Executive Functions in Preschool Children with ADHD and/or DBD

Assessment, Development and Role of Environment

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Executive Functions in Preschool Children with ADHD and DBD:
Assessment, Development and Role of Environment

Executieve Functies van kleuters met ADHD en DBD:
Onderzoek, Ontwikkeling en Rol van de Omgeving

(met een samenvatting in het Nederlands)

Proefschrift

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Kim Schoemaker

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Voor mama,
in herinnering aan papa

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General Introduction

Executive functions (EF) can be defined broadly as the top-down control of cognitive processes to achieve a purpose or goal (Séguin&Zelazo, 2005). EF include mental processes such as working memory, inhibition of inappropriate responses and flexibility in adaptation to changes (Nigg, 2006). Research has shown that EF are associated with various developmental outcomes such as academic achievement (Bull, Espy,&Wiebe, 2008), emotion regulation (Carlson&Wang, 2007) and theory of mind understanding (Hughes, 2011). Alongside, EF impairments are associated with different developmental disorders, such as autism (Robinson, Goddard, Dritschel, Wisley,&Howlin, 2009). School-age children and adolescents with attention deficit hyperactivity disorder (ADHD) and disruptive behavior disorder (DBD) also show EF impairments (Oosterlaan, Logan,&Sergeant, 1998; Willcutt, Doyle, Nigg, Faraone,& Pennington, 2005). However, knowledge is limited regarding preschool children, and especially clinically diagnosed preschool children with ADHD and DBD. This dissertation aims to study EF and the development of EF in preschool children with ADHD and/or DBD, as well as the role of the caregiving environment on the development of EF.

ASSESSMENT OF EF IN PRESCHOOL CHILDREN

EF tasks

There are several challenges related to the assessment of EF in the preschool period. Foremost, the characteristic behavior of young children, like distractibility and fluctuation in motivation levels, complicates the assessment of EF in preschoolers (Anderson& Reidy, 2012). Initially, tasks for EF in young children were adapted from EF tasks for adults, but the last decade tasks have been designed for preschoolers (Hughes, 2011). These child friendly tasks are appealing to young children and take into account the developmental level of children, like the amount of behavior responses and complexity of explanation of the task (Carlson, 2005; Espy, Kaufmann, McDiarmid& Glisky, 2001; Hughes, 2011). However, these requirements of novelty and age appropriateness are not limited to the individual tasks, but also apply to the whole assessment session. This has practical implications like the time of the day of the assessment and alternation in demands to keep the child's interest and attention (Anderson& Reidy, 2012).

Much research is needed to develop and organize a battery of EF tasks that encompass the range of EF abilities *and* are appealing to preschoolers (Anderson & Reidy,2012). Therefore, we chose to use EF tasks that were specifically developed for preschool children in the Developmental Cognitive Neuroscience Laboratory, Department of Psychology, University of Nebraska-Lincoln. In view of the specific aims of our study and the time constraints for the assessment of EF, Dr. Espy and Dr. Wiebe suggested a number of tasks to be used in our study, and provided training on how to administrate, score and interpret the tasks. We included EF tasks that were designed to measure working memory, inhibition and cognitive flexibility, varying in their motivational demands (Wiebe et al., 2011). The EF tasks were administrated in a fixed order, to alternate between specific EF components, between computer and non-computer tasks, between tasks with or without a reward, and between tasks with verbal and non-verbal responses.

Structure of EF

EF tasks also include non-executive abilities, such as language and motor skills. A number of studies have been conducted using a battery of tasks that share a requirement for EF, but the tasks differ in other stimulus and response demands. By using confirmatory factor analysis it is possible to parse common EF variance from that attributable to other sources and examine the latent construct representing a 'purified' EF measure (Wiebe et al.,2011). Studies using confirmatory factor analysis found only one unitary executive construct in preschool children (Hughes, Ensor, Wilson, & Graham, 2010; Wiebe et al., 2011; Willoughby, Blair, Wirth, & Greenberg, 2010). In contrast, studies using exploratory factor analysis identified three (Hughes, Dunn, & White, 1998) or four (Espy, Kaufmann, McDiarmid, & Glisky, 1999) separable EF components in preschool children.

In adulthood, Miyake et al. (2000) proposed that EF are a unitary construct with three dissociable components: working memory, inhibition and cognitive flexibility. Working memory refers to keeping information in mind over a delay and in more complex tasks manipulating that information. Inhibition refers to withholding or delay of a dominant or prepotent response. Cognitive flexibility refers to learning a rule and subsequently shifting to a new rule or mental set. Although the structure of EF that reflects both unity and diversity appears to be applicable from middle childhood onward (Wiebe et al.,2011), the structure during early childhood is not yet clearly defined. Studies indicate that in the preschool period EF may be characterized by a single factor structure and become increasingly differentiated with age (Hughes, 2011; Wiebe et al., 2011). In our study we examined the factor structure in a preschool sample with predominantly clinically diagnosed children.

DEVELOPMENT OF EF IN PRESCHOOL CHILDREN

In their review, Garon, Bryson and Smith (2008) propose that the EF components develop hierarchically during the preschool period. According to their model, the first developing EF component is working memory, followed by inhibition, and both skills are integrated later in cognitive flexibility. Empirical studies on the development of EF in typically developing (TD) young children showed the growth in performance on a general EF factor in children between the ages of three and six years (Hughes et al., 2010; Willoughby, Wirth, & Blair, 2012). Specifically, inhibition also improved in young children between two and five years at risk for externalizing behavior problems (Dennis, Brotman, Huang, & Gouley, 2007; Moilanen, Shaw, Dishion, Gardner, & Wilson, 2011).

EF IN PRESCHOOL CHILDREN WITH ADHD AND DBD

As EF are associated with goal-directed behavior, implications of EF deficits for impaired social functioning have been investigated. For example, poor executive working memory in school-aged children is found to be associated with peer rejection and poor overall social competence, as well as with specific social impairments, i.e., physical and relational aggression and poor conflict resolution skills (McQuade et al., 2013). Likewise, individual differences in inhibitory control are associated with children's ability to regulate their, positive as well as negative, emotional expression (Carlson & Wang, 2007). These studies, conducted with TD children, show that EF are related to normal variation in social and emotional functioning. We are specifically interested in how EF are related to the more extreme end of inadequate social functioning in preschool children, such as in those diagnosed with ADHD and/or DBD.

Although (mild forms of) externalizing behavior is a common feature in preschool children, there is increasing evidence that diagnoses of ADHD and DBD can be made reliably in preschool children (Keenan et al.,2011). ADHD is characterized by a pattern of hyperactivity, impulsivity and/or inattention. DBD encompasses oppositional defiant disorder (ODD) and conduct disorder (CD). ODD is defined as a pattern of negativistic, defiant, disobedient and hostile behavior toward authority figures, whereas CD is characterized a as a repetitive and persistent pattern of behavior in which the basic rights of others or major age-appropriate societal norms or rules are violated (DSM-IV-TR, 2000). The prevalence rates of ADHD and DBD in preschool children are similar to those later in childhood (Egger & Angold, 2006; Lahey,Miller,Gordon,& Riley,1999).The role of co-morbidity has been largely ignored, despite the fact that about half of the children with ADHD are also diagnosed with ODD, and the percentage of children with ODD who have co-morbid ADHD is even higher (Kutcher et al., 2004). There is a high correlation (0.80) between ADHD and DBD symptoms in preschoolers (Sterba, Egger, & Angold, 2007).

An important issue is the difficulty to differentiate between clinical and normative behavior in the preschool period. For example, 'losing temper' is a symptom of ODD but is not uncommon behavior in a preschooler (Keenan et al., 2007). Therefore, the clinical assessment of preschool externalizing behavioral problems is particularly complex and subtle (Wakschlag et al.,2005). The complexity of diagnosing DBD and ADHD in preschool children requires a combination of information about the child's behavior not only from a parent interview and a teacher/caregiver questionnaire, but also from an observation of the child (Wakschlag et al., 2008a; 2008b).

There is a large body of research on EF in school-age children and adolescents with ADHD and/or DBD. Impaired inhibition, working memory and cognitive flexibility performance have been shown in school-age children and adolescents with ADHD (Oosterlaan et al., 1998; Willcutt et al., 2005). Impaired inhibition has also been shown in children with DBD (Oosterlaan et al., 1998), specifically when motivational processes, i.e., reward and punishment, are involved (Fairchild et al., 2009; Matthys, Van Goozen, De Vries, Cohen-Kettenis, & Van Engeland, 1998; Matthys, Van Goozen, Snoek, & Van Engeland, 2004). Inconsistent results have been found regarding working memory and cognitive flexibility performance (Morgan & Lilienfeld, 2000; Oosterlaan, Scheres, & Sergeant, 2005). There is especially controversy about whether EF deficits in children with DBD are due to the presence of co-morbid ADHD (symptoms).

It is not clear whether preschool children with diagnosed externalizing clinical disorders show EF deficits similar to those observed in diagnosed school-age children, or whether such deficits do not emerge until later in development, particularly as assessing EF in preschoolers is not straightforward. Programs to train EF skills in young children have been developed and proved to increase EF performance (Diamond & Lee, 2011). Thus, if EF deficits occur already at preschool-age in children with externalizing behavior problems, these children may benefit particularly from such training. The last years there is an increasingly amount of research on EF in young children with externalizing behavior problems. Recently, the first meta-analysis was published in which neuropsychological deficits in preschoolers at risk for ADHD were examined (Pauli-Pott & Becker, 2011). We extended this work by conducting a meta-analysis in which we investigated externalizing behavior problems in preschool children from a broader perspective, i.e., in addition to young children with ADHD symptoms, we also included studies in young children with aggressive and oppositional symptoms. Furthermore, we conducted a study in preschool children with a clinical diagnosis of ADHD and/or DBD.

ROLE OF CAREGIVING ENVIRONMENT

Although the development of EF is assumed to be dependent on maturation of the cerebral cortex, a body of research in TD children has shown that caregiving environmental factors affect the development of EF as well. Indeed, positive parental control practices have been found to be associated with higher EF performance in TD children (Bernier, Carlson, & Whipple, 2010; Hughes & Ensor, 2009). Furthermore, maternal depression adversely affects children's EF performance (Hughes, Roman, Hart, & Ensor, 2013). It seems relevant to examine the relation between the caregiving environment and EF in preschool children with ADHD and DBD, since parents of children with ADHD and/or DBD tend to show high levels of negative controlling parenting practices, and high levels of depression, stress and ADHD symptoms (for reviews see Capaldi, DeGarmo, Patterson, & Forgatch, 2002; Deault, 2010; Johnston & Mash, 2001). Therefore, we examined the role of the caregiving environment on the development of EF in children with ADHD and/or DBD.

OUTPATIENT CLINIC

The aim of this dissertation was to study EF, the development of EF, and the role of the caregiving environment on the development of EF in preschool children with ADHD and/or DBD. To address these aims, we started an Outpatient Clinic for Preschool Children with Externalizing Behavioral Problems at the Department of Child and Adolescent Psychiatry, University Medical Center Utrecht in 2007. In this outpatient clinic, clinical practice and research were integrated resulting in mutual benefits. Children were assessed by a multidisciplinary team in one morning, and in order to evaluate their development they were re-assessed after 9 and 18 months. The advantage for the referred children and their parents was that no separate appointments for psychological and psychiatric assessments were needed, and that clinical decisions about starting or adapting interventions were based on longitudinal data. The advantage from the research perspective was that longitudinal data were collected, and, since clinical care was provided, the retention rate was very high.

Children were referred by general practitioners, well-baby clinics and pediatricians. Inclusion criteria were age between 3 years and 6 months and 5 years and 6 months and a score at or above the 90^{th} percentile either on the Attention Problems scale or the Aggressive Behavior scale of the Child Behavior Checklist completed by parents (CBCL/1,5 - 5) or the Child Teacher Report Form completed by teachers or day-care caregivers (C-TRF /1,5 - 5; both: Achenbach & Rescorla, 2000; Dutch version by Verhulst & Van der Ende, 2000). Furthermore, children with an IQ below 70 were excluded, estimated with the Raven colored progressive matrices (Raven, Court, & Raven, 1998) and the Peabody Picture Vocabulary-III-NL (Dunn & Dunn, 2005; Dutch translation by Schichtling, 2005).

Children were evaluated in a single, morning session. For an overview of the morning and the instruments see *Figure 1*. First, an interview with the parents regarding the child's behavior took place with the child psychiatrist or clinical child psychologist. Second, two measures of intellectual functioning were administered, followed by the EF tasks. We included EF tasks that were designed to measure working memory, inhibition and cognitive flexibility, for a detailed description of the EF tasks see chapter 3.

All tasks were administered individually by trained master students in a quiet room with a one-way mirror. One parent was in the room with the child and the assessment was recorded. If a second parent was present they could view the child from behind the one-way mirror. The parents completed questionnaires during the time the children performed the EF tasks, including questionnaires regarding parenting practices, parental stress, parental depressive symptoms and ADHD symptoms. The tasks were administered in a fixed order and lasted about two hours, including breaks. After another break the child's behavior was observed using the Disruptive Behavior Diagnostic Observation Schedule (DB-DOS; Wakschlag et al., 2008a, 2008b). The DB-DOS is a structured observation that evaluates the child's behavior during tasks systematically varying in the level of challenge and support. During the first part of the DB-DOS the child performed different tasks (including frustration, internalization of rules, compliance, and social play tasks) with the parent and the second part with a clinical child psychologist. Finally, a semi-structured DSM-IV based parent interview for the assessment of ADHD, ODD and CD in preschool children was administrated: Kiddie Disruptive Behavior Schedule (KDBD; Keenan et al., 2007). The studies on the clinical usefulness of the DB-DOS and KDBD were part of another doctoral dissertation by Tessa Bunte (Bunte et al., 2013a; Bunte, Schoemaker, Hessen, Van der Heidjen, & Matthys, 2013b).

Children were diagnosed with ADHD, DBD (i.e., ODD or CD) or ADHD+DBD on the basis of the strict application of the DSM-IV-TR criteria for these disorders (American Psychiatric Association, 2000). Consensus was reached between a child psychiatrist and a clinical child psychologist using the scores on the CBCL, C-TRF, the symptoms reported on the parent interview (KDBD), observation of the child's behavior during the DB-DOS, and the scores on the Child Global Assessment Schedule (C-GAS; Schaffer et al, 1983, Dutch translation by Bunte, Schoemaker, & Matthys, 2007), a measure of the impairment of the child's functioning, filled out by the parents as well as the teacher/caregiver. Three weeks after the assessment, the results of the psychiatric and psychological assessment were discussed with the parents, and interventions targeting the child, the family or the school were suggested, when needed.

The children were re-assessed after 9 and 18 months. The second assessment was shorter. It consisted of the EF tasks, the KDBD, the questionnaires to evaluate the child's behavior (e.g., CBCL, C-TRF, C-GAS), and questionnaire regarding the caregiving environment (questionnaires similar to the first assessment with exception of parental ADHD and depressive symptoms). The DB-DOS was not administrated. Although a formal DSM-IV-TR diagnosis was not given, the development of the child could be evaluated on the basis of the substantial information available. Results of the second assessment were discussed with the parents by phone or at the outpatient clinic, depending on the parents' preference. The assessment after 18 months was similar to the first assessment, with the exclusion of the IQ tasks and some questionnaires (regarding maternal depression and parental ADHD symptoms). The same procedure as the first assessment was followed for diagnosing the children. Three weeks after the assessment, the results of the third assessment were discussed with the parents.

To be able to compare the EF performance of clinically diagnosed children and their caregiving environment to typical development in this age period we also included a group of TD children. The TD group was recruited from regular primary schools and daycare centers. Children were excluded with a score at or above the 90^{th} percentile on the Attention Problems scale or the Aggressive Behavior scale of the CBCL and C-TRF. The same procedure was followed with the same measurements, but without the initial interviews with a child psychiatrist or clinical child psychologist. Parents received a letter with a short report of the assessment.

Over a period of 2½ years 208 referred children were clinically assessed. After the assessment, 146 children (70.2%) were diagnosed with ADHD and/or DBD and included in the study. It must be noted that an additional 47 children who were initially not diagnosed with ADHD or DBD (but with an estimated IQ above 70) were re-assessed after 9 and 18 months. These children were not included in this dissertation, but have been included in other studies (Bunte et al., 2013a; 2013b). A full-participation rate of 97% was achieved: four children did not come back for subsequent assessments (three parents refused to further participate and one moved to another country). Out of 142 children, 139 children participated in all three assessments and three children participated in two of the three assessments. In addition, 60 TD children were initially assessed; two children were excluded due to clinical level of symptoms on the KDBD. Out of 58 children, 57 children participated in all three assessments and one child participated in two of the three assessments.

Some children received treatment after the first or second assessment. Psychological interventions consisted of individual parent counseling at the clinic (N=73), individual counseling at home (N=33) and the Incredible Years parent program (Webster-Stratton, 2001a; N =7). Psychopharmacotherapy consisted of methylphenidate (N=48) after the first assessment. After the second assessment, an additionally 23 children were prescribed medication. After the second assessment methyphenidate was prescribed in most cases (N=66), but atomoxetine (N=3) and risperidon (N=1) were prescribed as well.

In weekly meetings clinical decisions were made in a multi-disciplinary team including child psychiatrists, clinical child psychologists, case-workers, research assistants, master students in clinical child psychology, and a school counselor. After parents' consent school counselors from the special education school associated with the Department of Psychiatry, the Prof F. Redl school, talked through the results of the clinical assessment with teachers and school psychologists from the child's school in view of implications for education and behavioral management. After the first assessment, these school counselors remained available for initiatives to support the child's cognitive and social development at school.

AIMS AND OUTLINE

The aim of this dissertation was to study EF (chapter 2 & 3), the development of EF (chapter 4), and the role of the caregiving environment on the development of EF (chapter 5) in preschool children with ADHD and/or DBD.

In **chapter 2**, a meta-analysis of studies of the concurrent relation between externalizing behavior problems and EF is reported. Due to the high co-morbidity rate and low number of studies examining distinct behavior problems we examined preschool children with (symptoms of) ADHD and/or ODD. The three research questions addressed in this meta-analysis are: (1) To what extent do preschool children with externalizing behavior problems exhibit overall EF impairments? (2) To what extent are the three EF components (working memory, inhibition, cognitive flexibility) related to externalizing behavior problems in preschool children? (3) Are there factors that may moderate the relationship between EF and externalizing behavior problems? The potential moderators included were sample features (child age and gender) and sampling method (community vs. referred samples).

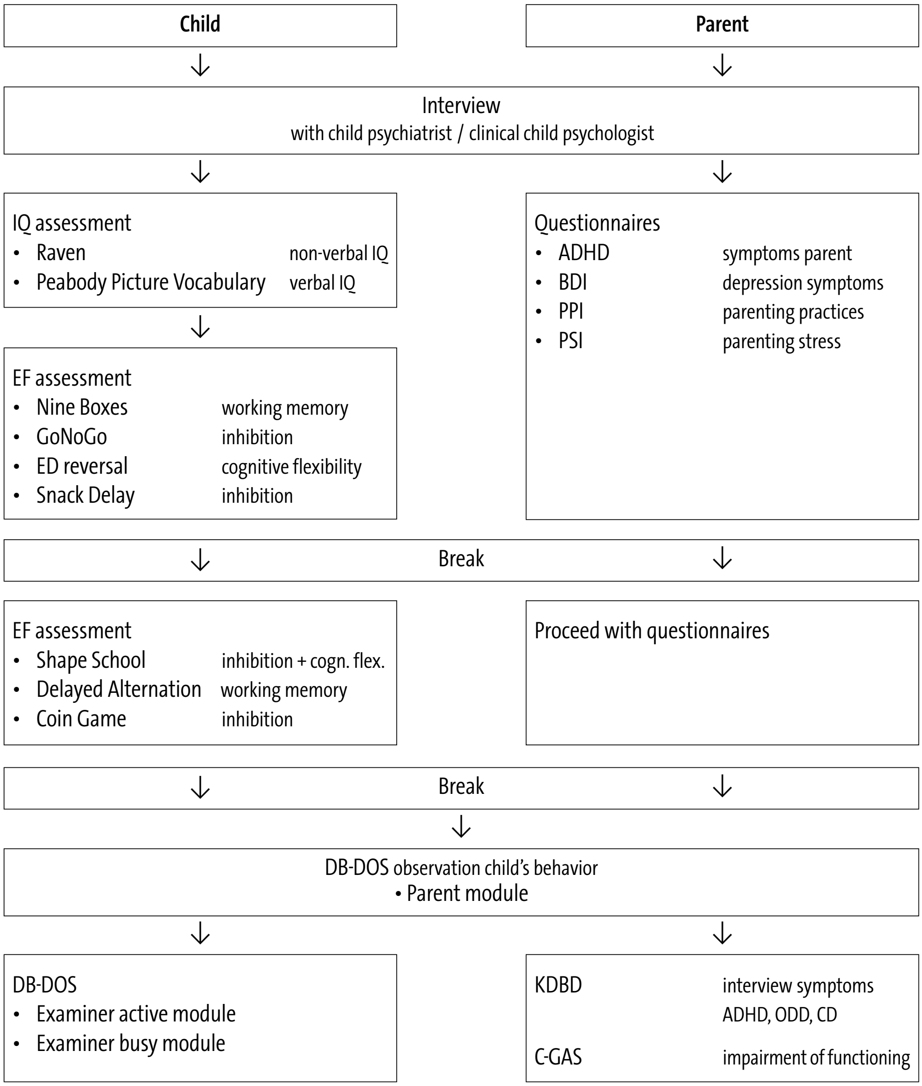
In **chapter 3** cross-sectional findings are presented. We examined the factor structure of EF, i.e., whether a one-factor or a two-factor (inhibition and working memory) model fitted the data. The preschool children with ADHD, DBD, ADHD+DBD and the TD children were compared on different EF tasks. We took into account co-morbidity between ADHD and DBD, by examining the co-morbid group, but also by accounting for the symptoms of the opposite disorder (e.g., accounting for DBD symptoms when examining ADHD impairments).

Chapter 4 describes the study of the longitudinal relation between EF and clinically diagnosed externalizing behavior problems. The first aim concerned the association between EF performance and a ADHD and/or DBD diagnosis across time. The second aim of the study was to examine the development of EF (inhibition as well as working memory) in clinically diagnosed preschool children compared to TD children.

Chapter 5 reports the study on the role of caregiving environment on the developmental change in EF in clinically diagnosed and TD preschool children. The caregiving environment was defined as parenting practices and maternal characteristics (maternal stress and maternal symptoms of depression and ADHD). We first examined if the caregiving environmental characteristics predicted change in the EF development over an 18 month period. Secondly, we tested if this effect differed for the four groups (ADHD, DBD, ADHD+DBD, TD). Third, we examined if the effect of maternal characteristics on the developmental change in EF performance was mediated by parenting practices.

Chapter 6 summarizes and discusses the findings of the studies. Furthermore, we discuss suggestions for further research and interventions.

Figure 1. Assessment Outpatient Clinic



C-GAS, Child Global Assessment Schedule; BDI, Beck Depression Inventory; DB-DOS, Disruptive Behavior Diagnostic Observation Schedule; KDBD, Kiddie Disruptive Behavior schedule; PPI, Parenting Practices Interview; PSI, Parenting Stress Index

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Executive Functions in Preschool Children with Externalizing Behavior Problems:

A Meta-Analysis

ABSTRACT

Deficits in executive functions (EF) have been found in school-age children and adolescents with externalizing behavior disorders. Present meta-analysis was carried out to determine whether these EF impairments can also be found in preschool children with externalizing behavior problems. Twenty-two studies were included with a total of 4021 children. Four separate meta-analyses were conducted, concerning overall EF, working memory, inhibition and cognitive flexibility. A medium correlation effect size was obtained for overall EF (ESzr =.22) and for inhibition (.24), whereas a small effect size was found for working memory (.17) and for cognitive flexibility (.13). Moderator analyses revealed a stronger effect for older preschoolers compared to younger preschoolers, and for children from referred samples compared to community samples. These results show that EF, especially inhibition, is related to externalizing behavior problems already in preschool years.

***Keywords*:** Executive Function, externalizing behavior problems, preschoolers, meta-analysis

INTRODUCTION

Although different definitions of executive functions (EF) exist, most authors agree that EF are the directing cognitive processes that enable purposeful and goal-directed behavior (Anderson, 2002; Oosterlaan, Logan, & Sergeant, 1998), i.e., the explicit control of thought, emotion and action (Séguin & Zelazo, 2005). EF include mental processes such as planning, working memory, inhibition of inappropriate responses, flexibility in adaptation to environmental changes, and decision making (Nigg, 2006). These functions serve to optimize behavior in changing environments. In their integrative framework, Miyake and colleagues (2000) proposed that in adulthood EF is a unitary construct with three partly dissociable components: working memory, inhibition and set shifting. Although the structure of EF that reflects both unity and diversity appears to be applicable from middle childhood onward (Wiebe et al., 2011), the structure during early childhood is not yet clearly defined.

Garon, Bryson, and Smith (2008) reviewed the development of EF in normally developing preschoolers, based on the integrative framework of Miyake et al. (2000). They proposed a model in which each EF component is built upon earlier developing functions in the first years of life. In their model, the first developing EF component is working memory (keeping information in mind over a delay and in some tasks manipulating that information), followed by inhibition (withholding or delay of prepotent response). Both skills are integrated in set shifting, also referred to as cognitive flexibility (learning a rule and subsequently shifting to a new rule).

Besides this theoretical model, factor analytic studies have been conducted to empirically test the structure of EF in young children. Earlier studies identified three or four separable EF components in younger children using exploratory factor analyses (Hughes, 1998; Espy, Kaufmann, McDiarmid & Glisky, 1999; Murray & Kochanska, 2002). More recently, however, studies used confirmatory factor analysis and found only one unitary executive construct in preschool children (Hughes, Ensor, Wilson, & Graham, 2010; Wiebe, Espy, & Charak, 2008; Wiebe et al., 2011; Willoughby, Blair, Wirth, & Greenberg, 2010). It must be noted that the latter three studies also examined a two-factor model (inhibition and working memory) which fitted the data equally well, but was rejected in favor of the one-factor model for reasons of parsimony. Thus, although the structure of EF in preschoolers still needs further investigation, recent studies indicate that EF may be characterized by a single factor structure in the preschool period and become increasingly differentiated with age (Hughes, 2011; Wiebe et al., 2011).

Over the last decade, there has been an increasing interest in EF of children with hyperactive, impulsive and aggressive behavior. Research on EF in children with externalizing behavior disorders, however, has focused mainly on school-age children and adolescents. First, regarding working memory, in meta-analyses concerning attention deficit hyperactivity disorder (ADHD) medium to large effect sizes were found, indicating that working memory function is impaired in school-age children and adolescents with ADHD (Martinussen, Hayden, Hogg-Johnson, & Tannock, 2005; Willcutt, Doyle, Nigg, Faraone, & Pennington, 2005). In contrast, there are inconsistent results regarding working memory performance in adolescents with the two disruptive behavior disorders (DBD), i.e., oppositional defiant disorder (ODD) and conduct disorder (CD). On the one hand, Séguin and colleagues (1995 and 1999) found a working memory impairment in physical aggressive children, also after controlling for ADHD. On the other hand, two more recent studies (Oosterlaan, Scheres, & Sergeant, 2005; Van Goozen et al., 2004) reported no differences between the ODD and normal control group.

Second, there is clear and convincing evidence of an inhibition deficit in school-age children and adolescents with ADHD with medium to large effect sizes, as has been repeatedly shown in meta-analyses (Alderson, Rapport, & Kofler, 2007; Pennington & Ozonoff, 1996; Willcutt et al.,2005). In a meta-analysis by Oosterlaan et al. (1998), however, deficits in inhibition were not uniquely associated with ADHD, but also with DBD. Other studies have shown deficiencies in inhibition with DBD, specifically when motivational processes, i.e., reward and punishment, are involved (Fairchild et al., 2009; Schutter, Van Bokhoven, Vanderschuren, Lochman, & Matthys, 2011).

Third, regarding cognitive flexibility, in meta-analyses concerning ADHD, medium effect sizes are reported, showing school-age children and adolescents with ADHD are at a disadvantage in this area (Pennington & Ozonoff, 1996; Willcutt et al., 2005). Small effect sizes were found in the relation between cognitive flexibility and antisocial behavior (Morgan & Lilienfeld, 2000). However, the possible role of ADHD co-morbidity was not examined in this study, although another meta-analysis revealed that EF deficits in CD are likely due to the presence of co-morbid ADHD (Pennington & Ozonoff, 1996). In sum, clear EF deficits have been documented in school-age children and adolescents with ADHD, but the results regarding DBD are less consistent.

Although this research provides valuable information on the role of EF in school-age children and adolescents with ADHD and DBD, it is important to examine whether deficits in EF may already be observed in preschool children with externalizing behavior problems, as chronic patterns of hyperactivity and behavior problems can already be identified in the preschool years (Shaw, Lacourse, & Nagin, 2005). Moreover, programs to train EF skills in young children have been developed and proved to increase EF performance (Diamond & Lee, 2011). Thus, if EF deficits occur already at preschool-age in children with externalizing behavior problems, these children may benefit particularly from such training. Since EF is related to later academic performance, improvement in EF may result in better academic performance (Diamond & Lee, 2011). Recently, the first meta-analysis was published in which neuropsychological deficits in preschoolers at risk for ADHD were examined (Pauli-Pott & Becker, 2011). A small effect size was found for working memory and a medium to large effect size for inhibition tasks in children with ADHD symptoms in comparison to typically developing preschoolers. A conclusion regarding cognitive flexibility was not drawn, as cognitive flexibility tasks had been included only in a limited number of studies.

The present meta-analysis extends this work by investigating externalizing behavior problems in preschool children from a broader perspective, i.e., in addition to young children with ADHD symptoms, we also included studies in young children with aggressive and DBD symptoms. Moreover, stricter inclusion criteria were applied (e.g., definition of preschool-age) and recent studies were included. Furthermore, in the current meta-analysis we included not only children with formal diagnoses, but also children with symptoms of ADHD and DBD. In many older studies, externalizing behavior problems in preschool children were viewed from a general perspective, e.g., 'hard to manage' children (Campbell, Pierce, March, Ewing, & Szumowski, 1994; Hughes, Dunn, & White, 1998) as clinicians were reluctant to diagnose young children with a psychiatric disorder. Only recently, instruments have become available to differentiate between normal development and psychopathology at preschool-age (e.g., Kiddie Disruptive Behavior Schedule, Keenan et al., 2007; Preschool Age Psychiatric Assessment, Egger & Angold, 2004). Although our initial aim was to examine the relation between EF performance and ADHD or DBD separately, there were unfortunately too few studies on DBD to examine this relation. However, as there is a correlation of .79 between symptoms of DBD and ADHD hyperactivity in preschoolers (Sterba, Egger, & Angold, 2007), it is presumably acceptable to examine ADHD and DBD combined.

The main aim of the current meta-analysis was to get specific insight in the relation between EF performance and externalizing behavior problems in preschool children. As there is an ongoing debate regarding the structure of EF in such young children and there are indications that EF may not be crystallized yet at this age, we studied EF at two levels; first overall EF, and second, the three EF components (working memory, inhibition, and cognitive flexibility) separately. Thus, the resulting two research questions are: (1) To what extent do preschool children with externalizing behavior problems exhibit overall EF impairments? (2) To what extent are the three EF components (working memory, inhibition, cognitive flexibility) related to externalizing behavior problems in preschool children? We hypothesize that possibly a smaller effect size for cognitive flexibility than for working memory and inhibition will be found, because cognitive flexibility develops later according to the developmental hierarchical model of Garon et al. (2008). Therefore, cognitive flexibility impairments might be less pronounced at this age.

The second aim of this meta-analysis was to examine whether there are factors that may moderate the relationship between EF and externalizing behavior problems. The strength of the association might depend in part on sample features (child age and gender) or sampling method. First, age may affect the relation between EF and behavior problems. Several studies confirm that EF performance in typically developing children improves rapidly with age during the preschool period (Carlson, 2005; Hughes et al., 2010; Wiebe et al., 2008), with a developmental spurt between three and five years (Garon et al., 2008). Thus, the relation between EF and externalizing behavior problems is expected to be less clear at the beginning of this developmental process. The third research question therefore is: Is there a difference in the strength of the relation for older preschool children (4½ to 6 years) compared to younger (3 to 4½ years) preschool children?

Second, the strength of the relationship between EF and externalizing behavior problems may be influenced by the severity of the behavior problems. In community samples behavior problems are less distinct compared to referred samples. Assuming that the contribution of neurobiological factors is larger compared to environmental factors when the behavior problems are more severe, the strength of the association between EF and externalizing behavior problems will be stronger in referred samples in contrast to community samples. The results of a meta-analysis in older children with ADHD (Willcutt et al., 2005) showed that effect sizes on EF measures were slightly smaller in community samples compared to clinical samples and concluded that weaknesses in EF were not restricted to clinical samples but were also present in the general population. Thus, the fourth research question is: Is the magnitude of the effect sizes found for children from referred samples different from the magnitude of the effect sizes for children from community samples?

Third, the relation between EF and behavior problems may be affected by the child's gender. Girls could differ from boys in their EF performance due to their more rapid developmental maturation (Keenan & Shaw, 1997). However, little is known about possible gender differences in the relation between externalizing behavior problems and EF in the preschool-age. Raaijmakers et al. (2008) found that there was a stronger relation between aggression and EF in boys compared to girls, whereas Thorell and Wåhlstedt (2006) found no gender difference in EF performance in children with ODD/ADHD symptoms. The fifth research question is: Does gender play a role in the relationship between externalizing behavior problems and EF?

METHOD

Retrieval of studies

A systematic computer search was performed in *Pubmed, Web of Science and PsychInfo*. The following combinations of keywords were used: Executive function (i.e., EF, working memory, inhibition, cognitive flexibility, neuropsychology), behavior problems (i.e., attention problems, attention deficit, hyperactivity, ADHD, oppositional, aggressive, externalizing, ODD, conduct problems, CD), and preschool (i.e., early childhood, young children). The reference lists of the retrieved articles were examined. Studies published before August 2011 were included. We limited our search to publications in English and in peer-reviewed journals. It should be noted that unpublished studies were excluded from this meta-analysis. Although limiting a meta-analysis to published documents introduces the potential for bias in favor of significant results, there is also a possibility of introducing a bias when searching for unpublished papers. It is not possible to systematically search for unpublished papers in a way that it is replicable. Moreover, there is no method available to calculate the magnitude of this bias (see also Kaminski, Valle, Filene, & Boyle, 2008). For this reason, we chose the standard and accepted method of computing the fail-safe number, rather than including unpublished studies with the risk of introducing an unknown source of bias.

Inclusion criteria

To be included studies had to meet the following criteria: (a) the study included children with externalizing behavior problems (clinical diagnosis / symptoms of ADHD, symptoms of ODD / CD, or aggressive or hard to manage children), (b) the mean age of the children in the study was between 3.0 and 6.0 years, and (c) EF tasks, aimed to measure working memory, inhibition or cognitive flexibility were administered. Studies were excluded which examined children with pediatric or neurological diseases.

In cases the article did not report sufficient information to permit calculation of the effect size, an attempt was made to contact the corresponding author for additional information. Data were obtained for four additional studies (Berwid et al.,2005; Raaijmakers et al.,2008; Tillman, Thorell, Brocki, & Bohlin, 2008; Willoughby et al., 2010). In case of longitudinal studies, the first assessment was chosen. This resulted in choosing Campbell et al. (1994), Berlin and Bohlin (2002), Dalen, Sonuga-Barke, Hall and Remmington (2004), and Thorell and Wåhlstedt (2006). Thus we excluded respectively Marakovitz and Campbell (1998), Berlin, Bohlin and Rydell (2003), Sonuga-Barke, Dalen, Daley, and Remington (2002) and Sonuga-Barke, Dalen and Remmington (2003), and Wåhlstedt, Thorell, and Bohlin (2008). If within an article two assessments were reported on the same sample, the first assessment was included (Antshel & Nastasi, 2008; Campbell et al., 1994). After correspondence with the authors it appeared that there was considerable overlap in sample between the studies of Berwid et al. (2005) and Marks et al. (2005). We included the study of Berwid, because the reported EF tasks resembled the other tasks included in this meta-analysis more.
The final sample included 22 studies (*Table 1*). Nineteen studies included one or more inhibition tasks, 13 studies included working memory tasks, and five studies included cognitive flexibility tasks. The included studies are marked with an * in the reference list.

Coding the Studies

Each study was coded with a detailed coding scheme for recording sample, study and methodological characteristics. All these study characteristics were coded by the first author. In case of any doubt, the first author of the article was contacted.

EF Tasks

The EF tasks were categorized, based on the definition and categorization of Garon et al. (2008), as working memory, inhibition or cognitive flexibility tasks. The classification of the EF tasks was coded by the first and second author independently. Inter-rater agreement was 99%. Although some EF tasks give multiple scores, it was often not possible to choose between different dependent variables from the same task, as most studies only reported a single dependent variable per task. This reflects the general heterogeneity in the assessment of EF which is characteristic of the field. Only in a few cases did we have the opportunity to choose between dependent variables. In those cases, the dependent variable was chosen which was most frequently used in the other included studies reporting on the same task (e.g., commission errors GoNoGo tasks). A table with the dependent variables of the EF tasks is displayed in the appendix on page 52.

Working memory tasks are tasks in which children have to keep information in mind over a delay and, in some tasks, have to update or manipulate that information. The following tasks were included: Digit/word span (n=5), Spatial memory (n=3), Word span backwards (n=2), Delayed alternation (n=2), Six/nine boxes (n=2), Selective reminding task (n=1), Sentence repetition (n=1), Span like task (n=1), Noisy book (n=1), Narrative memory (n=1), Delayed response (n=1), Dual request selective task (n=1), Multiple boxes test (n=1), and Picture learning (n=1).

Inhibition tasks require withholding or delay of a prepotent or automatic response or holding a rule in mind, responding according to this rule, and inhibiting a prepotent response. The following tasks were found in the literature: Stroop tasks (n =8), GoNoGo (n=7), NEPSY statue (n=5), Delay of gratification (n =3), Snack delay (n =2), Shape School-inhibit condition (n=3), Knock&tap (n=2), Stop-Signal task (n=1), Resistance to temptation (n=1), Delay aversion (n=1), Detour reaching box (n=1), Spatial conflict (n=1), Puppet says (n=1), Luria's handgame (n=1), Pencil tapping (n=1), and Tongue task (n=1).

Cognitive flexibility tasks require forming an arbitrary stimulus-response set in the first phase and shifting to a new stimulus-response set in the second phase, with attention to a new aspect of the same stimulus. The tasks included were Block sorting, Colour form test, Item selection, Object Classification Task, Set shifting, and Shape School-switch condition (for all tasks, n=1).

Externalizing behavior problems

Externalizing behavior problems were defined differently across studies, with some studies using a categorical approach, whereas others used a dimensional approach. In the categorical approach, used in 10 studies, a child was classified in the externalizing behavior problem group or the control group, according to either a cut-off score on a questionnaire or a judgment on the basis of a combination of different instruments. In these studies, the externalizing behavior problem group was defined as (high risk for) ADHD, 'hard to manage', aggressive, ODD or DBD. In the dimensional approach, used in 12 studies, symptoms or items on a continuous scale were used as outcome measures for behavior problems, including symptoms of ADHD, hyperactivity, attention problems, ODD, CD and aggression.

To assess externalizing behavior problems, a variety of instruments, informants and sampling methods were used across studies. Many studies used a combination of measurements and informants to define externalizing behavior problems. Sixteen different instruments were used to assess externalizing behavior problems, including eleven questionnaires and five semi-structured interviews. Many studies (k =10) used multiple informants (parents and a teacher/health visitor/research assistant). Eight of those studies reported on behavior problem groups, which were distinguished on the basis of reports from all informants and in the other two studies correlations were reported separately for different informants. In seven studies questionnaires were only filled out by parents and in vife studies questionnaires filled out by teachers were included. A table with information regarding the specific instruments, subscales, informants and how multiple measures were integrated is available upon request from the first author.

Differences in sampling method across studies were also noted. The majority of studies used a community sample (k =15), four studies reported on a clinically referred sample, and three studies used a procedure where they 'over selected' children with externalizing behavior.

Table 1. Studies Included in the Meta-Analysis, with Descriptors and ESzr for Each Task

Study	N	Externalizing behavior problems	Mean Age +range *years; months*	Gender *% boys*	Sampling method	Assess-ment	EF	EF task	ESzr task
Anthsel 2008	31 31	ADHD Control	4;9 4;9 R: all 4 years	60 59	Referred	Quest: P Interv: P	WM	Picture learning	.07
Berlin 2002	151	Hyperactive & CD	5;2	48	Community	Quest: T	Inh	GoNoGo	.38
Berwid 2005	16 42	High Risk ADHD Low Risk ADHD	4;8 4;8 R: 3;5–6;11	64 63	Community	Quest: P&T	Inh Inh	GoNoGo Day-Night stroop	.21 .16
Brocki 2007	72	ADHD and/or ODD	5;5 R: 4;1–7;0	83	⅓ Selected by psychologist	Quest: P&T	WM WM WM Inh Inh Inh	Digit span-forward Digit span-backward Spatial memory NEPSY statue Knock/tap/DNST GoNoGo	.13 .06 .07 .47 .42 .37
Campbell 1994	69 43	Hard2Manage Control	3;10 3;11 R: 2;6–4;6	100 100	¼ Selected by parents (several with diagnosis)	Quest: P&T	Inh Inh	Resistance to temptation Delay of gratification	.36 .19
Dalen 2004	19 19	ADHD Control	3;3 3;3 R: all 3 years	-	First selection by health visitor	Interv: P	Inh Inh Inh CF	Delay aversion Delay of gratification Puppet says Block sorting	.40 .92 .42 .32
Dennis 2003	37	Aggression	6;0 R: 4;5–7;6	51	Community (prevention trial)	Quest: P	Inh	Four inhibition tasks of effortful control battery	.39
Espy 2011	135 –194	ADHD and ODD symptoms	3;11 R: 2;5–6;0	44	Community	Quest: P	WM WM WM WM Inh Inh	Digit span Delayed alternation Delayed response Six boxes NEPSY statue Shape school- inhibit	.09 .11 .20 .03 .13 .12

Table 1. continued

Study	N	Externalizing behavior problems	Mean Age +range *years; months*	Gender *% boys*	Sampling method	Assess-ment	EF	EF task	ESzr task
Hughes 1998	40 40	Hard2Manage Control	4;4 4;2 R: 3;6–4;6	60 60	Community	Quest: P&T	WM Inh Inh CF	Noisy book Detour reaching box Luria's handgame Card sorting	.19 .30 .23 .17
Mahone 2005	40 40	ADHD Control	4;10 4;11 R: 3;0–6;6	85	Referred	Quest: P Interv: P	WM Inh	Multiple boxes task NEPSY statue	.22 .36
Mariani 1997	24 30	ADHD Control	5;0 5;1 R: 4;0–5;11	100 100	Referred	Quest: P or T Interv: P	WM WM WM CF	Digit span Selective reminding Spatial memory Color form test	.35 .42 .43 .23
Perner 2002	24 22	At risk ADHD Control	5;9 5;7 R: 4;9–6;6	-	Community	Quest: T Interv: P	Inh Inh Inh WM	NEPSY statue Knock & Tap GoNoGo Digit span -backward	.36 .21 .31 .12
Raaijmakers 2008	82 99	Aggressive behavior Control	4;3 4;4 R: all 4 years	72 65	Community	Quest : P	WM Inh Inh Inh CF CF	Digit span GoNoGo Day-Night stroop Shape School- inhibit Shape School- switch Object Classification task	.16 .23 .18 .24 .22 .04
Re 2010	23 23	ADHD Control	5;7 5;9	61 61	Community	Quest: P&T	WM	Dual Request Selective Task	.59
Rezazadeh 2011	31	Hyperactivity	4;9 R: 3–7	100	Community	Quest: P	Inh	Day-Night stroop	.07
Schoemaker 2012	61 33 52 56	ADHD DBD ADHD+DBD Control	4;7 4;4 4;6 4;8 R: 3;6–5;6	80 82 83 70	Referred	Quest: P&T Interv: P	WM WM Inh Inh Inh	Delayed Alternation Nine Boxes GoNoGo Snack Delay Shape School-inhibit	.20 .20 .37 .47 .41

Table 1. continued

Study	N	Externalizing behavior problems	Mean Age +range *years; months*	Gender *% boys*	Sampling method	Assess-ment	EF	EF task	ESzr task
Thorell 2006	17	ADHD	5;0	77	Community	Quest: T	WM	Digit span	.17
	13	ODD	R: 4;0–6;2	46			WM	Spatial memory	.17
	27	ADHD+ODD		70			Inh	Day-Night stroop	.30
	81	Control		40					
Tillman 2008	107	ADHD hyp/imp	5;1	63	Community	Quest: T	Inh	Stop signal task	.26
			R: 4;0–6;1				Inh	Stroop	.37
Von Stauf-fenberg 2007	776	Attention problems	4;6	49	Community	Quest: T	Inh	Delay of gratification	.16
							Inh	Day-Night stroop	.08
Will-oughby 2010	440 –873	ADHD	3;1	51	Community	Quest: P, T, RA	WM	Span task	.13
							Inh	Silly Sound stroop	.06
							Inh	GoNoGo	.05
							Inh	Spatial conflict	.12
							CF	Item selection	.12
Will-oughby 2011	738 –757	ADHD, ODD, aggression symptoms	4;6	50	Community (Head Start)	Quest: T	Inh	Pencil tapping	.12
			R: all 4 years				Inh	Snack delay	.13
							Inh	Tongue Task	.07
Young-wirth 2007	28	Hyperactive	4;9	61	Community	Quest: P	WM	Sentence repetition	.20
	14	ODD	R: 4;0–5;7	43			WM	Narrative memory	.21
	29	Hyp+ODD		72			Inh	NEPSY statue	.19
	123	Control		55					

ADHD	*Attention Deficit Hyperactivity Disorder*	*P*	*Parent report*
CF	*Cognitive flexibility task*	*Quest*	*Questionnaire*
DBD	*Disruptive Behavior Disorder*	*R*	*Age range*
EF	*Executive Function*	*RA*	*Research assistant*
Interv	*Interview*	*Selected*	*Selected from a community sample*
Inh	*Inhibition task*	*T*	*Teacher report*
ODD	*Oppositional Defiant Disorder*	*WM*	*Working memory task*

Differences in sampling method across studies were also noted. The majority of studies used a community sample (k=15), four studies reported on a clinically referred sample, and three studies used a procedure where they 'over selected' children with externalizing behavior problems in a community sample. This selection was done by identification of a child psychologist (Brocki, Nyberg, Thorell, & Bohlin, 2007), an observation of a health visitor (Dalen et al.,2004), or parent-identified (with several children in treatment for ADHD/ODD; Campbell et al.,1994). The definition of the externalizing behavior problems, informants and sampling method of each study is included in *Table 1*.

Moderators

Four moderators were included: age, sampling method, gender distribution, and impact factor of the journal. For age, we chose to split the mean age of the studies in the middle of the preschool period, resulting in a younger (3 years-0 months to 4 years-6 months) and older (4 years-7 months to 6 years-0 months) group. The sampling method was divided in referred/selected samples and community samples. Gender was coded as percentage of boys in the sample and analyzed as a continuous variable. The impact factor of a journal is a frequently used indicator of study quality in meta-analyses (e.g., Prinzie, Stams, Deković, Reijntjes, & Belsky, 2009). The impact factor of the journal (2010) was analyzed as a continuous moderator. Few studies reported on IQ or ethnicity, and therefore, the potential moderator effects of these variables could not be tested adequately with the present set of studies.

Data Analysis

Initially, a single effect size for each EF task within a study was calculated (see *Table 1*). Subsequently, when studies reported results on different types of externalizing behavior, we averaged effect sizes across behavior problems. For example, when results on EF tasks were reported separately for the ODD and ADHD group within one study, we calculated the effect size of each group and subsequently averaged the effect sizes. There were two levels of analyses, resulting in four separate meta-analyses. First, a meta-analysis of the overall EF was conducted. In this analysis, the weighted mean effect size across all EF tasks in a study was computed, because within some studies there was considerable variation in the number of children completing each task. Then, the weighted mean effect size across studies was computed as described below. Second, meta-analyses were conducted for the separate EF components, i.e., working memory, inhibition and cognitive flexibility. For these analyses, first, a weighted mean effect size for all tasks of the same component was computed within a study to be entered in the meta-analysis.

For each meta-analysis, a mean standardized effect size (random effects model) and the 95% confidence interval (CI) were computed. In 12 studies externalizing behavior problems were studied on a continuum, for these studies a correlation between the EF performance and externalizing behavior problems was reported. The other ten studies compared different groups, and subsequently reported means and standard deviations for each group or a Chi-square value (Hughes et al.,1998). We chose to convert all data to an effect size correlation (ESzr), as group comparisons are based on a dichotomy of an underlying continuous distribution of behavioral problems which is the same as in studies reporting correlations. All data were converted to ESzr with the transformations of Lipsey and Wilson (2001).

The fail- safe number (FSN) was calculated to indicate the robustness of findings. The FSN is the minimum number of studies with null results that are needed to reduce significant results to non-significance. If the FSN exceeds the critical value, i.e., five times the number of studies plus 10, findings are considered robust (recommended by Rosenthal,1995). The FSN also indicates sensitivity of the findings to publication bias (i.e., the tendency that non-significant findings often remain unpublished).

Homogeneity of the effect size distribution was examined with the Q-statistic. A significant Q-test indicates heterogeneity, which assumes that differences across effect sizes are due to sources other than sampling error, such as different study characteristics (Lipsey & Wilson, 2001). Moderator analyses were then conducted to try to explain heterogeneity across effect sizes. If the meta-analyses of one of the EF components (working memory, inhibition, cognitive flexibility) were heterogeneous, additional task specific meta-analyses were conducted.

Moderator analyses for the continuous moderator (i.e., percentage boys and impact factor) were analyzed with weighted regression analyses and categorical moderators (i.e., age and sampling method) were analyzed with an ANOVA-procedure (both macro's by Lipsey & Wilson,2001). An ANOVA yields two homogeneity estimates, the $Q_{between}$ and Q_{within}. A significant value for $Q_{between}$ indicates that the effects sizes are significantly different across different categories of the moderator variable, whereas a significant value for Q_{within} indicates that the effect sizes within a category of the moderator variable are heterogeneous (Lipsey & Wilson, 2001). The moderator variables were not related to each other, with the exception of gender and sample type. The clinical samples included studies with a higher percentage of boys ($F = 15.45$, $p = .001$).

Statistical meta-analyses and moderator analyses were performed using SPSS 16.0. Outliers were not excluded, considering the low number of studies. Correlation coefficient effect sizes can be classified as small (.10), medium (.25) or large (.40; Lipsey & Wilson, 2001).

RESULTS

Sample and study characteristics

The 22 studies included in the meta-analyses provided data on 4021 children, with sample sizes ranging from 31 to 873 (*Table 1*). The range of EF tasks included per study was one to six. The mean age of children across studies was 57 months (range 37 to 72 months). All studies reported on gender composition, with the exception of two studies. The percentage of boys ranged from 44% to 100%. Eight studies reported on the ethnicity of the children. The percentage of Caucasian ranged from 0% (Dennis & Brotman, 2003) to 100% (Campbell et al., 1994; Mariani & Barkley, 1997). Twelve studies reported a general IQ score, with the mean IQ across these studies ranging from 94 to 114.

All studies were published in peer reviewed journals between 1992 and August 2011. Twelve studies were conducted in the USA, four in Sweden, two in the United Kingdom, The Netherlands and Canada, and one in Italy and Austria. In all studies, EF performance and behavior problems were assessed concurrently, except for the study of Berlin and Bohlin (2002) in which the behavior problems were measured nine months after the EF assessment.

Effect sizes for overall EF

There was a significant relationship between the level of overall EF performance and externalizing behavior problems with a medium effect size of .22 ($p < .001$; see *Table 2*). This analysis included all 22 studies and 69 EF assessments, and the effect sizes ranged from .07

to .59. *Figure 1* shows the effect sizes and 95% CI for each study of the overall EF. The FSN was 547, which far exceeds Rosenthal's (1995) critical value (i.e., 120). The results can thus be considered robust against the file drawer effect.

The Q-statistic testing effect size heterogeneity was significant, $Q(21) = 41.53$, $p = .005$, indicating the effect sizes may be drawn from different populations. The results of the categorical moderator analyses are presented in *Table 3*. Moderator analyses revealed a significantly larger effect size for studies with older preschoolers (4½ to 6 years) relative to studies with younger preschoolers (3 to 4½ years). Studies examining a referred/selected sample reported significantly larger effect sizes compared to studies from community samples. Regression analyses showed a significant effect of gender distribution (ß= .44, p = .029), indicating that studies with a higher percentage of boys showed a higher effect size. There was no effect of the impact factor (ß = .15, p = .516).

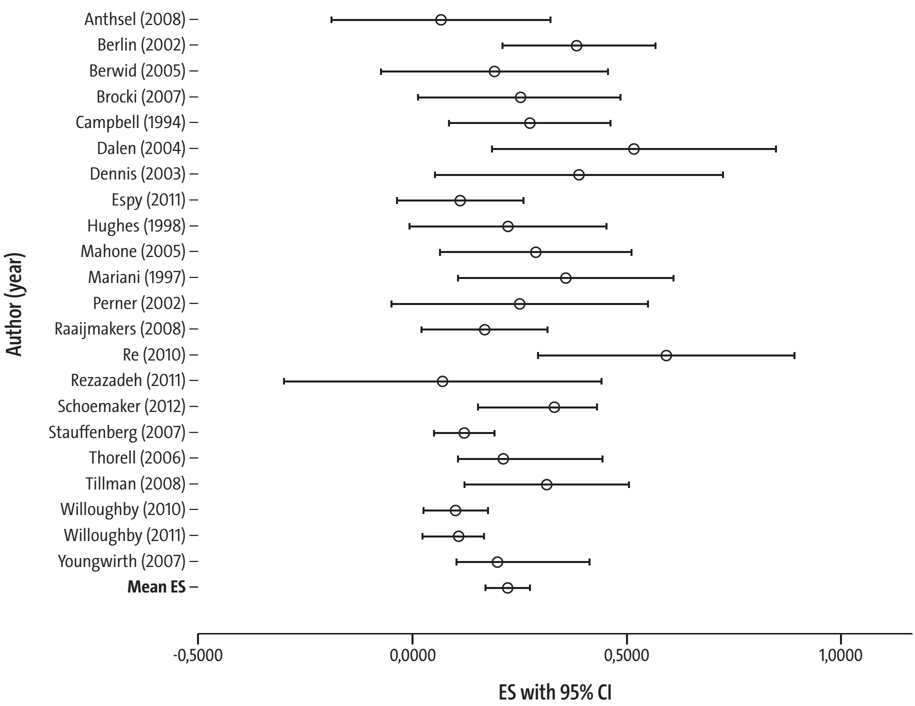
Table 2. Effect Sizes for Overall EF and for Separate EF Components

	k	N	ESzr	95 % CI	Q - statistic
Overall EF					
Overall EF	22	4.021	.22***	.17 – .27	41.53**
EF factors					
Working Memory	13	2.132	.17***	.12 – .23	15.06
Inhibition	19	3.795	.24***	.18 – .30	47.97***
Cognitive Flexibility	5	1.198	.13***	.08 – .19	2.24
Inhibition tasks					
GoNoGo	8	1.238	.26***	.15 – .38	22.58**
Stroop	7	1.785	.16***	.08 – .25	14.55*
Delay with motivation	5	1.875	.32***	.16 – .48	37.16***
Delay without motivation	5	614	.27***	.14 – .40	8.62

** p < .05, ** p < .01, *** p < .001*

CI = confidence interval, ESzr = mean correlation effect size, k = number of studies, N = number of participants

Figure 1. Forest Plot of Overall EF Tasks (k=22), Mean Effect Size with 95% CI



Effect sizes for EF components

Working memory

The meta-analysis of the working memory tasks included 13 studies and 23 assessments. A significant small effect size of .17 ($p < .001$) was found. Effect sizes ranged from .07 to .59 (*Figure 2* in the appendix on page 50). The FSN of 183 exceeded Rosenthal's criteria of 75. The Q-statistic was non-significant, $Q(12) = 15.06$, $p = .238$, indicating homogeneity.

Inhibition

In the meta-analysis of inhibition, 19 studies and 39 assessments were included. Results indicated a medium significant effect size of .24 ($p < .001$). Effect sizes ranged from .07 to .58

(see *Figure 3* in the appendix on page 51). Results were robust against the file drawer problem, as the FSN of 413 exceeded by far the Rosenthal's (1995) criteria of 105.
The distribution of effect sizes was heterogeneous, $Q(18) = 47.97$, $p < .001$, indicating the effect sizes may be drawn from different populations. Moderator analyses showed a significant effect of age and sample, with larger effect sizes found in older preschool children and children from referred/selected samples. Regression analyses showed a significant effect of gender distribution (ß = .48, p = .026), indicating that studies including a higher percentage of boys demonstrated a higher effect size. There was no effect of the impact factor (ß = .32, p = .194).

Table 3. Categorical Moderator Analysis

	$Q_{between}$	k	ESzr	95% CI	Q_{within}
Overall EF (k = 22)					
Age	20.90***				
Younger preschoolers (3.0 – 4.5)		7	.12***	.08 - .16	7.03
Older preschoolers (4.6 – 6.0)		15	.28***	.22 - .33	13.06
Sample	4.10*				
Community		15	.18***	.13 - .23	17.10
Referred/selected		7	.29***	.20 - .38	4.60
Inhibition (k = 19)					
Age	19.18***				
Younger preschoolers (3.0 – 4.5)		7	.14***	.09 - .19	9.92
Older preschoolers (4.6 - 6.0)		12	.31***	.25 - .37	8.13
Sample	13.31***				
Community		14	.18***	.13 - .23	14.08
Referred / selected		5	.39***	.29 - .49	2.55

** p < .05, ** p < .01, *** p < .001*

CI = confidence interval, ESzr = mean correlation effect size, k = number of studies, N = number of participants

Cognitive flexibility

The meta-analysis on cognitive flexibility included five studies and six assessments. Results indicated a small but significant effect size of .13 ($p<.001$). Effect sizes ranged from .12 to .32 (*Figure 4* in the appendix on page 51). There is possibly a file drawer problem while the FSN was 34 and Rosenthal's criterion was 35, so these findings should be interpreted with precaution. The Q-statistic was non-significant $Q(4)=2.24$, $p=.692$, indicating homogeneity.

Additional analyses: Effect sizes for inhibition tasks

An additional factor influencing heterogeneity in the inhibition component might be the wide variety of tasks administrated. Therefore, additional meta-analyses were conducted on task level. Four categories of common inhibition tasks were created, i.e., GoNoGo tasks, Stroop tasks, Delay tasks with motivation and Delay tasks without motivation.

GoNoGo tasks (Response inhibition)

In GoNoGo tasks children have to press a key when they see a target stimulus (Go condition) and inhibit that response when they see a non-target stimulus (NoGo condition). The meta-analysis for the GoNoGo tasks included eight tasks, including the computerized GoNoGo tasks (n=7) and a modified Stop Signal Task (Tillman et al., 2008). In case of the GNG/CPT task (Berwid et al., 2005), we requested the data from the 5 (Go) vs. 1 (NoGo) ratio and included it here as a GoNoGo task. Results indicated a medium mean effect size .26 ($p<.001$). The FSN was 55, which exceeds Rosenthal's critical value of 50. The results may thus be considered robust against publication bias. The Q-test indicated heterogeneity across samples, $Q(7)=22.58$, $p=.002$.

Stroop tasks (Interference suppression)

In the Stroop tasks children have to suppress an automatic response and activate a conflicting response. For example, in the day/night Stroop task, children must respond 'night' to a picture of the sun and 'day' to a picture of the moon. The Stroop analysis included seven tasks, including the day/night, boy/girl and silly sound Stroop tasks. The mean effect size was small but significant, .16 ($p<.001$). The FSN, 28, was under the critical value of Rosenthal, 45, indicating a possible file drawer problem. The Q-test indicated heterogeneity, $Q(6)=14.55$, $p=.024$.

Delay tasks (response delay with motivation)

In the delay tasks with tangible reward children had to stand (still) and wait for a signal before they could have a treat. The included tasks were Delay of gratification (n=3) and Snack delay (n=2). There was a medium significant effect size .32 ($p<.001$). The FSN of 22 was under the critical value of Rosenthal, 35, indicating a possible file drawer problem. The Q-statistic was significant, $Q(4) = 37.16$, $p<.001$.

Delay tasks (response delay without motivation)

In the delay tasks without tangible reward children had to stand (still) and wait for a signal before they were allowed to move again. The analysis included the five NEPSY statue tasks. There was a medium significant effect size .27 ($p<.001$). The FSN of 27 was just under the critical value of Rosenthal, 35, indicating a possible file drawer problem. The Q-test indicated homogeneity, $Q(4) = 8.62$, $p = .071$.

Additional Analyses: Separate analyses for ADHD and DBD symptoms

In order to investigate possible differences between ADHD and DBD symptoms, we conducted additional analyses, separately for studies that examined DBD symptoms and studies that examined ADHD symptoms. Studies were excluded if it was not possible to differentiate between ADHD and DBD (i.e., these studies included general categories of problem behavior, such as 'hard to manage'). There were too few studies to conduct moderator and task specific analyses separately. There were nine studies which examined DBD symptoms: two studies examining aggressive symptoms, three studies examined a DBD-only group (next to three other groups) and four studies reported a correlation with DBD symptoms (next to ADHD symptoms of the same group). Concerning the studies examining DBD symptoms, for overall EF we found an effect size of .19 ($p<.001$, 95% CI: .11-.26, k=9), for working memory .15 ($p<.001$, 95% CI: .07-.22, k=6), for inhibition .22 ($p<.001$, 95% CI: .13-.31, k=9), and for cognitive flexibility .13 (one study). There were 18 studies which examined an ADHD-only group or ADHD symptoms. Analyses for ADHD symptoms show for overall EF an effect size of .21 ($p<.001$, 95% CI: .16-.21, k=18), for working memory .17 ($p<.001$, 95% CI: .10-.24, k=11), for inhibition .24 ($p<.001$, 95% CI: .17-.31, k=13), and for cognitive flexibility .14 ($p<.001$, 95% CI: .06-.22, k=3). In sum, the effect sizes for DBD were very similar to the effect sizes for ADHD (range of differences .01 to .02), providing further support for the decision to conduct the analyses for ADHD and DBD symptoms together. However, this result needs to be interpreted with caution due to the low number of studies.

DISCUSSION

The purpose of the current meta-analysis was to determine the strength of the relationship between EF and externalizing behavior problems in preschool children. The unique contribution of this meta-analysis is the examination of the frequently co-occurring ADHD and DBD symptoms in preschoolers and the study of EF performance at different levels (i.e., overall, component, and task level) based on the latest insights in the structure of EF in preschoolers. The results indicate a medium mean effect size for the relationship between overall EF and externalizing behavior problems. However, results differed for the different EF components. A small effect size was found for working memory, a medium mean effect size was found for inhibition, and a small effect size for cognitive flexibility. The moderator analyses showed a stronger relation between EF and externalizing behavior problems for older preschool children (4½ - 6 year) compared to younger preschool children (3 - 4½ year) for the overall EF and inhibition component. There was a stronger relationship between EF and externalizing behavior problems for the children from referred/selected samples compared to community samples for the overall EF and inhibition component. Furthermore, there was a stronger relationship between EF and externalizing behavior problems in studies with a higher percentage of boys.

A medium effect size was found for **overall EF** (i.e., across all EF components and tasks). We chose to conduct a meta-analysis on overall EF given the findings of recent confirmatory factor analytic studies that EF may be best described as a single latent construct in very young children (Hughes et al., 2010; Wiebe, et al., 2008; Wiebe et al., 2011; Willoughby et al., 2010). However, the results of the present meta-analysis suggest that in preschool children with externalizing behavior problems there may not be just one EF factor, as there were differences in effect sizes across the different factors, with stronger impairments found for inhibition as compared to working memory and cognitive flexibility. If indeed there was only one overall EF factor, we would have expected the effect sizes of the different domains to be much more similar. Our finding is in line with the two-factor model of EF (inhibition and working memory) found in a recent study of preschoolers with ADHD and/or DBD (Schoemaker et al., 2012) and a three factor model in a sample with aggressive preschoolers (Raaijmakers et al., 2008). It might be that the EF factor structure is different in preschool children with externalizing behavior problems compared to typically developing children. However, the multi factor model is not necessarily at variance with the results of confirmatory factor analytic studies of typically developing preschool children (Wiebe et al., 2008; Wiebe et al., 2011; Willoughby et al., 2010), which reported that the two-factor model fitted equally well as the one-factor model. In fact, Wiebe et al. (2011) proposed that a multiple factor EF model might potentially be more ecologically valid than a single factor solution in preschoolers. The findings from the current meta-analysis seem to support this notion.

For **working memory**, a smaller effect size was found in the current study compared to meta-analyses of older children. Medium effect sizes were found for older children with ADHD, e.g., for spatial working memory and verbal working memory (d=.63 and .55, respectively; Willcutt et al., 2005), compared to a small effect size in our study. The medium effect size concerning the **inhibition** factor and separate inhibition tasks found in this meta-analysis is consistent with the results of meta-analyses of older children. For example, medium effect sizes were found for children with ADHD for the Stop signal task (Cohen's d=.61; Willcutt et al., 2005) and for children with antisocial behavior on the Stroop task (d=.43; Morgan & Lilienfeld, 2000). A similar pattern as working memory is observed for **cognitive flexibility**; we found a small effect size in contrast to meta-analyses of older children, which reported larger effect sizes. For example, on the Wisconsin Card Sorting Test a medium effect size was found for children with ADHD (d=.46; Willcutt et al., 2005) and a small to medium effect size for children with antisocial behavior (d=.24; Morgan & Lilienfeld, 2000). The cognitive flexibility component showed the lowest effect size compared to the other EF components. There was even a non-overlapping 95% confidence interval with inhibition. A possible explanation for this result may be that in the hierarchical model of Garon et al. (2008) cognitive flexibility is the last emerging EF component. As a result, it could be that at such young age children with cognitive flexibility impairments cannot be distinguished yet from typically developing children (see also Pauli-Pott & Becker, 2011).

Age, sampling method and gender distribution were studied as moderators in the relationship between EF and externalizing behavior problems. The results of the present meta-analysis show that there is a stronger relation between overall EF performance (and specifically inhibition performance) and externalizing behavior problems in older compared to younger preschool children. The age effect in the present meta-analysis may be due to several reasons. First, there are concerns regarding validity and measurement error of EF tasks, particularly for the younger preschoolers, even though in recent years a shift has been observed away from simplifying adult tests and toward more developmentally appropriate tasks for assessing EF in young children (Carlson, 2005; Espy et al., 1999; Hughes, 1998).

These child-friendly tasks are designed to minimize the complexity of instructions and responses. Despite these advantages, the individual variation in performance results not only from variation in EF ability but also from variation in non-executive abilities, i.e., language and motor skills (Wiebe et al., 2011). Second, among three year old children with medium to high levels of externalizing behavior problems, there is a subgroup of children who shows a decrease in these problem behaviors over the preschool period (Shaw et al., 2005). Possibly, the behavior problems of these three year old children are more associated with other risk factors (e.g., environmental) than with EF impairments, resulting in a weaker relation between EF and externalizing behavior problems. A longitudinal design is needed to test the hypothesis that EF impairment is a risk factor that plays a role in the stability of externalizing behavior problems in the preschool period.

Regarding sampling method, this meta-analysis clearly shows that EF impairments are not restricted to preschool children from clinical samples, but are also evident in preschool children from community samples. Small effect sizes were found in the community samples and medium to large effect sizes were found in the referred/selected samples. There was a significant difference between the sampling methods for the overall EF and inhibition domain. This result indicates a stronger relationship between EF performance and externalizing behavior problems when the behavior problems are more severe. Thus, the relation between EF (and especially inhibition task) performance and externalizing behavior problems may be nonlinear in nature. We assume that the role played by EF as a risk factor is less pronounced in community samples compared to referred samples.

Gender distribution of the sample appeared to have an effect on the relationship between EF and externalizing behavior problems. This result indicates a stronger relation between EF performance and externalizing behavior problems when studies included a higher percentage of boys. As it was not possible in this meta-analysis to directly compare EF performance between preschool boys and girls with behavior problems, further empirical studies are needed to examine this issue.

Recently, a meta-analysis was published investigating neuropsychological functioning in preschoolers with ADHD symptoms (Pauli-Pott & Becker, 2011). The data used for the current meta-analysis differs in a number of ways from the data used by Pauli-Pott and Becker (2011). First, we not only included studies measuring ADHD symptoms, but we also included studies of children with aggressive and DBD symptoms. Second, we added seven more recent studies. Third, we strictly defined preschool-age, therefore we limited ourselves to studies with a mean sample age until 6 years age, whereas Pauli-Pott and Becker(2011) included studies with a mean age until 7 years. Fourth, in contrast to the other meta-analysis, we included only the first of multiple assessments of the same sample to avoid dependency between data.

Even though additional studies and groups were included and stricter inclusion criteria were used, comparable results were found in the two meta-analyses, with slightly higher effect sizes found in the meta-analysis by Pauli-Pott and Becker (2011). However, the results of the current meta-analysis extend the findings of Pauli-Pott and Becker. First, additional evidence is provided on an overall EF and factor level instead of only on task level. Second, since the present meta-analysis includes EF data from preschool children with ADHD and DBD symptoms, this meta-analysis provides a broader perspective on EF impairments in preschoolers with externalizing behavior problems rather than specific for ADHD. This is important as in the preschool period ADHD and DBD symptoms are strongly associated (Sterba et al., 2007).

Finally, a number of limitations of this meta-analysis need to be considered. First, there was quite a broad range of operationalizations of EF (e.g., 16 different inhibition tasks) and externalizing behavior problems across studies. This could have influenced the strength and specificity of the results. Second, additional moderators could influence the strength of the relationship, e.g., Social Economic Status of the family, ethnicity, and IQ. Although these moderators were considered at the set-up of this meta-analysis, there was not enough information available in the studies to conduct these analyses. Third, in about half of the studies the sample size was relatively small. Fourth, studies were included in which the mean age of the children was between 3.0 and 6.0 years. Consequently, there were a number of studies in which some of the children were older than 6.0 years, therefore the finding should be interpreted with caution.

The findings of the present study have clinical implications. First, traditionally much attention has been given to environmental factors that affect young children with externalizing behavior problems (Campbell et al., 1994). The last years, however, more research has been conducted on child characteristics. This meta-analysis clearly shows that EF impairments can be identified in preschool children with externalizing behavior problems. Second, programs to train EF skills have been developed (Diamond & Lee, 2011). Such programs have been shown to increase EF performance, especially children with the initially poorest executive functions benefit most from these programs. EF predicts later academic performance, so improvement in EF may lead to improvement of school readiness and academic achievement (Diamond & Lee, 2011). Thus, early executive function training may reduce the achievement gaps later.

In conclusion, this meta-analysis shows that EF impairments in children with externalizing behavior problems can already be identified in the preschool period. However, more research needs to be undertaken to improve our understanding of the association between EF and externalizing behavior problems at this young age. An important limitation of the current literature is that little attention has been paid to differentiate EF deficits in DBD and in ADHD. Even though this is a challenge due to the high correlation between ADHD and DBD symptoms, future studies should assess EF performance for ADHD and DBD separately (e.g., Schoemaker et al., 2012; Youngwirth et al., 2007). Most importantly, longitudinal studies are needed to investigate the role of EF impairments in the stability of externalizing behavior problems.

APPENDIX

Figure 2. Forest Plot of Working Memory Tasks (K=13), Mean ES with 95% CI

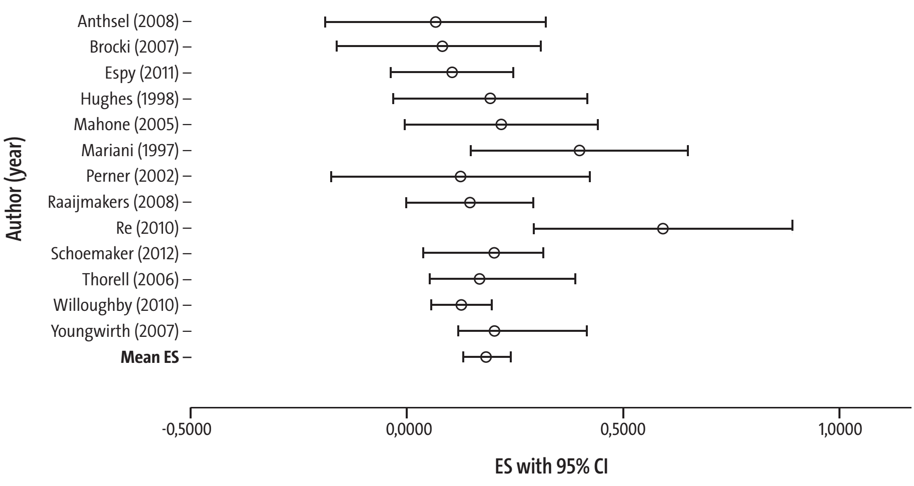


Figure 3. *Forest Plot of Inhibition Tasks (K=19) Mean ES with 95% CI*

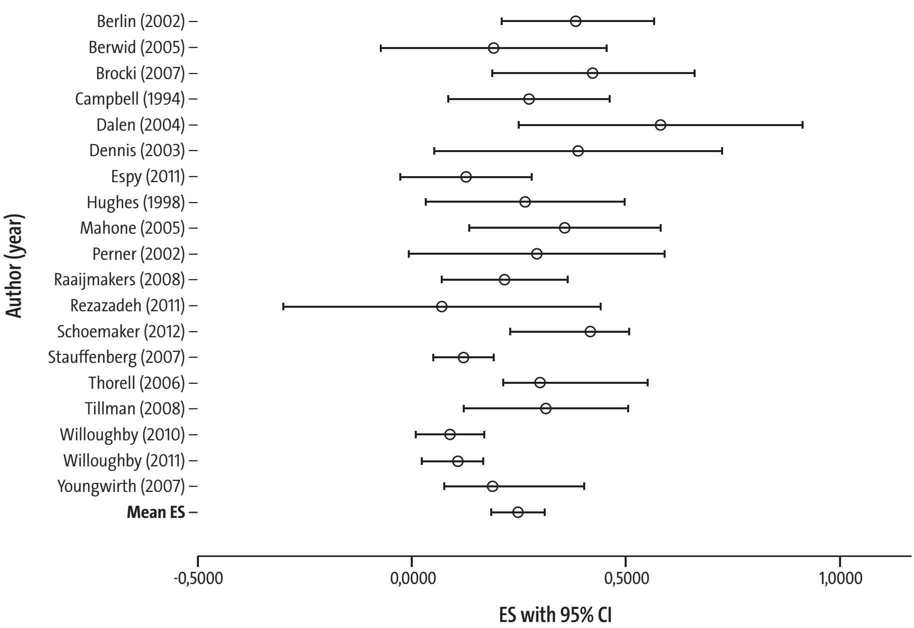
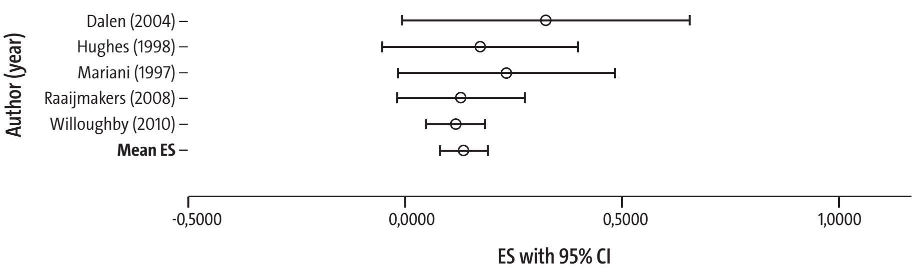


Figure 4. *Forest Plot of Cognitive Flexibility Tasks (K=5), Mean ES with 95% CI.*



Tabel 4. *EF Tasks Included in the Meta-Analysis with Description and Dependent Variable*

EF task	**Description**	**Dependent Variable**	**Study**
Working Memory tasks			
Digit/Word Span (Number recall)	Child is asked to repeat a list of words or digits.	# correct	Brocki (2007); Mariani (1997); Raaijmakers (2008); Thorell (2006)
		Max length of digit strings recalled	Espy (2011)
Spatial memory	Child has to recall the location of pictures within a matrix	# correct	Brocki (2007); Mariani (1997); Thorell (2006)
Digit span backwards	Child has to recall the words backward	# correct	Brocki (2007); Perner (2002)
Delayed Alternation	Child has to find treat underneath 1 of 2 cups, in alternating side	# correct	Espy (2011); Schoemaker (2012)
Six/nine boxes	Find a figure in different colored boxes that are scrambled after each trial	# correct / # total searches	Espy (2011); Schoemaker (2012)
Selective reminding	Child has to repeat a group of unrelated words	# correct	Mariani (1997)
Sentence repetition	Child has to recall exact words sentence	# correct [2=fully, 1 = partly]	Youngwirth (2007)
Span like	Remember animal drawing in house-drawing	Accuracy score	Willoughby (2010)
Noisy book	Child has to press the picture that is been called	Longest sequence on 2/3 trails, pass: median split	Hughes (1998)
Narrative memory	Child has to summarize story that just has been told.	# correct (free recall and cued recall)	Youngwirth (2007)
Delayed response	Find treat after delay	# correct	Espy (2011)

Tabel 4. continued

EF task	Description	Dependent Variable	Study
Dual Request Selective Task	E touches different cells in matrix, C has to recall first position	# correct	Re (2010)
Multiple boxes Test	C has to find all prizes in each box (boxes are not scrambled but out of sight between trials)	Total repetition errors	Mahone (2005)
Inhibition tasks *Delay Tasks*			
Picture Learning	C has to recall pictures in right order	# correct	Anthsel (2008)
NEPSY Statue	C has to stand still like a statue while E distracts	# Episodes without movement	Brocki (2007); Espy (2011)
		Movement per episode [3 point scale, 2= no errors, 0= more than 2 errors]	Perner (2002); Mahone (2005); Youngwirth (2007)
Delay Aversion	C has to choose between one 1 sweet directly or 2 sweets with delay	Eating per trial on 8 trials [3 point scale; 0 = sweet, 2 = waited]	Dalen (2004)
Delay of Gratification	C has to wait till E return in room and can eat larger pile of sweets	Time waited to eat treat	Stauffenberg (2007)
Delay of Gratification	C has to wait till E claps before retrieving cookie under cup	8 trails, 3 point scale [0=ate- 2=waited]	Dalen (2004); Campbell (1994)
Snack Delay	C has to stand still and wait for treat	Mean score across 3 trials, [4 point scale, 1 = eated, 4 = waited]	Willoughby (2011)
		# episodes without movement	Schoemaker (2012)
Resistance to temptation	C is engaged in toy, E then leaves the room and C may not touch the toy	# intervals touching toy	Campbell (1994)

Tabel 4. continued

EF task	Description	Dependent Variable	Study
***Inhibition tasks** Response inhibition*			
GoNoGo task	Press key when see target stimuli (go condition), don't press a key when see non-target stimuli (nogo condition)	Commission errors	Berlin (2002); Berwid (2005); Brocki (2007); Raaijmakers (2008); Schoemaker (2012)
		Accuracy hits minus false	Perner (2002)
		Accuracy score	Willoughby (2010)
Stop Signal Task	Press spacebar when a car appeared, except when stop sign (25%)	Commission errrors	Tillman (2008)
***Inhibition tasks** Interference – stroop*			
Day-Night stroop	Child has to say the opposite word, for example say day when picture of moon	# correct	Berwid (2005); Brocki (2007); Raaijmakers (2008); Thorell (2006); Tillman (2008)
		% incorrect	Stauffenberg (2007)
		Mean reaction time for correct responses	Rezazadeh (2011)
Silly-sound Stroop	Stroop like task: C has to make sound of dog when presented with picture of a cat , and visa versa	Accuracy score	Willoughby (2010)
***Inhibition tasks** Other inhibition tasks*			
Knock and Tap	When E knocks C has to tap on the table, or C has to not respond	# correct	Brocki (2007); Perner (2002)
Luria's handgame	When E shows fist, C has to point a finger and visa versa.	Pass: no more than 1 error over the 10 trials	Hughes (1998)
Pencil Tapping	If E taps pencil twice, C has to tap once (and visa versa)	# correct	Willoughby (2011)
Tongue Task	C has to place a M&M on their tongue and wait 40 seconds before they can eat it	# sec C waited before eating	Willoughby (2011)

Tabel 4. continued

EF task	Description	Dependent Variable	Study
Detour Reaching Box	Child has to retrieve marble from box but has to inhibit the predominant response	Pass: correct on 3 consec. trials, (max 15)	Hughes (1998)
Puppet says	C has to do what 1 puppet says, but has to inhibit response from other puppet	0/1/2 value on 8 NG trials	Dalen (2004)
Shape School – inhibition condition	Inhibit to name color when sad face	# incorrect inhibition trials	Raaijmakers (2008)
		# correct	Schoemaker (2012)
		Completion time	Espy (2011)
Spatial Conflict	Touch an item car/boat which is displayed on card	Accuracy score	Willoughby (2010)
Inhibition scale of Effortful control battery	Four tasks: Simon Says, Whispering on demand, taking turns while building a tower and matching game with distracters	Scores from Kochanska's effortful control battery	Dennis (2003)
Cognitive flexibility tasks			
Set shifting	Sort card according diff stimuli (3 rules, identity, color or size)	Pass: no more than 3 errors on any rule	Hughes (1998)
Colour form test	Child has to point to the response, switches from colour to shape	# errors	Mariani (1997)
Block sorting	Sort blocks according to different rules, color, shape etc	# correct	Dalen (2004)
Shape School - Switch condition	Child has to name figures according to opposite color	# errors	Raaijmakers (2008)
Object Classification Task for Children	Sort Pictures according different rules	# correct	Raaijmakers (2008)
Item selection	Select pictures according different dimensions (shape, color, size)	Accuracy score	Willoughby (2010)

C: child; E: examiner

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Executive Function Deficits in Preschool Children with ADHD and DBD

ABSTRACT

Background: Impairments in executive functions (EF) are consistently associated with attention deficit hyperactivity disorder (ADHD) and to a lesser extent, with disruptive behavior disorder (DBD), i.e., oppositional defiant disorder or conduct disorder, in school-aged children. Recently, larger numbers of children with these disorders are diagnosed earlier in development, yet knowledge about impairments in clinically diagnosed preschool children and the role of co-morbidity is limited. Therefore, the aim of the current study was to examine EF in clinically referred preschool children with a clinical diagnosis of ADHD, DBD, and ADHD+DBD. ***Method:*** Participants were 202 children aged 3,5 to 5,5 years, 61 with ADHD only, 33 with DBD only, 52 with co-morbid ADHD+DBD, and 56 typically developing children. Five EF tasks were administered. ***Results:*** Confirmatory factor analysis showed that the two-factor model (inhibition and working memory) fit the data better than a one-factor model in this clinical sample. Preschoolers with ADHD displayed inhibition deficits, also after controlling for IQ. Likewise, preschoolers with DBD displayed impaired inhibition, but when IQ was controlled differences were carried mostly by the effect on the task were motivational demands were high (i.e., when tangible rewards were used). This pattern was also found in the interaction between ADHD and DBD; impaired inhibition in the co-morbid group, however, was more severe than in the DBD group. Regarding working memory, few group differences were found. ***Conclusions:*** Clinically diagnosed preschool children with ADHD showed robust inhibition deficits, whereas preschool children with DBD showed impaired inhibition especially where motivational incentives were prominent. Severity of inhibition impairment in the co-morbid group was similar to the ADHD group.

Keywords*:** Executive functions, preschool children, DBD, ADHD ***Abbreviations: ADHD: attention deficit hyperactivity disorder, DBD: disruptive behavior disorder, EF: executive functions

INTRODUCTION

School-aged children with behavior problems show robust impairments in executive functions (EF; Oosterlaan, Logan, & Sergeant, 1998). EF can be defined broadly as the top-down control of cognitive processes to achieve a purpose or goal (Séguin & Zelazo, 2005). Miyake et al. (2000) proposed that, in adulthood, EF is a unitary construct with three dissociable components: working memory, inhibition and set shifting. Children with attention deficit hyperactivity disorder (ADHD) show EF deficiencies, especially in inhibition (Willcutt et al., 2005). In the meta-analysis by Oosterlaan et al. (1998), however, deficits in inhibition were not uniquely associated with ADHD, but also with the two disruptive behavior disorders (DBD), i.e., oppositional defiant disorder (ODD) and conduct disorder (CD). Other studies have shown deficiencies in inhibition in school-age children and adolescents with DBD, specifically when motivational processes, i.e., reward and punishment, are involved (Matthys, Van Goozen, De Vries, Cohen-Kettenis, & Van Engeland, 1998; Matthys, Van Goozen, Snoek, & Van Engeland, 2004; Schutter, Van Bokhoven, Vanderschuren, Lochman, & Matthys, 2011). Although these studies provided valuable information on the role of EF in school-aged children with ADHD and the DBD, chronic patterns of hyperactivity and behavior problems can already be identified in the preschool years (Shaw, Lacourse, & Nagin, 2005). Furthermore, there is increasing evidence that diagnoses of ADHD and DBD can be made reliably in preschool children (Keenan et al., 2007), and an increasing number of children are diagnosed clinically in preschool. It is not clear whether preschoolers with diagnosed, externalizing clinical disorders will show EF deficits similar to those observed in diagnosed school-age children or whether such deficits do not emerge until later in development, particularly as characterizing EF in preschoolers is not straightforward.

Indeed, there is substantial debate regarding the organization of EF in preschool children, i.e., whether EF is a unitary construct (Wiebe, Espy & Charak, 2008, in a community sample) or if separable components (e.g., working memory, inhibition, and shifting) can be identified at this young age (Hughes, Dunn & White, 1998, with hard to manage preschoolers). In their review, Garon, Bryson and Smith (2008) propose that the EF components develop hierarchically during the preschool period (working memory followed by inhibition followed by set shifting), although there is no specific evidence to date to support this developmental timetable.

In recent years there has been increasing interest in the study of EF, especially on inhibition and working memory, in preschool children with behavioral problems. In line with findings with older children, Mariani and Barkley (1997) found impairments on tasks designed to measure inhibition in clinically-diagnosed preschoolers with ADHD. Likewise, in community samples, inhibition impairments also have been observed, either when ADHD symptoms are defined categorically (Dalen, Sonuga-Barke, Hall, & Remington, 2004; Thorell & Wåhlstedt, 2006; Youngwirth, Harvey, Gates, Hashim, & Friedman-Weieneth, 2007) or continuously (Berlin & Bohlin, 2002; Sonuga-Barke, Dalen, Daley, & Remington, 2002; Tillman, Thorell, Brocki, & Bohlin, 2008; Von Stauffenberg & Campbell, 2007). For working memory, results are inconsistent, with some noting an impairment (Mariani & Barkley, 1997; Thorell & Wåhlstedt, 2006) and others not (Sonuga-Barke et al., 2002; Youngwirth et al., 2007).

While meta-analyses have confirmed the presence of impaired inhibition task performance in older children with DBD (Morgan & Lilienfeld, 2000; Oosterlaan et al., 1998), it is unclear whether such deficits also are evident in preschoolers. Results from several studies have revealed inhibitory deficits in preschoolers with DBD symptoms, but these were not robust after controlling for ADHD symptoms (Berlin & Bohlin, 2002; Sonuga-Barke et al., 2002; Thorell & Wåhlstedt, 2006; Youngwirth et al., 2007, but see Raaijmakers et al., 2008 for an exception). In contrast, preschoolers with symptoms of DBD do not appear to show deficits on working memory tasks (Raaijmakers et al., 2008; Thorell & Wåhlstedt, 2006; Youngwirth et al., 2007).

Attempts to delineate EF impairments in preschoolers with behavior problems at present are incomplete. Because most studies have used community samples with less severely disordered children, further investigation of EF is warranted in clinical samples of preschool children with ADHD and DBD, who have more severe behavior problems. For example, the lack of EF impairment found in preschoolers with DBD might be related to the lower DBD symptom severity in community samples studied thus far.

Furthermore, the role of co-morbidity has been largely ignored, despite the fact that about half of the children with ADHD are also diagnosed with ODD, and the percentage of children with ODD who have co-morbid ADHD is even higher (Kutcher et al., 2004). Only one study included a co-morbid ADHD+ODD group from a community sample (Youngwirth et al., 2007); these children exhibited deficits on both inhibition and working memory tasks.

The aim of the present study was to examine EF in clinically referred preschool children with a confirmed, clinical diagnosis of ADHD, DBD, and ADHD+DBD. A comparison group of typically developing (TD) children was also included. We capitalized on recent advancements in preschool EF assessment including tasks that were designed to preferentially measure inhibition or working memory, were developed specifically for use in this age range, and varied in their motivational demands. Based on results from studies with older diagnosed children, we expected that children with ADHD would show deficits on both inhibitory and working memory tasks, even when controlling for co-morbid DBD symptoms. Further, preschoolers with DBD were hypothesized to display selective impairments on inhibitory tasks (but not on those selected to measure working memory), also after controlling for ADHD symptoms, especially when motivational demands were more prominent. Furthermore, we expected that the co-morbid group would display inhibition as well as working memory deficits.

METHOD

Participants

Participants were 202 children aged 3,5 to 5,5 years with ADHD (N=61), DBD (N=33), ADHD+DBD (N=52), and TD children (N=56). Children with disorders were referred by general practitioners, well-baby clinics and pediatricians for clinical assessment at the Outpatient Clinic for Preschool Children with Behavioral Problems, Department of Child and Adolescent Psychiatry, University Medical Centre Utrecht. Children were diagnosed as ADHD, DBD (i.e., ODD or CD) or ADHD+DBD on the basis of the strict application of the DSM-IV-TR criteria for these disorders (American Psychiatric Association, 2000). Consensus was reached between a child psychiatrist and a clinical child psychologist using the following data sources: 1) the scores within the clinical range on the Attention Problems scale and the Aggressive Behavior scale of the Child Behavior Checklist completed by parents (CBCL/1,5–5) and the Child Teacher Report Form completed by teachers or day-care caregivers (C-TRF /1,5–5); (both: Achenbach & Rescorla, 2000; Dutch version by Verhulst & Van der Ende, 2000); 2) the symptoms reported on the Kiddie Disruptive Behavior Schedule (Keenan et al., 2007), a semi-structured DSM-IV based parent interview for the assessment of ADHD, ODD and CD in preschool children; 3) the scores on the Child Global Assessment Schedule (Shaffer et al., 1983), a measure of the impairment of the functioning of the child, filled out by the parents as well as the teacher/caregiver; and 4) the observation of the child's behavior using the Disruptive Behavior Diagnostic Observation Schedule (Wakschlag et al., 2008a, 2008b), a structured observation that evaluates the child's behavior during tasks systematically varying in the level of challenge and support.

The TD group was recruited from regular primary schools and daycare centers. Children with a score in the normal range on the Attention Problems scale and on the Aggressive Behavior scale of the CBCL and C-TRF were included.

All children with an IQ below 70, estimated by the average of the scores on the Raven Coloured Progressive Matrices (Raven, Court, & Raven,1998) and the Peabody Picture Vocabulary Test-III-NL, (Dunn & Dunn, Dutch translation by Schlichting, 2005) were excluded. None of the preschoolers in the clinical groups were on medication. The characteristics of the four groups are displayed in *Table 1*. The four groups did not differ on age ($p = .102$) or proportion of males ($p = .329$). There were group differences in estimated IQ ($p < .001$), with the TD group significantly outperforming the three clinical groups, who performed similarly to each other.

Procedure

Children were evaluated in a single, morning session. First, the two measures of intellectual functioning were administered, followed by the EF tasks. All tasks were administered individually by trained master's students in a quiet room with a one-way mirror. One parent was in the room with the child and the assessment was recorded. The tasks were administered in a fixed order and lasted about two hours, including breaks. After another break the child observation and parent interview were administrated. Parents received nominal financial compensation for participating and children received two small gifts. Written informed consent from the parents was obtained before participating and the study was approved by the Medical Ethical Review Committee of the University Medical Center Utrecht.

Table 1. Means (and SD) for the Demographics and Control Variables in the Four Groups

	TD (1) (n=56)		ADHD (2) (n=61)		DBD (3) (n=33)		ADHD+DBD (4) (n=52)		ANOVA	Post-hoc
	M	SD	M	SD	M	SD	M	SD	F/χ (3,202)	Bonferroni
Age (months)	55.66	7.18	55.20	7.41	51.88	8.29	54.12	6.80	2.10	–
% boys	69.60		80.30		81.80		82.70		3.44	–
IQ estimate	111.65	10.32	101.29	12.00	101.89	10.90	99.76	11.61	12.60*	1>2,3,4
CBCL attention	50.88	2.14	68.10	7.16	64.45	8.17	69.44	6.73	102.31*	1<2,3,4+3<2, 4
CBCL aggression	50.52	1.31	63.02	10.08	76.76	9.93	75.58	11.86	89.38*	1<2,3,4+2<3,4
TRF attention	52.05	3.85	70.93	10.96	59.69	7.41	70.88	12.77	50.67*	1<2,3,4+3<2, 4
TRF aggression	52.02	3.10	62.92	10.09	62.41	10.11	67.65	12.17	26.90*	1<2,3,4

* $p<.001$

Measures

The EF tasks used in this study were adapted from those used in Wiebe et al. (2011). The computerized tasks are administered through E-Prime version 1.2 (Psychology Software Tools, Pittsburgh, PA). All tasks were preceded by practice trials, to make sure the children adequately understood, and could perform, the tasks. Three tasks were considered to preferentially measure inhibitory skills, i.e., Go-No-Go, Modified Snack Delay, and Shape School – Inhibit Condition, and two more to preferentially assess working memory abilities, i.e., Nine Boxes and Delayed Alternation.

The ***Go-No-Go task*** is a computerized task where children were instructed to catch as many fish (Go stimuli, 75%) as possible by pressing the button when a fish appeared on the screen. They were instructed to let the shark (No-Go stimuli, 25%) swim by withholding the button press. Auditory feedback was provided when appropriately catching a fish or inappropriately catching a shark. Stimuli were presented for 1500 ms, with an inter-stimulus interval of 1000ms. The dependent variable was the proportion correct, the number of No-Go trials the child correctly did not press the button divided by the total number of No-Go trials.

Modified Snack Delay is a newly developed task that integrates the motivational context from the original Snack Delay paradigm (Kochanska, Murray, Jacques, Koenig, & Vandegeest, 1996) with the motor – inhibitory control demands of NEPSY statue (Korkman, Kirk, & Kemp, 1998). Children were instructed to stand still like a snowman with their hands on a mat, without talking or moving. In front of the child a glass with a treat underneath and a bell was placed. The child was instructed that when the examiner rang the bell, the child could move again and eat the treat. The task lasted 4 minutes, during which the examiner progressively distracted the child with various activities (e.g., dropping a pencil, knocking under the table) and culminating in leaving the room for 90 seconds. During each 5-second interval, four different behaviors categories were rated from DVD by trained raters: moving body, moving hands, talking and 'reward-related behavior' (e.g., eating treat, touching bell). Twenty percent were double coded to determine inter-rater reliability (mean inter-rater agreement = 88%). The dependent variable was the number of intervals that the child complied with all task rules (not moving, talking or 'reward-related behavior') divided by the total number of intervals (i.e., 48).

The ***Shape School - Inhibit Condition*** is a computerized task with cartoon figures with different shapes, colors, and expressions, where the naming rule differs in varying conditions. In the Inhibit condition, participants had to name the color of the figures with happy faces and suppress the prepotent color naming response when the figure had a sad/frustrated face. The dependent variable was the number of correct responses divided by the total number trials (i.e., N = 18).

In ***Delayed Alternation***, the child had to find a treat underneath one of two identical cups, where after a correct retrieval, the reward alternated to the opposite side in the next trial. There was a 10-second delay between trials, during which the treat was hidden out of the child's sight and the examiner actively distracted the child. A maximum of 16 trials was administrated or if the child made eight consecutive correct responses. The dependent variable was the number of correct retrievals divided by the by total trials.

In ***Nine Boxes***, children were instructed to find all 'Barbapapa' characters hidden in nine different colored boxes (with different shapes on the lid). The child was allowed to open one box per trial, and the boxes were shuffled behind a screen between trials during the 10 second delay. A maximum of 20 trials were administered, until the child found all characters or made five consecutive errors. The dependent variable was the number of correct retrievals divided by the total trials.

For the Go-No-Go and Snack Delay tasks, test retest was good, exceeding .80; the Shape School-Inhibit condition task was adequate (.71) and Delayed Alternation and Nine Boxes were less than desired (<.70) for use with individual children (Espy, Bull, Kaiser, Martin, & Banet, 2008; Espy, Wiebe, & Sheffield, 2009). Note that most reliabilities were calculated on somewhat different dependent variables than used here.

RESULTS

For all analyses, missing data (3.5%) in the EF measures were imputed, considering age, gender, IQ, groups assignment and performance on other EF tasks as auxiliary variables.

Factor analysis

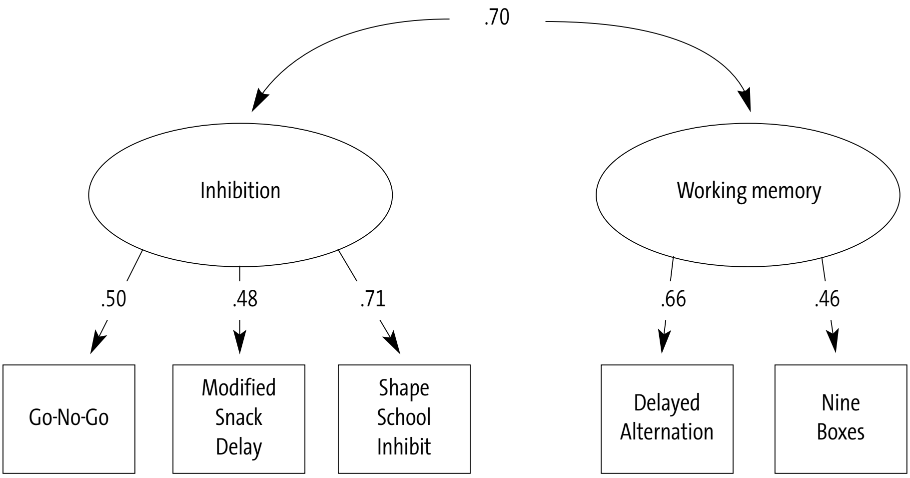
Before conducting factor analyses, the correlations between the EF tasks were calculated (*Table 2*). Confirmatory factor analyses were performed using the AMOS program, where both a one- and two-factor model were tested. In the one-factor model, all tasks loaded on one common factor. In the two-factor model, the Nine Boxes and Delayed Alternation tasks loaded on the working memory factor, and the Shape School, Go-No-Go and Snack Delay loaded on the inhibition factor (see *Figure 1*). The overall model fit was based on the χ^2 test, the root-mean-square error of approximation (RMSEA), comparative fit index (CFI) and Tucker and Lewis Index (TLI). Both models showed adequate fit to the data (one-factor model: χ^2 (5) =6.04, p = .302; RMSEA = .03, TLI = .98, CFI = .99; two-factor model: χ^2 (4) = .47, p = .976; RMSEA = .00, TLI = 1.09, CFI = 1.00). The models were compared with the χ^2 difference test. The two-factor model fitted significantly better than the one-factor model ($\Delta\chi^2$ = 5.57, df = 1, p =.018). Subsequently, the two-factor scores were computed using a two-factor exploratory factor analysis in SPSS (SPSS version 18.0, Chicago, IL, USA), the output was used as dependent variables.

Table 2. Correlation Between EF Tasks

	Go-No-Go	Modified Snack Delay	Shape School	Delayed Alternation	Nine Boxes
Go-No-Go	–	.25***	.36***	.21**	.17*
Modified Snack Delay		–	.34***	.23**	.18*
Shape School			–	.33***	.22**
Delayed Alternation				–	.31***
Nine Boxes					–

** p <.05, **p <.01, *** p <.001*

Figure 1. Two-Factor Model with Standardized Regression Weights (and correlation between factors)



Group differences

Our aim was to get a detailed picture of the EF performance in preschoolers with ADHD and/or DBD. Therefore, a 2 (ADHD vs. no ADHD) x 2 (DBD vs. no DBD) factorial design (utilizing two dummy variables to represent the factors) was used to compare the EF performance of children with ADHD only (ADHD=1, DBD=0), with DBD only (ADHD=0, DBD=1), co-morbid with both disorders (ADHD=1, DBD=1) and those with neither (TD controls, ADHD=0, DBD=0). This 2x2 multivariate analysis, instead of an analysis with four independent groups, is the recommended analytic design to test whether the performance of the group with two disorders could be described as an additive combination of the deficits found in the singly disordered groups or was due to an interaction of ADHD and DBD (for a similar procedure, see Willcutt et al., 2001). We studied different levels of dependent variables with this model: first at an overall level, second at a two EF factor level, and third at the individual task level, proceeding to the next level only when the preceding was significant.

The 2x2 groups differed in age (main effect DBD, $p = .023$), and therefore age was included as a covariate in all analyses. There is a debate regarding whether or not IQ should be included as a covariate in the analyses. On the one hand, including IQ as a covariate ensures that deficits in clinical groups cannot be explained by known group differences in intelligence. On the other hand, both ADHD and DBD typically are associated with mild IQ differences in comparison to individuals without these disorders. Controlling for IQ then may remove inappropriately the overlapping portion of the variance that is associated with the disorder. Because of these contrasting views, all results are reported both with and without controlling for IQ (see, for example also Willcutt et al., 2001). In the present study IQ accounted for 14% of the variance in the inhibition factor and 4% of the variance in the working memory factor.

First level: overall EF

First, to test whether either ADHD or DBD was associated with deficits at the overall EF level, repeated measure MANCOVAs were conducted with the two dummy coded diagnostic indicators as the between subjects variables and the inhibition and working memory factor as the within-subject variable. Results of the repeated measure MANCOVAs show that there was a significant main effect (controlling for age, but not IQ) of ADHD ($F = 33.56$, $p = .000$), DBD ($F = 8.04$, $p = .005$) and the respective interaction ($F = 6.02$, $p = .015$), indicating that children with ADHD, DBD, and ADHD+DBD diagnoses performed more poorly at the overall EF level (collapsed across the two EF factors). Adding IQ to the model as a covariate resulted in the same pattern of results, ADHD ($F = 23.04$, $p = .000$), DBD ($F = 4.59$, $p = .033$) ADHD x DBD ($F = 3.79$, $p = .053$). Therefore, further testing of the impact of diagnostic category on each EF factor was warranted.

Second level: EF factors

Second, to test whether either ADHD or DBD was associated with deficits on the EF measures independent of the other disorder, separate 2 x 2 (ADHD x DBD) MANCOVA were conducted for the EF factors, followed by planned contrasts. *Table 3* presents the unadjusted means of the four groups on the individual EF tasks and two-factor scores, together with the results of the 2 x 2 MANCOVAs controlling for age only. The analyses revealed a significant main effect of ADHD, a main effect for DBD and an interaction effect between ADHD and DBD for the inhibition factor, but not for the working memory factor. We added IQ to the model as an additional covariate besides age (see *Figure 2*). For the inhibition construct the main effect of ADHD ($F = 20.50$, $p = .001$), of DBD ($F = 4.65$, $p = .032$) and of ADHD x DBD ($F = 5.90$, $p = .016$) remained significant. For the working memory construct, the ADHD, DBD, and ADHD x DBD effects remained non-significant with IQ controlled.

Following planned contrasts were conducted, for the significant effects, in which the performance of each clinical group was directly compared with the performance of the TD group. Each clinical group differed significantly from the TD group ($p < .05$) on the inhibition factor with and without controlling for IQ.

Third level: EF tasks

The results at the level of the inhibition factor was significant, so follow-up MANCOVA analyses were conducted for the three inhibition tasks, to determine which tasks contributed to the observed group differences on the construct. There was an ADHD main effect and a DBD main effect evident on each of the three inhibition tasks. For the ADHD x DBD effect, the inhibition differences were carried mostly by a significant interaction effect on Modified Snack Delay (*Table 3*). Adding IQ to the model altered the pattern of results somewhat for the DBD main effect, now the inhibition differences were carried mostly by a significant effect on the Modified Snack Delay task ($F = 5.34$, $p = .022$) and not by the Go-No-Go and Shape School task anymore. The ADHD main effect remained significant for all three inhibition tasks and the ADHD x DBD effect remained significant for the Modified Snack Delay task only ($F = 4.57$, $p = .034$).

For the inhibition tasks planned contrasts were specified in which the performance of each clinical group was directly compared with the performance of the TD group. Each clinical group differed significantly from the TD group ($p < .05$) on the inhibition tasks. Adding IQ to the model the results remained the same, with one exception, the contrast between DBD and TD on Shape School-Inhibit Condition was marginally significant ($p = .058$).

Table 3. Unadjusted Means (and SD) of the EF Factors and Tasks in the Four Groups

	TD N=56		ADHD only N=61		DBD only N=33		ADHD+DBD N=52		Main Effect ADHD[a]	Main effect DBD[a]	Interaction ADHD x DBD[a]
	M	SD	M	SD	M	SD	M	SD	F (1, 197)	F (1, 197)	F (1, 197)
Inhibition factor	.72	.84	–.31	.92	–.14	.92	–.35	.93	29.11***	7.54**	8.18**
Go-No-Go	.88	.15	.71	.28	.72	.24	.70	.31	8.10**	3.94*	3.51
Modified Snack Delay	.34	.24	.13	.17	.17	.21	.11	.17	28.42***	7.12**	5.74*
Shape School inhibit	.90	.19	.71	.28	.71	.27	.69	.27	13.62***	4.08*	3.20
Working Memory factor	.24	.97	– .03	1.01	–.10	1.04	–.17	.99	2.82	0.52	0.03
Delayed Alternation	.69	.17	.59	.21	.65	.19	.61	.22	9.10**	0.19	0.49
Nine Boxes	.67	.16	.63	.16	.59	.16	.59	.14	1.35	4.40*	0.21

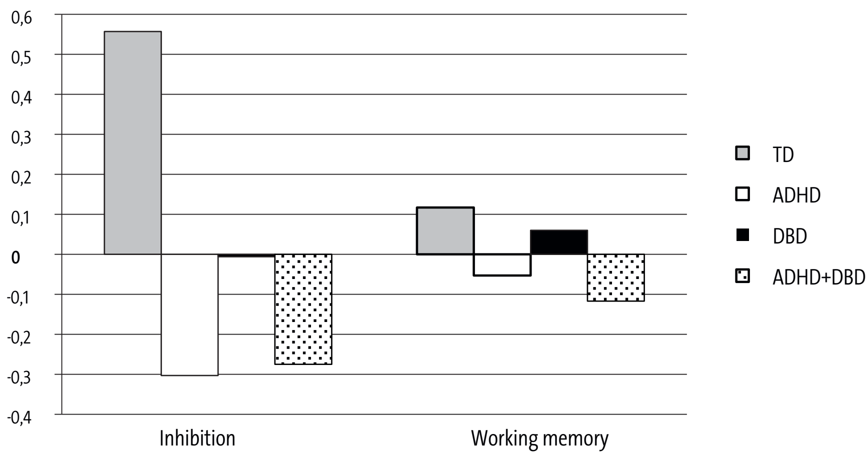
**p <.05, ** p< .01, ***p<.001*
[a] The results of analyses with age as covariate.
All variables of the EF tasks are proportion correct and the factor are Z-scores.

Controlling co-morbid symptoms

Finally, we used this model and added dimensional measures of behavior problems as a covariate. Thus, to test whether there were still significant main effects of ADHD and/or DBD when controlling for co-morbid symptoms, additional 2x2 MANCOVAs were conducted with the dimensional measures of the other disorder as a covariate (Aggressive Behavior and Attention Problems scores on CBCL/TRF), along with age and IQ.

First, the specificity of the association between ADHD and inhibition impairments was examined by adding CBCL aggression scores as a covariate. After controlling for age, IQ and CBCL aggression scores, the ADHD main effect remained significant for the inhibition factor ($F(1, 197) = 19.53$, $p < .001$). This warranted us to conduct the analyses for each individual task, the effects remained significant as well: Go-No-Go ($F(1, 197) = 4.44$, $p = .036$), Modified Snack Delay ($F(1, 197) = 19.95$, $p < .001$), and Shape School-Inhibit ($F(1, 197) = 9.21$, $p = .003$). Second, to test the specificity of the association between DBD and inhibition impairments was tested by adding CBCL attention scores as a covariate. When age, IQ and CBCL attention scores were included as covariates, the previously significant main effect was no longer significant for the inhibition factor. Similar pattern of results were found when TRF (instead of CBCL) aggression and attention problems scores were used as a covariate.

Figure 2. Marginal Means of the EF Factors Adjusted for Age and IQ



DISCUSSION

The aim of the current study was to investigate EF in preschool children with ADHD and DBD taking into account co-morbidity. A clinically referred sample of preschoolers with ADHD, DBD and both disorders was included as well as a TD group. When the structure of EF was examined in the present sample of clinically diagnosed preschoolers, a two-factor model (inhibition and working memory) fit the data better than a one-factor model. This pattern of findings is in contrast to those of Wiebe and colleagues (2008; 2011) who found a one-factor model in a large sample of typically developing children without frank psychopathology. The two-factor structure likely fit better in the clinically diagnosed children reflecting the underlying pattern of clinical impairments.

Regarding inhibition, results of the present study showed, first, that preschool children with ADHD (independent of DBD) consistently showed substantial and specific inhibition deficits (i.e. on the inhibition factor and on all three inhibition tasks), also after controlling IQ and dimensionally for DBD symptoms. These results are in line with studies in community samples (e.g., Berlin & Bohlin, 2002, Youngwirth et al., 2007) and the only study in a clinical sample (Mariani & Barkley, 1997). The latter study, however, did not account for DBD symptoms. The present study is the first one to show specific inhibition deficits in a clinical sample of preschoolers with ADHD irrespective of DBD co-morbidity, either examined from a categorical or a dimensional point of view.

Second, results of the present study showed an impairment on the inhibition factor in preschool children related to DBD diagnosis that was robust after controlling for IQ, and this DBD-related impairment was most evident on the Modified Snack Delay task. In comparison to typically developing children, DBD-only children performed more poorly on all inhibition tasks, in line with results from studies of older children (see meta-analyses: Morgan & Lilienfeld, 2000; Oosterlaan, et al., 1998). The Modified Snack Delay task, i.e., the inhibition task in which salient motivational factors (i.e., food reward) are included in addition to the motor inhibitory demands was particularly sensitive to differences to DBD in this preschool-age range. This finding may be consistent with results from studies in older children and adolescents using tasks in which reward and punishment are included (Fairchild et al., 2009; Matthys et al., 2004; Schutter et al., 2011), although the nature of these tasks differ substantially and the comparability of these tasks across age ranges is unknown. Because the DBD-related differences on the inhibition factor disappeared after controlling for ADHD symptoms (attention problems), it appears as though performance on inhibition tasks was associated with ADHD symptoms and not with the DBD diagnosis itself. Given the high prevalence of subclinical levels of ADHD symptoms that co-occur with frank DBD disorder, particularly in this age range, impaired inhibition can be considered a characteristic of children with DBD, even though the 'true' source of inhibition deficits might not be the DBD per se.

Third, the co-morbid (ADHD+DBD) group displayed inhibition deficits, also after controlling for IQ. The pattern of the inhibition impairment was similar to the pattern of the DBD group, i.e., differences remained only on the Modified Snack Delay task when IQ was controlled. Impaired inhibition in the co-morbid group, however, was more severe than in DBD group. In conclusion, in terms of severity of impairment the co-morbid group was similar to the ADHD group, whereas in terms of pattern of the inhibition impairment the co-morbid group was similar to the DBD group.

Regarding working memory, no differences among the groups were found on the working memory factor, which is consistent with the findings of other studies (e.g. Sonuga-Barke et al., 2002). Because these tasks, or similar variants, have been shown to be sensitive to process differences in preschoolers with other disorders, such as prematurity (Espy, McDiarmid, Cwik, Senn, Hamby, & Stalets, 2004), it does not appear to be a task measurement issue per se, although reliability of these tasks was lower than the inhibition tasks, which limits the true variance that can be attributed to variables of interest. The reasons for not finding a working memory deficit might be twofold, with different reasons for the DBD and ADHD group. Moffitt (1993) reviewed neuropsychological studies of children with CD and found that these children showed substantive verbal impairments; these impairments were more severe than performance impairments. So it might be that children with DBD especially show verbal working memory problems. Unfortunately, in the present study, the tasks were designed to enable performance of very young children, so the demands were not modality specific, but certainly were designed to assess spatial working memory to a greater degree. Secondly, a meta-analysis (Willcutt et al., 2005) showed that school-age children and adolescents with ADHD displayed working memory impairments. So it could be that working memory impairments become more apparent at later ages for children with ADHD.

Some limitations should be considered when interpreting the results. Concerning the assessment of EF, the difficulty of some tasks and the age of the sample restricted the broad assessment which we aimed. As a result, set shifting was unable to be assessed. The development of tasks appropriate to assess EF in preschool children, especially in the younger ones, remains a challenge. Another limitation is a relatively small sample that prevented us to examine specific developmental differences, which could have provided a more thorough characterization of impairments across the preschool period.

Despite these limitations, this study is the first on EF in clinically diagnosed preschool children with ADHD, DBD and both disorders co-morbidly. Results show inhibition impairments in all three groups already at this young age, which is most pronounced in the ADHD group. Future research should assess EF in children with ADHD and/or DBD longitudinally to gain insight in the development of EF over time in these disorders and the role of EF impairments as factors involved in the persistence of ADHD and/or DBD. Likewise, the effect of EF impairment on various domains of functioning, such as academic outcome and peer relations, needs to be examined. Thus, a better understanding of the role of EF in the psychopathology of ADHD and DBD ultimately may improve intervention strategies for these disorders.

Key points

- *Clinically diagnosed preschool children with ADHD show inhibition deficits.*
- *Clinically diagnosed preschool children with DBD show impaired inhibition especially when motivational factors are involved.*
- *Preschool children with ADHD and/or DBD display no working memory deficits.*

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Executive Functions in Preschool Children with ADHD and DBD:

An 18 month Longitudinal Study

ABSTRACT

In this longitudinal study, we examined the stability of the association between executive functions and externalizing behavior problems, and the developmental change of executive functions in a predominately clinically diagnosed preschool sample (N=200). Inhibition and working memory performance were assessed three times in 18 months. Across time, inhibition impairments in young children were associated with ADHD and DBD, and working memory impairments were associated with ADHD. Inhibition and working memory performance increased over time, especially in the early preschool period. The improvement of inhibition performance was more pronounced in the clinically diagnosed children compared to the typically developing children.

Keywords: Executive functions, preschool children, DBD, ADHD. ***Abbreviations:*** ADHD: attention deficit hyperactivity disorder, DBD: disruptive behavior disorder

INTRODUCTION

Over the last decade there has been an increasing interest in executive functions (EF), both in typically developing young children and in young children with externalizing behavior problems. EF can be defined as the top-down control of cognitive processes to achieve a purpose or goal, i.e., the explicit control of thought, emotion and action (Séguin & Zelazo, 2005). Miyake et al. (2000) proposed that, in adulthood, EF is a unitary construct with three dissociable components: working memory, inhibition and set shifting (also referred to as cognitive flexibility). The EF structure during early childhood, however, is not yet clearly defined. In their review, Garon, Bryson and Smith (2008) propose that the EF components develop hierarchically during the preschool period, i.e., working memory followed by inhibition followed by cognitive flexibility. To empirically test the structure of EF, using confirmatory factor analyses, most studies found only one unitary executive construct in preschool community samples (Hughes, Ensor, Wilson, & Graham, 2010; Wiebe, Espy, & Charak, 2008; Wiebe et al., 2011; Willoughby, Blair, Wirth, & Greenberg, 2010). However, a two-factor model, inhibition and working memory, was also found, including a study with clinically diagnosed preschool children (Miller, Giesbrecht, Müller, McInerney, & Kerns, 2012; Schoemaker et al., 2012).

In cross-sectional studies, impairments in EF have shown to be associated with externalizing behavior problems in young children. In a meta-analysis, Pauli-Pott and Becker (2011) examined neuropsychological deficits in preschoolers at risk for attention deficit hyperactivity disorder (ADHD). A medium to large effect size was found for inhibition and a small effect size for working memory tasks in children with ADHD symptoms in comparison to typically developing preschoolers. In another meta-analysis of EF in preschool children with externalizing behavior problems (Schoemaker, Mulder, Deković, & Matthys, 2013b), studies of children with aggressive behavior, children who are 'hard to manage', and children with symptoms of the two disruptive behavior disorders (DBD), i.e., oppositional defiant disorder (ODD) and conduct disorder (CD) were also included. When overall EF and inhibition were examined, a medium effect size was found in preschool children with externalizing behavior problems in comparison to typically developing children. Concerning working memory and cognitive flexibility, a small effect size was found. Moreover, there was a stronger relation between EF and externalizing behavior problems in older preschool children (4½-6 years) compared to younger preschool children (3-4½ years).

Although there is thus evidence, from cross-sectional studies, that impairments in EF are associated with externalizing behavior problems in young children, knowledge is limited concerning the stability of this association across time. To examine this association, we build upon a recent study in a clinically diagnosed preschool sample (Schoemaker et al., 2012). In this recent cross-sectional study, we examined EF in a preschool sample aged 3½ to 5½ years with a clinical diagnosis of ADHD, DBD, ADHD+DBD, as well as a typically developing (TD) group. When compared to TD children, preschool children with ADHD, DBD and ADHD+DBD showed inhibition deficits, but no working memory deficits. So far no longitudinal research has been conducted in clinically diagnosed preschool children. Studies in community samples, however, have demonstrated a longitudinal association between preschool inhibition performance and school-age symptoms of ADHD (Berlin, Bohlin, & Rydell, 2003; Brocki, Nyberg, Thorell, & Bohlin, 2007; Von Stauffenberg & Campbell, 2007), but no association with symptoms of DBD (Brocki et al., 2007). Working memory performance did not predict ADHD or DBD symptoms at school-age (Brocki et al., 2007; Kalff et al., 2002). Additionally, Wåhlstedt, Thorell and Bohlin (2008) showed that children with ADHD symptoms and an EF deficit displayed this deficit two years later as well.

The current study, thus, first aimed to investigate the stability of the association between EF and externalizing behavior problems. The questions we aimed to answer were: (a) Is there an association between EF performance and an ADHD and/or DBD diagnosis across time? Based on our cross-sectional study we expected to find inhibition deficits, but no working memory deficits, in clinically diagnosed children. (b) To what degree does EF in the preschool period predict a diagnosis of ADHD or DBD 18 months later (in the late preschool/early school-age period)? Based on the literature we expected that inhibition, but not working memory would predict a later diagnosis of ADHD and DBD.

The second aim of the study was to examine the development of EF in clinically diagnosed preschool children compared to TD children. With regard to the EF development in TD young children, the growth in performance on a general EF factor in children from 4 to 6 years was demonstrated by Hughes et al. (2010). Furthermore, Willoughby, Wirth, and Blair (2012) examined the functional form of change, and showed that the development of one latent EF factor was non-linear, with a larger change being observed between 3 and 4 years compared to between 4 and 5 years. The development of inhibition in particular was examined in two studies with young children at risk for externalizing behavior problems. Dennis, Brotman, Huang, and Gouley (2007) found that inhibition increased between 4 and 5 years and leveled off between 5 and 6 years. Before the age of 4, a linear growth was found, with consistent improvement between the ages of 2 and3, and the ages of 3 and 4 (Moilanen, Shaw, Dishion, Gardner, & Wilson, 2011).

With regard to our second aim, i.e., the developmental change of EF, we aimed to answer the following question: How do EF (inhibition and working memory) develop in preschool children with an early diagnosis of ADHD and/or DBD? We examined differences in development between four different groups of children (ADHD, DBD, ADHD+DBD and TD) and age (early and late preschool period). We divided the sample in two age groups to examine if there were differences in development for younger (between 3½ and 4½ years at the first assessment) and older (between 4½ and 5½ years) preschool children. Based on the literature that shows a faster improvement in the beginning of the preschool period, we expected that both inhibition and working memory develop over time for all groups with a faster improvement for the younger preschoolers compared to the older preschoolers. In sum, this is the first study to examine the stability of the association between EF and externalizing behavior problems and the developmental change of EF in a clinically diagnosed preschool sample.

METHOD

Participants

The 200 children (154 boys and 46 girls) in this study were participating in a longitudinal study conducted at the Outpatient Clinic for Preschool Children with Behavioral Problems, Department of Psychiatry, University Medical Center Utrecht. Children were assessed three times with a 9-month interval, consequently spanning 1½ years. At the first assessment the mean age of the children was 4 years; 7 months (SD = 7 months, range = 3;6 - 5;6). Children were diagnosed with ADHD (N=59), DBD (N=33) and ADHD+DBD (N=50) at the first assessment. We also followed 58 TD children. Between the four groups there was no difference in age (F=2.39, p=.070) or gender distribution at each assessment. *Table 1* shows the descriptive statistics of the four groups for the demographics and control variables. A full-participation rate of 97% was achieved; four children who only participated at the first assessment were excluded for this study. Three parents refused to further participate in the study and one family moved to another country. Four children (three from the DBD group and one from the TD group) participated in two of the three assessments, these children were not excluded.

Children with disorders were referred by general practitioners, well-baby clinics and pediatricians for clinical assessment. Children were diagnosed on the basis of the strict application of the DSM-IV-TR criteria for ADHD and DBD (American Psychiatric Association, 2000) at the first and third assessment. Consensus was reached between a child psychiatrist and a clinical child psychologist using the following data sources: 1) the scores within the clinical range on the Attention Problems scale and the Aggressive Behavior scale of the Child Behavior Checklist completed by parents (CBCL / 1½-5) and the Child Teacher Report Form completed by teachers or day-care caregivers (C-TRF / 1½-5); (both: Achenbach & Rescorla, 2000; Dutch version Verhulst & Van der Ende, 2000); 2) the symptoms reported on the Kiddie Disruptive Behavior Schedule (Keenan et al., 2007), a semi-structured DSM-IV based parent interview for the assessment of ADHD, ODD and CD in preschool children; 3) the scores on the Child Global Assessment Schedule (Shaffer et al., 1983, Dutch translation by Bunte, Schoemaker, & Matthys, 2007), a measure of the impairment of the functioning of the child, filled out by the parents as well as the teacher/caregiver; and 4) the observation of the child's behavior using the Disruptive Behavior Diagnostic Observation Schedule (Wakschlag et al., 2008a, 2008b), a structured observation that evaluates the child's behavior during tasks systematically varying in the level of challenge and support.

The TD group was recruited from regular primary schools and daycare centers. Children with a score in the normal range on the Attention Problems scale and on the Aggressive Behavior scale of the CBCL and C-TRF were included.

An IQ below 70 was defined as exclusion criteria, estimated by the average of the scores on the Raven Coloured Progressive Matrices (Raven, Court, & Raven, 1998) and the Peabody Picture Vocabulary Test-III-NL, (Dunn & Dunn, 2005; Dutch translation by Schichtling) at the first assessment. However, none of the children were estimated with an IQ below 70.

As this study was part of an Outpatient Clinic several children received treatment after the first assessment. Psychopharmacotherapy consisted of methylphenidate in most cases (N=48 after the first assessment; N=66 after the second assessment), but atomoxetine (N=3 after second assessment) and risperidon (N=1 after second assessment) were prescribed as well. If children were prescribed methylphenidate or atomoxetine, we asked parents to stop the medication 48 hours before the subsequent assessments. Psychological interventions consisted of individual parent counseling at the clinic or at home, the Incredible Years parent program (Webster-Stratton, 2001a), or a combination of these interventions. Two treatment variables were used as covariates, Psychopharmacotherapy (i.e., the number of months the child used medication between the first and third assessment) and psychological intervention (i.e., the number of consultations at the clinic or at home and/or number of sessions in the Incredible Years parent training).

Table 1. Means Scores and SD for the Demographics and Control Variables by Group

	TD (1) N=58		ADHD (2) N=59		DBD (3) N=33		ADHD+DBD (4) N=50		ANOVA	Post-hoc
	M	SD	M	SD	M	SD	M	SD	F/χ (3,202)	Bonferroni
Age at study entry (month)	55.98	7.18	55.08	7.50	51.88	8.29	53.92	6.75	2.39	–
% boys	67.2		79.7		81.8		82.0		4.49	–
IQ estimate	112.04	10.36	101.86	11.69	101.89	10.90	100.00	11.68	13.18*	1 > 2,3,4
Psychopharmacotherapy	50.88	2.14	68.10	7.16	64.45	8.17	69.44	6.73	102.31*	1 < 2,3,4 + 3 < 2, 4
Psychological Intervention	50.52	1.31	63.02	10.08	76.76	9.93	75.58	11.86	89.38*	1 < 2,3,4 + 2 < 3,4

** p<.001*
Psychological Intervention: number of psychological intervention sessions; Psychopharmacotherapy: months of medication

Procedure

At each assessment children were evaluated in a single, morning session. At the first assessment, the two measures of intellectual functioning were administered, followed by the EF tasks. All tasks were administered individually by trained master's students in a quiet room with a one-way mirror. One caregiver was in the room with the child and the assessment was recorded. The tasks were administered in a fixed order and lasted about two hours, including breaks. At the first and third assessment the child observation and parent interview were administrated after another break. At each assessment parents received nominal financial compensation for participating and children received two small gifts. Written informed consent from the parents was obtained before participating and the study was approved by the Medical Ethical Review Committee of the University Medical Center Utrecht.

Measures

The EF tasks used in this study were adapted from those used in Wiebe et al. (2011). The computerized tasks are administered through E-Prime version 1.2 (Psychology Software Tools, Pittsburgh, PA). All tasks were preceded by practice trials to make sure the children adequately understood and could perform the tasks. Three tasks were considered to preferentially measure inhibitory skills, i.e., Go-No-Go, Modified Snack Delay, and Shape School – Inhibit Condition and two more to preferentially assess working memory abilities, that is, Nine Boxes and Delayed Alternation.

The ***Go-No-Go task*** is a computerized task where children were instructed to catch as many fish (Go stimuli, 75%) as possible by pressing the button when a fish appeared on the screen. They were instructed to let the shark (No-Go stimuli, 25%) swim by withholding the button press. Auditory feedback was provided when children appropriately caught a fish or inappropriately caught a shark. Stimuli were presented for 1500 ms, with an interstimulus interval of 1000ms. The dependent variable was the d-prime, the z-score of correct Go-trials minus the z-score of correct No-Go trials.

Modified Snack Delay is a newly developed task that integrates the motivational context from the original Snack Delay paradigm (Kochanska, Murray, Jacques, Koenig, & Vandegeest, 1996) with the motor – inhibitory control demands of NEPSY Statue (Korkman, Kirk & Kemp, 1998). Children were instructed to stand still like a snowman with their hands on a mat, without talking or moving. A glass with a treat underneath and a bell was placed in front of the child. The child was instructed to move again and eat the treat when the examiner rang the bell. The task lasted 4 minutes, during which the examiner progressively distracted the child with various activities (e.g., dropping a pencil, knocking under the table) and culminating in leaving the room for 90 seconds. During each 5-second interval, four different behavior categories were rated from DVD by trained raters: moving body, moving hands, talking and 'reward-related behavior' (e.g., eating treat, touching bell). To measure pure motor inhibition only hand movement was used, where no movement was assigned a score of 1, some hand movement .5 and lots of movement 0. Twenty percent was double coded to determine inter-rater reliability (mean inter-rater agreement was 95%, 97.6% and 96.6 % respectively for the first, second and third assessment).The dependent variable was the total score of hand movement (range 0-48).

The ***Shape School- Inhibit Condition*** is a computerized task with cartoon figures in different shapes, colors, and expressions for which the naming rule differs in varying conditions. In the Inhibit condition, participants had to name the color of the figures with happy faces and suppress the prepotent color naming response when the figure had a sad/frustrated face. The dependent variable was the number of correct responses on the inhibit trials (i.e., N = 6 of the total 18 trials).

In ***Delayed Alternation,*** the child had to find a treat underneath one of two identical cups, where after a correct retrieval, the reward alternated to the opposite side in the next trial. There was a 10-second delay between trials, during which the treat was hidden out of the child's sight and the examiner actively distracted the child. A maximum of 16 trials was administrated or if the child made eight consecutive correct responses. The dependent variable was the number of correct retrievals divided by the total trials.

In ***Nine Boxes,*** children were instructed to find all 'Barbapapa' characters hidden in nine different colored boxes (with different shapes on the lid). The child was allowed to open one box per trial, and the boxes were shuffled behind a screen between trials during the 10-second delay. A maximum of 20 trials were administered, until the child found all characters or made five consecutive errors. The dependent variable was the number of correct retrievals divided by the total trials.

Data-analysis

For all analyses, missing data (3.9 %) in the EF measures were imputed, using age, gender, IQ, group assignment at the first assessment and performance on other EF tasks as auxiliary variables. Prior to analyses we computed inhibition and working memory composite scores for each assessment. The inhibition score at each assessment is the mean of the Z–scores across all assessments of the 3 inhibition tasks (i.e., Go-No-Go, Shape school- Inhibit and Modified Snack Delay) and the working memory scores are the mean of the two working memory tasks (i.e., Delayed Alternation and Nine Boxes). This structure was chosen based on the results of the two-factor structure found at the first assessment (Schoemaker et al., 2012).

Our first aim was to determine the stability of the association between EF and diagnosis. We examined this in two different ways. First, we examined if the EF impairments were present across the three assessments using repeated measure analysis. Second, we examined if the EF domains at the first assessment predicted a diagnosis of ADHD or DBD at the third assessment, using logistic regression analysis. The outcome variable was the diagnosis at the third assessment (the dummy variables diagnosis vs. no diagnosis). We performed separate analyses for the diagnosis of ADHD and DBD independently of co-morbidity. The EF scores of the first assessment were used as predictors. In the first step of the analyses we included the control variables, i.e., age of the child at study entry and the treatment variables. In the regression analyses comparing DBD vs. No-DBD group both treatment variables were used (i.e., psychopharmacotherapy and psychological intervention). In the regression analyses comparing ADHD vs. No ADHD only psychological intervention was included; psychopharmacotherapy could not be used as predictor because it was only prescribed in the ADHD group, so it was a characteristic of one of the groups. In the second step, we included the EF variables (inhibition and working memory scores).

Our second aim was to examine how children with a diagnosis develop over time in their inhibition and working memory performance. We performed, for both EF domains, a 3-way repeated measure analysis of covariance, with the inhibition and working memory scores as the within factors and the four (diagnostic) groups (ADHD, DBD, ADHD+DBD, TD) and the two age groups (young and old) as the between-subjects factors. Treatment (psychopharmacotherapy and psychological intervention) was used as a covariate.

It is currently debated whether or not IQ should be included as a covariate in the analyses. On the one hand, including IQ as a covariate ensures that deficits in clinical groups cannot be explained by known group differences in intelligence. On the other hand, both ADHD and DBD typically are associated with mild IQ differences in comparison to individuals without these disorders. Controlling for IQ then may inappropriately remove the overlapping portion of the variance that is associated with the disorder. Because of these contrasting views, all results are reported both with and without controlling for IQ (also see for example Willcutt et al., 2001). Statistical analyses were conducted using SPSS (version 18).

RESULTS

Stability of EF impairments

Table 2 shows the mean and standard deviation of the four groups on the inhibition and working memory composite score at the three assessments. Repeated measure analyses showed that, for inhibition, the main effect of ***diagnosis*** was significant, $F(3,190) = 21.45$, $p < .001$. Post-hoc analyses showed that all three clinical groups performed significantly worse than the TD group. For working memory, there was a main effect for ***diagnosis***, $F(3,190) = 4.27$, $p = .006$, with the TD group outperforming the ADHD and co-morbid group, no difference was found between the TD and DBD group. When IQ was added as a covariate the main effect for inhibition remained significant but the main effect for working memory did not.

Predictive value of EF on diagnosis

At the third assessment 109 children were diagnosed with ADHD, and the non-ADHD group included 88 children. It must be noted that there was a high stability rate of the ADHD diagnosis; 102 (93.6%) children who were diagnosed with ADHD at the first assessment were also diagnosed with ADHD at the third assessment, irrespective of co-morbidity with DBD. The first step of the logistic regression model was significant, $\chi(2) = 11.30$, $p = .004$. The control variables (age and psychological intervention) correctly predicted 70.1 % of the children. The second step, including executive functions, was also significant, χ (2) = 54.29, p <.001. Inhibition was a significant predictor, whereas working memory was not. The total model was significant, χ (4) = 65.60, p <.001, predicting 75.1 % of the children correctly. We performed the analyses with additionally IQ as a covariate; IQ was a significant predictor in the first step of the model, but in the second step the results remained the same, with age, psychological intervention and inhibition as significant predictors.

At the third assessment 60 children were diagnosed with DBD and the non-DBD group included 137 children. The stability rate of a DBD diagnosis was 64,2 % (N=52) irrespective of co-morbidity with ADHD. The first step of the logistic regression model was significant, χ (3) = 26.43, p <.001. The control variables correctly predicted 70.1 % of the children. The second step, including executive functions, was also significant, χ (2) = 6.02, p = .049. Again, only inhibition was a significant predictor. The total model was significant, (5) = 32.45, p <.001, predicting 70.1 % of the children correctly. We performed the analyses with additionally IQ as a covariate; IQ was not a significant predictor. In the second step both the treatment variables remained significant predictors, whereas there was only a marginal trend (p = .068) for inhibition when IQ was included in the model.

Table 2. Mean Scores and SD for the Inhibition and Working Memory Composite Score by Group and Assessment

EF component	Assessment	TD		ADHD		DBD		ADHD+DBD	
		M	SD	M	SD	M	SD	M	SD
Inhibition	1	.39	.48	–.52	.89	–.53	.94	–.78	.91
	2	.51	.40	–.04	.56	.07	.69	–.28	.88
	3	.70	.37	.09	.56	.27	.53	–.13	.77
Working memory	1	.01	.71	–.40	.87	–.37	.82	–.50	.81
	2	.39	.61	.04	.74	.04	.81	–.13	.85
	3	.51	.63	.20	.76	.05	.68	–.05	.75

Table 3. Results of Logistic Regression Analyses Predicting Diagnosis at 3rd Assessment From Executive Functions at 1st Assessment

	ADHD vs. No ADHD			DBD vs. No DBD		
	B	Exp. (B)	CI Exp. (B)	B	Exp. (B)	CI Exp. (B)
Step 1						
Age	– 0.01	1.01	[0.97, 1.05]	– 0.04	0.97	[0.92, 1.01]
Psychopharmacotherapy	–	–	–	0.09***	1.09	[1.04, 1.15]
Psychological Intervention	0.13**	1.14	[1.05, 1.25]	0.11**	1.12	[1.04, 1.21]
Step 2						
Age	0.14***	1.15	[1.08, 1.23]	0.01	1.01	[0.95, 1.07]
Psychopharmacotherapy	–	–	–	0.07*	1.07	[1.02, 1.12]
Psychological Intervention	0.12 *	1.13	[1.02, 1.24]	0.12**	1.12	[1.04, 1.21]
Inhibition	– 1.68***	0.19	[0.10, 0.34]	– 0.53*	0.59	[0.37, 0.94]
Working memory	– 0.38	0.68	[0.42, 1.10]	– 0.11	0.89	[0.56, 1.43]

** p <.05, ** p <.01, *** p <.001*
CI : 95 % confidence interval;
Psychological Intervention: number of psychological intervention sessions; Psychophatmacotherapy: months of medication

Development of EF

Inhibition

Repeated measure analyses showed that for inhibition the main effect for ***assessment*** was significant, F (2, 340) = 53.48, p <.001, indicating an improvement over time. Post-hoc analyses showed that children performed significantly better at each subsequent assessment. The significant main effect of ***age group***, F (1, 190) = 82.95, p < .001, indicated that the older group performed significantly better than the younger group.

There was a significant interaction effect between the ***assessment and diagnosis***, $F(6, 340) = 2.48$, $p = .028$, indicating that the performance over the three assessments differed for the four groups (see *Figure 1a*). Although performance of all four groups increased over assessments this increase was more pronounced for the three clinical groups compared to the TD group, especially between the first and second assessment. There was a significant interaction effect between the ***assessment and age group***, $F(2, 340) = 22.53$, $p < .001$, indicating that increase was more pronounced for the younger group compared to the older group, especially between the first and second assessment (see *Figure 2a*). There was a significant interaction effect between the ***diagnosis and age group***, $F(3, 190) = 3.87$, $p = .010$, indicating that the difference between the younger and older groups was larger for the clinical groups than for the control group. When IQ was added as a covariate, all main effects remained significant and the post-hoc results remained the same. The interaction between assessment and diagnostic groups became non-significant: $F(6, 341) = 1.40$, $p = .218$, indicating that, when the influence of IQ was included in the model the degree of increase in inhibition was similar for control and clinical groups.

Figure 1a. Marginal Means for Inhibition Composite Score by Group and Assessment Adjusted for the Treatment Variables

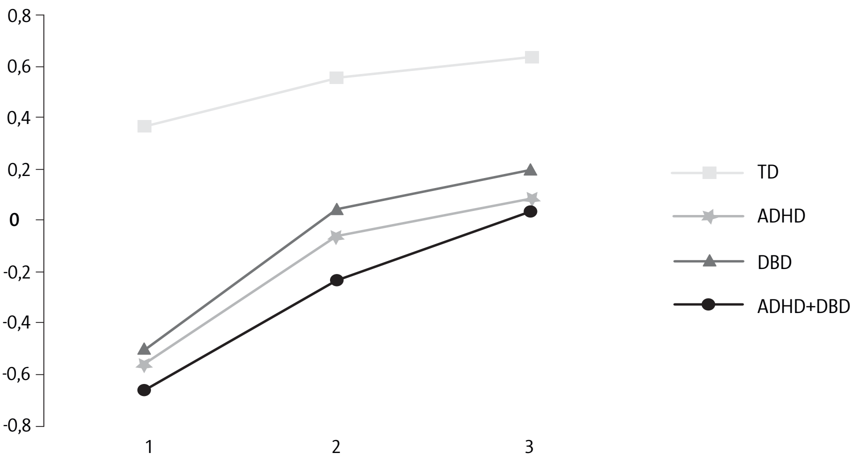


Figure 1b. Marginal Means of the Working Memory Composite Score by Group and Assessment Adjusted for the Treatment Variables

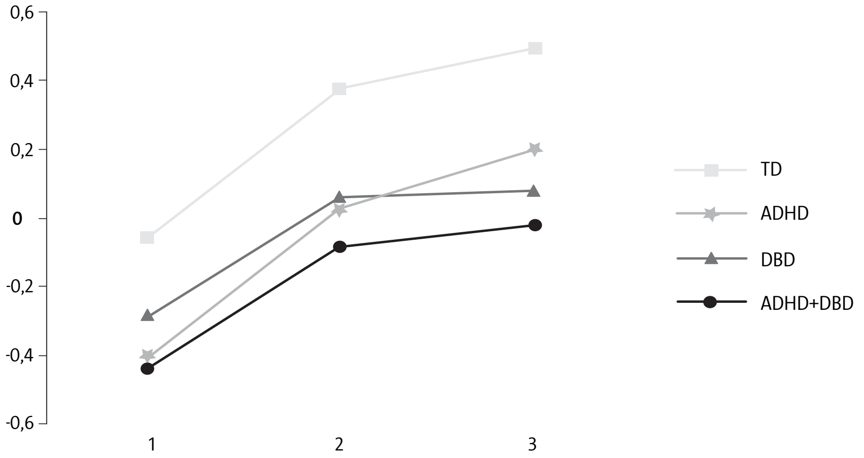


Figure 2a. Marginal Means for the Inhibition Composite Score by age group and Assessment Adjusted for the Treatment Variables

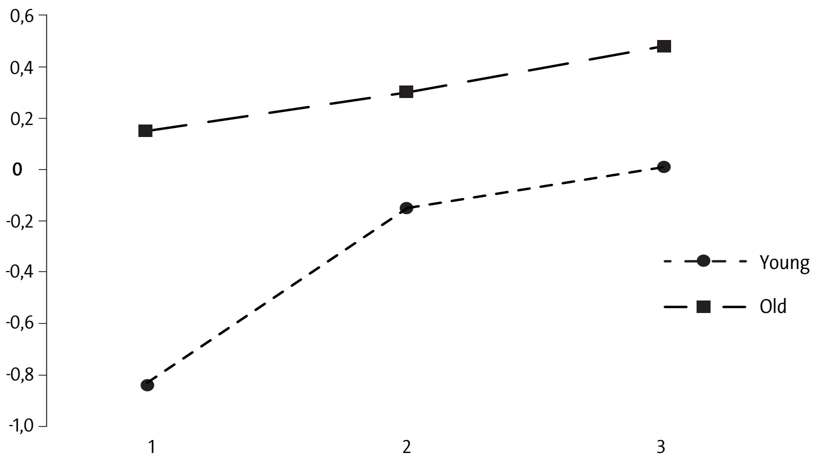
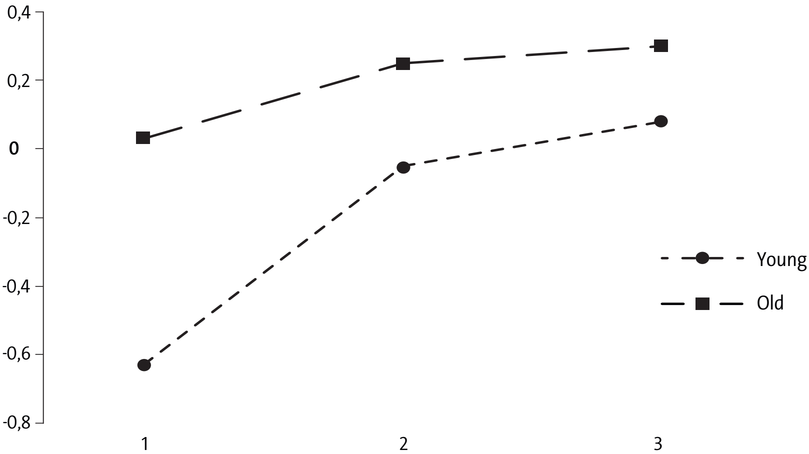


Figure 2b. Marginal Means for the Working Memory Composite score by Age Group and Assessments Adjusted for the Treatment Variables



Working memory

For working memory the main effect for ***assessment*** was significant, F (2, 380) = 14.67, p <.001. Post-hoc analyses showed a better performance at the second and third assessment compared to the first assessment. There was a main effect for ***age group***, F (1, 190) = 23.33, p < .001, the older children outperformed the younger children.

The interaction between ***assessment and age group*** was significant, F (2, 380) = 6.48, p =.002, showing that there is a faster increase in the younger group (*Figure 1b*). The remaining interaction effects were not significant, ***assessment and diagnosis*** F (6, 380) = 0.40, p =.880 (*Figure 2b*), and ***diagnosis and age group*** F (3, 190) = 1.07, p =.363. When IQ was added as a covariate, only the main effect of age group remained significant, F (1, 189) = 19.11, p <.001.

DISCUSSION

The present study aimed to examine, first, the stability of the association between EF impairments and diagnoses of ADHD and DBD in preschool children, and, second, the development of two primary EF functions, inhibition and working memory, in these children. Regarding inhibition we found that across the three assessments the three clinical groups performed worse than the TD group. This result extends the cross-sectional findings of an inhibition deficit in the three clinical groups at the first assessment (Schoemaker et al., 2012). Additionally, inhibition performance at the first assessment was associated with ADHD and DBD diagnoses 18 months later. This result is in line with studies in community samples showing that inhibition performance at preschool-age predict ADHD symptoms in the school-age (Berlin et al., 2003; Brocki et al., 2007; Stauffenberg & Campbell, 2007). The results of the present study show that inhibition was not only a predictor for an ADHD diagnosis, but also for a DBD diagnosis. This relation was not found in a community sample study (Brocki et al., 2007). This might indicate that the relation between inhibition and DBD symptoms is stronger when the behavior problems are more severe. It must be noted that the predictive validity of inhibition is significant, but relatively small.

Regarding the stability of the association between working memory and ADHD, interestingly across the three assessments the ADHD and co-morbid group performed worse than the TD group. This result is in contrast to the findings of the first assessment where we did not find a working memory impairment (Schoemaker et al., 2012). Furthermore, in the present study working memory performance at the first assessment was not associated with a diagnosis of ADHD 18 months later. It might be that working memory impairments in children with ADHD become more salient with age. Other studies in preschoolers did not find working memory impairments (e.g., Sonuga-Barke, 2002), whereas in school-age children and adolescents these impairments become apparent (e.g., Willcutt et al., 2005).

The children with DBD-only did not show working memory impairments across time, and working memory was not longitudinally associated with a DBD diagnosis. These results are in line with results of the cross-sectional study (Schoemaker et al., 2012) and findings of community studies which also failed to find a longitudinal association between working memory and DBD symptoms (Brocki et al., 2007; Kalff et al., 2002). However, it might be that the children with DBD especially show verbal working memory impairments (Moffitt,1993). It must be noted that the EF tasks we administered had a greater spatial than verbal working memory demand.

Regarding the development of EF an improvement with age was demonstrated for inhibition as well as for working memory performance in children with a clinical diagnosis and TD children. These results are similar to studies with TD children (Hughes et al., 2010; Willoughby et al., 2012) and at-risk children (Dennis et al., 2007; Moilanen et al., 2009). Importantly, the improvement of inhibition performance in children with a clinical diagnosis was even more pronounced than the improvement of TD children. Apparently, the clinical diagnosed children seem to catch up a part of their delay. In contrast, there were no differences between groups regarding the rate of development of working memory performance. As described before, overall there were larger group differences in inhibition compared to working memory. As a result, the clinically diagnosed children probably had less chance to catch up with their typically developing peers on their working memory performance.

The younger children showed a faster improvement on inhibition and working memory performance compared to the older children. Together with the result of a faster improvement between the first and second assessment compared to between the second and third assessment in all children, this could be interpreted as a faster improvement at the beginning of the preschool period. The more rapid change between the ages of 3½ and 4½ years in our sample may be associated with maturation and/or the transition to school; in The Netherlands, school-age entry is age 4. During the transition to school significant changes in children's daily experiences appear, including more complex executive control demands (e.g., remembering instructions, inhibiting impulses). These experiences could accelerate the maturation of EF (Hughes et al., 2010). The findings of other studies (Dennis et al., 2007; Willoughby et al., 2012) also suggest a non-linear development of EF. However, the results differ at which age the EF development is slowing down. This might be due to the different age at which children go to school in the USA and the UK. Alternatively, the age differences across studies can be due to the various EF tasks that were administered or a ceiling effect of certain tasks.

In general, the somewhat differential results for inhibition and working memory underline the importance of examining the different EF factors separately at this young age, instead of as one general factor. This is especially important for children with externalizing behavior problems. It might be that discrepancies between EF factors become apparent in children with externalizing behavior problems, but not in TD children (Wiebe et al., 2011; Schoemaker et al., 2012).

It must be noted that a few results changed when IQ was taken into account. Even though a somewhat lower IQ is a characteristic of children with ADHD and DBD, controlling for IQ seems appropriate given the relatively high IQ of the TD group. Results suggest that when taking IQ into account some diagnosis related differences are removed.

Some limitations of the current study are worth mentioning. First, concerning the assessment of EF, it is a challenge to develop EF tasks that are appropriate for the whole age range from 3 to 7 years. For example, cognitive flexibility tasks were administered, but these might be too difficult for the younger children. On the other side, for older preschool children more complex measures are probably necessary to detect subtle impairments (Anderson & Reidy, 2012). Second, the groups are relatively small and the time-span relatively short. If larger groups were included and if the time span between assessments had been larger perhaps more pronounced differences would have been found.

The results of this study indicate an improvement in EF performance in preschool children with an ADHD and/or DBD diagnosis. In spite of this, EF training for these children may be considered since their performance is impaired when compared to TD children. Programs to train EF skills in young children have been developed and have shown to increase EF performance (Diamond & Lee, 2011). Especially children with the initially poorest EF benefit most from these programs. Importantly, EF performance in the preschool years predicts later academic performance (Bull, Espy, & Wiebe, 2008; Clark, Pritchard, & Woodward, 2010). So, improvement in EF may lead to improvement of school readiness and academic achievement (Diamond & Lee, 2011). However, when planning training programs for 3½ to 4½ year old children one should take into account that a fast development of EF skills is characteristic for this age group.

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The Effect of Environmental Characteristics on the Developmental Change in Executive Functions in Preschool Children with ADHD and/or DBD

ABSTRACT

Background: The caregiving environment is known to be associated with executive function performance in typically developing children. However, this relation has not been studied yet in preschool children with attention deficit hyperactivity disorder (ADHD) and disruptive behavior disorder (DBD). The aim of this study was to examine the effect of caregiving environment characteristics on the developmental change in executive function performance in a sample of predominantly clinically diagnosed preschool children. ***Methods:*** Participants were 199 preschool children (mean age 4;6 years at baseline), 58 diagnosed with ADHD, 33 with DBD, 50 with ADHD+DBD and 58 typically developing children. Inhibition and working memory performance were assessed at baseline and at 18 month follow-up, using a battery of tasks especially developed for preschool children. Mothers completed questionnaires on parenting practices and maternal characteristics at baseline. ***Results:*** Maternal stress was associated with a relatively smaller increase in inhibition performance in all children. Physical punishment and harsh and inconsistent discipline mediated the relation between maternal stress and working memory performance in all children. A high level of praise and incentives was associated with a relative larger increase in working memory performance in children with DBD. ***Conclusions:*** Even over a relatively brief period of 18 months caregiving environmental characteristics were associated with young children's developmental changes in executive functions, also in children with ADHD and DBD.

Keywords: caregiving environment, parenting practices, executive functions, ADHD, DBD;
Abbreviations: ADHD: attention deficit hyperactivity disorder; DBD: disruptive behavior disorder

INTRODUCTION

Executive functions (EF) in children have increasingly received attention in research literature. EF can be defined as the top-down control of cognitive processes to achieve a purpose or goal, i.e., the explicit control of thought, emotion and action (Séguin & Zelazo, 2005). Although the development of EF is assumed to be dependent on maturation of the cerebral cortex, a body of research has shown that environmental factors such as parenting control practices affect the development of EF as well. In preschool children positive parental control practices refers to encouraging and guiding the child's behavior, whereas negative control refers to harshness, criticism, and intrusive control (Karreman, Van Tuijl, Van Aken, & Deković, 2006). In the literature different dimensions of positive parental control have been found to be associated with higher EF performance in typically developing (TD) children. Scaffolding (i.e., guiding the child to achieve levels of problem solving that they could not reach on their own) is associated with higher general EF performance longitudinally (Bernier, Carlson, & Whipple, 2010), as well as with growth in general EF performance (Hughes & Ensor, 2009). Furthermore, limit setting and scaffolding have been shown to predict growth in inhibition (Lengua, Honorado, & Bush, 2007). In contrast, parenting practices that involve negative control are expected to suppress the development of children's EF. Results of studies, however, are inconsistent. On the one hand, inconsistent parenting has an adverse effect on the development of EF performance (Hughes & Ensor, 2009). On the other hand, negative affect (i.e., being overcritical towards the child) was not related to inhibition performance six months later (Lengua et al., 2007).

In addition to parenting practices, disadvantageous family factors have been considered. Maternal depression adversely affects children's EF performance over a four year period (Hughes, Roman, Hart, & Ensor, 2013). Besides, maternal symptoms of attention deficit hyperactivity disorder (ADHD) were concurrently associated with inconsistent parenting practices (Mokrova, O'Brien, Calkins, & Keane, 2010). The relation between maternal characteristics and EF may be mediated by parenting practices, but specific studies addressing this issues are lacking. Several studies, however, have examined the mediating role of parenting practices in the relation between other more distal disadvantageous family risk factors and EF. For example, supportive parenting practices have been found to mediate the longitudinal relation between cumulative risk (e.g., poverty, maternal depression, family history of problems) and inhibition (Lengua et al., 2007). Likewise, maternal positive engagement and negative intrusiveness mediated the longitudinal relation between growing up in poverty and EF (Rhoades, Greenberg, Lanza, & Blair, 2011).

The majority of research concerning the relation between the caregiving environment and EF was conducted in community samples with predominantly TD children. It is, however, important to also examine this relation in children with ADHD and disruptive behavior disorder (DBD). Different meta-analyses have shown that children with (symptoms of) ADHD (Pauli-Pot & Becker, 2011; Willcutt et al., 2005, Schoemaker, Mulder, Deković, & Matthys, 2013b) and DBD (Oosterlaan et al., 1998; Schoemaker et al., 2013b) exhibit EF impairments. Parents of these children tend to show high levels of negative controlling parenting practices and higher levels of depression, stress and ADHD symptoms (Capaldi, DeGarmo, Patterson, & Forgatch, 2002; Johnston & Mash, 2001). We should, however, acknowledge the reciprocal relationship between the caregiving environment and the child's (externalizing) behavior (Gross, Shaw, & Moilanen, 2008; Yates, Obradović, & Egeland, 2010).

To our knowledge, only two studies investigated the relation between the caregiving environment and EF in clinical samples. In the first study with a school-age ADHD sample, maternal stress was concurrently negatively related to EF (Joyner, Silver, & Stavinoha, 2009). In the second concurrent study (Schroeder & Kelley, 2009), parenting practices were not related to EF in the ADHD group, whereas positive control (i.e., limit-setting) was related to EF in the TD group. The latter results suggest that a different relation between parenting practices and EF may exist in TD children compared to children with ADHD.

The aim of the current study was to examine the effect of caregiving environmental characteristics on the developmental change in EF in young children with ADHD and/or DBD. In prior studies, we found that preschool children diagnosed with ADHD and/or DBD show EF impairments (Schoemaker et al., 2012), and that these children's EF performance increases over time (Schoemaker et al., 2013a). In the present study we first examined if caregiving environmental factors predicted change in EF performance over an 18 month period. Based on literature with TD children, caregiving environmental factors were expected to be associated with change in EF performance over time in the whole sample. Secondly, we tested if this effect differed for the four groups (ADHD, DBD, ADHD+DBD, TD). We had no specific hypotheses with regard to possible differences among groups. On the one hand, the environmental characteristics may have a stronger effect on the development of EF in the clinical groups relative to the TD group. On the other hand, underlying genetic factors in ADHD and DBD may play a more important role in the maturation of the cerebral cortex in the clinical groups than in the TD group (Shaw et al., 2007). Third, we examined if the effect of maternal characteristics on the developmental change in EF performance was mediated by parenting practices.

METHOD

Participants

The 199 children (153 boys and 46 girls) in this study were participating in a longitudinal study conducted at the Outpatient Clinic for Preschool Children with Behavioral Problems, Department of Psychiatry, University Medical Center Utrecht. Children were assessed three times with a 9-month interval, consequently spanning 1½ years. In this study we used the data from the first assessment (referred to as baseline) and third assessment (referred to as follow-up). At baseline the mean age of the children was 4 years; 6 months (SD = 7 months, range = 3;6 –5;6). Children were diagnosed with ADHD (N =58), DBD (N=33) and ADHD+DBD (N=50) at baseline. We also followed 58 TD children. *Table 1* provides demographic variables by group, groups did not differ in age or gender distribution. A full-participation rate of 97% was achieved.

Children with disorders were referred by general practitioners, well-baby clinics and pediatricians for clinical assessment. Children were diagnosed on the basis of the strict application of the DSM-IV-TR criteria for ADHD and DBD (American Psychiatric Association, 2000) at the baseline assessment. Consensus was reached between a child psychiatrist and a clinical child psychologist using the following data sources: 1) the scores within the clinical range on the Attention Problems scale and the Aggressive Behavior scale of the Child Behavior Checklist completed by parents (CBCL/1,5–5) and the Child Teacher Report Form completed by teachers or day-care caregivers (C-TRF/1,5–5); (both: Achenbach & Rescorla,2000; Dutch version by Verhulst & Van der Ende, 2000); 2) the symptoms reported on the Kiddie Disruptive Behavior Schedule (Bunte, Schoemaker, Hessen, Van der Heijden,

& Matthys, 2013b; Keenan et al., 2007), a semi-structured DSM-IV based parent interview for the assessment of ADHD, ODD and CD in preschool children; 3) the scores on the Child Global Assessment Schedule (Shaffer et al.,1983), a measure of the impairment of the functioning of the child, filled out by the parents as well as the teacher; and 4) the observation of the child's behavior using the Disruptive Behavior Diagnostic Observation Schedule (Bunte et al., 2013a; Wakschlag et al., 2008a, 2008b), a structured observation that evaluates the child's behavior during tasks systematically varying in the level of challenge and support. The TD group was recruited from regular elementary schools and daycare centers. Children with a score in the normal range on the Attention Problems scale and on the Aggressive Behavior scale of the CBCL and C-TRF were included.

An IQ below 70 was defined as an exclusion criteria, estimated by the average of the scores on the Raven Coloured Progressive Matrices (Raven, Court, & Raven, 1998) and the Peabody Picture Vocabulary Test - III- NL, (Dunn & Dunn, Dutch translation by Schichtling, 2005) at baseline assessment. However, none of the children were estimated with an IQ below 70.

Table 1: Means Scores and SD for the Demographics Variables by Group

	TD (1) N=58		ADHD (2) N=58		DBD (3) N=33		ADHD+DBD (4) N=50		ANOVA	Post-hoc
	M	SD	M	SD	M	SD	M	SD	F/χ (3,197)	Bonferroni
Age at baseline (months)	55.98	7.18	55.07	7.56	51.88	8.29	53.92	6.75	2.37	–
% boys	67.2		79.3		81.8		82.0		4.42	–
IQ estimate	112.04	10.36	101.74	11.76	101.89	10.90	100.00	11.68	13.21*	1 > 2,3,4
Psychopharmacotherapy	0.00	0.00	5.12	6.36	1.24	3.55	9.74	7.19	35.91*	4 > 1,2,3; 2 > 1, 3
Psychological Intervention	0.00	0.00	3.19	3.77	3.82	5.42	3.90	5.27	11.39*	1 < 2,3,4

** p<.001*
Psychological Intervention: number of psychological intervention sessions; Psychopharmacotherapy: months of medication

As this study was conducted at an outpatient clinic several children received treatment after the baseline assessment (*Table 1*). Psychological interventions consisted of individual parent counseling at the clinic or at home, the Incredible Years parent program (Webster-Stratton, 2001a), or a combination of these interventions. Psychopharmacotherapy consisted of methylphenidate in most cases (N=48 after baseline assessment; N=66 after the second assessment), but atomoxetine (N=3 after second assessment) and risperidon (N=1 after second assessment) were prescribed as well. If children were prescribed methylphenidate or atomoxetine, we asked parents to stop the medication 48 hours before the follow-up assessment. The questionnaires were completed by the primary caregiver, i.e., biological mothers (N=191), biological fathers (N=3), foster mothers (N=3) or adoptive mothers (N=2). We further refer to all primary caregivers as mothers.

Procedure

At both assessments children were evaluated in a single, morning session. At the baseline assessment, the two measures of intellectual functioning were administered, followed by the EF tasks. All tasks were administered individually by trained master students in a quiet room with a one-way mirror. One caregiver was in the room with the child and the assessment was recorded. The tasks were administered in a fixed order and lasted about two hours, including breaks. At the baseline assessment caregivers completed the questionnaires during the time the children performed the EF tasks. The child observation and parent interview were administrated after another break. At each assessment parents received nominal financial compensation for participating and children received two small gifts. Written informed consent from the parents was obtained before participating and the study was approved by the Medical Ethical Review Committee of the University Medical Center Utrecht.

Measures

Executive Functioning

The EF tasks used in this study were adapted from those used in Wiebe et al. (2011) and were reported in Schoemaker et al. (2012). The computerized tasks are administered through E-Prime version 1.2 (Psychology Software Tools, Pittsburgh, PA). Three tasks were considered to preferentially measure inhibitory skills, i.e., Go-No-Go, Modified Snack Delay, and Shape School-Inhibit Condition and two more to preferentially assess working memory abilities, that is, Nine Boxes and Delayed Alternation.

The ***Go-No-Go task*** is a computerized task where children were instructed to press the button when a fish appeared (Go stimuli, 75%) and withholding the button press when a shark appeared (No-Go stimuli, 25%). Auditory feedback was provided when appropriately catching a fish or inappropriately catching a shark. The dependent variable was the d-prime, the z-score of correct Go-trials minus the z-score of correct No-Go trials.

In ***Modified Snack Delay*** children were instructed to stand still like a snowman with their hands on a mat, without talking or moving. In front of the child a glass with a treat underneath and a bell was placed. The child could move again and eat the treat when the bell was rang, after 4 minutes. The examiner progressively distracted the child (e.g., dropping a pencil, knocking under the table) and culminating in leaving the room. During each 5 second interval, four different behaviors categories were rated by trained raters. To measure pure motor inhibition only hand movement was used, where no movement was assigned a score of 1, some hand movement .5 and lots of movement 0. Twenty percent were double coded to determine inter-rater reliability (i.e.,95%, and 96.6% respectively for baseline and follow-up). The dependent variable was the total score of hand movement (range 0-48).

The ***Shape School-Inhibit Condition*** is a computerized task with cartoon figures with different shapes, colors, and expressions, where the naming rule differs in varying conditions. In the Inhibit condition, participants had to name the color of the figures with happy faces and suppress the prepotent color naming response when the figure had a sad/frustrated face. The dependent variable was the number of correct responses on the inhibit trials (i.e., N=6 of the total 18 trials).

In ***Delayed Alternation***, the child had to find a treat underneath one of two identical cups, where after a correct retrieval, the reward alternated to the opposite side in the next trial. During the 10-second delay between trials, the treat was hidden out of the child's sight and the examiner actively distracted the child. A maximum of 16 trials was administrated or if the child made eight consecutive correct responses. The dependent variable was the number of correct retrievals divided by the by total trials.

In ***Nine Boxes***, children were instructed to find all ‘Barbapapa’ characters hidden in nine different colored boxes. The child was allowed to open one box per trial, and the boxes were shuffled behind a screen between trials during the 10-second delay. A maximum of 20 trials were administered, until the child found all characters or made five consecutive errors. The dependent variable was the number of correct retrievals divided by the total trials.

Caregiving Environment

The ***Parent Practices Interview*** (PPI; Webster-Stratton, 2001b) was designed to measure dimensions of parenting practices of parents of young children. Mothers reported on their response to misbehavior or appropriate behavior on a seven-point scale, ranging from ‘not (likely) at all’ to ‘always/very likely’. We excluded opinions on general statements about parenting (two with regard to the dimension Positive verbal discipline and four with regard to Praise and incentives), because we were interested in the actual behavior of the parents. Drawing on the distinction between positive and negative control parenting practices, we used the dimensions in this questionnaire representing these control practices. To assess positive control we used the dimensions Positive verbal discipline (e.g., discussing the problem with the child, 4 items, α=0.81) and Praise and incentives (e.g., giving a hug or compliment, 10 items, α= 0.64). Four items, originally from Positive verbal discipline scale, that contained the word ‘praise’, we placed into the scale Praise and incentives. To assess negative control we used the scales Harsh and inconsistent discipline (e.g., threatening, but not punishing, 15 items, α=0.83) and Physical punishment (e.g., slapping or hitting when misbehavior occurs, 6 items, α=0.88). Summary scales were computed, consequently each scale ranged from 0 to 7.

The ***Beck Depression Inventory*** (BDI; Beck, Ward, Mendelson, Mock, & Erbaugh, 1961) was designed to assess general depressive symptoms. Mothers reported on 21 items regarding their depressive feelings on a four-point scale. A total score was derived (α=.84).

The ***Parenting Stress Index*** (PSI; Abidin, 1983; Dutch translation de Brock, Vermulst, Gerris, & Abidin, 1992*)* was designed to measure parental stress perception with raising their child (2 to 13 years). To asses parental stress we used four subscales of the parental characteristic domain including restriction imposed by parental role, social isolation, relationship with spouse and parental health. Mothers responded to 26 questions (α=.92) on a six-point scale ranging from ‘totally disagree’ to ‘totally agree’. It yields a total stress score with a possible range from 26 to 156.

The self-report ***ADHD symptoms*** (Kooij & Buitelaar, 1997) was designed to measure symptoms of hyperactivity and inattention during childhood and adulthood. Mothers rated their own ADHD symptoms on a four-point scale ranging from 'never or seldom' to 'very often'. The number of ADHD symptoms in adulthood (α= .92) was used, with a maximum of nine inattention and nine hyperactivity symptoms.

Data-analysis

For all analyses, missing data (3.9%) in the EF measures were imputed, using age, gender, IQ, group assignment at the first assessment and performance on other EF tasks as auxiliary variables. Prior to analyses we computed inhibition and working memory composite scores for each assessment. The inhibition score at each assessment is the mean of the Z–scores across all three assessments of the three inhibition tasks (i.e., Go-No-Go, Shape School-Inhibit and Modified Snack Delay) and the working memory scores are the mean of the two working memory tasks (i.e., Delayed Alternation and Nine Boxes). This structure was chosen based on the results of the two-factor structure found at the first assessment (Schoemaker et al., 2012). We made sure mothers filled out the questionnaires on the day of assessment, consequently there were no missing questionnaires and a low percentage (1.9%) of missing questions.

First, we examined the effect of the caregiving environment on the developmental change in EF in four hierarchical regression analyses, with parenting practices and maternal characteristics as separate predictors, and inhibition and working memory at follow-up as separate dependent variables. We controlled for age in the analyses as age was significantly correlated to EF performance. We did not control for any treatment variables, as treatment was inevitable related to behavior problems (i.e., treatment was only given in the clinical groups). In the first step, age and EF performance (inhibition or working memory respectively) at baseline were entered. In the second step four parenting practices, respectively three maternal characteristics, were entered simultaneously. The caregiving environment variables were centered.

Secondly, we examined if the diagnostic group (ADHD, DBD, ADHD+DBD) was a moderator of the effect of the caregiving environment on the developmental change in EF. To this aim, we added two more steps to the hierarchical regression analyses. In the third step diagnostic groups (i.e., three dummy variables with the TD group as the reference group) were added. In the fourth step the interaction terms (i.e., the dummy variables by caregiving environment) were entered simultaneously. In these models, a statistically significant interaction term supported the moderation hypothesis.

Third, we investigated if parenting practices mediated the effect of maternal characteristics (independent variable) on the developmental change in EF (dependent variable) for the total sample. The following assumptions must be met for mediation (Baron & Kenny, 1986); (a) significant correlations between independent variable, mediator and dependent variable and (b) a significant reduction of the relation between the independent variable and the dependent variable when controlling for the mediator. To test the significance of reduction the 95% confidence interval (CI) was examined (macro of Preacher & Hayes, 2008). If the interval does not include zero, the reduction can be considered significant

RESULTS

Preliminary analyses

The four groups were first compared on parenting practices, maternal characteristics and EF performance (*Table 2*). The differences between the clinical groups and the TD group were in the expected direction, with the exception of Praise and incentives. Contrary to expectations, mothers of the children with ADHD+DBD reported they used more often Praise and incentives compared to the mothers of the TD children. Bivariate correlations between caregiving environment characteristics and EF are displayed in *Table 3*.

Effect of the caregiving environment on the developmental change in EF

Regression analyses were performed to examine whether parenting practices or maternal characteristics predicted change in EF performance. Analyses showed that the parenting practices (*Table 4*) did not explain an additional percentage of the variance either in inhibition performance or in working memory performance. At the level of individual predictors, Praise and incentives was significantly associated with a smaller increase in inhibition and working memory performance. Maternal characteristics (*Table 5*) explained an additional 3% of the variance in inhibition performance. Maternal stress was associated with a smaller increase in inhibition performance. Maternal characteristics were not associated with changes in working memory performance.

Moderation of the groups

Three dummy variables for diagnostic group, added in the third step, contributed significantly, as expected given the results shown in *Table 1*, to changes in inhibition and working memory performance. The fourth step of the regression analyses, containing interaction terms, did not add significantly to the explanation of variation in inhibition performance (ΔR^2=.02, *ns*).

Table 2. Mean scores and SD for Caregiving Environment Characteristics and Executive Functions by Group

	TD (1)		ADHD (2)		DBD (3)		ADHD+DBD (4)		ANOVA	Post-hoc
	M	SD	M	SD	M	SD	M	SD	F/χ (3,197)	Bonferroni
Parenting practices										
Positive verbal discipline	6.04	.91	5.88	1.04	5.73	1.27	5.56	1.19	1.91	–
Praise & incentives	4.10	.67	4.26	.64	4.44	.58	4.51	.64	4.34**	1 < 4
Physical punishment	1.17	.27	1.49	.59	1.90	1.16	1.62	.62	9.55***	1 < 2,3,4; 2<3
Harsh&inconsistent discipline	2.46	.48	2.84	.70	3.06	.80	2.90	.72	7.14***	1 < 2,3,4
Maternal characteristics										
Depressive symptoms	4.29	3.96	8.79	5.43	9.55	5.85	9.66	6.47	11.87***	1 < 2,3,4
Stress symptoms	49.41	15.35	70.17	24.06	73.42	20.34	76.82	21.09	19.64 ***	1 < 2,3,4
ADHD symptoms	1.34	3.03	2.52	3.01	3.00	2.96	2.94	3.89	2.89*	–
Executive functions										
Inhibition (baseline)	.39	.50	–.53	.90	–.53	.95	–.78	.91	22.17***	1 > 2,3,4
Working memory (baseline)	.01	.71	–.41	.87	–.37	.82	–.50	.81	4.51**	1 >2,4
Inhibition (follow-up)	.70	.37	.09	.56	.27	.53	–.13	.77	20.81***	1 >2,3,4; 3 >4
Working memory (follow-up)	.51	.63	.21	.76	.05	.68	–.05	.75	6.20***	1 >3,4

** p<.05,**p<.01,*** p<.001*

The lack of significant interaction effects indicate that the diagnostic groups were not a moderator of the relations between caregiving environment variables and changes in inhibition.

For working memory, however, the fourth step was significant (ΔR^2=.11, p =.011). Four significant interactions were found. First, there was an interaction between ADHD and Positive verbal discipline (ß=.22, p=.039). The significance of the simple slopes were tested and the results showed a marginally significant slope in the ADHD group (ß=.24, p=.054), indicating that a high level of Positive verbal discipline was associated with a larger increase in working memory performance in the ADHD group. Second, there was an interaction

Table 3. Bivariate Correlation between Parenting Practices, Maternal Characteristics and EF (N=199)

	1	2	3	4	5	6	7	8	9	10	11
1 Age											
2 Positive verbal discipline	.06	–									
3 Praise and incentives	–.13	.25***	–								
4 Physical punishment	–.19**	–.17*	.01	–							
5 Harsh and inconsistent discipline	–.01	–.14*	–.04	.47***	–						
6 Depression symptoms	–.07	.01	.09	.31***	.32***	–					
7 Stress symptoms	–.12	–.06	.09	.27***	.34***	.69***	–				
8 ADHD symptoms	–.09	–.03	.12	.21**	.24**	.54***	.36***	–			
9 Inhibition (baseline)	.57***	.15*	–.24**	–.26***	–.10	–.09	–.13	–.07	–		
10 Working memory (baseline)	.48***	.03	–.17*	–.26***	–.23**	–.10	–.21**	–.20**	.45***	–	
11 Inhibition (follow-up)	.34***	.16*	–.23**	–.14*	–.10	–.18**	–.25***	–.10	.60***	.42***	–
12 Working memory (follow-up)	.22**	.08	–.18*	–.20**	–.14*	–.08	–.16*	–.05	.32***	.38***	.38***

** p<.05,**p<.01,*** p<.001*

Table 4. Results of Hierarchical Multiple Regression Analyses Predicting Change in EF Performance from Parenting Practices

	Inhibition			Working memory		
	b(SE)	ß	ΔR²	b(SE)	ß	ΔR²
1. Age	.00 (.01)	.01	.36***	.00 (.01)	.04	.15***
EF baseline	.42 (.05)	.59***		.33 (.07)	.36***	
2. Age	.00 (.01)	.02	.02	.00 (.01)	.02	.03°
EF baseline	.38 (.05)	.55***		.28 (.07)	.32***	
Positive verbal discipline	.07 (.04)	.11°		.06 (.05)	.09	
Praise and incentives	–.13 (.06)	–.13*		–.17 (.08)	–.15*	
Physical punishment	.05 (.06)	.05		–.09 (.08)	–.09	
Harsh and inconsistent disc.	–.06 (.06)	–.06		–.03 (.08)	–.02	
Total F (6,192)			19.42***			7.14***

**p<.10, * p<.05, **p<.01*

Table 5. Results of Hierarchical Multiple Regression Analyses Predicting Change in EF Performance from Maternal Characteristics

	Inhibition			Working memory		
	b(SE)	ß	ΔR²	b(SE)	ß	ΔR²
1. Age	.00 (.01)	.01	.36***	.00 (.01)	.04	.15***
EF baseline	.42 (.05)	.59***		.33 (.07)	.36***	
2. Age	.00 (.01)	–.01	.03*	.00 (.01)	.04	.01
EF baseline	.40 (.05)	.58***		.32 (.07)	.36***	
Depressive symptoms	.00 (.01)	–.02		–.00 (.01)	–.01	
Stress symptoms	–.01 (.00)	–.17*		–.00 (.00)	–.10	
ADHD symptoms	.00 (.01)	.02		.01 (.02)	.06	
Total F (5,193)			24.36***			7.24***

**p<.10, * p<.05, **p<.01*

between DBD and Praise and incentives (ß=.19, p=.020), showing that a high level of Praise and incentives was associated with a larger increase in working memory performance in the DBD group (ß=.40, p=.042). The last two interactions were between Harsh and inconsistent discipline and ADHD (ß=.28, p=.026) as well as DBD (ß=.28, p=.015). There was a significant relation in the TD group (ß=-.29, p=.043), indicating that in TD group a high level of Harsh and inconsistent discipline was associated with a smaller increase in working memory performance. This relationship was not significant in the ADHD or DBD group. Regarding maternal characteristics, no moderation effects were found for either inhibition (ΔR^2=.03, *ns*) or working memory (ΔR^2=.04, *ns*).

Mediation analyses

The bivariate correlations (*Table 3*) showed that there were four possible mediated relations. The two negative parental control dimensions (Physical punishment and Harsh and inconsistent discipline) were a potential mediator in the relation between maternal stress and working memory. Furthermore, physical punishment was a potential mediator in the relation between both maternal stress and depression, on one hand, and inhibition, on the other hand. We performed hierarchical regression analyses, controlling for age and baseline level of EF, to analyze if the contribution of the independent variable reduced when the potential mediator was included. The contribution of maternal stress on working memory performance was reduced significantly (ß=-.08 to ß=-.06; 95% CI -.005 to -.001) when Physical punishment was added to the regression model. Similarly, the contribution of maternal stress on working memory performance reduced significantly (ß=-.08 to ß=-.07; 95% CI -.006 to -.001) when Harsh and inconsistent discipline was added. There was no significant mediation in the two other potential mediated relations.

DISCUSSION

The main aim of the study was to examine the effect of the caregiving environment on the developmental change in EF in young children with ADHD, DBD, ADHD+DBD, and TD over a period of 18 months. In spite of the moderate stability of EF performance and the relatively brief period of 18 months, some caregiving environmental characteristics were associated with changes in EF performance. For the whole sample, and therefore, also for the ADHD and DBD subsamples, maternal stress was associated with a relatively smaller increase in inhibition performance. This finding specifies the Joyner et al. (2009) study that reported a concurrent association between maternal stress and maternal reported EF in an ADHD sample. In the present study, however, maternal stress and EF were assessed independently, and the longitudinal relation was studied in terms of changes in EF performance. Furthermore, for the whole sample negative parenting control practices, i.e., Physical punishment and Harsh and inconsistent discipline, mediated the relation between maternal stress and working memory performance. This finding is in line with studies showing that parenting practices mediate the relation between a combination of less proximate risk factors (e.g., poverty) and child EF (Lengua et al.,2007; Rhoadas et al.,2011). The relevance of negative parenting control practices became also apparent in the association between harsh and inconsistent discipline and a smaller increase in working memory performance, although this was found only in the TD group. In sum, maternal stress was associated with reduced EF development, also in children with ADHD and DBD, and negative parenting control strategies mediated this relation.

Positive parenting control practices also appeared to be relevant. For the whole sample, contrary to expectations a high level of Praise and incentives was associated with a relatively smaller increase in inhibition and working memory performance. In contrast, in children with DBD a high level of Praise and incentives was associated with a relative larger increase in working memory performance. Studies of the effect of praise on noncompliance also are mixed, possibly due to the complexity of praise. For example, without the added reward of positive nonverbal responses, praise alone may not be effective; in general, praise has the potential for certain negative effects (for a review see Owen, Slep,& Heyman, 2012). Further, a high level of Positive verbal discipline, the other positive parenting control practice, in the ADHD subsample was associated with a relatively larger increase in working memory performance, although this association only approached significance. In this context, Schroeder and Kelley (2009) compared correlations and found a significant concurrent relation between positive control and EF in the TD group, but no significant relation in the ADHD group; the magnitude of the correlation, however, was similar, but probably due to the small sample size of the ADHD group the correlation did not reach significance. In sum, although results of positive parenting control practices in the present study are less consistent than those of negative parenting control practices, a high level of Praise and incentives was associated with increased working memory development in the DBD group.

The study has several methodological limitations. First, parenting practices were assessed with self-report questionnaires instead of observations. Second, the relatively small sample size of each group resulted in limited power to detect significant differences between the groups. Third, there is a bidirectional influence between the caregiving environment and the child's externalizing behavior, and possibly also child's cognitive functioning. Therefore, the direction of effects cannot be exactly determined.

In sum, even over a relatively brief period of 18 months caregiving environmental characteristics were associated with young children's developmental changes in EF, also in children with ADHD and DBD. Further research is needed to examine possible differences in developmental change between inhibition and working memory, and between clinical groups. Psychological interventions should target maternal stress and both negative and positive parenting control practices, in order to promote the development of EF in children with ADHD and DBD.

Keypoints

- Maternal stress is associated with a relatively smaller increase in inhibition performance in all children.
- Physical punishment and harsh and inconsistent discipline mediate the relation between maternal stress and working memory performance in all children.
- A high level of praise and incentives is associated with a relative larger increase in working memory performance in children with DBD.

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6

General Discussion

The aims of this dissertation were threefold. We aimed to study EF, the development of EF, and the role of the caregiving environment on the development of EF in preschool children with ADHD and/or DBD. First, we focused on the *assessment* of EF in these groups by (a) using newly developed age-appropriate EF tasks and examining the EF factor structure; (b) conducting a meta-analysis regarding the relation between EF and externalizing behavior problems in preschool children (chapter 2); (c) conducting a cross-sectional study to examine differences in EF performance in preschool children diagnosed with ADHD, DBD and ADHD+DBD (chapter 3). Second, the *development* of EF in these children was examined in a 1½ year longitudinal study (chapter 4). Third, the *role of the caregiving environment* on the development of EF was investigated (chapter 5). In this concluding chapter we address the main findings of the studies and provide a general discussion. Additionally, recommendations for future research and interventions are discussed.

ASSESSMENT OF EF IN PRESCHOOL CHILDREN

EF tasks

As outlined in the introduction, there are several challenges related to the assessment of EF in the preschool period. Therefore, we chose to use EF tasks that were specifically developed for preschool children and worked together with the developers (i.e., Dr. Espy and Dr. Wiebe). We assessed inhibition with the Go-No-Go, Shape School and Modified Snack Delay, and working memory was assessed using Delayed Alternation and Nine Boxes. Despite an extensive collaboration we still experienced some challenges regarding the assessment of EF in young children.

Firstly, the tasks which aimed to measure cognitive flexibility (ED reversal and the switch and inhibit-switch condition of the Shape School task) did not seem appropriate at this young age. In particular, there was no indication of an age effect or an association with the other EF tasks. As a consequence, we were not able to measure cognitive flexibility. It must be noted that when reviewing the literature (chapter 2), it appeared that the number of studies using cognitive flexibility tasks in preschoolers was very limited. Furthermore, it is theorized (Garon, Bryson, & Smith, 2008) that cognitive flexibility is the last EF component to develop. This might indicate that adequate assessment of cognitive flexibility in preschool children is probably not feasible.

Secondly, we were especially interested in inhibition and associated decision making deficits when motivational processes, i.e., reward and punishment, are involved. Studies in older children and adolescents with DBD indeed did show deviances in these functions (Fairchild et al., 2009; Matthys, Van Goozen, Snoek, & Van Engeland, 2004). Therefore, we included a preschool gambling task, i.e., the Coin Game. It appeared, however, that this task was too complex for preschoolers. To study inhibition within a motivational context we used another task, i.e., the Modified Snack Delay. In this task, the amount of movements children display while attempting to refrain from eating a tempting snack is considered a measure of inhibition. However, it is unclear whether the amount of movements is specifically dependent on the reward (i.e., do children move more because they have difficulty inhibiting impulses to eat the snack, or do children move more because they have a general tendency to do so?). Despite this limitation, in our view the Modified Snack delay can be considered a motor inhibition task within a motivational context.

Structure of EF

In this dissertation we found a two-factor structure of EF (**chapter 3**): an inhibition and a working memory factor. Studies using confirmatory factor analysis in typically developing (TD) preschool children found only one unitary EF construct (Hughes, Ensor, Wilson, & Graham, 2010; Wiebe, Espy, & Charak, 2008; Wiebe et al., 2011; Willoughby, Blair, Wirth, & Greenberg, 2010). It might be that a two-factor structure fit the data better in a sample with children with externalizing behavior problems, as it may reflect the underlying pattern of clinical impairments. Moreover, it has been noted that in children assessment of different EF components might be useful as the components operate and develop more independently in comparison to adults when the system is fully developed (Wasserman & Wasserman, 2013). Importantly, the assessment of specific EF components in children allows to study the possible differential relationship with academic school readiness, social skills and behavior problems (Wiebe et al., 2011).

However, we must be careful with definite conclusions about the EF factor structure. Two issues complicate the examination of the EF factor structure. First, the majority of EF tasks measure multiple processes, executive and non-executive. This refers to the task impurity problem (Miyake et al., 2000), in which task are considered to measure one EF component, but also include demands on other EF components or non-executive skills such as processing speed (Anderson & Reidy, 2012; Van der Ven, Kroesbergen, Boom, & Leseman, 2013). Second, the EF factor structure depends on the choice of dependent variables of the EF tasks that are included in the model. The latter is shown in a study by Miller, Giesbrecht, Müller, McInerney and Kerns (2012) who used similar EF tasks as Wiebe et al. (2008) did. First, they used similar dependent variables and found a one-factor model as Wiebe et al., did. Secondly, they choose dependent variables that were more specifically representative of working memory and inhibition (e.g., with the GoNoGo task commission errors instead of hit ratio) and found that a two-factor model fitted the data better than the one-factor model found by Wiebe et al. (2008).

EF in children with externalizing behavior problems: a meta-analysis

We conducted a meta-analysis to examine the status of the research field (**chapter 2**) regarding the relation between EF performance and externalizing behavior problems in preschool children. Our aim was to determine if EF impairments can already be identified in preschool children with externalizing behavior problems. As the number of studies investigating EF performance in preschool children with behavior problems has accumulated over the past decade, the time was right for a meta-analysis to statistically review those studies.

In the meta-analysis 22 studies were reviewed with children from community and referred samples, and children with symptoms of ADHD and/ or DBD. Only four studies, including our study, used a clinically referred sample. A wide range of operationalizations of behavior problems and a diversity of EF measurements were used. A medium effect size was found for the relation between overall EF and externalizing behavior problems. Separate analyses were conducted regarding the three EF factors, with different results. A medium effect size was found for inhibition. This finding is in line with the meta-analysis of Pauli-Pott and Becker (2011) who found a medium effect size for the relation between inhibition and ADHD (symptoms) in preschoolers. The medium effect size for inhibition was similar to meta-analyses of older children (e.g., Willcutt et al., 2005). For working memory and cognitive flexibility a small effect size was found, whereas effect sizes were in the medium range in older children (Morgan & Lilienfeld, 2000; Willcutt et al., 2005. With regard to working memory, it appeared that short term memory tasks were often used in studies of preschool children instead of tasks of the central executive (i.e., the EF component of working memory; Baddeley, 1986), making comparisons between younger and older children difficult. With regard to cognitive flexibility, as mentioned above, adequate assessment of this EF component in preschool children is a challenge.

Moderator analyses for overall EF and inhibition showed that there was a stronger relation for older (4½ - 6 years) preschoolers and for children from referred samples compared to respectively younger (3 - 4½ years) preschoolers and children from community samples. There was also a stronger relation between overall EF and inhibition and externalizing behavior problems in studies with a higher percentage of boys. In the meta-analysis it was not possible to directly compare the results of boys and girls, for studies often do not report separate findings by gender. For future research it would be useful to examine EF separately for boys and girls, as a greater neurobiological vulnerability in boys may explain higher prevalence of externalizing behavior problems in boys compared to girls (Matthys, Vanderschuren, & Schutter, 2013).

Inhibition in clinically diagnosed children

To further investigate the topic of which EF component is impaired in which group of children with externalizing behavior problems (ADHD and/or DBD) while taking into account co-morbidity, we conducted a study with clinical diagnosed preschool children (**chapter 3**). For preschool children with ADHD we found convincing evidence regarding an inhibition deficit; an inhibition impairment held for tasks with or without a motivational demand, after controlling for IQ, and after taking aggressive symptoms into account. In sum, results are in line with those from studies in older children (Willcutt et al., 2005) and show that inhibition impairments in ADHD already manifest in the preschool period.

Preschool children diagnosed with DBD-only showed an inhibition impairment as well, also after controlling for IQ. However, this impairment was less consistent than for preschool children with ADHD. When examining each inhibition task separately, the children with DBD were impaired, but after controlling for IQ the children with DBD were only impaired on the inhibition task with a motivational demand. These results are in line with studies of older children in that results of studies in DBD are less inconsistent when motivational factors are involved (Matthys et al., 2013). Importantly, EF studies in DBD suffer from the assumption that ODD and CD are similar disorders as recent research suggests that ODD is a separate disorder to be distinguished from CD (Matthys et al., 2013). In addition, ODD is a heterogeneous disorder with an irritability (emotional) symptom cluster and a defiant/ headstrong (behavioral) symptom cluster, and therefore different associations between these symptom clusters and EF impairments may be expected (Ezpelata et al., 2012; Matthys et al., 2013).

Children diagnosed with both ADHD+DBD showed impaired inhibition performance, also after controlling for IQ, especially on the inhibition task with a motivational demand. In terms of severity of impairment, the co-morbid group was similar to the ADHD group, whereas in terms of pattern of the inhibition impairment, the co-morbid group was similar to the DBD group. There is an ongoing debate whether the EF profile in the co-morbid group is a combination of ADHD and DBD impairments (Thorell & Wahlstadt, 2006) or whether it characterizes a separate neurocognitive disorder (Luman et al., 2009). The findings of this study do not support the hypothesis of the co-morbid group as a separate disorder.

It is important to note is that in children with ADHD (with or without an additional diagnosis of DBD) when we controlled for DBD symptoms, the inhibition impairment was still apparent. In contrast, in children with DBD (with or without an additional diagnosis of ADHD) when we controlled for ADHD symptoms, the inhibition impairment disappeared. Thus, from a theoretical perspective one may argue that the 'true source' of inhibition deficits in preschool children with DBD may not be the DBD per se. On the other hand, from a clinical perspective impaired inhibition can be considered a characteristic of preschool children with DBD, given the high prevalence of subclinical levels of ADHD symptoms that occur with DBD in this age range.

Working memory in clinically diagnosed children

In the cross-sectional study, children with ADHD and/or DBD did not show overall working memory impairments (**chapter 3**). However, when considering separate working memory tasks, there was an ADHD impairment on the Delayed Alternation task and a DBD impairment on the Nine Boxes task. When we examined a slightly different sample and computed the working memory factor in a different way (i.e., using z-scores across the three assessments), the ADHD and co-morbid group showed impairments compared to the TD group at the baseline assessment (chapter 5). Moreover, across the three assessments, the ADHD and co-morbid group performed worse than the TD group on the overall working memory factor (chapter 4). In sum, results of the present study suggest working memory impairments in preschool children with ADHD, however the evidence for impairment is less consistent compared to studies of older children (Willcutt et al., 2005).

For DBD, there was only an impairment on the Nine Boxes task. It should be noted that the working memory tasks we administered had a greater spatial than verbal working memory demand. This is important as it has been suggested that children with DBD show especially verbal working memory impairments (Moffitt, 1993). In addition, McQuade et al. (2013) showed that impairments in central executive working memory, as opposed to storage working memory, are related to higher levels of aggression. In sum, further study of working memory, and specifically verbal working memory and the central executive, is needed in preschool children with DBD.

DEVELOPMENT OF EF IN PRESCHOOL CHILDREN

The second aim of this dissertation was to investigate the development of EF in preschool children. The preschool period is characterized by rapid changes in multiple areas of development, but children differ in their timing and rate of development. In chapter 2 and 3 we presented cross-sectional evidence for EF impairments in preschool children with ADHD and/or DBD (symptoms). Children with ADHD and/or DBD may show a delay in EF as compared to TD children at one point in time, but may catch up part of their delay across development. In **chapter 4** we examined the development of EF in clinically diagnosed preschoolers.

Regarding the development of EF, an improvement with age was demonstrated for inhibition as well as for working memory performance in children with a clinical diagnosis and TD children. Importantly, the improvement of inhibition performance in children with a clinical diagnosis was even more pronounced than the improvement of TD children. In contrast, there were no difference between groups regarding the rate of development of working memory performance. Furthermore, the younger preschool children showed a faster improvement on inhibition and working memory performance compared to the older children.

Our findings showed that also for children with ADHD and/or DBD their inhibition and working memory performance developed in the preschool period. However, it has been suggested that the age span for which impairments in different components of EF are detectible, differs between the components due to differences in their normative development (Tillman, Brocki, Sørensen, & Lundervold, 2013). For instance, although inhibition, working memory and cognitive flexibility all start developing in the preschool years, inhibition develops most up until age 11, whereas working memory and cognitive flexibility develop more slowly and well into middle adolescence (Garon et al., 2008; Huizinga, Dolan, & Van der Molen, 2006). One may speculate that the longer developmental period for these EF components is due to the more complex demands that are required with planning or decision making when children are getting older. To determine whether children with ADHD and/or DBD show impairments in these functions, it is important to study them in a developmental period where they have matured for (most) TD children. Studying working memory, in addition to cognitive flexibility, in early childhood may underestimate the true impairment when TD children perform more poorly than their full potential due to uncompleted maturation. We therefore support the view that ADHD and DBD are developmental disorders in terms of cognitive (in addition to behavioral) functioning (Tillman et al., 2013).

ROLE OF CAREGIVING ENVIRONMENT

The third aim of this dissertation was to investigate the role of the caregiving environment on the development of EF performance (**chapter 5**). Our study is unique in investigating change in EF performance in clinical groups next to a TD group. From the findings in this study we can first conclude that we did not find a *pattern* in the associations between various factors of the caregiving environment and inhibition or working memory performance. In order to identify such a pattern studies should examine EF during a longer period of time. The results that we did find suggest that both positive and negative caregiving characteristics are associated with changes in EF development. Concerning the positive effect, the use of praise and incentives was related to an increase in working memory performance in children with DBD. This result supports the usefulness to encourage positive parenting practices which is a main goal in behavioral parent training for children with externalizing behavior problems (e.g., Incredible Years; Webster-Stratton, 2001a). Regarding the negative environmental characteristics, maternal stress was associated with a smaller increase in inhibition performance in all children. Moreover, physical punishment and harsh and inconsistent discipline mediated the relation between maternal stress and working memory performance. In behavioral parent training parents learn using positive and consistent strategies instead of harsh and inconsistent strategies to affect their children's behavior, and maybe their EF development.

In sum, even over a relatively brief period of 18 months and despite the moderate stability of EF performance, caregiving environmental characteristics were associated with preschool children's developmental changes in EF, also in children with ADHD and DBD. Even more important, we showed this despite the assumption that the EF development depends on the maturation of the cerebral cortex, and that underlying genetic factors in ADHD (and possibly also in DBD) play an important role in the maturation of the cerebral cortex (Shaw et al., 2007).

METHODOLOGICAL CONSIDERATIONS

STRENGHTS, LIMITATIONS AND FUTURE DIRECTIONS

When we started this study we considered several issues. First, we aimed to capture severe externalizing behavior problems. As the diagnosis of ADHD and DBD can already be made reliable in the preschool years (Keenan et al., 2007), we included clinically diagnosed preschoolers. Second, we aimed to include a variety of EF tasks (presumably assessing different aspects of EF, with or without motivational reward) that have been developed especially for preschool children (Espy et al., 2011; Wiebe et al., 2011). Third, we chose a longitudinal design to examine the fast changing period in the preschool years on the behavior level (clinical diagnosis) as well as the cognitive level (EF). Fourth, we applied a broad view by not only focusing on child characteristics, but also on environmental characteristics. These considerations proved to be effective. Our studies showed differential results for different behavior problems and EF components. These findings encourage to keep studying separate behavior disorders and distinct EF components.

Despite these methodological strengths, there are also some limitations worth mentioning. First, regarding the EF tasks, if we were able to include age-appropriate cognitive flexibility tasks, we could have tested if a three-factor model fitted the data the best, similar to older children or adults. Furthermore, if working memory tasks with a higher verbal demand were included we could have tested if children with DBD were impaired on verbal working memory. Finally, one task in which reward and punishment was included appeared to be too complex, and therefore was not used; as a result, we were limited in studying DBD preschool children's EF functioning in a motivational context. Second, parenting practices were assessed with self-report questionnaires instead of observations. Questionnaires have the potential caveat of social desirable answers and poor self-reflection. However, questionnaires capture parenting practices on a daily basis and on a longer period of time when compared to observations. For example, physical punishment was reported, this would presumably not be observed. Third, the relatively small sample size of each group resulted in limited power to detect significant differences between ages or diagnostic groups. Fourth, the relative short time span made it more difficult to capture developmental changes in EF. In sum, to expand the knowledge regarding EF in the rapid developing preschool period future studies need to include a diverse battery of EF tasks (including cognitive flexibility tasks, verbal working memory tasks and tasks with a strong motivational demand) and study a narrow age range.

OPPORTUNITIES FOR INTERVENTION

Consensus guidelines recommend that psychosocial interventions constitute first-line treatment of ADHD and DBD in preschool children (Gleason et al., 2007). Recently, a meta-analysis was conducted regarding psychosocial treatment such as behavioral parent training in young children with disruptive behavior problems (Comer, Chow, Chan, Cooper-Vince, & Wilson, 2013). Psychosocial treatment in preschoolers has a large and sustained effect;

effects are larger for problems of oppositionality and noncompliance than for problems of impulsivity and hyperactivity. Yet there is an increasing reliance on medication for DBD and ADHD and a decreasing reliance on psychological intervention for young children (Comer et al., 2013). This increasing subscription of medication can be due to the minimal availability of effective psychosocial treatments and the relative convenience to subscribe psychopharmacological treatment. As an alternative for pharmacological treatment and psychosocial interventions there has been increasing efforts to target EF deficits in children and adolescents with ADHD. In a recent review the Cogmed Working Memory Training for elementary school children and adolescents with ADHD was evaluated applying evidence-based treatment criteria. This review shows mixed findings and concludes that the training is a 'possible efficacious treatment' (Chacko et al., 2013). Cogmed Working Memory Training is not advised as a treatment possibility at this moment; further research is necessary before it can be considered a clearly efficacious treatment. To our knowledge, a review on the effectiveness of EF training in preschool children with ADHD (or DBD) symptoms has not been conducted.

When considering EF training in preschool children with ADHD and/or DBD it is important to take into account, first, that improvement of EF performance in these children is fast in the beginning of the preschool years (3-4½ years), as was shown in chapter 4. Second, in chapter 5 it appeared that the caregiving environment is associated with change in EF performance. Therefore, when starting an EF training it seems appropriate to also support the family. As discussed above, behavioral parent training assists parents using appropriate parenting strategies which may affect their children's EF development.

Various EF trainings have been developed that aim improving EF performance or broader achievement levels (Diamond & Lee, 2011; Hughes, 2011). For example, Head Start REDI is an early intervention with demonstrated effects across problem behavior domains and improvement in task orientation (Bierman et al., 2008). It is important to improve EF performance in the preschool years, as such an improvement has consequences for school readiness and later academic performance (Bull, Espy, & Wiebe, 2008; Clark, Pritchard, & Woodward, 2010; Diamond & Lee, 2011).

GENERAL CONCLUSION

The aim of this dissertation was to study EF, the development of EF, and the role of the caregiving environment on the development of EF in clinically diagnosed preschool children with ADHD and/or DBD over a period of 1½ years. A two-factor EF model (inhibition and working memory) was found; this two-factor model may be more eligible in young children with externalizing behavior problems than a one-factor model. Children with ADHD, and to a lesser extent children with DBD, were impaired on inhibition performance. Working memory impairments became more apparent with age in children with ADHD. Inhibition and working memory performance increased over time for children with ADHD and/or DBD, especially in the early preschool period. The caregiving environment seems to be associated with changes in EF performance over time. Overall, we found more similarities than differences in EF performance between children with ADHD and DBD.

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Nederlandse samenvatting

Bij sommige heel drukke kinderen wordt na onderzoek de diagnose ADHD (Aandachtstekort stoornis met hyperactiviteit) gesteld, bij heel driftige en agressieve kinderen de diagnose ODD (Oppositioneel-opstandige stoornis) of CD (gedragsstoornis). ADHD wordt gekenmerkt door een patroon van hyperactiviteit, impulsiviteit en/of aandachtsproblemen in een mate die niet past bij het ontwikkelingsniveau. ODD wordt gedefinieerd als een patroon van negatief, opstandig, ongehoorzaam gedrag en vijandig gedrag naar volwassenen. CD wordt gekarakteriseerd door een repetitief en volhardend patroon van gedrag waarin de basis rechten van anderen of belangrijke leeftijdspassende sociale normen of regels worden overtreden. De psychiatrische stoornissen ODD en CD worden samen ook wel Disruptive Behavior Disorder (DBD) genoemd.

Voornamelijk in de kleuterleeftijd is het ingewikkeld om een onderscheid te maken tussen een psychiatrische stoornis en bij de leeftijd passend ongewenst gedrag. Driftbuien zijn bijvoorbeeld een symptoom van ODD, maar zijn zeker niet ongewoon bij een kleuter. Deskundig onderzoek, waarbij observaties van verschillende personen (leerkracht, ouder, specialist) worden meegenomen, is dan ook een zeer belangrijk om deze diagnosen te stellen. Tijdens het onderzoek dat ten grondslag ligt aan deze dissertatie hebben we de jonge kinderen zorgvuldig gediagnosticeerd en hun ontwikkeling 1½ jaar lang gevolgd.

Bij kinderen vanaf de schoolleeftijd en adolescenten met ADHD en DBD is het al langer bekend dat er bij hen sprake is van neuropsychologische tekorten. Echter, de kennis ten aanzien van kleuters is beperkt. Dit geldt in het bijzonder voor kleuters met een klinische diagnose van ADHD en/of DBD. Het is niet duidelijk of bij kleuters deze tekorten al aanwezig zijn of dat deze zich pas op latere leeftijd manifesteren. Dit komt voornamelijk omdat onderzoek naar executieve functies van kinderen in de kleuterleeftijd ingewikkeld is. Deze dissertatie gaat specifiek over het neuropsychologisch functioneren, of wel de executieve functies, van kleuters met ADHD en/of DBD.

Wat zijn Executieve Functies?

Executieve functies (EF) zijn een verzamelterm voor alle cognitieve vaardigheden die het mogelijk maken om een doel te kunnen bereiken. De vaardigheden zoals werkgeheugen, impulsen beheersen en cognitieve flexibiliteit zijn de voornaamste EF. Deze drie EF hebben wij onderzocht. EF spelen bijvoorbeeld een belangrijke rol bij gedrag, het reguleren van emoties en schoolprestaties.

Bedenken Plannen Uitvoeren Evalueren

De bovenstaande afbeeldingen en onderschriften illustreren de stappen die ondernomen moeten worden om een doel te bereiken. Binnen deze illustraties is het doel het bouwen van een kasteel. Er zijn specifieke vaardigheden, executieve functies, vereist om de stappen te doorlopen en het doel te kunnen bereiken. Ten eerste werkgeheugen, dit is het onthouden van informatie zoals het doel, het uit het geheugen ophalen van mogelijke wegen om het doel te bereiken, het vasthouden van de belemmeringen hierbij, en het ophalen van mogelijke oplossingen hiervoor. Ten tweede, inhibitie, dit is het onderdrukken of vertragen van een reactie. Voor bovenstaande situatie geldt dat het kind waarschijnlijk zijn impuls moet beheersen om de blokjes niet te snel te stapelen. Ten derde, cognitieve flexibiliteit, dit is het leren van een regel of het voor ogen hebben van een doel en vervolgens wisselen naar een andere regel of doel. In bovenstaande situatie; als je maar twee vlaggetjes hebt en je was aan plan een kasteel te bouwen met drie vlaggetjes, moet je iets anders bedenken. Deze drie componenten zijn vanaf de leeftijd van 6 á 7 jaar al redelijk goed ontwikkeld, bij jongere kinderen zijn deze drie componenten in mindere mate ontwikkeld.

Hoe worden EF onderzocht?

Er zijn verschillende uitdagingen bij het onderzoeken van EF bij kleuters. Zo is het bijna onmogelijk om in één taak maar één EF component te meten, vaak zijn meerdere vaardigheden vereist. Bij de taken die cognitieve flexibiliteit meten, heeft een kind ook zijn werkgeheugen nodig om de regel te onthouden. Daarnaast geldt dat de uitingsmogelijkheden van kleuters beperkter zijn dan die van oudere kinderen; bij de ontwikkeling van taken moet daarom rekening worden gehouden met de verbale en motorische mogelijkheden van kleuters. Ook kan de reeks van taken niet te lang zijn, moeten de taken voldoende afwisselend zijn en aansluiten bij de belevingswereld van een kind. Wij hebben ervoor gekozen om taken te gebruiken die ontwikkeld en getoetst zijn door ervaren neuropsychologen (Dr. K. Espy en Dr. S. Wiebe). Vervolgens hebben wij intensief met hen samengewerkt, onder andere door een training te volgen over de afname en interpretatie van de taken. In de zeven taken die wij hebben afgenomen, bestond de afwisseling onder meer in het gebruik van een computer en het toekennen van een beloning. Verder vereisten de taken afwisselend een verbale of motorische reactie van het kind.

Een voorbeeld van een inhibitie taak die wij hebben afgenomen is de Go-No-Go taak. Het kind zit hierbij voor een computer met daarvoor een grote knop waarop het kan drukken. Het kind wordt gevraagd om op de knop te drukken als het een vis ziet op het scherm, zodat het kind de vis kan vangen. Als het kind op de knop drukt verschijnt op het beeldscherm een vis in een netje, vergezeld van een zacht geluid. Als het kind niet op de knop drukt zwemt de vis voorbij. Als het kind een haai ziet op het scherm mag het juist niet op de knop drukken, dan gaat het netje namelijk kapot. Drukt het kind toch op de knop dan verschijnt er op het beeldscherm een gebroken net met een haai erin en een hard geluid. Er zwemmen meer vissen dan haaien in de zee. Dus het kind moeten vaak op de knop drukken, maar deze impuls beheersen als het een haai ziet.

Een werkgeheugen taak die wij hebben afgenomen is de Nine Boxes taak. Hierbij worden de negen leden van de Barbapapa familie in negen doosjes verstopt. Het kind moet alle figuurtjes weer vinden. De doosjes hebben ieder een andere kleur met een andere vorm erop. De doosjes worden na iedere beurt van plek gewisseld buiten het zicht van het kind. Het kind moet dus onthouden welke kleur doosje het al heeft opengemaakt.

Doel van deze dissertatie

Het doel van deze dissertatie was om meer inzicht te verkrijgen in EF bij kleuters met ADHD en DBD. In deze dissertatie stond niet alleen het onderzoek naar EF, maar ook de ontwikkeling van EF en de rol van de omgeving op de ontwikkeling van EF centraal.

Spreekuur voor jonge kinderen met gedragsproblemen

Op het Universitair Medisch Centrum Utrecht (UMC Utrecht) hebben we een spreekuur voor jonge kinderen met gedragsproblemen opgezet bij de afdeling kinder - en jeugdpsychiatrie. Kinderen van 3½ jaar tot 5½ jaar werden verwezen door consultatiebureauartsen, kinderartsen of huisartsen wanneer er vanwege gedragsproblemen een vermoeden bestond van ADHD, ODD of CD. Binnen dit spreekuur zijn ook gunstig ontwikkelende kinderen onderzocht, om deze kinderen te vergelijken met kinderen met een diagnose. Deze kinderen werden geworven via kinderdagverblijven en scholen. Alle kinderen werden onderzocht in één ochtend door een multi-disciplinair team en werden heronderzocht na 9 en 18 maanden.

Het onderzoek bestond zowel uit een psychologisch als psychiatrisch onderzoek. Tijdens het eerste onderzoek bestond het psychologisch onderzoek uit de afname van twee taken om de intelligentie in te schatten en zeven EF taken. Het psychiatrisch onderzoek bestond uit de afname van een gedragsobservatie (Disruptive Behavior Diagnostic Observation Schedule; DB-DOS) en een interview met de ouders gericht op het voorkomen van symptomen (Kiddie Disruptive Behavior Schedule; KDBD). De bruikbaarheid in de praktijk van deze psychiatrische instrumenten wordt in de dissertatie van Tessa Bunte beschreven. Verder vulden de ouders een aantal vragenlijsten in, niet alleen over het gedrag van hun kind, maar ook over hun eigen opvoedingsvaardigheden, symptomen van stress, depressie en ADHD. Voor een gedetailleerde weergave van de onderzoeksochtend zie figuur 1 in hoofdstuk 1 (pagina 23).

De kinderen werden gediagnosticeerd met ADHD, DBD of beide. Kinderen met een andere of geen diagnose zijn niet meegenomen in deze dissertatie. Na drie weken werden de bevindingen en advies voor behandeling besproken met de ouders. De behandeling kon gericht worden op het kind (medicatie), de ouders (ouder training/cursus, thuis begeleiding, ouderbegeleiding in het ziekenhuis) of de school (begeleiding van de leerkracht, organiseren van extra hulp), of een combinatie hiervan.

De tweede onderzoeksochtend, na negen maanden, was korter. Het onderzoek bestond voor het kind uit het uitvoeren van de EF taken en voor de ouders uit het invullen van een aantal vragenlijsten en een ouderinterview. Het derde onderzoeksmoment was gelijk aan het eerste onderzoek, met uitzondering van de intelligentietaken en een aantal vragenlijsten. Over een periode van 2½ jaar werden er in totaal 208 verwezen kinderen onderzocht. Na het eerste onderzoek werden 146 verwezen kinderen (70.2%) gediagnosticeerd met ADHD en/of DBD en geïncludeerd in de studie. Er werden ook 60 zich gunstig ontwikkelende kinderen onderzocht, twee kinderen hadden een klinisch niveau van ADHD of DBD en zijn derhalve niet geïncludeerd in de studie.

ONDERZOEK VAN EF

Bij het eerste doel, onderzoek naar EF, staan de volgende drie vragen centraal:(a) Wat zijn de mogelijkheden om EF te onderzoeken bij jonge kinderen en wat is de EF structuur? (b) Wat is de EF prestatie bij jonge kinderen met ADHD en DBD symptomen? (c) Is er verschil in EF prestaties te meten tussen groepen kinderen met AHD, DBD en ADHD+DBD?

EF taken

Hoewel de meeste taken die we gebruikt hadden geschikt bleken om EF te meten om deze jonge leeftijd, hebben wij toch moeilijkheden ondervonden met sommige EF taken. Zo bleken de taken die cognitieve flexibiliteit beoogden te meten en een inhibitie taak waarin beloning een grote rol speelt, niet geschikt voor de leeftijd van de kinderen van ons onderzoek. Om deze reden zijn deze taken zijn dan ook niet gerapporteerd in deze dissertatie.

EF structuur

De theorieën over de EF structuur (uit welke en hoeveel componenten bestaan EF), kunnen getoetst worden als verschillende taken zijn afgenomen bij dezelfde groep kinderen. In deze dissertatie vonden we een twee factor structuur van EF, een inhibitie en een werkgeheugen factor (**hoofdstuk 3**). De drie taken die beoogden inhibitie te meten vielen onder de inhibitiefactor. De twee taken die beoogden werkgeheugen te meten vielen onder de werkgeheugen factor. Studies met zich gunstig ontwikkelende kinderen vonden over het algemeen één algemene EF factor. Wellicht past een twee factor structuur beter in een steekproef van kinderen met ADHD en DBD, aangezien de twee factoren het onderliggende patroon van de tekorten kan weergeven.

Meta-analyse

Ons doel was om te onderzoeken of EF tekorten ook al gevonden kunnen worden bij kinderen met ADHD en DBD in de kleuterleeftijd. De laatste jaren zijn meer onderzoeken naar EF bij kleuters met (symptomen van) ADHD en DBD gedaan. Wij hebben een meta-analyse uitgevoerd. Dit is een methode om de resultaten van alle eerder uitgevoerde studies statistisch samen te vatten. In **hoofdstuk 2** worden de resultaten van de meta-analyse besproken.

In de meta-analyse zijn 22 studies betrokken die drie tot zes jarige kinderen met symptomen van ADHD en /of DBD hadden onderzocht. Deze studies waren voornamelijk gericht op kinderen met symptomen (maar geen diagnose) van ADHD en/of DBD, afkomstig uit de algemene bevolking (dus niet afkomstig uit de hulpverlening).

De sterkte van de relatie tussen de gedragsproblemen van de onderzochte kinderen en hun algemene EF was matig groot. Er werden aparte analyses uitgevoerd voor de drie EF factoren, met verschillende resultaten. Er werd ook een matig sterke relatie gevonden tussen inhibitie en gedragsproblemen. Er werd een zwakke relatie gevonden voor de relatie van gedragsproblemen met werkgeheugen en met cognitieve flexibiliteit.

Vervolgens hebben we moderator analyses uitgevoerd ten aanzien van algemene EF en inhibitie. Moderator analyses zijn analyses om te onderzoeken of sommige relaties sterker zijn voor bepaalde groepen. De analyses wezen uit dat er een sterkere relatie was tussen gedragsproblemen en algemene EF en inhibitie voor oudere kleuters (4½-6 jaar) in vergelijking met jongere kleuters (3-4½ jaar). Daarnaast bleek dat de relatie sterker was in studies met kinderen die verwezen waren naar specialisten in vergelijking met studies die kinderen uit de algemene populatie hadden onderzocht. Ook was de relatie sterker tussen algemene EF en gedragsproblemen en tussen inhibitie en gedragsproblemen als er binnen een studie een hoger percentage jongens was onderzocht.

Inhibitie bij kleuters met ADHD/DBD

Om specifiek te onderzoeken of kinderen met ADHD en/of DBD een tekort hadden op een bepaalde EF factor, hebben we een studie uitgevoerd met klinisch gediagnosticeerde kleuters (**hoofdstuk 3**). Er is een hoge mate van co-morbiditeit tussen beide stoornissen. De helft van de kinderen met ADHD heeft ook een diagnose van DBD, omgekeerd is het percentage zelfs hoger. Desondanks wordt in onderzoek de rol van co-morbiditeit veelal onvoldoende onderkend. In ons onderzoek hebben we een groep kinderen met alleen ADHD, alleen DBD en een co-morbide groep met ADHD+DBD onderzocht.

Voor kleuters met de diagnose ADHD vonden we overtuigend bewijs van een inhibitie tekort. De inhibitie problemen hielden namelijk stand voor taken met en zonder een beloning en na correctie voor intelligentie en agressieve symptomen.

Kinderen met alleen een DBD diagnose lieten ook een verminderde inhibitiefunctie zien, maar dit tekort was minder consistent dan voor kinderen met alleen een ADHD diagnose. Als we de inhibitie taken apart onderzochten lieten de kinderen met DBD ook een verminderde prestatie zien. Maar nadat gecorrigeerd werd voor intelligentie, vertoonden de kinderen met DBD alleen een tekort op de taken met een beloning.

Kinderen met een diagnose ADHD+DBD lieten ook een inhibitie tekort zien, ook na correctie voor intelligentie, in het bijzonder op taken met een beloning. Qua mate van tekort leek de co-morbide groep meer op de ADHD groep, maar qua patroon van tekorten leek de co-morbide groep meer op de DBD groep.

Werkgeheugen bij kleuters met ADHD/DBD

In de studie besproken in **hoofdstuk 3** vonden we geen verminderde prestatie op de algemene werkgeheugen factor voor kinderen met ADHD en/of DBD. Wanneer we echter de taken apart onderzochten lieten de kinderen met ADHD een verminderde prestatie zien op de Delayed Alternation taak en de kinderen met DBD op de Nine Boxes taak. Als de werkgeheugen factor op een andere manier berekend wordt en de steekproef iets is aangepast, laten de kinderen met ADHD en ADHD+DBD wel een verminderde prestatie zien op de werkgeheugen factor tijdens het eerste onderzoek (hoofdstuk 5). Bovendien laten de kinderen met ADHD en ADHD+DBD een werkgeheugen tekort zien over de gehele periode van het onderzoek (hoofdstuk 4). Concluderend kan gezegd worden dat in deze studie kinderen met ADHD een verminderde werkgeheugen functie laten zien. Deze resultaten zijn echter minder consistent dan in andere studies met oudere kinderen zijn gevonden.

Kinderen met DBD lieten alleen een verminderde prestatie zien op de Nine Boxes taak bij het eerste onderzoek. Er werd verder geen verminderde werkgeheugen prestatie gevonden bij kinderen met DBD. Het moet opgemerkt worden dat de werkgeheugen taken een grotere ruimtelijke dan verbale reactie van kinderen vereiste. Dit is belangrijk omdat is gesuggereerd dat oudere kinderen met DBD voornamelijk tekorten hebben in hun verbale werkgeheugen.

ONTWIKKELING

Het tweede doel van deze dissertatie was om de ontwikkeling van EF bij kleuters met een diagnose van ADHD en/of DBD te onderzoeken (**hoofdstuk 4**). Eerder onderzoek met zich gunstig ontwikkelde kinderen toonde een verbetering in presteren aan op zowel inhibitie als werkgeheugen taken naarmate de kinderen ouder werden. Ons onderzoek heeft aangetoond dat dat zich ook een verbetering voordeed bij kinderen met ADHD en/of DBD. Het was zelfs zo dat de klinisch gediagnosticeerde kinderen zich sneller ontwikkelden op het gebied van inhibitie dan de zich gunstig ontwikkelende kinderen. Ondanks de snellere ontwikkeling presteerden de zich gunstig ontwikkelende kinderen beter dan de klinisch gediagnosticeerde kinderen bij het derde onderzoek. Daarentegen waren er geen groepsverschillen in de snelheid van ontwikkeling op het gebied van werkgeheugen. Verder lieten de jongere kleuters (3-4½ jaar) een snellere ontwikkeling zien in inhibitie en werkgeheugen in vergelijking met oudere kleuters (4½-6 jaar).

ROL VAN DE OMGEVING

Het derde doel van deze dissertatie was om de rol van de opvoedingsomgeving op de ontwikkeling van EF bij kleuters met ADHD en DBD te onderzoeken (**hoofdstuk 5**). We onderzochten in hoeverre opvoedingsvaardigheden en kenmerken van de moeder (stress, depressie en ADHD symptomen) gemeten bij het eerste onderzoek, de ontwikkeling voorspelden van EF (van het eerste onderzoek tot 18 maanden later). De resultaten die we hebben gevonden suggereren dat zowel positieve als negatieve opvoedingskenmerken gerelateerd zijn aan verandering in EF. Wat betreft de positieve effecten, het gebruik van beloningen hing samen met betere prestaties op taken voor werkgeheugen bij kinderen met DBD. Wat betreft de negatieve effecten, stress symptomen van moeder hingen samen met geringere prestaties op taken voor inhibitie bij alle kinderen. Bovendien medieerde fysiek straffen en hard en inconsequent opvoeden de relatie tussen stress en werkgeheugen. Dit betekent dat stress, via deze negatieve opvoedingsvaardigheden, een ongunstige invloed heeft op de ontwikkeling van werkgeheugen. Samenvattend, zelfs over een korte periode van 18 maanden en de stabiliteit van EF prestaties in acht nemend, blijkt dat de opvoedingsomgeving invloed heeft op de ontwikkeling van EF, ook bij kinderen met ADHD en/of DBD. Dit resultaat heeft belangrijke implicaties voor psychologische interventies.

AANBEVELINGEN

In **hoofdstuk 6** hebben we de resultaten samengevat en bediscussieerd tegen de achtergrond van resultaten van andere studies. Daarnaast hebben we sterke punten en beperkingen van het onderzoek besproken. Verder noemen we twee suggesties voor verder onderzoek. Ten eerste, onderzoek naar de (voorspellende) rol van EF in de persistentie van ADHD en DBD (gedurende een periode van drie tot vijf jaar) is belangrijk. Ten tweede, nader onderzoek naar andere EF gebieden, zoals verbaal werkgeheugen en cognitieve flexibiliteit bij kleuters met ADHD en DBD, is aangewezen.

Zijn de resultaten ook relevant voor de behandeling? Psychologische interventies, in het bijzonder de training in opvoedingsvaardigheden, zijn de eerste keuze behandeling voor ADHD en DBD. Daarnaast heeft onderzoek ook het effect van methylfenidaat bij kleuters met ADHD en DBD aangetoond. Mogelijk kan de training van EF een bijdrage leveren aan de behandeling. Er zijn aanwijzingen voor een effect van de training van werkgeheugen bij kinderen in de schoolleeftijd en adolescenten met ADHD. Verder onderzoek naar het effect van training van werkgeheugen en andere EF, zoals inhibitie, met name bij kleuters is aangewezen. Hierbij mag de rol van de directe omgeving, de ouders en de leerkracht, niet worden onderschat.

CONCLUSIE

Door ons onderzoek hebben we getracht de centrale vraagstelling of EF tekorten ook al gevonden kunnen worden bij kinderen met ADHD en DBD in de kleuterleeftijd te kunnen beantwoorden. Concluderend kunnen we zeggen dat kleuters met ADHD, en in iets mindere mate met DBD, al tekorten vertonen op het gebied van inhibitie. Werkgeheugen problemen bij kinderen met ADHD werden duidelijker naarmate de kinderen ouder werden. Inhibitie en werkgeheugen prestaties verbeterden over de periode van 1½ jaar voor kinderen met ADHD en DBD, voornamelijk in het begin van de kleutertijd. De opvoedingssituatie lijkt bij zowel kinderen met ADHD als kinderen met DBD invloed te hebben op de ontwikkeling van EF. Over het algemeen vonden we meer overeenkomsten dan verschillen tussen kinderen met ADHD en DBD.

Dankwoord

Ik ben lekker stout

Ik wil niet meer, ik wil niet meer!
Ik wil geen handjes geven!
Ik wil niet zeggen elke keer:
Jawel mevrouw, jawel meneer...
nee, nooit meer in m'n leven!
Ik hou m'n handen op m'n rug
en ik zeg lekker niks terug!

Ik wil geen vieze havermout,
ik wil geen tandjes poetsen!
Ik wil lekker knoeien met het zout,
ik wil niet aardig zijn, maar stout
en van de leuning roetsen
en schipbreuk spelen in de teil
en ik wil spugen op het zeil!

En heel hard stampen in een plas
en dan m'n tong uitsteken
en morsen op m'n nieuwe jas
en ik wil overmorgen pas
weer met twee woorden spreken!
En ik wil alles wat niet mag,
de hele dag, de hele dag!

En ik wil op de kanapee
met hele vuile schoenen
en ik wil aldoor gillen: nee!
En ik wil met de melkboer mee
en dan het paardje zoenen.
En dat is alles wat ik wil
en als ze kwaad zijn, zeg ik: Bil!

Uit: 'Ik ben lekker stout', Annie M.G. Schmidt, 1955

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Curriculum Vitae

Kim Schoemaker was born on December 14th, 1980 in Alphen aan den Rijn, The Netherlands. In 1999, she graduated from high school at the Groene Hart Lyceum in Alphen aan den Rijn. Before starting her study pedagogical and educational sciences at VU University Amsterdam she travelled for a year with the educational non-profit organization Up With People. During her study she completed her clinical internship at Medisch Kindertehuis Antonius in Bakkum. She completed her research for her Master's thesis on attachment behavior at the University of Western Ontario, Canada. This thesis received an award from the Association of Educationalists (Nederlandse Vereniging van Pedagogen en Onderwijskundigen; NVO). After graduating cum laude in 2005 she combined her work as a researcher with clinical work. As a research assistant at VU University she participated in a study on attachment to foster parents. The clinical work comprised of working with teenage mothers at Altra, a centre of expertise for education, youth and upbringing in Amsterdam. In 2007 she started her PhD project at the department of Child and Adolescent Psychiatry at the University Medical Center in Utrecht. Together with two psychiatrists she set up an outpatient clinic for preschool children with disruptive behavior. She was trained to administrate the executive function tasks (University of Nebraska, Lincoln) and the psychiatric observation (University of Illinois, Chicago) and subsequently trained colleagues at the outpatient clinic. Here she coordinated the assessments and worked as clinical practitioner (orthopedagoog) in a multidisciplinary team. Furthermore, she gained teaching experience as she supervised various clinical internships and Master's theses. From September 2009 she worked at the research group Clinical Child and Family Studies at Utrecht University to finish her dissertation. She aspires to continue combining clinical practice and scientific research.

Publications

MANUSCRIPTS IN THIS DISSERTATION

Schoemaker, K., Bunte, T., Espy, K.A., Deković, M. & Matthys, W. (2013). The effect of environmental characteristics on the developmental change in executive functions in young children with ADHD and/or DBD. *Manuscript in preparation for submission*

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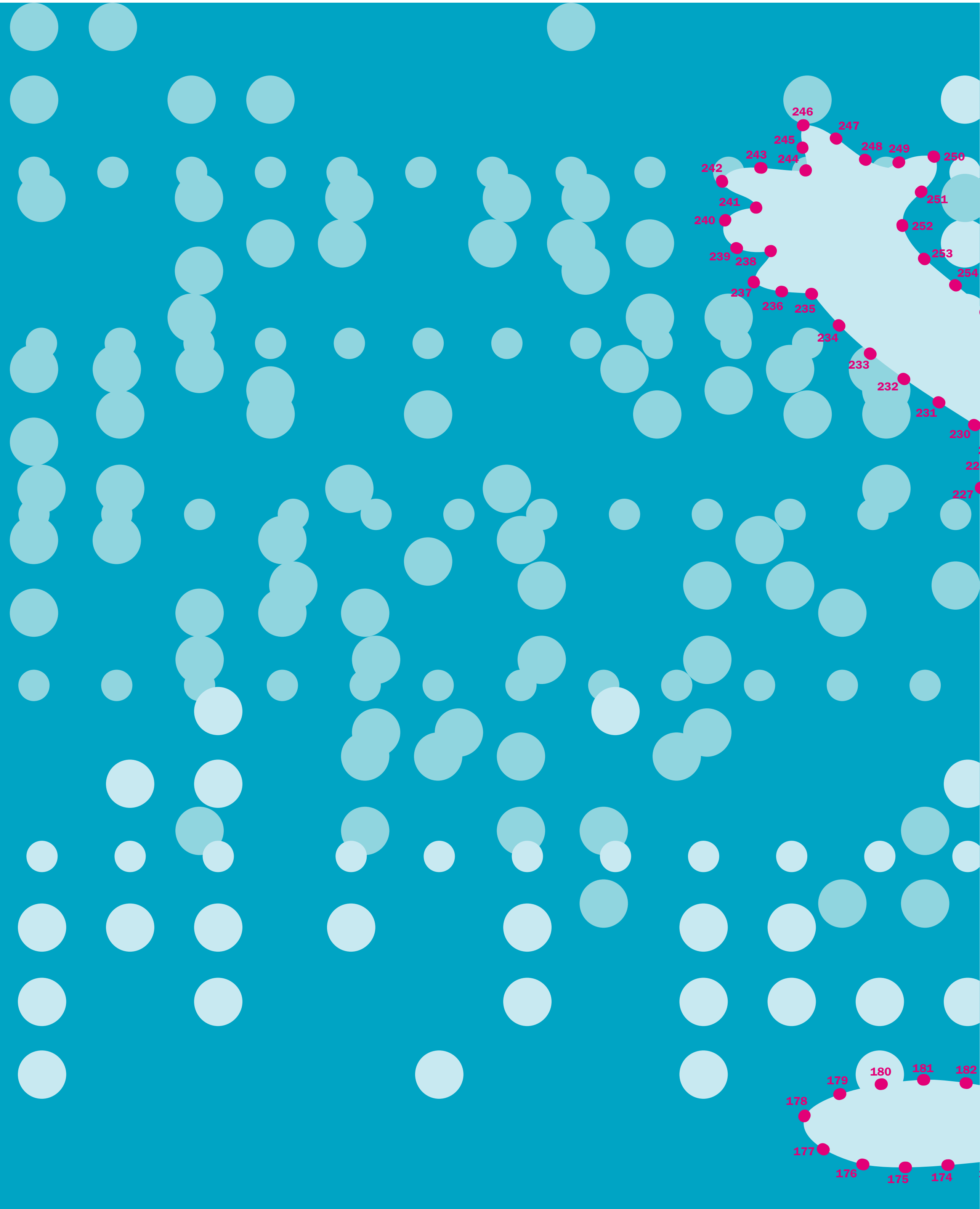

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