

**PHD Dissertation**



**PARENT TRAINING FOR EARLY ADHD**

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## List of papers

- 1) Rimestad, M.L., Lambek, R., Christiansen, H.Z. & Hougaard, E. (2016). Short- and long-term effects of parent training for preschool children with or at-risk of ADHD: a systematic review and meta-analysis. *Journal of Attention Disorders*, e-pub ahead of print, DOI: 10.1177/1087054716648775.  
*Published*
- 2) Rimestad, M.L., Trillingsgaard, T. & Hougaard, E. Combining parent and teacher training for early ADHD: a randomized study of effectiveness. *Pending the decision of the editor in Journal of Child and Family studies following revisions on initial submission*
- 3) Rimestad, M.L., O'Toole, M.S. & Hougaard, E. Mediators of change in a parent training program for early ADHD difficulties: the role of parental strategies, parental self-efficacy and therapeutic alliance. *Under review in Journal of Attention Disorders.*

## **List of abbreviations**

**ADHD:** Attention-Deficit Hyperactivity Disorder

**ECBI:** Eyberg Child Behaviour Inventory

**CD:** Conduct Disorder

**CRS:** Conners Ratings Scale - Index

**ES:** Effect size

**FU:** Follow-Up

**IY:** Incredible Years

**ODD:** Oppositional Defiance Disorder

**PT:** Parent Training

**RCT:** Randomized controlled trial

**SESBI-R:** Sutter-Eyberg Student Behaviour Inventory

**SDQ:** Strengths and Difficulties Questionnaire

**SMD:** Standardized Mean Difference

**TT:** Teacher Training

Childhood ADHD is among the most commonly diagnosed childhood disorder, estimated to be prevalent in 5.29 % of children world-wide (Polanczyk, de Lima, Horta, Biederman & Rhode, 2007). Besides core deficits in attention, hyperactivity, and impulsivity, ADHD is associated with low self-esteem (Mazzone et al., 2013), peer rejection (Bagwell et al., 2001) and increased risk of developing subsequent conduct problems (Beauchaine, Hinshaw & Pang, 2010). Parenting a child with ADHD is challenging, and high levels of conflict and low parental self-efficacy often characterize families of children with ADHD. Parent training (PT) is a treatment originally developed for families with children with conduct problems (Patterson, 2002), and PT has been studied since the 1980's (e.g. Webster-Stratton, 1981) and established as an effective treatment of early behavioral and conduct problems (e.g. Lundahl, Risser & Lovejoy, 2006; Menting, de Castro & Matthys, 2013). Around the turn of the millennium, an expanding number of studies emerged (e.g. Sonuga-Barke et al., 2001; Bor, Sander & Markie-Dadds, 2002; Jones, Daley, Bywater & Eames, 2007), investigating the efficacy of PT for ADHD and reporting promising indication of the efficacy of PT for this population as well. This dissertation contributes to this field of research. We wanted to review and synthesize the present body of studies that investigated ADHD for a preschool population in order to assess the overall efficacy of this relatively new intervention for ADHD (Paper 1). Furthermore, as prior studies had not succeeded in establishing a transfer of efficacy from the parent-child relation to other contexts as daycare or school, we wanted to investigate the relative effectiveness of adding a component of teacher training (TT) to PT (Paper 2). Finally, the field of research on PT for ADHD is relatively new, and little research on active ingredients and mediators of change was available. Thus, we aimed at investigating putative mediators of change in PT as well (Paper 3).

# ADHD

## Diagnosis

Attention-deficit hyperactivity disorder (ADHD) is a neurodevelopmental, pervasive disorder with childhood onset. Two diagnostic systems exist; the American *Diagnostic and Statistical Manual* (DSM) and the formal European diagnostic system by World Health Organization (WHO), ICD-10.

In the DSM 5, ADHD is a neurodevelopmental disorder, implying that the disorder has onset the developmental period and that it produces impairment in personal, social, and academic functioning (American Psychiatric Association, 2013). ADHD is according to diagnostic criteria in the DSM 5<sup>th</sup> Ed. (DSM-5) divided into two dimensions; a dimension of inattention and a dimension of hyperactivity and impulsivity. In order to meet the requirements for a diagnosis of ADHD, a child must display six or more symptoms from the inattention dimension and/or six or more symptoms from the hyperactivity/impulsivity dimension. Furthermore, several of these symptoms should be present before the child is 12 years old, and the symptoms should be cross-situational, which means that they should be present in more contexts, such as home and school. Additionally, the symptoms should be present for minimum six months prior to the assessment. Furthermore, the DSM –5 states that these symptoms should be impairing, which implies that they must hinder normal functioning in social, academic or occupational contexts. Finally, comorbid disorder of schizophrenia and mood disorders are exclusion criteria in the DSM-5. The diagnosis is divided into three different presentations, characterized by predominant type of symptoms. With six or more symptoms from the inattention dimension, the presentation is labeled predominantly inattentive. With six or more symptoms from the hyperactivity/impulsivity dimension, the presentation is labeled predominantly hyperactive /impulsive, and with six or more symptoms from both dimensions of inattention and hyperactivity present, the presentation is labeled combined. The diagnosis is divided into categories of symptom severity, labeled mild, moderate, and severe.

Some aspects of the diagnosis were changed in the transition to DSM-5. Research had indicated that evidence of clear-cut subtypes was questionable and further that the subtypes seemed to change over time (e.g. Willcutt et al., 2012) which led to a change of *subtypes* in DSM-4 to presentations in DSM 5. Criteria for age of onset was changed from 7 to 12 years old, and the specifier of symptom severity was added. Finally, the exclusion criterion of a comorbid diagnosis of autism spectrum disorder from the DSM-4 was omitted in the DSM-5.

In ICD-10, the corresponding diagnosis to ADHD is Hyperkinetic Disorder, and the requirements for the diagnosis are less inclusive than in the DSM. In order to meet diagnostic criteria for Hyperkinetic Disorder, a child must have impairing and age-inappropriate symptoms of both inattention and hyperactivity. Furthermore, these symptoms should be present before the age of 6 and exclusion criteria are comorbid disorders of anxiety, mood disorders and pervasive developmental disorders as autism spectrum disorder (WHO, 1990). Most literature and research in the field applies the DSM version of the diagnosis, and it is the applied diagnosis in this dissertation, both regarding applied literature and in the three studies.

Children with ADHD represent a heterogeneous group, displaying large variation in both symptoms, age of onset, severity and comorbidity (Steinhausen, 2009). Although the categorical aspect is inherent in the concept of diagnosis, a dimensional understanding of the disorder is widely accepted and is consistent with available evidence of the disorder (Roberts, Milich & Barkley, 2014).

### **Prevalence and gender**

Prevalence of ADHD is a widely debated topic (Scioutto & Eisenberg, 2007), with heterogeneous findings across studies (Polanczyk et al., 2007). Two recent meta-analyses have investigated the prevalence of childhood ADHD, Polanczyk and colleagues (2007) and Willcutt (2012). In their 2007 systematic review and meta-analysis, Polanczyk and colleagues identified 102 studies

investigating prevalence of ADHD. When pooling estimates from all studies across countries, the worldwide prevalence of ADHD was estimated to be 5.29%. Results from a multivariate meta-regression model indicated that geographical location was not significantly associated with prevalence rates when controlling for methodological variables. A recent meta-analysis by Willcutt (2012) confirmed this finding. Willcutt (2012) also found that the prevalence appears comparable across assessment from parents, teachers, and clinical diagnosis or self-reports for adolescents and adults.

More boys than girls are diagnosed with ADHD. Estimates vary across studies and sampling methods, however most estimates of gender ratios range from 4:1 to 2:1 (Owens, Cardoos & Hinshaw, 2015). Polanczyk et al. (2007) found that boys seemed to present with more severe symptoms, compared to girls. This finding is, however, contradicted in other studies (e.g. Graetz, Sawyer & Baghurst, 2005). Some have claimed that girls with ADHD are clinically under-recognized, possibly due to divergent clinical presentations or cultural expectancies (McGee & Feehan, 1991), however evidence of clear-cut differences between boys and girls is scarce (Owens et al., 2015). There is a tendency towards heightened risk of internalizing difficulties in girls with ADHD, although this is true for girls in general, compared to boys (Owens et al., 2015).

### **Comorbidities**

ADHD is a highly comorbid disorder, with estimates of up to 80% of children with ADHD having at least one other disorder (Pliszka, 2009). As such, a child with ADHD alone is a more rare case than a child with ADHD combined with another disorder. Oppositional Defiance Disorder (ODD) and conduct disorder (CD) are the two most frequent comorbid disorders, mirrored in estimates of 45-84% of children with ADHD meeting diagnostic criteria for ODD alone or combined with CD. Pliszka (2009) point to extensive evidence of the combination of ADHD and ODD/CD as a severe condition, with more parental psychopathology, more conflicts in the parent child relation and with

higher rates of divorce in the family, compared to ADHD alone. Furthermore, there is indication that the trajectory from ADHD to later substance abuse is mediated by CD (August, Winters, Realmuto, Fahnhorst, Botzet, & Lee, 2006).

Depression and anxiety are also comorbid disorders for childhood ADHD, with estimates of 33% of children with ADHD also meeting criteria for depression and 25-50% for anxiety (Angold, Costello & Erkanli, 1999). These estimates vary across studies, depending on methods and samples. Causal mechanisms of these comorbid disorders are complex, albeit with some evidence that depression and anxiety can arise in children with ADHD as a consequence of difficulties with functioning in peer relations as well as in relation with significant adults (Angold et al., 1999).

Finally, autism spectrum disorder (ASD) is comorbid with ADHD as well. Among children with ASD, estimates of diagnostic criteria for ADHD range from 30-80%, just as children with ADHD have traits of ASD above levels in the general population (Rommelse, Franke, Geurts, Hartman & Buitelaar, 2010). The co-existence of ASD and ADHD has become increasingly acknowledged in the last decade (Rommelse et al., 2010), of which the omitted exclusion criterion of ASD in children with ADHD in the DSM-V bears testimony.

#### **ADHD related difficulties**

In addition to the core deficits inherent in the ADHD diagnosis, children with ADHD generally display further difficulties related to the disorder. As a group, children with ADHD perform worse on standardized tests of intelligence, compared to children with normal development (Frazier, Demaree & Youngstrom, 2004). Academic performance is lowered as well (Barkley, DuPaul & McMurray, 1990) and specific learning disorders are more likely in a sample of children with ADHD compared to children with normal development (Barkley, 2015). Motor development deficit is evident in up to 50% of children with ADHD (Kadesjö & Gillberg, 2003), and sleep disturbance is a common problem in children with ADHD, occurring in up to 70% of children diagnosed with

ADHD (Virring, Lambek, Thomsen, Møller & Jennum, 2016). Furthermore, children with ADHD often suffer from dysfunctional emotion regulation (Shaw, Stringaris, Nigg & Leibenluft, 2014), and have impaired social functioning, both when assessed by parents, teachers and peers (DuPaul, McGoey, Eckert & Van Brakle, 2001).

### **Preschool ADHD**

Although most children with ADHD are diagnosed in the school age around 7 years (Sonuga-Barke et al., 2006), ADHD symptoms will most often emerge before this age (Lahey et al., 1998).

Although diagnosis of preschool ADHD requires close attention to discrimination between developmental changes and ADHD symptoms (Spira & Fischel, 2005), a growing body of evidence point to the validity of preschool ADHD (e.g. Lahey et al, 1998; Lahey et al., 2004) and further that the preschool version of ADHD resembles the school-aged version of ADHD (Lahey et al., 1998).

The pattern of comorbidity and functional impairment in preschool ADHD also seems to mirror comorbidity in the school-age version of ADHD (Wilens et al., 2002). Approximately two thirds of children with developmentally inappropriate levels of ADHD symptoms in preschool will meet symptom criteria for ADHD later in life (Harvey, Youngwirth, Thakar, & Errazuriz, 2009; Lahey et al., 2004).

### **Etiology**

The etiology of ADHD is complex and findings are not conclusive (Thapar, Cooper, Eyre & Langley, 2013). The fact that ADHD is regarded as a neurodevelopmental disorder has established consensus, which is mirrored in the re-allocation of the ADHD diagnosis from disruptive behavior disorders in the DSM-4 to neurodevelopmental disorders in the DSM-5. It seems that the causal mechanisms of ADHD are manifold, with intricate interactions between genetic dispositions and perinatal as well as environmental features. A gene  $\times$  environment framework in understanding the etiology is frequently adopted (Thapar et al., 2013).

### *Heredity*

Heredity of ADHD is well established. In a recent family study with 79 German children diagnosed with ADHD, parental ratings on self-report questionnaires of ADHD indicated that 40% of the children had at least one parent with clinical levels of ADHD (Starck, Grünwald & Schlarb, 2016). Conversely, a study found that a child of parents with ADHD has 57% risk of ADHD (Biederman, Faraone, Mick, Spencer, Wilens, Kiely et al., 1995). Twin studies have shown that heritability explains up to 70-80 % of the variance in ADHD symptoms (Rhee, Waldman, Hay & Levy, 1999), and these estimates of high heritability have prompted a host of research on specific genetic variations causing the disorder, of which results indicate that ADHD is a polygenic condition (Barkley, 2015).

### *Pre-and perinatal factors*

Regarding pre-and perinatal conditions, there is consistent evidence of the association between low birth weight and ADHD difficulties later in life (Mick, Biederman, Faraone, Sayer & Kleinman, 2002). Furthermore, there are links between maternal smoking (Thapar et al., 2003) and alcohol consumption (Mick et al., 2002) as well as high levels of lead and development of ADHD (Nigg et al., 2008), although the latter three appear to be less consistently shown in the literature (e.g. Thapar et al., 2013; Obel et al., 2016).

### *Neuropsychological deficits*

Neuropsychologically, children with ADHD present as a heterogeneous groups as well. Studies have found that children with ADHD appear to have deficits in executive functions, working memory, inhibition, planning and problem solving (Barkley, 2015). However, some studies have found that not all children with ADHD exhibit these difficulties (e.g. Lambek, Tannock, Dalsgaard, Trillingsgaard, Damm & Thomsen, 2011). Furthermore, research have pointed to impaired reward

systems in children with ADHD and thus increased preference of immediate gratification, leading to a hyperresponsiveness to reinforcement (Luman, Oosterlaan & Sergeant, 2005; Sonuga-Barke et al., 2010). Moreover, Kohls, Herpertz-Dahlmann and Konrad (2009) found that children with ADHD were especially responsive to and motivated by social rewards, such as praise and positive facial expressions.

### *Parenting*

As will be described and discussed further in this dissertation, there are associations between child ADHD and parental strategies, stress and self-efficacy, just as there may be more or less beneficial environments for children with ADHD or ADHD difficulties (Johnston & Jassy, 2007). ADHD can challenge parental ability to provide responsive and consistent parenting, which may exacerbate the behavior of the child (Johnston & Mash, 2001). There are some studies pointing to a link between parental negative and harsh reactions to child ADHD symptoms possibly leading to child behavioral problems (e.g. Johnston & Mash, 2001; Johnston & Jassy, 2007). However, there is no substantial evidence of these rearing practices as causal mechanisms of development of ADHD alone (Barkley, 2015; Thapar et al., 2013).

To summarize, converging evidence indicate that genetic and neurological features are causal mechanisms in development of the disorder (Thapar et al., 2013). This biological disposition may affect the appearance of certain neuropsychological dysfunctions, such as inhibitory difficulties and executive dysfunction. These difficulties can then cause child behavior to be disordered by inattention, hyperactivity, and impulsivity. Family characteristic such as parenting are not likely to cause ADHD in itself, but may have implications for the child's behavior and development.

In the next section, characteristics of families with ADHD and relations between parenting variables and child features will be described.

## **Families of children with ADHD**

As described above, ADHD is pervasive, impairing, and characterized by a variety of difficulties for the child. Naturally, having a child in the family with ADHD or ADHD symptoms will affect the family as well. Parents of children with ADHD experience more stress and have more dysfunctional interactions with their children, compared to parents of normally developed children (Sollie, Mørch & Larsson, 2016). Furthermore, up to half of mothers of children with ADHD have experienced clinical levels of depression, and an even larger part display subclinical levels of depressive symptoms (Johnston & Chronis-Tuscano, 2015). Moreover, relationships between children with ADHD and their siblings are also characterized by conflicts and dysfunctional interaction patterns, although the research on this matter is scarce (Mikami & Pfiffner, 2008). Finally, parenting a child with ADHD can cause a great strain on the marital relationship between the parents, and divorce is more frequent in these parents compared to parents of children with normal development (Wymbs, Pelham, Molina, Gnagy, Wilson & Greenhouse, 2008).

Children with ADHD are more demanding of parent attention, often more negative in their interactional style and more non-compliant than children with normal development (Mash & Johnston, 1982). Consequently, it is difficult and challenging to parent a child with ADHD. Studies have found that ADHD symptoms can elicit negative and controlling parental strategies (Johnston and Mash, 2001; Daley, Sonuga-Barke & Thompson, 2003). In turn, this negative and controlling parenting can influence the child in an adverse manner, as this type of parenting constitutes a risk factor in eliciting child defiance and conduct problems (e.g. Patterson, 2002). The combination of child ODD/CD and family problems is severe, as longitudinal studies have shown that this combination predicted more adverse outcomes in adolescents, compared to ADHD symptoms alone (Johnston & Mash, 2001). Furthermore, mothers of children with both ADHD and ODD/CD show

more elevated levels of parenting stress than mothers of children with ADHD alone. Thus, there is an important link between child ADHD, parental response and development of conduct problems.

Parental cognitions are also influenced by child characteristics. Parenting sense of competence, self-efficacy and satisfaction in the parent role are lowered in samples of mothers of children with ADHD (Mash & Johnston, 1983; Johnston and Chronis-Tuscano, 2015). Since parental self-efficacy is associated with maternal warmth and consistent use of discipline (Sanders & Woolley, 2005), and that low sense of parental self-efficacy is associated with negative parenting, these cognitions may have significant influence on parental support in child development and in parent-child relations (Mash et al., 1983).

Seeing that ADHD is a highly hereditary disorder, ADHD in both parent and child is a relatively common combination. High levels of parental ADHD can have consequences for the ability to maintain a consistent, calm and organized every day for the child (Johnston et al., 2015). Seeing that children with ADHD can be difficult to parent, this may be particularly challenging for parents who themselves have difficulties in planning and in regulating emotional responses. Some studies have pointed to a possible advantage of parental ADHD in interaction with child ADHD, the so-called similarity-fit hypothesis, allegedly due to an increased tolerance and understanding of child difficulties (Psychogiou, Daley, Thomson & Sonuga-Barke, 2007; Psychogiou, Daley, Thomson & Sonuga-Barke, 2008). However, other studies have failed to confirm this hypothesis (e.g. Johnston and Lee Flynn, 2011).

In summary, evidence from prior research has established that families of children with ADHD are challenged in multiple areas. Specifically, parent-child interactions appear important in development of subsequent conduct problems. However, most research in the area is correlational in methodology (Johnston & Mash, 2001). Seeing that mechanisms in exacerbation of ADHD symptoms and development of conduct problems between children and parents are most likely

transactional in nature, correlational research does not provide a causal clarification of this relationship.

## **Interventions for ADHD**

In the next section of the dissertation, two of the prevailing interventions, pharmacological treatment and PT, are described.

### **Pharmacological treatment**

Pharmacological treatment of ADHD can be either stimulant medication (medication improving dopaminergic and noradrenergic transmission in the central nervous system, i.e. methylphenidate or amphetamine) or nonstimulant medication (most often medication blocking norepinephrine reuptake, as atomoxetine) (Connor, 2015). Stimulant medication is the most frequent pharmacological treatment as well as the most thoroughly researched medicament for ADHD (Connor, 2015). The efficacy is explored in several reviews and meta-analyses (e.g. Banaschewski et al., 2006; Storebø et al., 2015) with the Cochrane review and meta-analysis from Storebø and colleagues (2015) most recently published. The extensive meta-analysis systematically identified between-group studies and crossover studies investigating the efficacy of methylphenidate for children and adolescents with ADHD (mean age 9.7 years). In addition to extraction of relevant efficacy outcome, studies were coded on reports of adverse effects and methodological quality. In total, 185 randomized studies were included. On teacher rating of child ADHD, there was a standardized mean difference (*SMD*) of -0.77 between treatment and comparison, favoring methylphenidate. Moreover, there was an overall efficacy of methylphenidate on parent-rated quality of life corresponding to a *SMD* of 0.61, and an overall efficacy on teacher –reported general behavior of corresponding to a *SMD* of -0.87. However, Storebø and colleagues (2015) also found that the methodological quality of the studies was generally low, and that there was considerable risk of bias in the form of possible vested interests from pharmacological companies. Furthermore,

methylphenidate was associated with increased risk of non-serious adverse effects as sleep problems and loss of appetite, and serious adverse effects could not be estimated due to too few studies reporting this (Storebø et al., 2016a).

Stimulant medication is the recommended treatment for school-aged children and adolescents with moderate to severe impairment (NICE Guidelines, 2008/2013). However, the efficacy of stimulant medication is less consistent for preschool children (Greenhill et al., 2006). Furthermore, as became increasingly evident with the Storebø et al. (2015) meta-analysis, adverse effects associated with stimulant medication may be more extensive than previously assumed. Furthermore, adverse effects seem to be more frequent in preschool children with ADHD (Greenhill et al., 2006). Stimulant medication is a somewhat controversial topic in research on treatment of ADHD (e.g. Banaschewski, 2016; Storebø, Simonsen & Gluud, 2016b; Mulder, Hazell, Rucklidge & Malhi, 2016) and is not recommended as first-line treatment for preschool ADHD (NICE, 2013).

### **Parent Training**

PT is an intervention where parents are trained to manage their child's difficulties more effectively, aiming at alleviating symptoms in the child. By teaching parents about their child's difficulties and training them in new ways of interacting with their child on a daily basis, the parents are thus included in the treatment as the agent of change (Schaefer & Briesmeister, 1989). Children themselves are usually not participating in PT, but are indirect recipients of the intervention through their parents. As parents learn how to structure everyday life for children in adaptive ways, the aim is to improve child and family functioning and ameliorate child symptoms.

PT is a structured, manualized training course, distributed either in groups (Webster-Stratton, 2001; Barkley, 2013) or individually (Thompson et al., 2009; Thijssen, Vink, Muris, & Ruiter, 2016).

Most frequently applied PT programs are Triple-P Positive Parenting Program (Bor, Sanders and Markie-Dadds, 2002), Parent Management Training-Oregon (PMTO; Thijssen et al., 2016), Defiant

Children (Barkley, 2013), Incredible Years (IY- Webster-Stratton, 2001), Preventing Program for Externalizing Problem Behaviour (PEP – Hanisch et al., 2010), Parent-Child Interaction Therapy (PCIT - Niec, Barnett, Prewett & Shanley Chatham, 2016) and the New Forest Parent Training Program (NFPP; Thompson et al., 2009).

Historically, PT has been developed to treat behavioral and conduct problems (e.g. Patterson, 2002; Webster-Stratton, 1982), and meta-analyses have pointed to efficacy of PT on ODD/CD, both short- and long term (e.g. Lundahl, Risser & Lovejoy, 2006; Menting, de Castro & Matthys, 2013).

Emerging studies have found that PT appears to reduce symptoms of ADHD as well (e.g. Jones, Daley, Hutchings, Bywater & Eames, 2007; Webster-Stratton, Beauchaine & Reid, 2011).

Moreover, specialized programs for ADHD are developed (e.g. New Forest Parent Training Program) with specific focus on targeting the neurodevelopmental difficulties underlying ADHD (Thompson et al., 2009; Sonuga-Barke, Thompson, Abikoff, Klein & Brotman, 2006). Furthermore, some of the more generic programs, originally developed for ODD/CD, have integrated special practices for ADHD in the treatment (Webster-Stratton et al., 2011).

Overall, there are several common factors in PT treatment of early conduct problems and ADHD.

All programs apply some form of behavioral contingency in their therapeutic content, aiming at increasing the frequency of desired child behavior and decreasing the undesired behavior.

Furthermore, parents are taught about scaffolding and limit-setting across the programs, and emotion regulation and problem solving is on the training schedule. Modeling of the new beneficial parental strategies is a central part of PT, and is done in video clips and role-plays. Parents either train these new strategies with the child on-the-spot, if the child is included in the training (as in PCIT, where the training is lab-based and the therapist guides the parents from behind a one-way mirror, e.g. Niec et al., 2016) or, more often, the new strategies are trained in role-plays with parents during group sessions and practiced at home in between sessions.

Programs vary with respect to length of training course, within a typical range of 8 sessions (as in the individually distributed New Forest Parenting Program, Thompson et al., 2009) to 20 sessions (e.g. in the group-based Incredible Years Parenting program, Webster-Stratton et al., 2011). A few studies have investigated the PT with a flexible course, where the parents only proceed to new elements when the therapist perceives that the trained parenting skills are acquired (e.g. Matos, Bauermeister & Bernal, 2009).

### **Incredible Years**

Incredible Years (IY) is a PT program developed by Carolyn Webster-Stratton and colleagues at University of Washington in Seattle in the 1970-ies (Webster-Stratton, 2011). The program series include PT, Teachers Classroom Training, Child Training, Baby Program and Toddler Programs as well as programs for Autisms Spectrum Disorder (Webster-Stratton, 2011). The PT treatment is the cornerstone in the IY series, and is based on the BASIC series (Webster-Stratton, 2008) and the ADVANCED series (Webster-Stratton, 2011). BASIC is a 12-session group-based PT course where parents are taught to implement praise, play, social skills, emotion coaching, and limit setting as well as rewards and natural consequences of child behavior. In the ADVANCED series, which can be added to the BASIC course with additional sessions, skills as interpersonal communication, marital wellbeing, parental anger management, and depression are implemented.

In the IY two group leaders conduct groups of parents of 6-7 children. A key point in the IY is that the therapeutic learning in the PT group is collaborative, and that the parents themselves are to be considered experts on their child (Webster-Stratton, 2011). As such, the group leaders do not lecture on topics and act as experts; however they do give short presentations when introducing new skills and strategies. Most learning is based on group discussion, and topics are supported by extensive use of video clips (“vignettes”) modeling effective and non-effective strategies (Webster-Stratton, 1981; 1982). Group leaders facilitate discussions and reflections on the video-vignettes, as well as initiate exercises to learn new strategies and ways of handling the child’s difficulties. Group leaders

aim at eliminating the social stigma related to having a child with ODD/CD, ADHD or both, and facilitate social support across parents and couples in the group (Webster-Stratton, 2011).

Children themselves are not participating in the IY PT, but parents are instructed in training the acquired skills and strategies at home in interaction with the child. IY PT underlines the importance of making it as easy as possible for the parents to participate in the groups. Thus, a special arrangement strongly suggested by Webster-Stratton is to arrange for baby-sitting while the parents receive PT, both for the focus-child as well as siblings. Furthermore, meals should be offered to both parents and children during training, and the PT is arranged to take place in times of the day where parents are able to attend regardless of type of work, i.e. in the afternoon after working hours or during the daytime, for unemployed parents or parents with nighttime jobs (Webster-Stratton, 2001; 2006; 2011).

#### **Theoretical background of IY PT**

IY PT was developed for preventing ODD/CD and targeting behavioral difficulties (Webster-Stratton, 2001). The program has been extended to also include children with ADHD difficulties (Webster-Stratton, Beauchaine & Reid, 2011; Cassøe, Bæk Bomme, Møller & Straarup, 2014). As such, very little theoretical literature on IY PT or PT for ADHD in general, explicitly focuses on how ADHD is targeted in the training courses (Oord & Daley, 2015). Below, a summary of elements in IY PT that theoretically corresponds to treating ADHD with PT are presented.

IY PT is underpinned by tenets of social learning theory (Bandura, 1973), model learning (Bandura, Ross & Ross, 1961), attachment theory (Bowlby, 1988), as well as developmental psychology on the authoritative parent (Baumrind, 1966). In a social learning theoretical framework, behavioral difficulties in children are viewed as functional responses to environments and interactions marked by high levels conflict as well as modeled imitations of parental behavior (Patterson, 2002; Patterson, Reid, & Dishion, 1998). In families of children with conduct problems, patterns of

aggressive behavior resulting in the *Coercive Cycle* are frequent (Patterson, 2002). The Coercive Cycle is a behavioral pattern where the child's negative and aggressive behavior is escalated as a functional response to parental harsh and aggressive reaction. If the child learns that escalating aggression further can lead to desired outcome, e.g. if the parents withdraw from demands or give in to the child's persistent requirements despite an initial "no" from the parent, then a potential pattern of coercion can be established and reinforced with increasing aggression from both child and parent. This pattern can occur with teachers, siblings and peers as well, and is associated with adverse outcomes in adolescents and adulthood, such as deviant developmental trajectories and substance abuse (Patterson, 2002). Thus, PT seek to interrupt the coercive cycle by teaching parents to identify and reinforce the positive, prosocial and collaborative child behaviors as well as negatively reinforce the aggressive and anti-social behaviors (Webster Stratton, 2011). In PT, parents learn to reinforce positive behavior by extensive use of praise, positive affect, and verbal identification (e.g. "*I see that you helped your little brother there. It was a very nice thing to do and you make me proud by doing that*") and rewards in the form of incentive schedules (Webster-Stratton, 2011). Thus, as the coercive cycle is broken and negative interactions are reduced, the positive interactions between parent and child are increased, forming a more collaborative relationship between parent and child. As parental competencies are increased, this is theorized to prevent and reduce child behavioral problems (Patterson, 2002; Webster-Stratton, 1998). Seeing that children with ADHD are often more non-compliant than children with normal development (Mash & Johnston, 1983), and that ADHD in itself constitutes a risk factor of developing ODD and CD (Beauchaine, Hinshaw & Pang, 2010), it is reasonable to target these difficulties in children with ADHD or ADHD difficulties.

This basis of positive interactions and a strengthened relation between parent and child are important for attachment as well. IY PT aims to improve attachment between parent and child by introducing daily play activities largely based on child-centered play, strengthening the relation in

the dyad (Webster-Stratton, 2011). Seeing that children with ADHD may be in risk of insecure attachment to primary caregivers (Storebø, Darling Rasmussen & Simonsen, 2013), this focus in IY could be essential.

Moreover, parental self-efficacy is theorized to be increased as parents are empowered with renewed moral support as well as a series of practical strategies acquired to handle difficult child behavior. As the parents witness that the child improves in wellbeing and behavior, parents may experience mastery of the parenting role, thus increasing perceived competence. This, in turn, will lead to a more motivated parenting style, which again will lead to more consistent, predictable parenting (Bandura, 1993; Webster-Stratton, Reid & Hammond, 2001). Children with conduct problems and especially ADHD may need guiding and scaffolding as well as highly structured surroundings in order to alleviate the impairment from the symptoms, and thus, parental self-efficacy is theorized to be essential.

Furthermore, teaching parent to coach their child is a substantial part of IY PT, particularly in an ADHD sample (Webster-Stratton et al., 2011). Emotion coaching is theoretically aimed at improving child emotion regulation, which is particularly compromised in childhood ADHD (Shaw et al., 2014). Emotion coaching in IY PT can be defined as guiding the child emotionally by verbally describing own actions and emotions as well as emotions and actions of the child. Discussing emotions with the child and guiding the child to stay calm may improve emotion regulation, which is important to function adequately in both peer relations and in social relations generally (Webster-Stratton, Reid & Hammond, 2001; Ellis, Alisic, Reiss, Dishion & Fisher, 2014). Parents are also taught to increase persistence in attention and on tasks. Teaching the child to sustain attention and persist on difficult tasks aim at targeting the core deficits of inattention, impulsivity, and hyperactivity in ADHD. Parents are taught to describe and verbally acknowledge the child's endeavors in sustaining attention and task persistence by repeatedly describing the

internal state of concentration and calm as it occurs ( e.g. “ *you are so calm on your chair, concentrating really nicely, your breath is calm, you are doing great, one more minute*” etc.).

Theoretically, if the efforts of the child to sustain attention and persist on difficult tasks is guided, praised and encouraged by the parent, the child’s motivation to continuously engage in tasks that are challenging may be reinforced and increased (Luman et al, 2005; Sonuga-Barke et al., 2010), correspondingly increasing child self-efficacy and perhaps eventually self-esteem (Webster-Stratton, 2011). As described in the section on etiology pp. 14, children with ADHD are in general more responsive to social rewards. Albeit not an explicit part of theories of IY, the reliance of IY on behavioral reinforcement and social interaction as praise and rewards between parent and child could thus be hypothesized to alter the child motivation and possibly subsequently ameliorate the symptoms of ADHD (Luman et al., 2005).

Finally, psychoeducation on ADHD is presumed to increase understanding of the child’s difficulties, thereby optimizing parental scaffolding (Vygotsky, 1978). Inattention, hyperactivity and impulsivity as well as possible deficits in executive functioning can cause child difficulties in structuring and completing everyday activities. Parentally induced structure and routines, which are central in IY PT, may ensure that everyday activities are predictable and thus may reduce child impairment. Furthermore, if parents are able to mentalize child difficulties, they are more likely to assist the child adequately in managing symptoms and difficulties, and are less likely to put forth unrealistic demands, which could be harmful to child self-esteem and put a strain on the parent-child relation (Ferrin et al., 2014).

In summary, elements from IY PT theoretically could be hypothesized to target ADHD symptoms as well as conduct problems and difficulties related to the two disorders, although literature on this matter is scarce (Oord & Daley, 2015)

## **Empirical background of PT for ADHD**

### **Efficacy**

In the last two decades, there has been an upsurge in studies assessing the efficacy of PT for ADHD. Two recent meta-analyses on the efficacy of PT for ADHD have been published. In 2013, Sonuga-Barke and colleagues synthesized the evidence of non-pharmacological treatments for ADHD in a meta-analysis, discriminating between what they termed the most proximal assessment of outcomes, which they defined as rated by individuals close to the treatment and thus unblinded to treatment condition, and the probably blinded assessment, as reported by raters blinded to treatment condition. The authors summarized the efficacy of non-pharmacological interventions, such as restrictive elimination diets; free fatty acids supplement trials and behavioral interventions. In the category of behavioral interventions for ADHD, 15 randomized controlled trials were included, of which eight interventions were PT alone, six were PT combined with teacher and/or child training and one study was child training alone. The meta-analysis found that, applying most proximal assessment of ADHD, there was significant efficacy corresponding to a small to moderate ES ( $SMD = 0.40$ ). However, when applying probably blinded assessment, the efficacy was negligible and nonsignificant ( $SMD = 0.02$ ), and the authors concluded that there was limited empirical support for the use of behavioral interventions on ADHD symptoms (Sonuga-Barke et al., 2013). In 2014, Daley and colleagues (Daley, Oord, Ferrin, Danckaerts, Doepfner, Cortese & Sonuga-Barke, 2014) published a meta-analysis on behalf of the European ADHD Guidelines group, where the main aim was to investigate the efficacy of behavioral interventions utilizing the same inclusion criteria as Sonuga-Barke et al., (2013). The authors added analyses of the efficacy of PT on several supplementary variables as parenting practices, child academic achievement and social skills, parental self-concept and parental mental health. Daley and colleagues included 32 studies of behavioral intervention with children age 3-18, of which parent training was included in some form in all cases. The ESs of PT on ADHD were in line with the findings from Sonuga-Barke et al. (2013), with no efficacy on blinded measures of ADHD ( $SMD = 0.02$ ) and a slightly smaller ES

( $SMD = 0.35$ ) on proximal assessment of ADHD, compared to the 2013 meta-analysis by Sonuga-Barke and colleagues. Efficacy of most proximal assessment on conduct problems was  $SMD = 0.26$  and  $SMD = 0.31$  on probably blinded assessment. There was a most proximal assessed  $SMD$  of 0.47 on social skills, and 0.28 on academic achievement. There was efficacy of behavioral interventions on positive parenting (most proximal  $SMD = 0.68$ , probably blinded  $SMD = 0.63$ ) and efficacy on negative parenting as well (most proximal  $SMD = 0.57$ , probably blinded 0.43) (Daley et al., 2014). On parental self-concept, most proximal  $SMD$  was 0.37. Efficacy of PT on parental mental health was negligible ( $SMD = 0.09$ ). The authors conclude that there is beneficial efficacy of PT for ADHD related difficulties, despite no apparent efficacy on objective measures on core deficits. The meta-analysis was accompanied by an editorial in same journal issue by Pfiffner (2014), proposing that a singular emphasis on core deficits in ADHD as in the 2013 meta-analysis is too narrow a focus when evaluating the advantages of intervening with behavioral interventions. This argument has also been proposed in a Letter to the Editor as a response to the 2013 meta-analysis (Chronis-Tuscano, Chacko & Barkley). The editorial by Pfiffner (2014) similarly concludes that there is beneficial efficacy of behavioral interventions for ADHD related difficulties. Thus, PT is regarded as an effective treatment for ADHD, although as yet, the documentation of efficacy of PT on blinded measures of ADHD is limited.

Our meta-analysis (paper 1) was conducted in order to quantify the efficacy of PT for the particular period of preschool ADHD (2.5-6 years). As ADHD is a neurodevelopmental disorder, this implies that development is impaired in several domains. Neuroplasticity, defined as the brain's ability to reorganize itself, is more pronounced in the preschool years (Halperin, Bédard, & Curchack-Lichtin, 2012). Thus, it has been proposed that this period may be a critical window of intervening early. Intervening with preschool children could possibly be more efficacious and prevent a more severe development of the disorder, compared to intervening later (Halperin et al., 2012; Sonuga-Barke et al., 2013). Early intervention might also prevent the development of secondary difficulties

to ADHD, as conduct problems and peer problems in school. Furthermore, long-term effects of PT for preschool ADHD had not previously been meta-analyzed. A more detailed description of empirical background for PT for ADHD is available in paper 1.

### **Change processes in parent training for ADHD**

As outlined, empirical findings support at least the parent-rated efficacy of PT on ADHD and behavioral problems, and Daley et al. (2014) found that there appeared to be parentally assessed efficacy on several parenting variables as well. Furthermore, the underlying principle of intervening with PT is that parents are agents of change, bringing about change in child outcomes by altering parental strategies. However, very little research on formal mediation and mechanisms of change has been conducted. According to Kazdin (2007), knowledge on how the interventions work is essential in evidence-based psychotherapy research in order to provide optimal conditions and focus on active ingredients. Traditional approaches to statistical mediation have used either the Baron and Kenny (1986) ‘causal steps’ approach, or the ‘product-of-coefficients’ approach suggested by Sobel (1982) (See Figure 1). Statistical mediation according to the causal steps approach requires 1) that the independent variable (i.e., the intervention) causes change in the proposed mediator (the *a* path in Figure 1); 2) that change in the proposed mediator is associated with change in the dependent variable (i.e. outcome; the *b* path); and 3) a reduction of the association between treatment and outcome (from *c* to *c'*) after controlling for the contribution of the proposed mediator (*b*). The product-of-coefficients approach investigates the indirect mediational path as indicated by the interaction of path *a* and path *b* ( $a \times b$ ); theoretically corresponding to the *c* minus *c'* pathway in the causal steps approach (Preacher & Hayes, 2008). However, none of these approaches to statistical mediation takes account of the timeline of change in variables that is necessary for conclusions as to the direction of causality. Kazdin (2007) recommends time-lagged analyses of repeated measurements of both process and outcome as an optimal approach to intervention research on mediation.

**Figure 1.** The Mediation Model

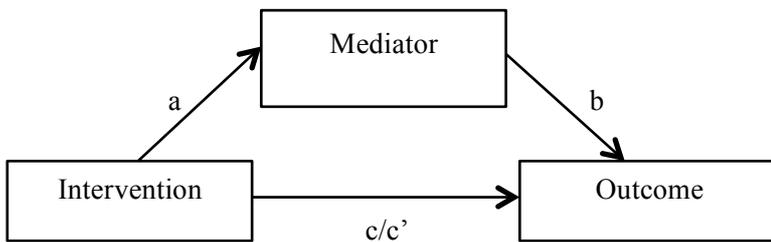


Figure 1 The Mediation model

Forehand, Lafko, Parent, and Burt (2014) reviewed the evidence of parenting as a mediator of PT on child outcomes in 25 studies investigating PT for externalizing difficulties. Generally, few studies had investigated parenting as mediator in treatment or prevention studies. The authors concluded that regarding conduct problems, composite measures of parenting and discipline received support as mediating variables. No conclusions could be drawn for ADHD, since no included studies had formally tested parenting as mediator in PT for ADHD. A more detailed background of studies investigating change processes in PT for ADHD is presented in Paper 3.

## **The Ph.D. project**

### **Research questions**

In this project, we wanted to investigate PT for early ADHD. The main purpose was three-fold.

First, 1) in a systematic review and meta-analysis, we wanted to assess the overall short-and long-term efficacy of PT for preschool ADHD on reductions in ADHD, ODD/CD and negative parenting, both on parental ratings and blinded assessments. Second, in a randomized-controlled trial, we wanted to explore 2) if IY PT in a Danish setting combined with Teachers Training (TT) based on the same principles as the IY PT was more effective in reducing ADHD and ODD/CD, both at home and in kindergarten/school, compared to IY PT alone. Finally, 3) we wanted to

explore the mediating mechanisms in the effectiveness study of reduced negative parenting, increased positive parenting and a more appropriate disciplinary style as well as parental self-efficacy and working alliance in PT on the changes in parentally assessed symptoms of ADHD and ODD/CD. These research questions are reflected in the three papers included in this thesis.

## **Methods**

### **Study setting, data collection procedures, and sample characteristics for paper 2 and 3.**

The study setting for data-collection in paper 2 and 3 was Center for ADHD, an independent non-profit clinic funded at the time of the study by Edith and Godtfred Kirk Christiansen Foundation. The Centre is situated in the City of Aarhus, Denmark. The Centre was established in 2010, a time when the wait-lists of assessment in the child psychiatry sector were lengthy and treatment resources were limited. The aim of the Center is to provide easy access to preventive treatment for families of children with or at risk of ADHD, regardless of child diagnostic status. A formal ADHD diagnosis is not an inclusion criterion to receive treatment at the Clinic. Parents self-refer to the Centre with concerns of child hyperactivity, impulsivity, inattention, and behavioral difficulties, and a psychologist screens parents for inclusion by telephone. If assessed as eligible for inclusion, parents fill out three validated questionnaires. Parents are visited at home by two psychologists from the Centre, who conduct a semi-structured clinical interview with the parents. Finally, questionnaires and clinical interview are reviewed at a following treatment conference at the Centre, and it is decided if parents should be offered treatment in the form of IY PT at the Centre.

For inclusion in the RCT in this study, we followed the clinic inclusion criteria. When assessing parents as eligible for inclusion, we obtained informed consent of participating in the research projects from parents and permission to contact schools and kindergarten of the child in focus. Psychologists from the Centre and MLR then contacted the school/ kindergarten of every child offered PT treatment at the Centre and invited them to join the study. The two teachers/ kindergarten professionals primarily associated with the child were invited to join, and they were

informed on the randomized character of the study, implying that half of the schools /kindergartens would be offered treatment and half would be providing only assessment of child symptoms on online questionnaires. After teachers had accepted participation in the study, MLR conducted the randomization using an online random number generator (<https://www.random.org/>). If two children came from the same kindergarten or school, we randomized them together in order to avoid teachers having to respond to children differently. Parents and teachers of 34 children were randomized to PT and TT, and parents and teachers of 30 children were randomized to PT only. Parents who declined participation in the study ( $n= 2$ ) and parents of children whose school/kindergarten declined participation ( $n= 13$ ) were still offered PT treatment at the center.

Data collection took place during the two-year period from august 2013-august 2015. Parents of 84 children self-referred for treatment at the center, and parents and teachers of 64 children participated in the study (See paper 2 for extensive flowchart of inclusion). Parents filled out questionnaires of child symptoms as well as self-reported questionnaires of parental variables before, during and after treatment as well as at six months FU. Teachers filled out questionnaires before and after treatment as well as at six months FU. No parents dropped out of PT, and teachers of four children dropped out of treatment due to busy schedules ( $n=3$ ) and illness ( $n=1$ ).

In the participant sample of children, 15 (24.4 %) were girls. The mean child age was 6.4 years, and 26 children (40.6 %) had already been diagnosed with ADHD prior to parental self-referral. Out of these, 11 had a comorbid diagnosis to ADHD; ODD/CD in nine children, autism spectrum disorder in one child and tic disorder in one child. Fifty-four (84.4 %) of the families had had prior contacts with the public services regarding their child's difficulties (e.g. school psychologist, child psychiatrist or general practitioner), and 14 of the children (21.9 %) received medical treatment for their ADHD. The mean age of the mothers participating was 36 years, and 50 (78 %) were married or co-living; 14 (22%) were single mothers.

Summaries of the three papers are presented below.

### **Paper 1**

**Purpose:** The main aim of the study was to systematically investigate and synthesize the short- and long-term efficacy of PT for preschool children (2.5-6 years) with ADHD. Moreover, a number of moderator analyses were planned, including methodological quality of the studies; difference in efficacy between generic vs. specific programs; individually vs. group distributed PT programs, and difference in efficacy between PT for children with a formal ADHD diagnosis and children at-risk of ADHD, included in trials on ADHD rating scales.

**Methods:** Inclusion criteria for studies were that the study should be published in a Peer reviewed journal, written in English. The study should be randomized, and control conditions should be no or minimal intervention (i.e. wait-list, minimal intervention or treatment as usual). Children should be 2.5-6 years old, and should be included on a basis of a formal ADHD diagnosis or with symptoms on a validated rating scale. Studies comparing PT to medication were excluded, but medication could be part of the treatment as usual condition. Electronic databases of PubMed and PsycInfo were searched systematically and independently by the first and third author. 2231 records were screened, and 16 studies matched the inclusion criteria. A distinction was made between parentally assessed and objectively assessed efficacy. Studies were subjected to systematic review and meta-analysis using Comprehensive Meta-analysis version 3.2.00089. The ES was computed as a change score variant of Hedges' *g*.

**Results:** There was significant efficacy of PT on parentally assessed reductions of ADHD, corresponding to a moderate ES (0.51,  $p < .001$ ), however this was not the case for independently assessed ADHD (0.12,  $p = .325$ ). For conduct problems, the ES for parentally assessed efficacy was moderate and significant (0.44,  $p = .001$ ), whereas the ES for independently assessed efficacy was small and insignificant (0.31,  $p = .117$ ). For negative parenting, efficacy was moderate to large and

significant ( $0.63, p < .001$ ); for independently assessed reductions in negative parenting, the efficacy remained significant in the small to moderate range ( $0.33, p = .001$ ). Efficacy of parentally assessed symptom reduction and negative parenting were sustained at follow-up. There was significant heterogeneity in 4 out of six analyses, in the moderate to large range. Ratings of methodological quality did not predict magnitude of efficacy. There was no significant difference in efficacy neither between individually distributed and group-based PT, nor between generic and specific PT programs, as well as between children included on the basis of a clinical ADHD diagnosis vs. children included on validated ADHD rating scales.

**Conclusions:** The results partially supported the efficacy of PT for preschool ADHD, seeing that parentally assessed efficacy on ADHD, ODD/CD and reductions in negative parenting was significant and moderate. However, only reductions in negative parenting were significant following analyses of independent assessment. This may be due to rater bias in parents both participating in treatment and assessing the efficacy, alternatively due to methodological difficulties in valid objective assessment of ADHD. PT may be favorable for family functioning, however future studies need to investigate the high levels of heterogeneity and explore the long-term efficacy of preventive PT.

## **Paper 2**

**Purpose:** The main aim of the study was to investigate the relative effectiveness of combining IY PT for 3-8 year old children with ADHD or ADHD difficulties with a TT program specifically developed for early ADHD and conduct problems, compared to IY PT alone.

**Methods:** This study took place at a Danish clinic, Centre for ADHD. Children of parents who had self-referred to Centre for ADHD were screened for inclusion in the RCT. Schools and kindergartens of the children were invited to participate in the study. In total, 64 families and schools fit the inclusion criteria and accepted participation. Families were randomly allocated to

either PT + IY ( $n = 34$ ) or PT only ( $n = 30$ ). Teachers and day-care professionals of 34 children were included in the TT program. Parents and teachers of the children in both groups assessed the child symptoms before intervention, after intervention and at six months FU. Two sets of analyses were conducted separately for both parents and teachers; comparison of differences in change between the two groups from pre intervention to post intervention and from pre interventions to FU. Furthermore, ESs in the form of Cohen's  $d$  were computed based on within-group changes. Finally, percentages of children with statistical and clinical significant change were computed.

**Results:** There were no significant difference in effectiveness between the PT+TT group and the PT only group on neither parental nor teacher-rated outcomes ( $ps > .05$ ). ESs of within-group change based on teacher ratings were significant and moderate for reductions in ODD/CD in the PT+TT group, whereas they were negligible in the PT only group. Mother-reported ESs were in line with those generally reported in the literature. However, mother-reported ESs were generally larger in the PT only group, indicating no evidence of an incremental effectiveness of intervening both at home and in kindergarten/school. Satisfaction with the interventions was high, both for parents and teachers. Despite no drop-out in PT and low drop-out in TT, there was substantial non-completion of questionnaires.

**Conclusions:** Seeing that there were no statistical significant differences between the two groups on neither parental nor teacher reports, there was no apparent evidence of an incremental effectiveness of supplementing PT with TT. However, there were distinctly larger teacher-reported ESs in the PT+TT group, possibly suggesting a context-dependent effectiveness of the TT. Statistical analyses might not detect differences in the small to moderate range due to insufficient power in this small-scale study. Possibly, some effects of a school-based intervention especially likely to generalize across contexts, as peer-acceptance and self-esteem, are too early to detect at 6 months FU. Because of limitations in statistical power and non-completion of questionnaires, caution must be made as to

firm conclusions on the results.

### **Paper 3**

**Purpose:** The primary aim was to explore mediational mechanisms in PT for 3-8 year old children with ADHD or ADHD difficulties. More specifically, we investigated the mediating role of negative parenting, positive parenting, appropriate discipline, parental self-efficacy and working alliance between parent and group leader on reductions in child ADHD and conduct problems.

**Methods:** This study was conducted with the same sample as in Paper 2, where data were collected at Center for ADHD. Mothers of 64 children receiving PT were included in this study. Mothers assessed parental strategies, parental self-efficacy and working alliance as well as child outcome of ADHD and conduct problems before, during, and after PT. Indirect effects of the putative mediators were investigated in multi-level models with product-of-coefficient mediational analysis, and significant mediators were subjected to time-lagged analysis in order to explore the causal relations between mediator and outcome.

**Results:** Reduced negative parenting and increased parental self-efficacy, not positive parenting and appropriate discipline, mediated the efficacy on ADHD and conduct problems. Increased parental self-efficacy appeared to be the strongest mediating variable, explaining 81% of the variance in ADHD symptoms and 28 % of variance in conduct problems. Time-lagged analysis showed that change in mediators did not significantly predict change in symptom outcomes, and a reversed pattern was detected for ADHD and parental self-efficacy, indicating that child ADHD symptom reduction from pre to mid significantly predicted increase in parental self-efficacy from mid to post treatment. Change in therapeutic alliance significantly explained 7 % of the variance in ADHD, not conduct problems, but when applying standard analyses in the area of alliance research, the robustness of these results was negligible.

**Conclusion:** Parental self-efficacy and reduced negative parenting appear to mediate of the efficacy of PT for ADHD and conduct problems, however the causal relationship between these mediators and child symptom outcomes need to be further examined with more fine-grained analysis, as almost all change in child symptom outcomes had already taken place mid-treatment in this study. Therapeutic alliance may not mediate the efficacy of PT, possibly due to a more didactic and educational form than traditional psychotherapy.

## **General discussion**

Across the three papers, results on different aspects of PT for preschool and early school year ADHD raise issues for discussion. In the following section, first a general discussion, then methodological issues and limitations of the present study will be presented.

### **Efficacy of PT**

In Paper 1, the efficacy of preschool PT for ADHD was synthesized, and results partially supported PT as an efficacious treatment, seeing that parentally assessed efficacy was significant with ESs in the moderate range for reductions in ADHD, conduct problems, and negative parenting.

Furthermore, exploratory analyses found that there seemed to be no difference in efficacy between the programs originally developed for reducing conduct problems and the ADHD, just as there seemed to be no difference in efficacy between individual and group-based PT or between children included in trials with clinically diagnosed children, compared to children included with validated rating-scales. The efficacy of PT for preschool children in our meta-analysis (0.51) was comparable to what Daley and colleagues (2014) found (0.35 on most proximal assessment). Though numerically larger, the efficacy of PT was not significantly larger in preschool children than in children aged 3-18, which was the age-range in Daley et al. (2014). Results were also comparable on negative parenting and conduct problems, although the independent assessment of conduct

problems was not significant in our meta-analysis ( $ES = 0.31, p = .117$ ). It could be argued that an effective intervention should be provided as early as possible, to prevent further distress for children and families. If parents alter their parenting strategies to a less negative and inconsistent parenting style early, this may prevent coercive interaction cycles, improving the relation and possibly preventing further development of conduct disorder. Discussion on parental ratings vs. blinded assessment of ADHD is presented in the methodological discussion later in the dissertation (pp. 41).

When analyzing child clinical recovery following PT in paper 2, defined as the percentage of children who had Reliable Change as outlined by Jacobson & Truax (1991) as well as a score of 1 SD below pre-treatment mean at post intervention, it is evident that PT was not a curative treatment for child ADHD symptoms. Less than a third of the children had recovered on parentally rated ADHD at 6 months FU in both groups, suggesting that ADHD symptoms persevered even though parents reported significant reductions in these from pre to FU. Considering that ADHD is a neurodevelopmentally-rooted disorder, defined by chronic and persistent symptoms, it is perhaps not surprising that training parents in handling their child's difficulties does not alleviate all problems for the child.

### **Effectiveness in a kindergarten/school setting.**

In the study presented in Paper 2, there was no statistically significant difference between teacher-reported child ADHD and conduct problems over time between teachers that did and teachers that did not receive a 4\*3 hours intervention based on the core principles of IY PT. Thus, effectiveness of adding TT to PT was not convincing. Possibly, the TT intervention was too short to achieve notable effectiveness. Kindergarten/day-care and school are complex contexts with many children, teachers and agendas. Plausibly, interventions aiming at causing change, not on an overall classroom level but on symptoms at the level of individual children, need to be more comprehensive. However, although not significant in between-group analysis, there was significant within-group change on teacher reports corresponding to moderate ESs in reductions in conduct

problems as measured by the SESBI-R at post intervention and at FU. In the PT only group, there was negligible teacher-reported within-group change on the SESBI-R. This could indicate that there was a teacher-reported effect on child conduct problems when training the teachers, albeit in a smaller scale than could be detected by the statistical power available in this relatively small-scale study. Why would there be change in conduct problems and not ADHD? An explanation could be that, in kindergarten and early school years, teachers and kindergarten professionals less easily detect ADHD symptoms because the scholastic learning is limited and children have more unstructured play-time. In contrast, conduct problems are perhaps more easily detected as aggression and oppositional behavior cause obvious difficulties in interactions with teachers and peers. Indeed, conduct problems cause difficulties in the day-care setting and school for peers and teachers and the affected children themselves (Biederman et al., 1996; Harada, Yamazaki & Saitoh, 2002). It is, however, very likely that ADHD symptoms were simply not reduced in the school setting. Other studies with more extensive TT (e.g. Kern et al., 2007) did not find efficacy on ADHD symptoms either, as described more detailed in Paper 2.

Regarding the effectiveness of PT alone, prior reviews (Rajwan, Chacko & Moeller, 2012) have pointed to an apparent lack of transfer of PT to other contexts (e.g. kindergarten or school), and this was evident in our paper 2 as well. Teachers of children who had received PT alone did not report reductions in child ADHD symptoms, despite the fact that parents of this group of children reported reductions in ADHD corresponding to moderate to large ES from pre to FU. In fact, teachers in the PT alone group perceived ADHD symptoms as increasing from pre intervention to FU. However, caution must be warranted when interpreting this, as this group of teachers could have been demoralized by the fact that they did not receive an intervention. Alternatively, teachers could have increased their attention to child ADHD problems during the intervention period and as such, this increase in ADHD symptoms could be a reflection of this increased focus. Regardless of

interpretations, effects of PT did not generalize to the kindergarten or school context, despite teachers knowing that parents were receiving PT intervention.

### **Mediators of change in PT**

In the analysis of mediators of change in Paper 3, we found that reduced negative parenting and increased parental self-efficacy was associated with reductions in ADHD and conduct problems. Positive parenting and appropriate discipline were not significantly associated with child symptom reductions, and therapeutic alliance only weakly and inconsistently mediated change in both child and parent variables. These findings are in line with results of the relatively few studies that have investigated mediating processes of PT for ADHD, although there is some variation in previous results. Seabra-Santos and colleagues (2016) found reductions in negative parenting and increased parental self-efficacy to be statistical mediators of the treatment effect in their study of IY PT for early ADHD, fully in line with findings in the present study. Mediation analyses from the Multimodal Treatment of Attention Deficit Hyperactivity Disorder Study (MTA) study by Hinshaw et al. (2000) found that reduced negative parenting, and not increased positive parenting, mediated the efficacy of PT. Results are however not entirely comparable, as this pattern was only evident in the PT+ medication group, and on teacher reports on social skills. Hanisch et al. (2014) found statistical mediation on both reduced negative parenting and increased positive parenting following a preventive intervention for both ADHD and conduct problems.

That reductions in negative parenting should mediate improvement in child outcomes is in accordance with social learning theory of development of externalizing difficulties, considering how negative and coercive parenting, possibly elicited from the stressful conditions of parenting a child with ADHD and conduct problems, can form a basis of negative interactions with the child and escalation of aggression. Similarly, although ADHD is a disorder with strong biological disposition, a family context characterized by high levels of conflict and stress could be assumed to

exacerbate child symptoms of ADHD (Johnston et al., 2001). Thus, reducing adverse and harsh parenting in order to reduce child symptoms of both ADHD and conduct problems is concordant with the theoretical basis of PT. Increased parental self-efficacy was also associated with reductions in conduct problems and ADHD, explaining as much as 81 % of the variance in symptoms of ADHD over time. Mothers who have high levels of parental self-efficacy are more consistent in their parenting, display high levels of maternal warmth and have high satisfaction with their parenting, which may improve the parent-child relationship further.

However, in Paper 3 we wanted to explore the timeline of change in order to further elucidate the causal relations between reductions in child symptoms and parenting variables; something that had not been done in prior studies investigating mediational processes in PT for ADHD. Here, the only significant relationship established was the reversed finding that reductions in ADHD symptoms from pre intervention to mid intervention significantly explained the increase in parental self-efficacy from mid to post intervention. Thus, as such, this paper could not establish a timeline of change between the mediators that we found significantly associated with change in child symptoms. It is reasonable to assume that parenting a child who displays fewer symptoms is easier, thus improving parental sense of mastery and self-efficacy. However, theoretically, parental competences and self-efficacy should also assist in reducing child difficulties, maybe as a consequence of the less negative parenting taught in the PT, as proposed by Seabra-Santos et al. (2016). This reciprocal relationship cannot be established, however, without a timeline of change.

### **Is one year enough time to capture the full effects of early intervention?**

One of the benefits of training primary adults surrounding the child may be the possibility of adults continuing to utilize the acquired skills and strategies after the intervention has ended; a possible advantage over pharmacological intervention. The theoretical base of PT views dysfunctional interactions as cyclically consolidated patterns over time (Patterson, Reid & Dishion, 1998; Patterson, 2002). It may thus similarly take some time to alter these interactions and consequently

see the full results unfold in reductions of child difficulties. Thus, assessing the effectiveness of PT and PT combined with TT at post treatment and at FU 6 months later (as in paper 2) or even at 12 months later (as was the longest period of FU in the meta-analysis of Paper 1) may be too soon to observe the full effectiveness. In the meta-analysis, within-group analysis from post to FU showed that there was slight and marginally significant increase in efficacy on all parent reported measures, and on parent-reported effectiveness on ADHD (CRS) and on conduct problems (ECBI) in Paper 2, symptoms were reduced further from post to FU. Seeing that parents continue to perceive effectiveness of PT, then possibly the FU at 6 months after PT was ended was too early to see the full range of the effectiveness. Drugli, Larsson, Fossum and Mørch (2010) investigated the effects of PT for ODD/CD in a sample of 4-8 years 5-6 years after termination of PT. Where all children qualified for a diagnosis pre-treatment, two-thirds of the children who participated at 5-6 years FU did not qualify for an ODD/CD diagnosis. This speaks to the strengths of PT for long-term effectiveness. This may, however, not be generalizable to an ADHD population, seeing that ODD/CD is a more interactionally rooted disorder compared to ADHD, viewed as a chronic disorder and not typically outgrown (e.g. Lahey et al., 2004).

Furthermore, as discussed in Paper 2, there could be aspects of the TT that simply take more time to unfold, as peer acceptance and self-confidence. If these effects had not unfolded by the six months FU, we do not know if these could have generalized out of the school context to the home setting and thus have supported an incremental effectiveness of adding PT to TT. In this way, the study may not have been successful in evaluating the effects of combining early interventions in more settings.

## **Methodological issues and limitations**

A number of methodological issues and limitations have been raised throughout the three papers constituting this dissertation, and below, some of them are further addressed.

### **Limitations from the meta-analysis (Paper 1)**

There are some methodological limitations in Paper 1. Seeing that only English-language papers were included and that only two databases were searched, it is possible that some studies could have been left out. Although we also searched the reference lists of the included studies, it is uncertain what studies could have been found in other databases. Furthermore, the Jadad protocol for quality assessing the included studies may have been too limited, only including dropout, randomization and blinding. Seeing that there was little variance in the Jadad scores on the included studies, a more comprehensive grading may have revealed other aspects. The adapted version of the Jadad was chosen as it had been developed and used previously in the specific field by Daley et al. in their comprehensive meta-analysis (2014), however a more rigorous version revealing other aspects could have been used to further explore and evaluate the overall effectiveness of PT for preschool ADHD. Finally, the ES metric in the form of Hedges  $g$  may not be intuitively interpretable for clinicians. Although very frequently used in psychological research, other measures could have been applied that more directly reported on clinical difference.

### **Validity of parent-reported questionnaires vs. blinded assessment**

A substantial methodological issue raised in Paper 1 was the apparent discrepancy between efficacy on parent-assessed and independently assessed outcomes. When relying on blinded ratings, only reductions in negative parenting were significant. There are several interpretations to this finding. First, it could be that parents were positively biased, naturally being both emotionally involved with their child and wishing to see a positive change. Seeing that parents were both the active agents in treating their child as well as assessors of the efficacy on their child, it is clear that parents are not

neutral informants on this matter. Following this argument, the only valid efficacy of PT would be reduced harsh and negative parenting, as this was evident on blinded, independent assessment as well.

Conversely, it could also be that the validity of the objective measures of child symptoms was compromised. Assessment of child symptoms of ADHD in the studies included in the meta-analysis was generally conducted with observations of children in a lab, live or audiotaped, lasting 5-30 minutes. These relatively short observations conducted before and after PT may not be enough to validly capture ADHD symptoms. Furthermore, it could be hypothesized that children with ADHD difficulties could be performing an extraordinary effort in solving the tasks they were presented with while being observed in a lab in a relatively short period of time. If this were the case, these observations would not mirror the actual symptomatic state of children while problem solving or playing outside the experimental lab. Knowledge of the validity of observational measures is limited (Pelham et al., 2005), and the heterogeneity of measures was high, which makes it difficult to conclude on this matter. Considering that parents observe and interact with their child during extensive amounts of time and across context, they may be more valid observers of child difficulties. However, successfully blinding parents to experimental conditions is of course impossible in investigating PT. The obvious limitation drawn from this methodological discussion is that paper 2 and 3 were relying solely on un-blinded parental and teacher assessment. This is a substantial limitation in this study, as we cannot infer about the degree as to which these assessments were biased, with no comparison to observational measures of neither child nor parental outcomes.

### **Discrepancy between parental ratings and teacher ratings on ADHD**

In Paper 2, there was an apparent discrepancy between teacher and parent ratings of child ADHD at baseline. Teachers rated the children somewhat lower on ADHD ( $M = 17.24$ ,  $SD = 6.3$ ) than parents ( $M = 23.27$ ,  $SD = 4.44$ ). We contacted the teachers/day care professionals after parents had self-

referred to the Clinic in order to receive treatment. This implies that children were included on parental initiative, not on teacher referral. The somewhat lower teacher-rating of child ADHD at baseline could indicate that teachers were not utterly concerned with the ADHD difficulties of the child. If teachers did not perceive children as suffering from severe ADHD symptoms, the null finding of the TT intervention on ADHD is not surprising. Teachers did perceive children as displaying substantial conduct problems (group  $M = 155$ , clinical cutoff = 151 on the SESBI-R) and there was significant teacher-rated within-group change on conduct problems (SESBI-R) in the PT +TT group corresponding to a moderate ES (0.61 at post treatment), compared to no significant change in the PT only group. Maybe the relatively young age of children and the corresponding limited scholastic demands, in particular in kindergarten, did not allow for teacher detection of ADHD as readily as ODD/CD. A study investigating the prognostic validity of teacher-, clinician- and parent- ratings of ADHD in preschool found that clinician-and-parent-rated ADHD in preschool, not teacher-ratings, predicted ADHD at age six (O'Neill et al., 2014). Clinician-ratings were the best predictor of symptoms of ADHD in lab observations at age six (O'Neill et al., 2014). However, studies have also pointed to low agreement between parent-and teacher-ratings of ADHD in school-age children, with teachers rating ADHD higher than parents (Wolraich et al., 2004). Conclusion on the relative validity of parent and teacher reports is generally a difficult topic (Pelham et al., 2005).

### **Following Clinic inclusion criteria in the RCT**

Children included in the study reported in Paper 2 and 3 were not included based on a formally assessed ADHD diagnosis, but on the basis of parental concern, validated rating scales (but not necessarily above clinical cutoff) and semi-structured interviews with parents. The methodological implication of this inclusion is that we cannot know if children would, in fact, qualify for a formal ADHD diagnosis. This decision not to require a formal ADHD diagnosis is rational from a clinical perspective, bearing in mind the preventive aspect of the treatment and the intention of the clinic to

provide easy-access treatment to children and families early, before child difficulties are further consolidated through negative interactions with family, teachers and peers. However, it is a limitation from a research point of view, as some of the children in the sample might not qualify for an ADHD diagnosis. The combination of clinician assessment and parent ratings of child ADHD symptoms in preschool has been found to be a valid predictor of ADHD at age six in the study by O'Neill et al. (2014), which lends support to including children in our study with clinical interviews and rating scales. Furthermore, the moderator analysis from the meta-analysis in Paper 1 did not find significant differences in efficacy between children with formal diagnosis and children who had symptoms of ADHD on a rating scale. Trillingsgaard et al. (2014) benchmarked results of PT at Centre for ADHD against results from a rigorous RCT conducted by Webster Stratton and colleagues in Seattle (2011). They found that the self-referred Danish sample treated with IY PT at the same clinic as in paper 2 and 3 in our study were comparable to the American RCT with formal ADHD diagnosis of participant children. Thus, although children in this sample were not included on formal diagnosis, the majority of them may have a high likelihood of receiving the diagnosis if evaluated.

### **Statistical power, non-completion of questionnaires and lack of passive control comparison**

An important methodological issue and central limitation in paper 2 was the relative lack of statistical power. The number of participants in the study was based on practical considerations. The maximum period for data collection in a PhD project was considered 2 years. The capacity of the clinic was to treat 84 children over the two-year period that constituted the active data-collection. Out of these 84 families, 64 fit the inclusion criteria. A sample of 64 children denotes some restraints regarding statistical power. When comparing two active groups, as was the case for mother reports in this study, small differences may likely be the upper bound of change that could be expected. A priori power calculations showed that 120 participants would be needed to obtain

statistical power at the recommended .80 level. Thus, on mother-reports, our study was underpowered to detect differences in the small range.

For detection of differences corresponding to moderate ESs, a priori power calculations had showed that in order to obtain statistical power at the 0.80 level, 28 participants should be enough. This means that the sample size of 64 (and even 42, as was number of teachers completing post treatment assessment) should be sufficient in order to obtain enough power to detect moderate differences. However, a small sample is of course not very robust regarding error and variance. Considering the moderate ES on the SESBI-R pre-post and pre-FU that teachers in the PT+TT group reported, compared to negligible difference in the PT alone group, restricted power may likely have played a role here.

In the meta-analysis, statistical power was also a problem in the moderator analyses with few studies in the pairwise comparisons, thus warranting caution in interpretation of the moderator analyses in Paper 1.

Moreover, the fact that about 19 % of the mothers failed to complete questionnaires post intervention is a considerable limitation in the study. This non-completion occurred despite the comprehensive reminder protocol and the attempt to reward completion of questionnaires with LEGO ® boxes at FU. Non-completion of assessment in this range is not uncommon in the field of PT for ADHD (e.g. Östberg & Rydell, 2012). On teacher reports, 34 % of the teachers failed to complete questionnaires at post intervention. Almost 50% of the teachers in the control comparison (PT only) failed to complete questionnaires despite our reminder procedures and attempts to reward completion of questionnaires with LEGO ® boxes for completing teachers. This indeed compromised the quality of the data. When investigating the context-specific effectiveness of the TT in school, losing so many teacher-assessments both compromises the data itself and statistical power as well. Teachers in the PT only group may have been demoralized after being randomized

to control-condition and thus lost the motivation to complete questionnaires. Furthermore, there had been lockouts between schools and teachers a few months before starting the RCT, and a new large-scale educational reform was implemented parallel with our study. Substantial changes in the organization of the school and increased requirements for teachers' working hours were part of the reform, and it was generally unpopular among teachers. This was of course unfortunate as a context for implementing our study, as teachers may likely have had fewer resources to complete questionnaires etc. Furthermore, this may also explain why 13 out of 60 schools declined to participate in the study, thereby bypassing the supposed attractive opportunity to receive a free specialized course. The reform and the particular circumstances surrounding the context of the study must be taken into consideration when interpreting the negative results from paper 2.

Finally, we had no passive control-group in paper 2 and paper 3. Without a passive control-group, we cannot claim a "true" effect of PT because we cannot attribute changes to the intervention solely. Although ESs were in line with those reported in prior studies (e.g. Webster-Stratton et al., 2011), we cannot know if children would have had reductions in symptoms over time without intervention. This claim is also relevant in the meta-analysis, where there was no passive comparison in the FU. Some child difficulties can be outgrown or simply be a product of regular developmental changes, and without a passive control group, we cannot infer about the relative proportion of child difficulties that would simply diminish over time. On the other hand, passive control groups in child research should be applied with caution (Herbert et al., 2015). For a distressed family seeking help for child difficulties, waiting half a year or longer to receive treatment is a long time and can have consequences for family and child functioning. Long-term FU with passive comparison groups are normally ethically not justifiable, and thus very rarely conducted. The lack of control-groups does warrant some caution from a research perspective as to firm conclusions on effectiveness of PT in this study. However, other studies with wait-list groups (as Webster-Stratton et al., 2011) have found that child difficulties do not seem to diminish over

time. Especially ADHD symptoms, viewed as chronic and persistent, are not likely to change. Thus, although not rigorously controlled, the effectiveness of PT from this study can still contribute to the growing field of field of research on PT for ADHD.

## **Conclusions, implications and future perspectives**

Taken together, we concluded that there is partial support for intervening with PT for early ADHD, based on the meta-analysis on studies investigating PT for early ADHD. There was significant efficacy on parental ratings of ADHD symptoms, conduct problems and negative parenting, and results were stable at FU. However, only negative parenting was supported in efficacy when analyzing data from blinded raters, warranting caution of possible bias in parental ratings (Paper 1). Furthermore, there was no incremental effect of adding TT to PT, as there were no statistical difference in effect between the two groups on neither parental nor teacher assessment of child difficulties. However, teachers that had received TT reported significant change in child conduct problems corresponding to a moderate ES, compared to negligible change reported by the teachers that had not received TT. This could indicate a school-context specific advantage of training teachers, although the limited statistical power and the high non-completion of assessment of teachers in the PT only group preclude conclusions (Paper 2). Finally, in paper 3, the reductions of parental ratings of ADHD and conduct problems following PT appeared to be mediated by reductions in negative parenting and increased parental self-efficacy, explaining up to 81 % of the variance. However, time-lagged analyses did not support the hypothesized relationship between changes in mediators causing change in outcome. On the contrary, it appeared that reductions in ADHD caused increased parental self-efficacy. However, it is important to note that more time-points could possibly have revealed a different timeline.

Considering findings from prior research and presents results, PT is a favorable intervention for parents of children with ADHD difficulties. Parents report less ADHD symptoms and less conduct problems among their children. They also report their own parenting as less harsh and negative and feel more efficacious in their role as parents. Seeing that families of children with ADHD are at risk of or present with high levels of conflict, stress and divorce, these results are promising. However, as is also evident from the results, PT is not a curative treatment of neither ADHD nor conduct problems. Results of reliable change (RCI) and clinically significant change (a score of 1 SD below pre-treatment mean) in Paper 2 showed that less than a third of all children had recovered, (i.e. had both statistical and clinical change) suggesting that PT does not eliminate all difficulties. Bearing in mind the biological foundation and chronic and persistent character of ADHD, this may likely not be the case for any treatment of ADHD. Plausibly, the realistic aim of PT is not to eliminate child symptoms, but rather to alleviate symptoms and possibly prevent a more severe condition without clinical levels of comorbid ODD/CD (Sonuga-Barke et al., 2013; Webster-Stratton et al., 2011). In addition to child symptom reduction, parenting also changed during PT, with less harsh and negative parenting and perceiving higher levels of self-efficacy in their parenting, plausibly leaving parents more satisfied in their role as parents. This is of clinical relevance, as parenting a child with ADHD difficulties appears to increase the risk of parental stress and strain. Positive parenting also appears to increase following PT, probably also contributing to a more pleasant environment at home for both parents, children and siblings, despite no evidence as mediating effect on reductions of child symptoms.

Concordant with previous findings (e.g. Rajwan et al., 2012), effects of PT may not generalize to a kindergarten/ school setting. This implies that PT should not be a stand-alone treatment if the child displays substantial difficulties in the school/kindergarten setting. On the other hand, the lack of significant difference in adding TT to PT compared to PT alone, both in the home context and partly in kindergarten/school, suggest that this combination is not effective either, although firm

conclusions on this matter cannot be drawn, taking into consideration the small scale nature of this study and the degree of non-response of the teachers in the PT only group.

Results from this project raise several implications for future research. Since there appears to be a pattern of discrepancy between efficacy on parental ratings and blinded assessment of ADHD symptoms, future studies should investigate the validity and sensitivity of blinded measures of ADHD in order to validly evaluate the outcomes of treatments. More research is also needed on interventions in the school and combination with PT, preferably in larger samples and with more extensive training of the teachers. Similarly, the long-term effects of intervening early in both family and daycare/school for early ADHD symptoms are largely unknown. Considering the transactional aspects of child developmental difficulties, both in relation to parents, teachers and peers, interventions may need longer time to fully unfold. Finally, further mediational research on the active ingredients in PT may result in therapeutic optimization of PT. In particular, future efficacy studies should implement multiple data-points in order to establish a time-line of change, thus facilitating conclusive findings of causal relations between mediators and outcome in training parents of children with ADHD.

This project was motivated by the prospects of improving clinical practice by studying what happens when training families and teachers of children with ADHD or ADHD difficulties. The struggles that these children face tend to add a heavy burden on early family life and gradually transfer to become school difficulties as well. Families of children with ADHD tend to have more frequent conflicts and negative interactions (Sollie et al., 2016; Mash et al., 1982). Parents of children with ADHD often feel that their parenting is insufficient and they display low levels of parental self-efficacy (Mash & Johnston, 1983; Johnston & Chronis-Tuscano, 2015). As evident from the current study findings, parents perceive their parenting as less negative and have more self-efficacy in their role as parents following PT. As illustrated by this quote from a mother of a 6-

year-old boy in an interview after participating in the IY PT at the clinic, conducted in the spring 2013:

*“I don’t think of the situation as hopeless anymore. When I am having a difficult conflict, I know I have some tools I can use [...] some additional strategies. Many times before, I have felt like a bad mother to [boy’s name], I was really often yelling at him, and I actually did not feel that I gave him what I ought to. I don’t have that feeling anymore, so things have become easier, it is generally more pleasant to be a mother”.*

PT will not eliminate ADHD symptoms, and due to the chronic, persistent nature of ADHD, further intervention later in life may still be important. PT will also not eliminate ADHD related difficulties from affecting children’s lives in the school or daycare setting. PT may, however, prevent exacerbation of child difficulties and reduce distress in the family, altering negative and harsh interaction patterns. With the end goal being optimal coordination and transfer of training effects across family and school/daycare, this thesis represent the evaluation of one first such step. It remains important, and potentially fruitful, to continue the development and evaluation of programs that aim to improve this transfer. Hopefully, future research will continue such further steps, thereby helping both children, families and teachers to reduce adverse interactions and improving sense of efficacy and self-worth while effectively ameliorating the stressors and stress from children living with ADHD difficulties.

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## Dansk resume

**Baggrund:** ADHD optræder hos ca. 5.29 % af alle børn på verdensplan (Polanczyk et al., 2007).

Ud over selve kernesymptomerne uopmærksomhed, hyperaktivitet og impulsivitet oplever børn med ADHD ofte vanskeligheder i form af lavt selvværd (Mazzone et al., 2013), social afvisning hos jævnaldrene (Bagwell et al., 2001) og øget risiko for at udvikle adfærdsvanskeligheder (Beauchaine, Hinshaw & Pan, 2010). Det er udfordrende at være forælder til et barn med ADHD eller ADHD-lignende vanskeligheder, og i familier med børn med ADHD er der ofte mange konflikter. Forældrene oplever tillige ofte lav forældre-self-efficacy. Forældretræning (FT) er en intervention der retter sig imod at ændre forældrenes strategier og redskaber for at afhjælpe barnets vanskeligheder. **Formål:** Formålet med dette forskningsprojekt var at undersøge effekten af FT over for tidlig ADHD. For det første ville vi undersøge den samlede, hidtil offentliggjorte evidens for forældretræning til førskolebørn i alderen 3-6 år (Artikel 1). For det andet ville vi undersøge om der er større virkning af at træne både lærere og forældre, i forhold til kun at træne forældrene (Artikel 2). Afslutningsvis ville vi undersøge hvilke processer i FT der medierede den forældre-rapporterede symptomreduktion hos børnene (Artikel 3). **Metode:** For at evaluere den samlede evidens for effekten af FT til førskole ADHD søgte vi systematisk i de elektroniske databaser PsycInfo og PubMed med centrale søgeord for at identificere de relevante udgivne studier. Data fra de identificerede artikler blev samlet i en metaanalyse, hvor vi separat analyserede forældrenes rapporteringer og uafhængige bedømmelser af effekten af FT (Artikel 1). For at undersøge effekten af både at træne lærere og forældre inkluderede vi 64 familier med børn i alderen 3-8 år, der havde selv-henvist sig til Center for ADHD, en uafhængig, non-profit privat klinik i Århus. Familierne blev randomiseret til enten FT med forældretræningsprogrammet De Utrolige År ® kombineret med et fire-sessions kursus baseret på principperne fra IY til to af de tætteste lærere/pædagoger til barnet, eller FT alene. Mødrene rapporterede om barnets symptomer og egne forældrevariable før, under, efter og 6 måneder efter FT; lærerne rapporterede om børnenes symptomer før, efter og 6

måneder efter (Artikel 2). Mødrenes rapporteringer om deres barns symptomer, forældrestrategier og forældre-self-efficacy blev undersøgt i statistiske mediations-analyser, der tog forbehold for tidsfølgen i udviklingen af forældrevariable og disses påvirkning på børnenes symptomer (Artikel 3). **Resultater:** Meta-analysen fandt en samlet, moderat forældre-rapporteret effekt af FT til førskolebørn med ADHD, både på ADHD symptomer, adfærdsvanskeligheder og negativ forældrestil; dog var kun reduktion i negativ forældrestil signifikant i de uafhængige bedømmelser. Den forælderreporterede effekt var stabil i follow-up perioder op til 1 år efter endt FT (Artikel 1). Der var ingen signifikante forskelle på læreres og forældres rapporteringer af børnenes symptomer imellem den gruppe der havde modtaget både FT og lærertræning, og dem der havde modtaget FT alene. Det kunne tyde på at der ikke var en øget effekt af også at træne børnenes lærere/pædagoger . Dette nul-fund skal dog tolkes med forsigtighed, da der var en lærer/pædagog-vurderet moderat effektstørrelse på adfærdsvanskeligheder i den gruppe af børn der havde fået kursus modsat ingen effekt hos de lærere der ikke modtog kursus. Dette kunne muligvis kunne indikere en kontekst-specifik effekt af kurset i skolen/børnehaven (Artikel 2). I mediationsanalyserne fandt vi, at reduceret negativ forældrestil og øget forældre-self-efficacy, ikke øget positiv forældrestil og en mere passende disciplin, lod til at mediere effekten på barnets symptomer på ADHD og adfærdsvanskelighed. Tidsserieanalyserne kunne dog ikke etablere, at ændringen i mediatorerne skulle gå forud for ændringen i symptomerne hos barnet. Der så ud til at være et omvendt forhold mellem forældre-self-efficacy og ændring i ADHD symptomerne, således at reduktionen i ADHD-symptomerne hos børnene gik forud for en øget forældre-self-efficacy. Terapeutisk alliance var kun svagt og inkonsistent forbundet med ændring i både forældre-og-børnevariable. **Konklusioner:** På baggrund af metaanalysen i artikel 1 findes der delvis støtte til FT som indsats til ADHD hos førskolebørn, om end der kun var blindet dokumentation for effekten på negativ forældrestil. Ud fra resultaterne i artikel 2 lader der ikke til at være en øget effekt af at tilføje lærertræning til FT. Der kan dog måske være tale om en kontekst-afhængig effekt i skolen på adfærdsvanskeligheder. Der er

brug for mere forskning med flere deltagere og muligvis mere udvidet kursus til lærere/pædagoger for at kunne konkludere på involvering af lærere/pædagoger i FT til tidlig ADHD. Endeligt lod reduceret negativ forældretil og øget forældre-self-efficacy i artikel 3 til at være forbundet med reduktion af symptomer hos barnet, men der er brug for mere forsknings med flere datapunkter for at kunne konkludere yderligere på de kausale forhold i virkningsmekanismer i FT.

## **English summary**

**Background:** Childhood ADHD is prevalent in 5.29 % of children worldwide (Polanczyk et al., 2007) and is associated with low self-esteem (Mazzone et al., 2013), peer rejection (Bagwell et al., 2001) and increased risk of developing subsequent conduct problems (Beauchaine, Hinshaw & Pang, 2010). Parenting a child with ADHD is challenging, and high levels of conflict and low parental self-efficacy often characterize families of children with ADHD. Parent training (PT) is an intervention aiming at altering parenting strategies in order to improve child difficulties.

**Objectives:** The aim of the study was to explore PT for early ADHD. First, we wanted to investigate the combined evidence of the efficacy of intervening with PT for children aged 3-6 years (Paper 1). Second, we aimed to investigate the relative effectiveness of adding teachers training (TT) to PT, compared to PT alone (Paper 2). Finally, we wanted to explore which processes in PT that mediated the parent-rated reductions of child outcomes (Paper 3). **Methods:** To assess the combined evidence of the efficacy of PT for preschool ADHD (Paper 1), the electronic databases of PubMed and PsycInfo were searched systematically with relevant search terms in order to identify published studies that had investigated PT for preschool ADHD. The data were subjected to meta-analysis, and separate analyses were conducted for parent ratings and independent observations of child symptoms following PT. In order to examine the relative effectiveness of adding TT to PT (Paper 2), we included 64 families of children aged 3-8 years, who had self-referred to Center for ADHD, a Danish non-profit independent clinic. Families were randomly allocated to either PT with Incredible Years ® PT (IY) (Webster-Stratton, 2011) in combination with a four-session TT course based on IY principles for two of the child's teachers, or to PT alone. Parents assessed child symptoms and parenting variables before, during and after PT as well as six months later; teachers assessed child symptoms before, after and six months later. Mother-reports of both child outcomes and parenting variables were subjected to both statistical mediation and time-lagged analysis to investigate the causal relations between mediator and outcome (Paper 3).

**Results:** The meta-analysis found an overall moderate parent-rated effect of PT on ADHD symptoms, conduct problems and negative parenting; however only negative parenting was significant on independent observations. Parent reported results were stable in follow-up periods up to 1 year after treatment had ended. There were no significant differences on ratings of child ADHD and conduct problems between the teachers and parents that had received both PT and TT and the teachers and parents in the PT only group. This indicates no incremental effect of adding TT to PT. However, a moderate effect size (ES) on teacher ratings of child conduct problems in the PT +TT group compared to no teacher-reported change in the PT alone group, could point to a school context-specific effect of TT. When investigating mediators of change in PT on parental ratings from all 64 mothers before, during and after PT, reduced negative parenting and increased parental self-efficacy, not increased positive parenting or appropriate discipline, appeared to mediate the reductions in child ADHD and conduct problems. However, time-lagged analyses could not detect a change in the statistical mediators as explaining subsequent reductions in child symptoms. A reversed pattern was found for parental-self efficacy and child ADHD, indicating that as child symptoms decreased, parental self-efficacy increased. Therapeutic alliance was only weakly and inconsistently related to change in both child and parent outcomes. **Conclusions:** On the basis of the meta-analysis in Paper 1, we conclude that there is partial support for intervening with PT for early ADHD, although blinded evidence was only found for reduction in negative parenting. Our data from paper 2 does not support an incremental effect of adding TT to PT. Cautious interpretation of these data is warranted, as there may be a context-specific effect of TT on reductions of child conduct problems. More research with larger samples and possibly more extensive TT is needed. Finally, based on results in paper 3, reductions in negative parenting and increased parental self-efficacy may mediate the effect of PT on child outcomes. Future research investigating PT should include more data points to establish causal relations between parenting variables and child outcomes.

## Appendices

## Appendix A

**Paper 1:** Rimestad, M.L., Lambek, R., Christiansen, H.Z. & Hougaard, E. (2016). Short- and long-term effects of parent training for preschool children with or at-risk of ADHD: a systematic review and meta-analysis. *Journal of Attention Disorders*, e-pub ahead of print, DOI: 10.1177/1087054716648775.

# Short- and Long-Term Effects of Parent Training for Preschool Children With or at Risk of ADHD: A Systematic Review and Meta-Analysis

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## Abstract

**Objective:** The aim of the study was to synthesize the evidence of parent training (PT) as an early intervention for preschool children aged 2.5 to 6 years with ADHD or ADHD symptoms. **Method:** A systematic review and meta-analysis was conducted. **Results:** Sixteen studies including 1,003 children were analyzed. Parent-rated outcomes revealed moderate effect sizes (ESs; Hedges'  $g$ ) of 0.51 for ADHD symptoms, 0.44 for conduct problems, and 0.63 for negative parenting. Based on independent assessment, results were only significant for negative parenting. Parent-rated outcomes were sustained at follow-ups of 3 to 12 months. Program type, intervention modality, and child diagnostic status did not moderate the effect. **Conclusion:** PT was partially supported as an efficacious intervention for preschool children with ADHD or ADHD symptoms with moderate ESs on parent-rated outcomes, but no significant results on independently assessed ADHD symptoms. (*J. of Att. Dis.* XXXX; XX(X) XX-XX)

## Keywords

ADHD, parent training, early intervention, meta-analysis

## Introduction

ADHD is a neurodevelopmental disorder characterized by age inappropriate, persistent, and pervasive inattention and/or hyperactivity-impulsivity that interferes with daily functioning at home, school, or work (American Psychiatric Association, 2013). The disorder is associated with a host of problems, including emotion dysregulation (Shaw, Stringaris, Nigg, & Leibenluft, 2014), neuropsychological dysfunction (Pauli-Pott & Becker, 2011), social problems (Bagwell, Molina, Pelham, & Hoza, 2001), and academic underachievement (DuPaul & Stoner, 2014). ADHD may also increase the risk of developing oppositional defiant disorder (ODD) and conduct disorder (CD), disorders that are highly related to school dropout, delinquency, and crime (Beauchaine, Hinshaw, & Pang, 2010; Offord & Bennett, 1994). Even though the diagnostic criteria for ADHD were originally developed with a primary focus on school-age children, ADHD is also diagnosed in preschool children (Greenhill, Posner, Vaughan, & Kratochvil, 2008; Lahey et al., 2004; Sonuga-Barke, Auerbach, Campbell, Daley, & Thompson, 2005) with an estimated 2% to 6% of preschoolers meeting the criteria for ADHD (Lavigne, LeBailly, Hopkins, Gouze, & Binns, 2009). Given the negative impact

of ADHD, there is an obvious need for an intervention that works. The question is when and how to intervene.

The risks associated with ADHD appear to increase with early onset (Barkley et al., 2000), just as the possibility of change may be greater early in life as the brain's ability to reorganize itself (i.e., neuroplasticity) is more pronounced in younger children (Halperin, Bédard, & Curchack-Lichtin, 2012). From a family perspective, the presence of ADHD symptoms may increase the likelihood of parents responding to their children with a negative parental style, which in turn might maintain or even enhance the disruptive difficulties of the child (Johnston & Mash, 2001). Therefore, early intervention that aims at altering inadequate parenting strategies before they consolidate might be beneficial (Thompson et al., 2009). If the developmental trajectory of the child is altered early on, some of the secondary difficulties related to ADHD such as peer rejection, low self-esteem, and

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comorbid ODD/CD might be prevented (Beauchaine et al., 2010; Halperin et al., 2012; Sonuga-Barke & Halperin, 2010), possibly resulting in a less severe condition (Sonuga-Barke et al., 2013).

ADHD can be treated pharmacologically (Banaschewski et al., 2006), but the efficacy is less consistent and associated with more adverse effects in preschool children (Kollins & Greenhill, 2006). Parent training (PT) is an early intervention that aims to alter inadequate parenting strategies, promote positive parent-child relationships, and in some cases target the underlying neuropsychological problems characteristic of ADHD (e.g., Abikoff et al., 2015; Sonuga-Barke, Daley, Thompson, Laver-Bradbury, & Weeks, 2001). Currently, PT programs fall into two categories, more generic programs that have been developed from ODD/CD PT programs and adapted for ADHD (Bor, Sanders, & Markie-Dadds, 2002; Jones, Daley, Hutchings, Bywater, & Eames, 2007; Webster-Stratton, Reid, & Beauchaine, 2011) and programs specifically developed for ADHD such as the Revised New Forest Parenting Programme (Sonuga-Barke et al., 2001; Thompson et al., 2009). Programs also differ with respect to delivery modus, that is, whether PT is delivered in groups or individually. The various programs do, however, have theoretical and methodological similarities. For instance, they all include tenets from social learning theory; apply psychoeducation; promote a positive focus on the child; train parents in the importance of structure, routines, and predictability in the everyday life of the child; and use praise and rewards as incentives.

Two prior meta-analyses by Sonuga-Barke et al. (2013) and Daley et al. (2014) investigated the effectiveness of behavioral interventions (including PT) for children and adolescents with ADHD aged 3 to 18 years. Both meta-analyses pointed to a discrepancy in the results depending on the assessor perspective; interventions were efficacious for parental reported ADHD symptoms but not for ADHD symptoms rated by independent assessors. A recent meta-analysis investigating PT for preschool ADHD (2.5-6 years; Mulqueen, Bartley, & Bloch, 2015) found a moderate effect on parental reported ADHD symptoms, the only outcome analyzed, based on a small number of studies ( $n = 8$ ).

The primary aim of this article is to evaluate the effectiveness of PT programs, at post-treatment and follow-up, for preschool children with or at risk of ADHD (2.5-6 years) by means of a systematic review and meta-analysis. ADHD symptoms, child oppositionality and conduct problems (henceforth conduct problems), and negative parenting, parent-reported or rated by independent assessor, were examined. Moderator analyses were planned for methodological quality of studies, type of program (generic vs. specific PT programs), modality (group vs. individual intervention), and ADHD diagnostic status (diagnosis vs. ADHD symptoms).

## Method

The study was conducted in accordance with the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines (Moher, Liberati, Tetzlaff, Altman, & The PRISMA Group, 2009).

### Inclusion Criteria

Randomized controlled trials (RCTs) of PT programs and interventions published in peer-reviewed English language articles were included in the review. Participants were parents of preschool children aged 2.5 to 6 years with ADHD symptoms according to a formal diagnostic system or rated on a validated rating scale. Studies comparing PT with systematic medication were excluded, although use of medication as part of treatment as usual was allowed. Control conditions had to be no or minimal intervention groups (e.g., waitlist, treatment as usual, minimal intervention, or attention control).

### Search Strategy and Data Extraction

The electronic databases of PsycInfo and PubMed were searched in May 2015 using the following keywords: "Attention\* Deficit\* Disorder\*" or "Attention\* Deficit\* Disorder\* with Hyperactivity\*" or "adhd" or "hyperkin\* disorder\*" and "parent AND train\*" or "interven\*." Relevant Medical Subject Headings (MESH terms) were included in the PubMed search. The search was not restricted by year. Article titles, abstracts, and in some cases full texts were screened to evaluate eligibility, just as reference lists of included studies were screened to identify published follow-ups of studies included in the review, separate searches were conducted. The first author (MLR) and the third author (HJC) independently conducted the literature search, and disagreements were resolved through discussion ( $n = 3$ ). An excel spreadsheet with coding variables was created and data were extracted independently by MLR and HJC regarding program, diagnosis or rating scale symptoms, age of the children, percentage of girls, intervention modality, control condition, duration of intervention as well as measures of ADHD, conduct problems, and negative parenting. Furthermore, the PT program was coded according to whether it was initially and specifically developed for treating ADHD. If more than one active intervention was reported in a study, the most ADHD specific intervention was analyzed. Intention to treat (ITT) data were analyzed if they were available, alternatively completer data were analyzed.

### Outcome Measures

The following outcomes were included in the analyses: (a) child ADHD symptoms, (b) child conduct problems, and (c) negative parenting; each measured through two separate

informants, parents and independent raters. Child symptoms reported in semi-structured interviews with parents (e.g., Parental Account of Child's Symptoms, PACS; Chen & Taylor, 2006) were coded as parent-reported outcomes. Independent assessment was conducted by raters probably blinded to intervention condition based on naturalistic observations or on observations in experimental setups (e.g., "Fun Park"; Sonuga-Barke et al., 2001). Because only a few studies reported teacher ratings ( $n = 4$ ), they were omitted from the meta-analysis.

### Methodological Quality Assessment

Methodological quality of studies was assessed independently by the first author (MLR) and the third author (HZC) according to the Jadad criteria (Jadad et al., 1996), where studies are coded according to randomization, blinding, and description of withdrawal and dropout. Furthermore, the scoring was adjusted for multiple outcomes following Daley et al. (2014) so that studies with at least one blinded outcome receive a score of 1 on the blinding item. The scores range from 0 to 4, with a score of three indicative of acceptable methodological quality. Disagreements between the first and third author with respect to scores ( $n = 2$ ) were solved through discussion.

### Statistical Strategy

For post-treatment outcomes, effect sizes (ESs) were calculated from the differences between the pre- and post-scores for the intervention and control group standardized by use of change scores. The change score strategy is less sensitive to pre-treatment differences between groups than ESs based on post-scores (Borenstein, Hedges, Higgins, & Rothstein, 2009). Calculation of change score ESs requires, however, knowledge of the pre-post correlation of measures, which is typically not reported in the studies. In case of no information, we imputed a conservative estimate of  $r = .5$  for the pre-post correlation, a value that would make the ES comparable with a standardized mean difference based on post-scores (Borenstein et al., 2009). A Hedges'  $g$  correction was applied to correct for possible bias in small samples (Hedges & Olkin, 1985). In case of several measures within a given outcome category, measures were standardized and averaged. Outcomes at follow-ups were calculated as within-group ESs from post-treatment to follow-up in the intervention group, as most participants receive interventions in the follow-up period. Because independent assessment is rare at follow-up, these analyses were based on parental report alone.

All analyses were conducted within the inverse variance random effects model where individual studies are treated as if they were a random sample from a larger population. This model is generally preferable to a fixed model because

it allows for generalizations (Borenstein et al., 2009). Heterogeneity was examined using  $Q$  and  $I^2$  statistics (Higgins & Thompson, 2002).  $Q$  statistic calculates the probability value for heterogeneity of studies. Significant heterogeneity is indicated by  $p \leq .05$ .  $I^2$  statistic estimates the amount of variance in a pooled ES that can be accounted for by heterogeneity in the sample of studies.  $I^2$  values of 25%, 50%, and 75% indicate low, moderate, and high degrees of heterogeneity, respectively. Publication bias was examined through Eggers' test (Egger, Davey, Schneider, & Minder, 1997). If Eggers' test was significant, indicating risk of publications bias, the Trim and Fill method was applied to impute missing studies to adjust estimates (Duval & Tweedie, 2000). Following the Cochrane guidelines on publication bias, only analyses including more than 10 studies were subjected to Eggers' test (Higgins & Green, 2011).

The association between ES and study methodological quality (i.e., Jadad score) was investigated using meta-regression. The other planned moderator analyses (generic vs. specific programs, group vs. individual treatment, diagnosis vs. rating scale) were conducted by use of meta-ANOVA. All moderator analyses were based on parental reports on ADHD symptoms, since parental reports are most widely used as outcome measures in PT, and ADHD symptoms may be considered a primary outcome in this meta-analysis. However, generic versus specific programs as a moderator was also analyzed on independent assessments of ADHD symptoms.

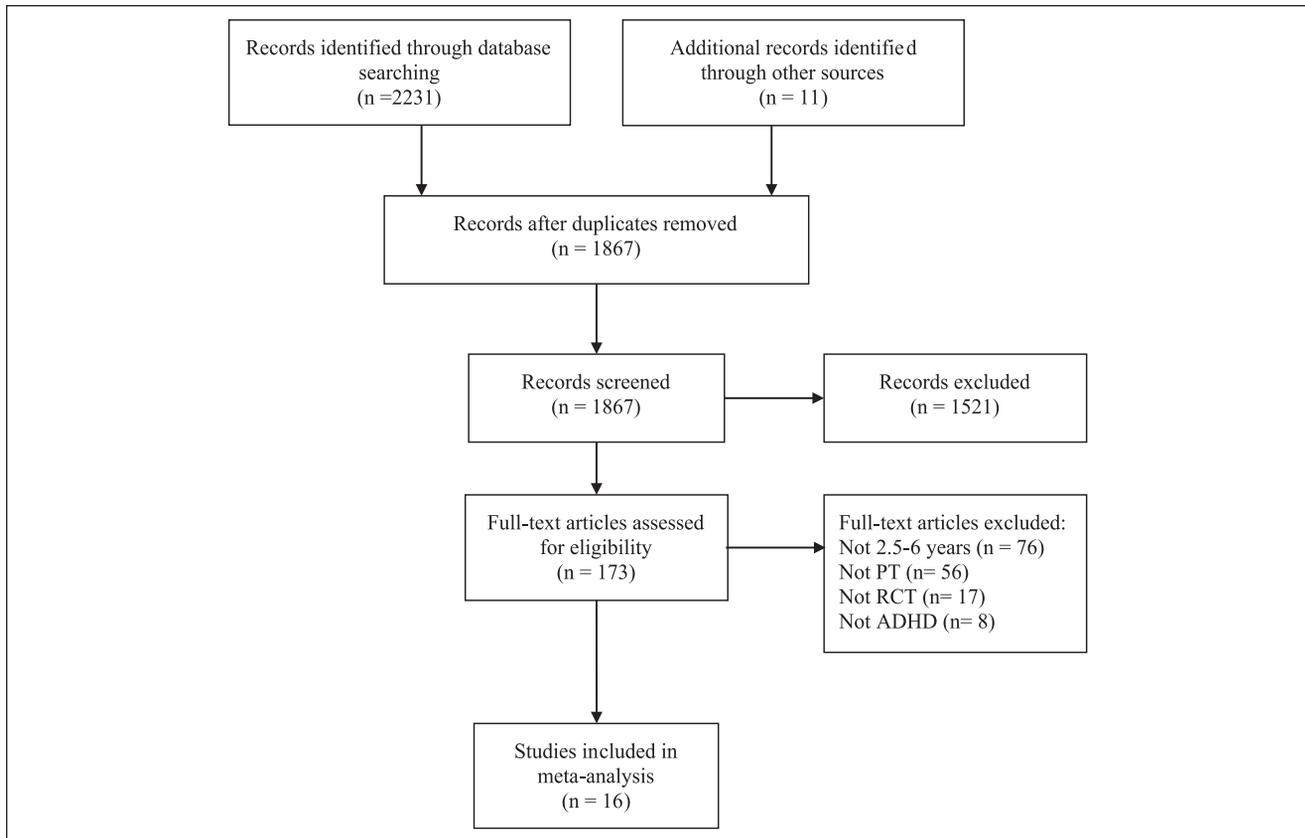
All analyses were conducted using the Comprehensive Meta-Analysis program, version 3.2.00089.

## Results

In total, 16 studies were included in the analysis. The selection process is illustrated in Figure 1.

### Characteristics of Studies

Characteristics of studies are seen in Table 1. The total number of children included in the meta-analysis was 1,003. Study sample sizes ranged from 22 to 149 children, with a mean sample size of 77. In nine studies (Studies 1, 4, 7, 9, 10, 12, 13, 15, 16; see Table 1), children were diagnosed with ADHD; in six studies (2, 3, 5, 6, 8, 14), children had symptoms above clinical cut-off on a validated rating scale. In one study (11), ADHD was measured but was not an inclusion criterion. The two most frequently adopted interventions were the Incredible Years Parent Training Program ( $n = 6$ ; 2, 6, 8, 9, 11, 16) and The New Forest Parent Training Programme ( $n = 4$ ; 1, 12, 13, 15). Most PT programs were delivered in groups ( $n = 9$ ; 2, 3, 5, 6, 8, 9, 10, 11, 16), as compared with individually ( $n = 6$ ; 1, 4, 7, 12, 13, 15); in one case (14), both delivery approaches were applied. Specific programs were delivered in four studies (1, 12, 13,



**Figure 1.** PRISMA flowchart illustrating the search process.

Note. PRISMA = Preferred Reporting Items for Systematic Reviews and Meta-Analyses.

15), all in the form of the Revised New Forest Parenting Programme. Parent-reported measures were most often provided by mothers; only one study (16) presented father-reported data as well. The methodological quality of studies was generally acceptable with a mean Jadad score of 3. For follow-up studies, time points ranged from 3 to 12 months, with five studies (2, 4, 6, 8, 16) reporting 12 months' follow-up, one study (1) reporting 6 months' follow-up, and two studies (10, 12) reporting approximately 3 months' follow-up.

## Outcomes

### Post-intervention

**ADHD symptoms.** The ES of PT on parent-reported ADHD symptoms compared with WL, PLA, or TAU ( $n = 15$ ) was significant and moderate (0.51, CI = [0.33, 0.69],  $p < .001$ ; see Figure 2). Heterogeneity was significant and large ( $Q = 51.5$ ,  $df = 14$ ,  $p < .001$ ,  $I^2 = 72.8$ ). Egger's test (Egger et al., 1997) was insignificant ( $p = .189$ ). The ES of PT on independently assessed ADHD symptoms ( $n = 9$ ) was insignificant and small (0.12, CI = [-0.12, 0.36],  $p = .325$ )

with significant and moderate to large heterogeneity ( $Q = 13.9$ ,  $df = 5$ ,  $p = .016$ ,  $I^2 = 64.2$ ).

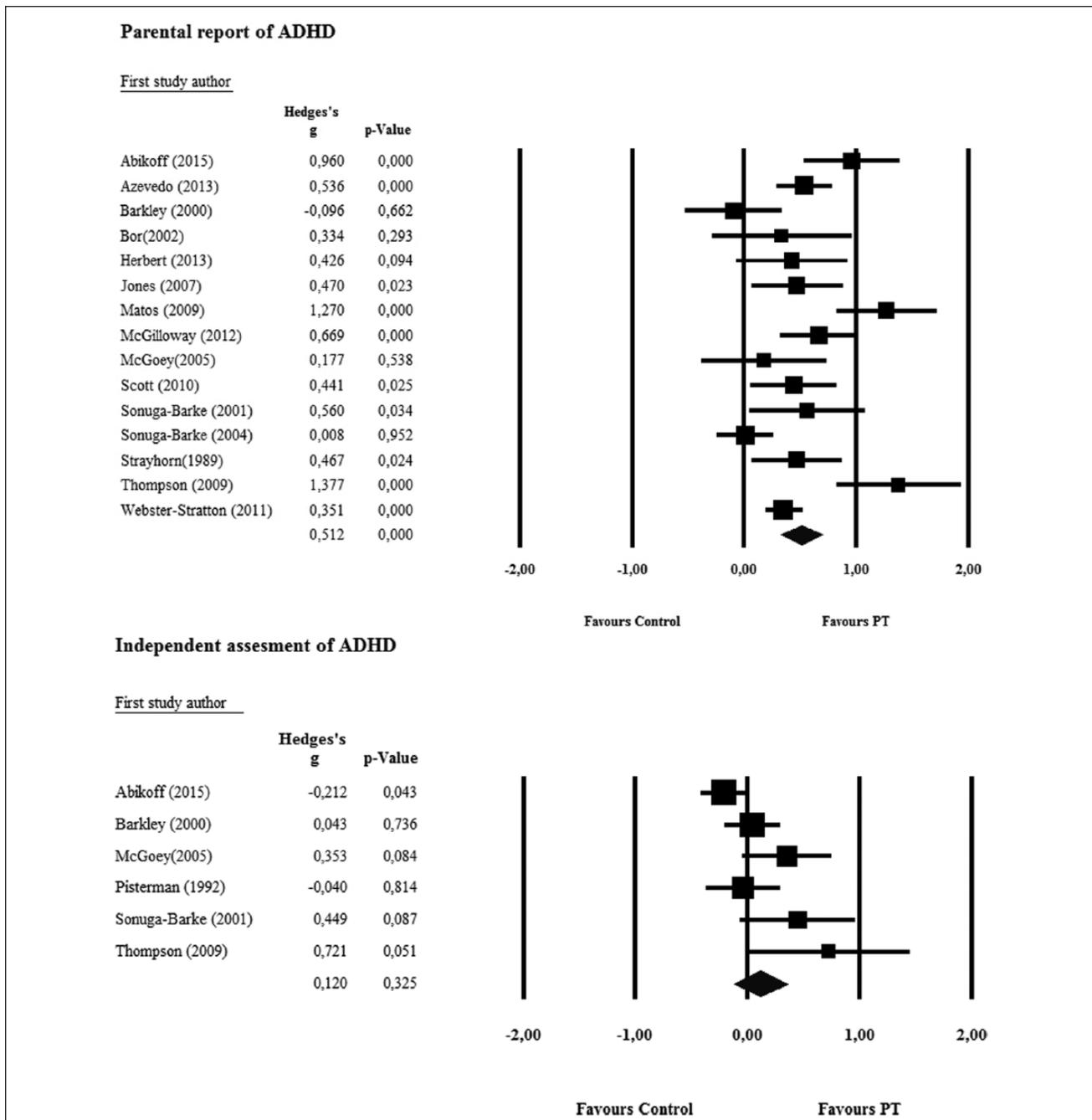
**Conduct problems.** The ES of PT on parent-reported conduct problems ( $n = 13$ ) was small to moderate and significant (0.44, CI = [0.17-0.70],  $p = .001$ ; see Figure 3). Heterogeneity was large and significant ( $Q = 67.1$ ,  $df = 12$ ,  $p < .001$ ,  $I^2 = 82.1$ ). Egger's test was insignificant ( $p = .242$ ). Analysis of independently assessed child conduct problems ( $n = 5$ ) revealed a small and insignificant ES (0.31, CI = [-0.07-0.69],  $p = .117$ ) and heterogeneity was insignificant and small to moderate ( $Q = 8.04$ ,  $df = 4$ ,  $p = .091$ ,  $I^2 = 50.0$ ).

**Negative parenting.** The ES of PT on self-reported negative parental style ( $n = 8$ ) was significant and moderate to large (0.63, CI = [0.32-0.93],  $p < .001$ ; see Figure 4). Heterogeneity was significant and large ( $Q = 28.9$ ,  $df = 7$ ,  $p < .001$ ,  $I^2 = 75.2$ ). The ES on independently assessed negative parental style ( $n = 9$ ) was significant and small (0.33, CI = [0.13-0.53],  $p = .001$ ) and heterogeneity was insignificant and small to moderate ( $Q = 13.8$ ,  $df = 9$ ,  $p = .139$ ,  $I^2 = 34.8$ ).

**Table 1. Characteristics of Included Studies (N = 16).**

Study	n (% girls/M age of child in years)	Intervention (modus)	Control condition	Duration of intervention in weeks/FU in months	ADHD measure (informant)	Oppositional and conduct measure (informant)	Negative parenting measure (informant)	Jadad score
(1) Abikoff et al. (2015)	101 (26.4/3.57)	The Revised New Forest Parenting Programme (I)	WL	8/6	CPRS (P,T,C) PLAY PARK (P)	NYPRS (P) NYTRS (T)	PPI (P) GIPCI (R)	4
(2) Azevedo, Seabra-Santos, Gaspar, and Homem (2013, 2014)	100 (28/4.65)	Incredible Years (G)	WL	14/12	WWP (P) PKSB (P, T) PACS (P)	PKBS (P, T) PACS (P)	PPS (P)	2
(3) Barkley et al. (2000)	81 (36/N/A)	Defiant Children (G)	NTCG	10	CBCL (P, T) CPT (R)	CBCL (P,T)	PPS (P)	3
(4) Bor, Sanders, and Markie-Dadds (2002)	58 (32/3.47)	Triple-P-enhanced (I)	WL	12/12	ECBI (P)	ECBI (P)	FOS-RII (R) PS (P)	3
(5) Herbert, Harvey, Roberts, Wichowski, and Lugo-Candelas (2013)	31 (24.8/4.56)	Parenting Your Hyperactive Preschooler (G)	WL	14	DBRS-I, (P) DBRS-h-i (P)	DBRS-odd (P)	Naturalistic observation (R) PS (P)	4
(6) Jones Daley, Hutchings, Bywater, and Eames (2007)/ Jones Daley, Hutchings, Bywater, and Eames (2008)	79 (32/3.86)	Incredible Years (G)	WL	12/12	CPRS (P)	N/A	N/A	4
(7) Matos, Bauermeister, and Bernal (2009)	22 (N/A/N/A)	Parent-Child Interaction Training Programme (I)	WL	Flexible, max 17.	BASC-PRS-h (P) DBRS-h (P) CPRS (P)	DBRS-odd (P) ECBI-problem (P) ECBI (P)	PPI (P)	1
(8) McGilloway et al. (2012)/ McGilloway et al. (2014)	149 (49/4.76)	Incredible Years (G)	WL	14/12	CPRS (P)	ECBI (P)	DIPCS (R)	4
(9) McGoey, DuPaul, Eckert, Volpe, and Van Brakle (2005)	57 (14/4.0)	Incredible Years + Teachers Training (G)	CCG	21	PDT (R) PSA (R) PKBS-o/i (P,T)	PDT (R) PSA (R) PKBS-o/a (P,T)	PDT (R) PSA (R)	2
(10) Pieterman et al. (1992)	45 (19/4.1)	Group Parent Training Programme for Compliance (G)	WL	12/3	PDT (R)	PDT (R)	PDT (R)	3
(11) Scott et al. (2010)	112 (29.5/5.21)	Incredible Years (G)	HL	28	PACS (P)	PACS (P)	EE (R)	4
(12) Sonuga-Barke, Daley, Thompson, Laver- Bradbury, and Weeks (2001)	78 (38.5/N/A)	New Forest Parent Training Programme (I)	PACS	8/3.5	PACS (P) FF (R)	ECBI (P) PACS (P)	PPI-Neg (P) N/A	3
(13) Sonuga-Barke, Thompson, Daley, and Laver- Bradbury (2004)	89 (N/A)	The Revised New Forest Parent Training Programme (I)	WL	8	PACS (P) WWP (P)	BCL (P)	N/A	2
(14) Stryhorn and Weidman (1989)	89 (56/3.75)	Training Exercises for Parents of Preschoolers (I + G)	NTCG	NR	BPB (P, T) DSM-III (P)	BPB (P,T) DSM-III (P)	PPS (P)	2
(15) Thompson et al. (2009)	41 (26.8/4.26)	New Forest Parent Training Programme (I)	TAU	8/1.5	WWP (P) PACS (P) FY (R)	N/A	EE-neg (R)	4
(16) Webster-Stratton, Reid, and Beauchaine (2011)/ Webster-Stratton, Reid, and Beauchaine (2013)	99 (25/5.23)	Incredible Years (G) + Incredible Years Dinosaur Child Training (G)	WL	20/12	CPRS (P,T)	ECBI (P,T)	DIPCS (R)	3

Note: FU = follow-up; I = individually administered program; CPRS = Conners' Rating Scale-Revised; P = parent rated; T = teacher rated; C = clinician rating; NYPRS/NYTRS = New York Rating Scale Parent or Teacher; PPI = Parenting Practices Interview; GIPCI = Global Impressions of Parent-Child Interactions-Revised; G = group administered program; R = observational rating; WWP = Werry-Weiss-Peters Activity Scale; PACS = Parental Account of Child's Symptoms; ; DIPCS = Dyadic Parent-Child Interaction Coding System; PS = Parenting Scale; CBCL = Child Behaviour Checklist; CPT = Continuous Performance Test; PPS = Parenting Practices Scale; ECBI = Eyberg Child Behaviour Inventory; FOS-RII = Revised Family Observation Schedule; DBRS = Disruptive Behaviour Inattention subscale; DBRS-i-h = Disruptive Behaviour Inattention-Hyperactivity subscale; DBRS-odd = Oppositionality Subscale; PDT = Parent Directed Task; PSA = Parent Supervised Activity; PKBS = Preschool and Kindergarten Behavior Scales Overactivity/Inattention subscale; PKBS-o/a = Preschool and Kindergarten Behavior Scales Oppositional/Aggressive subscale; EE = expressed emotions; PPI-Neg = Parental Practices inventory negativity subscale; BPB = Behaviour Checklist hyperactivity factor; N/A = not available; BPB = Behav Preschool Behaviour; DSM-III-R = parent ratings on *Diagnostic and Statistical Manual of Mental Disorders* (3rd ed., rev.; DSM-III-R; American Psychiatric Association, 1987); FY = FarmYard Observation; EE-neg = Expressed emotions negativity subscale; WL = Waitlist; NTCG = No Treatment Control Group; CCG = Community Control Group; HL = Helpline; BASC-PRS-h = Hyperactivity and Aggression Subscales of the Behavioral Assessment System for Childre-Parent Rating Scale.



**Figure 2.** Parental and independent assessment of ADHD.

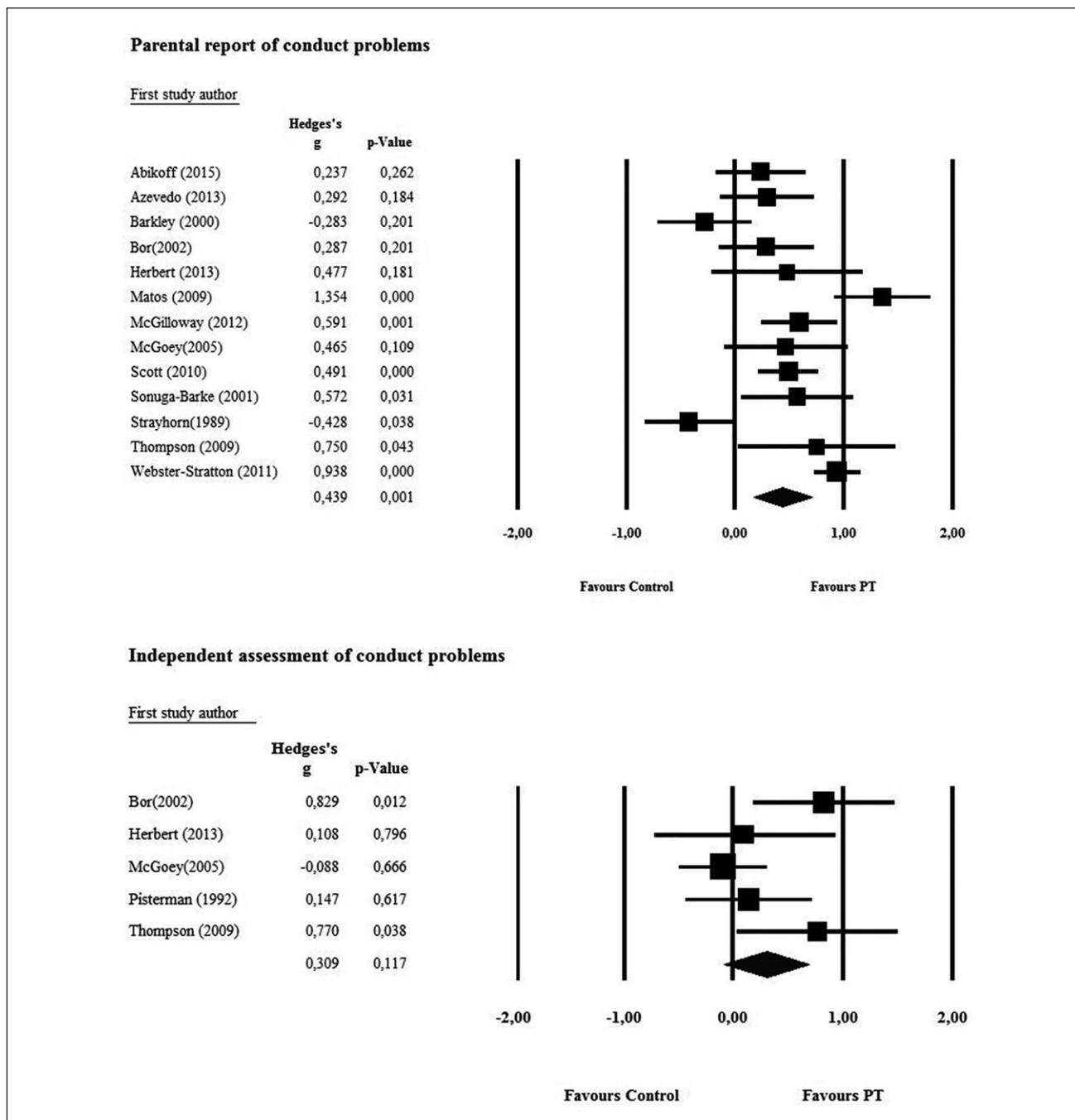
#### Follow-up

**ADHD symptoms.** The ES of PT on parent-rated ADHD symptoms from post-treatment to follow-up ( $n = 8$ ) was slightly positive, and marginally significant (0.07, CI =  $[-0.01, 0.15]$ ,  $p = .059$ ). There was no indication of heterogeneity ( $Q = 4.9$ ,  $df = 7$ ,  $p = .664$ ,  $I^2 = 0.0$ ).

**Conduct problems.** The ES of PT on parent-rated conduct problems from post-treatment to follow-up ( $n = 8$ ) was

slightly positive, but insignificant (0.07, CI =  $[0.01, 0.15]$ ,  $p = .103$ ). There was no indication of heterogeneity ( $Q = 1.1$ ,  $df = 7$ ,  $p = .98$ ,  $I^2 = 0.0$ ).

**Negative parenting.** The ES of PT on parent-rated negative parenting from post-treatment to follow-up ( $n = 5$ ) was small, but positive and marginally significant (0.12, CI =  $[-0.01, 0.24]$ ,  $p = .059$ ). There was no indication of heterogeneity ( $Q = 1.1$ ,  $df = 4$ ,  $p = .895$ ,  $I^2 = 0.0$ ).



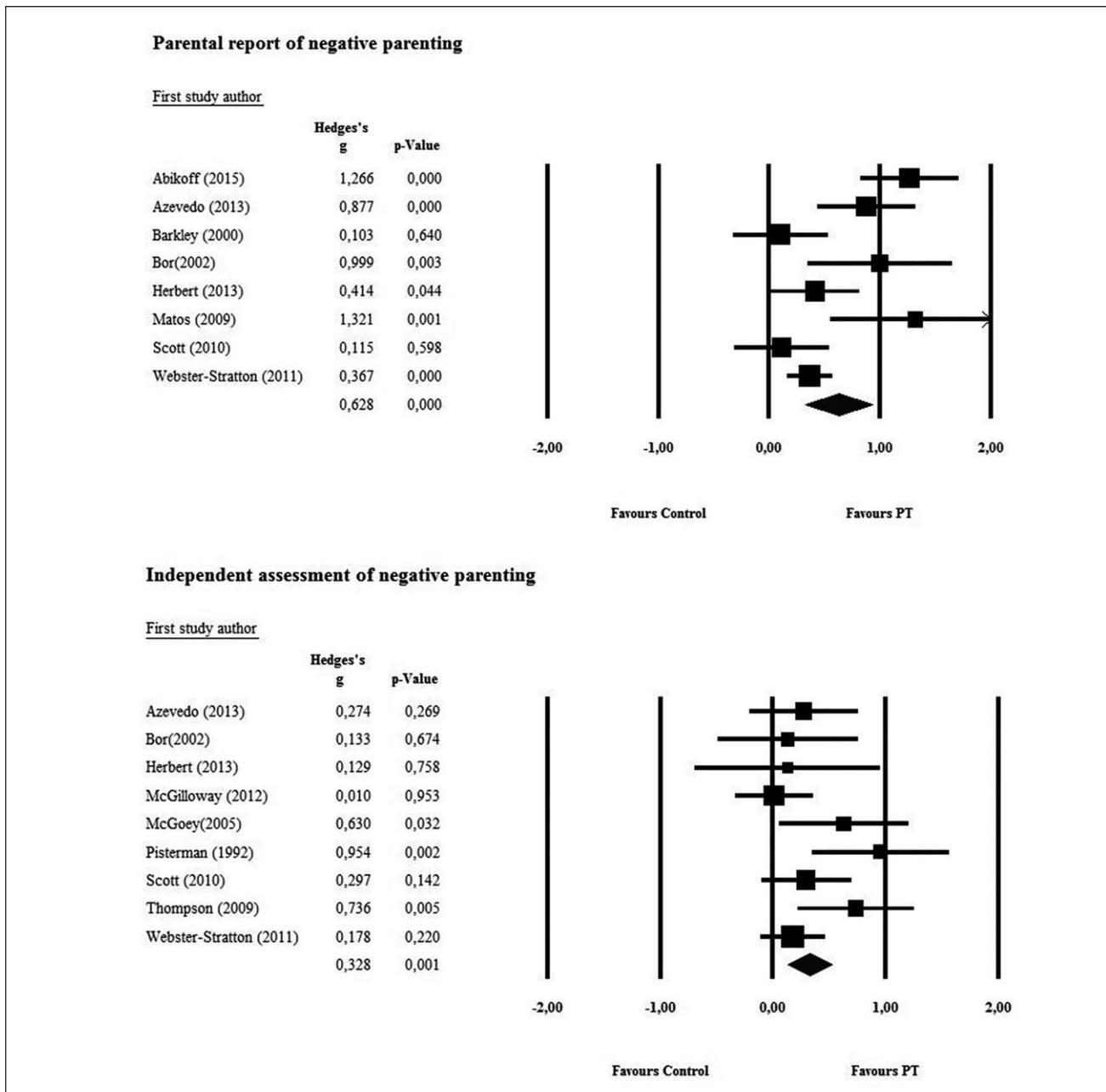
**Figure 3.** Parental and independent assessment of conduct problems.

**Moderators**

*Study quality.* No significant association between the Jadad score and effect on parental reported ADHD symptoms was found ( $\beta = .12, p = .320$ ).

*Specific vs. generic programs.* There was no significant difference in parent-reported reduction in ADHD symptoms ( $Q = 0.697, df = 1, p = .404$ ) between the four studies on

specific PT programs (ES = 0.65, CI = [0.27, 1.04],  $p < .001$ ) and the 11 studies applying generic PT programs (ES = 0.47, CI = [0.24, 0.69],  $p < .001$ ). Furthermore, there was no significant difference on independently assessed reduction in ADHD symptoms ( $Q = 0.06, df = 1, p = .803$ ) between the specific programs (ES = 0.18, CI = [-0.23, 0.58],  $p = .398$ ) and the generic programs (ES = 0.11, CI = [-0.26, 0.48],  $p = .569$ ).



**Figure 4.** Parental and independent assesment of negative parenting.

**Modality of intervention.** There was no significant difference in parent-reported ADHD symptoms on delivery modus ( $Q = 1.0$ ,  $df = 1$ ,  $p = .320$ ) between the eight interventions being delivered in a group ( $ES = 0.38$ ,  $CI = [0.12, 0.65]$ ,  $p = .004$ ) and the six interventions delivered individually ( $ES = 0.72$ ,  $CI = [0.40, 1.94]$ ,  $p < .001$ ).

**Diagnostic status.** When analyzing efficacy of PT on parent-reported ADHD symptoms, there was no significant difference ( $Q = 0.76$ ,  $df = 1$ ,  $p = .385$ ) between the

eight studies with children diagnosed with ADHD ( $ES = 0.60$ ,  $CI = [0.32, 0.71]$ ,  $p < .001$ ) compared with the seven studies with children with symptoms above clinical cut-off on a validated rating scale ( $ES = 0.43$ ,  $CI = [0.41, 0.71]$ ,  $p = .003$ ).

## Discussion

The present meta-analysis partially supports the effectiveness of PT for preschool children with ADHD or ADHD

symptoms and a probable risk of developing ADHD. The ESs based on parental reports were in the moderate range (0.44-0.63), comparable with ESs often reported for psychological interventions for psychiatric disorders (Lambert, 2013). An ES = 0.51 for parent-reported ADHD symptoms could be estimated to correspond to a difference between PT and control conditions of 25% units (Rosenthal & Rubin, 1982), so that, for instance, 62.5% children in PT achieved favorable outcome, compared with 37.5% in the control conditions. The parental reported outcomes were stable at follow-ups of 3 to 12 months. ESs based on independent assessments were, however, all small, and only significant for negative parenting. Independently assessed ADHD symptoms achieved a statistically insignificant ES of 0.12.

Our results for parental reported ADHD symptoms are in line with Mulqueen et al.'s (2015) meta-analysis on PT for ADHD preschoolers. Their meta-analysis was conducted on parent-reported ADHD symptoms in studies with clinician provided diagnoses with an ES of 0.61, almost exactly the same as the ES found for diagnosed children in our study (0.60; there was an overlap of six studies).

ESs on outcome provided by most proximal assessors in Daley et al.'s (2014) meta-analysis on behavioral interventions for children aged 3 to 18 years were in line with our parent-reported ES for negative parenting (0.57 vs. 0.63), but numerically (although not significantly) smaller than ours for ADHD symptoms (0.35 vs. 0.51) and conduct problems (0.26 vs. 0.44). Daley et al. (2014) found age to moderate ESs for their most proximal assessment with larger ESs for younger children. Most proximal outcomes in their study could include both assessment by parents and by involved teachers, but in case both were reported, parent ratings were preferred. Similar to our study, Daley et al. found significant results for independently assessed negative parenting but not for ADHD symptoms. Differently, they found a significant effect for independently assessed conduct problems, although their ES (0.31) is similar to ours (0.31). In contrast to Daley et al. (2014), we did not include teachers as independent assessors, because the teachers' blinded status of treatment condition can be uncertain. Differences between the studies may reflect the broader range of behavioral interventions and ages of children included (3-18 vs. 2.5-6) in Daley et al. (2014), as well as, of course, differences in sample sizes influencing significance of results.

The current meta-analysis found consistent effect of PT on negative parenting across assessment perspectives, although the ES was numerically smaller when rated by independent assessors (0.33 vs. 0.63). Parents are the direct recipients of PT, and therefore, changes in parenting should be considered necessary if changes in off-spring behavior are to occur. Hinshaw et al. (2000) found that a reduction in negative parenting partially mediated the effect of PT on

children in the Multimodal Treatment Studies of ADHD (MTA). Although there is no evidence of parenting practices as fundamental causes of ADHD (Barkley, 2014; Johnston & Mash, 2001), harsh and negative parenting has been hypothesized to maintain or exacerbate child ADHD and conduct problems in a developmental psychopathology perspective (Mash & Johnston, 2005).

Because all moderator analyses (study methodological quality, specific vs. generic programs, individual vs. group delivered interventions, ADHD diagnosis vs. symptoms on rating scales) were insignificant, they could not assist in clarifying the high levels of heterogeneity in most analyses. Although the small number of studies limits the power of these analyses, differences in pairwise comparisons only in one case exceeded a small ES of 0.20; individually conducted interventions achieved a numerical higher ES than group-based interventions (0.72 vs. 0.38).

No differences were found between specific and generic programs on either parent-reported or independently assessed ADHD symptoms; both types of programs achieved small and insignificant ESs based on independent assessment (0.18 vs. 0.11). There was overlap between this analysis and the analysis of modality, because the specific program, in all cases, was individually delivered, while generic programs in most cases were group based. It should be noted that the small number of studies in the moderator analyses warrants caution in the interpretations of these results.

The discrepancy between parental and independently rated assessment of child ADHD symptoms revealed in the present meta-analysis was also found by Sonuga-Barke et al. (2013) and Daley et al. (2014). It raises the question, whether parental reports or independent assessment most adequately reflect the results on ADHD symptoms. On one hand, parents of preschool children may be the most ecologically valid assessors of children's symptoms, because they observe them across different contexts during extensive amounts of time. This stands in contrast to the generally brief independent assessment periods in the PT studies, ranging from 5 to 30 min, and often conducted in laboratory settings. Consequently, PT could have an effect on ADHD symptoms, albeit one that is difficult to measure objectively. On the other hand, and as previously discussed by Zwi, Joens, Thorgaard, York, and Dennis (2011) and Sonuga-Barke et al. (2013), parent-reported outcomes in PT could be inflated due to satisfaction with treatment or the parents' active role in the treatment of their children. If independent assessment is considered most valid, the lack of efficacy could suggest that ADHD symptoms are difficult to change by psychological means, possibly because they are rooted in a neurological dysfunction. A similar discrepancy between proximal ratings and independent assessment has been found in studies on cognitive training for ADHD (Cortese et al., 2015).

The stable parent-reported outcomes from post-intervention to follow-up may indicate that results are not just temporary “honey-moon effects” (LaBarbera & Dozier, 1985) due to immediate satisfaction with treatment or gratitude to therapists. Furthermore, the sustained efficacy after termination of the intervention may point to an advantage of PT over for instance medication.

The findings in this meta-analysis on preschool PT are generally in line with those of Daley et al. (2014) of behavioral interventions for children aged 3 to 18. Daley et al. (2014) underlined the broad range of beneficial effects of behavioral interventions on families with ADHD children including more efficient parenting and reduction in children’s conduct problems, problems that may seriously stress families. Our study also found consistent evidence across rater perspectives for such beneficial effects, although results for independently assessed conduct problems only bordered on significance, maybe due to insignificant power (ESs were quite similar in the two studies).

The proposal that ADHD may be most efficiently treated in the preschool years was not directly supported in our study, since results were similar to those of behavioral interventions for a broader age span in Daley et al. (2014). Parent-rated outcomes were, however, numerically higher in our meta-analysis. It could be argued that an efficacious intervention should be provided as early as possible to prevent further distress. The finding of parent-reported stable positive outcomes in follow-up periods of up to 1 year also points to the benefits of early PT for ADHD. Longer follow-up periods are necessary to investigate possible long-term effects of such interventions in the prevention of negative developmental paths with further disabilities and comorbid ODD or CD, although it could be difficult to conduct such studies without adequate control conditions.

This meta-analysis has some limitations. First, there are relatively few PT studies. The high degree of heterogeneity among the studies could not be explained through the moderation analyses, maybe due to low statistical power. Moreover, the follow-up analyses could not be based on between-group analyses and were conducted on parent rating alone with 1 year constituting the longest follow-up period. Finally, no grey/unpublished literature was included in the review, which could possibly have excluded any unpublished studies revealing little or no efficacy.

## Conclusion

Taken together, the present meta-analysis partially supports PT as treatment for preschool children with ADHD with moderate parent-reported ESs for ADHD symptoms, conduct problems, and negative parenting. These results were stable at follow-ups of 3 to 12 months. For independently assessed outcomes, a significant result was found for negative parenting, while conduct problems bordered on

significance. In line with previous meta-analyses, there was no effect of PT on independently assessed ADHD symptoms. Early PT interventions may be beneficial for family functioning and child conduct problems, but thus far, there is no objective documentation of an effect on ADHD symptoms. Further studies are needed to investigate study heterogeneity and the possible long-term preventive effects of early PT interventions on negative developmental trajectories.

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## **Appendix B**

**Paper 2:** Rimestad, M.L., Trillingsgaard, T. & Hougaard, E. Combining parent and teacher training for early ADHD: a randomized study of effectiveness.

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**COMBINING PARENT AND TEACHER TRAINING FOR EARLY ADHD: A RANDOMIZED STUDY  
OF EFFECTIVENESS**

**Abstract**

*Parent training (PT) has been documented as an effective intervention for early ADHD based on parental assessment, but documentation of transfer of its results outside the family context has been limited. Due to the cross-contextual impairments typically caused by ADHD, it has been proposed that PT in combination with an intervention in kindergarten or school based on systematic teacher training (TT) would be better suited. The objective of this study was to evaluate the effectiveness of a combination of PT and TT in a community sample of children with early ADHD problems. Families of 64 3-8-year old children were randomized to either PT alone with “The Incredible Years” PT or to a combined PT + TT intervention. Moderate to large effect sizes on mother-reported outcomes were revealed, in line with those generally reported in the literature on PT interventions for early ADHD. The study found no statistically significant differences between the two groups on any of the outcome measures rated by parents or teachers. There was, however, a non-significant trend favoring PT + TT on teacher-reported conduct problems, possibly indicating a context-specific effect of TT at school/kindergarten. In conclusion, the study found no support for better results of a combined PT + TT intervention for early ADHD problems compared to PT alone, although there were insignificant indications of context-dependent improved results at school for the combined PT + TT group.*

**Keywords:** *early intervention, parent training, attention deficit hyperactivity disorder, teacher training, Incredible Years.*

Attention Deficit Hyperactivity Disorder (ADHD) is a heterogeneous neurodevelopmental disorder characterized by age inappropriate, impairing patterns of inattention, hyperactivity and impulsivity (American Psychiatric Association, 2013). The worldwide prevalence of ADHD is estimated to be around 3-5% (Polanczyk, Silva de Lima, Horta, Biederman & Rohde, 2007) and it is among the most commonly diagnosed disorders in childhood. Children with ADHD often suffer from social skills deficits (Landau & Moore, 1991), emotional dysregulation (Shaw, Stringaris, Nigg & Leibenluft, 2014), peer rejection (Bagwell, Molina, Pelham & Hoza, 2001), and academic difficulties (DuPaul & Stoner, 2014). ADHD takes its toll on families, too, and families with children with ADHD are often suffering from dysfunctional interaction patterns (Daley, Sonuga-Barke & Thompson, 2003) as well as high levels of family stress and conflict (Deault, 2010; Johnston & Mash, 2001). Research points to a link between the stressful and challenging nature of child ADHD and parental negativity and inconsistency as an evoked consequence, possibly causing exacerbation in child ADHD difficulties and development of secondary behavioral difficulties, as oppositional defiance disorder (ODD) or conduct disorder (CD) (Hinshaw, Owens, Wells, Kraemer, Abikoff, Arnold . . . Wigal, 2000; Johnston and Mash, 2001; Mash & Johnston, 2005). Consequently, there is an obvious need for an early intervention for families with children suffering from ADHD.

Parent Training (PT) is an intervention where parents are trained to manage problematic child behavior, most often based on behavioral contingency principles. Acknowledging parents as the most significant agents in child development, PT includes them as active participants and co-therapists in the management of their child's difficulties. PT aims to lower negative and heighten positive parent-child interaction and parents are taught to structure daily routines in a predictable manner to make them more manageable for the child. Recent meta-analyses have documented efficacy of PT for ADHD in youth (3-18 years; Daley et al., 2014) as well as in preschoolers (2.5-6 years; Rimestad, Lambek, Christiansen & Hougaard, 2016), although not on blinded assessment of ADHD symptoms. Some PT programs, such as the Incredible Years (IY; Webster-Stratton, Reid & Beauchaine, 2011), have been developed from ODD/CD PT programs, and some have specifically been developed for ADHD, such as the Revised New Forest Parenting Program (Thompson et al., 2009). Rimestad and colleagues (2016) did not find differences in outcomes between PT programs originally developed for ODD/CD and programs specifically developed for ADHD.

The IY ® PT Program is a group-based program for parents of young children (2-8 years) developed by Carolyn Webster-Stratton and co-workers at the University of Washington (Webster-Stratton, 2011; Webster-Stratton & Reid, 2010). As mentioned, the IY was originally intended for children with ODD/CD but it has later been adapted for ADHD (Webster-Stratton, Beauchaine and Reid, 2011). The IY is well documented as efficacious for children with disruptive disorders (Menting, Oribio de Castro & Matthys, 2013) but it has also been found efficacious for ADHD in a handful of studies (Azevedo, Seabra-Santos, Gaspar & Homem, 2013; Jones, Daley, Hutchings, Bywater & Eames, 2007; McGilloway et al., 2012; Webster-Stratton et al., 2011). A benchmark study of a Danish version of the IY conducted at the same clinic as the present study (Trillingsgaard, Trillingsgaard & Webster-Stratton, 2014) found effect sizes in line with those reported in an American RCT on IY (Webster-Stratton et al., 2011).

However, an important point has been made regarding the limited transfer of outcome of PT from the home setting to other contexts (Rajwan, Chacko & Moeller, 2012). PT programs may therefore have little or no visible effects in a preschool or school setting. Since ADHD is characterized by impairment across different contexts (APA, 2013), children with ADHD will display difficulties outside the home, notably in preschool/school settings (Pffiffner & DuPaul, 2014). Many school and kindergarten teachers have difficulties with structuring their teaching in ways that are sensibly adapted to children with ADHD and behavioral difficulties (Arcia, Frank, Sanchez-LaCay & Fernandez, 2000). Teacher Training (TT) and classroom intervention in a school age population are considered well-established interventions for ADHD (DuPaul, Eckert & Vilaro, 2012; Pelham & Fabiano, 2008). Incredible Years Teacher Classroom Management (Webster-Stratton, 2011) is a part of the IY training series, though primarily aimed at strengthening school readiness and improving classroom atmosphere. Recognizing that ADHD causes difficulties in academic functioning as well as peer relations, it may be important to empower teachers with knowledge of and tools to manage ADHD difficulties in the school or kindergarten (DuPaul and Stoner, 2014; Tarver, Daley & Sayal, 2015).

Based on such considerations, it has been suggested that an intervention addressing child difficulties both at home and at school would be more effective than an intervention in only one of these settings (Pffiffner, Villodas, Kaiser, Rooney & McBurnett, 2013). PT at home may be especially suitable to help with problems in the family, while TT could possibly ameliorate some of the difficulties children experience at school, such as conduct problems here, as well as peer rejection and academic failure with consequently lowered self-esteem (McQuade & Hoza, 2014). PT and TT could thus be speculated to supplement each other with outcomes

primarily in different contexts, or perhaps even to synergize with a mutual intensification of outcomes at both places.

There are rather few studies addressing the combination of PT and TT for early ADHD. Östberg and Rydell (2012) found that combining PT and TT in a school age population (age 7-10 years) resulted in reduced ADHD symptoms compared to a no-intervention control group. Corkum, McKinnon and Mullane (2005) combined a PT intervention with a low-intensity TT (weekly mails with educational guidelines) in a preschool setting. The combined PT +TT intervention had better outcomes than PT alone. In both of the above-mentioned studies, parent- and teacher-ratings were combined into composite scores, which precludes analysis of possible differential outcomes of the interventions at home and in school. Kern and colleagues (2007) compared a 20 session group-based PT program with the PT program combined with individualized interventions, both at home and in school. Both groups revealed improvement, with no significant between-group differences. McGoe, DuPaul, Eckert, Volpe & Van Brakle (2005) compared a combined IY PT program and TT for 3-5 year old preschoolers with or at risk of ADHD to a group receiving treatment as usual, finding no significant differences in effect between the two groups at post intervention. Thus, there are plausible arguments for a combination of PT and TT as intervention for ADHD, but findings from previous studies are mixed and inconclusive.

The aim of this study is to evaluate the effectiveness of a combination of IY PT and a TT program specifically developed at the clinic where the study took place. We hypothesized, that children who received both PT and TT would have better outcomes; especially in the school or kindergarten context where the TT was delivered.

## **Methods**

All participants signed a consent form after they were given oral and written information on the study. The study was conducted in compliance with standards from the regional ethical committee and approved by the Danish Data Protection Agency.

### *Participants*

Participants were parents of children self-referred to the Centre for ADHD, where the study took place. The Centre for ADHD is a non-profit private clinic funded by Edith and Godtfred Kirk Christiansen's Foundation, a Danish private foundation supporting several family-oriented projects for common welfare. The Centre for ADHD is situated in the city of Aarhus, Denmark, providing parent-involved treatment for early ADHD difficulties. The aim of the Centre is to provide early treatment to families with young children with or at risk

for ADHD based on the IY PT program. Children receiving treatment must have ADHD symptoms, but a formal ADHD diagnosis is not required for the family to receive treatment at the clinic. Parents self-refer to the center, and families are included on the basis of an overall clinical assessment, based on a clinical semi-structured interview with the family as well as validated questionnaires (see below). The intervention was free of charge, but parents paid 600 DKKR for the IY PT book and handouts. The intervention was announced on the clinic website as well as in flyers distributed in health clinics in the local area.

In accordance with the inclusion criteria at the Centre for ADHD, families in the study had to have a child between 3 and 8 years with ADHD symptoms, but not necessarily with a formal ADHD diagnosis. Moreover, for inclusion in this study it was decided that families should not have received prior PT. In total, 64 children were included.

### *Measures*

*Demographic questionnaire:* This questionnaire, specifically constructed for the study, consisted of 26 questions regarding background information on the child, medication status, possible prior diagnosis, family structure, parental marital status and level of education.

*Conners' 3 ADHD Index Parent Rating Scale (CPRS; Conners, 2009).* The CPRS is a measure of frequency of ADHD symptoms. It is a questionnaire to be filled out by parents with 10 items, each rated on a 4-point Likert-scale from 0 = *never or almost never* to 3 = *Very often or always*. The CPRS is a short version of the full form of the Conners' ADHD Rating scales, including the items that most effectively differentiated youth with ADHD from nonclinical controls (Westerlund, Ek, Holmberg, Näswall & Fernel, 2009). The scale has been found to have acceptable psychometric properties, with a Cronbach's  $\alpha$  of .90 (Westerlund et al., 2009). In the present study, Cronbach's  $\alpha$  was .76 at pre-treatment.

*Conners' 3 ADHD Index Teacher Rating Scale (CTRS; Conners, 2009).* The CTRS is the teacher version of the CPRS, measuring teacher-reported ADHD symptoms. The scale has demonstrated acceptable psychometric properties, with a Cronbach's  $\alpha$  of 0.94 (Westerlund et al, 2009). In the present study, Cronbach's  $\alpha$  was .96 at pre-treatment.

*Eyberg Child Behavior Checklist (ECBI; Eyberg & Pincus, 1999).* The ECBI is a questionnaire assessing child ODD/CD symptoms. The questionnaire consists of two subscales, an Intensity Scale and a Problem Scale. The Intensity Scale has 36-items concerning frequency of opposition and conduct problems, with each item rated from 1 = *Never* to 7 = *Always*. The Problem Scale is a categorical scale where each of the 36 problem behaviors is scored as to whether it poses a problem to the parent (1= *Yes*; 0 = *No*). In this study, only the intensity scale

was used. The scale has been found to have acceptable psychometric properties with a Cronbach's alpha of .93 (Reedtz, Bertelsen, Lurie, Handegård, Clifford & Mørch, 2008). In this study, Cronbach's alpha was .86 at pre-treatment.

*Sutter-Eyberg Student Behavior Inventory-Revised (SESBI-R (Eyberg & Pincus, 1999).*

The SESBI-R is a teacher version of the ECBI with 38 items, measuring teacher-rated ODD and CD. It has been found to have acceptable psychometric properties, with a Cronbach's alpha of .96 (Kirkhaug, Drugli, Mørch & Handegård, 2012). In the present study, Cronbach's alpha was .94 at pre-treatment.

*Strengths and Difficulties Questionnaire (SDQ) (Goodman, 1997).* The SDQ measures child difficulties and strengths within different behavioral domains. The SDQ has 34 items, each scored from 0 = *Not true*, 1 = *Somewhat True* or 2 = *Certainly true*. It can be filled out by parents, teachers or older (> 11 years) children. The scale has four subscales on conduct problems, hyperactivity/inattention, emotional difficulties, and social competences. Two broad symptom dimensions, internalizing and externalizing difficulties, can be calculated from the scale (Goodman, Lamping & Ploubidis, 2010), and these dimensions were applied in the study. The scale has been found to have acceptable psychometric properties in a Scandinavian population, with a mean alpha of 0.73 (Malmberg, Rydell & Smedje, 2003). In the present study, Cronbach's  $\alpha$  in the parent version was .67 on the internalizing subscale pre-treatment, and .74 on the externalizing subscale; in the teacher version, the respective  $\alpha$ -values were .62, and .79.

*Experiences of Service Questionnaire (ESQ; Attride-Stirling, 2002).* The ESQ assesses degree of participant satisfaction with the intervention. The scale is a 10-items questionnaire with three answer categories for each item: 1 = *not true*; 2 = *partly true*; 3 = *certainly true*. Parents filled out the questionnaire no later than four weeks after termination of the intervention. In the present study, the scale had an internal consistency of  $\alpha = .79$ .

*Treatment satisfaction questionnaire:* This questionnaire was developed by Centre for ADHD to assess the teacher satisfaction with the intervention. The questionnaire consisted of 9 items, with answer ranging from 1 to 7, with higher ratings indicating more satisfaction.

### *Procedures*

After the families had contacted the Centre, a clinical psychologist screened them for suitability via telephone. If eligible, two clinical psychologists arranged a home-based intake interview with the parents on the child's problem behavior as well as on aspects of their family life, and the parents filled out the CPRS, the ECBI, and the SDQ as well as the demographic questionnaire. Based on the information from the interview, it was decided if the families should be offered treatment at a following conference at the Centre. All families offered treatment

were informed about the study and asked for permission to contact their child's kindergarten or school. If the parents had given informed consent and the kindergarten or school accepted to participate in the study, families were randomized to either PT combined with TT or PT alone. The first author (MLR) conducted randomization by use of a computerized random number generator, and results of the randomization were hidden from the Centre until start of the treatment. If two children came from the same school or kindergarten, they were randomized together in order to avoid teacher obligation to respond differently to the children. Both mothers and fathers were encouraged to participate in the PT course, regardless of their marital status. 64 mothers and 45 fathers participated in the courses. Childcare facilities were arranged for the children (including siblings) during the PT intervention and the families were provided with free meals.

The two teachers primarily associated with the child were invited to participate in the teacher intervention. All expenses of the interventions were covered by Centre for ADHD.

Data were collected at three points; pre, post and follow-up (FU). Pre-test scores from parents were obtained no later than 2 weeks before PT start. Teachers' pre-test scores were obtained within a week before PT start and no later than 2 weeks after. The two interventions ran parallel time-wise for approximately five months, with one session of PT every week and 4 weeks in between the teachers training sessions. Post measures were obtained no later than four weeks after termination of the interventions. FU measures were obtained six months after the end of the program. Parents and teachers received a LEGO box (sponsored by LEGO ®; value about 600 DKKR) after completing the last FU measurement.

Parent-rated pre-measures were collected in paper form; all other data were collected via email using an online data collection platform. In case parents did not respond to the questionnaires, they were reminded four times in total; two times by email, one time by text message and one time by telephone call. In case teachers did not respond to the questionnaires, they were reminded four times by email.

### *Interventions*

#### *Parent training*

The PT intervention was an 18 session group based PT program consisting of the 12 session Incredible Years Parent Training Program ® BASIC version combined with 3 sessions from the ADVANCE Program (Webster-Stratton, 2011) as well as three additional sessions. The BASIC program is designed for 2-8 year old children and it aims to enhance attachment and positive parent-child interactions, and reduce a negative and promote a positive parenting style. Parents are trained in integrating play and positive labeling in their parental practices,

along with structuring difficult everyday situations as well as routines and house rules. Furthermore, parents are taught how to use behavioral contingency principles in the form of rewards and consequences, such as loss of privileges and time out. Sessions consisted of video vignettes showing effective and ineffective parental strategies, group discussions and exercises where parents practice new ways of managing their child's difficulties. The three extra sessions from the ADVANCE program cover problems with the parents' own reactions, such as inefficient communication between partners and management of their own anger and depressed feelings. Finally, a network session (e.g. with grandparents, neighbors, and leisure time instructors), an individual session, and a session where all current parent groups met, were included in the course.

To assist the IY PT, the Centre for ADHD has developed an ADHD specific guide that is applied throughout the program (Cassøe, Bæk Bomme, Møller & Straarup, 2014). The guide consists of psychoeducation on ADHD and some supplementary exercises, especially focusing on dealing with ADHD problems, such as clear and direct communication with the child as well as encouraging the child's endeavors in sustaining attention. Parents of 6-7 children and two group leaders participated per group. All eight group leaders were clinical psychologists accredited or under education in IY. A treatment fidelity checklist was filled out by the group-leaders at the end of each session regarding content covered as required by the IY PT. The checklists were screened for treatment adherence by IY certified Peer Coaches. All group sessions were videotaped and there was weekly supervision with participation of a certified IY mentor.

#### *Teacher training*

The TT program was a manualized program developed at the Centre for ADHD (Bertelsen & Straarup, in press). The program consisted of 4 x 3 hours group sessions and a 2-hour individual supervision session dealing with the teachers' experiences with their focus-child. Group sessions were conducted with approximately 15 school or kindergarten teachers and two group leaders. The program was inspired by the IY TT program and based on the core principles from the IY Basic Program (Webster-Stratton, 2011), but with a specific focus on ADHD. The manual included psychoeducation on ADHD and disruptive behavior, strategies to promote inclusion of children with special needs in a kindergarten/school setting, use of behavioral contingency principles to motivate the children to cooperate, and, generally, to focus on positive aspects of the child's behavior. Group sessions were conducted in accordance with IY principles, utilizing video vignettes, exercises, role-play and group discussions. The two teacher program developers conducted all TT group sessions.

*Analytical procedure*

Group differences in baseline participant characteristics were examined using, as appropriate, Pearson's  $\chi^2$ , or Student's *t*-test. Exploratory post-hoc regression analyses of the influence of pretreatment variables that differed between groups were conducted (on parent-reported ADHD symptoms only). Two repeated-measures ANOVAs were applied for analyzing between-group differences in outcome; one from pre- to post-treatment; and one from pre-treatment to follow-up. These analyses were conducted separately for each outcome variable as rated by both parents and teachers. Within-group changes were analyzed with paired *t*-tests, and effect sizes (ES) were calculated as standardized mean differences from pre- to post treatment and pre to FU based on *SD* difference scores (a variant of Cohen's *d*: Borenstein, Hedges, Higgins and Rothstein, 2009). The percentage of children with statistical significant change (improvement) on ADHD and ODD/CD symptoms was calculated based on the Reliable Change Index (Jacobson & Truax, 1991) based on psychometric values from the study. A score  $\geq 1$  *SD* below pre-treatment *M* at post was regarded as clinical significant change (Ogles, Lambert & Masters, 1996). Children with statistical significant change may be considered improved, while children with both statistical and clinical significant change may be considered recovered.

When more than one teacher had filled in the questionnaires, the answers were averaged. For parental ratings, only the mother reports were analyzed. Missing item-level data on scales (c. 3.5 %) were handled by mean substitution; a procedure that performs well if few items are missing per individual on a scale, and if scales have acceptable internal consistency (Shafer and Graham, 2001). Due to the high rate of participants not completing assessment at post and follow-up, only completer analyses were performed. Statistical analysis was conducted using SPSS version 22.

**Results***Participant flow*

Families of 64 children participated in the study. Thirty-four children were randomized to the PT + TT condition, and 30 children to PT alone (See Fig. 1). Out of the 22 families that were excluded from the study, 19 received PT at the clinic and three were recommended other forms of treatment for clinical reasons. There was no dropout in the PT intervention in either of the two groups, but four of the teachers of the 34 children discontinued the TT program; three gave as reason a too busy schedule, and one illness. Despite low participant dropout, there was considerable non-completion of questionnaires for both parents and teachers.

Post hoc analysis revealed no difference in parent-reported child symptom severity of ADHD at baseline between the mothers that did complete post assessment ( $M = 23.05$ ,  $SD = 4.67$ ) and the mothers that did not ( $M = 24.16$ ,  $SD = 3.27$ ) ( $p = .442$ ). Furthermore, mothers not completing post-assessment were equally distributed between the two groups ( $\chi^2 = 0.06$ ,  $p = .810$ ). Children with teachers who did not complete post-assessment were not rated significantly different on teacher-reported ADHD symptoms at baseline ( $M = 18.79$ ,  $SD = 6.01$ ) compared to children with teachers that did complete post assessment ( $M = 16.98$ ,  $SD = 4.16$ ) ( $p = .489$ ). There was no statistically significant difference in the distribution of non-completing teachers between the two groups,  $\chi^2 = 2.82$ ,  $p = .114$ , albeit with numerically more non-responders in the PT only group (at post, 8 teachers did not complete post assessment in PT+TT vs. 13 teachers in PT only).

\*\*\* Insert Figure 1 about here\*\*\*

#### *Sample characteristics*

For the whole group of participants, the mean age was 6.37 years ( $SD = 1.73$ ), with 15 girls (24.4 %). Twenty-six of the children (40.6 %) had received an ADHD diagnosis prior to the intervention; 15 children ADHD alone and 11 ADHD and a comorbid disorder. Comorbid diagnoses were conduct disorder in nine children, autism spectrum disorder in one child and tic disorder in one child. Fifty-four (84.4 %) of the families had had prior contacts with the public services regarding their child's difficulties (e.g. school psychologist, child psychiatrist or general practitioner), and 14 of the children (21.9 %) received medical treatment for their ADHD. Mean number of attended PT sessions was 14, and there was no significant difference in attendance across the two groups ( $p = .367$ ).

Characteristics of the two groups are presented in Table 1. The two groups differed significantly at baseline on gender with more girls in the PT + TT group ( $p = .017$ ), and on mother-rated SDQ- externalizing subscale, with more symptoms in the PT+TT group ( $p = .047$ ). There was a non-significant trend ( $p = .083$ ) towards a difference in ODD/CD as measured by the mother-reported ECBI, with more symptoms in the PT+TT group. There were no significant differences at baseline with respect to child or mother age, level of mother or teacher reported ADHD-symptoms or internalizing difficulties as measured by the SDQ. There was also no difference in medication status at baseline. Post-hoc regression analysis revealed that neither gender ( $\beta = -.04$ ,  $p = .775$ ) nor mother reported pre-level SDQ externalizing subscale ( $\beta = -.04$ ,  $p = .817$ ) significantly predicted the magnitude of the effect post-treatment on mother-reported ADHD symptoms.

\*\*\* Insert Table 1 about here\*\*\*

*Between-group analyses*

At post-treatment and FU, a 2 (Group) × 2 (Time) ANOVA revealed no significant interaction effects on neither mother-reported nor teacher-reported outcome measures, indicating no difference in effectiveness between the two interventions (See Table 2).

\*\*\* Insert Table 2 about here\*\*\*

*Within-group analyses*

Within group changes in the two groups are seen in Table 3 and Figure 2. All mother-reported measures revealed significant and moderate to large ESs in both the PT and the PT + TT group with comparable ESs in the two groups. The pre-FU ESs in both groups were in line with pre-posttreatment ESs - even with numerical higher FU ESs on 6 of the 8 measures – which shows the gains from the interventions to be durable assessed from the mothers' perspective.

On the teacher-reported measures in the PT+TT group, there were significant and moderate ESs (0.42-0.63) on three of the four measures post treatment, and two at FU. No significant changes were found on ADHD symptoms (CTRS) at post treatment or FU. Significant change was only revealed for the PT alone group on one of the four measures at post-treatment, and on none at FU.

\*\*Insert Table 3 about here\*\*

\*\* Insert Fig. 2 about here\*\*

*Improvement and recovery*

Percentage of children with statistically significant improvement and statistical and clinical recovery on measures of ADHD and conduct problems are seen in Table 4. For both parents and teachers, improvement and recovery on ODD/CD exceeded improvement and recovery on ADHD. Recovery for mother-reported ADHD at FU was comparable across the two groups, with 22.2 percent in the PT and TT group compared to 25.9 percent in the PT alone group. For ODD/CD, 51.9 percent of the children had recovered at FU in the PT alone group, compared to 37 percent in the PT and TT group. On teacher ratings, 16 percent had recovered on ADHD symptoms in the PT and TT group at FU, compared to none in the PT alone group. On ODD/CD, 20 percent of the children had recovered at FU in the PT and TT group, compared to none in the PT alone group.

\*\*\*Insert Table 4 about here\*\*\*

#### *Treatment satisfaction*

Generally, both parents and teachers were highly satisfied with the treatments. The mean score pr. item on the parent-rated ESQ was 2.82 (out of max 3); 74 % of the parents had experienced the PT program as certainly (score 3) helpful for their child's difficulties, and 94 % would certainly (score 3) recommend the program to a friend. The mean score pr. item on the teacher-rated satisfaction questionnaire was 6.1 (out of max 7); 75.6 % of the teachers reported that they thought that the intervention had helped to reduce the child's difficulties (score 6 or 7), and 97.8 % of the teachers would certainly (score 7) recommend the program to a colleague.

## **Discussion**

The two intervention groups achieved moderate to large ESs on mother-reported outcomes in line with those generally reported in the literature on PT interventions for early ADHD. ESs were thus comparable to those achieved in the Trillingsgaard et al. (2014) prior study at the Clinic and in the Webster-Stratton et al. (2011) RCT. For instance, the pre-post and pre-FU ESs on mother-reported ADHD symptoms in the study were 0.57 and 0.75, respectively, compared to 0.8 and 0.7 in Trillingsgaard et al. (2014), and 0.8 and 0.7 in Webster-Stratton et al. (2011). It should be noted, however, that rather few of the children (16.7-25.9 % rated by parents at FU) could be considered recovered on ADHD symptoms after treatment with both statistical and clinical significant change, indicating that PT rarely eliminates all difficulties for ADHD children and their families.

There were, however, no significant between-group differences in outcomes between the PT only and the PT + TT group. None of the ANOVAs revealed significant interaction effects on any of the outcome measures. Mother-reported ESs were even numerically higher for the PT only than for the TT + PT group on three of the four outcome measures, both posttreatment and at FU. The hypothesis of a general incremental outcome of adding TT to PT in the treatment of young children with ADHD or ADHD symptoms was thus not supported. The study is, of course, a small-scale one, only powered to detect large differences in outcome, but the fact that mother-reported ESs generally were larger in the PT only group indicates that the failure to support the hypothesis probably was not due to limited power. One explanation of the result could be that the TT program with its only 4x3 hour sessions was too short to achieve a notable difference for the children. On the other hand, there is limited empirical support for the value of TT as a supplement to PT for young children with ADHD, and some studies also failed to find incremental outcome of adding TT to PT, even with a more intensive TT intervention (Kern et al., 2007).

Another possibility, as indicated in the introduction, could be that outcomes of interventions for ADHD are context dependent with little transfer from one setting to another. This could, perhaps, explain why an intervention in a school/kindergarten context could have little influence on family interaction, and therefore would not reveal any effect on parent-reported outcomes. The hypothesis that the supplementary TT intervention especially would achieve its outcomes in the school or kindergarten context, where it was delivered, could in the study only be tested on teacher-reported outcomes. As mentioned, there were no interaction effects in the between-group analyses on teacher-reported outcomes. However, here power problems might more plausibly have played a role, since within-group analyses revealed numerically larger ESs on all teacher-reported measures for the combined PT + TT group. Thus, for conduct problems, plausibly more easily observed and influenced than ADHD symptoms in a kindergarten/early school context, there were moderate to large ESs in the PT + TT group based on teacher-reported outcome (0.61 posttreatment; 0.52 at FU) compared to ESs close to zero in the PT alone group. ADHD symptoms may be less disturbing in kindergarten or first years of primary school with a limited focus on scholastic learning. In fact, teachers rated children's initial ADHD symptoms lower than parents ( $M = 17.24$ ;  $SD = 6.3$  vs.  $M = 23.27$ ;  $SD = 4.44$ ). If teachers did not observe severe child ADHD difficulties, the finding of little change in such difficulties is not surprising.

The importance of special education for teachers on how to include children with atypical development and/or behavioral problems in school is generally acknowledged. Overall, it seems unlikely that some form of systematic training of teachers in how to deal with children with ADHD difficulties would not benefit such children. As mentioned, the study found some indication of better outcome of the combined PT and TT program on teacher-reported conduct problems, although there were no significant differences between the two groups. That there was no indication of better results of supplementary TT on parent-reported outcomes could possibly be due to the fact that some of the suggested benefits of interventions at school would not immediately be noticeable in the family. Improvement in peer relations and self-confidence, changes that might more easily generalize to an out-of-school context, may need more time to fully unfold. Instances of latency of effects of PT, both at home and in a school setting, has been described in the field (Strayhorn and Weidman, 1991; Shaw, Dishion, Supplee, Gardner and Arnds, 2006). Therefore, 5 months of intervention and 6 months of FU may not be enough time to reveal the full impact of the TT intervention.

The study has several limitations. The absence of a passive control group hinders strict conclusions on the effect of both interventions, although pre-post ESs were in line with those found in other studies investigating PT for early ADHD. The study is underpowered to detect difference in small to moderate ESs, most likely the upper

bound for what could be expected in a comparison of two active interventions. There was no independent assessment in the study, and both parent- and teacher-ratings could be biased. Generally, provision of valid objective assessment has been difficult within ADHD intervention research (Rimestad et al., 2016; Sonuga-Barke, Brandeis, Cortese, Daley, Ferrin, Holtmann, ...Sergeant, 2013). Last, but not least, there was a high degree of non-completion of questionnaires among the participants in spite of reminders. Non-completion of assessment among parents in studies on ADHD is not uncommon in the literature, possibly due to high levels of family stress (Östberg and Rydell, 2012; Chronis-Tuscano, O'Brien, Johnston, Jones, ...Seymour, 2011). The high degree of teacher non-completion could have been influenced by an unpopular educational reform forced on the teachers by the Danish Government after lockouts taking place immediately before the start of the study. Although the rate of missing assessments is a drawback of the study, post-hoc analyses showed that the two groups were equally affected. This suggests that while the rate of non-completion did reduce the statistical power to detect a difference, it most likely did not otherwise bias the comparison of the groups.

## Conclusion

The study found no significant differences between a PT program for early ADHD based on The Incredible Years © and the combination of the PT program with a TT program delivered to teachers in kindergarten or school. The hypothesis of an incremental effect of supplementing PT with TT in the treatment of ADHD was thus not supported. Within group analyses revealed, however, noticeably larger effect sizes on teacher-reported conduct problems for the PT + TT group, perhaps indicating a school context dependent advantage of the combined interventions. Some of the benefits of a school-based intervention especially prone to generalize to other contexts, such as improved peer relations and self-esteem, might take longer time to unfold. The study was underpowered with a high degree of non-completion of questionnaires. Further studies on TT for ADHD are needed; preferably with larger samples and covering more extended time periods.

*All parents and teachers participating in this study had given informed consent, and the study was conducted in compliance with ethical standards. The authors declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.*

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**Table 1**  
*Pre-treatment characteristics of the groups*

Characteristics	PT only <i>n</i> = 30 <i>M</i> ( <i>SD</i> ) or <i>n</i> (%)	PT+ teacher training <i>n</i> = 34 <i>M</i> ( <i>SD</i> ) or <i>n</i> (%)	<i>p</i>
Child age (in months)	72.47 (19.79)	75.50 (20.70)	.553
Number of girls in group	3 (10 %)	12 (35.3 %)	<b>.017</b>
Mother age (in years)	35.73 (5.31)	36.09 (6.06)	.805
Number of children diagnosed with ADHD	13 (43.3 %)	13 (38.2 %)	.393
Number of children receiving medication	7 (23.33%)	7 (20.58 %)	.791
Mother-reported CPRS	22.62 (4.75)	23.82 (4.15)	.288
Teacher-reported CTRS	18.90 (6.26)	17.52 (6.88)	.698
Mother-reported ECBI	153.00 (21.28)	163.64 (26.36)	.083
Teacher-reported SESBI	162.32 (32.20)	151.03 (28.05)	.207
Mother-reported SDQ-internalizing.	5.55 (3.34)	6.48 (3.48)	.362
Mother-reported SDQ-externalizing.	12.43 (2.95)	13.94 (2.98)	<b>.047</b>

**Table 2***Results from ANOVAs comparing two groups over time*

Interaction effects (Time * group)						
	Pre-post			Pre-FU		
	<i>F</i>	<i>df</i>	<i>P</i>	<i>F</i>	<i>df</i>	<i>P</i>
<i>Mother-reported</i>						
CPRS	0.52	49	.472	1.14	51	.289
ECBI	0.17	43	.684	0.25	52	.616
SDQ ext	0.26	43	.610	0.36	52	.551
SDQ int.	2.35	43	.132	1.19	52	.279
<i>Teacher-reported</i>						
CTRS	1.46	38	.235	1.98	36	.167
SESBI-R	1.06	38	.310	2.57	36	.117
SDQ ext.	0.00	40	.987	0.02	36	.879
SDQ int.	0.64	40	.429	3.26	36	.079

Note: CPRS: Conner's Rating Scale 3AI, CTRS: Conner's Teacher Rating Scale 3 AI, ECBI: Eyberg Child Behavior Inventory, SESBI-R: Sutter-Eyberg Student Behavior Inventory, SDQ int.: Strengths and Difficulties internalizing subscale, SDQ ext.: Strengths and Difficulties externalizing subscale.

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**Table 3**  
*Outcome variables and effect sizes across groups*

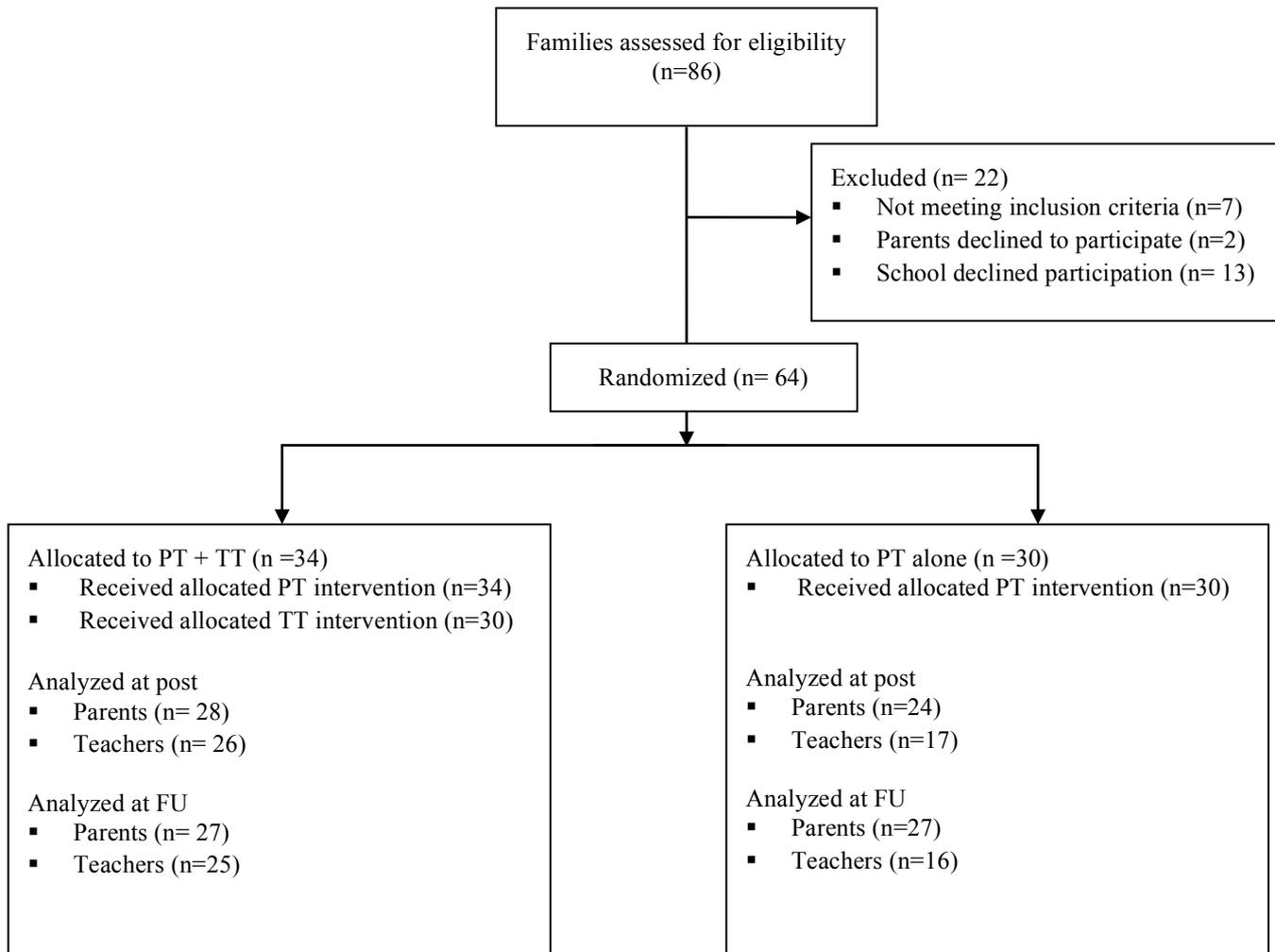
	PT + TT <i>M (SD)</i>				PT alone <i>M (SD)</i>					
	Pre (n=34)	Post (n=28)	FU (n=27)	<i>d</i> pre-post	Pre (n=29)	Post (n=24)	FU (n=27)	<i>d</i> pre-post	<i>d</i> pre-FU	
<i>Mother-reported</i>										
CPRS	23.78 (4.29)	21.5 (6.38)	20.85 (6.07)	0.40*	0.51*	22.17 (5.06)	18.69 (7.80)	17.50 (6.00)	0.57*	0.75**
ECBI	164.10 (24.36)	140.92 (22.57)	135.15 (25.89)	0.75**	0.87**	155.77 (19.48)	127.00 (26.02)	123.26 (23.73)	1.16**	1.42**
SDQ int.	9.36 (3.76)	6.45 (3.09)	6.70 (3.98)	0.79**	0.73**	7.43 (3.34)	5.96 (3.26)	5.44 (2.67)	0.59*	0.61*
SDQ ext.	14.23 (2.68)	11.41 (3.14)	11.44 (3.27)	0.93**	0.70**	12.52 (3.04)	10.17 (3.49)	9.33 (2.92)	0.75*	0.97**
<i>Teacher-reported</i>										
CTRS	17.33 (7.16)	15.35 (8.34)	16.50 (8.21)	0.20	0.14	18.50 (6.93)	18.59 (6.84)	20.15 (5.16)	-0.22	-0.28
SESBI-R	152.08 (29.27)	139.39 (38.71)	141.81 (33.66)	0.61**	0.52*	161.46 (26.32)	155.81 (26.09)	164.46 (24.52)	0.09	-0.03
SDQ int.	6.32 (3.57)	5.16 (3.48)	4.84 (3.12)	0.42*	0.59*	5.24 (3.16)	4.71 (3.11)	5.47 (2.81)	0.25	0.01
SDQ ext.	13.44 (5.44)	10.0 (5.76)	11.43 (5.61)	0.53*	0.44	16.35 (5.96)	13.44 (4.97)	14.56 (5.61)	0.52*	0.42

Table notes: \* Significant at  $\alpha = .05$ , \*\* Significant at  $\alpha = .001$ , CPRS: Conner's Parent Rating Scale 3AI, CTRS: Conner's Teacher Rating Scale 3 AI, ECBI: Eyberg Child Behavior Inventory, SESBI-R: Sutter-Eyberg Student Behavior Inventory, SDQ int.: Strengths and Difficulties internalizing subscale, SDQ ext.: Strengths and Difficulties

**Table 4**  
*Percentage of children with improvement and recovery across groups*

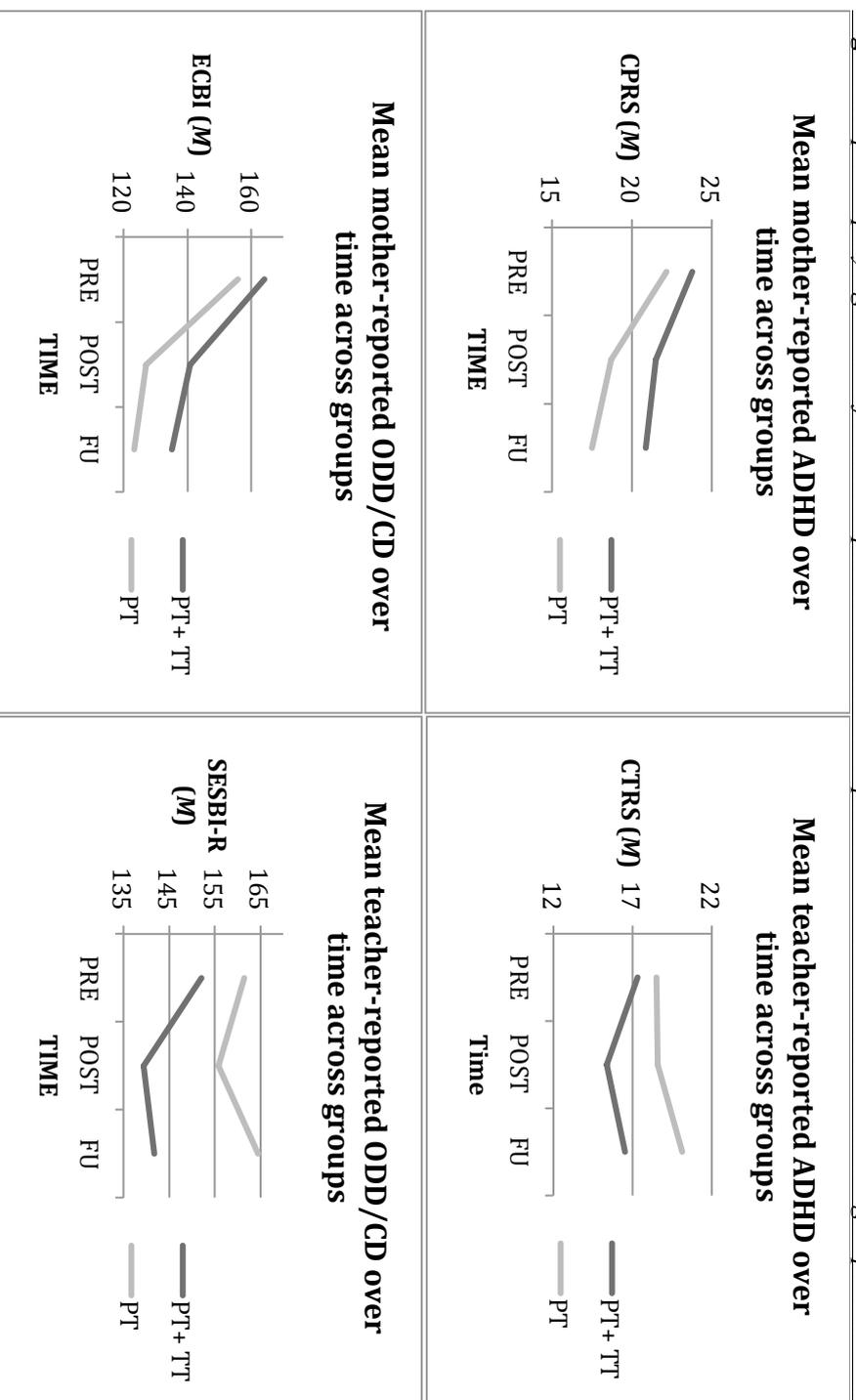
	PT + TT		FU		PT alone		FU	
	Post Improvement	Post Recovery	Post Improvement	Post Recovery	Post Improvement	Post Recovery	Post Improvement	Post Recovery
<i>Parent-reported</i>								
CPRS	34	25	22.2	22.2	30	16.7	34.6	25.9
ECBI	40.9	17.9	44.4	37	47.8	37.5	59.3	51.9
<i>Teacher-reported</i>								
CTRS	33.3	23.1	27.3	16	18.8	5.9	18.8	0
SESBI-R	41.7	23.1	40.9	20	25	6.6	31.3	0

Abbreviations: CPRS: Conner's Rating Scale 3AI, CTRS: Conner's Teacher Rating Scale 3AI, ECBI: Eyberg Child Behavior Inventory, SESBI-R: Sutter-Eyberg Student Behavior Inventory.

**Figure1.** *Participant flow*

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Fig. 2: Graphs displaying means of mother-reported and teacher-reported ADHD and ODD/CD across groups over time



## **Appendix C**

**Paper 3:** Rimestad, M.L., O'Toole, M.S. & Hougaard, E. Mediators of change in a parent training program for early ADHD difficulties: the role of parental strategies, parental self-efficacy and therapeutic alliance.

*Submitted to Journal of Attention Disorders*

**TITLE: MEDIATORS OF CHANGE IN A PARENT TRAINING PROGRAM FOR EARLY ADHD  
DIFFICULTIES: THE ROLE OF PARENTAL STRATEGIES, PARENTAL SELF-EFFICACY AND  
THERAPEUTIC ALLIANCE**

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### Abstract

**Objective:** The aim was to explore mediators of change in parent training (PT) for 3-8 year-old children with ADHD difficulties. **Methods:** Parents of 64 children received PT with Incredible Years ® and assessed child ADHD symptoms and conduct problems and their parenting strategies, parental self-efficacy and therapeutic alliance before, during and after PT. Product-of-coefficients mediation analyses in multi-level models were applied, and causal relations between mediators and outcome were investigated in time-lagged analyses. **Results:** Increased parental self-efficacy and reduced negative parenting statistically mediated reductions in ADHD and conduct problems in the product-of-coefficient analyses. However, time-lagged analyses were unable to detect a causal relation between prior change in mediators and subsequent child symptom reduction. There was limited evidence of therapeutic alliance as mediator of child symptom reduction or change in parenting variables. **Conclusion:** Parental self-efficacy and reductions in negative parenting may mediate change in PT, but more fine-grained time-lagged analyses are needed to establish causality.

**Keywords:** *parent training; ADHD; conduct problems; mediation; incredible years; treatment*

Attention-deficit hyperactivity disorder (ADHD) is a neurodevelopmental disorder, characterized by age-inappropriate, persistent and impairing symptoms of inattention, hyperactivity and impulsivity (American Psychiatric Association, 2013). The prevalence of childhood ADHD is estimated to be about 5-7 %, and it is among the most commonly diagnosed childhood disorders (Willcutt, 2012). Children with ADHD are often suffering from emotional dysregulation (Shaw, Stringaris, Nigg & Leibenluft, 2014), social problems (Bagwell, Molina, Pelham & Hoza, 2001) and low self-esteem (Mazzone et al., 2013). Parenting a child with ADHD is challenging, as affected children are

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typically more non-compliant, more negative in interactional style and harder to manage, compared to children with normal development (Mash & Johnston, 1982). Consequently, families of children with ADHD are often distressed (Theule, Wiener, Tannock & Jenkins, 2013), have high levels of conflicts, and negative parent-child interactions (Deault, 2010). A negative parenting style may increase the risk of maintaining or exacerbating ADHD symptoms and is a risk factor of development of oppositional defiance disorder (ODD) and conduct disorder (CD) (Johnston & Mash, 2001). Therefore, early interventions targeting both child and family difficulties have been developed and evaluated.

Parent training (PT) programs have been found to be effective interventions for ADHD and ADHD symptoms according to parental assessment (Daley, van der Oord, Ferrin, Danckaerts, Doepfner, Cortese & Sonuga-Barke, 2014; Rimestad, Lambek & Hougaard, 2016). Some PT programs, such as the Incredible Years (IY) program (Webster-Stratton et al., 2011), were developed from PT programs for ODD and CD; others, like the New Forest Parent Training Program (Abikoff, et al., 2015; Thompson et al., 2009), were specifically developed for ADHD. Rimestad and colleagues (2016) did not find differences in effectiveness between the two types of programs in their meta-analysis of PT for early (2½-6 years) ADHD.

However, little is known about change processes or mechanisms in IY or other PT programs. As stressed by Kazdin (2007), knowledge of *how* an intervention works should help in optimizing therapeutic change by providing optimal conditions and focusing on active ingredients. Studies on change processes are now increasingly conducted as mediation studies. A mediator or mediating variable may be defined as a variable that statistically accounts for, and thereby possibly explains the relationship between the dependent and independent variable (Kazdin, 2007; p. 3). Traditional approaches to statistical mediation have either used the Baron and Kenny (1986) ‘causal steps’ approach or the ‘product-of-coefficients’ approach suggested by Sobel (1982). However, none of

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these approaches to statistical mediation takes account of the timeline of change in variables that is necessary for conclusions as to the direction of causality. Kazdin (2007) recommends time-lagged analyzes of repeated measurements of both process and outcome as an optimal approach to intervention research on mediation.

The assumption in PT programs is that a child's difficulties can be changed through a change in parenting practices. Potential mediating parental practices have been suggested to be both an increase in positive parenting and a decrease in negative/ineffective parenting as well as use of appropriate discipline. Positive parenting includes strategies such as praise, joint play and use of reasoning, humor and incentives (Gardner, Sonuga-Barke & Sayal, 1999), whereas negative parenting concerns critical comments, blame, hostility, disapproval and use of harsh discipline (Hinshaw et al., 2000). Another proposed mediator is parental self-efficacy, defined as parental beliefs about competence in one's parenting (Coleman & Karraker, 2003). A high degree of parental self-efficacy has been found to be associated with parental responsiveness, warmth and persistence in parental strategies (Sanders & Woolley, 2005). Conversely, a low degree of parental self-efficacy is associated with coercive parent-child interactions (Sanders et al., 2005).

The above-mentioned parental practices and self-efficacy have been found to change in PT interventions for children with ADHD (Daley et al., 2014), but there is currently limited empirical knowledge about their mediating role in treatment outcome (Forehand, Lafko, Parent and Burt, 2014). Three prior studies have investigated mediational processes in PT for early ADHD, all in the form of statistical mediation without taking account of the time-line of change in variables. One large study ( $n=579$ ) compared medication, behavior therapy, their combination and a community control condition with school children in the age range of 7-10 years (Multimodal Treatment Study of Children with ADHD [MTA]; Hinshaw et al., 2000). Regression analyses (time x parenting x treatment condition) revealed a mediational effect of parent-rated negative/ineffective treatment

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(but not of positive parenting) on teacher-rated ADHD/oppositional symptoms and social skills for the combined group compared to community control (but not for the other condition comparisons). Hanisch, Hautmann, Plück, Eichelberger and Döpfner (2014) investigated the mediating effects of a preventive PT program (PEP) in a randomized controlled trial (RCT) in a product-of-coefficient analysis for both child conduct problems and ADHD difficulties, compared to a passive control group in a sample of 155 preschool children age 3-6 with or at risk of ADHD and behavioral problems. They investigated dysfunctional parenting, positive parenting, parental self-efficacy and parental warmth as mediators on child outcomes of ODD/CD and ADHD symptoms. The study found that changes in both ADHD and ODD/CD symptoms were mediated primarily by a reduction in dysfunctional parenting but also by an increase in positive parenting to a lesser extent. There were no mediating effects of parental warmth or parental self-efficacy. Seabra-Santos and colleagues (2016) investigated the mediating processes of IY PT in an RCT with 124 preschool children with or at risk of ADHD and behavioral problems. The study applied causal-steps mediation analysis and found that reductions in negative parenting as well as increased parental self-efficacy mediated the effect of IY PT on child externalizing behavior. Although the three studies were able to establish statistical mediation, it should be noted that the causal relations between mediator and outcome were not addressed in time-lagged analyses.

Besides parenting variables, other putative mediators in PT could be related to the therapeutic process itself. The therapeutic or working alliance is the most studied process variable in research on psychotherapy with adults (Horvath, Del Re, Flückiger & Symmonds, 2011). The alliance refers to the quality of the collaboration between therapist and patient; often specified as an emotional bond between the partners, and agreement on aims and tasks of the therapy (Bordin, 1979). The alliance has most often been considered a rather stable indicator of the quality of the therapeutic relationship, although it has also been suggested that an increase in alliance quality during therapy

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may be related to better outcome (e.g., Patterson & Chamberlaine, 1994). Most studies have simply investigated the correlation between alliance and outcome (Horvath et al., 2011), although some have corrected outcome variables for change prior to the time point of measurement of the alliance (11 studies in a recent meta-analysis by Crits-Christoph, Gibbons & Mukherjee, 2013).

The therapeutic alliance has also been found to predict outcome in psychotherapy with youths (McLeod, 2011; Shirk, Karver & Brown, 2011). In the largest meta-analysis by McLeod (2011;  $k=38$ ), the mean weighted effect size ( $r$ ) between alliance and outcome was small ( $r=.14$ ) with no significant difference between results for therapist-child ( $r=.12$ ) and therapist-parent alliance ( $r=.15$ ); a somewhat smaller effect size (ES) than that found in the adult alliance literature ( $r = .28$  in Horvath et al., 2011).

There has, however, been little research on the therapeutic alliance in PT, and only one study specifically dealing with the therapeutic alliance in PT for children with ADHD could be localized. In the study by Lerner, Mikami and McLeod (2011), early (3<sup>rd</sup> session) observer-rated therapist-parent alliance and change in the alliance during an 8-session group-based PT (Parental Friendship Coaching) predicted improvement in several parenting and child behaviors observed during a 1-hour lab-based playgroup. In addition, some studies have investigated the alliance in PT for conduct problems. Kazdin and Whitley (2006) found that self-reported parent-therapist alliance in PT for children with oppositional and antisocial behavior was associated with self- and therapist-reported positive change in parenting variables. In contrast to these positive findings, however, Hukkelberg and Ogden (2013) in a large ( $n = 331$ ) naturalistic study found that self-reported therapist-parent alliance *negatively* predicted change in children's problem behavior in individually delivered PT for children with externalizing behavioral problems.

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Taken together, few studies have investigated mediators in PT for children with ADHD or ADHD difficulties, and results are somewhat mixed. The aim of this study was to explore mediating processes of the IY PT program for children aged 3-8 with ADHD or ADHD symptoms. We investigated parental discipline, positive and negative parenting, parental self-efficacy and therapeutic alliance as putative mediators.

### **Methods**

#### *Participants*

The sample consisted of 64 families. These families took part in an RCT, in which they were randomized to PT with IY alone, or to IY PT combined with a brief Teachers Training (TT) (results concerning the effectiveness can be found in Rimestad, Trillingsgaard & Hougaard, 2016). Since the study found no significant differences in outcome between the PT only and the PT + TT group, we treated the whole sample as a unified group in the mediational analyses.

Participants were parents of 3-8 year old children, who had self-referred to the Centre for ADHD in Aarhus, Denmark. The Centre for ADHD is a non-profit private clinic funded by Edith and Godtfred Kirk Christiansen's Foundation. The Clinic offers treatment to families with young children with or at risk for ADHD. The intervention was announced at the Clinic's website as well as in flyers distributed in health clinics in the local area.

Families were accepted for treatment on the basis of an overall clinical assessment including a semi-structured interview with parents and validated questionnaires (see below). In accordance with the inclusion criteria at the Centre for ADHD, families in the study had to have a child between 3 and 8 years with ADHD symptoms, but a formal ADHD diagnosis was not necessary. Families who had received PT prior to the intervention were excluded.

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*Measures**Outcome measures*

The intervention study applied several outcome measures (Rimestad et al., 2016), but in this study only the two parent rating scales on ADHD symptoms and conduct problems were used, as these were the primary outcomes.

*Conners' 3 ADHD Index Parent Rating Scale (CPRS; Conners, 2009).* The CPRS is a questionnaire to be filled out by parents assessing child ADHD symptoms. The scale consists of 10 items, each rated on a 4-point Likert-scale from 0 = *never or almost never* to 3 = *Very often or always*. The CPRS is a short index version of the full form of the Conners' ADHD Rating scales, including the items that most effectively differentiated youth with ADHD from nonclinical controls (Westerlund, Ek, Holmberg, Näswall & Fernel, 2009). The scale has been found to have good psychometric properties, with a Cronbach's  $\alpha$  of .90 (Westerlund et al., 2009). In the present study, Cronbach's  $\alpha$  was .76 at pre-treatment.

*Eyberg Child Behavior Checklist (ECBI; Eyberg & Pincus, 1999).* The ECBI is a questionnaire assessing child ODD/CD symptoms (henceforth labeled conduct problems). The questionnaire consists of two subscales, an Intensity Scale and a Problem Scale, but in this study only the intensity scale was used. The Scale has 36-items concerning frequency of opposition and conduct problems, with each item rated from 1 = *Never* to 7 = *Always*. The scale has been found to have good psychometric properties with a Cronbach's alpha of .93 (Reedtz, Bertelsen, Lurie, Handegård, Clifford & Mørch, 2008). In this study, Cronbach's alpha was .86 at pre-treatment.

*Mediator measures*

*Parenting Sense of Competence (PSOC) (Johnston & Mash, 1989).* The PSOC is a questionnaire assessing parental competence or self-efficacy in their role as parents. The scale has 16 items, each

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rated on a 6-point Likert scale from 1= *Strongly Disagree* to 6= *Totally Agree*, with higher scores indicating greater parental self-efficacy. The scale has been found to have acceptable psychometric properties, with a Cronbach's alpha of .80 (Ohan, Leung & Johnston, 2000). The pre-treatment Cronbach's' alpha was 0.83.

*Parenting Practices Index* (PPI; Conduct Problems Prevention Research Group, 1996). The PPI is a questionnaire that assesses the frequency of specific parental practices. Items are rated on 7-point Likert scales, ranging from 1= *never/not likely* to 7 = *always/almost 100% likely*. The scale has several subscales, of which three were used here; Appropriate Discipline Subscale (APP DISC) (16 items, Cronbach's alpha = .80 in this study), Harsh and Negative Parenting (NEG PAR) (14 items, Cronbach's alpha = .70) and Positive Parenting (POS PAR) (consisting of 15 items, Cronbach's alpha = .81).

*Working Alliance Inventory - Short Form - Patient version* (WAI-SF-Pt; Tracey & Kokotovic, 1989). The WAI-SF-Pt is a widely used 12-item version of the original 36-item WAI (Horvath & Greenberg, 1989) measuring the therapeutic alliance between therapist (here group leader) and patient (participant parent) from the patient/parent perspective. Each of the 12 items of the scale are scored on a 7-point Likert scale from 1= *never* to 7 = *all the time*. The full scale WAI is a measure of the overall quality of the alliance, and the scale has sub-scales for the three dimensions of Bordin's (1979) working alliance concept, therapeutic bond, agreement on therapeutic goals, and agreement on therapeutic tasks. In the present study, only the full scale was used. The scale has been found to have good psychometric properties, with a Cronbach's alpha of .93 for the full scale (Tracey & Kokotovic, 1989). Cronbach's' alpha pretreatment in this study was .91.

### *Procedures*

After the families had contacted the Centre, a clinical psychologist screened them for eligibility via the telephone. If eligible, two clinical psychologists arranged a home-based intake interview with

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the parents concerning the child's problem behavior and aspects of their family life. The parents also completed the CPRS and the ECBI. Based on the intake interview and the parental scores on the questionnaires, it was decided if the families should be offered treatment at a following clinical conference. Both mothers and fathers were encouraged to participate in the PT course, regardless of their marital status; 64 mothers and 45 fathers participated in the courses. Childcare facilities were arranged for the children (including siblings) during the PT intervention, and the families were provided with free meals during the treatment. The intervention was free of charge, but parents paid for the IY PT book and handouts.

Data were collected pre-, mid- and post-treatment. Six-month follow-up data from the effectiveness study (Rimestad et al., 2016) were not used in the analyses, since therapeutic processes are most meaningfully investigated in the intervention period. Except for WAI, all measures were collected at all three data points. Pre-test scores on outcome and process measures were obtained no later than 2 weeks before PT start; except for WAI that was collected after 3 sessions of PT. Mid treatment scores were obtained after the 10<sup>th</sup> session, approximately two months after treatment start. Post measures were obtained no later than four weeks after termination of the interventions.

Parent-rated pre-measures were collected in paper form; all other data were collected via email using an online data collection platform. In case parents did not complete the questionnaires, they were reminded up to four times in total; two times by email, one time by text message and one time by telephone call.

All participants signed a consent form after receiving verbal and written information on the study. The study was conducted in compliance with standards from the regional ethical committee and approved by the Danish Data Protection Agency.

*Intervention*

The PT intervention was an 18-session group based PT program, consisting of the Incredible Years Parent Training Program ® BASIC version as well as some content from the ADVANCE program (Webster-Stratton, 2011) and additional ADHD specific content added by the Clinic (cf. Cassøe, Bæk Bomme, Møller & Straarup, 2011) The program is designed for 2-8 year old children, aiming to enhance attachment and positive parent-child interactions as well as reducing negative parenting. Parents are trained in integrating play and positive appraisal in their parental practices, along with structuring difficult everyday situations. Furthermore, parents are taught how to use behavioral contingency principles in the form of rewards and negative consequences, such as loss of privileges and time-out. A supplementary ADHD specific guide developed by the Clinic (Cassøe et al., 2011) was implemented, covering psychoeducation on ADHD and supplementary exercises focusing on dealing with ADHD problems. All sessions consisted of video vignettes showing effective and ineffective parental strategies, group discussions and exercises where parents train new ways of managing their child's difficulties. Groups consisted of parents of 6-7 children and two group leaders. All eight group leaders were clinical psychologists accredited or under education in IY. Treatment fidelity checklists were filled out by the group leaders at the end of each session and screened for treatment adherence by IY certified Peer Coaches. All group sessions were videotaped and reviewed during weekly collegial supervision with participation of a certified IY mentor.

Teachers of the 34 children who were randomized to PT + TT received a 4\*3 hours of training course and an individual supervision session dealing with the teacher's experiences with their focus-child (Bertelsen & Straarup, in press). The course was held in groups of 10-15 teachers and two group leaders, and it was based on the core principles of the IY PT. The manual included psychoeducation on ADHD and disruptive behavior, strategies to promote inclusion of children

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with special needs, use of behavioral contingency principles to motivate the children to cooperate, and, generally, to focus on positive aspects of the child's behavior.

*Analytical procedure*

Missing data at the item level were low (3.5 %) and handled by mean imputation; a procedure that performs well if few items are missing per individual on a scale, and if scales have acceptable internal consistency (Shafer & Graham, 2002). For changes in variables over time, ESs were calculated as standardized mean differences between variables based on *SD* difference scores (a variant of Cohen's *d*; Borenstein, Hedges, Higgins and Rothstein, 2009).

We conducted two sets of mediational analyses. First, we analyzed all proposed mediators separately by use of the product-of-coefficients method; for all variables that here met the criterion for mediation. Secondly, we conducted time-lagged analyses of change to address the causal relationship between the variables.

We used the product-of-coefficients approach in the analyses of statistical mediation, as it has been recommended over the causal-steps approach due to more statistical power and lower risk of type 1 error (MacKinnon, Lockwood, Hoffman, West & Sheets, 2002). The product-of-coefficients approach investigates the indirect mediational path as indicated by the interaction ( $a \times b$ ) of path *a* (from the independent variable to the mediator) and path *b* (from the mediator to the dependent variable) in Baron and Kenny's (1986, p. 1176) mediational figure (see Figure 1). Path *c* in figure 1 indicates the direct association between the independent and dependent variable (outcome).

**\*\*Insert Figure 1\*\***

With no passive control group in the study, time served as the independent variable in the product-of-coefficients analyses. The product-of-coefficients was determined in multilevel models (MLMs)

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(cf. Krull & MacKinnon, 2001). These MLMs were two-level models, where time was nested within individuals. After obtaining the coefficients, results were bootstrapped, since this method provides an empirical approximation of the sampling distribution of  $ab$  as normal, thereby making it possible to construct confidence intervals for the indirect effect (Preacher & Hayes, 2008). The bootstrapping was conducted with 5000 iterations in order to obtain both standard errors and confidence intervals. A significant mediating effect was established when the 95% bootstrapped confidence interval did not include the value zero. ESs in the analyses were expressed as the proportion of the total effect of the intervention accounted for by the proposed mediator, that is the mediated effect / total effect based on absolute values of the variables (Kenny, Korchmaros & Bolger, 2003; MacKinnon et al., 2007). Since separate models were specified for each of the proposed mediators, the effect sizes do not sum to 100%.

As mentioned, time-lagged analyses were conducted on all variables that fulfilled the criteria for statistical mediation in the product-of-coefficients analyses. First, it was analyzed if the proposed mediator at mid-treatment (controlling for pre-intervention value) predicted the outcome at post intervention (controlling for mid-treatment value). Secondly, the reverse path was investigated, that is, if the outcome at mid-treatment (controlling for pre-treatment value) predicted the mediator post-treatment (controlling for mid-treatment value). Controlling for the preceding time points partials out stable aspects and prior changes of variables (Pek & Hoyle, 2016).

Because the therapeutic alliance is most often considered a rather stable indicator of the quality of the therapeutic relationship, we here also conducted analyses of the relationship between alliance (at the two data points) and residualized pre-post change in outcome and process variables. In case mid-treatment alliance predicted residualized pre-post change in a criterion variable, we also analyzed its association with mid-post change.

All statistical analyses were conducted in STATA v. 14.

## Results

### *Study inclusion*

In total, families of 84 children were screened for eligibility of inclusion, and 64 were included (for further information of the inclusion process, see Rimestad et al., 2016). No families dropped out of treatment, but there was some non-completion of questionnaires. Mothers of 55 and 52 children completed questionnaires at the mid-intervention and post-intervention assessment, respectively. Post hoc analysis revealed no difference in parent-reported child symptom severity of ADHD as measured on the CRS at baseline between the mothers that did complete post assessment ( $M = 23.05$ ,  $SD = 4.67$ ) and the mothers that did not ( $M = 24.16$ ,  $SD = 3.27$ ) ( $p = .442$ ) (Rimestad et al., 2016).

### *Sample characteristics*

For the whole group of participants, the mean age of the child was 6.4 years ( $SD = 1.7$ ), with 15 girls (24.4 %). Twenty-six of the children (40.6 %) had received a formal ADHD diagnosis prior to the intervention. Of these children, 15 only had a diagnosis of ADHD and 11 had ADHD and a comorbid disorder. Comorbid diagnoses were CD in nine children, autism spectrum disorder in one child and tic disorder in one child. Fifty-four (84.4 %) of the families had had prior contacts with the public services concerning their child's difficulties (e.g. school psychologist, child psychiatrist or general practitioner), and 14 of the children (21.9 %) received medical treatment for their ADHD. Mean number of attended PT sessions was 14.

### *Within changes over time*

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Mean scores and ESs on measures across the three time points are presented in Table 1. There was a significant reduction in both ADHD and conduct problems from pre- to post-treatment, corresponding to moderate and large ESs, respectively. The largest changes, however, were from pre-to mid-treatment. There were also positive and significant changes from pre- to post-treatment in all variables except for appropriate discipline. The largest ESs were found for positive parenting and reduction in negative parenting (ESs > 1.0). Changes in mediation variables were also generally largest in the first half of intervention period.

\*\*\* *Insert Table 1 about here*\*\*\*

### *Product-of-coefficients analyses*

Corresponding to the within change analyses reported above, time significantly predicted changes in both ADHD, as measured by the CRS ( $B=-1.5, p<.001$ ), and conduct problems, as measured by the ECBI ( $-12.4, p<.001$ ). Results of the mediational analyses are seen in Table 3. Two of the parent variables, parental self-efficacy (PSOC) and negative and harsh parenting (PPI NEG PAR) showed a significant indirect ( $a \times b$ ) effect on both outcomes; parental self-efficacy explained 81% and 28% of the total effect on ADHD symptoms and conduct problems, respectively; negative and harsh parenting explained 29% in both cases. WAI also showed an indirect effect on ADHD symptoms explaining 7% of the effect.

### *Time-lagged analyses*

Neither of the three variables with significant indirect effect in the product-of-coefficients analyses, that is parental self-efficacy (PSOC), negative and harsh parenting (PPI NEG PAR) or therapeutic alliance (WAI), predicted later outcomes in the time-lagged analyses ( $ps>.05$ ). However, in the reverse analyses, exploring the opposite direction of causality, ADHD symptoms at mid-treatment

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significantly predicted self-efficacy at post-treatment (after controlling for the variables' prior values) ( $B=-0.4$ ,  $p=.037$ ). This was not the case for conduct problems ( $B<0.1$ ,  $p=.132$ ) or therapeutic alliance ( $B<0.1$ ,  $p=.133$ ). None of the two outcome measures predicted negative parenting ( $ps>.05$ ).

### *Further analyses on therapeutic alliance*

Therapeutic alliance (WAI) measured in session 3 only significantly predicted pre-post residualized change in one of the six criterion variables, appropriate discipline ( $z=2.54$ ;  $p=.011$ ;  $r=.32$ ); and measured mid-treatment it only predicted positive parenting ( $z=2.31$ ;  $p=.021$ ;  $r=.29$ ). Mid-treatment alliance did not predict mid-post change on positive parenting ( $z=1.28$ ;  $p=.201$ ;  $r=.16$ ). Range of ESs (Pearson's  $r$ ) for the non-significant analyses was  $-.04$  to  $.22$ .

## **Discussion**

The product-of-coefficient analyses revealed indirect mediational effects of two of the four parenting variables, namely increases in parental self-efficacy (PSOC) and reductions in harsh and negative parenting (PPI NEG PAR), on both symptoms of ADHD (CRS) and conduct problems (ECBI). Parental self-efficacy appeared to be the strongest mediating variable, explaining 81% of the change in ADHD symptoms. There was no significant indirect effect of appropriate discipline or positive parenting on child outcomes.

These findings largely correspond to previous findings in studies of statistical mediation, although there is some variation in previous results. Reductions in negative parenting and increased parental self-efficacy were also found to be statistical mediators of the treatment effect in the Seabra-Santos et al. (2016) study of IY PT for early ADHD as in the present study. Hinshaw et al. (2000) in the MTA study, found that reduced negative parenting, and not increased positive parenting, mediated

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the efficacy of PT, albeit only in the PT+ medication group. Hanisch et al. (2014) found statistical mediation on both reduced negative parenting and increased positive parenting.

Considering that IY PT (and most other PT programs) specifically targets harsh and negative parenting and aims to increase self-efficacy in parents, it is coherent with the underlying assumptions in PT that these parenting variables will be associated with the outcome on child symptoms (Tarver, Daley & Sayal, 2015). Parental self-efficacy is generally associated with secure attachment feelings towards the child (Lovejoy, Verda & Hays, 1997), persistence in strategies (Bandura, 1977), greater satisfaction with the parental role (Sanders et al., 2006) and increased likelihood of engaging in difficult parenting tasks (Tarver et al., 2015), all contributing to positive parent-child interactions. Likewise, reductions in negative parenting may improve parent-child interactions over time and prevent aggression and coercion cycles (Patterson, 2002).

Appropriate discipline did not improve over the PT course, contrary to what could be expected. However, appropriate discipline did not improve in the Danish study by Trillingsgaard and colleagues (2014) either, whereas it increased in Webster-Stratton et al. (2011), possibly suggesting that disciplinary styles diverge due to cultural differences.

In the time-lagged analyses, taking the time-line of change into consideration, none of the statistical mediators in the product-of-coefficients analyses predicted subsequent reductions in child ADHD or conduct problems. On the contrary, one of the two reversed time-lagged analyses showed that reductions in ADHD symptoms from pre to mid-intervention significantly predicted increased parental self-efficacy from mid- to post-intervention. None of the other reversed analyses revealed significant results.

The negative findings in the time-lagged analyses of the proposed mediators may, likely, be due to the fact that almost all of the change in both outcome variables took place from pre- to mid-

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intervention. With small and insignificant change in outcomes from mid- to post-intervention, there is limited possibility of mediational influence.

The reverse finding that decline in ADHD symptoms predicted improvement in parental self-efficacy indicates that when child symptoms were reduced, mothers perceived their parenting as more efficacious and competent. A child with less ADHD symptoms may be easier to parent (Theule et al., 2013) and since parental self-efficacy increases with a sense of successful mastery of the parental role (Coleman & Karraker, 1998) it is plausible that more successful parenting and thus more parental self-efficacy increased following child symptom reductions.

Since the children did not receive any other active treatment than PT (and ineffective TT), it seems unlikely that change in parental variables were not involved in the children's improvement. A possible speculation could be that changes in parental practices early in treatment led to symptomatic decline, which again led to improved self-efficacy. Such a reciprocal causal relation between mediators and outcome has been found in other studies with reversed time-lagged analyses (e.g., in a study on self-efficacy as a mediator in cognitive behavior therapy for panic disorder; Fentz et al., 2013). The design of the present study precludes, however, a test of this possibility.

In the product-of-coefficients analyses, change in therapeutic alliance over the intervention period came out as a statistical significant mediator of child ADHD symptoms, although only explaining 7 % of the variance. This was not the case for conduct problems. In the correlational analyses, 3<sup>rd</sup> session alliance only significantly predicted residualized pre-post change on one of eight outcomes and parental criteria variables, appropriate discipline; and mid-treatment alliance only predicted change in positive parenting. Due to the explorative character of the analyses, we did not make use of Bonferroni correction. Had we done so (e.g., dividing  $\alpha$ -values by 2 and 4, respectively, for the 2 outcome and 4 parent variable analyses), only the correlation between mid-treatment alliance and

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positive parenting would be significant ( $p = .012$ ). As mentioned in the introduction, ESs for the association between therapist-parent alliance and outcome in psychotherapy with children have generally been smaller than those reported for therapist-patient in the adult literature (McLeod, 2011), and one study even found a negative association between alliance and outcome in PT for children with externalizing problems (Hukkelberg & Ogden, 2013). It could be that the therapeutic alliance is less important in PT, being a rather didactic/educational intervention, than in more traditional forms of psychotherapy. However, it should be kept mind that parents generally rated the alliance high (e.g., a mean of 6.0 out of max 7 per item post-intervention), corresponding to the high degree of satisfaction with the PT intervention (Rimestad et al., 2016).

There are several limitations to this study. The lack of a passive control group hinders a firm conclusion as to the effectiveness of the PT intervention. Concerning missing data, there was a considerable non-completion of questionnaires at mid and post, corresponding to 14% and 19%, respectively. Furthermore, the study had only three data points thereby not capturing the change dynamics during the early sessions, which precluded conclusive time-lagged analyses. In addition, all measures were parent-reported with potential biases due to shared method variance of process and outcome variables, and to parents themselves being actively involved in treatment. Valid objective assessment of variables would have heightened the methodological quality of the study; however, objective assessment of ADHD symptoms within the field may be difficult to provide (e.g. Sonuga-Barke et al., 2013; Rimestad et al., 2016).

### **Conclusion**

Statistical mediation was found for two of the four proposed parental mediators, self-efficacy and negative and harsh parenting. Improved self-efficacy explained 81% of the change in ADHD symptoms and 28 % in conduct problems; reduced negative and harsh parenting explained 29% of

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the change in both outcome variables. However, time-lagged analyses with pre-, mid- and post-intervention data points did not find that prior change in mediators predicted subsequent change in children's symptoms. For one of the proposed mediators, a reversed pattern of mediation was found, indicating that parental self-efficacy increased as a consequence of prior reduction in ADHD symptoms. Therapeutic alliance between parents and group-leaders was only weakly related to parental variables or to outcome. The study is the first to apply time-lagged analyses of mediators in PT for ADHD. However, the few data points in the study preclude any firm conclusions as to causal mediation, especially concerning change early in the intervention. Future mediation studies would profit from more fine-grained time-lagged analyses with more (optimally session-wise) data points.

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## MEDIATION IN PARENT TRAINING FOR EARLY ADHD

**Table 1**

Within-group changes over time

	Pre-intervention		ESs pre- mid	Mid-intervention		ESs mid- post	Post-intervention		ESs pre- post
<i>Outcome</i>	M	(SD)		M	(SD)		M	(SD)	
<i>measures</i>									
CRS	23.27	(4.44)	0.63*	20.51	(6.26)	0.03	20.38	(7.14)	0.48*
ECBI	158.66	(24.52)	0.77**	141.02	(22.04)	0.30	134.50	(24.99)	0.94**
<i>Proposed mediators</i>									
WAI	68.41 <sup>a</sup>	(11.57)	0.14	70.66	(11.20)	0.27	71.85	(0.04)	0.33*
PSOC	61.90	(9.57)	0.45*	66.69	(10.54)	0.42*	69.47	(10.91)	0.76**
PPI APP DISC	3.65	(0.77)	0.36*	3.52	(0.69)	-0.28	3.63	(0.71)	-0.31
PPI POS PAR	4.51	(0.75)	1.06**	5.31	(0.68)	0.18	5.41	(0.62)	1.16**
PPI NEG PAR	2.51	(0.49)	0.44*	2.22	(0.55)	0.40*	2.05	(0.43)	1.0**

Note: All ESs are adjusted so that a positive change is indicated by a positive ES and a negative change is indicated by a negative sign. \*= significant at alpha = .05; \*\*= significant at alpha = .001.

<sup>a</sup>: measured after 3<sup>rd</sup> session

CRS: Conners Parent Rating Scale- Index; ECBI: Eyberg Child Behavior Inventory; PPI: Parental Practices Index; PPI APP DISC: Appropriate Discipline Subscale, PPI NEG PAR: Negative /harsh parenting subscale; PPI POS PAR: Positive Parenting Subscale; PSOC: Parental Sense of Competence; WAI: Working Alliance Inventory.

## MEDIATION IN PARENT TRAINING FOR EARLY ADHD

**Table 2**  
Product-of-coefficients analysis

	B	BSSE	95% BSCI	Total effect explained by mediator
<b>CRS</b>				
WAI	<b>-.10</b>	<b>.08</b>	<b>-.35 - &lt;-.01</b>	<b>7 %</b>
PSOC	<b>-.63</b>	<b>.17</b>	<b>-1.04 - -.33</b>	<b>81 %</b>
PPI APP DISC	<-.01	.05	-.13 - .11	<1 %
PPI POS PAR	-.25	.28	-1.16 - .24	17 %
PPI NEG PAR	<b>-.41</b>	<b>.20</b>	<b>-.81 - -.03</b>	<b>29 %</b>
<b>ECBI</b>				
WAI	-.07	.43	-.99 - .75	1 %
PSOC	<b>-3.30</b>	<b>1.35</b>	<b>-6.43 - -1.12</b>	<b>28 %</b>
PPI APP DISC	<-.01	.20	-.50 - .36	<1 %
PPI POS PAR	-.55	1.23	-3.23 - 1.72	5 %
PPI NEG PAR	<b>-3.34</b>	<b>1.03</b>	<b>-5.65 - -1.57</b>	<b>29 %</b>

Note: Significant mediators are in bold. BSSE: Bootstrapped standard error; BSCI: Bootstrapped confidence interval; CRS: Conners Parent Rating Scale- Index; ECBI: Eyberg Child Behavior Inventory; PSOC: Parental Sense of Competence; WAI: Working Alliance Inventory; PPI APP DISC: Parental Practices Index - Appropriate Discipline Subscale; PPI POS PAR: Parental Practices Index- Positive Parenting Subscale; PPI NEG PAR: Parental Practices Index- Negative and Harsh Parenting Subscale.

**Figure 1** The mediational model

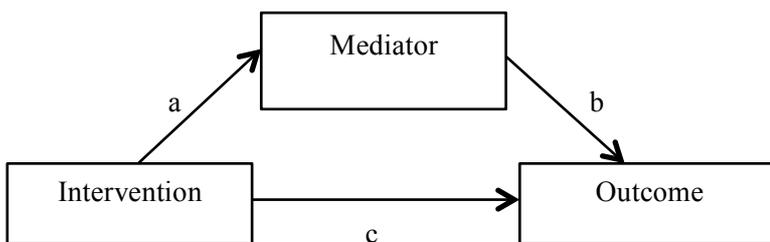


Figure 1. The Mediational model

## **Appendix D**

Author contribution statements

## Co-authorship declaration

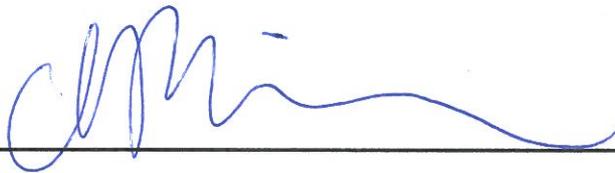
Manuscript title: "Short-and long term efficacy of parent training for preschool ADHD"

Authors: Marie Louise Rimestad (MLR), Rikke Lambek (RL), Helene Zacher Christiansen (HZC), Esben Hougaard (EH).

I hereby declare my contribution to the manuscript to be as described below:

MLR	RL	HZC	EH	
X			X	Conception and design of study
X		X		Collection and assembly of data
X	X		X	Data analysis and interpretation
X	X	X	X	Manuscript writing
X	X	X	X	Final approval of manuscript

MLR:



RL:



HZC:



EH:



## Co-authorship declaration

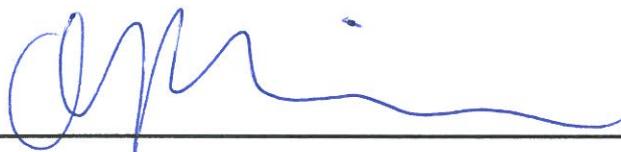
Manuscript title: "Combining parent and teacher training for early ADHD: a randomized study of effectiveness"

Authors: Marie Louise Rimestad (MLR), Tea Trillingsgaard (TT), Esben Hougaard (EH).

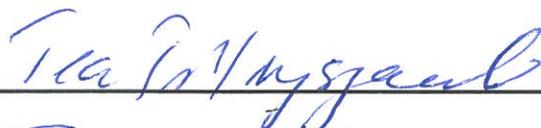
I hereby declare my contribution to the manuscript to be as described below:

MLR	TT	EH	
X	X	X	Conception and design of study
X			Collection and assembly of data
X	X	X	Data analysis and interpretation
X	X	X	Manuscript writing
X	X	X	Final approval of manuscript

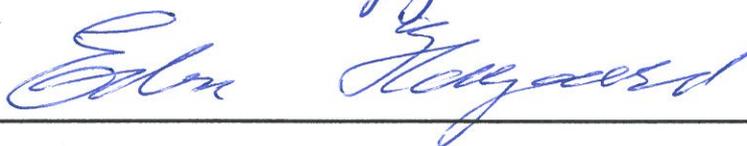
MLR:



TT:



EH:



## Co-authorship declaration

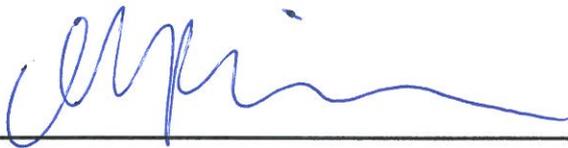
Manuscript title: "Mediators of change in a parent training program for early ADHD difficulties: the role of parental strategies, parental self-efficacy and therapeutic alliance"

Authors: Marie Louise Rimestad (MLR), Mia Skytte O'Toole (MSO), Esben Hougaard (EH).

I hereby declare my contribution to the manuscript to be as described below:

MLR	MSO	EH	
X		X	Conception and design of study
X			Collection and assembly of data
X	X	X	Data analysis and interpretation
X	X	X	Manuscript writing
X	X	X	Final approval of manuscript

MLR:



MSO:



EH:



