the total score of the PUTS, but also considered whether an urge was present at all.

3.4.4 ADHD Rating Scale (ADHD-RS)

Parents and teachers of all children completed the ADHD Rating Scale (ADHD-RS), which is a questionnaire covering ADHD symptoms comprising subscales of inattention and hyperactivity/impulsivity (DuPaul, Power, Anastopoulos, & Reid, 1998). An additional eight questions were included in the Danish version covering symptoms of CD. Each item is scored on a Likert-scale from 0-3 representing frequency. Teacher ratings were considered when establishing ADHD related impairment in school, and parent ratings were included in our analyses as T-scores based on Danish norms.

3.4.5 Child Behavior Check-List (CBCL)

Parents completed the Child Behavior Check-List (CBCL) for ages 6-18, which is a questionnaire identifying various problem behaviors in children during the past six months (Achenbach & Rescorla, 2001). In addition to a scale of "Total problems" and two general scales of "Internalizing" and "Externalizing" problems, the CBCL generates eight empirically derived subscales and six DSM-oriented subscales. Employing the CBCL in the clinical assessment, we used the derived Deficient Emotional Self-Regulation (DESR) profile in the validation study (paper 1).

The DESR combines the scores of three of the empirically derived scales (Aggressive Behavior, Anxious/Depressed, and Attention Problems) to generate a measure reflecting difficulties in ER, which has been found to be associated with high rates of disruptive behavior (Biederman et al., 2012; Spencer et al., 2011).

3.4.6 Behavior Rating Inventory of Executive Function (BRIEF)

The Behavior Rating Inventory of Executive Function (BRIEF) is a parent- and teacher-completed rating scale assessing executive function in children and adolescents aged 5-18 (Gioia, Isquith, Guy, & Kenworthy, 2000). Parents and teachers are asked to rate the child's problematic behaviors during the past six months. The BRIEF questionnaire consists of 86 questions which comprise eight clinical scales (Inhibit, Shift, Emotional Control, Initiate, Working Memory, Plan/Organize, Organization of Materials, and Monitor). We used The BRIEF in the general assessment of the child and specifically focused on the Emotional Control Scale (EC), which represents the influence of executive function on ER, in the validation study (paper 1).

3.4.7 Wechsler Intelligence Scale for Children, fourth edition (WISC-IV)

The Wechsler Intelligence Scale for Children – fourth edition (WISC-IV) is a test of intelligence for children and adolescents aged 6-16 (Wechsler, 2004). In the present study, the WISC-IV served as part of the clinical assessment of the child and constituted one of the exclusion criteria, as children scoring below 80 were excluded (the full scale IQ has a mean of 100 and a standard deviation of 15). The WISC-IV is administered by psychologists and produces a full scale IQ score along with four index scores (Verbal Comprehension, Perceptual Reasoning, Working Memory, and Processing Speed). We used the full scale IQ in the clinical characterization of the children and the full scale IQ along with the Verbal Comprehension Index (VCI) and the Perceptual Reasoning Index (PRI) in the validation study (paper 1). The former index reflects aspects of intelligence related to verbal concept formation and the latter index is a non-verbal measure of aspects of intelligence reflected in fluid reasoning and spatial processing.

3.5 Measures of emotion regulation

Psychologists or a trained research nurse administered the tests of ER. The Tangram Emotion Coding Manual (TEC-M) and the Picture Reappraisal Task (PRT) are both experimental tasks of ER, but with different approaches to the concept. Whereas the former assesses clinician-rated behavior during a frustration task with unguided and spontaneously occurring ER, the latter is a computer task of a learned ER strategy (cognitive reappraisal) which the participants are explicitly asked to use before ultimately rating the efficacy of the strategy through self-report. Both measures are described in detail below.

3.5.1 The Tangram Emotion Coding Manual (TEC-M)

The TEC-M was developed as a clinical instrument for evaluating profiles of ER in children in the context of parent-child-interactions. The primary outcome of the test is an evaluation of the child's overall ability of ER, whilst also providing information on other emotional processes such as expression of emotions, strategy use, and measures of parental behavior. Whereas the

Figure 4. The Tangram Puzzle with figure example.

present manual and concomitant outcome is new, the task itself is not. Originally, the Tangram Task made use of the ancient Chinese Tangram puzzle (Figure 4) in assessing parent-child interactions in the context of child anxiety (Hudson & Rapee, 2001, 2002), and later the task was used with a focus on attachment relations under the name Co-construction Task (Steele et al., 2007). Finally, the most recent version of the task, the Tangram Construction Task, has been applied once again in the field of child anxiety with an accompanying manual for coding primarily aspects of the parent-child dyad (Esbjørn, Christiansen & Steele, unpublished manuscript). For the purpose of studying ER in children, we developed a novel manual with an enhanced focus on the child and his or her specific ER profile with parental influence as a secondary measure. We based this manual on the process model of emotion regulation (Gross, 2014) with the five regulatory processes constituting the skeleton of the scoring sheet (Figure 5). Each item is associated with one of the regulatory processes, although this division generally serves the organization of items, as in theory the processes are interrelated and not necessarily chronologically ordered. Items were created through a theoretical-empirical approach in which they emerged from going through numerous videos and were then situated in the theoretical landscape of the process model of emotion regulation.

The present manual enables coding of behavior during the Tangram Construction Task in its most recent form, which is a five-minute complex puzzle serving as a frustration task. To increase the ecological validity, the child and one parent/primary caregiver are alone during the five minutes, while the task is video filmed. Before leaving the room, the test administrator shows the child the puzzle and six pictures of possible figures created from the Tangram pieces and says: *"Look, here is a puzzle. With this puzzle you can make a lot of different figures. It is your job to make as many of these figures with these jigsaw pieces as you can during the next five minutes"*. The test administrator next addresses the parent: *"You are here to support your child, but you are only to help if it is truly necessary. You get the solutions for you to see, but you are not to show them to the child"*. Instructions were provided verbatim to minimize the influence of having multiple people administering the task. One coder (JH) subsequently coded

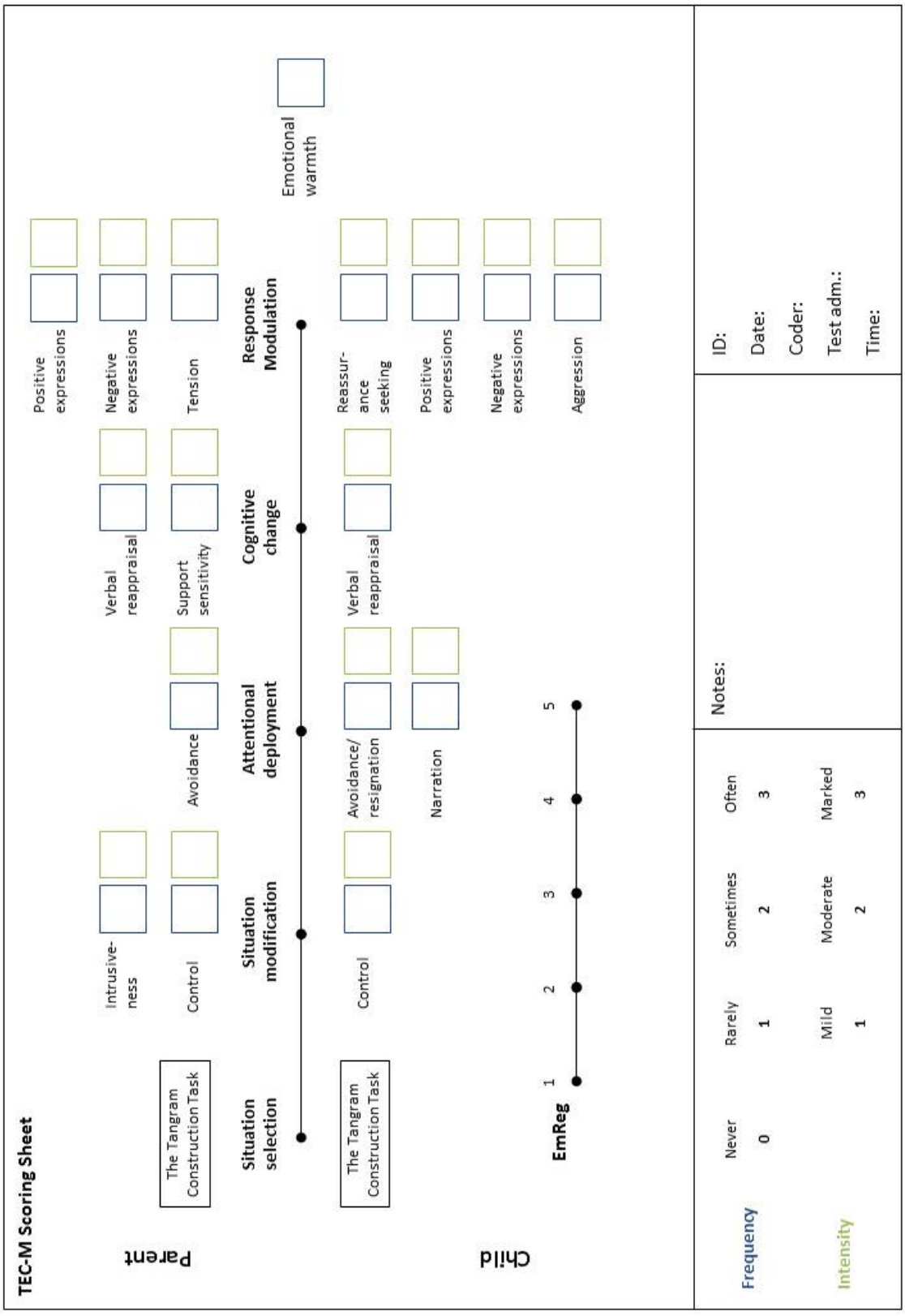

Figure 5. The TEC-M Scoring Sheet.

all videos with the TEC-M, which comprises information on the content of individual items and a guide for interpretation.

The manual comprises eight items on parental behavior (intrusiveness, control, avoidance, verbal reappraisal, support sensitivity, positive expressions, negative expressions, and tension) and nine items on child behavior (control, avoidance/resignation, narration, verbal reappraisal, reassurance-seeking behavior, aggression, positive expressions, negative expressions, and the overall emotion regulation scale (EmReg)). Finally, one item is scored on the parent-child dyad (emotional warmth). With the exception of the EmReg score, which is scored on a five-point Likert scale (1 = Very poor ER, 2 = Poor ER, 3 = Moderate ER, 4 = Good ER, 5 = Excellent ER), all items are scored on frequency (0 = Never, 1 = Rarely, 2 = Sometimes, 3 = Often) and intensity (1 = Mild, 2 = Moderate, 3 = Marked); the latter is only scored when frequency scores are above 1.

3.5.2 Picture Reappraisal Task (PRT)

The Picture Reappraisal Task (PRT) is an experimental computer task assessing participants' ability to cognitively reappraise negative affect when viewing unpleasant images. The task simultaneously generates a measure of emotional reactivity represented as the difference in negative affect following neutral images versus negative images, respectively. Emotional reactivity represents an expression of a child's threshold, intensity, and duration of arousal, and the target of regulation (Jahromi et al., 2012). The PRT has been used in different variations in several previous studies in typically developing populations comprising both children and adults as an experimental measure of reappraisal (McRae, Gross, et al., 2012; Ochsner, Bunge, Gross, & Gabrieli, 2002; Ochsner et al., 2004; Silvers et al., 2012).

In the present study, psychologists or a trained research nurse administered the PRT and provided instructions verbatim to decrease instruction bias. The task consisted of a total of 15 neutral images and 30 negative images selected from the International Affective Picture System (Lang, Bradley, & Cuthbert, 2008). Participants were asked to look at the images and respond naturally ("Look"), or to consciously try to decrease any negative affect following the viewing of the picture ("Decrease"), and to verbalize their efforts to allow for subsequent coding of strategies. For neutral images, the cue would always be "Look". For negative images, one half was preceded by the cue "Look", and the other half was preceded by the cue "Decrease", thus creating three conditions: 1) "Look neutral", 2) "Look negative", and 3) "Decrease negative" (Figure 6). Pictures were displayed in blocks of five aversive images and 10 neutral images, alternating in sequence in a pseudorandomized manner. We counterbalanced images between the two negative conditions with similar valence ratings for both groups of pictures. When reacting naturally, the participant was instructed to look at the picture quietly and be attentive to any arisen feelings. Examples of reappraisal could be imagining that the picture was taken from a movie to challenge the authenticity or to suggest solutions that would make the situation better. Participants were trained on how to react naturally and how to reappraise in a standardized manner. If a child did not initiate reappraisal within the first few seconds, formalized verbal prompts were provided by the test administrator. Finally, participants reported their experienced level of negative affect on a scale from one to five (1 = no negative affect, 2 = mild

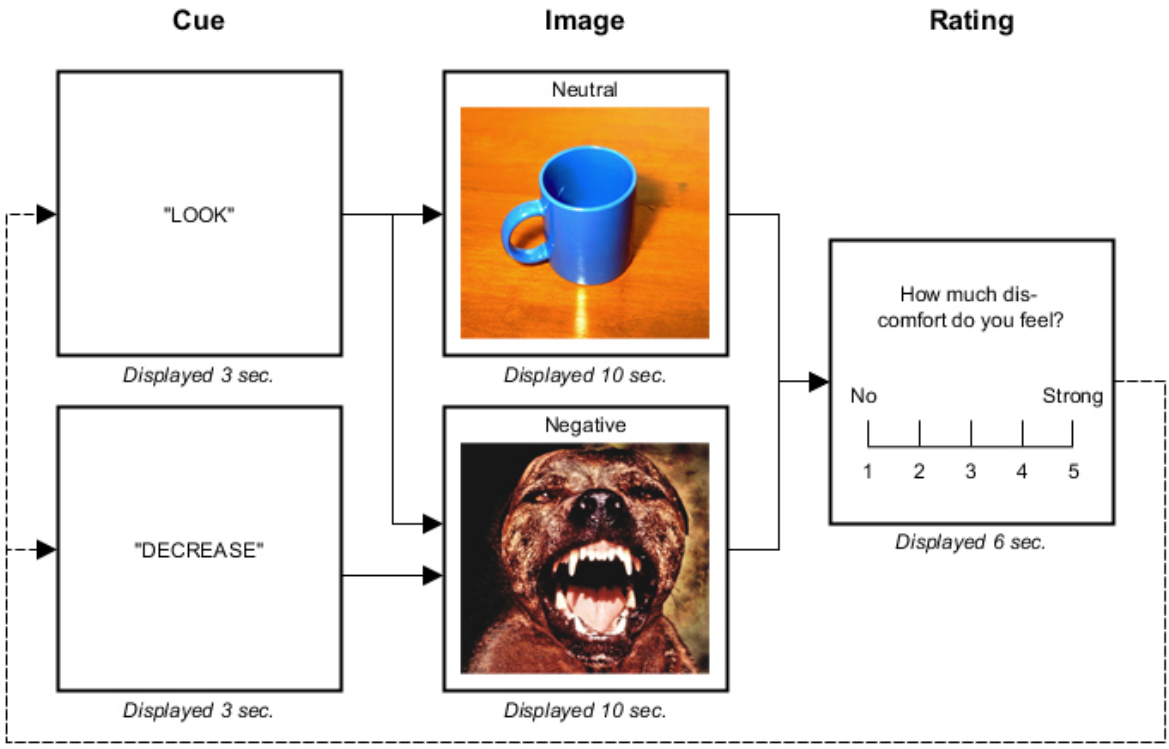

Figure 6. Picture Reappraisal Task.

negative affect, 3 = moderate negative affect, 4 = marked negative affect, 5 = strong negative affect).

All verbalizations during reappraisal were subsequently coded by an assessor (JH) following modified procedures from the Reappraisal Tactic Coding Guide (McRae, Ciesielski, et al., 2012), which describes ten categories within cognitive reappraisal: "Explicitly positive", "Change current circumstances", "Reality challenge", "Change future consequences", "Agency", "Distancing", "Technical-analytic-problem solving", "Acceptance", "Non-specific reappraisal", and "Failure to reappraise". Based on the collected data, we added two categories: "Negating the negative" and "No negative affect". The former category bears resemblance to suppression by removing or replacing the element causing discomfort, and the latter category was used when children explicitly stated that the picture was not perceived as unpleasant at all. Unlike the original coding guide, which estimated the degree to which each category was employed, we here documented the specific strategy for each reappraisal.

3.6 Ethical considerations

The protocol for the research presented in this thesis was approved by the Regional Committee on Health Research Ethics (journal number H-2-2013-085) and the Danish Data Protection Agency (journal number 2017-58-0015). Written informed consent was obtained from legal

caretakers of all participants in accordance with the declaration of Helsinki. It was pivotal to the study design to maintain an awareness that we were working with children, many of whom with vulnerable mental states. We obtained verbal consent from all children, which would overrule parental consent if the child should wish to withdraw from the study. We explicated that participants were free to withdraw from the study at any time and that the assessment and potential treatment offered would in no way be affected by withdrawal. All procedures were thoroughly explained by experienced staff including confidentiality and any potential risks. Participants were offered an inconvenience reimbursement for transportation and time spent of DKK 750, approximately 100 euros, which also covered participation in an MR brain scan on a third day. There were no significant risks associated with participation, but the time spent at the hospital in connection with the assessment could be inconvenient to families, just as some neuropsychological and ER tests could cause mild distress. We prioritized a thorough debriefing following neuropsychological tests, which had all previously been applied with children in the same age range. It was our clear belief that any potential discomfort caused by the lengthy days of testing would be outweighed by the value of the research, which hopefully could benefit other families affected by the same neuropsychiatric disorders.

3.7 Data analyses

We conducted all data analyses using SPSS version 22.0 adopting a significance threshold of .05 and using two-tailed tests. We applied the Bonferroni correction to control for family-wise error rate in multiple comparisons. Nonparametric tests were used when data did not meet test specific assumptions with few exceptions when a test was deemed robust to certain violations. For all papers, we computed differences in age and IQ with a one-way ANOVA with Bonferroni corrected pairwise comparisons where relevant (independent samples t-test for paper 1 with only two groups). Group differences in SES, sex, comorbidity, and ADHD subtype were computed with the chi-square test of homogeneity or Fisher's exact test. The remaining analyses for each paper are described below.

3.7.1 Paper 1: The Puzzle of Emotion Regulation – Development and Evaluation of the Tangram Emotion Coding Manual for Children

We assessed inter- and intrarater reliability of items on the TEC-M between two raters for a sample of 30 participants coded by both raters and 10 participants coded twice by the same rater with minimum three months between coding sessions. We tested for absolute agreement with average-measures ICC's, applying a two-way mixed model (Hallgren, 2012), and interpreted the clinical relevance of reliability coefficients as $< .40$ = poor, .40-.59 = fair, .60-.74 = good, and $> .75$ = excellent (Cicchetti, 1994). For the analysis of convergent and discriminant validity, we chose the measure of overall ER (EmReg score) for comparison with the EC scale, the DESR profile, and IQ measures. As the EmReg score is an ordinal variable and presented with a non-normal distribution, we used Spearman's rank-order correlation for the analyses (EmReg score versus EC, DESR, total IQ, VCI, and PRI, respectively). We assessed content validity by having eight experts in the field evaluate items as "Essential", "Useful, but not essential", or "Not necessary", in regard to whether the specific item reflected the theoretical domain satisfactorily (Lawshe, 1975). These ratings allowed for a calculation of a content validity ratio

(CVR). The critical value of the CVR is dependent on the number of panelists; in this case 0.75. Items rated as essential by more than half of the panelists held some content validity (Lawshe, 1975). We conducted Mann-Whitney U tests for the investigation of group differences, as item scores were ordinal variables, and calculated effect sizes for significant results.

3.7.2 Paper 2: Emotion Regulation in Children with Tourette Syndrome – Associations with ADHD and Tic Severity

To investigate a potential latent structure in the items of the TEC-M, we performed an exploratory factor analysis (EFA) using principal axis factoring with an orthogonal rotation (varimax) for parent and child frequency items separately. We included all items from the TEC-M in the analysis and accepted item loadings above 0.3 as pertaining to a factor. We applied the Kaiser Criterion of eigenvalues above one (Kaiser, 1960) in deciding the number of factors to retain. Group differences in ER were examined only for frequency scores due to the low number of intensity scores (which were only scored when frequency was scored higher than 1). We assessed differences in proportions of scores between groups on all items of the TEC-M with the chi-square test of homogeneity followed by pairwise comparisons with the Holm-Bonferroni method. We created two levels of frequency, "Low" and "High", by combining never/rarely and sometimes/often, while the EmReg score retained five levels. We analyzed group differences in factor scores nonparametrically, due to skewness, with the Kruskal-Wallis H test followed by pairwise comparisons where relevant with Dunn's procedure with a Bonferroni correction. For the explorative analysis of correlations between the EmReg score and symptoms pertaining to TS and ADHD, we used Pearson's product-moment correlation and the point-biserial correlation. We explored EmReg correlations with a) YGTSS total score; b) YGTSS score minus impairment; c) PUTS total score; d) any presence of premonitory urges represented by a score of 3 or 4 on the PUTS in any of the first six questions representing different urges; e) ability to hold back tics represented by a score of 3 or 4 on the final question of the PUTS ("I am able to stop my tics, even if only for a short period of time"); f) ADHD-RS subscale inattention score; g) ADHD-RS subscale hyperactivity/impulsivity score; and h) ADHD-RS subscale conduct disorder score. Only children with TS were included in analyses a-e, whereas correlations with ADHD-RS scores were conducted for the total sample.

3.7.3 Paper 3: Reappraisal is an Effective Emotion Regulation Strategy in Children with Tourette Syndrome and ADHD

We calculated the effect of reappraisal (regulation success) on the PRT as the percent decrease in negative affect from "Look negative" to "Decrease negative" (("Look negative" – "Decrease nega-tive")/"Look negative" x 100) and reactivity as the percent increase in negative affect comparing "Look neutral" to "Look negative" (("Look negative" – "Look neutral")/"Look neutral" x 100) (Silvers et al., 2012). Performing a Friedman test, as data were not normally distributed, with Bonferroni corrected pairwise comparisons, we investigated differences in ratings of negative affect from condition one ("Look neutral") to condition two ("Look negative"), and condition two to condition three ("Decrease negative") for all participants and within each group; the former comparison representing emotional reactivity, and the latter representing ER. For the analysis of correlations between age and reappraisal/reactivity, we conducted a Pearson's product-moment correlation. We used an ANCOVA with age as a covariate for the

analysis of regulation success across groups, and a one-way ANOVA for group differences in reactivity. Looking further into the specific verbal strategies encompassed in reappraisal, we examined group differences in frequency of choice of a specific strategy with the Kruskal-Wallis H test, as data were not normally distributed, looking at each category separately, as well as categories one to nine combined, representing all the presumed efficacious strategies. Finally, we conducted Spearman's rank-order correlations to explore the relationship between the specific verbal strategies and age, regulation success, and reactivity, respectively.

3.7.4 Additional analysis employed in the discussion

For discussion purposes, we incorporated a few correlation analyses which were not part of the papers. Specifically, we investigated the correlation between reappraisal and reactivity appertaining to paper 3 with the Pearson's product-moment correlation. Furthermore, we examined correlations between the EmReg score of the TEC-M and clinical outcome measures of irritability from the K-SADS-PL with Spearman's rank-order correlations as the K-SADS classifications are scored on an ordinal scale.

4. Results

4.1 Participant characteristics for the total sample

Although participant samples varied slightly across the three papers, characteristics matched the total sample. The two groups of children in paper 1 and the four groups of children in papers 2 and 3 did not differ significantly on age or sex, nor were the three clinical groups different in regard to comorbidity and ADHD subtype for the last two papers. Groups did, however, differ significantly on SES and IQ, with the highest scores in the control group followed by the TS group, the TS + ADHD group, and finally the ADHD group (Table 2). Group differences in IQ ($F(3, 165) = 5.986$, $p = .001$) were driven by the ADHD group presenting with significantly lower scores than the control group ($MD = 9.048$, 95% CI [2.52, 15.57], $p = .002$) and the TS group ($MD = 9.048$, 95% CI [2.52, 15.57], $p = .002$).

4.2 Paper 1: The Puzzle of Emotion Regulation - Development and Evaluation of the Tangram Emotion Coding Manual for Children

4.2.1 Inter- and intrarater reliability

The interrater reliability analysis of items on the TEC-M resulted in excellent agreement for 11 items, good agreement for 13 items, fair agreement for six items, and poor agreement for four items (Table 3). This resulted in an average ICC of .61. ICC's for the intrarater reliability analyses were excellent for 23 items for rater one and 24 items for rater two, good for five items for both raters, fair for three (rater one) and four (rater two) items, and poor for one item, resulting in average ICC's of .82 and .79, respectively.

4.2.2 Convergent and discriminant validity

The analysis of convergent validity of the TEC-M produced mixed results. For the association between the EmReg score and the EC scale, the correlation coefficient was small and non-significant, r_s = -.20, $p = .079$. The correlation coefficient between the EmReg score and the

Table 3. Interrater and intrarater reliability of TEC-M items.

		Interrater	Interater 1	Interrater 2
Parent items	Intrusiveness, F	0.71	0.88	0.84
	Intrusiveness, I	0.76	0.83	0.91
	Avoidance, F	0.79	1.00	0.90
	Avoidance, I	0.78	1.00	0.85
	Control, F	0.63	0.63	0.82
	Control, I	0.76	0.52	0.71
	Verbal reappraisal, F	0.89	0.94	1.00
	Verbal reappraisal, I	0.73	0.89	0.91
	Negative expressions, F	0.50	0.74	0.83
	Negative expressions, I	0.48	0.78	0.76
	Positive expressions, F	0.64	0.83	0.96
	Positive expressions, I	0.58	0.94	0.54
	Tension, F	0.55	1.00	1.00
	Tension, I	0.75	1.00	1.00
	Support sensitivity, F	0.75	0.57	0.65
	Support sensitivity, I	0.22	0.69	0.53
Child items	Situation rejection, F	Z.V.	Z.V.	Z.V.
	Situation rejection, I	Z.V.	Z.V.	Z.V.
	Control, F	0.66	0.89	0.74
	Control, I	0.51	0.37	0.40
	Avoidance/resignation, F	0.61	0.95	0.66
	Avoidance/resignation, I	0.70	0.91	0.36
	Narration, F	0.64	0.76	0.80
	Narration, I	0.67	0.86	0.80
	Verbal reappraisal, F	0.60	1.00	0.84
	Verbal reappraisal, I	0.71	0.97	0.85
	Reassurance seeking, F	0.83	0.83	0.90
	Reassurance seeking, I	0.40	0.69	0.68
	Tension, F	Z.V.	Z.V.	Z.V.
	Tension, I	Z.V.	Z.V.	Z.V.
	Aggression, F	-0.07	Z.V.	0.78
	Aggression, I	-0.07	Z.V.	0.78
	Negative expressions, F	0.72	0.47	0.93
	Negative expressions, I	0.62	0.73	1.00
	Positive expressions, F	0.78	0.78	0.80
	Positive expressions, I	0.76	1.00	0.83
	Incongruent positive affect, F	Z.V.	Z.V.	Z.V.
	Incongruent positive affect, I	Z.V.	Z.V.	Z.V.
	EmReg	0.78	0.81	0.79
Shared	Emotional warmth	0.33	1.00	0.54

Note. F = Frequency, I = Intensity. Z.V. = Zero variance. Dark blue represents good or excellent reliability, light blue represents fair reliability, and grey represents poor reliability.

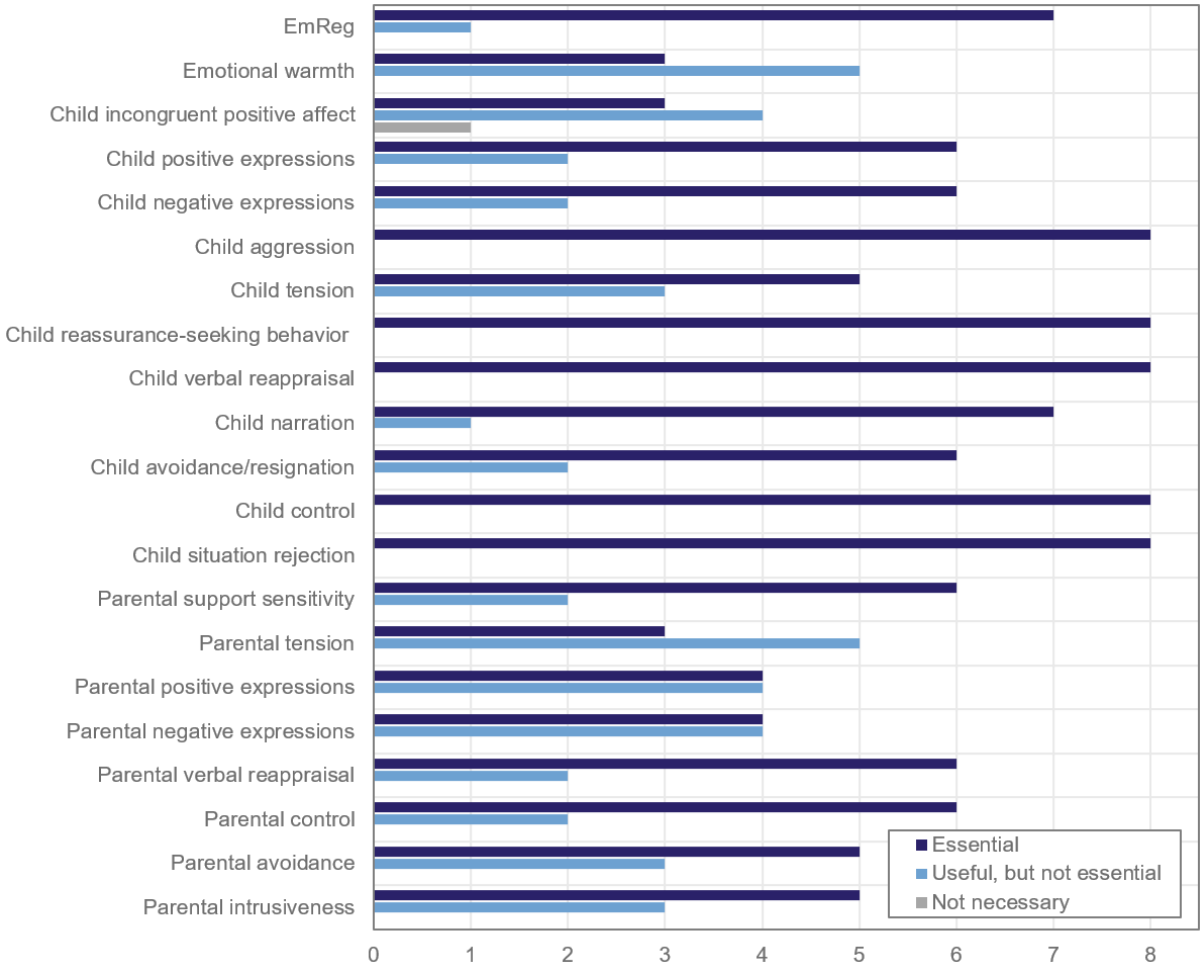

Figure 7. Content validity. Rating count from eight panelists.

DESR profile, however, was close to medium and significant, r_s = -.28, p = .016. The examination of discriminant validity resulted in medium or close to medium, significant correlation coefficients between the EmReg score and the full scale IQ (r_s = .30, p = .011), the VCI (r_s = .27, p = .026), and the PRI (r_s = .28, p = .018).

4.2.3 Content validity

Seven TEC-M items out of a total of 21, as frequency and intensity items were rated together, reached the required CVR of 0.75 (situation rejection, control, narration, verbal reappraisal, reassurance-seeking behavior, aggression, and the EmReg score). Eighteen items were rated as essential by at least half the panelists, and only a single item, which was ultimately removed, was categorized as unnecessary by one panelist (Figure 7).

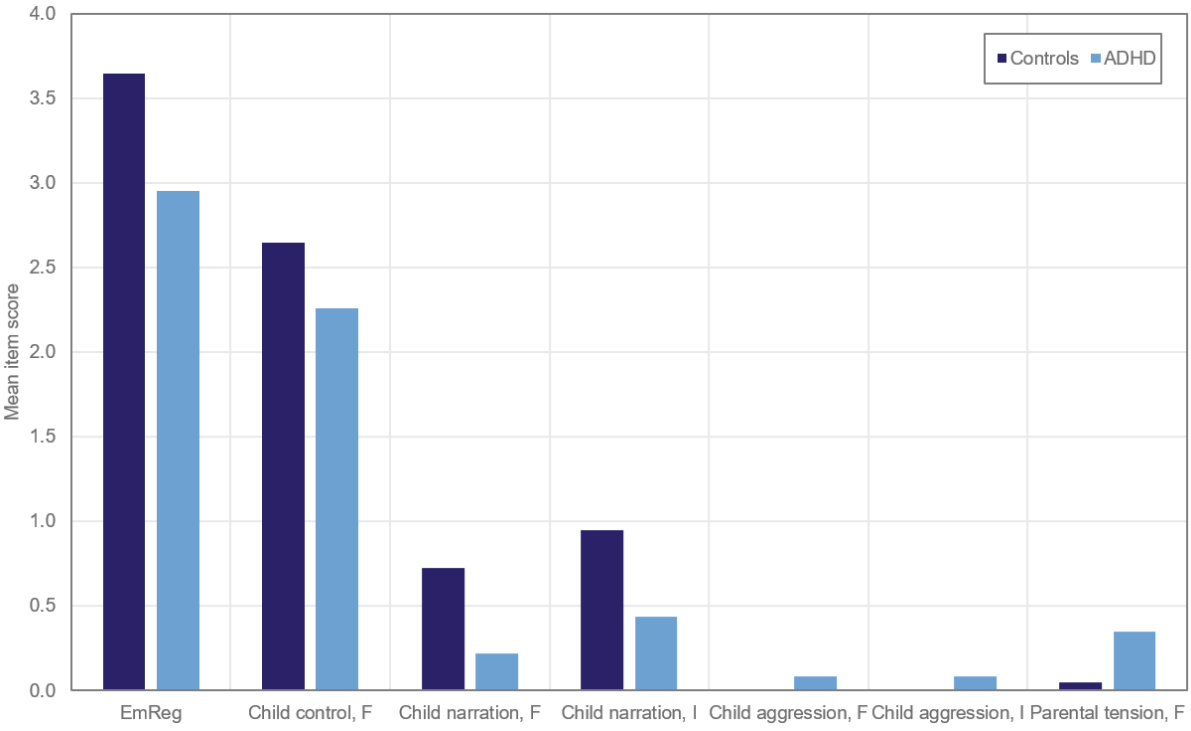

Figure 8. Mean score for items that differed significantly between the ADHD group and the control group. Note that the EmReg score has a range of 1-5, whereas the remaining items have ranges of 0-3.

4.2.4 Group differences

The ADHD group and the control group differed on seven items of the TEC-M (Figure 8). On the primary outcome of the TEC-M, the EmReg score, children from the ADHD group scored significantly lower than children from the control group, U = 414.5, z = -3.122, p = .002, r = -0.34. The same pattern presented for frequency of child control, U = 409, z = -2.566, p = .010, r = -0.28, and frequency, U = 452, z = -2.885, p = .004, r = -0.31, and intensity of child narration, U = 499.5, z = -2.366, p = .018, r = -0.26. Conversely, the ADHD group scored higher on frequency and intensity of child aggression than the control group, U = 775, z = 2.366, p = .019, r = 0.26, and on frequency of parental tension, U = 808.5, z = 1.982, p = .047, r = 0.21.

4.3 Paper 2: Emotion Regulation in Children with Tourette Syndrome – Associations with ADHD and Tic Severity

4.3.1 Exploratory factor analysis

An exploratory principal axis factoring analysis revealed three meaningful factors for the child items in the TEC-M, which explained 53% of the variance in total (Table 4). Items loading on factor 1 were control and EmReg with positive loadings, and avoidance, reassurance-seeking, and negative expressions with negative loadings, and we thus named this factor ”Emotional

Table 4. Orthogonally rotated factor loadings for child items.

Item	**Factor 1:** Emotional control	**Factor 2:** Emotion communication	**Factor 3:** Verbal emotion regulation strategies
Control	0.539		
Reassurance-seeking	-0.354		
Avoidance	-0.533		
EmReg	0.851	0.383	
Negative expressions	-0.397	-0.363	
Emotional warmth		0.645	
Aggression		-0.563	
Positive expressions		0.306	
Narration			0.679
Verbal reappraisal			0.368
Eigenvalue	2.483	1.594	1.284
% variance explained	24.83	15.94	12.84

Note. Items below 0.30 were suppressed.

control". Items loading onto factor 2 were positive expressions, emotional warmth, and EmReg with positive loadings, and aggression and negative expressions with negative loadings. We named the second factor "Emotion communication". Narration and verbal reappraisal loaded onto the third factor, which was named "Verbal ER strategies". The EFA for parent items did not render a valid factor solution and was not subject to further analyses.

4.3.2 Group differences in emotion regulation on the TEC-M

Groups differed on the primary outcome of the TEC-M, the EmReg score, $p = .019$. Pairwise comparisons revealed no significant difference between the control group and the TS group, whereas both groups including children with ADHD scored significantly lower than the two former groups (controls > ADHD, $p = .012$; controls > TS + ADHD, $p = .010$; TS > ADHD, $p = .037$; and TS > TS + ADHD, $p = .029$). Groups differed in a similar manner on factor 1 (Emotional control; $H = 14.819$, $p = .002$). The control group presented with higher scores of ER than the ADHD group ($p = .037$) and the TS + ADHD group ($p = .031$), and the TS group achieved significantly higher scores than the TS + ADHD group ($p = .041$). Finally, level of parental tension was different across groups, $p = .001$. Seventeen percent of parents of children with ADHD presented with high frequency of tension, which was significantly higher than the control group, in which high frequency of parental tension was absent, $p = .028$.

4.3.3 Associations between emotion regulation, symptom severity, and premonitory urges

Tic severity (YGTSS) or presence of premonitory urges (PUTS) did not correlate with ER ability as measured with the EmReg scale. Severity of inattention and hyperactivity/impulsivity (ADHD-RS), however, were inversely correlated with ER ability represented by small, significant correlation coefficients (Table 5). Severity of conduct disorder (ADHD-RS) and ER ability were not significantly correlated.

Table 5. Associations between the EmReg score and scores from the YGTSS, the PUTS, and the ADHD-RS.

	EmReg	
	r (df)	p
YGTSS – Total score	-0.107 (63)	0.397
YGTSS – Minus impairment	-0.009 (63)	0.941
PUTS – Total score	0.111 (63)	0.377
PUTS – Presence of any urge	-0.064 (63)	0.612
PUTS – Hold back tics	0.058 (63)	0.644
ADHD-RS – Inattention	-0.234 (147)	**0.004**
ADHD-RS – Hyperactivity/impulsivity	-0.211 (147)	**0.010**
ADHD-RS – Conduct disorder	-0.152 (147)	0.065

Note. YGTSS = Yale Global Tic Severity Scale. PUTS = Premonitory Urge for Tics Scale. ADHD-RS = ADHD Rating Scale.

4.4 Paper 3: Reappraisal is an Effective Emotion Regulation Strategy in Children with Tourette Syndrome and ADHD

4.4.1 Main effects of condition and associations with age

We found a main effect of reappraisal in the PRT represented by significantly different mean ratings across conditions, $\chi^2 = 245.116$, $p < .001$. Post hoc analysis revealed that ratings of negative affect following a natural response to negative images ($Mdn = 2.50$) were significantly higher than for neutral images ($Mdn = 1.00$, $p < .001$). Furthermore, children rated negative pictures as less unpleasant following reappraisal ($Mdn = 2.13$) than following a natural response, $p < .001$. Regulation success correlated significantly with age, $r = .227$, $p = .004$, whereas reactivity did not, $r = .101$, $p = .203$).

4.4.2 Group differences in regulation and reactivity

Mirroring the total sample, all groups presented with an effect of test condition with increased ratings when pictures were negative as opposed to neutral, and a decrease following reappraisal. Differences between test conditions were significant for all groups, with the exception of children in the ADHD group whose affect after reappraisal was not significantly lower than the affect following reacting naturally to a negative image. We did not find a difference between groups on regulation success when we included age as a covariate, $F(3, 155) = 1.063$, $p = .367$, partial $\eta^2 = .020$, nor did we see any difference in reactivity across groups, $F(3, 156) = 1.964$, $p = .122$.

4.4.3 Reappraisal strategies

Similar to regulation success, groups did not differ on choice of specific strategy within the context of reappraisal. We documented the degree to which participants applied a theoretically efficacious strategy, as opposed to using simpler forms of ER such as suppression, and found a medium-sized correlation between number of effective strategies and regulation success, $r_s = .322$, $p < .001$. The most frequently used type of reappraisal, "Change current circumstances", appeared to be driving this association. The use of the suppression associated strategy "Negating

the negative" was not associated with regulation success, r_s = -.103, p = .300. Whereas participants' reactivity did not correlate with frequency of use of theoretically efficacious strategies, it did correlate significantly with use of "Negating the negative", r_s = .259, p = .008.

4.4.4 Additional analysis employed in the discussion

The analysis including reactivity and regulation success resulted in a small, significant correlation (r = .293, p < .001). The EmReg score correlated significantly and inversely with irritability from the ODD screening, (r_s = -.262, p = .002), but not with irritability pertaining to depression (r_s = -.107, p = .219) or mania (r_s = -.002, p = .977).

5. Discussion

5.1 Main findings and contributions to the field

The purpose of the present work was two-fold. First, we aimed to develop a generic test of ER to be used across diagnostic groups and in non-clinical populations. Second, we investigated ER abilities in children with TS, including clinical contrast groups with ADHD and TS + ADHD, respectively, and a control group of typically developing children to elucidate the specificity of findings. The aim was to increase the understanding of the emotional component in TS by employing two experimental tests of ER; the first assessing clinician-rated, spontaneous ER, and the second assessing self-evaluated ER success using the learned strategy of cognitive reappraisal. The theoretically embedded, novel measure of ER, the TEC-M, showed an overall good interrater reliability, excellent intrarater reliability, and acceptable psychometric properties. Applying this test in the four groups of children showed that children with TS were not impaired in ER ability compared to typically developing controls, when they did not have a comorbid disorder of ADHD. This finding is in line with previous literature suggesting a specific role of ADHD in dysregulated behavior in TS, but the present study is the first to demonstrate this relationship with an experimental measure in a large group of children; thereby bridging experimental findings and clinical impressions. Children did not differ according to group on the second measure of ER focusing specifically on the strategy of reappraisal, nor did they differ on reactivity. Findings across groups, however, supported the effectiveness of reappraisal as an ER strategy.

This present work has contributed to the study of ER through the development of a novel instrument with a potential applicability in research as well as in the clinic. As illustrated in this thesis, the field has suffered from a scarcity of appropriate experimental measures of ER in children and adolescents. The TEC-M makes it possible to study spontaneously occurring ER experimentally, in a design that incorporates the connection between emotional outbursts and parent-child interactions by taking measures of parental behavior into account in the assessment of children's ER ability. To the best of our knowledge, this study is the first to examine ER ability experimentally in children with TS. We find that children with TS perform equally to controls on both measures of ER, as well as on IQ, which contradicts the earlier mentioned

"umbrella tendency" of including wildly differing aspects of behavior into the diagnosis of TS. In children with TS without ADHD, we do not see any significant differences compared to controls. Of interest for patterns of ER in children with ADHD, we demonstrate impairments in spontaneous regulation, but an ability to learn and employ the cognitively demanding strategy of reappraisal, which is an interesting dissociation that may impact the otherwise rather bleak views on ER ability in children with ADHD.

The following sections will center on the overarching aim of the study, discussing the accumulated findings across subsections. These subsections will comprise a discussion of the experimental study of ER, our findings regarding ER in children with TS and the role of ADHD, as well as more general discussions regarding the potential role of ER as a transdiagnostic marker and a discussion of the heterogeneity of TS, and what the present findings may offer to a dimensional approach to TS.

5.2 The experimental study of emotion regulation

As mentioned earlier in this thesis, the greatest challenge of studying ER lies in its immense complexity and fluid transitions between different processes, which are difficult to disentangle. Not only are at least five groups of processes part of ER (Gross, 2014), but numerous nuances are present within each group, as well as differing individual incentives for each specific approach. In addition, further processes precede the regulation itself, including emotion recognition, emotion reactivity, and emotion generation. The relatively fluid boundaries between these emotional processes impede the deciphering of the basis of a certain type of behavior as related to the emotion generation process or the regulation process (Bunford et al., 2018). Assuming that ER theoretically is a delimited ability, this ability is hardly observable in full. An inherent limitation to the experimental study of ER thus becomes a reliance on mere observable behavior and an acceptance of this premise.

Focusing on examples from coding with the TEC-M (papers 1 and 2), behavior involving a child leaning away from the puzzle with a sad expression, stopping the work temporarily, would be characterized as avoidance/resignation and negative expressions, cf. the TEC-M scoring sheet (Figure 5). This behavioral expression may, however, be based on a multitude of underlying emotional states, such as fatigue, boredom, sadness related to performance on the task or to the relationship with the parent, sadness related to something completely different from the task or the dyad, frustration directed at the task, the parent, or at the child him- or herself, or mounting inattention; the latter involving yet another process to be considered, namely the potential influence of cognitive ability. Any sign of negative or positive affect would be coded accordingly, but it would not be evident from the behavioral manifestation, whether high emotional reactivity or low regulation was responsible for the response. Regardless of whether an intense negative emotional expression is caused by a low reactivity threshold or poor ER abilities, coding this behavior as a negative expression with the TEC-M will most likely still elucidate aspects of ER. In the PRT (paper 3), high reactivity did not affect regulation success (reappraisal) negatively; on the contrary, the individual ranks between groups on reactivity were identical to ranks on regulation success. Interestingly, although this was not presented in the third paper, reactivity and regulation success correlated significantly with each other ($r(158) = .293$, $p < .001$), poten-

tially pointing to a positive relationship between the two concepts with high reactivity increasing the awareness of emotional states and thereby the regulation of these states. Although this hypothesis is not in line with previous findings of high reactivity (Fogleman et al., 2018) and poor ER (Graziano & Garcia, 2016) in children with ADHD, this discrepancy could easily be caused by differences in methods of assessment. Reactivity did, however, correlate positively with the strategy resembling suppression the most, suggesting that children high in reactivity are more likely to employ this strategy. This finding is interesting in light of the earlier described association between suppression and activity in the amygdala. Returning to the TEC-M, the subjective threshold for an individual to generate a response on items, such as seeking reassurance with a parent, remains unknown to the coder. For other items, understanding the background for potentially ambiguous behavior, such as a parent smiling while correcting the child's work, or commenting that something is incorrect, requires knowledge regarding the general dynamic of the dyad. Finally, some items are more directly applicable to the assessment of ER, for example the spontaneous occurrence of verbal reappraisal. Accepting a lack of insight into some aspects and simply coding observable behavior thus remains a premise of the task and is also a way of simplifying the interrelated processes by simply looking at the behavioral response in a frustrating situation.

As illustrated, the association between observable behavior and the underlying emotion is complex (Bunford et al., 2018) and although many ER tasks allow for stimulus control, measurement precision remains a domain of compromise. In support of the TEC-M, however, results from the inter- and intrarater reliability analysis (paper 1) illustrated that two raters quite reliably were able to reach the same conclusion, even though the underlying process remained unknown.

In addition to the challenges associated with the conceptual complexity in the experimental study of ER, further factors are likely to influence the process of regulation. Although we strived to increase the ecological validity of the frustration task (paper 1) as much as possible, many participants would most likely still be aware that they were part of a staged situation, in which their behavior would be evaluated (Bunford et al., 2015). Relevant to both experimental tasks (papers 1, 2, and 3), successful ER has been shown to depend not only on the ability to apply an efficacious strategy, but also on participants' beliefs about emotions being malleable (Gutentag, Halperin, Porat, Bigman, & Tamir, 2017). Parent-child interactions have quite consistently been implicated in the development of psychopathology with established associations between dyadic relations and aggression and externalizing behavior in childhood (Lewis et al., 2006; Mackler et al., 2015). In paper 2, we demonstrated increased rates of parental tension in parents of children with ADHD compared to all other groups, and this group also presented with the poorest ER ability. These separate findings are not necessarily related, but may be indicative of an interpersonal and potentially reciprocal effect of low tolerance for frustration in children and parental tension, respectively. Finally, the development of cognitive abilities and maturation of certain structures in the brain are known to undergo substantial changes during childhood and adolescence (Ahmed et al., 2015), which may further complicate findings of ER ability given potentially differing individual trajectories.

5.2.1 Validation of the TEC-M

The validation of a psychological instrument involves multiple processes, which were previously viewed as distinct entities, but are increasingly being incorporated into the unifying concept of construct validity, encompassing convergent validity, discriminant validity (Strauss & Smith, 2009), and content validity (Cronbach & Meehl, 1955). The basis for supporting construct validity of a test is its theoretical foundation, which must influence the selection of items from the beginning (Strauss & Smith, 2009); in this study the process model of emotion regulation (Gross, 2014). Convergent validity is obtained through robust associations between the outcome of the new test and a theoretically related measure, whereas discriminant validity is represented by smaller associations with measures that are unrelated to the new instrument (Evans et al., 2016). Finally, content validity is an estimation of the degree to which items on an empirical measure adequately reflect their theoretical framework (Carmines & Zeller, 1979).
The distinction between specific types of ER and different results depending on the source of information is relevant to the validation of the TEC-M. With the aim of testing the convergent validity of the instrument, we set out to find a measure of comparison that was theoretically related to the TEC-M to illustrate the capacity of the latter to measure ER. Analyses of convergent validity produced only small correlation coefficients (-.20 and -.28) between the primary outcome (the EmReg score) and existing measures of ER, namely the EC scale from the BRIEF and the DESR profile from the CBCL. However atypical for analyses of convergent validity, these results were not entirely surprising. Although we expected correlation coefficients in the medium range (i.e., above .30), the premise for developing the TEC-M was to design an instrument that was different from existing measures and was able to capture aspects of dysregulated behavior that questionnaires could not. The correlation with the DESR profile was significant and close to medium, suggesting that the output of the TEC-M was more representative of the DESR profile than the EC scale. The EC scale carries a marked executive component, and the more behaviorally oriented DESR profile may better reflect the content of the primary outcome of the TEC-M representing the overall ER ability (the EmReg score). The association with the DESR profile further indicated a connection between clinician-rated and parent-rated conceptions of child behavior; however, the methodological variation between measures (on the one hand parent-report of general behavior in a six months' timeframe and on the other hand clinician-rated behavior in a staged, frustration-eliciting context) may easily explain the low correlation coefficients for both measures.

The examination of discriminant validity produced small to medium correlation coefficients between the EmReg score and full scale IQ, the VCI, and the PRI. These results were consistent with the TEC-M's hypothesized association with a measure of intelligence.

Overall, content validity ratings were satisfactory. Though only few measures reached the CVR required to officially establish content validity (.75 with eight panelists), this result possibly reflected the inclusion of parent items in a measure of children's ER, which were consequently classified by some members of the expert panel as only "Useful, but not essential".

5.2.2 The transferability of empirical knowledge to everyday disruptive behavior

The challenge of producing ecologically valid measures of ER remains a constant point for improvement. As evident from the present work, elements of frustration or negativity were included in both experimental measures of ER, as this is essential to create responses that can be coded or measured in a defined period of time. This premise, however, markedly limits the transferability of empirical findings to everyday behavior, as only regulation of negative emotions is addressed and moreover in an artificial setting. Changing the perspective to the disruptive behavior manifesting in everyday life, substantial variation in developmental trajectories within disorders are evident, further obscuring direct comparisons. To discuss externalizing disorders collectively entails a wide compilation of associated behaviors. In the example of ADHD, the distinction between symptoms of inattention and hyperactivity can predict differing problematic behaviors. Inattention has been linked with academic underachievement and social isolation, and hyperactivity has been associated with peer rejection and aggression (Mullin & Hinshaw). Thus, it is not only a matter of the experimental test to be able to mimic everyday behavior, but also which aspect of behavior the test should elucidate.

Although we did not include any correlations between the EmReg score and clinical measures of anger and irritability in the papers, these analyses are interesting to incorporate in the context of transferability. The K-SADS-PL interview, as described in the methods section, addresses aspects of irritability as part of the screening for depression, mania, and ODD; each with measures of the frequency and intensity of this behavior resulting in a collective rating from one to three. In depression, the question involves subjective feelings of irritability with mild if any behavioral manifestations. In mania, subjective feelings of anger or irritability, often resulting in aggressive behavior, are addressed. Finally, the screening for ODD focuses on loss of control resulting in rage attacks. Looking into the associations between these three clinical representation of anger and irritability, we find that only the latter type encompassed in the ODD screening, significantly and inversely correlated with the EmReg score ($r_s(132)$ = -.262, p = .002) with the remaining categorizations of irritability showing no direct association with the TEC-M's overall measure of ER ability. This finding is interesting in showing on the one hand that the TEC-M is relevant to actual clinical manifestations of the type of behavior it is intended to measure, practically adding to its validation, and on the other hand showing the diversity in representations of disruptive behavior, as not all types of irritability shared the same potentially underlying ER deficit.

5.3 The emotional component in TS

As evident from the systematic search presented in the introduction (Table 1), subtle impairments in social cognition have been reported in individuals with TS, though the majority of studies accepted varying comorbidity and medication and were carried out in small samples. The few studies of emotional processes in TS-alone have indicated only mild if any impairments compared to controls. The same picture emerged when looking into the literature on prevalence of rage attacks. Children with TS-alone did not present with a higher occurrence of rage attacks than controls, but displayed a wide range of behavioral and emotional problems when tics were

accompanied by one or more comorbid disorders. ER, however, has not been studied in children with TS, although the phenomenological similarities between rage attacks and tics warrant a more exhaustive investigation of the connection.

In the present thesis, we employed two tests of ER addressing different aspects of regulation. An examination of different types of ER could potentially reveal underlying differences in patterns of regulation in individuals with TS with comorbidity and without. Regulation assessed with the TEC-M can be characterized as spontaneous and unguided, as children were only instructed to solve puzzles and were under no regulations regarding further behavior. In the second test, children specifically practiced the strategy of reappraisal and received instructions and guidance from a test administrator. The latter setup substantially narrowed the behavioral outcome by tapping a type of regulation that is qualitatively different from responding naturally to frustration in the presence of a parent. In relation to the process model of emotion regulation (Gross, 2014, 2015) and the associated conceptualization of ER, the boundaries for solving the Tangram Construction Task may include up to four out of the five groups of strategies (situation modification, attentional deployment, cognitive change, and response modulation) as evident from the scoring sheet (Figure 5). The task may further include the first process of situation selection if a child should refuse to participate in the task or choose to leave during the task. Displays of ER may occur automatically or in a controlled manner and thus allow for a broad look into specific profiles of ER. Conversely, the PRT is directed at only one group of the process model, namely cognitive change, but may also include strategies within attentional deployment and response modulation; the former exemplified in a child automatically looking away from an aversive image or simply ignoring the element causing discomfort by using suppression, and the latter in a child trying to calm oneself after viewing an unpleasant picture. In the following, results from papers 2 and 3 will be discussed as respectively unguided and guided regulation.

5.3.1 Unguided emotion regulation

As expected, children with TS were not impaired on ER ability represented by the EmReg score, and severity of tics, and presence of PU's were not associated with ER as measured with the TEC-M (paper 2). Children with ADHD on the other hand presented with significantly lower scores of ER than the control group and the TS group, in line with hypotheses and previous findings.

We performed an EFA based on the assumption that the latent structures in the observable variables of the TEC-M would reveal components that were not observable on their own and that factors would relate to underlying psychological processes that contributed to the variation in data (Bosten et al., Yong & Pearce). This analysis resulted in three factors for the child items of the TEC-M, of which groups differed significantly on the factor 'emotional control'. This factor consisted of positive loadings from control (structuring the task and taking initiative) and the EmReg score and negative loadings from seeking assurance with a parent, avoiding the task, or producing verbal or non-verbal negative expressions. Similar to results on the EmReg score, there was no significant difference between children from the TS groups and the control group. Children with ADHD or TS + ADHD presented with significantly lower emotional control than children from the control group, and the TS + ADHD group scored significantly lower than

children from the TS group. Whereas groups differed on the EmReg score, emotional control, and parental tension as mentioned above, the remaining items did not differentiate the groups. Thus, specific profiles of ER did not emerge that could contribute to the understanding of ER difficulties in some children with TS, besides examining their comorbidity status.

Results from the TEC-M corroborated the apparent connection between dysregulated behavior and ADHD, by not only showing group differences compared to controls and children with TS on a categorical level, but also by showing a dimensional relationship between symptom severity, as measured with the ADHD-RS, and the EmReg score. The more severe inattention or hyperactivity/impulsivity, the lower the EmReg score, i.e., the poorer the regulation ability.

5.3.2 Guided emotion regulation: Cognitive reappraisal

On our measure of regulation success following the guided use of cognitive reappraisal, groups did not differ (paper 3). This was expected for the TS and control groups, but was surprising in regard to the ADHD groups, possibly indicating a potential for learning ER strategies even in children with attention problems, in whom dysregulated behavior is often part of the clinical manifestation. Choice of strategy within the framework of reappraisal was coded as one of nine potentially effective strategies with extra options for coding strategies resembling suppression, a failure to reappraise, or children stating that the picture did not cause discomfort. Choice of strategy did not differ across groups. In the only study of ER in individuals with TS, adults with TS reported more frequent use of suppression than controls, but use of reappraisal did not differ between groups (Drury et al., 2016). This distinction was not evident from the present work in children.

Looking beyond groups, the results of paper 3 demonstrated the effectiveness of cognitive reappraisal and a tendency for reappraisal ability to improve with age. Although groups did not differ in choice of strategies applied, the individual strategies differed greatly in regard to their association with final ER success. The most widely applied reappraisal strategy involved a reinterpretation of the depicted circumstances to decrease the experience of negative effect. This strategy was executively demanding, as children had to come up with an authentic reinterpretation and verbalize it in only 10 seconds, while the image appeared on the screen. This strategy additionally showed the highest correlation with ER success. The fact that the seemingly most cognitively demanding task resulted in the best regulation through reappraisal may hint to the connection between emotional and executive functions by demonstrating that higher cognitive involvement is adaptive to the ER process.

Although reappraisal was effective across all participants, the majority of children found it difficult to reappraise and ended up relying on the suppression-like strategy which was less efficient. Thus, an explanation of the lack of group findings could be that children were too young to fully utilize the strategy of reappraisal, and that potential group differences would have emerged more clearly in older groups.

5.3.3 Comparing group performances across types of emotion regulation

The comparison of findings across tests is of substantial interest to the understanding of the emotional component in TS, because the same groups of children perform differently on tasks of spontaneous ER and guided ER, respectively. This discrepancy is interesting to the theoretical understanding of ER and to the understanding of ER ability in these groups of children. Looking at the results from the PRT (paper 3) in isolation, one may argue that the children with ADHD represent an atypical group because they are capable of reappraising similarly to controls. The same group, however, is challenged when it comes to regulation their emotions spontaneously, thereby creating a dissociation between different aspects of regulation. This discrepancy is not strange when considering the complexity of the concept of ER. For the TS group, the fact that they perform similarly to controls on both measures of ER is an indication of their intact ER abilities when they do not have a co-existing diagnosis of ADHD. The heterogeneity of TS and ADHD is also relevant to consider in relation to the findings. With the diagnostic system consisting of various symptoms for each disorder and requirements to present with a fixed number of symptoms out of a larger group, clinical manifestation may differ widely within the same diagnosis (Gross & Jazaieri, 2014). Two children from the ADHD group may be markedly different in terms of specific symptoms, impairment, and comorbidity, thus compromising the grounds for comparison.

5.4 A transdiagnostic perspective on emotion regulation

Current systems of classifying mental disorders, such as the DSM-5 and the ICD-10, are built on the presence or absence of symptoms specific to distinct psychopathological entities (Fernandez et al., 2016; Stephan et al., 2016). This categorical view does not encompass the underlying dimensionality of mental disorders, which can result in widely differing clinical manifestations between individuals within one disorder in regard to subjective suffering, impairment, and symptom severity (Fernandez et al., 2016; Krueger & Eaton, 2015; Stephan et al., 2016). Nor are classifications unequivocally supported by findings from genetics and neuroscience (Insel et al., 2010). Symptoms often overlap between disorders and "comorbidity is the rule, rather than the exception" (Krueger & Eaton, 2015, p. 27). As previously seen with the case of irritability, this symptom spans at least six diagnoses (Stringaris, 2015), suggesting that various mental disorders are manifestations of relatively few, underlying dimensions (Krueger & Eaton, 2015). Although classifications are helpful to the understanding of psychological problems and may serve as a starting point for treatment efforts, the separation of diagnostic entities entails an associated division of expertise, research, and treatment, which may benefit more from adopting a wider perspective (Fernandez et al., 2016).

Recently, a focus on transdiagnostic factors has become increasingly popular to the understanding of processes that are involved in the development and maintenance of psychopathology and which apply to multiple mental disorders (Aldao, Gee, De Los Reyes, & Seager, 2016). The basic division in internalizing and externalizing disorders represents an early approach to a transdiagnostic perspective (Krueger & Eaton, 2015) and more recently, the Research Domain Criteria (RDoC) project has developed a new framework for conceptualizing mental disorders (Insel et al., 2010). The RDoC project represents a novel approach to the understanding of mental dis-

orders. It is not intended to replace current diagnostic systems, but rather to supplement the understanding of the spectrum from mental health to mental illness, by seeking to describe all basic functions underlying behavior in units of analysis spanning from genes and brain circuits to behavioral paradigms and self-report (Research Domain Criteria, RDoC, 2019). Given the role of ER and dysregulated emotional behavior in numerous disorders of mental health, ER has recently been suggested as an additional domain in the RDoC matrix (Fernandez et al., 2016).

The research in affective processes in psychopathology has been carried out under the label clinical affective science which has as its main focus the role of ER in the development and maintenance of various types of psychopathology (Aldao, 2016). Recent studies have found that the use of maladaptive strategies predicted psychopathology more than the relative use of adaptive strategies was related to mental well-being (Aldao et al., 2010; Martins, Freire, & Ferreira-Santos, 2016). The use of adaptive ER strategies, such as problem solving and acceptance, has been hypothesized to be a protective factor against psychopathology, and maladaptive ER strategies have been implicated as risk factors (Aldao et al., 2016). Risk factors and treatment in particular are relevant in a transdiagnostic perspective as they often apply to more than one disorder (Fernandez et al., 2016). A dimensional and transdiagnostic approach to impairments may further prove a more efficient guide for treatment than a categorical approach (Stephan et al., 2016).

Although ER is not (yet) an official RDoC domain it is well in line with the objective of the RDoC to conceptualize ER as a transdiagnostic factor. The role of ER in multiple types of psychopathology has been well-established in the present thesis, but prior research has predominantly been carried out in units of analysis other than the behavioral domain. The hope for the TEC-M is for this instrument to contribute to a transdiagnostic approach to mental disorders, by adopting a generic, diagnosis non-specific option for assessing ER abilities objectively by clinicians across disorders (paper 1). The TEC-M adopts a dimensional approach to the assessment of ER by grading level of impairment as opposed to the binary approach of establishing either presence or absence of dysregulated emotional behavior. This approach is in line with recommendations in the field of ER (Aldao, 2016; Fernandez et al., 2016).

The collective findings from papers 2 and 3 suggest that a dimensional account of ER is not directly relevant to TS. ER is an exceedingly relevant transdiagnostic factor to the vast majority of psychiatric disorders, and the finding of subtle deficits in TS in tasks of social cognition justifies further clarification. The results from the experimental tasks of ER in isolation, however, do not support ER deficits as part of the central TS symptomatology.

5.5 A dimensional approach to Tourette syndrome

In line with a transdiagnostic perspective on mental disorders, researchers and clinicians have increasingly started to challenge the unitary classification of TS by offering new perspectives to the understanding of tics and their relation to varying accompanying symptom manifestations (Robertson, 2015b). Although the only symptoms required for a diagnosis of TS are tics, the well-established associations between TS and other psychiatric disorders, illustrated by comorbidity rates of 90%, initially led to an unofficial subdivision of TS in pure TS or TS-alone

(representing TS without comorbidity) and TS+ (representing TS with mixed comorbidity). Experts in the field have proposed clinical and neurological heterogeneity in TS and emphasized that the frequent variation in clinical presentation and comorbidity renders the entire diagnosis controversial (Robertson, 2015b, 2015a). This heterogeneity has been assessed in factor analytic studies with TS. One study assessed clusters emerged from data from 638 children and adults with TS and found three subtypes, explaining less than half the variance in total, but pointing to TS as a phenotypically heterogeneous disorder (Cavanna et al., 2011). Emerged subtypes included patients with complex motor tics and echo-/pali-phenomena, patients with attention problems, hyperactivity, and aggression, and patients with complex vocal tics and coprophenomena (Cavanna et al., 2011). A more recent study reached the same conclusion of widespread heterogeneity in TS by applying hierarchical ascendant clustering to a dataset including information on tics, comorbidity, and developmental milestones for 174 children and adolescents with TS (Cravedi et al., 2018). This analysis likewise resulted in three groups, with different phenotypes than the previously described factor analytic study, however. Individuals in the first group presented with complex phenotypes comprising varying comorbidity and a history of pre- and perinatal complications. The second group manifested as pure TS with patients presenting with only the core symptoms of TS. Finally, the third group revealed a less familiar subtype of children with school problems relating to both ADHD, nonspecific attentional problems, writing problems, and early contact with the healthcare system. Interestingly, severity or onset of tics did not differ significantly across emerged groups, and OCD was frequent in all three groups; possibly indicating a shared genetic background between TS and OCD (Cravedi et al., 2018). The authors suggest that the classification of TS should encompass a grading from simple to complex TS in the future.

Findings regarding the shared genetic etiology between TS and other psychiatric disorders have been inconclusive. The finding that a comorbid diagnosis of ADHD was associated with more ecco- and copro-phenomena in individuals with TS (Robertson et al., 2015) could indicate a shared genetic background between the two disorders. This account was contradicted by a review of findings rejecting the hypothesis of a shared genetic etiology (Banaschewski, Neale, Rothenberger, & Roessner, 2007). More recent accounts, however, have supported the view of shared genetic risks contributing to much of the cooccurrence in neurodevelopmental disorders (Thapar & Rutter, 2015) and specifically TS, ADHD, OCD, and disruptive behavior (Hirschtritt et al., 2015).

The question remains whether TS is best represented by continuing with the current diagnostic criteria of motor and vocal tic(s), or whether a diverse description of subtypes within the syndrome would provide a more optimal depiction. In a disorder with only 10% of individuals presenting solely with tics, it is difficult to decide whether it is more meaningful to look at TS-alone, or whether studies should accommodate the diverse clinical representations and examine TS in samples with mixed comorbidity. In an area that has not been studied experimentally, we believed it to be an important first step to decipher difficulties in ER as relating to either TS, ADHD, or a combination, to enable a reflection of potential diagnosis-specific difficulties. From the present findings, we acquit the children with TS of being impaired in their ER ability, but this does not change the fact that many children with TS continue to have as their primary concern the uncontrollable rage attacks – regardless of whether these outbursts are related to

comorbid ADHD. The group differences found with the TEC-M (paper 2) demonstrate that ER difficulties are present in the ADHD group and the group of children with TS + ADHD, indicating a need for thorough assessment of symptoms and comorbidity, when children first present with tics, to ensure investigators are aware of behaviors associated with ADHD and are able to provide support at an early point. With at least half of the children with TS presenting with comorbid ADHD (Banaschewski et al., 2007), emotional dysregulation remains an area of relevance to children with TS; only now we have further indications that the dysregulated behavior is not related to presence or severity of tics.

5.6 Methodological considerations

5.6.1 Strengths and limitations

The strengths of the present work include a relatively large group of children with TS and a matched control group recruited through the Danish registers. Further, in line with recommendations in the field (Aldao, 2016), the inclusion of clinical contrast groups of children with ADHD and TS + ADHD add to the specificity of findings by enabling an examination of whether findings are generally associated with child psychopathology, or whether they are restricted to a specific diagnostic entity. All children in the study were medication-naïve, and thus any potential effects of medication did not interfere with their performance. Children's diagnostic status was thoroughly assessed through a multi-method approach, which systematically assessed accounts of behavior through a semi-structured diagnostic interview with the child and the parent separately, as well as through questionnaire-based parent- and teacher reports of behavior. Finally, the present study applied an experimental, ecologically valid measure that allowed for a direct assessment of ER instead of relying solely on information gathered from questionnaires.

A number of limitations were present in this work. Besides the TS group and the control group, groups differed in size, and the small clinical contrast groups may have resulted in less power to detect differences between groups, and thus more type II errors. The clinical presentation of the TS + ADHD group was interesting, as the addition of two diagnoses did not lead to a higher level of impairment. On the contrary, this group presented with higher IQ and resembled the control group the most in terms of regulation success in paper 3. These findings could almost hint to a protective capacity of the diagnosis of TS, although the small group size severely limits any general statements.

Throughout the study, we decided not to include IQ as a covariate in statistical analysis, although the groups differed significantly on this measure. It is well-established that lower IQ scores are part of the presentation in ADHD (Frazier, Demaree, & Youngstrom, 2004). Thus, statistically controlling for a feature, which is inherently part of the symptomatology in any neuropsychological tests, would lead to a misrepresentation of that group. Whereas the link between TS and IQ has not been fully established, it generally shows that children with TS, who do not have a comorbid disorder, have the same level of IQ and rate of learning disabilities as children without neurodevelopmental disorders (Pruitt & Packer, 2013), as we also find in the present sample. Matching the ADHD group on IQ to that of the TS group and the control group would result in unrepresentative groups, as the ADHD group would present with higher

IQ than their population mean, and the control group would have IQ scores below the norm (Dennis et al., 2009). Studies have been inconsistent regarding the role of SES in the moderation of children's inherited intelligence (Hanscombe et al., 2012). We wanted to avoid over-correcting the sample and potentially correcting for IQ by proxy, and thus, SES was not controlled for.

The presence of comorbidity in 14 children from the TS group was both a strength and a limitation. The comorbidity limits the degree to which we can comment on children with TS-alone. On the other hand, and as mentioned above, only 10% of children with TS present with TS-alone, and thus a TS-alone group is not representative of the general population of individuals with TS. Though comorbidity was present in some children, only four children received a diagnosis of ODD, which could be associated with a vulnerability to dysregulated behavior. The remaining children presented with OCD or anxiety disorders. We excluded children with severe comorbidity such as psychosis or autism, but accepted other types of comorbidity and still ended up with a relatively "pure" TS group, which enabled potential differences to emerge through the inclusion of the clinical contrast groups.

The procedures for choosing parametric versus nonparametric tests were not followed consistently across tests and papers. For every test, assumptions were checked, and a violation of normality for example, would lead to the choice of a nonparametric test. At other times, however, a test has been deemed robust to the violations, such as running parametric tests in the final two papers due to the increase in sample size compared to paper 1. Although this inconsistency does not appear to change findings, the procedures regarding choice of test ought to have been followed consistently.

A major limitation mentioned in all papers was the lack of blinding, as the assessor (JH) also carried out many of the clinical assessments. Studying children with tics further complicates blinding procedures, as tics would sometimes be noticeable from the videos. To accommodate the overlap in assessments and codings, all videos would be placed randomly with only the identification number visible so that there would be no clues regarding the diagnostic status of the child. These codings were carried out several months after seeing the participants, and for many children more than a year would have passed. Thus, in practice the diagnostic affiliation for the majority of participants was not clearly evident, and, naturally, every effort was made to disregard diagnostic clues in the coding process.

Finally, the cross-sectional nature of the study hinders a clarification of whether our findings represent transitory phenomena or stable traits.

5.6.2 Reflections on selection bias

When conducting studies in which participants are to represent different populations, groups should be representative to enable generalizability of findings to the total population of individuals that the samples represent. In the present study, we did not systematically document information about the individuals who did not wish to participate and thus cannot rule out any selection bias. We did, however, include a large group of children with TS to ensure representativeness, and recruited an equally large group of typically developing, matched controls.

Although we did not match on IQ and socioeconomic status (SES), the TS group and the control group presented with almost identical means on these measures.

One could argue that recruitment through a pediatric clinic with a nationwide responsibility for the care and management of children TS would result in patients having more pronounced difficulties than children from primary or secondary care. We did not, however, see the otherwise well-established high rates of comorbidity with ADHD, as many of the referred children presented with pure TS, which does not speak to a selected group in terms of severity. We chose to recruit typically developing children through the registers to reduce selection bias by receiving a randomly generated extract of children. This, however, does not eliminate selection bias. For the TS sample and the control sample in particular, there could be a bidirectional bias in participation. On the one hand, families with the possibility of prioritizing their child's participation in research, even when this entailed two full days of neuropsychological testing, might be a selected group beforehand in terms of better resources. On the other hand, the TS and the control groups may include children whose parents suspect the child's behavior to lie within a psychiatric spectrum and who want to explore this in an unofficial, yet standardized context. As the children with TS were recruited from a pediatric clinic, a psychiatric assessment was not part of their clinical routine. This was not the case for the children with ADHD who were already in the child and adolescent psychiatric outpatient clinic with a referral of ADHD and where the choice of whether to enter a research project only had a minimal effect on the procedures of assessment.

5.7 Clinical implications

Disruptive behavior in children is one of the most frequent causes of mental health referrals and common to numerous psychiatric disorders, emphasizing a need for a better understanding of the basis of dysregulated emotional behavior and improved methods of treatment. Impairments in ER ability appear to show resistance to treatment (Rosen et al., 2018) and become increasingly resistant with age, thus emphasizing a need for early identification of these difficulties (Lewis et al., 2006). A first step in managing disruptive behavior will most often entail an assessment of comorbidity requiring intervention, to see if rage attacks will diminish consequently. Psychosocial interventions such as anger control training and parent managing training have proven effective in treating disruptive behavior (Piasecka et al., 2018). A recent group intervention program for managing frustration in children with ADHD has also shown promise in decreasing ER deficits and difficulties with externalizing behavior (Rosen et al., 2018).

Knowledge is still lacking about the cause and nature of impairments in ER and their associations with disruptive behavior. In TS, rage attacks may be viewed as types of tics, as reactions to exhausting days of suppressing tics, or as stemming from comorbid disorders. Further, they may be conceived as central to the child's diagnosis, stemming from the emotional consequences of living with a mental disorder, or as unrelated to psychopathology as mere manifestations of individual variation in the typical development of ER. Considering the confusion surrounding the concept of emotional dysregulation, a transdiagnostic approach is beneficial to supersede the boundaries of diagnoses and ensure a focus on ER difficulties regardless of diagnosis. Recently, Fernandez and colleagues (2016) have encouraged clinicians to focus on ER as a factor that may

maintain or exacerbate maladaptive behavior instead of relying on diagnostic classifications in the understanding of psychopathology. Specifically, clinicians should specify which part(s) of ER the child is struggling with, such as the identification of a relevant goal, the correct selection of an appropriate strategy, the implementation of the strategy in question, or the continued monitoring that is required to modify strategies according to subjective goals. Such an approach provides targeted points for intervention, which may benefit the specific ER impairment as well as aspects of psychopathology that were potentially maintained by the impairment (Fernandez et al., 2016).

While these points are immensely valid to enhance the understanding of ER difficulties and appropriate interventions, they may prove difficult to implement in the clinical practice. In this regard, the TEC-M may provide a useful, quick, and simple supplement to assessing ER difficulties by providing a starting point for identifying the process(es) in need of intervention and further gaining insight into the interplay between the child and the parent. The parent may play a role in the maintenance of children's dysregulated behavior, which may further maintain psychopathology. With the TEC-M, parents' response to the child's dysregulated emotional behavior can be examined, e.g., whether the behavior is rewarded or punished. Furthermore, videos may later on provide illustrative examples of adaptive and maladaptive behaviors related to the child, the parent, and their dynamics. The task may also serve as a method of evaluation prior to and following a therapeutic intervention.

Our findings that children from the ADHD groups were capable of learning and applying the strategy of reappraisal equally to controls (paper 3) was surprising and holds substantial clinical relevance by illustrating a potential for training specific ER abilities. Conversely, and of equal clinical importance, the same groups of children were impaired in spontaneous ER (paper 2), which underscores previous accounts of emotional dysregulation in ADHD. This finding points to a need for examining potential difficulties in ER early in the assessment of children with ADHD, but also of children with TS who may already have a comorbid diagnosis of ADHD and who are in any case vulnerable to developing attentional problems. Furthermore, special attention should be given to scores of irritability in diagnostic screenings, as we have previously demonstrated an association hereof with ER difficulties. The opposite may also be relevant, namely to maintain an awareness of signs of ODD when the score on the TEC-M indicates poor ER abilities.

Finally, the present findings are relevant to understanding the associated behaviors that many parents and some clinicians attribute to TS. One may argue that terminology and causation are irrelevant and that clinicians should merely deal with the behavior at hand. Some parents may even find comfort in attributing the presence of rage attacks to TS, thereby explaining the hugely distressing behavior as something that is caused by a well-known condition and not by family dynamics, for example. The demarcation of TS and emotional dysregulation is relevant, however, to counteract nosological misconceptions and keeping an evidence-based approach to diagnostic entities. Studies, including the present, have repeatedly demonstrated that difficulties with ER are not an inherent part of TS without comorbidity, and this message is important to convey to individuals with TS and the clinicians helping them. Instead of accepting certain characteristics as a natural part of TS, it is important to know that there is no apparent direct link between TS-alone and emotional dysregulation and that the presence of such in children with

TS should prompt an assessment of comorbidity as well as other factors potentially underlying the behavior.

5.8 Implications for research and recommendations for future studies

Applying a transdiagnostic approach is as relevant to research efforts at it is to clinical approaches. By adopting a dimensional and flexible perspective to symptoms, facets of psychopathology may emerge that would not be evident from a categorical perspective, in line with the RDoC approach (Aldao, 2016; Fernandez et al., 2016). We believe the TEC-M could contribute to this endeavor and have identified a number of ideas in the process that would be ideal to include in future studies of ER in children.

An obvious addition to the TEC-M is the inclusion of physiological measures before, during, and after performing the task. Physiological measures do not provide information about the individual threshold for emotional stimuli, but they could compliment behavioral findings by providing information on change in bodily measures of frustration from resting state to task solving. As part of another study, we have recently included measures of HRV in children while participating in the Tangram Construction Task to see how these measures may contribute to the understanding of individual reactions to frustration. Another point, which has also recently been included in the mentioned study, was to include a visual analog scale as a subjective measure of frustration to relate to the behavioral manifestation. Generally, given the complexity of the concept under investigation, the more measures of control that can be incorporated, the better. Specifically, children could report how much they felt they were being evaluated, and how much they generally experience to be in control of their feelings (Bunford et al., 2018; Gutentag et al., 2017). Building on the theoretical validation of the TEC-M, it would be interesting to see if scores on this instrument could prospectively indicate emotional problems later in life, thereby providing support to its potential role in estimating the need for early intervention.

The inclusion of additional clinical contrast groups would profit the clarification of potential diagnosis-specific difficulties and would elucidate how difficulties with ER manifest across child psychiatric disorders. Considering the heterogeneity that is evident within multiple types of psychopathology, diagnostic categories could be further subdivided; for example the inattentive versus hyperactive/impulsive subtype in ADHD or a primarily obsessive contra compulsive nature in OCD, although this would require a substantial number of participants in each group. The inclusion of a self-report questionnaire of ER abilities would contribute to a multi-method approach. In the present study, we did include a self-report measure administered by a clinician, but unfortunately had to discard these data, as children had not been able to fully understand the questions, thus rendering answers invalid. Future studies including self-report questionnaires with children should ensure their appropriateness for the age group in question by piloting them before commencing the study. The well-established yet still unclear relationship between ER ability and executive functions could easily be pursued further to increase the understanding of the executive component in ER and how separable the concepts are.

The lack of difference between children with TS and children from the control group on ER and cognitive measures would be interesting to explore further. While the present findings, and multiple studies before this, do not find individuals with this particular neurodevelopmental disorder to perform (markedly) differently to typically developing controls, an interesting aspect to address are potential underlying differences, that do not manifest on a behavioral level. Adding an fMRI layer to cognitive and emotional tests would potentially reveal a compensation on a neural level by employing more network activity resources than controls to reach the same behavioral level. This hypothesis is in line with the view that individuals with TS perform well on measures of cognition because they constantly train inhibitory abilities through tic suppression.

Finally, a longitudinal perspective to ER abilities in children would be the ideal goal to pursue and something we hope to implement later on. Much could be gained from following patterns of ER, neuropsychological profiles, and psychopathology over time, ideally premorbidly, to see how they each co-develop. Hopefully, a longitudinal approach could aid the identification of specific points for intervention targeted the various domains of ER, neuropsychology, and psychopathology, respectively. An exploration of ER over time would additionally allow for a characterization of this ability as more state-like or trait-like, considering the natural developmental trajectories.

6. Conclusion

The present thesis posits that children with TS-alone do not appear to be impaired on experimental measures of ER ability. Children with TS + ADHD and children with ADHD show difficulties in spontaneous regulation, but not in ER when using the strategy of reappraisal. This latter finding is relevant to therapeutic approaches to dysregulated behavior in demonstrating a potential for learning ER strategies under guidance. In support of a dimensional examination of ER, we offer a novel, validated approach to this end, the TEC-M. The aim for the TEC-M is to function as a measure of ER in research as well as in clinical settings. ER is a multifaceted concept and the present findings indicate that ability in different facets of the domain can vary within individuals. ER may not be a relevant transdiagnostic factor to study in relation to the 10% of individuals with TS-alone. However, for the remaining 90% with comorbidity, widely faceted clinical presentations, and potentially accompanying disruptive behavior, ER should be carefully assessed to hopefully enable interventions targeted dysregulated emotional behavior before any long-term effects manifest. The ultimate objective is the alleviation of impairing explosive outbursts that cause substantial distress for children and their families. The present thesis offers a first step to a better understanding of the role of ER in TS and ADHD, with future research needed to benefit treatment efforts in the long term.

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A. Paper 1

The Puzzle of Emotion Regulation – Development and Evaluation of the Tangram Emotion Coding Manual for Children.

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Manuscript in review.

The Puzzle of Emotion Regulation: Development and Evaluation of the Tangram Emotion Coding Manual for Children

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Abstract

The ability to regulate one's emotions is crucial to engaging successfully in social contexts and difficulties in emotion regulation are seen in multiple psychiatric disorders, prompting an increased interest in the concept. Suitable methods for assessing emotion regulation, however, are lacking. In this study, we investigated the interrater and intrarater reliability, construct validity, and content validity of a new observational method of evaluating children's emotion regulation abilities (a complex puzzle task) in a sample of 62 children without psychiatric disorders and 23 children with attention-deficit/hyperactivity disorder (ADHD) aged 7-12, using intra-class correlation coefficients for the reliability analyses and Spearman's rank-order correlations for analyses of convergent and discriminant validity. A panel of experts examined the content validity of the test, and the authors used Mann-Whitney U-tests to investigate the ability of the test to differentiate the non-clinical group from the ADHD group. Results showed a high level of inter- and intrarater reliability of the test. There was mixed evidence for convergent and discriminant validity as expected due to the novelty and experimental nature of the test making it difficult to compare with questionnaire-based measures. Content validity analysis was satisfactory, and the group comparison showed that the test differentiated the groups on the primary outcome measure. Overall, the measure demonstrated high feasibility and satisfactory psychometric properties. The generic nature of the test makes it suitable for use across psychiatric disorders and age groups with potential relevance in both research and clinical settings.

1 Introduction

Interest in the concept of emotion regulation (ER) has increased markedly over the years making it one of the fastest growing areas in the field of psychology (Gross, 2015a). ER is an essential concept in psychological research and crucial for understanding the dimensions of mental illness (Cole et al., 2004; Gross, 2015a/b; Gross & Thompson, 2007; Thompson, 1994), due to its pertinence on a continuum from normal development to severe psychopathology throughout life. Specifically, the ability to adequately regulate emotional responses is crucial for development and for the capacity to engage in a social context (Lewis et al., 2006). Also, disturbances in the development of self-regulatory control (including ER) most likely play a part in the development of a wide range of psychiatric disorders (Marsh et al., 2009) and emotional dysregulation is involved in a great number of both externalizing and internalizing disorders, such as attention-deficit/hyperactivity disorder (ADHD; Aldao et al., 2016), anxiety, and depression (Klemanski et al., 2017). Finally, a key component in the growing interest in ER is its potential role in a dimensional approach to psychopathology (Aldao, 2016; Fernandez, Jazaieri, & Gross, 2016), exploring pathological processes in a transdiagnostic perspective to disentangle which impairments span multiple disorders and which are disorder-specific (Aldao, 2016). Despite the relevance and popularity of the concept, various definitions of both emotions and their regulatory processes exist in the vast literature. One widely used definition describes ER as the attempt to influence the experience and expressions of one's emotions through the activation of instinctive or explicit goals which impacts the modification of an emotional response (Gross, 2015a/b; Gross & Jazaieri, 2014). Despite the growing interest in the field, research on the subject is sparse, possibly due to a lack of proper behavioral instruments of assessment (Lewis et al., 2006). A number of instruments for the assessment of ER exist, but these measures are either developed for younger children or adults or rely on indirect measures, such as parent-reported questionnaires. While self-report measures exist in several versions and remain the norm (Robinson & Clore, 2002), this type of measure is far from ideal when examining ER (Zelkowitz & Cole, 2016), as self-report relies on the metacognitive abilities of the informant and is known to be affected by mood. Various neurobiological ways of investigating ER exist as well; however, more precise experimental measures are needed that allow for direct observation of the child's behavior in a naturalistic setting to assess ER ability or screen for the lack of it.

We thus developed the Tangram Emotion Coding Manual (TEC-M) to contribute to the field with a clinical instrument to evaluate children's ER profiles within the framework of parent-child interactions. The primary outcome of the test is the child's overall emotion regulation ability, but other facets such as communication of emotions, specific regulatory strategies, and parental behavior are part of the instrument as well. The inspiration for this manual was found in the Tangram Construction Task Manual (Esbjørn et al., unpublished manuscript), which expands on previous methodology in the field of child anxiety (Tangram Task; Hudson & Rapee, 2001; 2002) and attachment relations (Co-Construction Task; Steele et al., 2007). The current instrument, however, is based on the process model of emotion regulation (Gross, 2015a; Gross & Jazaieri, 2014), which is a widely used framework for understanding and organizing ER. The process model of emotion regulation centers on five sequential processes of ER, namely "situation selection", "situation modification", "attentional deployment", "cognitive change", and "response modulation" (Gross, 2015a; Gross & Thompson, 2007) that may range from adaptive to maladaptive depending on the specific individual, the context, and the emotions in question (Gross & Jazaieri, 2014). Situation selection refers to choosing whether to engage in a given situation depending on the emotion that it is likely to elicit, whereas situation modification refers to directly altering the physical environment in a given situation. Attentional deployment captures the possibility of directing attention towards or away from specific stimuli to influence one's emotions. Cognitive change refers to how individuals appraise a situation to alter its emotional significance. Finally, response modulation occurs late in the emotion-generative process and captures the individual's

experiential, physiological, or behavioral responses to the situation eliciting a given emotion (Gross & Jazaieri, 2014). This model shaped the development of the coding manual by elucidating specific aspects of the regulatory process that may occur during regulation of emotions, thus providing a framework for items to be included. This allowed for a combined theory- and data-driven process of item generation, in which we included items representing observable aspects of each process.

As part of the validation of the instrument we investigated the construct validity (comprising convergent and discriminant validity) of the TEC-M, which reflects the degree to which an instrument measures what it purports to measure (Strauss & Smith, 2009). Convergent validity deals with the association between the test under investigation and a theoretically related variable (Evans et al., 2016), whereas discriminant validity is supported by the finding of smaller or non-existing associations with scores from instruments that are not directly theoretically related (Evans et al., 2016), thus supporting the uniqueness of a new measure (Holton et al., 2007). Several studies have suggested a link between executive functions and ER. From a neural perspective, both tasks require activation of the frontal lobes and one hypothesis states that executive planning strategies interact with the individual's underlying affective characteristics to facilitate successful ER (Fox, 1994). Zelazo and Cunningham (2007) suggested that the conscious and voluntary form of ER simply is an executive task making the two constructs isomorphic. Based on the association between executive functions and ER, we hypothesized that the Emotional Control Scale from the Behavior Rating Inventory of Executive Function (BRIEF; Gioia et al., 2000) would correlate well with the overall ER outcome of the instrument. Additionally, we chose the Deficient Emotional Self-Regulation Profile (Biedermann et al., 2012; Spencer et al., 2011) from the Child Behavior Checklist (CBCL; Achenbach & Rescorla, 2001) and expected a similar correlation, as this scale reflects elevated scores on a number of symptoms assumed to reflect general dysregulation of emotions. We chose IQ for the analysis of discriminant validity. Based on the nature of the test we expected some correlation between ER and the intellectual abilities of the child. Previous studies have demonstrated associations between children's parent-reported ER skills and later academic success (Graziano et al., 2006) as well as ER abilities and kindergarten achievement (Howse et al., 2003). One study with adults, however, reported only weak correlations between IQ and emotional intelligence scores (Checa & Fernández-Berrocal, 2015). We thus expected some overlap in IQ and ER as measured with the TEC-M, but assumed this association weaker than that between ER and theoretically related questionnaire measures.

The main objectives of the present study were to describe the development of a new clinical instrument that accommodates the need for observational, clinician rated evaluation of children's ER profiles and to assess the inter- and intrarater reliability of the instrument as well as its convergent, discriminant, and content validity. Finally, we examined the instrument's ability to differentiate emotion regulation profiles in a group of children with ADHD from a typically developing control group. Children with ADHD are known to present with dysregulated behavior (Graziano & Garcia, 2016) and we were interested in whether the test would be sensitive to this. We hypothesized that the group of children with ADHD would achieve lower scores of overall emotion regulation ability.

2 Methods

2.1 Participants

Participants were 85 children aged 7-12 (M = 9.42, SD = 1.36). The vast majority lived in the Capital Region (N = 77) with the remainder living in other regions of Denmark. Of the 85 included children, 62 were healthy controls and 23 children had a diagnosis of ADHD, eight of which of the predominantly inattentive type (Table 1). Five children with ADHD additionally met diagnostic criteria for

oppositional-defiant disorder (ODD). The gender distribution showed a predominance of boys due to the fact that the majority of control participants were matched to a population of children with neuropsychiatric disorders presenting with a natural preponderance of boys. All participants underwent a thorough clinical assessment with the Kiddie-Schedule for Affective Disorders and Schizophrenia, Present and Lifetime Version (K-SADS-PL; Kaufman et al., 1997). Control participants were excluded if they met DSM-IV (American Psychiatric Association, 2000) criteria for any lifetime psychiatric diagnosis with the exception of enuresis and encopresis. Children with ADHD were excluded if a comorbid autism spectrum disorder or psychotic disorder was present. All participants were Caucasian except for one participant of South Asian descent and no participants were taking or had previously been taking psychotropic medications.

Participants were drawn from two larger studies carried out in the Mental Health Services, Capital Region of Denmark. Twenty-one children with ADHD and all children aged 8-12 (N = 70) were part of a study investigating ER in children with neuropsychiatric disorders, whereas the remaining 13 control participants and two participants with ADHD were part of a control group of seven-year-old children from a high risk-study (Thorup et al., 2015). Children with ADHD were recruited from the local outpatient clinic when referred for diagnostic assessment and before starting any treatment, with the exception of two children with ADHD who were initially recruited as controls, but met criteria for a diagnosis during the assessment. We chose to include participants from two separate studies to increase the age span. Participants were recruited from 2013-2016. All healthy controls from the two studies were randomly selected and recruited via the Danish Civil Registration System (Pedersen, 2011) to reduce selection bias.

2.2 Measures

2.2.1 The Tangram Construction Task

The Tangram Construction Task is a five-minute test that is administered with the child and one caregiver present. The test has been carried out successfully with children from the age of seven to twelve, but could potentially be used in both younger and older age groups. During the five minutes, the child is instructed to solve as many puzzles as possible from a total of six puzzles. The parent is instructed to help only if it is "truly necessary" and is given the solution booklet to aid the process without being allowed to show the solutions to the child. The test administrator leaves the room after having instructed the child and parent. The five-minute test is video-taped for subsequent coding. Debriefing at the end of the test emphasizes for the child that the task is extremely difficult even for adults and that the objective was to see how children carry on with their work even when it poses great challenges. The brevity of the test makes it suitable for research and in clinical assessment when mapping children's abilities to regulate emotions. Furthermore, the presence of a parent increases the ecological validity by mimicking a real-life situation where the child is alone with a parent instead of facing an unfamiliar test administrator.

The present manual (available upon request) comprises eight items for parental behavior (intrusiveness, control, avoidance, verbal reappraisal, support sensitivity, positive expressions, negative expressions, and tension), nine items for child behavior (control, avoidance/resignation, narration, verbal reappraisal, reassurance-seeking behavior, aggression, positive expressions, negative expressions, and the overall emotion regulation scale (EmReg)) and one item on the parent-child dyad (emotional warmth). Each regulatory process of the process model of emotion regulation is represented by at least one item that is placed accordingly on the scoring sheet, although in theory the boundaries between the five processes can be fluid and some items may be applicable to multiple processes. Although several facets

of parental behavior are scored, the primary focus of the test is child behavior. The purpose of the parent items is to allow for the possibility of studying the quality of interaction (represented by the item "emotional warmth") and to have access to parental measures of potential interest in clinical settings such as negative attitudes or level of support. Also, parental behavior is taken into account when assessing the child's overall ER ability by weighing child behavior against the displayed parental support or lack thereof. Three additional items for child behavior (situation rejection, tension, and incongruent positive affect) appear in the present analyses but were ultimately removed as a result of the work presented in this article. All items except for the EmReg are scored on a four-point Likert-scale on frequency (never, rarely, sometimes, often) and on a three-point Likert-scale on intensity (mild, moderate, marked). The EmReg score is an overall measure of the child's perceived emotion regulation skills when taking into account all factors, such as parental influence, perceived difficulties etc., and it is scored on a five-point scale with 1 representing very poor ER skills and 5 representing excellent ER skills. The remaining items cover aspects of emotion regulation such as the use of specific strategies, but also more simple emotional behavior such as emotion communication as well as behavior comprising a more executive component. This way of coding allows for a quantification of the child's abilities and a descriptive insight into the personal ER style. The inclusion of the EmReg score allows for measuring changes over time, for example before and after therapy.

2.2.2 Behavior Rating Inventory of Executive Function (Parent Form) – Emotional Control Scale

The Behavior Rating Inventory of Executive Function (BRIEF) is a parent-completed rating scale consisting of 86 questions that assess executive function in children and adolescents aged 5-18 (Gioia et al., 2000). Parents are asked to rate their child's problematic behaviors during the past six months. The Emotional Control Scale (EC) is one of eight clinical scales in the BRIEF and reflects the influence of executive function on ER.

2.2.3 Child Behavior Check-List – "Deficient Emotional Self-Regulation profile"

Parents completed the Child Behavior Check-List (CBCL) for ages 6-18, which is a questionnaire identifying various problem behaviors in children (Achenbach & Rescorla, 2001). Parents were asked to rate their child's behavior during the past six months. The Deficient Emotional Self-Regulation (DESR) profile from the CBCL combines the scores of three symptom scales (Aggressive Behavior, Anxious/Depressed, and Attention Problems) and has been found to reflect difficulties in regulation of emotions, as well as being associated with high rates of disruptive behavior (Biedermann et al., 2012; Spencer et al., 2011).

2.2.4 Wechsler Intelligence Scale for Children

The Wechsler Intelligence Scale for Children – fourth edition (WISC-IV) is an intelligence test for children aged 6-16, which generates a full scale IQ and a number of index scores (Wechsler, 2004). In the present study, we used the full scale IQ along with the Verbal Comprehension Index (VCI) and the Perceptual Reasoning Index (PRI). The former measures aspects of intelligence reflected in verbal concept formation and the latter is a non-verbal measure of facets of intelligence reflected in fluid reasoning, spatial processing, and visual-motor integration.

2.3 Procedures

The study was approved by the Regional Committee on Health Research Ethics (journal number H-2-2013-085) and the Danish Data Protection Agency (journal number 2017-58-0015). Written informed consent was obtained from parents of all participants in accordance with the Declaration of Helsinki. All subjects participated in the test as part of a large test battery. Psychologists, medical doctors, or a

research nurse administered the test and gave instructions verbatim in order to decrease instruction bias. Two trained raters (JH and KSS) conducted the coding of the videos during a period of three months.

2.4 Data analyses

Statistical analyses were performed using SPSS version 22.0. The datasets for the analysis in this manuscript are available upon request, without reservations, to all researchers.

We used the intra-class correlation coefficient (ICC) for the analysis of interrater reliability (IRR). This is one of the most frequently used statistics for reporting IRR for ordinal variables (Hallgren, 2012). Two trained raters (JH and KSS) coded a subset of 30 participants for the IRR analysis. For the intrarater reliability analysis, each rater coded 10 videos twice with approximately three months in between. We applied a two-way mixed model testing for absolute agreement with average-measures ICCs. Reliability coefficient values were interpreted based on the guidelines presented in Cichetti (1994).

We conducted Spearman's rank-order correlations (Spearman, 1910), as EmReg is both an ordinal variable and presented with a non-normal distribution. We tested associations between the EmReg score and the EC Scale, the DESR profile, total IQ, VCI, and PRI. For the comparison with IQ, only the 70 participants drawn from the study with children with neuropsychiatric disorders were included, as the 15 children from the high risk study were not tested with the WISC-IV. Values of ±0.1 represent a small effect, ±0.3 is a medium effect, and ±0.5 is a large effect (Field, 2009). We chose only the EmReg score for comparison as this representation of overall ER ability is the primary outcome of the TEC-M.

The concept of content validity centers on the theoretical construct that is being measured and whether a test is able to capture all aspects of that construct (Carmines & Zeller, 1979). Lawshe (1975) proposed a method of testing content validity, which makes use of a panel of experts in the field who evaluate individual items of a test as to whether they reflect the theoretical domain in a satisfactory manner. In this method the panel rates each item as either ‘Essential’, ‘Useful, but not essential’, or ‘Not necessary’. From these ratings, it is possible to calculate a content validity ratio (CVR), which is held against a critical value depending on the number of panelists. In the present study, eight panelists at the doctoral and master’s level rated the items, requiring a CVR of 0.75. Any item perceived essential by more than half of the panelists is thought to hold some degree of content validity, and the more who perceive it to be essential, the greater its content validity (Lawshe, 1975).

We examined the ability to differentiate the ADHD group from the control group for all items. We used Mann-Whitney U tests (Mann & Whitney, 1947) to investigate group differences, as items were scored on an ordinal scale, and calculated effect sizes for significant results. Differences in age and gender between the control group and the ADHD group were assessed with an independent samples t-test and the chi-square test of homogeneity, respectively.

3 Results

3.1 Inter- and intrarater reliability

ICCs showed excellent agreement for 11 items, good agreement for 13 items, fair agreement for six items, and poor agreement for four items (Table 2). The intrarater reliability analyses showed excellent agreement for respectively 23 and 24 items, good agreement for five items, fair agreement for

respectively three and four items, and poor agreement for one item with some variation in items between raters. For items with zero or low variance an ICC could not be calculated. Overall, the ICCs reflected a high level of inter- and intrarater agreement.

3.2 Convergent and discriminant validity

For convergent validity, we found a weak and only trend level, inverse correlation between EC and the EmReg score, $r_s(78) = -.20$, $p = .079$ (Table 3). Regarding the DESR profile compared to EmReg we found a significant, close to medium, inverse correlation, $r_s(73) = -.28$, $p = .016$. For the comparison with EC and DESR, data were missing from five and ten participants, respectively, whose parents had not filled out the questionnaires. We found a medium, positive correlation between total IQ and EmReg score, $r_s(68) = .30$, $p = .011$ for the analysis of discriminant validity. Weak positive correlations were found between EmReg and the VCI ($r_s(68) = .27$, $p = .026$) and the PRI ($r_s(68) = .28$, $p = .018$).

3.3 Content validity

For the content validity analysis (Table 4), seven items out of a total of 21 reached the critical value of 0.75 (situation rejection, control, narration, verbal reappraisal, reassurance-seeking behavior, aggression, and the EmReg score). At least half the panelists perceived eighteen items in total as essential, suggesting broad basis for content validity. For three items, more than half the panelists rated 'Useful, but not essential', whereas only one item was perceived as 'Not necessary' by only one panel member, and this item was ultimately removed.

3.4 Sensitivity between groups

There was homogeneity of variances for age as assessed by Levene's test for equality of variances ($p = .497$). The two groups did not differ on age and gender distributions (Table 1). Mann-Whitney U tests were run to determine which items could differentiate the ADHD group from the control group. Distributions of item scores for children with ADHD and those in the control group were similar when assessed by visual inspection. For all significant scores, both the median and the mean rank are reported, as the Likert-scale format of the ratings can lead to identical medians for some items, even when there is a difference between groups. Item scores differed significantly between the groups on seven out of a total of 40 items: Item score on frequency of parental tension was significantly higher in the ADHD group ($Mdn = 0.00$, mean rank = 47.15) than in the control group ($Mdn = 0.00$, mean rank = 41.46), $U = 808.5$, $z = 1.982$, $p = .047$, $r = 0.21$. Item score on frequency of child control was significantly higher in the control group ($Mdn = 3.00$, mean rank = 46.60) than in the ADHD group ($Mdn = 2.00$, mean rank = 33.30), $U = 409$, $z = -2.566$, $p = .010$, $r = -0.28$. For frequency of child narration, the control group ($Mdn = 1.00$, mean rank = 47.21) scored significantly higher than the ADHD group ($Mdn = 0.00$, mean rank = 31.65), $U = 452$, $z = -2.885$, $p = .004$, $r = -0.31$, and the same was true for intensity of child narration where the control group ($Mdn = 1.00$, mean rank = 46.44) scored significantly higher than the ADHD group ($Mdn = 0.00$, mean rank = 33.72), $U = 499.5$, $z = -2.366$, $p = .018$, $r = -0.26$. For frequency and intensity of child aggression (identical results), the ADHD group scored higher ($Mdn = 0.00$, mean rank = 45.70) than the control group ($Mdn = 0.00$, mean rank = 42.00), $U = 775$, $z = 2.366$, $p = .019$, $r = 0.26$. Finally, children from the control group achieved significantly higher scores on the EmReg ($Mdn = 4.00$, mean rank = 47.81) than the ADHD group ($Mdn = 3.00$, mean rank = 30.02), $U = 414.5$, $z = -3.122$, $p = .002$, $r = -0.34$.

4 Discussion

The purpose of this study was two-fold. Firstly, we described the theoretical background and development of the TEC-M; a novel coding manual for an experimental test of ER in children that allows for direct, observational measures of overall ER ability as well as a number of ER characteristics and quality of the parent-child interaction. Secondly, we examined the manual on a number of parameters, such as reliability, validity, and sensitivity to group differences.

Analyses showed good or excellent interrater agreement for the majority of items pointing to a substantial robustness of the items and indicating that training and consulting the manual can produce reliable ratings on most items even between two professional groups (a psychologist and a medical doctor, respectively). The items reaching only poor or fair agreement point to a need for further clarification in the manual and further standardized assessment of the respective scores. Results from the intrarater reliability analysis showed that the two raters succeeded in similar or identical ratings at two points in time for the vast majority of items. For the few items that reached poor or fair agreement (only one item per rater showed poor agreement), the majority were intensity scores, suggesting a need for further clarification of the different levels of intensity of a specific behavior. Although reliability ratings were satisfactory, it should be emphasized that thorough training in the coding procedures is central to decreasing bias in clinician ratings by creating a common framework for coding. Additionally, regular co-ratings with experienced coders would be preferable to strengthen the accuracy of clinicians' ratings. Three items showed zero variance in the distribution of scores (child situation rejection, child tension, and incongruent positive affect), which led to the decision of removing them from the manual completely as the rarely displayed behavior pertaining to these items was well-represented by other items.

There are no clear guidelines as to which scores might serve as cut-off for establishing construct validity when interpreting correlations. Cichetti (1994) accurately points out that the ideal correlation value depends on what the new test purports to measure in relation to an existing one: "We know for sure that we would hope for a correlation of neither 1.00 nor 0. In the first case, the new test could be considered a veritable clone of the one with which it is being compared. In the second case, the construct validity of the very concept being measured would be called into question" (Cichetti, 1994, p. 5). Both correlations for convergent validity (EmReg compared to EC and DESR) were weak; however, the comparison with DESR was close to medium and significant. Due to the explorative nature of these tests we did not correct for multiple comparisons, but it would be interesting to see if future studies would be able to replicate these findings. Although traditionally correlations testing for convergent validity are higher, it is not surprising that these correlations were not. First of all, the questionnaires used for comparison were parent-reports of child behavior assessed over a period of six months. This type of assessment, though theoretically intended to measure ER, is in clear contrast to the outcomes of the TEC-M which aims at direct and instantaneous observation of ER in a concrete setting within a limited timeframe. The purpose of developing the TEC-M was to create an instrument that was able to capture aspects of ER that previous studies had not been able to, due to a lack of tests or relying on parent- or self-report measures. Future studies of the validity of the TEC-M would benefit from more similar measures of comparison; however, the current lack of experimental tests in the area complicates the possibility for investigating convergent validity further. In regard to the examination of discriminant validity, the correlation between IQ (full scale IQ, VCI, and PRI) was in the medium range, suggesting some association between performance on the test (ER ability) and general intellectual abilities as shown in previous studies. We expected that the experienced level of frustration would to some degree depend on how difficult the child would find the task, thus explaining the association with IQ. Interestingly, the correlations between the EmReg score and VCI and PRI, respectively, were

almost identical. We expected to find a slightly higher correlation between EmReg and PRI as the tasks covered in this index are somewhat similar to the Tangram Construction Task. The TEC-M, however, also includes aspects related to verbal abilities, such as narration and verbal reappraisal. The fact that verbal comprehension and perceptual reasoning correlated equally well with EmReg supports the role of the EmReg score not as a measure of construction skills, but as a measure of ER ability.

For the examination of content validity, more than half the panelists rated the majority of items 'Essential', displaying a satisfying degree of content validity, although the critical CVR was only reached for seven items. All items except for one were rated either 'Essential' or 'Useful, but not essential' supporting the relevance of the selected items for the reflection of the theoretical construct of ER. The panelists were asked to rate the relevance of each item in regard to its ability to adequately cover ER, leading to some panelists finding the parent items to be only useful, but not essential as they were not direct measures of the child's abilities. The same was the case for the item covering 'Emotional warmth', which is a shared measure for the child and the parent and as such does not capture the specific ER abilities of the child. Still, we find the inclusion of parent-items relevant for elucidating the bigger picture of children's ER characteristics and the influence of parental behavior on these behaviors, an association that has been established in research previously (Lewis et al., 2006).

We examined the test's ability to differentiate children with ADHD from the control children in an explorative manner and found significant differences in scores between the groups on seven items with effect sizes in the medium range. These findings corresponded well with existing knowledge in the field of ADHD pointing to reduced emotion regulation abilities, an increased occurrence of aggressive behavior, as well as deficits in executive functioning (Melnick & Hinshaw, 2000). Firstly, the ADHD group scored significantly lower on the EmReg score, which is a measure of overall ER ability. Secondly, the ADHD group scored significantly higher on both frequency and intensity of aggression. This finding, however, must be interpreted with caution due to the low number of participants scoring higher than zero on these items. Frequency of parental tension was significantly higher in the ADHD group than in the control group; a finding that is in line with research showing a strong association between parenting styles and ADHD symptom severity (Muñoz-Silva & Lago-Urbano, 2016). For the control group, scores were significantly higher on frequency of control, as well as frequency and intensity of narration. An explanation for this might be that both the autonomy of taking control of the task and the employment of narration to facilitate task solving rest on higher executive functions which may be impaired in children with ADHD.

The TEC-M is not intended as a diagnostic test, but as a transdiagnostic tool to characterize children's individual ER abilities through systematic rating. Although in the present study the test has only been applied to children with ADHD, the TEC-M is generic and can be used across diagnostic groups with a dimensional approach to dysregulation of emotions. The frustration element of the puzzle is necessary to facilitate the elicitation of codable behavior, but this premise also limits the degree to which one can make statements regarding general behavior. Interpreting the displayed behavior as part of assessment in a child psychiatric setting must therefore include a consideration of the limited context of the task and must always be viewed in connection with the complete assessment. In a clinical context, the test may serve as an evaluation before and after therapy or as a measure to assess the need for psychoeducation in the family. Additionally, using instructive examples from the videos to illustrate maladaptive and adaptive behaviors on behalf of both the parent and the child could prove useful in therapy. Finally, the exploration of potential profiles of ER corresponding to various disorders could help accommodate a dimensional approach to child psychopathology and an individualized approach to treatment.

5 Limitations

There are a number of limitations to this study, such as a preponderance of boys and a limited age span in the sample. However, due to its feasibility, the test could easily be used in both younger and older age groups and a general expansion of the participant sample in regard to gender, age, and clinical grouping represents an important direction for future research. Moreover, future measures for convergent and discriminant validity should be chosen carefully beforehand to assure these correlations in the full sample of participants. One major limitation of the study was the lack of systematic blinding. As the raters had administrated the test themselves to some of the participants, it was impossible to achieve total blinding. Many of the participants, however, had been instructed by others than the raters and in the case of a rater having instructed a particular participant, several months would have passed between contact with the patient and the actual rating.

Five of the co-authors in the present study were simultaneously part of the expert panel for the examination of content validity. The main purpose of the rating, however, was not to reach a high content validity ratio, but rather to evaluate the instrument in an unbiased manner as part of its development. Additionally, the panelists were kept in the dark as to how their ratings would translate into an assessment of content validity.

A final limitation to be discussed is the inherent challenge in measuring emotion regulation on a behavioral level, as the regulation itself will often be invisible to the rater. The TEC-M is designed to code the actual behavior exhibited, but it cannot declare the degree of regulation or the intensity of the emotion that is being regulated. The use of explicit strategies such as verbal reappraisal or narration is directly codable, whereas the implicit or nonverbal regulation will always be an approximation. The key goal of the EmReg score is to function as such an approximation by taking into account all information gathered from the TEC-M.

6 Conclusion

The findings of the present study support the use of the TEC-M, a coding manual for an observational test of emotion regulation in children, by demonstrating satisfactory psychometric properties. This contribution is relevant as dysregulation of emotions is evident in numerous mental disorders and a further understanding of these processes might help improve treatment options. The TEC-M differs from existing measures in regard to its ecological validity and systematic observation of the child and its generic methodology will hopefully be able to contribute to a transdiagnostic approach to psychopathology focusing on a fundamental dimensionality in psychiatric disorders.

7 Conflict of interest

The authors have no conflicts of interest to disclose.

8 Author Contributions

JH collected the data, developed the instrument, and wrote the manuscript with input from all authors. KS co-developed the instrument and both KS and KM contributed to the collection of data. BC helped drafting a part of the text, and BH developed the original instrument on which this work was based. JJ and KP supervised the development of the instrument and contributed to the interpretation of the results, and SV and KP were involved in designing the study. All co-authors critically revised the article for intellectual content and approved the final version.

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Table 1. Participant characteristics

ADHD = Attention-deficit/hyperactivity disorder. [a]Chi-square test of homogeneity. [b]$t(83) = 0.697$. [c]Four children with ADHD additionally met criteria for oppositional defiant disorder (ODD), one participant had both ODD and separation anxiety disorder, and one participant met criteria for generalized anxiety disorder, separation anxiety disorder, and transient tic disorder in addition to ADHD.

Characteristic	ADHD (n = 23)	Controls (n = 62)	Test of group differences
Male, N (%)	15 (65.2)	49 (79.0)	$p = .190$[a]
Mean age (*SD*)	9.59 (1.25)	9.36 (1.40)	$p = .488$[b]
Comorbid disorders[c], N (%)	6 (26.1)	0 (0)	

Table 2. Inter- and intrarater reliability

CI = Confidence interval. ICC = Intra-class correlation coefficient. Z.V. = Zero variance. N/A = Not applicable. IPA = Incongruent positive affect. EmReg = Overall emotion regulation scale. All ICC values of good or excellent agreement are marked in bold (Cichetti, 1994).

	Item (F = frequency, I = intensity)	Distribution of scores (%)				Interrater reliability		Intrarater Reliability (rater 1)		Intrarater Reliability (rater 2)	
		0	1	2	3	ICC	95% CI	ICC	95% CI	ICC	95% CI
Parent items	Intrusiveness F	30	33	15	22	**0.71**	[0.53, 0.89]	**0.88**	[0.51, 0.97]	**0.84**	[0.41, 0.96]
	Intrusiveness I	30	43	18	8	**0.76**	[0.31, 0.90]	**0.83**	[0.33, 0.96]	**0.91**	[0.62, 0.98]
	Avoidance F	73	25	2	0	**0.79**	[0.57, 0.90]	**1.00**	N/A	**0.90**	[0.62, 0.97]
	Avoidance I	73	20	7	0	**0.78**	[0.54, 0.89]	**1.00**	N/A	**0.85**	[0.29, 0.96]
	Control F	0	5	28	67	**0.63**	[0.22, 0.83]	**0.63**	[-0.43, 0.91]	**0.82**	[0.29, 0.95]
	Control I	0	13	67	20	**0.76**	[0.50, 0.88]	0.52	[-0.63, 0.87]	**0.71**	[-0.02, 0.93]
	Verbal reappraisal F	73	18	8	0	**0.89**	[0.76, 0.95]	**0.94**	[0.77, 0.98]	**1.00**	N/A
	Verbal reappraisal I	73	18	7	2	**0.73**	[0.43, 0.87]	**0.89**	[0.59, 0.97]	**0.91**	[0.68, 0.98]
	Negative expressions F	50	40	10	0	0.50	[-0.05, 0.76]	**0.74**	[0.07, 0.93]	**0.83**	[0.33, 0.96]
	Negative expressions I	50	40	8	2	0.48	[-0.10, 0.75]	**0.78**	[0.20, 0.94]	**0.76**	[-0.07, 0.94]
	Positive expressions F	3	28	35	33	**0.64**	[0.26, 0.83]	**0.83**	[0.29, 0.96]	**0.96**	[0.86, 0.99]
	Positive expressions I	3	50	43	3	0.58	[0.12, 0.80]	**0.94**	[0.77, 0.98]	0.54	[-1.01, 0.89]
	Tension F	93	3	2	2	0.55	[0.03, 0.78]	**1.00**	N/A	**1.00**	N/A
	Tension I	93	2	5	0	**0.75**	[0.47, 0.88]	**1.00**	N/A	**1.00**	N/A
	Support sensitivity F	2	12	30	57	**0.75**	[0.39, 0.89]	0.57	[-1.02, 0.90]	**0.65**	[-0.26, 0.91]
	Support sensitivity I	2	25	62	12	0.22	[-0.61, 0.62]	**0.69**	[-0.08, 0.92]	0.53	[-1.27, 0.89]
Child Items	Situation rejection F	100	0	0	0	Z.V.	N/A	Z.V.	N/A	Z.V.	N/A
	Situation rejection I	100	0	0	0	Z.V.	N/A	Z.V.	N/A	Z.V.	N/A
	Control F	0	7	43	50	**0.66**	[0.28, 0.83]	**0.89**	[0.52, 0.97]	**0.74**	[0.00, 0.93]
	Control I	0	32	63	5	0.51	[-0.02, 0.76]	0.37	[-1.89, 0.85]	0.40	[-2.03, 0.86]
	Avoidance/resignation F	38	28	28	5	**0.61**	[0.16, 0.82]	**0.95**	[0.80, 0.99]	**0.66**	[-0.49, 0.92]
	Avoidance/resignation I	38	27	35	0	**0.70**	[0.33, 0.86]	**0.91**	[0.66, 0.98]	0.36	[-1.03, 0.83]
	Narration F	52	32	17	0	**0.64**	[0.25, 0.82]	**0.76**	[0.04, 0.94]	**0.80**	[0.27, 0.95]
	Narration I	52	13	35	0	**0.67**	[0.31, 0.84]	**0.86**	[0.42, 0.96]	**0.80**	[0.27, 0.95]
	Verbal reappraisal F	70	25	3	2	**0.60**	[0.17, 0.81]	**1.00**	N/A	**0.84**	[0.33, 0.96]
	Verbal reappraisal I	70	17	13	0	**0.71**	[0.39, 0.86]	**0.97**	[0.88, 0.99]	**0.85**	[0.43, 0.96]
	Reassurance-seeking F	10	27	43	20	**0.83**	[0.65, 0.92]	**0.83**	[0.32, 0.96]	**0.90**	[0.57, 0.98]
	Reassurance-seeking I	10	30	55	5	0.40	[-0.28, 0.72]	**0.69**	[-0.18, 0.92]	**0.68**	[-0.38, 0.92]
	Tension F	98	2	0	0	Z.V.	N/A	Z.V.	N/A	Z.V.	N/A
	Tension I	98	2	0	0	Z.V.	N/A	Z.V.	N/A	Z.V.	N/A
	Aggression F	96	3	0	0	-0.07	[-1.35, 0.50]	Z.V.	N/A	**0.78**	[0.17, 0.94]
	Aggression I	97	3	0	0	-0.07	[-1.35, 0.50]	Z.V.	N/A	**0.78**	[0.17, 0.94]
	Negative expressions F	3	33	40	23	**0.72**	[0.41, 0.87]	0.47	[-0.77, 0.86]	**0.93**	[0.73, 0.98]
	Negative expressions I	3	42	48	7	**0.62**	[0.22, 0.82]	**0.73**	[-0.14, 0.93]	**1.00**	N/A
	Positive expressions F	7	28	42	23	**0.78**	[0.53, 0.90]	**0.78**	[0.08, 0.95]	**0.80**	[0.15, 0.95]
	Positive expressions I	7	45	45	3	**0.76**	[0.49, 0.89]	**1.00**	N/A	**0.83**	[0.37, 0.96]
	IPA F	98	2	0	0	Z.V.	N/A	Z.V.	N/A	Z.V.	N/A
	IPA I	98	2	0	0	Z.V.	N/A	Z.V.	N/A	Z.V.	N/A
Emotional warmth		0	3	20	77	0.33	[-0.23, 0.66]	**1.00**	N/A	0.54	[-1.01, 0.89]
EmReg (score (n))		1 (1), 2 (11), 3 (31), 4 (32), 5 (10)				**0.78**	[0.53, 0.89]	**0.81**	[0.21, 0.95]	**0.79**	[0.10, 0.95]

Table 3. Spearman's rank-order correlations

EmReg = Overall emotion regulation scale. EC = Emotional Control Scale. DESR = Deficient Emotional Self-Regulation profile. VCI = Verbal Comprehension Index. PRI = Perceptual Reasoning Index.

		EC	DESR	Total IQ	VCI	PRI
EmReg	Correlation	-.20	-.28	.30	.27	.28
	p value	.079	.016	.011	.026	.018

Table 4. Content validity

*'Essential'; 'Useful, but not essential'; 'Not necessary'. CVR = Content Validity Ratio. EmReg = Overall emotion regulation scale.

Item	Rating count*	CVR
Parental intrusiveness	5; 3; 0	0.25
Parental avoidance	5; 3; 0	0.25
Parental control	6; 2; 0	0.5
Parental verbal reappraisal	6; 2; 0	0.5
Parental negative expressions	4; 4; 0	0
Parental positive expressions	4; 4; 0	0
Parental tension	3; 5; 0	-0.25
Parental support sensitivity	6; 2; 0	0.5
Child situation rejection	8; 0; 0	1
Child control	8; 0; 0	1
Child avoidance/resignation	6; 2; 0	0.5
Child narration	7; 1; 0	0.75
Child verbal reappraisal	8; 0; 0	1
Child reassurance-seeking behavior	8; 0; 0	1
Child tension	5; 3; 0	0.25
Child aggression	8; 0; 0	1
Child negative expressions	6; 2; 0	0.5
Child positive expressions	6; 2; 0	0.5
Child incongruent positive affect	3; 4; 1	-0.25
Emotional warmth	3; 5; 0	-0.25
EmReg	7; 1; 0	0.75

B. Paper 2

Emotion Regulation in Children with Tourette Syndrome – Associations with ADHD and Tic Severity.

Julie Hagstrøm, Katrine S. Spang, Jens Richardt Møllegaard Jepsen, Signe Vangkilde, Katrine Maigaard, Liselotte Skov, Anne Katrine Pagsberg, Kerstin Jessica Plessen.

Manuscript in review.

Running head: Emotion Regulation in Tourette Syndrome

Emotion Regulation in Children with Tourette Syndrome – Associations with ADHD and Tic Severity

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Conflict of Interest

The authors have no conflicts of interest to disclose.

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Emotion Regulation in Tourette Syndrome

Abstract

Background: Explosive outbursts occur in 25-70% in children with Tourette syndrome (TS) and may cause more distress than the tics themselves. Previous studies indicate that a concurrent diagnosis of attention-deficit/hyperactivity disorder (ADHD) is associated with disruptive behavior in children with TS; however, this relationship has primarily been studied using parent-reported questionnaires.

Methods: We examined emotion regulation (ER) with a direct, observational measure in 150 medication-naïve children aged 7-12 allocated to four groups: Forty-nine with TS, 23 with ADHD, 16 with TS + ADHD, and 62 typically developing controls. We assessed participants' ER ability and aspects of the regulatory process, as well as parent-child interactions within the context of a complex puzzle task and coded behavior with the Tangram Emotion Coding Manual. We examined group differences in ER and potential associations between efficacy of ER and symptom severity pertaining to TS and ADHD, respectively.

Results: Children with TS and controls did not differ concerning their ER ability; however, children with ADHD and those with TS and comorbid ADHD achieved significantly lower scores of ER than those with TS only and controls. A similar pattern emerged for a factor covering executive emotional control on the same task. Finally, parents of children with ADHD displayed more signs of tension during the experimental task. Scores of ER ability were not associated with tic severity or premonitory urges; however, they correlated inversely with symptoms of ADHD (inattention and hyperactivity/impulsivity).

Conclusions: This study is the first to experimentally evaluate ER ability in children with TS. Our findings support earlier studies by showing impaired ER in children with ADHD, but not in TS without comorbid ADHD. These findings increase the understanding of the specific processes that are

impaired in this population. A better understanding of the phenomenology of dysregulated behavior will hopefully contribute to optimizing treatment possibilities.

Keywords: Emotion regulation, Tourette syndrome, ADHD, tic severity, premonitory urges.

Abbreviations

ADHD = Attention-deficit/hyperactivity disorder
ADHD-RS = ADHD Rating Scale
CBCL = Child Behavior Checklist
EFA = Exploratory factor analysis
EmReg = Overall emotion regulation scale
ER = Emotion regulation
OCD = Obsessive-compulsive disorder
ODD = Oppositional defiant disorder
PUTS = Premonitory Urge for Tics Scale
YGTSS = Yale Global Tic Severity Scale
SES = Socioeconomic status
TEC-M = Tangram Emotion Coding Manual
TS = Tourette syndrome

Emotion Regulation in Tourette Syndrome

Twenty-five to 70% of children with Tourette syndrome (TS) present with dysregulated emotional behavior (Chen et al., 2013); the large span of this estimate possibly indicating a lack of systematic assessment (Budman, Bruun, Park, Lesser, & Olson, 2000). Disruptive behavior has been described as the most impairing "comorbidity" (Leclerc, O'Connor, Forget, & Lavoie, 2011) and parents of children with TS often experience the explosive outbursts as more impairing than the tics per se (Dooley, Brna, & Gordon, 1999). In a larger perspective, disruptive behavior and aggression are common to numerous psychiatric disorders and frequent causes of referrals to psychiatric evaluations. This also holds true for TS, where emotional dysregulation often causes the families to seek help in the first case (Stephens & Sandor, 1999). As many as 90% of children with TS also meet criteria for at least one other psychiatric diagnosis, such as attention-deficit/hyperactivity disorder (ADHD) or obsessive-compulsive disorder (OCD; Robertson, Cavanna, & Eapen, 2015). Although diagnostic criteria for TS are limited to the presence of tics, the high occurrence of comorbidity and appertaining symptomatology makes it challenging to separate disorder specific behavior from symptoms due to other comorbidities; thus pointing to a potential multifactorial etiology of disruptive behavior in children with TS.

Factor analytic studies have suggested an aggressive phenotype in TS (Robertson et al., 2015) and the nature of explosive outbursts in TS bears resemblance to tics, pointing to disruptive behavior as a potential tic subtype (Leclerc et al., 2011). Unlike the type of aggression seen in oppositional-defiant disorder (ODD), the explosive outbursts in TS occur suddenly without apparent premeditation and is followed by a sense of loss of control (Budman et al., 2000). Likewise, patients with TS have described feeling tense preceding a rage attack with a subsequent sense of relief, which is comparable to the sequence of premonitory urges preceding a tic and relief following execution of the tic (Budman et al., 2000). Current views on disruptive behavior in TS, however, delineate the "pure" TS (i.e., TS without any comorbidities) from TS with comorbidities by showing that children with pure TS

resemble typically developing children in regard to aggressive behavior (Stephens & Sandor, 1999), and by pointing to a significant impact of ADHD on the presence of explosive outbursts. ADHD is associated with emotional dysregulation (Graziano & Garcia, 2016) and the prevalence of disruptive behavior and explosive outbursts in children with TS is higher when they have a simultaneous diagnosis of ADHD (Budman et al., 2000; Chen et al., 2013; Robertson et al., 2015), but the underlying processes remain obscure.

Hence, a growing body of evidence suggests that comorbid ADHD creates a vulnerability for the development of disruptive behavior in children with TS. Most studies, however, have relied on parent-reported questionnaires or have focused merely on establishing the occurrence of this behavior. In this study, we aimed to observe mechanisms of emotion regulation (ER) that potentially underlie dysregulated behavior to complement previous findings with a direct observational measure of ER in medication-naïve groups of children with TS, ADHD, TS and ADHD, and typically developing controls. Rage attacks are most often targeted against a parent (Budman et al., 2000) and a recent review of ER strategies across psychopathologies emphasized the need for assessment of "spontaneous emotion regulation in ecologically valid contexts" (Aldao, Nolen-Hoeksema, & Schweizer, 2010). We thus asked the children in the presence of a parent to solve a complex task eliciting frustration applying a recently validated manual for coding child ER (Hagstrøm et al., in review) to examine potential differences across groups. We hypothesized that we would find a difference in the capacity of ER between children with ADHD and the remaining groups. We additionally performed an exploratory factor analysis to investigate the latent structure of the test items. Finally, we explored the potential associations between overall ER ability and the severity of tics, premonitory urges, and ADHD symptoms, since previous studies have reported mixed results regarding the relationship between tic severity and disruptive behavior.

Emotion Regulation in Tourette Syndrome

Methods

Participants

We included 150 medication-naïve children aged 7-12 (M = 9.58, SD = 1.34) in the study: Forty-nine children with TS, 23 children with ADHD, 16 children with TS + ADHD, and 62 typically developing controls (Table 1). We assessed children adopting a multi-informant approach with the semi-structured diagnostic interview Kiddie-Schedule for Affective Disorders and Schizophrenia, Present and Lifetime Version (K-SADS-PL; Kaufman et al., 1997) for parents and children and the ADHD Rating Scale (ADHD-RS; DuPaul, Power, Anastopoulos, & Reid, 1998). The ADHD-RS is a parent- and teacher-rated questionnaire with Danish norms covering symptoms of inattention, hyperactivity/impulsivity, and conduct disorder. Children presenting with tics were additionally assessed with the Yale Global Tic Severity Scale (YGTSS; Leckman et al., 1989), which evaluates number, frequency, intensity, complexity, and interference of tics as well as tic-related impairment, and with the Premonitory Urge for Tics Scale (PUTS; Woods, Piacentini, Himle, & Chang, 2005), which assesses type, frequency, and control of premonitory urges. Twenty-four percent of children in the TS group, 26% of children in the ADHD group, and 18% of children in the TS + ADHD group had comorbid OCD, ODD, or anxiety disorders, but no severe psychopathology (e.g., autism spectrum disorder or psychotic disorders). Children in the control group did not meet DSM-IV (American Psychiatric Association, 2000) criteria for any lifetime diagnosis apart from enuresis and encopresis not related to any other psychopathology. Exclusion criteria for all groups were IQ < 80 and birth at gestational age < 37 weeks. The socioeconomic status (SES) of participants' parents was classified using the International Standard Classification of Education (Unesco, 2012). Groups did not differ in age or sex, nor did the clinical groups differ in comorbidity or ADHD subtype except for a borderline significantly different distribution of ODD (Table 1). IQ and SES scores differed across groups with the

control group achieving the highest scores followed by the TS group, the ADHD + TS group, and finally the ADHD group.

The Tangram Emotion Coding Manual (TEC-M)

The Tangram Emotion Coding Manual (TEC-M) offers a standardized assessment of children's ER ability and parent-child-interactions during a frustration task and has recently been developed and validated (Hagstrøm et al., in review). Eighty-five of the 150 children in the present study were also part of the validation study (the current ADHD and control group). The manual is theoretically founded on the process model of emotion regulation (Gross, 2014) and offers a novel coding system for the Tangram Construction Task; a task which has previously been applied in the field of child anxiety (Esbjørn, Christiansen & Steele, unpublished manuscript; Hudson & Rapee, 2001). In the five-minute complex puzzle task, the child tries to solve as many puzzles as possible in the presence of a parent, who holds the solution booklet, but is instructed to help only if it is "truly necessary". The test is video-taped to allow for subsequent coding of eight items covering parental behavior (intrusiveness, control, avoidance, verbal reappraisal, support sensitivity, positive expressions, negative expressions, and tension), nine items for child behavior (control, avoidance/resignation, narration, verbal reappraisal, reassurance-seeking behavior, aggression, positive expressions, negative expressions, and the overall emotion regulation scale (EmReg)), and one item on the parent-child dyad (emotional warmth). Items are scored on frequency (never, rarely, sometimes, often) and intensity (mild, moderate, marked) with the primary outcome being the EmReg score, which is scored on a five-point Likert scale with 1 representing very poor ER skills and 5 representing excellent ER skills. The EmReg score represents the child's overall ER ability when all factors are considered, such as parental behavior and perceived level of difficulty. The remaining items cover aspects of regulation, emotional expressions, and emotional behavior related to executive control.

Procedures

This research was approved by the Regional Committee on Health Research Ethics (journal number H-2-2013-085) and the Danish Data Protection Agency (journal number 2017-58-0015). We obtained written informed consent from parents of all participants in accordance with the declaration of Helsinki. We recruited participants with TS and ADHD from the local child and adolescent psychiatric outpatient clinic and a pediatric hospital clinic, both at Capital Region University Hospitals, Denmark, from 2013 to 2016. Children from the control group were randomly selected and recruited via the Danish Civil Registration System (Pedersen, 2011) and were matched on age and sex with the TS group. Psychologists or a medical doctor conducted the clinical assessments. JH completed the coding of all videos.

Data analyses

We aimed to detect medium effect sizes as a minimum between our primary groups, the TS group and the control group, with an a priori alpha level of .05 and an expected power of .80. This required a sample size of 51, which we exceeded for the control group and almost reached for the TS group.

We performed an exploratory factor analysis (EFA) using principal axis factoring with an orthogonal rotation (varimax) as this rotation technique is recommended when first exploring the dataset (Kline, 2014; Yong & Pearce, 2013). Factor scores were obtained with the Anderson-Rubin method (DiStefano, Zhu, & Mîndrilă, 2009). We included all items from the TEC-M in the final analysis and used the Kaiser Criterion of eigenvalues above one (Kaiser, 1960) in deciding the number of factors to retain. Item loadings above 0.3 were accepted as pertaining to a specific factor. Separate EFA's were conducted on the parent and the child items, respectively, with the shared item

"Emotional warmth" being included in both analyses. EFA's were solely conducted on frequency items as the inclusion of both frequency and intensity items created multicollinearity.

We used the chi-square test of homogeneity to investigate differences in proportions of frequency scores between groups on all ER items. We pooled the frequency scores of never/rarely and sometimes/often, thereby creating two levels of "low" and "high" frequency, whereas the EmReg score retained five levels. We applied the Holm-Bonferroni method for pairwise comparisons and conducted all subsequent group-wise comparisons with non-parametric procedures since four of the six derived factor scores showed marked skewness. We made initial overall comparisons using the Kruskal-Wallis H test on the factor scores and, where this proved significant, we performed the six pairwise comparisons using Dunn's procedure with a Bonferroni adjustment.

We assessed associations between the EmReg score as an overall measure of ER ability and symptom severity with Pearson's product-moment correlation and the point-biserial correlation.

Results

Exploratory factor analysis

The Kaiser-Meyer-Olkin (KMO) measure of sampling adequacy and Bartlett's test of sphericity supported the use of factor analysis (Yong & Pearce, 2013) for parent items (KMO = 0.61, Bartlett's test: χ^2= 161.07, *df* = 36, $p < .001$) and child items (KMO = 0.62, Bartlett's test: χ^2= 250.37, *df* = 45, $p < .001$). Not all coefficients in the correlation matrix were above 0.3, but all items were retained in the final analysis. We chose to deviate from this convention (Yong & Pearce, 2013) as the analysis was explorative and as we had a priori decided to include all items in the factor matrix. Furthermore, the objective of the EFA was to identify underlying variables and thus we were not concerned with any potential item redundancy. The factor analysis for parent items did not render a valid factor solution and was not subject to further analyses. The factor analysis for the child items re-

vealed three theoretically meaningful factors, which explained 53% of the variance in total (Table 2). All child items loaded on the three factors with two items cross-loading ("Negative expressions" and "EmReg"). We named the first factor "Emotional control" as the items loading positively on this factor were "Control" and "EmReg" with "Avoidance", Reassurance-seeking", and "Negative expressions" loading negatively on the factor. The second factor, "Emotion communication", consisted of positive loadings from "Positive expressions", "Emotional warmth", and "EmReg" and negative loadings from "Aggression" and "Negative expressions". The final factor represented "Verbal ER strategies" with loadings from "Narration" and "Verbal reappraisal".

Group differences in emotion regulation on the TEC-M

The initial analysis revealed significant differences between groups on parental intrusiveness, parental tension, the EmReg score, and child factor 1 (Emotional control). The primary outcome of overall ER ability of the child, the EmReg score, differed across groups (p = .019). Post-hoc analyses indicated that the control group scored significantly higher than the ADHD group (p = .012) and the TS + ADHD group (p = .010) but not the TS only group, which presented with significantly higher scores than the ADHD group (p = .037) and the TS + ADHD group (p = .029) and no significant difference compared to the control group (Appendix A). Group scores on child factor 1 (Emotional control) differed across groups ($H(3) = 14.819$, p = .002; Appendix A). Pairwise comparisons showed that the control group achieved higher scores than the ADHD group (p = .037) and the TS + ADHD group (p = .031), and the TS group scored significantly higher than the TS + ADHD group (p = .041). Six to eight percent of parents in the three clinical groups were high on frequency of intrusive behavior, whereas this behavior was present in 25% of parents to children in the control group; a statistically significant difference of p = .030 (Appendix B); however, this result did not survive a control for multiple comparisons. High frequency of parental tension was present in 2% of parents in the TS group and not present at all in the control group and the TS + ADHD group, whereas 17% of

parents in the ADHD group were tense "sometimes" or "often". This overall difference was significant (p = .001), and pairwise comparisons showed that the ADHD group scored significantly higher than the control group (p = .028).

Associations between emotion regulation, symptom severity, and premonitory urges

We did not find any association between children's ability to regulate emotions and TS related symptoms such as severity of tics and premonitory urges (Table 3). ADHD symptoms of inattention and hyperactivity/impulsivity were both inversely correlated with the EmReg score with small, but significant correlation coefficients (r(147) = -.234, p = .004 and r(147) = -.211, p = .010, respectively), whereas conduct disorder was not significantly correlated with ER ability (Table 3).

Discussion

We evaluated children's ability to regulate their emotions with an experimental task in four groups of medication-naïve children with TS, ADHD, TS + ADHD, and typically developing controls. Our primary groups, the TS group compared with the control group, did not differ in overall ER ability. Conversely, children with ADHD received significantly lower ER scores than children with TS and controls. These findings were mirrored in the results for the child factor "Emotional control", partly due to the EmReg score loading significantly onto this factor. Parents of children with ADHD displayed increased tension compared to all other groups. Finally, ER difficulties were not correlated with TS symptoms, but correlated inversely with severity of inattention.

The absence of a deficit in ER in children with TS suggests that explosive outbursts in TS are not a subtype of tics but are related to other factors. Previous studies have suggested that the presence of explosive outbursts in children with TS is largely accounted for by comorbid ADHD (Budman et al., 2000; Chen et al., 2013). We investigated whether these findings would emerge when using an experimental task as an alternative to questionnaires and when assessing general ER ability in an eco-

logically valid situation, which potentially underlies the disruptive behavior manifesting in everyday life. As no previous studies have directly investigated ER in children with TS, the basis for comparison is limited. To our knowledge, only one previous study has evaluated ER in adults with TS (Drury, Wilkinson, Robertson, & Channon, 2016). The authors examined self-reported ER in 20 adults with pure TS and 20 healthy controls and found that the group with TS was similar to the control group in the use of reappraisal as an ER strategy, but that they utilized strategies of suppression more often than the controls (Drury et al., 2016). These experimental results support earlier findings by showing that children with ADHD, but not TS, present with difficulties in ER. We could thus confirm with a novel approach that dysregulation of emotions is not inherently part of the presentation in TS in children who do not have a concurrent diagnosis of ADHD.

Parents of children with ADHD showed a higher level of tension. Abundant literature has examined the role of the dyad in child symptoms and parental stress, with mixed results regarding the causality of these associations (Choe, Olson, & Sameroff, 2013; Deault, 2010; Muñoz-Silva, Lago-Urbano, Sanchez-Garcia, & Carmona-Márquez, 2017). Our results support an association between parental behavior and child symptoms; still without addressing causality, i.e., whether parents with high levels of tension are more likely to have children with ADHD, or whether child ADHD creates tense parents. An interesting endeavor for future studies would be to further explore the directionality of this relationship through longitudinal studies to increase options for ameliorating distress in both the parent and the child.

ER difficulties did not correlate with TS symptoms in the present study, whereas earlier studies concerning the association between symptom severity in TS and various aspects of disruptive behavior have produced conflicting results. In the original article on the development of the PUTS, the authors demonstrated significant correlations between various internalizing symptoms and the aggressive subscale measured with the Child Behavior Checklist (CBCL; Achenbach & Rescorla, 2001)

and the YGTSS for children above 10 years of age, but not for younger children (Woods et al., 2005). In an earlier study, CBCL scores (including aggressive behavior) were related to tic severity in individuals with TS and ADHD (Nolan, Sverd, Gadow, Sprafkin, & Ezor, 1996), whereas studies including individuals with TS without comorbid ADHD could not establish a link between tic severity and explosive outbursts (Budman et al., 2000; Stephens & Sandor, 1999). In a large study of the prevalence of intermittent explosive disorder (IED; American Psychiatric Association, 2013) in individuals with TS in the US and Costa Rica, explosive outbursts were associated with both early age of tic onset and higher YGTSS score in the total sample, but not in the US sample in isolation (Chen et al., 2013). Together these findings point to a need for further systematic investigations into the relationship between tic severity and ER difficulties. Our results, however, support the notion that difficulties in ER are not related to the severity of tics and offer an important point to share with families of children with TS. Unlike symptoms of TS, severity of inattention and hyperactivity/impulsivity showed significant, inverse correlations with ER ability. This specific association with ADHD symptoms is interesting and deserves further examination. An interesting focus for future research would be to study this phenomenon longitudinally in individuals with TS to see if ER ability and ADHD symptoms fluctuate in unison, or if one precedes the other.

The identified factors from the EFA were theoretically and clinically meaningful and contributed to the further clarification of emotional dysregulation by pointing to different aspects of the regulatory process. The child items displayed that some items related directly to the regulation of emotions (the verbal strategies and the emotionally laden control), whereas the third factor contained items more simply related to expression of emotions.

Several methodological limitations were apparent in the present study. Although the TS group and the control group were matched on age and sex, the four groups in total were not comparable regarding SES and IQ. In line with other studies, children with ADHD display lower mean IQ com-

pared with typically developing children; however, controlling for IQ in children with neurodevelopmental disorders may be misleading, as scores can become overcorrected and unrepresentative for the general population (Dennis et al., 2009). Also, both ADHD groups had higher rates of comorbid ODD than the TS group, which may have contributed to these children's lower scores of ER ability. The overall rates of ODD, however, were quite low with only nine children in total presenting with comorbid ODD. The four groups differed in size and the groups of children with ADHD and TS + ADHD limit the power of our analyses. The comparison of multiple clinical groups, however, is an important part of understanding the disorder specific contributions to challenges in ER. It should be noted that 12 children in the TS group had additional diagnoses of ODD, OCD, or anxiety disorders causing the group to only partly represent the "pure" TS as the group was only "pure" with respect to ADHD. We decided not to exclude for these comorbidities to ensure representativeness in a disorder with such high comorbidity rates. The majority of studies on TS in all research areas include children who have multiple comorbid disorders, which can pose a challenge to understanding the unique symptomatology of TS. In contrast, the direct clinical relevance of a "pure" TS group can be questioned as the transferability to patient needs will only have direct relevance to a minority of patients. A major limitation was the lack of blinding in the coding of some videos, as the coder had also assessed some of the children. Efforts were made to mask the diagnostic status of these participants, who had been assessed several months or more than a year prior to coding. Generally, blinding is an inherent problem when studying children with TS as they often present with tics during assessment. As a final issue, the factor analysis should be interpreted with caution due to various methodological limitations that were accepted due to its explorative nature. The inclusion of items in the analysis with correlation coefficients lower than .30 and in a factor with correlations below .70 is controversial, as well as circumventing the cut-off for the anti-image matrices of .50 for a few items. The presence of variables with low correlation coefficients may indicate a lack of patterned relationship, which can be problematic in a factor analysis (Yong & Pearce, 2013). Furthermore, it should be not-

ed that a few items cross-loaded on more than one factor, which we believe is partly due to the complex construct that we measure. Although we applied the EFA to a heterogeneous sample as recommended, we did not unequivocally obtain a simple structure of factors (Kline, 2014).

A number of strengths are in evidence for the present study. We examined a large, medication-naïve and clinically well-described group of children through a multi-method approach and with a control group that was matched to the TS group with minimal selection bias due to recruitment via the Danish registers. By employing a novel task in the assessment of ER in children we have contributed to the understanding of the emotional processes that underlie disruptive behavior in developmental TS and ADHD with the objective of ultimately benefitting treatment options. The finding that emotional dysregulation is seemingly distinct from TS in a direct experimental measure furthers our knowledge of the clinical manifestation and a contingent presence of explosive outbursts should prompt a systematic examination to assess comorbidity. For children with ADHD, an effort targeted training of ER competence seems ideal as the present findings once again point to a component of impaired ER in these children.

In conclusion, the present study is the first to directly evaluate ER ability in children with TS while additionally considering the impact of comorbidity and symptom severity. We demonstrate impaired ER ability in children with ADHD with and without TS, but not in children with TS in the absence of ADHD. These findings contribute to the understanding of dysregulated child behavior by using an experimental measure to assess children's ER in a context that has largely been dominated by parent-reported questionnaires, thereby allowing a multi-method approach to investigating ER.

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Table 1

Participant characteristics

Characteristic	TS (n = 49)	ADHD (n = 23)	TS + ADHD (n = 16)	Controls (n = 62)	*p*-value
Mean age, years (*SD*)	9.9 (1.3)	9.6 (1.3)	9.3 (1.1)	9.4 (1.4)	.122[a]
Male, n (%)	39 (79.6)	15 (65.2)	13 (81.2)	49 (79.0)	.510[b]
Mean IQ (*SD*)	102.1 (9.3)	94.7 (8.9)	96.6 (11.2)	103.2 (11.1)	.004[a]
Mean SES (*SD*)	5.7 (1.5)	4.4 (1.8)	5.1 (1.8)	6.1 (1.6)	.010[c]
Comorbidity, n					
ODD (%)	2 (4.1)	5 (21.7)	2 (12.5)	0 (0)	.045[d]
OCD (%)	3 (6.1)	0 (0)	1 (6.25)	0 (0)	.505[d]
GAD (%)	1 (2)	0 (0)	0 (0)	0 (0)	1.000[d]
SAD (%)	2 (4.1)	1 (4.3)	0 (0)	0 (0)	1.000[d]
Specific phobia (%)	4 (8.2)	0 (0)	0 (0)	0 (0)	.379[d]
ADHD subtype, n					
Combined (%)	NA	17 (73.9)	9 (56.3)	NA	.312[d]
Inattentive (%)	NA	6 (26.1)	5 (31.3)	NA	.734[d]
Hyperactive/ impulsive (%)	NA	0 (0)	2 (12.5)	NA	.162[d]

Note. TS = Tourette syndrome. ADHD = Attention-deficit/hyperactivity disorder. SES = Parental socioeconomic status. ODD = Oppositional defiant disorder. OCD = Obsessive compulsive disorder. GAD = Generalized anxiety disorder. SAD = Separation anxiety disorder. [a]One-way ANOVA. [b]Chi-square test of homogeneity. [c]Fisher-Freeman-Halton exact test of independence. [d]Fisher's exact tests.

Table 2

Orthogonally rotated factor loadings for child items

Item	Factor 1: Emotional control	Factor 2: Emotion communication	Factor 3: Verbal emotion regulation strategies
Control	.539		
Reassurance-seeking	-.354		
Avoidance	-.533		
EmReg	.851	.383	
Negative expressions	-.397	-.363	
Emotional warmth		.645	
Aggression		-.563	
Positive expressions		.306	
Narration			.679
Verbal reappraisal			.368
Eigenvalue	2.483	1.594	1.284
% variance explained	24.83	15.94	12.84

Note. Loadings below .30 were suppressed.

Table 3

Correlations between emotion regulation (EmReg score) and symptom severity

	EmReg	
	r (df)	*p*
YGTSS - Total	-.107 (63)	.397
YGTSS minus impairment	-.009 (63)	.941
PUTS – Total	.111 (63)	.377
PUTS – Presence of any urge	-.064 (63)	.612
PUTS – Hold back	.058 (63)	.644
ADHD-RS – Inattention	-.234 (147)	**.004**
ADHD-RS – Hyperactivity/impulsivity	-.211 (147)	**.010**
ADHD-RS – Conduct disorder	-.152 (147)	.065

Note. YGTSS = Yale Global Tic Severity Scale. PUTS = Premonitory Urge for Tics Scale. ADHD-RS = ADHD Rating Scale. ADHD-RS scores are T-scores based on Danish norms. EmReg correlations with a) YGTSS total score; b) YGTSS score minus impairment; c) PUTS total score; d) any presence of premonitory urges represented by a score of 3 or 4 on the PUTS in any of the first six questions representing different urges; e) ability to hold back tics represented by a score of 3 or 4 on the final question of the PUTS ("I am able to stop my tics, even if only for a short period of time"; Woods et al., 2005); f) ADHD-RS subscale inattention; g) ADHD-RS subscale hyperactivity/impulsivity; and h) ADHD-RS subscale conduct disorder. For correlations with YGTSS and PUTS, only the 65 children with TS in total were included in the analysis, whereas correlations with ADHD-RS were conducted for the entire sample except for one control participant with missing data.

Emotion Regulation in Tourette Syndrome

Key points

- Tourette syndrome (TS) is associated with a high occurrence of explosive outbursts, seemingly related to comorbid ADHD.
- Difficulties in emotion regulation (ER) may underlie disruptive behavior, but has previously only been examined through questionnaires in children with TS.
- Using a novel, observational task we examined ER in children with TS, ADHD, TS + ADHD, and typically developing controls.
- We found no difference between the TS group and the control group on ER ability, whereas ER ability in children with ADHD was significantly lower.
- These findings support earlier work with a new methodological framework and point to ADHD as central to the presence of dysregulated behavior, while children with TS perform similarly to controls.

Appendix A

Descriptive statistics for child items

Child items, frequency scores, n	Level	TS (n = 49)	ADHD (n = 23)	TS + ADHD (n = 16)	Controls (n = 62)	*p*
Control (%)	Low	3 (6.1)	3 (13.0)	1 (6.2)	3 (4.8)	.601
	High	46 (93.9)	20 (87.0)	15 (93.8)	59 (95.2)	
Avoidance (%)	Low	36 (73.5)	14 (60.9)	9 (56.2)	48 (77.4)	.236
	High	13 (26.5)	9 (39.1)	7 (43.8)	14 (22.6)	
Narration (%)	Low	41 (83.7)	23 (100.0)	14 (87.5)	52 (83.9)	.226
	High	8 (16.3)	0 (0.0)	2 (12.5)	10 (16.1)	
Verbal reappraisal (%)	Low	43 (87.8)	22 (95.7)	12 (75.0)	56 (90.3)	.231
	High	6 (12.2)	1 (4.3)	4 (25.0)	6 (9.7)	
Reassurance-seeking (%)	Low	21 (42.9)	8 (34.8)	4 (25.0)	23 (37.1)	.627
	High	28 (57.1)	15 (65.2)	12 (75.0)	39 (62.9)	
Aggression (%)	Low	48 (98.0)	23 (100.0)	16 (100.0)	62 (100.0)	.557
	High	1 (2.0)	0 (0.0)	0 (0.0)	0 (0.0)	
Negative expressions (%)	Low	19 (38.8)	7 (30.4)	3 (18.8)	25 (40.3)	.385
	High	30 (61.2)	16 (69.6)	13 (81.2)	37 (59.7)	
Positive expressions (%)	Low	23 (46.9)	8 (34.8)	3 (18.8)	26 (41.9)	.226
	High	26 (53.1)	15 (65.2)	13 (81.2)	36 (58.1)	

EmReg (%)	1	0 (0)	1 (4.3)	0 (0)	0 (0)	**.019**
	2	7 (14.3)	5 (21.7)	5 (37.5)	6 (9.7)	
	3	14 (28.6)	12 (52.2)	7 (43.8)	19 (30.6)	
	4	22 (44.9)	4 (17.4)	2 (12.5)	28 (45.2)	
	5	6 (12.2)	1 (4.3)	1 (6.2)	9 (14.5)	
Factor 1: Emotional control (mean (*SD*))		0.17 (1.02)	-0.49 (0.93)	-0.58 (0.96)	0.20 (0.92)	**.002**
Factor 2: Emotion communication (mean (*SD*))		-0.11 (1.22)	-0.05 (0.93)	-0.16 (1.52)	0.15 (0.59)	.677
Factor 3: Verbal ER strategies (mean (*SD*))		0.15 (1.20)	-0.48 (0.73)	-0.01 (0.94)	0.07 (0.89)	.066

Note. TS = Tourette syndrome. ADHD = Attention-deficit/hyperactivity disorder. EmReg levels: 1 = Very poor, 2 = Poor, 3 = Moderate, 4 = Good, 5 = Excellent.

Appendix B

Descriptive statistics for parent items

Parent items, frequency scores, n	Level	TS (n = 49)	ADHD (n = 23)	TS + ADHD (n = 16)	Controls (n = 62)	*p*
Intrusiveness (%)	Low	45 (91.8)	21 (91.3)	15 (93.8)	46 (74.2)	**.030**
	High	4 (8.2)	2 (8.7)	1 (6.2)	16 (25.8)	
Avoidance (%)	Low	47 (95.9)	23 (100.0)	14 (87.5)	59 (95.2)	.337
	High	2 (4.1)	0 (0.0)	2 (12.5)	3 (4.8)	
Control (%)	Low	5 (10.2)	1 (4.3)	1 (6.2)	6 (9.7)	.829
	High	44 (89.8)	22 (95.7)	15 (93.8)	56 (90.3)	
Verbal reappraisal (%)	Low	45 (91.8)	22 (95.7)	14 (87.5)	52 (83.9)	.384
	High	4 (8.2)	1 (4.3)	2 (12.5)	10 (16.1)	
Negative expressions (%)	Low	49 (100.0)	21 (91.3)	15 (93.8)	56 (90.3)	.178
	High	0 (0.0)	2 (8.7)	1 (6.2)	6 (9.7)	
Positive expressions (%)	Low	18 (36.7)	10 (43.5)	3 (18.8)	24 (38.7)	.426
	High	31 (63.3)	13 (56.5)	13 (81.2)	38 (61.3)	
Tension (%)	Low	48 (98.0)	19 (82.6)	16 (100.0)	62 (100.0)	**.001**
	High	1 (2.0)	4 (17.4)	0 (0.0)	0 (0.0)	
Support sensitivity (%)	Low	1 (2.0)	0 (0.0)	0 (0.0)	4 (6.5)	.325
	High	48 (98.0)	23 (100.0)	16 (100.0)	58 (93.5)	

Note. TS = Tourette syndrome. ADHD = Attention-deficit/hyperactivity disorder.

C. Paper 3

Reappraisal is an Effective Emotion Regulation Strategy in Children with Tourette Syndrome and ADHD.

Julie Hagstrøm, Katrine Maigaard, Anne Katrine Pagsberg, Liselotte Skov, Kerstin Jessica Plessen, Signe Vangkilde.

Manuscript in review

Reappraisal is an Effective Emotion Regulation Strategy in Children with Tourette Syndrome and ADHD

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Abstract

Difficulties in emotion regulation (ER) have been associated with a number of psychiatric disorders emphasizing a need for further clarification of the concept. One type of ER is cognitive reappraisal which involves reframing an emotion to alter the way it is experienced. In the present study, we examined reactivity and reappraisal ability with an experimental computer task in 160 medication-naïve children aged 8-12 divided into four groups: Fifty-eight children with Tourette syndrome (TS), 26 children with attention-deficit/hyperactivity disorder (ADHD), 19 children with TS and ADHD, and 57 typically developing controls. Children rated negative affect following neutral and negative images while being asked to verbally reappraise their experienced negative affect on half the negative trials, allowing for subsequent coding of type of strategy employed. The learned ER strategy of reappraisal was effective across all participants and the ability to reappraise was positively correlated with age, whereas reactivity was not. Groups did not differ in reactivity, regulation success or use of ER strategies. Finally, the use of efficacious strategies was correlated with regulation success, whereas use of a less adaptive strategy related to suppression was associated with reactivity, but not regulation of emotions. Our findings indicate that children regardless of diagnostic status are able to learn and employ an adaptive ER strategy when instructed in the technique which is highly relevant to therapeutic approaches to dysregulated behavior.

Keywords**:** Emotion regulation, Tourette syndrome, ADHD, reappraisal, reactivity

Introduction

The capacity of adaptive emotion regulation (ER) in childhood and adolescence is crucial for engaging in demanding social environments, and recently ER has obtained a key role in models of psychopathology (Aldao, Nolen-Hoeksema, & Schweizer, 2010). In children with Tourette syndrome (TS) explosive outbursts have been described as more disruptive than tics (Leclerc, O'Connor, Forget, & Lavoie, 2011), and in attention-deficit/hyperactivity disorder (ADHD), dysregulated emotions are recognized as a central part of the clinical presentation (Graziano & Garcia, 2016). Further examination of the role of ER ability in these neuropsychiatric disorders is crucial to understanding the processes that underlie disruptive behavior.

ER is a complex concept comprising multiple processes. One widely used conceptualization is the process model of emotion regulation which identifies five regulation strategies that alter the emotion-generative process: Situation selection, situation modification, attentional deployment, cognitive change, and response modulation (Gross, 2014). Known strategies for regulating emotions can be directly related to this model such as distraction as part of attentional deployment, expressive suppression associated with response modulation, and reappraisal as part of cognitive change (McRae, 2016). Cognitive reappraisal is recognized as a potentially highly effective strategy for regulating emotions (Aldao et al., 2010; Dennis & Hajcak, 2009; Hendricks & Buchanan, 2016; Ochsner, Bunge, Gross, & Gabrieli, 2002) and describes the complicated process of "...rethinking the meaning of affectively charged stimuli or events in terms that alter their emotional impact" (Ochsner & Gross, 2008). Contrary to other types of regulation, which may occur automatically and unconsciously, reappraisal is a top-down process closely related to cognitive control. Studies have shown that children down to the age of three and four are capable of reappraising (Hua, Han, & Zhou, 2015; Mischel & Baker, 1975) and that this ability increases with age (McRae, Gross, et al., 2012; Silvers et al., 2012). Delineating reappraisal from the less effective strategy of expressive suppression, previous studies have demonstrated that reappraisal modifies the physiological and emotional response, whereas suppression only reduces the outward response, leaving physiological and self-report measures of negative affect unaffected (Aldao et al., 2010; Hendricks & Buchanan, 2016). Importantly, this distinction is warranted on a neural level as well. Whereas reappraisal is one of the later strategies in the process model, it happens relatively early in the emotion-generative process, with frontal engagement reducing activity in amygdala and insula. Conversely, the frontal engagement in suppression occurs later and leads to an increased activity in the amygdala and the insula (Ochsner & Gross, 2008).

Preceding the process of regulation is the generation of emotions which is largely dependent on an individual's emotional reactivity. Reactivity carries a two-fold meaning reflecting both the threshold at which a stimulus elicits a response in the individual as well as the nature of that response (Carthy, Horesh, Apter, Edge, & Gross, 2010; Silvers et al., 2012). Theories on child temperament highlight the interplay between reactivity and regulation with individual ER style interacting with the natural response to an emotional stimulus (Dennis & Hajcak, 2009). A vast body of literature has studied reactivity in children with ADHD whereas very few studies have examined reactivity in TS. Research into reactivity styles in ADHD points to an increased intensity of emotions and higher reactivity behaviorally and physiologically (Fogleman, Leaberry, Rosen, Walerius, & Slaughter, 2018). In a study of adults with TS, results indicated different interpersonal reactivity styles compared to a control group (Eddy, Macerollo, Martino, & Cavanna, 2015); however, this study was limited by varying comorbid disorders and mixed medication status in the TS group. Regarding ER, both TS and ADHD have been associated with explosive behavior (Graziano & Garcia, 2016) with recent studies pointing to ADHD as decisive in this dysregulation of emotions (Budman & Olson, 2000; Chen et al., 2013).

The aim of the current study was to investigate ER, specifically cognitive reappraisal, and reactivity in four medication-naïve groups of children with TS, ADHD, TS and ADHD combined, and typically developing children as the formation of adaptive ER skills is a crucial part of normal development. We investigated reappraisal as a learned ER strategy and reactivity with an experimental task that has previously been used in typically developing populations. We hypothesized that we would discover a main effect of employing the strategy of reappraisal across all participants and that the groups would show a staircase effect with the control group achieving the highest regulation success scores followed closely by the TS group and ultimately the groups including children with ADHD. For reactivity scores we expected the ADHD groups to score higher than the remaining groups. Finally, we were interested in examining in an explorative manner the effectiveness of different verbal reappraisal strategies and whether the use of strategies differed across groups.

Methods

Participants

We included a total of 160 medication-naïve participants aged 8-12 (M = 9.84, SD = 1.29) in the study: Fifty-eight children with TS, 26 children with ADHD, 19 children with TS and ADHD, and 57 typically developing children (Table 1). As no previous studies could guide the selection of an appropri-

ate sample size, we aimed to be able to detect at least medium sized possible differences in the primary clinical group (children with TS) when compared to the control group (typically developing children). With an a priori alpha value of .05 and assumed power of .80, this required a sample size of at least 51 in each of these two groups. We allocated participants to one of the four groups following systematic multi-informant clinical assessment with the semi-structured diagnostic interview Kiddie-Schedule for Affective Disorders and Schizophrenia, Present and Lifetime Version (K-SADS-PL; Kaufman et al., 1997), the Yale Global Tic Severity Scale (YGTSS; Leckman et al., 1989), and the ADHD Rating Scale (ADHD-RS; DuPaul, Power, Anastopoulos, & Reid, 1998). The YGTSS is a clinician-rated instrument that evaluates number, frequency, intensity, complexity, and interference of tics, and the ADHD-RS is a parent- and teacher-rated questionnaire addressing ADHD symptoms. Psychologists or a medical doctor performed the interviews with the parent(s) and the child separately and reached a diagnosis in consultation with a specialist in child and adolescent psychiatry. Children were eligible for inclusion in one of the clinical groups if they met DSM-IV (American Psychiatric Association, 2000) criteria for either TS or ADHD. Additional diagnoses were acceptable, but we excluded children in the case of severe comorbidity (autism spectrum disorder and psychotic disorders). Further exclusion criteria were birth at gestational age < 37 weeks, IQ below 80, and presence of any neurological condition. IQ was assessed with the Wechsler Intelligence Scale for Children – fourth edition (WISC-IV; Wechsler, 2004). We recruited participants for the clinical groups from the local child and adolescent outpatient clinic and from a pediatric department in the Capital Region of Denmark in the period from 2013 to 2016. Children in the control group did not meet criteria for any lifetime psychiatric disorder, except for enuresis and encopresis, and were randomly selected via the Danish Civil Registration System (Pedersen, 2011) and matched on age and sex with the TS group. We registered the socioeconomic status of the parents of each participant using the International Standard Classification of Education (Unesco, 2012). In the case of different scores for each parent, the highest score would go into the analysis.

Picture Reappraisal Task

Variations of the Picture Reappraisal Task (PRT) have been used in several previous studies (McRae, Gross, et al., 2012; Ochsner et al., 2002, 2004; Silvers et al., 2012) as an experimental measure of reappraisal. In our study, participants were shown 15 neutral images and 30 negative images on a computer screen using E-prime software (Psychology Software Tools, Pittsburgh, PA) and were asked to either look at the pictures and react naturally or to actively decrease negative emotions. The cue for neutral images was always “Look” (responding naturally), whereas the cue for negative images alternat-

ed between “Look” (reactivity) and “Decrease” (reappraisal) creating a total of three conditions: 1) “Look neutral”, 2) “Look negative”, and 3) “Decrease negative”. The cue screen was displayed for three seconds followed by the picture for 10 seconds. Finally, a rating screen would appear for six seconds asking the participants to rate their level of negative affect on a scale from one to five, with one representing complete absence of negative affect and five representing strong negative affect. Contrary to former versions of the task we decided to display the images in blocks of 10 pictures with the cue “Look” and five with the cue “Decrease” to avoid accidentally measuring trial-by-trial switching instead of reappraisal. Aversive pictures were counterbalanced between the two negative conditions to achieve similar normative ratings of valence for each condition (3.34 and 3.37, respectively). This created four versions of the experiment (A, B, C, and D) as a result of combining two distributions of negative images with two sequences (starting with “Look” or “Decrease”, respectively). Six participants were tested on a version of the task that was different from versions A-D regarding order of images and cue time. These children saw the same pictures, and their measures of reactivity and regulation success were not significantly different from the remaining participants. There were no significant differences in distribution of versions across all participants nor within the groups. Pictures were selected from the International Affective Picture System (IAPS pictures 1050, 1120, 1201, 1300, 1321, 1930, 2120, 2130, 2688, 2780, 2810, 2900, 3022, 3230, 3280, 5470, 5820, 5970, 6190, 6300, 7000, 7002, 7004, 7009, 7010, 7041, 7090, 7100, 7140, 7150, 7224, 7380, 7595, 7950, 9050, 9250, 9404, 9421, 9470, 9480, 9490, 9582, 9594, 9600, 9611; Lang, Bradley, & Cuthbert, 2008) and had all previously been used in similar studies with corresponding age groups.

Procedures

This study was part of a larger project approved by the Regional Committee on Health Research Ethics (journal number H-2-2013-085) and the Danish Data Protection Agency (journal number 2017-58-0015). Written informed consent was obtained from caregivers of all participants in accordance with the Declaration of Helsinki. All subjects participated in a large test battery. Psychologists or a research nurse administered the PRT and provided instructions verbatim to decrease instruction bias. All participants underwent a thorough practice session with instructions on how to react naturally and how to reappraise. Suggested strategies were the same for all participants. For all trials in the “Decrease negative” condition the test administrator would give one or two prompts to ensure the child answered timely and to act as a reminder for the children with attention problems: “How can you make this picture better” or “How can you make this picture less unpleasant”. The task was filmed to allow for subsequent coding of

reappraisal strategies. JH conducted the coding of the videos according to the previously published Reappraisal Tactic Coding Guide (McRae, Ciesielski, & Gross, 2012) with the following modifications: In addition to the ten categories from the Coding Guide ('Explicitly positive', 'Change current circumstances', 'Reality challenge', 'Change future consequences', 'Agency', 'Distancing', 'Technical-analytic-problem solving', 'Acceptance', 'Non-specific reappraisal', and 'Failure to reappraise') we added two extra categories ('Negating the negative' and 'No negative affect'). The former category can be associated with suppression strategies as the tactic involves simply saying the picture would be better if the elements causing discomfort weren't there or were replaced with something better. The latter category was included for whenever children explicitly stated that they did not feel any discomfort whatsoever. Whereas the original coding guide used a system of estimating the degree to which each category is used, we directly coded the strategy for each of the 15 reappraisals.

Data Analyses

Statistical analyses were performed using SPSS version 22.0. The raw data employed in the analyses is available upon request, without reservations, to all researchers. We excluded participants with less than two thirds of ratings per condition available (number of missing ratings per condition >5). We assessed differences in number of missing ratings across conditions with the Friedman test with a Bonferroni correction for multiple comparisons and differences in number of missing ratings between groups with the Kruskal-Wallis H test as data was not normally distributed.

Between-group differences in age and sex were assessed with a one-way ANOVA and the chi-square test of homogeneity, respectively. Difference in SES score was assessed with the Fisher-Freeman-Halton exact test of independence. We examined variations in the presence of comorbid disorders in the clinical groups with Fisher's exact test, and for the two groups including children with ADHD we tested for differences in ADHD subtype with the chi-square test of homogeneity where sample size was sufficient (combined subtype) and Fisher's exact test for the remaining two subtypes. Finally, we conducted a one-way ANOVA to examine differences in full scale IQ with Bonferroni-adjusted pairwise multiple comparisons.

We examined regulation and reactivity across all participants. Following the procedure of Silvers and colleagues (2012) we calculated regulation as the percent decrease in negative affect from "Look negative" to "Decrease negative" (("Look negative" – "Decrease negative")/"Look negative" x 100) and reactivity as the percent increase in negative affect comparing "Look neutral" to "Look negative"

("Look negative" – "Look neutral"/"Look neutral" x 100). Due to a non-normal distribution, we performed a Friedman test with Bonferroni corrected pairwise comparisons to investigate differences between ratings of negative affect on conditions one ("Look neutral"), two ("Look negative"), and three ("Decrease negative"). We expected to find a higher rating of negative affect in the "Look negative" condition compared to the "Look neutral" as a representation of emotional reactivity, and we expected lower ratings on the "Decrease negative" condition compared to the "Look negative" as a representation of ER. We examined correlations between age and reappraisal and reactivity, respectively, using Pearson's product-moment correlation and expected to find a medium correlation between age and reappraisal ability. Finally, although the PRT is not a speeded task, we examined differences in reaction times (the time from the picture disappears from the screen until the participant presses a button with his or her rating of negative affect) across all participants with a one-way repeated measures ANOVA to assess an overall effect of condition on reaction time and post hoc analyses with a Bonferroni correction where relevant. We expected an increasing staircase effect with condition one showing the shortest reaction times followed by condition two, indicating that the negative affect associated with looking at aversive images would result in longer reaction times. Ultimately, we hypothesized that the process of reappraisal in condition three would result in the longest reaction times.

Following the procedures for the investigation of main effects across all participants, we examined ratings across conditions within groups to see if the individual groups mirrored the total participant sample. We furthermore conducted an ANCOVA controlling for age for the analysis of group differences in regulation success, as previous studies have suggested that ER improves with age up to late adolescence (Silvers et al., 201, and ran a one-way ANOVA for the analysis of reactivity across groups.

As data was not normally distributed, we conducted a Kruskal-Wallis H test to examine differences between groups in the use of strategies. We examined group differences for each individual category and for categories one to nine combined representing the total use of a potentially advantageous strategy and not including the strategies 'Failure to reappraise', 'Negating the negative', or 'No negative affect'. Due to a non-normal distribution of data, we conducted Spearman's rank-order correlations including all participants in the analyses to assess the relationship between regulation success, reactivity, and age and the use of effective and ineffective strategies. Videos were missing from 14 participants (five with TS and three from each of the remaining groups) who could thus not be included in the analyses.

Results

Participant Characteristics

The four groups did not differ significantly on age or sex (Table 1). Groups differed concerning parental SES with the control group presenting with the highest SES followed by the TS group, the TS + ADHD group, and the ADHD group, $p = .001$. The three clinical groups did not differ significantly on comorbidity and for the two groups including children with ADHD, there was no significant difference in ADHD subtype. IQ was significantly different across groups ($F(3, 156) = 4.919$, $p = .003$). Post hoc analyses revealed that this result was driven by significantly lower scores in the ADHD group compared to the control group ($MD = 9.048$, 95% CI [2.52, 15.57], $p = .002$) and the TS group ($MD = 7.501$, 95% CI [0.99, 14.01], $p = .015$). These differences remained significant when controlling for SES. Five participants in total were excluded due to missing ratings (two from the TS group, two from the TS + ADHD group, and one from the control group). One additional participant from the control group was excluded due to a markedly deviant response profile which suggested that the rating had been reversed. There was no significant difference in number of missing ratings across conditions or between groups.

Main Effects of Condition and Associations with Age and Reaction Times

Ratings of negative affect were significantly different across conditions, $X^2(2) = 245.116$, $p < .001$. Bonferroni corrected post hoc analysis revealed significantly different median ratings of negative affect following a natural response to neutral images ($Mdn = 1.00$) compared to negative images ($Mdn = 2.50$; $p < .001$). As expected, ratings of negative pictures following reappraisal ($Mdn = 2.13$) were lower than ratings following a natural response to negative images ($p < .001$). We found a small, but significant correlation between age and reappraisal ability ($r(158) = .227$, $p = .004$) and no correlation between age and reactivity ($r(158) = .101$, $p = .203$). For the analysis of reaction times, the assumption of sphericity was violated as assessed by Mauchley's test ($X^2(2) = 22.058$, $p = < .001$) and we thus applied a Greenhouse-Geisser correction for the one-way repeated measures ANOVA ($\varepsilon = .885$). Reaction times were significantly different for the three conditions ($F(1.769, 281.341) = 212.721$, $p < .001$, partial $\eta^2 = .572$) with reaction time increasing from looking at neutral pictures ($M = 1712.91$, $SD = 33.73$), to negative pictures ($M = 2001.02$, $SD = 37.21$), to finally reappraising negative pictures ($M = 2446.53$, $SD = 42.99$). Pairwise comparisons revealed that reaction time increased significantly from "Look neutral" to "Look negative" ($M = 288.11$, 95% CI [216.65, 359.57], $p < .001$) and from "Look negative" to "De-

crease negative" (M = 445.51, 95% CI [358.05, 532.98], $p < .001$). For all groups, mean reaction times increased with condition (Table 3).

Group Differences in Regulation and Reactivity

Ratings of negative affects differed between conditions for all groups, similar to the results for the total sample (Figure 1). For all groups, ratings were significantly higher when looking at negative pictures compared to neutral pictures and significantly lower when reappraising negative pictures compared to looking at negative pictures. As the only exception, children in the ADHD group did not significantly decrease their ratings of negative affects when asked to reappraise compared to reacting naturally.

For the analysis of differences in regulation we first addressed the assumptions of the ANCOVA. We did not find a linear relationship between the dependent variable and the covariate as assessed by visual inspection of a scatterplot, however, there was homogeneity of regression slopes as the interaction term was not statistically significant, $F(3, 152) = 0.335$, $p = .800$. Standardized residuals were normally distributed as assessed by Shapiro-Wilk's test ($p > .05$) and there was homoscedasticity as assessed by visual inspection of the standardized residuals plotted against the predicted values. Levene's test showed homogeneity of variance ($p = .764$) and there were two outliers in the data defined as cases with standardized residuals smaller or greater than three standard deviations. We found no reason to exclude these cases based on behavior during the test and the overall result did not change markedly when running the analysis without outliers. After adjusting for age, we did not find a statistically significant difference between groups on regulation, $F(3, 155) = 1.063$, $p = .367$, partial $\eta^2 = .020$ (Table 2).

Reactivity scores were not normally distributed, as assessed by Shapiro-Wilk's test ($p < .05$) and there were some outliers based on visual inspection of a boxplot. We decided to carry on with the ANOVA, as it is robust to violations of normality and outliers. There was homogeneity of variances as assessed by Levene's test ($p = .320$). We did not find a statistically significant difference between groups on reactivity, $F(3, 156) = 1.964$, $p = .122$ (Table 2).

Reappraisal Strategies

We did not find any group differences in choice of strategies when asked to reappraise. However, we did find a significant, positive medium sized correlation across all participants between total number of theoretically effective strategies employed and regulation success (Table 4). This correlation

was driven primarily by the specific strategy ‘Change current circumstances’. We did not find a correlation between the theoretically ineffective suppression-like strategy ‘Negating the negative’ and regulation success, nor did we find a correlation between number of effective strategies employed and reactivity. Higher reactivity scores were associated with more frequent use of the strategy reflecting suppression. In accordance with the previously described correlation between age and regulation success, we also found a significant correlation between age and number of effective strategies employed, which was once again driven by the specific strategy ‘Change current circumstances’ (Table 4).

Discussion

In the present study we examined profiles of emotion regulation and reactivity in four groups of clinically well-described and medication-naïve children aged 8-12 years with TS, ADHD, TS and ADHD, and typically developing controls to increase our understanding of the mechanisms underlying disruptive behavior in these disorders. The inclusion of more than one diagnostic group is important to systematically identify the processes that are associated with general psychopathology and the symptoms specific to one disorder (Aldao, 2016). The results for all participants corroborate the position of reappraisal as an effective strategy for ER. Unlike simpler forms of regulation such as suppression and distraction, reappraisal is a cognitively demanding strategy which is highly reliant on age (McRae, Gross, et al., 2012; Silvers et al., 2012). The current study supports earlier findings by demonstrating a correlation between age and regulation success, but not between age and reactivity. As expected, we did not find any statistically significant group differences in regulation ability between the TS group and the control group; nor did we find an overall difference between the groups with ADHD and the control group, which was unexpected. We did see, however, that only the ADHD group did not decrease negative affect significantly following reappraisal, which may be indicative of a reduced ability to regulate emotions. We did not find elevated reactivity in the ADHD groups as hypothesized. For specific regulation strategies, there were no differences between the groups in types of strategies employed, but for all participants we found that the summated use of effective strategies correlated with ultimate regulation success.

From the unadjusted descriptives it appears that we do in fact see the expected pattern of regulation success with the control group achieving the highest score, the ADHD group the poorest score, and the TS groups positioned along this spectrum. As the task itself requires a certain level of attention (Ochsner & Gross, 2008) it is plausible that the children with ADHD would be additionally challenged.

Interestingly, however, we did not find any significant differences across groups in regulation ability when controlling for age. One explanation for this might be that the images were not sufficiently unpleasant and therefore unable to differentiate the groups. Our study's age span made it challenging to select images that would create negative affect in a twelve-year-old without traumatizing an eight-year-old and, as mentioned, all pictures had been pre-screened by parents in a previous study thereby removing some of the more aversive ones. Another explanation might stem from the nature of this specific type of ER. Unlike spontaneously occurring regulation in everyday life or studies applying experimental tasks that allow for unguided regulation, the PRT addresses reappraisal as the sole potential strategy, furthermore incorporating an element of learning. It may be that children regardless of group are able to learn to apply this strategy when explicitly instructed how to do so. This finding is in line with an earlier study of similarly guided reappraisal in children with anxiety where the authors did not find any difference in reappraisal ability compared to a control group (Carthy et al., 2010). Previous studies have demonstrated maladaptive ER in children with ADHD (Lugo-Candelas, Flegenheimer, Harvey, & McDermott, 2017; Shushakova, Ohrmann, & Pedersen, 2018), but our results could indicate that children with attentional problems are capable of training their regulation ability when guided, which may prove relevant in treatment or in demanding settings such as school. Although we lack a baseline measure of reappraisal ability preceding the practice session, this hypothesis could further delineate the cognitively demanding reappraisal from children's spontaneous ability to regulate, which was identified as an important question to pursue in a recent article (Ahmed, Bittencourt-Hewitt, & Sebastian, 2015).

Contrary to our hypothesis, the ADHD groups did not display heightened reactivity; in fact, the control group presented with the highest mean level, although this was not significantly different from the other groups. An explanation for the lack of finding may be that heightened reactivity is part of adolescence even without comorbidity (Ahmed et al., 2015) and that high reactivity does not necessarily pose a problem if the individual is able to adaptively regulate these emotions to regain emotional equanimity. One possible explanation for the low reactivity scores in the ADHD groups could be that the emotional stimuli need to be more powerful to create a response in the ADHD group. Specifically, children with ADHD are generally more exposed to negative interactions with parents and teachers (Johnston & Jassy, 2007) and peers (Landau, Milich, & Diener, 1998) which in turn might create a higher threshold for negative input due to habituation. It would be interesting for future studies to look further into reactivity styles, as our results were not able to replicate the findings of increased reactivity in chil-

dren with ADHD, and into the relationship between reactivity and regulation to better understand their individual contributions to everyday disruptive behavior.

The finding that effective strategy use was directly associated with regulation success further supports reappraisal as an adaptive strategy. Specifically, we found an interesting dissociation, where the most highly used advantageous reappraisal strategy "Change current circumstances" predicted reappraisal success, but not emotional reactivity, while the use of the typically maladaptive, suppression-associated strategy "Negating the negative" predicted emotional reactivity, but not reappraisal success. Results indicate that different strategies might vary in their efficiency. In our study, the strategy of reinterpreting the meaning of the emotionally laden situation ("Change current circumstances"), which is in many ways the essence of reappraisal, proved most efficient, whereas another frequently used strategy of challenging the reality of the scene by thinking the unpleasant stimulus was fake or staged by actors, did not have a direct effect on regulation success. It might be that challenging the reality is a less demanding strategy not different from merely negating the negative stimulus and that the higher cognitive involvement is more efficient in modifying negative affect. Although we did not see any group differences in the use of strategies, an interesting meta-analysis on emotion regulation strategies across psychopathology found that the frequency of use of maladaptive strategies (rumination, avoidance, and suppression) was positively correlated with level of psychopathology, and conversely that adaptive strategies (acceptance, reappraisal, and problem solving) were associated with less psychopathology (Aldao et al., 2010). One reason we did not find this may be that these differences in spontaneous regulation are attenuated to some degree under guidance, once again pointing to a potential gain from practicing adaptive strategies in children with dysregulated behavior.

A premise for studying emotion regulation in a developmental sample is the concurrent individual cognitive and brain structural and functional development that affects regulation ability. Reappraisal is thought to rely on interactions between prefrontal and cingulate regions involved in cognitive control and the insula and amygdala involved in emotional processing (Ochsner & Gross, 2008); structures which undergo developmental changes during adolescence (Ahmed et al., 2015). Additionally, the development in the brain parallels the development of cognitive processes involved in ER such as executive functions and it would be interesting to further investigate the relationship between ER strategies and neurocognitive abilities (Ahmed et al., 2015). Specifically, it would contribute to the understanding of the cognitive/executive component in ER to examine whether the capacity of reappraisal co-develops with specific cognitive domains. Furthermore, it would be of great interest to investigate ER abilities

considering brain maturation to explore whether certain levels of maturation are required for certain types of regulation and whether this relationship is mediated by enhanced cognitive abilities following brain maturation.

There were a number of limitations to the current study such as difference in group sizes and the small combined TS + ADHD group. While such a group could provide important insights into the shared and specific contributions to regulation ability in a comorbid group compared to single diagnosis groups, the small sample size severely limits the power of analyses directly involving the group. The groups differed significantly on SES and IQ, but we chose not to include IQ as a covariate as this may lead to unrepresentative groups by either having a neurodevelopmental sample with IQ higher than the population mean or a control group with IQ lower than the population mean (Dennis et al., 2009). One major limitation was the lack of blinding. Test administrators were not blinded to diagnostic status, and the coder was not blinded when assessing strategy use. Ratings of negative affect, which were the primary outcome, were not affected by the lack of blinding as this measure was self-evaluated by the participants. Self-report is fundamental to understanding the child's emotional situation and earlier studies have found that self-reported negative affect following reappraisal was associated with corresponding physiological and neural changes (Silvers et al., 2012). However, this method also created some bias as self-report is known to be susceptible to mood and requires metacognitive abilities, and the experience of the emotion can be mixed with the experience of the regulation. Moreover, one study with adults found that test participants often relied on another strategy than the one they were instructed to use (Demaree, Robinson, Pu, & Allen, 2006). Future studies would benefit from a multi-method approach incorporating physiological measures as a more objective basis for comparison. An additional limitation related to the task was the attempt to create an appropriate amount of negative affect in all age groups which led to some pictures simply not producing the desired negative affect in all children. Ideally, aversive images should be tailored to the child's individual arousal threshold to ensure all children experience the same level of negative affect.

In conclusion, although research has suggested that the symptomatology of ADHD and to a lesser extent TS is frequently accompanied by dysregulated emotional behavior, we did not in the present study find any group differences in reappraisal ability when children were trained in the procedure. The lack of group differences in guided ER ability is of special relevance to therapeutic approaches to children with these disorders who experience difficulties with spontaneous, unguided emotion regulation,

although from the present study we are unable to comment on the long-term effects of this training or how it might transfer to everyday problems with disruptive behavior.

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Conflict of interest

The authors have no conflicts of interest to disclose.

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Table 1

Participant Characteristics

Characteristic	TS (n = 58)	ADHD (n = 26)	TS + ADHD (n = 19)	Controls (n = 57)	*p*
Mean age (*SD*)	9.9 (1.3)	9.6 (1.1)	9.6 (1.3)	9.9 (1.3)	.570
Male, n (%)	48 (82.8)	19 (73)	14 (73)	47 (82.5)	.627
Mean IQ (*SD*)	102.2 (9.7)	94.7 (8.6)	99.6 (10.8)	103.7 (11.4)	.003
Mean SES (*SD*)	5.7 (1.4)	4.4 (1.8)	4.9 (1.8)	6.0 (1.4)	.001
Comorbidity					
ODD, n (%)	4 (6.9)	5 (19)	4 (21)	0 (0)	.103
Conduct disorder, n (%)	0 (0)	1 (3)	1 (5)	0 (0)	.188
OCD, n (%)	3 (5.2)	0 (0)	2 (10)	0 (0)	.293
GAD, n (%)	2 (3.4)	0 (0)	0 (0)	0 (0)	1.000
SAD, n (%)	2 (3.4)	1 (3)	0 (0)	0 (0)	1.000
Specific phobia, n (%)	4 (6.9)	0 (0)	0 (0)	1 (1)	.380
Tics NOS, n (%)	0 (0)	1 (3)	0 (0)	0 (0)	.437
ADHD subtype					
Combined, n (%)	NA	20 (76)	11 (57)	NA	.173
Inattentive, n (%)	NA	6 (23)	6 (31)	NA	.524
Hyperactive/ impulsive, n (%)	NA	0 (0)	2 (10)	NA	.173

Note. TS = Tourette syndrome. ADHD = Attention-deficit/hyperactivity disorder. SES = Parental socio-economic status. ODD = Oppositional defiant disorder. OCD = Obsessive compulsive disorder. GAD = Generalized anxiety disorder. SAD = Separation anxiety disorder. NOS = Not otherwise specified. NA = Not applicable.

Table 2

Mean Scores of Reactivity and Regulation

		Reactivity		Regulation			
				Unadjusted		Age-adjusted	
Group	N	M	SD	M	SD	M	SE
TS	58	46.46	17.81	9.33	19.13	9.09	2.31
ADHD	26	42.65	21.47	4.57	18.26	6.19	3.57
TS + ADHD	19	44.51	21.52	13.81	15.27	14.20	4.05
Controls	57	52.24	18.04	13.91	16.00	12.22	2.36

Note. TS = Tourette syndrome. ADHD = Attention-deficit/hyperactivity disorder.

Table 3

Mean Reaction Times Across Conditions

	TS	ADHD	TS + ADHD	Controls
Mean RT (ms), "Look neutral" (SD)	1680.1 (426.7)	1969.6 (436.5)	1765.0 (480.4)	1609.3 (356.3)
Mean RT (ms), "Look negative" (SD)	2012.6 (488.6)	2121.0 (486.2)	2036.4 (457.0)	1919.5 (445.2)
Mean RT (ms), "Decrease negative" (SD)	2454.9 (561.4)	2451.7 (624.3)	2443.9 (424.8)	2435.8 (534.7)

Note. TS = Tourette syndrome. ADHD = Attention-deficit/hyperactivity disorder. RT = Reaction time.

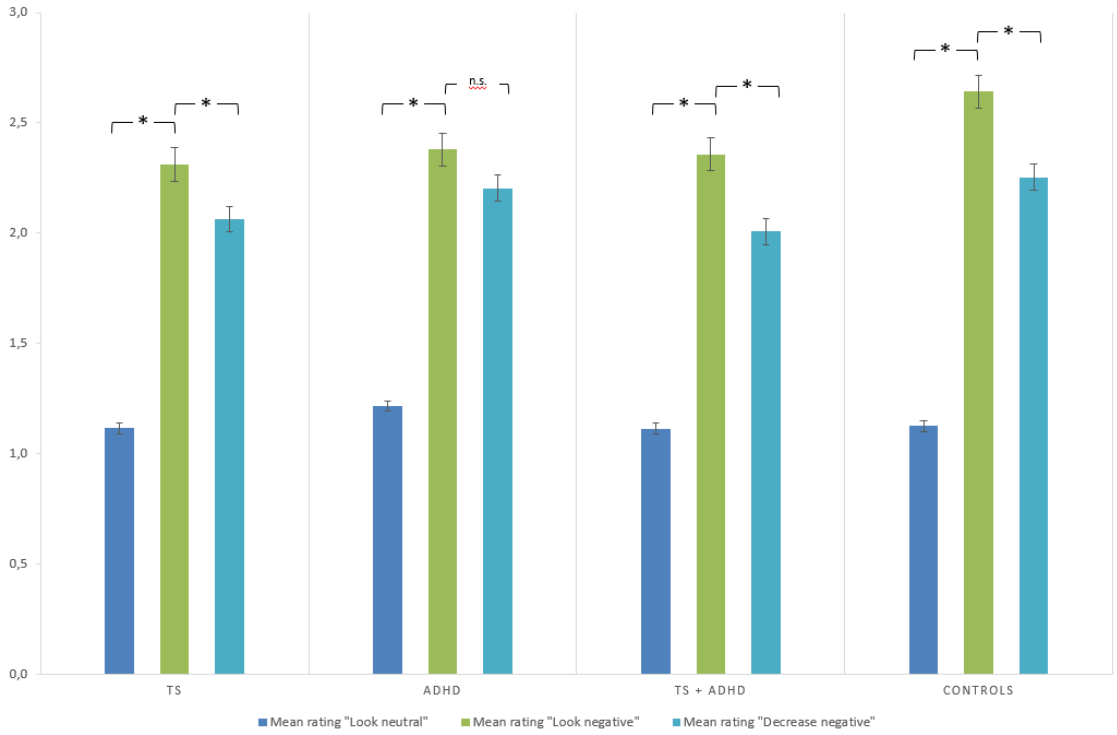
Table 4

Correlations Between Regulation Success, Reactivity, and Age and Reappraisal Strategies

	Total reappraisal		'Change current circum-stances'		'Negating the negative'	
	r_s (df)	*p*	r_s (df)	*p*	r_s (df)	*p*
Regulation success	.322 (144)	**<.001**	.317 (103)	**.001**	-.103 (102)	.300
Reactivity	.019 (144)	.818	.188 (103)	.055	.259 (102)	**.008**
Age	.261 (144)	**.001**	.297 (103)	**.002**	-.120 (102)	.225

Note. Total reappraisal = Strategies from category one to nine combined. df = Degrees of freedom.

Figure 1. Mean ratings across groups. TS = Tourette syndrome. ADHD = Attention-deficit/hyperactivity disorder. N.s. = Not significant. Error bars represent the standard error.



D. Co-authorship declarations

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DECLARATION OF CO-AUTHORSHIP

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Title of PhD thesis:
Emotion Regulation in Children with Tourette Syndrome

This declaration concerns the following article:
The Puzzle of Emotion Regulation – Development and Evaluation of the Tangram Emotion Coding Manual for Children

The PhD student's contribution to the article: *(please use the scale (A,B,C) below as benchmark*)*	(A,B,C)
1. Formulation/identification of the scientific problem that from theoretical questions need to be clarified. This includes a condensation of the problem to specific scientific questions that is judged to be answerable by experiments	B
2. Planning of the experiments and methodology design, including selection of methods and method development	B
3. Involvement in the experimental work	B
4. Presentation, interpretation and discussion in a journal article format of obtained data	C

**Benchmark scale of the PhD student's contribution to the article*		
A. refers to:	*Has contributed to the co-operation*	*0-33 %*
B. refers to:	*Has contributed considerably to the co-operation*	*34-66 %*
C. refers to:	*Has predominantly executed the work independently*	*67-100 %*

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This declaration concerns the following article:
Emotion Regulation in Children with Tourette Syndrome – Associations with ADHD and Tic Severity

The PhD student's contribution to the article: *(please use the scale (A,B,C) below as benchmark*)*	(A,B,C)
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Emotion Regulation in Children with Tourette Syndrome

This declaration concerns the following article:
Reappraisal is an Effective Emotion Regulation Strategy in Children with Tourette Syndrome and ADHD

The PhD student's contribution to the article: *(please use the scale (A,B,C) below as benchmark*)*	(A,B,C)
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