University of Groningen

Home-based Behavioral Parent Training for children with ADHD

Nobel, Ellen

DOI:
[10.33612/diss.182262024](https://doi.org/10.33612/diss.182262024)

IMPORTANT NOTE: You are advised to consult the publisher's version (publisher's PDF) if you wish to cite from it. Please check the document version below.

Document Version
Publisher's PDF, also known as Version of record

Publication date:
2021

[Link to publication in University of Groningen/UMCG research database](#)

Citation for published version (APA):

Nobel, E. (2021). *Home-based Behavioral Parent Training for children with ADHD: a follow-up treatment for non-responders to routine clinical treatment?* [Thesis fully internal (DIV), University of Groningen]. University of Groningen. <https://doi.org/10.33612/diss.182262024>

Copyright

Other than for strictly personal use, it is not permitted to download or to forward/distribute the text or part of it without the consent of the author(s) and/or copyright holder(s), unless the work is under an open content license (like Creative Commons).

The publication may also be distributed here under the terms of Article 25fa of the Dutch Copyright Act, indicated by the "Taverne" license. More information can be found on the University of Groningen website: <https://www.rug.nl/library/open-access/self-archiving-pure/taverne-amendment>.

Take-down policy

If you believe that this document breaches copyright please contact us providing details, and we will remove access to the work immediately and investigate your claim.

Downloaded from the University of Groningen/UMCG research database (Pure): <http://www.rug.nl/research/portal>. For technical reasons the number of authors shown on this cover page is limited to 10 maximum.

Home-based behavioral parent training for children with ADHD

A follow-up treatment for non-responders
to routine clinical treatment?

Ellen Nobel

The research described in this thesis was financially supported by the Netherlands Organization for Health Research and Development (ZonMw, nr 15700.3010).

The author thanks the participating families, clinicians, and the health care centers Accare, Jonx Lentis, and GGZ In de Bres.

Copyright © Ellen Nobel, 2021

Design cover: proefschriften.net

Printed by proefschriften.net

All rights are reserved. No part of this book may be reproduced, stored in a retrieval system, or transmitted in any form or by any means, without the written permission from the author.

rijksuniversiteit
 groningen

Home-based behavioral parent training for children with ADHD

A follow-up treatment for non-responders to routine clinical treatment?

Proefschrift

ter verkrijging van de graad van doctor aan de
Rijksuniversiteit Groningen
op gezag van de
rector magnificus prof. dr. C. Wijmenga
en volgens besluit van het College voor Promoties.

De openbare verdediging zal plaatsvinden op

woensdag 27 oktober 2021 om 14.30 uur

door

Ellen Nobel

geboren op 30 oktober 1983
te Groningen

Promotor(es)

Prof. dr. P.J. Hoekstra

Prof. dr. B.J. van den Hoofdakker

Beoordelingscommissie

Prof. dr. J.K. Buitelaar

Prof. dr. S.A. Reijneveld

Prof. dr. R.A. Schoevers

Table of contents

Chapter 1	General introduction	7
Chapter 2	Home-based parent training for school-aged children with attention-deficit/hyperactivity disorder and behavior problems with remaining impairing disruptive behaviors after routine treatment: a randomized controlled trial	27
Chapter 3	Effects on parenting parameters of a home-based parent training for children with attention-deficit/hyperactivity disorder with remaining disruptive behaviors after routine treatment: a randomized controlled trial	57
Chapter 4	Which improvements in parenting mediate effects of home-based behavioral parent training for school-aged children with attention-deficit/hyperactivity disorder?	77
Chapter 5	Parent-clinician agreement in rating presence and severity of attention-deficit/hyperactivity disorder symptoms	93
Chapter 6	Summary and general discussion	111
	References	127
	Samenvatting in het Nederlands	139
	Dankwoord	149
	Curriculum Vitae	157

Chapter 1

General introduction

The main goal of the trial, described in this dissertation, was to investigate the short- and long-term effectiveness of a *home-based* behavioral parent training (BPT) as a treatment for *school-aged children* with attention-deficit/hyperactivity disorder (ADHD) *who had responded insufficiently to evidence-based treatments* (i.e., pharmacotherapy and/or clinic-based BPT). This introductory chapter presents an outline of the relevance of this trial. The chapter starts with a brief overview of adverse consequences for present and future functioning of children with ADHD, illustrating the need for treatment. Further, the effectiveness of current treatments for school-aged children with ADHD is described. Next, the choice of home-based BPT as follow-up treatment for children who responded insufficiently to evidence-based treatments, and the content of the newly developed treatment is motivated. Finally, the aims of the trial, the study design, and the outline of the dissertation are described.

Attention-deficit/hyperactivity disorder: impairment and comorbidity

Children with ADHD are characterized by persistent inattentiveness and/or hyperactivity-impulsivity across different situations, starting in childhood (American Psychiatric Association, 2013). These characteristics are present to an extent beyond what would be expected for the child's developmental level; to warrant a diagnosis the symptoms should be associated with significant impairment in different settings. ADHD is one of the most common mental disorders in childhood and adolescence (Polanczyk, de Lima, Horta, Biederman, & Rohde, 2007; Wittchen et al., 2011). Multiple systematic reviews established a population prevalence of around 2.2-7.2% (Polanczyk et al., 2007; Willcutt, 2012; Erskine et al., 2013; Polanczyk, Willcutt, Salum, Kieling, & Rohde, 2014; Polanczyk, Salum, Sugava, Caye, & Rohde, 2015; Thomas, Sanders, Doust, Beller, & Glasziou, 2015). For up to 50-65% of the children with ADHD the disorder persists into adulthood (Lara et al., 2009; Faraone, Biederman, & Mick, 2006).

ADHD may be associated with a broad range of functional impairments, such as academic underachievement (Barbarese, Katusic, Colligan, Weaver, & Jacobsen, 2007; McGee, Prior, Williams, Smart, & Sanson, 2002; Erskine et al., 2016), lower socio-

economic status (Erskine et al., 2016), negative social interactions (DuPaul, McGoey, Eckert, & VanBrakle, 2001), impaired relationships with peers (Hinshaw, 2002; Mrug, Hoza, & Gerdes, 2001; Mrug et al., 2009), and disturbances in family functioning (Johnston & Mash, 2001); all of which may lead to a reduced quality of life (Danckaerts et al., 2010; Faraone et al., 2015). Children with ADHD are also at increased risk of trauma-related events due to accidents (Ertan, Özcan, & Pepele, 2012; Leibson et al., 2006; Shilon, Pollak, Aran, Shaked, & Gross, 2012), bullying (Holmberg & Hjern, 2008), and sexual abuse (Cengel-Kültür, Cuhadaroglu-Cetin, & Gökler, 2007; Briscoe-Smith & Hinshaw, 2006). There is a greater long-term risk of suicide attempts (Bussing, Mason, Bell, Porter, & Garvan, 2010) and delinquency as well (Erskine et al., 2016; Bussing et al., 2010; Vreugdenhil, Doreleijers, Vermeieren, Wouters, & Van den Brink, 2004).

ADHD is associated with a significant social and economic burden. The disorder is one of the top 100 of causes of global burden (objectified with disability-adjusted life years; Erskine et al., 2014) and in Europe the per-person ADHD related costs for children and adolescents are around €9,860- €14,483 equivalent with total national annual ADHD-related costs of €1,1041- €1,529 million (Le et al., 2014). Notably, only 8-25% of these costs are related to child mental health care, while about 30% come from family members' health care and productivity loss. Potentially, more effective treatments can have positive effects on these ADHD-related costs.

The comorbidity-rate for ADHD is very high; approximately 75% of individuals with ADHD have a comorbid disruptive disorder or internalizing disorder, and around 60% have more than one comorbid disorder. Approximately, 35-60% of the children with ADHD have a comorbid oppositional defiant disorder (ODD), 30-50% have a comorbid conduct disorder (CD), and 20-40% have an anxiety and/or mood disorder (Jensen & Steinhausen, 2015; Taurines et al., 2010; Biederman, Newcorn, & Sprich, 1991; Wells et al., 2006). ADHD is also a risk factor for developing a substance use disorder (Szobot et al., 2007). Comorbid anxiety disorders and ODD are prevalent in childhood (Taurines et al., 2010), whereas mood disorders and CD often develop towards the end of the elementary school years. During adolescence and adulthood, ADHD is often associated with personality disorders (Sobanski et al., 2010; Biederman et al., 1991) and substance abuse or dependence (Erskine et al., 2016).

In sum, ADHD in children is associated with a range of adverse consequences in present and future functioning, including the development of comorbid disorders. Therefore, treatment in early stages is important, as this may have a positive effect on a downward developmental pathway of functioning.

ADHD and parenting

Having a child with ADHD is associated with parents missing/altering work, avoiding social activities, and increased parental worries/stress (Fridman et al., 2017, Anastopoulos, Guevremont, Shelton, & DuPaul, 1992; Pimentel, Vierira-Santos, Santos, & Vale, 2011); all of this despite the use of ADHD pharmacotherapy (Fridman et al., 2017). Compared to parents with children with other neurodevelopmental disorders (autism spectrum disorder, learning disorders or language impairments), parents of children with ADHD (together with those of children with autism spectrum disorder) report the highest levels of parenting stress (Craig et al., 2016). In parents of children with ADHD, parenting stress is associated with children's ADHD symptom severity (Anastopoulos et al., 1992; Fridman et al., 2017; Craig et al., 2016), especially the severity of the hyperactive-impulsive symptoms (Graziano, McNamara, Geffken, & Reid, 2011). Also, the coexistence with comorbid problems and maternal psychopathology may influence the level of parenting stress (Anastopoulos et al., 1992). Furthermore, the combination of a child with ADHD and comorbid behavioral problems and a parent with a rejective parenting style is negatively associated with parenting stress levels (Pimentel et al., 2011).

Studies showed that parental attitudes and parenting practices have a stronger relationship with children's comorbid behavior problems than with ADHD symptoms. Parental negative attitudes to their child, lack of warmth, and positive parenting, negative discipline strategies, and parental intrusiveness are predictive for the presence of oppositional-defiant and conduct behavior (Deault, 2010; Kaiser, McBarnett, & Piffner, 2011). Parental overprotectiveness and lack of positive parenting towards children with ADHD are associated with child anxiety, while inconsistent parenting is a risk factor for depression (Deault, 2010). Moreover, better parenting skills are associated

with positive social functioning and better academic achievement of the child (Deault, 2010; Kaiser et al., 2011).

In conclusion, parenting stress, attitudes and practices are associated with the severity of the child's ADHD, the presence of comorbidity, and the social and academic functioning of the child. Therefore, it seems desirable (especially in severe and complex cases) - and is already common practice - that treatments not only focus on the child's behavior and well-being; also these parenting parameters deserve attention.

Treatment of school-aged children with ADHD

Pharmacotherapy

Pharmacotherapy is a widely used treatment for school-aged children with ADHD. The efficacy of medical treatment (especially of psychostimulants) is well established through multiple systematic reviews and meta-analyses. These studies found large reductions on the severity of ADHD symptoms (effect sizes [ESs] varied between .52-1.02) and relevant functional impairments (Cortese et al., 2018; Keen & Hadjikhouri, 2011; Schachter, Pham, King, Langford, & Moher, 2001; Riera et al., 2017; Banaschewski et al., 2006; Van de Loo-Neus, Rommelse, & Buitelaar, 2011).

However, medical treatment for school-aged children with ADHD has its limitations. First, the response to pharmacotherapy is not always sufficient. Psychostimulants rarely lead to normalization of ADHD behavior (Banaschewski et al., 2006) and are less effective when the child has (multiple) comorbid problems and when the child is older (Riera et al., 2017). Moreover, results of meta-analyses suggest that the long-term effects of medication may be less pronounced (Riera et al., 2017), or hardly lead to normalization on various domains of functioning (Shaw et al., 2012). Furthermore, some studies reported no significant results for medical treatment on outcomes such as social functioning, academic achievement, disruptive/aggressive symptoms, internalizing problems, and the parent-child relationship (e.g., Jensen et al., 2001; The MTA Cooperative Group, 1999; Van de Loo-Neus et al., 2011; Shaw et al., 2012). Meanwhile, as outlined before, these functional impairments and comorbid problems are very commonly reported in children with ADHD and may contribute to the economic burden of the disorder. Second, medical

treatment is not always an option, because of adverse effects (such as insomnia, reduced appetite, slow growth, stomach ache, headache, and dizziness; Cortese et al., 2018; Schachter et al., 2001; Graham et al., 2011) or negative attitudes that parents and clinicians may have on this type of treatment (Schatz et al., 2015; Berger, Dor, Nevo, & Goldzweig, 2008).

In sum, the efficacy for pharmacotherapy for school-aged children with ADHD is well established. However, in clinical practice, pharmacotherapy is sometimes not viable and often not sufficient. Consequently, there is a need to look for other effective treatments beyond pharmacotherapy.

Behavioral Parent Training

BPT is a well-established treatment for school-aged children with ADHD. The objective of the treatment is to reduce ADHD symptomatology and noncompliant behavior of the child by teaching parents behavioral management principles to address these at home. Typically, traditional BPT programs are manualized, provided as group format, and exist of eight to twelve sessions on a weekly basis performed in an outpatient setting (e.g., Barkley's Defiant Children [Barkley, 1987]). Programs usually start with providing psychoeducation about ADHD, child disruptive problems, and general behavioral management principles. This is followed by teaching parents behavioral techniques, such as reinforcement skills, (e.g., positive attending and the use of comprehensive, token/point systems). Also, the use of planned ignoring and punishment strategies is discussed, for example the implementation of response cost and time out. Between sessions, parents practice with these skills through homework assignments and are given feedback at the next session.

In the last decades, some BPT programs for parents of school-aged children with ADHD were developed for specific target groups, e.g., the STEPP (Strategies to Enhance Positive Parenting) program for single-mothers (Chacko et al., 2009), the COACHES (Coaching Our Acting-out Children: Heightening Essential Skills) program for fathers (Fabiano et al., 2012) and a program for mothers with mild depression (Chronis-Tuscano et al., 2013). In addition, alternative delivery methods were developed, such as a guided self-help program (Dose et al., 2017) or treatment by video conference (Xie et al., 2013).

Effectiveness of BPT

To our knowledge, no systematic reviews or meta-analyses on behavioral parent training for children with ADHD have focused specifically on school-aged children. Most systematic reviews and meta-analyses included preschool and school-aged children together, mostly with an overrepresentation of preschool children (e.g., Evans, Owens, Wymbs, & Ray, 2018; Pelham & Fabiano, 2008; Sonuga-Barke et al., 2013; Daley et al., 2014; Coates, Taylor, & Sayal, 2015). A meta-analysis of Daley and colleagues (2014) showed that ADHD and conduct problems improved more in younger than in older children after behavioral treatment, whereas most interventions in this meta-analysis included a BPT component. Therefore, the conclusions from the systematic reviews and meta-analyses on the effectiveness of BPT that included preschool as well as school-aged children were perhaps too positive for school-aged children. This is why we focus here on randomized controlled trials (RCT) that have investigated the effectiveness of BPT for our specific age group (i.e., Chacko et al., 2009; Chronis-Tuscano et al., 2013; Corkum, Mckinnon, & Mullane, 2005; Fabiano et al., 2012; Hoath & Sanders, 2002; Dose et al., 2017; Rajwan, Chacko, Wymbs, & Wymbs, 2014; Van den Hoofdakker et al., 2007; Xie et al., 2013). Table 1 provides a summary of treatment characteristics, inclusion criteria, and design specifications of these nine trials. Six of these trials investigated BPT without child training (Chronis-Tuscano et al., 2013; Corkum et al., 2005; Dose et al., 2017; Hoath & Sanders, 2002; Van den Hoofdakker et al., 2007; Xie et al., 2013), whereas three trials evaluated programs in which BPT was combined with child-therapy (targeting mostly social skills; Chacko et al., 2009; Fabiano et al., 2012; Rajwan et al., 2014). Only two of the nine studies determined the effectiveness of a traditional BPT program compared to a waiting list (Hoath & Sanders, 2002; Van den Hoofdakker et al., 2007). The other seven studies investigated the effectiveness of a target-group-adjusted-program. Most of these studies compared the adjusted treatment with a traditional BPT, except the studies of Fabiano and colleagues (2012) and Dose and colleagues (2017) that compared adjusted BPT with a waiting list. The study of Chacko and colleagues (2009) compared a specially developed BPT with traditional BPT and a waiting list. Most investigated BPTs were group-based, only Dose and colleagues (2017) examined an individualized format. All studies targeted children with ADHD and common comorbidities and medication use was

Table 1. Overview of randomized controlled trials that investigated the effectiveness of behavioral parent training (BPT) for school-aged children with ADHD

Study	Characteristics BPT					Inclusion criteria			Design		
	BPT alone ¹	Traditional /adjusted ²	Group/ individual	Timing of treatment ³	Delivery location	Sample type	Comor-bidity	Pharmaco-therapy?	N	Control group	Blind treatment assignment?
Chacko et al., 2009	-	Adjusted	Group	First-line	Clinic-	Non-referred	✓	✓	120	Active ⁴ & Waiting list	-
Chronis-Tuscano et al., 2013	✓	Adjusted	Group	First-line	Guided self-help	Non-referred	✓	✓	98	Active ⁴	-
Corkum et al., 2005	✓	Adjusted	Group	First-line	Clinic	Referred	✓	✓	30	Active ⁴	-
Dose et al., 2017	✓	Adjusted	Individual	First-line	Clinic	Referred	✓	✓	103	Waiting list	-
Fabiano et al., 2012	-	Adjusted	Group	First-line	Clinic	Non-referred	✓	✓	55	Waiting list	-
Hoath et al., 2002	✓	Traditional	Group	First-line	Clinic	Referred	✓	✓	20	Waiting list	-
Rajwan et al., 2014	-	Adjusted	Group	First-line	Clinic	Non-referred	✓	Unknown	80	Active ⁴	-
Van den Hoofdakker et al., 2007	✓	Traditional	Group	First-line	Clinic	Referred	✓	✓	94	Waiting list	-
Xie et al., 2013	✓	Adjusted	Group	First-line	Video conference	Referred	✓	✓	22	Active ⁴	-
This study	✓	Adjusted	Individual	Second-line	Home	Referred	✓	✓	73	Active & Waiting list	Partial

Note: ✓ = yes; - = no; ¹ BPT not in combination with other psychosocial intervention; ² Traditional BPT was defined as BPT designed for all children with ADHD, Adjusted BPT was defined as BPT developed for a specific subgroup of children with ADHD; ³ BPT offered as first- or second-line nonpharmacological treatment; ⁴ Active control group was traditional BPT.

allowed during the studies (mostly with the constraint that medication had to remain stable during the trial).

*Child outcomes

Considering the above mentioned RCTs, the following child outcomes were reported: first, four of the five trials that compared BPT with a waiting list (all using parent ratings) found no difference in the reduction of ADHD symptoms between the BPT group and the control group (Van den Hoofdakker et al., 2007; Dose et al., 2017; Chacko et al., 2009; Hoath & Sanders, 2002) and one did not include an ADHD outcome measure (Fabiano et al., 2012). Of the trials that compared adjusted BPT to traditional BPT, two found a significant effect on ADHD symptom reduction in favor of the adjusted BPT (Corkum et al., 2005; Xie et al., 2013), one found no difference between the treatment conditions (Chacko et al., 2009), and two did not include the severity of ADHD symptoms as outcome measure (Chronis-Tuscano et al., 2013; Rajwan et al., 2014). Second, all nine studies reported that BPT was effective on the reduction of oppositional and conduct problems (ES's varying between .43-.75) compared to the control condition, except for the study of Corkum and colleagues (2005); in all studies this considered parent rated measures. Third, only Xie and colleagues (2013) investigated the effect of BPT on social functioning and found a significant improvement of videoconference mediated BPT compared to traditional BPT. Fourth, four studies investigated the effect on overall impairment of the child (parent-rated: Chacko et al., 2009, Chronis-Tuscano et al., 2013, and Dose et al., 2017; clinician-rated: Xie et al., 2013) and all found significant effects of BPT compared to the control condition (ES's varying between .68-.81), except for the study of Dose and colleagues (2017), which found no significant effect. Fifth, Van den Hoofdakker and colleagues (2007) and Xie and colleagues (2013) reported significant effects of BPT compared to control on parent-rated internalizing problems of the child. At last, Van den Hoofdakker and colleagues (2007) also found that the control group (routine clinical care only) received significantly more polypharmacotherapy than the group that was assigned to BPT with routine clinical care.

In conclusion, traditional BPT for school-aged children with ADHD is effective on parent-rated externalizing and internalizing problems and general functioning of the child. In this

specific age group, there seem to be no significant effects of traditional BPT on ADHD symptoms. Moreover, BPT programs that were adjusted for specific target groups (depressed mothers, fathers, single-mothers) seem to have additional value for the reduction of externalizing and internalizing problems and improvement of general functioning of the child.

* Parent outcomes

The aim of BPT is to improve the child's behavior and functioning by teaching parents behavioral management skills. Therefore, most trials included parenting outcome measures, such as parenting practices/skills, parenting stress, and parental mental health. Looking again at the previously mentioned RCTs that investigated the effectiveness of BPT for school-aged children, six of the nine RCTs investigated the effectiveness of BPT on parenting skills and they all found a positive effect of BPT compared to a control condition on negative parenting (ES's varying between .19-.68) and almost all on positive parenting (ES varying between .54-.81; Fabiano et al., 2012; Chacko et al., 2009, Hoath & Sanders, 2002; Xie et al., 2013; Chronis-Tuscano et al., 2013; Dose et al., 2017). Three of these six studies that found positive effect of BPT on parenting skills were trials that compared BPT for a specific target group with traditional BPT. Two trials investigated the effect of BPT on parenting stress, leading to inconsistent results: Van den Hoofdakker and colleagues (2007) did not find an effect, whereas Chacko and colleagues (2009) found significant results by comparing a BPT for a specific target group with traditional BPT and a waiting list group. Only one study investigated the effect of BPT, compared to waitlist, on parenting competence and found a significant effect in favor of BPT (Hoath & Sanders, 2002). Furthermore, Chacko and colleagues (2009) and Hoath and Sanders (2002) did not find an effect on parental mental health, but in a study in which BPT was combined with a cognitive-behavioral treatment for depressed mothers, improvement of maternal depression was established (Chronis-Tuscano et al., 2013).

In conclusion, BPT improves parenting practices and has probably a positive effect on parenting competence. Inconsistent results were found for the effects of BPT on parenting stress. Again, BPT programs that were designed for a specific target group (i.e., depressed mothers, fathers, single-mothers) seem to have additional value for the

reduction of negative parenting and parenting stress, and the improvement of positive parenting and parental mental health.

Combination of pharmacotherapy and BPT

It is reasonable to presume that the combination of pharmacotherapy and BPT is best recommended for school-aged children with ADHD, in particular for severe cases with high levels of functional impairment and comorbid problems. Various studies, including systematic reviews and meta-analyses, confirm this line of thought: combined treatment has shown to be more effective than medication management or behavioral/psychological treatment (in most cases at least BPT) alone on ADHD symptoms, disruptive behaviors, internalizing problems, social functioning, academic achievement, and the parent-child relationship (Jensen et al., 2001; The MTA Cooperative Group, 1999; Keen & Hadjikhouri, 2011; Reale et al., 2017; Catalá-López et al., 2017). A positive effect of combined treatment on behavioral functioning, evaluated by a clinician, has not only been found for children with ADHD alone, but also for ADHD children with comorbid disorders ($ES = .75$), especially for children with comorbid ODD ($ES=.98$) or anxiety ($ES=1.61$; Reale et al., 2017), and when a child with ADHD has both of these comorbidities (Jensen et al., 2001). As a result, clinical guidelines for ADHD advice a combined treatment of medication and BPT for school-aged children (Pliszka, 2007; American Academy of Pediatrics, 2011; CADDRA Canadian ADHD Resource Alliance, 2018; Taylor et al., 2004; NHS Quality Improvement Scotland, 2009).

Treatment for children with remaining impairments

Providing pharmacotherapy and BPT for school-aged children with ADHD is not always sufficient. Clinicians see a sizable proportion of parents who still ask for help with the behavioral problems of their child *at home*. These families did not benefit from previous treatments for a variety of reasons, including insufficient short-term response to treatment, restricted long-term effectiveness, or problems getting the treatment organized (Koerting et al., 2013; Assenany & McIntosh, 2002). The question then arises what treatment should be offered in those cases.

Other treatments

Although it is reasonable to look for a different treatment when BPT and pharmacotherapy were insufficient, different trials investigating these various treatment-options showed disappointing results. For example, for treatments directed at the child (such as social skills training, play therapy, cognitive therapy, or neurofeedback) minimal or no treatment effects on ADHD symptoms or functioning level were found (especially when trials used probably blinded observations; e.g., Pelham & Fabiano, 2008; Sonuga-Barke et al., 2013; Cortese et al., 2016). For the relatively new meditation-based therapies for ADHD (such as mindfulness and yoga), a review concluded that most trials were single armed, and therefore no definitive conclusions on their effectiveness could be made. Two of the three controlled studies that were included in this review showed no effect of meditation-based therapies on the severity of ADHD symptoms (Evans et al., 2018).

Summer treatment programs, in which children receive multiple, daily therapies during several weeks, are other well-established treatments for school-aged children with ADHD (Pelham & Fabiano, 2008). However, these are quite intensive treatment programs and as far as we know not available in the Netherlands. Finally, a meta-analysis showed the limited effectiveness of dietary treatments on probably blinded ratings of severity of ADHD symptoms, namely food color exclusion ($ES=.42$) and free fatty acid supplements ($ES=.16$). The effect size of the food color exclusion was likely an overestimation of the efficacy due to selection bias; only studies were included in which children with ADHD had an established sensitivity for food colors. The effect size of free fatty acid supplements was small and interpreted as clinically irrelevant (Sonuga-Barke et al., 2013).

Recommendations for enhancing BPT

As the effects of other treatments for ADHD are disappointing, they are not viable follow-up treatments when children with ADHD keep problems at home after at least an attempt of the known evidence-based treatments pharmacotherapy and/or BPT. Then it may be reasonable to try to strengthen the BPT, of which we know it is an effective treatment for at least a portion of the children.

Two meta-analyses investigated whether specific treatment elements strengthened the effect of BPT in preschool and school-aged children with externalizing behaviors.

These meta-analyses showed that teaching parents to positively interact with their child and practicing with their child during treatment sessions were associated with improvements in parenting skills and children's externalizing behaviors. Also teaching parents skills such as positive reinforcement, praise, time-out and other non-violent discipline techniques and responding consistently to the child were associated with improvements in child behaviors. Teaching parents emotional communication and problem-solving skills, and informing parents about ancillary services were all related to improvements in parenting skills (Kaminski, Valle, Filene, & Boyle, 2008; Leijten et al., 2018).

Although understudied, an important barrier for the effectiveness of BPT is poor attendance to the therapy. It is not uncommon that up to 50% of the parents with children with disruptive behavioral disorder do not fully attend the treatment (Chacko et al., 2016; Assenany & McIntosh, 2002; Koerting et al., 2013;). Several factors have been found to influence treatment attendance. First, several family characteristics have shown to be associated with low attendance of BPT: low family income, low maternal education level, single-parenthood, and large family size. Also poor housing, unemployment, and belonging to a minority ethnicity may influence attendance (Chacko et al., 2016; Cunningham et al., 2000; Assenany & McIntosh, 2002). Families with lower social-economic status experience more difficulties in fitting with the organizational demands of clinic-based BPT, because they often have inflexible and demanding work schedules, lack of childcare coverage and unreliable transportation (Koerting et al., 2013; Assenany & McIntosh, 2002). In line with this, Cunningham, Bremner, and Boyle (1995) found that families with children with more severe behavioral problems and families from different cultural backgrounds were more likely to attend parent training if it was located in the neighborhood school rather than in local clinics. Moran and Ghate (2005) advised that parenting programs should include home visits to enhance attendance. Another factor that has found to be associated with lower attendance is family dysfunction: parental psychopathology, marital violence, and adverse and harsh child rearing practices. Also, a negative image of the child by the parents is associated with lower attendance (Cunningham et al., 2000; Assenany & McIntosh, 2002). Finally, the severity of the child's externalizing behavior (especially conduct disorder symptoms) is associated with lower attendance to BPT (Assenany & McIntosh, 2002).

In sum, to enhance BPT outcomes it may be crucial to make it easier for families to attend the treatment and to prevent early treatment dropout. Lowering organizational demands for these families could help overcome the practical problems of transportation, inflexible working hours, and lack of childcare coverage. When sessions take place at home, in vivo practice with the child can also be done more easily and frequently. In addition, the home setting is particularly suitable to provide flexible and individually tailored programs, which parents highly appreciate (Koerting et al., 2013)

Behavioral Parent Training Groningen @HOME

We developed a home-based BPT specifically designed for families with a child with ADHD and disruptive behavior who did not benefit sufficiently from (kept remaining problems) after psychopharmacotherapy and clinic-based BPT, irrespective of the cause of this insufficient response (insufficient short-term response to treatment, restricted long-term effectiveness, or problems in getting the treatment organized). This home-based treatment, called Behavioral Parent Training Groningen at HOME (BPTG@HOME), is a manualized, cognitive-behavioral home-based treatment for children with ADHD and behavior problems. The treatment consists of fourteen to sixteen weekly home visits (90-120 minutes per session), spread over four months. Between every home visit, the therapist and parents have two contacts through telephone or email in which homework assignments are evaluated. In most home visits, parents receive feedback using video recordings on the skill they have been practicing that week. Furthermore, psychoeducation and cognitive interventions are included in the sessions.

BPTG@HOME is divided into different treatment modules. The first module is an assessment module in which the therapist determines which treatment modules and how many sessions per module should be given. This is based on treatment goals of the family, rating scales, and the therapist's assessment during the first session. The subsequent three basic modules Following, Leading, and Compliance are provided to all families. These modules include three to six sessions; the number of sessions depends on whether the therapist considers the parents need more practice of a certain skill. The module Following aims to improve the parent-child relationship through implementing

Table 2. Outline of the content of BPTG@HOME

Treatment module	Obligatory sessions	Additional sessions
Basic modules		
Assessment 2 sessions	1. Assessment 2. Treatment plan	
Following 3-6 sessions	1. Play time 2. Parental cognitions 3. Practicing play time	4. Practicing play time extended 5. Practicing play time extended 6. Practicing play time extended
Leading 3-6 sessions	1a. Providing structure 2. Praise 3a. Communication skills	1b. Providing structure extended 3b. Communication skills extended 4. Providing structure/Praise/ Communication skills extended
Compliance 3-5 sessions	1. Assessment of non-compliant behavior 2a. House rules 3a. Reward and punishment	2b. House rules extended 3b. Reward and punishment extended
Additional modules		
Disagreement between parents regarding handling the child 0-2 sessions		1. Dealing with conflicts 2. Communication with spouse
Aggressive parental behavior to the child 0-2 sessions		1. Dealing with parental frustration 2. Dealing with parental frustration extended
Anxious or depressed child 0-2 sessions		1. Dealing with an anxious child 2. Dealing with a child with depressive symptoms
Other problematic behaviors of the child 0-x sessions		1a. Dealing with specific behavioral problem 1 1b. Dealing with specific behavioral problem 1 extended 2a. Dealing with specific behavioral problem 2 2b. Dealing with specific behavioral problem 2 extended ...
Final basic module		
Maintenance training 3 sessions	1. Evaluation 2. Follow-up 1 3. Follow-up 2	

daily playtime with the child. In this module, parents are taught to establish a positive interaction with their child through praise, being friendly and attentive, and avoiding instructions, questions, and criticism. In the module Leading, parents learn to prevent disruptive behaviors of their child. The last basic module, Compliance, teaches parents

how to cope with noncompliant behavior of their child. After the basic modules, the therapists can select additional treatment modules, which focus on remaining parenting problems, such as disagreement between parents regarding handling the child or dealing with their child's internalizing problems. After a maximum of fifteen sessions, the treatment is finished with a final session in which the therapy is evaluated and in which parents plan future goals. Within three months, two follow-up sessions at home are planned aimed at maintaining learned skills. Table 2 shows the outline of the treatment in detail.

The basic modules of BPTG@HOME are based on the principles of Barkley's Defiant Children (Barkley, 1987), the program of Forehand & McMahon (1981, *Helping the Noncompliant Child*), and Parent-Child Interaction Therapy (Eyberg & Funderburk, 2011). The additional modules include basic behavioral interventions (such as exposure techniques for child anxiety and behavior activation for depressive symptoms) and couples' therapy techniques.

Enhancements implemented in BPTG@HOME

One important goal of BPTG@HOME was to make the treatment more accessible for families and to minimize dropout rates, using the recommendations discussed above. Offering the sessions at home should solve transportation problems and lacking childcare coverage. To prevent treatment dropout and to optimize adherence of homework assignments, two telephone or email contacts between sessions at home were scheduled. Every session ended with making an inventory of possible practical and emotional barriers that could interfere negatively with the implementation of the assignment, and these potential issues were resolved.

A second goal of BPTG@HOME was to optimize the content of the treatment. First, the home situation made it possible to enable parents to frequently practice new skills with their child during treatment (with video-feedback). Second, the treatment could be tailored to (if present) multiple comorbid problems of the child (additional modules Anxious or depressed child and Other problematic behavior of the child) or to parental dysfunction (additional modules Disagreement between parents regarding handling the child and Aggressive parental behaviors to the child). The treatment was also tailored to parents' individual needs by adapting the intensity of certain treatment modules to

parents' capabilities. This way, we aimed to improve the self-efficacy of parents. Third, in BPTG@HOME we combined behavioral management techniques with relationship enhancement techniques, because a meta-analysis showed that a combination of these techniques is more effective for children with disruptive disorders (including ADHD) than behavioral management techniques alone (Leijten et al., 2018). Therefore, the treatment starts with the module Following, with the primary aim to improve the quality of interaction between parent and child. In the modules Leading and Compliance, special attention is paid to the consistency of a parent, and the consistency between the parents.

Outline of this dissertation

Effectiveness of BPTG@HOME

The aim of the trial described in this dissertation was to investigate the short- and long-term effectiveness of BPTG@HOME as a second-line nonpharmacological treatment for school-aged children with ADHD who had responded insufficiently to pharmacotherapy and/or clinic-based BPT. We investigated the effectiveness on child parameters, including ADHD symptoms and comorbid problems of the child (chapter 2), and explored the effects on a range of measures of parental functioning (chapter 3). The effectiveness of a home-based BPT for school-aged children with ADHD has never been investigated as far as we know.

We included school-aged children with ADHD who had responded insufficiently to pharmacotherapy and/or clinic-based BPT, irrespective of the cause of this insufficient response. We set up our study as a pragmatic effectiveness trial, i.e., fully embedded in routine clinical practice and with broad inclusion criteria regarding insufficient improvement through routine treatment. These broad inclusion criteria, for example included allowing common comorbidities and medication use, was in line with most other studies that investigated the effectiveness of BPT for school-aged children with ADHD (see table 1).

Within this randomized controlled design, the effectiveness of BPTG@HOME was compared with a waiting list condition, as well as with a care-as-usual home-based treatment. The comparison with the waiting list arm enabled the investigation of the

effectiveness of this second-line treatment. The comparison with the care-as-usual home-based treatment arm enabled to investigate its effectiveness over and above non-specific effects of home-based treatment.

Mediating role of parenting factors

The rationale of a behavioral parent training is that by improving parenting, changes in child behaviors and functioning will be achieved, suggesting mediation effects of parenting parameters on child outcomes. However, studies investigating such mediation effects are scarce (Forehand, Lafko, Parent, & Burt, 2014; Sandler, Schoenfelder, Wolchik, & Mackinnon, 2011). To our knowledge, only a few studies included children with ADHD (e.g., Katzmann et al., 2017; Vander Stoep et al., 2017; Haack, Villodas, McBurnett, Hinshaw, & Pfiffner, 2017; Heath, Curtis, Fan, & McPherson, 2015; Chronis-Tuscano et al., 2013). Given this limited amount of research, we also investigated whether parenting variables mediated child outcomes of BPTG@HOME (chapter 4).

Parent-ratings versus blinded clinician-ratings

Recent meta-analyses recommended blinded raters (e.g., clinicians, teachers) in studies aimed at evaluating BPTs effectiveness in order to control for the potential bias in parent-reported measures (Daley et al., 2014; Sonuga-Barke et al., 2013). However, it is not certain that these blinded raters will capture the changes in children's behaviors in the home environment. Possibly, these measures would be more suitable to establish the generalization of treatment effects to other settings. Furthermore, clinicians will use mostly parent-reports to make treatment decisions. Therefore, we did not include blinded raters in our trial but only used parent-reports. To evaluate whether the use of non-blinded parent-ratings is sufficiently valid, we investigated the agreement between a parent-report questionnaire and blinded clinical judgement when assessing severity of ADHD symptoms (chapter 5).

Chapter 2

Home-based parent training for school-aged children with attention-deficit/hyperactivity disorder and behavior problems with remaining impairing disruptive behaviors after routine treatment: a randomized controlled trial

E. Nobel

P.J. Hoekstra

J.A. Brunnekreef

E.H. Messink-de Vries

B. Fischer

P.M.G. Emmelkamp

B.J. van den Hoofdakker

This chapter is published in European Child & Adolescent Psychiatry, 2019; <https://doi.org/10.1007/s00787-019-01375-9>

Abstract

Objective: To investigate the effectiveness of home-based behavioral parent training for school-aged children with attention-deficit/hyperactivity disorder (ADHD) and behavior problems with remaining impairing disruptive behaviors after routinely offered treatments in clinical practice.

Method: In a randomized controlled study including 73 referred children with ADHD and impairing disruptive symptoms after routine clinical pharmacotherapy and/or clinic-based parent training had been tried or, at least, offered, home-based behavioral parent training ($n=26$) was compared to a waiting list ($n=23$) and a care-as-usual home-based treatment ($n=24$). It was unknown to families which of the home-based treatments they received. Using mixed models for repeated measures, we examined the effectiveness on the primary outcome measure of children's severity of disruptive behaviors, and on a number of secondary outcome measures (the degree to which parents experienced the disruptive behaviors as troublesome, ADHD symptoms, oppositional-defiant disorder [ODD] symptoms, and internalizing problems).

Results: Compared to the waiting list, children receiving home-based parent training improved significantly more regarding severity of disruptive behaviors ($ES=.75$), ADHD symptoms ($ES=.89$), ODD symptoms ($ES=.65$), and internalizing problems ($ES=.60$). Compared to care-as-usual, home-based parent training was more effective in reducing disruptive behaviors ($ES=.57$), ADHD symptoms ($ES=.89$), and ODD symptoms ($ES=.88$). Significantly more reduction of children's internalizing problems was not found.

Conclusions: Children with ADHD and residual behavioral problems after routine treatment may benefit from home-based behavioral parent training.

Introduction

Children with attention-deficit/hyperactivity disorder (ADHD) frequently have co-occurring disruptive behaviors (Bendiksen et al., 2014; Jensen & Steinhausen 2015; Steinhausen et al., 2006), that typically form the most impairing symptoms for parents (Johnston & Mash, 2001) and therefore are an important focus of treatment. Guidelines for the management of ADHD indicate medication and behavioral parent training as first choice evidence-based treatments (e.g., American Academy of Pediatrics, 2011; NICE, 2008; Taylor et al., 2004). While these interventions may effectively target children's ADHD symptoms and associated disruptive behaviors (Daley et al., 2014; Reale et al., 2017; Sonuga-Barke et al., 2013), a sizable proportion of children and families do not benefit from these treatments in clinical practice, due to a variety of reasons. Besides insufficient treatment response, this may also be due to problems of parents getting a clinic-based behavioral parent training organized, which may form a barrier to enroll in parent training programs or lead to high drop-out rates (Assenany & McIntosh, 2002; Chacko et al., 2016; Koerting et al., 2013). Other factors may also play a role in less successful behavioral parent training, such as the capacity of parents to apply the learned techniques in daily life.

The question arises what treatment should be offered to children with ADHD who still have impairing disruptive symptoms after clinic-based behavioral parent training has been offered. For this purpose, we developed a home-based behavioral parent training, based on existing evidence-based behavioral parent training programs for children with disruptive behaviors (i.e., Barkley's Defiant Children (Barkley, 1987), Helping the Noncompliant Child of Forehand & McMahon (1981), and Eyberg and Funderburk's Parent-Child Intervention Therapy (2011)) for our specific target group, delivery mode, and setting. By offering treatment at home, we aimed to increase the attendance. Cunningham, Bremnes, and Boyle (2015) already reported higher attendance rates when parent training was located in neighborhood schools than in local clinics. Also, a home-based behavioral parent training program for preschool children with ADHD had higher attendance than a clinic-based group behavioral parent training (Sonuga-Barke et al., 2018).

Besides better attendance rates, training parents in the home setting may have other advantages. A meta-analysis of Kaminski, Valle, Filene, and Boyle (2008) showed that letting parents practice with their own child during treatment sessions is associated with better outcome of parent training. This in vivo practice can be done more easily and more frequently when sessions take place at home. Also, research on barriers to parent training programs has shown that parents highly appreciate flexible and individually tailored programs (see for review Koerting et al., 2013). The home setting is particularly suitable to provide such a flexible and individually tailored approach, which may also facilitate the ability of parents to apply the behavioral techniques to their child in difficult to handle daily life situations.

The home-based treatment included not only regular behavior management techniques, but combined these with relationship enhancing strategies (following principles of Parent-Child Interaction Therapy (Eyberg & Funderburk, 2011)). Recent meta-analyses showed the superior effectiveness of parenting programs for reducing disruptive child behavior that integrate relationship enhancement with behavior management, compared to behavior management alone (Leijten et al., 2018). Kaminski and colleagues also showed that teaching parents to positively interact with their child is associated with improvement in parenting skills and the child's externalizing behavior (Kaminski et al., 2008).

The overall aim of the present study was to investigate the effectiveness of home-based behavioral parent training for school-aged children with ADHD and behavior problems who have impairing disruptive symptoms after routine clinical pharmacotherapy and/or clinic-based parent training have been tried or, at least, offered. In a three-arm randomized controlled design, we compared the effectiveness of our home-based parent training with two different control conditions: a waiting list condition and a non-specific, care-as-usual home-based treatment. Comparison with the waiting list condition was used to investigate the effectiveness of our home-based behavioral parent training, with the expectation that this treatment would be more effective than the waiting list condition. We examined the effectiveness on children's severity of disruptive behaviors, ADHD symptoms, oppositional-defiant disorder (ODD) symptoms, and internalizing problems and on the degree to which parents experienced the disruptive behaviors as troublesome. As these outcome measures could suffer from

parent-expectation bias, we included a comparison of our home-based behavioral parent training with a non-manualized home-based treatment and also did not inform parents about the nature of the allocated home-based treatment. We expected to find effectiveness over and above non-specific and parent-expectation bias effects of the non-manualized home-based treatment. An additional aim was to investigate the long-term effects of the home-based parent training by comparing this treatment with the non-specific, care-as-usual home-based treatment at a six months follow-up, with the expectation to also find long-term effectiveness. We set up our study as a pragmatic effectiveness trial, i.e., fully embedded in routine clinical practice and with broad inclusion criteria, including children with and without medication, families who were not able to start or complete clinic-based parent training, as well as families who finished parent training but of whom the child did not improve sufficiently.

Methods

Participants and procedure

This was a three-arm, parallel, multicenter randomized controlled superiority trial with balanced randomization (1:1:1). All participants had been referred to three large child mental health care organizations in the North of the Netherlands (Accare, Jonx, and GGZ In de Bres), together including 16 outpatient locations. The largest group had been referred to Accare ($n=60$, 82.2%); just a small portion of the sample came from other sites (Jonx $n=4$, 5.5%; GGZ In de Bres $n=9$, 12.3%). Children had to meet the following inclusion criteria: (1) at time of referral a diagnosis of ADHD (all comorbid disorders allowed) as obtained from medical records (based on clinical interviews with the parents and teacher); (2) a Global Assessment of Functioning score of < 55 , according to the DSM-IV-TR (American Psychiatric Association, 2000); (3) current Eyberg Child Behavior Inventory (ECBI) ratings in the clinical range (i.e., Intensity scale > 131 and Problem scale > 3 ; Eyberg & Pincus, 1999); (4) a full scale, verbal and performance IQ > 70 as established within the previous two years (in 94.5% of the cases based on Wechsler Intelligence Scale for Children-III-NL); (5) had previously been offered and/or received routine treatments including ADHD medication and/or clinic-based behavioral parent training; and (6)

Figure 1. Subject Flow

Note: n = number of children; BPTG@HOME = behavioral parent training Groningen at home; CAU = care-as-usual home-based parent training; T1 = baseline assessment; T2 = assessment directly after treatment or waiting list; T3 = follow-up assessment six months after both treatments.

^a (1) parents thought treatment was no longer necessary after 1 or 2 sessions (n=4), (2) parents had other expectations of treatment (n=1), and (3) parents found treatment too intensive (n=1). ^b (1) parents did not return questionnaires and (2) too many missing data. ^c parents decline to participate in assessment.

attending primary school and aged six to thirteen at time of inclusion in the trial. Children were excluded from the study if (1) they had a medical condition that prohibited participation in the study; (2) their parents were unable to understand or follow instructions e.g., due to intellectual disability of the parents; or (3) their family had received home-based treatment in the previous year.

Participants who had given consent, after being informed by a member of the research project, were randomly assigned to the manualized home-based treatment ($n=26$), a care-as-usual home-based treatment ($n = 24$), or a waiting list of four months ($n=23$). Any ongoing treatment, including pharmacotherapy, was allowed across all study arms. An assistant who was not involved in the study conducted the randomization, using a computerized random number generator, and subsequently informed a member of the research team about the outcome of the randomization. Subsequently, families were provided with a sealed envelope containing a letter stating either the randomization outcome active home treatment (but not which treatment) or waiting list. Parents who were randomized to our home-based parent training or to the care-as-usual treatment were not explicitly informed about the nature of the allocated home-based treatment. Furthermore, no information about the differences between the treatments were publicized on a website and the therapists who performed the care-as-usual treatment were not informed about the content of the other home-based treatment. Moreover, when parents had questions about the randomization and the other treatment, all therapists were instructed not to answer the question, but to refer to the research team. The flow of subjects from initial recruitment through final analysis is presented in Figure 1. Table 1 contains child and family characteristics.

The Medical Ethical Committee of the University Medical Center in Groningen provided ethical approval for the study (METC nr 2010.289). The trial has been registered at <http://www.trialregister.nl>: Home-based behavioral treatment for ADHD; NTR3021. Because of the slow recruitment, we changed from a single-center to a multi-center study.

Measures

The primary outcome measure of our study was the ECBI, a parent-report measure of child disruptive behaviors, such as noncompliant, aggressive, hyperactive, and impulsive

Table 1. Child and family characteristics ($N = 73$)

Child characteristics			
Age child (mean in years (<i>SD</i>) range)	8.8	(1.5)	6.01-12.05
Total IQ child (mean in years (<i>SD</i>) range) ^a	94.5	(12.3)	71-119
Male child (<i>no</i> (%))	52	(71.2)	
Caucasian (<i>no</i> (%))	68	(93.2)	
ADHD diagnoses assessed by clinician at time of referral (<i>no</i> (%))			
ADHD combined type	48	(65.8)	
ADHD inattentive type	4	(5.5)	
ADHD hyperactive/impulsive type	11	(15.1)	
ADHD Not otherwise specified	10	(13.7)	
Family characteristics			
Family composition (<i>no</i> (%))			
Two biological parents	34	(46.6)	
One biological, one stepparent	11	(15.0)	
Single parent	28	(38.4)	
Education level mothers (<i>no</i> (%)) ^b			
Low	27	(37.0)	
Middle	38	(52.1)	
High	8	(11.0)	
Unknown	0	(0.0)	
Education level fathers (<i>no</i> (%)) ^b			
Low	18	(37.5)	
Middle	24	(50.0)	
High	5	(10.4)	
Unknown	1	(2.1)	
Siblings with identified psychopathology (<i>no</i> (%)) ^c	21	(28.8)	
Treatment history			
Pharmacotherapy for ADHD (<i>no</i> (%))	68	(93.2)	
Received parent training ^d (<i>no</i> (%))	61	(83.5)	
Finished parent training (<i>no</i> (%))			
Finished training	36	(49.3)	
Drop-outs	30	(41.1)	
Unknown	7	(9.6)	
Years between parent training and inclusion in trial ($n=50$); mean (<i>Sd</i>) range)	1.8	(1.6)	0.04-6.54
Other psychological treatment	38	(52.1)	
Child characteristics and received treatment during trial			
Comorbidity – externalizing problems (<i>no</i> (%)) ^e			
ODD	34	(46.6)	
CD	6	(8.2)	
Comorbidity – internalizing problems			
CBCL Internalizing T-score (mean (<i>SD</i>) range)	63.5	(8.4)	39-82
CBCL Internalizing			
Normal range (<i>No</i> (%))	19	(26.0)	
Threshold range (<i>No</i> (%))	12	(16.4)	

Clinical range (<i>no</i> (%))	40	(54.8)	
Current medication (<i>no</i> (%))	63	(86.3)	
Change in medication during trial (<i>no</i> (%))	20	(27.3)	
Other current psychosocial care at T1 (mean frequency of sessions per month (<i>SD</i>) range)	2.9	(4.7)	0-24
Care focused on parents	1.9	(4.3)	0-24
Care focused on child	.9	(1.9)	0-8

Note: *SD* = standard deviation; *no* = number of cases; ADHD = attention-deficit/hyperactivity disorder, ODD = oppositional defiant disorder; CD = conduct disorder; CBCL = Child Behavior Checklist.

^a Full scale IQ. ^b Classification according to a Dutch education classification system (Centraal Bureau voor de Statistiek, 2015) which is related to the International Standard Classification of Education (Unesco Institute for Statistics, 2014). ^c Established by asking parents if other siblings were diagnosed with psychopathology. ^d All remaining parents had only attempted to start behavioral parent training. ^e Established with Parent Interview of Child Symptoms (Schachar et al., 1995).

behaviors as well as attention problems. The Intensity scale assesses the frequency of 36 behavioral problems at home in the past week on a scale ranging from never (1) to always (7). The Problem scale measures whether the parents experienced these disruptive behaviors as troublesome in the past week (0 = no; 1 = yes). The ECBI has generally good psychometric properties (supported by more than 20 studies across cultures and countries; Funderburk, Eyberg, Rich, & Behar, 2003; Sivan, Ridge, Gross, Richardson, & Cowell, 2008). We used the ECBI as primary outcome measure, because it appears to be sensitive in measuring treatment effects (Eisenstadt, Eyberg, McNeil, Newcomb, & Funderburk, 1993; Nixon, Sweeney, Erickson, & Touyz, 2004) and describes a broad range of disruptive behaviors (not only ODD and conduct disorder symptoms, but ADHD symptoms as well) which are the main focus of the treatment.

Secondary outcome measures included: (1) severity of ADHD symptoms (18 items) and (2) severity of ODD symptoms (8 items), both rated by parents with the Swanson, Nolan, and Pelham Questionnaire (SNAP-IV) on a four-point scale, ranging from not at all to very much (Swanson, 1992). We used the SNAP-IV to measure specific ADHD symptoms and ODD symptoms separately. As the SNAP-IV is a frequently used measure in ADHD studies (e.g., McCann et al., 2014; The MTA Cooperative Group, 1999; Yang, Gao, Li, & Zhao, 2015; Wagner, Vallerand, & McLennan, 2014), comparison with other trials and samples may be possible. (3) Internalizing problems were assessed with the Internalizing subscale of the Child Behavior Checklist (CBCL; Achenbach, 1991). Prior randomized controlled trials have found significant effects of clinic-based behavioral parent training

on parent-rated internalizing problems of the child (Van den Hoofdakker et al., 2007; Xie et al., 2013).

All outcome measures were administered before randomization (T₁; baseline) and directly after the treatment or waiting list period (T₂). At T₁, we also assessed ODD and CD comorbidity with the Parent Interview for Child Symptoms (P.I.C.S.-4; Schachar, Ickowicz, & Wachsmuth, 1995). The time between T₁ and T₂ for the waiting list and our home-based treatment groups was around four months. The time between T₁ and T₂ for the care-as-usual treatment group was much longer and had greater variation, as the treatment duration was flexible. The primary outcome measure (ECBI) was also administered six months after both active treatment conditions (T₃), but not in the waiting list group. Due to ethical restrictions we could not withhold home-based treatment from families allocated to the waiting list for longer than four months (after the waiting period, families received one of the two home-based treatments). All assessments took place at home of the families. A week before the assessment date, parents received a letter with the questionnaires. In the assessment visit, we checked if the questionnaires were completed and if not, we asked parents to fill them in during the visit. The primary caretaker filled in the questionnaires, in most cases (97.3%) this was the mother.

Treatments, therapists and treatment integrity

We developed a manualized, cognitive-behavioral home-based treatment for children with ADHD and behavior problems (called “behavioral parent training Groningen at home [BPTG@HOME]”). The treatment consisted of fourteen to sixteen weekly home visits (90-120 minutes per session), spread over four months. Between every home visit, the therapist and parents had two contacts through telephone or email in which homework assignments were evaluated. In most home sessions, parents received feedback using video recordings on the skill they had been practicing that week. Furthermore, psychoeducation and cognitive interventions were given.

The treatment was divided in different treatment modules. The first module was an assessment module in which the therapist determined which treatment modules and how many sessions per module should be given. This was based on treatment goals of the family, rating scales, and the therapist’s assessment during the first session. The

subsequent three basic modules Following, Leading, and Compliance were provided to all families. These modules included three-to-six sessions, the number of sessions depending on whether the therapist considered the parents needed more practice of a certain skill. The module Following aimed to improve the parent-child relationship through daily playtime with the child. In this module, parents were taught to establish a positive interaction with their child through praise, being friendly and attentive, and avoiding instructions, questions, and criticism. Eight families (31%) received the minimum number of three sessions, for fifteen (57%), two (8%), and one (4%) families this was extended with one, two, and three sessions respectively. In the module Leading parents learned to prevent disruptive behavior of their child. Twelve families (46%) received the minimum number of three sessions, for eleven (42%) and three (12%) families this was extended with one and two sessions respectively. The last basic module, Compliance, taught parents how to cope with noncompliant behavior of their child. Eleven families (42%) received the minimum number of three sessions, for eleven (42%) and four (16%) families this was extended with one and two sessions respectively. After the basic modules, the therapist could select additional treatment modules, which focused on remaining parenting problems, such as disagreement between parents regarding how to handle their child or how to deal with their child's internalizing problems (see table 2). Twelve families (46%) received one additional treatment module and four families (15%) received two modules. After a maximum of fifteen sessions, the treatment was finished with a final session in which the therapy was evaluated and in which parents planned future goals. Within three months, two follow-up sessions at home, aimed at maintenance of learned skills, were planned. Table 2 shows the outline of the treatment and in the appendix a more detailed overview of the treatment is presented.

The basic modules were based on the principles of Barkley's Defiant Children (1987), the program of Forehand & McMahon (Helping the Noncompliant Child; 1981), and Parent-Child Interaction Therapy (Eyberg & Funderburk, 2011). The optional modules included basic behavioral interventions (such as exposure for child anxiety and behavior activation for depressive symptoms) and couples' therapy techniques. Seven families (27%) received the additional module Aggressive parental behaviors towards the child, five families (19%) received the module Other problem behaviors of the child, three families (12%) received the module Anxious or depressed child, and one family (4%)

Table 2. Outline of the content of BPTG@HOME

Treatment module	Obligatory sessions	Additional sessions
Basic modules		
Assessment 2 sessions	1. Assessment 2. Treatment plan	
Following 3-6 sessions	1. Play time 2. Parental cognitions 3. Practicing play time	4. Practicing play time extended 5. Practicing play time extended 6. Practicing play time extended
Leading 3-6 sessions	1a. Providing structure 2. Praise 3a. Communication skills	1b. Providing structure extended 3b. Communication skills extended 4. Providing structure/Praise/ Communication skills extended
Compliance 3-5 sessions	1. Assessment of noncompliant behavior 2a. House rules 3a. Reward and punishment	2b. House rules extended 3b. Reward and punishment extended
Additional modules		
Disagreement between parents regarding handling the child 0-2 sessions		1. Dealing with conflicts 2. Communication with spouse
Aggressive parental behaviors to the child 0-2 sessions		1. Dealing with parental frustration 2. Dealing with parental frustration extended
Anxious or depressed child 0-2 sessions		1. Dealing with an anxious child 2. Dealing with a child with depressive symptoms
Other problem behaviors of the child 0-x sessions		1a. Dealing with specific problem behavior 1 1b. Dealing with specific problem behavior 1 extended 2a. Dealing with specific problem behavior 2 2b. Dealing with specific problem behavior 2 extended ...
Final basic module		
Maintenance training 3 sessions	1. Evaluation 2. Follow-up 1 3. Follow-up 2	

received the module Disagreement between parents regarding handling the child.

Psychologists who provided BPTG@HOME were experienced in regular behavioral parent training (years of experience in regular parent training $M=3.5$, $SD=1.5$, range 2-6). However, they had no experience with home-based parent training. They all had received

a two-day training and weekly supervision from an experienced cognitive-behavioral therapist. During the study, the therapists and study staff met weekly to monitor treatment integrity; problems with adherence to the treatment manual were discussed and resolved. Furthermore, therapists completed a treatment integrity checklist after each session. Parts of the protocol that had not been covered were scheduled for the next session. Adherence to the protocol was high: the content covered in the treatment sessions was 94.3%. Non-adherence was mainly a result of the clinician's judgment that a small part of a treatment session required adjustment or was not applicable to the case at hand.

Families in the care-as-usual condition received an uncontrolled home-based treatment, which was eclectic and had no restrictions in duration, number of sessions, or content. In the Netherlands this care-as-usual treatment is a regular treatment for families with children with various behavior problems. However, the effectiveness of this treatment has not yet been investigated in a randomized controlled trial. The treatment started with a four-six week observation at home, aimed at building a therapeutic relationship with the family, assessing the problems of child, parents, and siblings, and determining treatment goals. This was followed by a non-manualized treatment phase, in which different techniques and interventions could be used, such as psycho-education, techniques from solution-focused and family therapy, and skills training (e.g., training of parenting skills or children's social skills). Therapists were free to choose techniques and interventions. In general, the therapist visited the family once to twice every week, for 1.5-2 hours. Continuation or discontinuation of the treatment was decided jointly by parents and therapist every three months. The care-as-usual treatment was performed by experienced social workers (years of experience with home-based treatment: $M=4.4$ $SD=2.0$, range 1.5-8.0), who had weekly peer supervision meetings.

During the four-month waiting period the families did not receive any form of home-based treatment.

Sample size

We originally aimed to include $n=40$ families in each treatment arm. With this sample size we would have been able to detect between-group differences with an effect size (Cohen's d) of .4 with 90% power. Unfortunately, the recruitment was much more

problematic than we had thought, due to lower referral to the study than expected, resulting in $N=73$ included patients after three years (March 2011 – May 2014) of recruiting. Still, with this sample size the power was above 80% as recalculated with GLIMMPSE (Kreidler et al., 2013) for most outcome measures (ECBI Intensity scale .52, ECBI Problem scale .90, secondary outcome measures .81-.99).

Statistical analysis

Handling of missing data and outliers

When participants had more than 10% missing values on a questionnaire or 20% missing values on a relatively small subscale (e.g., the SNAP ODD subscale), the total scores were not calculated but regarded as missing. In case of less missing values, the missing values were replaced by the average rating-per-item (sub)scale score, with which total scores were calculated. Using Cook's distance, we assessed potential outliers on all outcome measures.

Baseline differences between study arms

We presented the change of extra care during trial (medication and other care) for each study arm. With one-way analyses of variance (ANOVA; $p < .05$) for continuous variables and Chi-square tests ($p < .05$) for categorical variables we determined if there were significant differences between study arms with regard to basic demographic features (children's age and IQ, and educational level of the primary caregiver), baseline scores on outcome measures, characteristics of treatment (duration of treatment, number of sessions), medication change during trial, and extra care received during the trial.

Effectiveness analyses

Outcomes of the treatments were analyzed with linear mixed effects models for repeated measures (MMRM), following the intent-to-treat paradigm. We investigated the short-term effectiveness for all outcome measures by comparing the mean changes from baseline to T2 of the home-based behavioral parent training with those of the waiting list and of the care-as-usual treatment. Long-term effectiveness was explored by the mean change on the ECBI from baseline to T3 of BPTG@HOME compared with that of the care-

as-usual treatment, comparing ECBI ratings in the BPTG@HOME group with the care-as-usual home-based group at T₃. In the MMRM analyses the variables treatment, observation time, and the interaction between observation time and treatment (time*treatment) were entered as fixed effects, with the intercept specified as a random effect, using an unstructured covariance matrix for within-patient correlation ($p < .05$). The effectiveness of the treatment was assessed by interpreting the time*treatment interaction effect, reflecting the short- or long-term change in outcome measure due to a specific treatment. The random intercept in turn controls for the variety present in the baseline scores on the outcome measures, ensuring that we do not confound patient heterogeneity with treatment effects. Effect sizes were computed by dividing the mean differences between BTPG@HOME and the waiting list/care-as-usual groups at T₁ and T₂/T₃ by the pooled sample standard deviation (Taylor, 2015).

Robustness checks

We performed two sensitivity analyses. First, when significant differences were found in children's age and IQ, educational level of the primary caregiver, the duration of the treatment, the number of sessions, frequency of medication changes during the trial, and/or amount of extra care received between study arms, additional MMRM analyses were performed by re-estimating the model with these variables as additional fixed effects. According to Peduzzi and colleagues (Peduzzi, Concato, Kemper, Holford, & Feinstein, 1996), a maximum of two variables can be included given the small sample size. When more than two variables showed a significance difference between study arms, we prioritized differences in treatment characteristics (duration of treatment and number of session). When necessary, the other significant variables were included in additional analyses combined with one of the two treatment characteristics. Second, we repeated the MMRM analyses using the subsample of $N = 61$ children who previously received parent training.

Table 3. Participant and treatment characteristics for each study arm

	BPTG@HOME	Care-as-usual	Waiting list
Age of child in years (<i>M</i> (<i>SD</i>) [range])	8.2 (1.5) [6.0-11.1]	9.0 (1.3) [6.0-11.1]	9.4 (1.4) [7.0-12.1]*
Full scale IQ of child (<i>M</i> (<i>SD</i>) [range])	93.4 (11.1) [71-119]	84.8 (14.0) [73-118]	95.5 (12.0) [72-119]
Educational level mother (frequency of those with low, middle, and high level)	11, 13, 2	8, 14, 2	8, 11, 4
Duration of treatment in months (<i>M</i> (<i>SD</i>) [range])	5.3 (1.7) [1.3-8.8]	13.9 (5.4) [5.7-28.8]*	4.3 (0.4) [3.8-5.5]*
Total number of sessions (<i>M</i> (<i>SD</i>) [range])	13.1 (5.1) [1-17]	29.8 (18.0) [1-87]*	-
Changes in medication use during treatment (frequency of lowered and increased dosage, and change of type of medication)	5, 2, 4	2, 2, 1	1, 3, 0
Other psychosocial care at pre-treatment (mean frequency of sessions per month (<i>SD</i>) range)	3.6 (4.7) [0-16.5]	2.5 (4.4) [0-16]	2.4 (5.1) [0-24]
*Care focused on parents	2.4 (3.9) [0-13]	1.8 (4.0) [0-16]	1.6 (5.1) [0-24]
*Care focused on child	1.2 (2.1) [0-8]	0.8 (2.0) [0-8]	0.8 (1.4) [0-4]
Other psychosocial care at post-treatment (mean frequency of sessions per month (<i>SD</i>) range)	3.7 (4.5) [0-20]	4.8 (7.10) [0-23]	-
*Care focused on parents	2.7 (4.1) [0-17]	4.2 (7.1) [0-23]	-
*Care focused on child	0.9 (1.9) [0-8]	0.6 (1.5) [0-5]	-
Other psychosocial care at follow-up (mean frequency of sessions per month (<i>SD</i>) range)	3.8 (3.9) [0-16]	3.0 (2.9) [0-8]	-
*Care focused on parents	2.2 (3.7) [0-16]	1.2 (1.4) [0-4]	-
*Care focused on child	1.6 (2.1) [0-6]	1.8 (3.0) [0-8]	-

Note: *M*= mean; *SD* = standard deviation; *no*= number of cases.

* Significant difference ($p < 0.05$) with the BPTG@HOME group according to ANOVA. All other comparisons (according to ANOVA or chi-square test) were not significant.

Results

Outliers

No evidence of outliers was found, as Cook's distances varied between .00 and .09 (ECBI intensity scale), .00 and .08 (ECBI problem scale), .00 and .06 (SNAP ADHD scale), .00

and .10 (SNAP ODD scale), and 0.00 and 0.15 (CBCL internalizing scale). This is well below the threshold of 1 (Cook & Weisberg, 1982).

Baseline differences between study arms

Table 3 shows basic demographic features (child's age, total IQ, and educational level of the primary caregiver), treatment characteristics (duration of treatment and number of sessions), and change of extra care during trial (medication and other care) of the three study arms. There were no significant differences between BPTG@HOME and the other study arms on the baseline ratings of the outcome measures. Also, there were no differences in medication use during treatment between the three study arms and in extra care received at T1, T2, and T3. However, the care-as-usual treatment lasted significantly longer than BPTG@HOME and consisted of significantly more sessions, and the BPTG@HOME condition had younger children (see Table 3).

Effectiveness analyses

Table 4 shows the results from the MMRM analyses of short- and long-term effectiveness of BPTG@HOME compared to the waiting list condition and the care-as-usual treatment. On the short run (T2), compared to the waiting list condition, BPTG@HOME was associated with larger reductions of the ECBI Intensity scale, the SNAP ADHD scale, the SNAP ODD scale, and the CBCL Internalizing scale. Except for the CBCL Internalizing scale, similar larger reductions on these scales were observed of the BPTG@HOME group compared to the care-as-usual treatment. Of further note, the T2 mean scores on the ECBI Intensity scale (133.3) in the BPTG@HOME group but not those in the other arms (143.0 and 141.8 in the care-as-usual treatment and waiting list, respectively), neared the clinical cut-off score (<131), indicating that the disruptive behaviors post-BPTG@HOME treatment were almost below the clinical range. The T2 mean score of the BPTG@HOME group and the waiting list group on the ECBI Problem scale, but not those of the care-as-usual treatment arm reached the clinical cut-off score (<15). On the long run (T3) and compared to the care-as-usual treatment, in the BPTG@HOME group we observed a significantly larger reduction in the ECBI Problem scale. Furthermore, the T3 mean scores of the BPTG@HOME treatment group on the ECBI Intensity and the ECBI Problem scale both dropped below the clinical cut-off score.

Table 4. Outcome of BPTG@HOME versus waiting list and care-as-usual treatment, analyzed with linear mixed model for repeated measures.

	BPTG@HOME	Care-as-usual	Waiting list	BPTG@HOME vs. waiting list				BPTG@HOME vs. Care-as-usual			
				β	p	95% CI	ES	β	p	95% CI	ES
Baseline (<i>M</i> (<i>SD</i>) [range])											
ECBI Intensity scale	156.1 (26.6) [103-209]	156.0 (22.1) [117-204]	149.7 (18.5) [110-182]								
ECBI Problem scale	19.9 (7.4) [3-30]	21.3 (5.7) [9.3-32]	18.0 (6.6) [5-34]								
SNAP ADHD scale	34.3 (9.6) [16-46]	31.4 (8.9) [16-51]	30.6 (9.6) [5-51]								
SNAP ODD scale	13.2 (5.2) [5-22]	11.0 (4.6) [3-22]	11.9 (5.2) [0-22]								
CBCL Internalizing scale	14.6 (6.6) [6-36]	15.2 (8.6) [1-39]	11.5 (7.4) [0-27]								
After treatment (<i>M</i> (<i>SD</i>) [range])											
ECBI Intensity scale	133.3 (27.1) [78-188]	143.0 (28.0) [99-192]	141.8 (22.1) [92-178]	17.8	.048*	5.0-30.5	.75	13.7	.009**	0.1-27.2	.57
ECBI Problem scale	14.2 (9.0) [0-26]	17.4 (8.4) [5-31]	14.6 (7.7) [3-34]	2.8	.167	-1.1-6.7	-	2.2	.300	-1.9-6.4	-
SNAP ADHD scale	24.5 (11.0) [6-50]	31.6 (11.6) [11-50]	18.5 (9.1) [7-50]	8.7	.001**	3.8-13.6	.89	9.3	.001**	4.1-15.5	.89
SNAP ODD scale	7.5 (4.7) [0-17]	10.1 (5.2) [2-19]	9.1 (4.5) [0-16]	3.2	.028*	.4-6.1	.65	4.4	.005**	1.4-7.4	.88
CBCL Internalizing scale	8.7 (5.0) [1-18]	12.1 (8.3) [2-29]	9.8 (8.0) [1-36]	4.4	.011*	1.1-7.6	.60	2.6	.139	-0.9-6.1	-
6 months follow-up (<i>M</i> (<i>SD</i>) [range])											
ECBI Intensity scale	125.1 (21.4) [87-160]	133.0 (22.62) [94-170]	-	-	-	-	-	9.9	.130	-2.9-22.9	-
ECBI Problem scale	8.7 (7.4) [0-23]	13.8 (6.3) [5-23]	-	-	-	-	-	4.6	.039*	0.4-8.8	.65

Note: *M* = mean; *SD* = standard deviation; 95% CI = 95% confidence interval; ES = effect size; ECBI = total score on the subscales of the Eyberg Child Behavior Checklist; SNAP = total score on the subscales of the Swanson, Nolan and Pelham-IV; CBCL = total score on the subscale of Child Behavior Checklist.

* $p < .05$; ** $p < .01$.

Robustness checks

The care-as-usual treatment lasted significantly longer than BPTG@HOME and consisted of significantly more sessions (see Table 3). Therefore, we repeated the MMRM analyses, now controlling for treatment duration and the number of sessions. At T2 we again found significantly larger reductions in the BPTG@HOME group on the ECBI Intensity scale ($\beta = 14.8$; $p = .047$; 95% CI = .2-29.4), on the SNAP ADHD scale ($\beta = 9.3$; $p = .002$; 95% CI = 3.9-14.6), and on the SNAP ODD scale ($\beta = 4.6$; $p = .008$; 95% CI = 1.3-7.8). The significant decrease at T3 in the ECBI Problem scale also persisted ($\beta = 4.8$; $p = .024$; 95% CI = .6-9.0). As an additional sensitivity analysis, we replaced either the covariate 'duration of treatment' or 'number of sessions' with 'age'. All the results were unchanged under these new specifications.

The analysis with the subsample of $N = 61$ children who had previously received clinic-based parent training resulted in similar findings, except for the comparison between the BPTG@HOME and the care-as-usual treatment: the change on the SNAP ODD scale was no longer significant ($\beta = 3.7$, $p = .061$).

Discussion

This study investigated the effectiveness of home-based behavioral parent training, as follow-up treatment for school-aged children with ADHD and behavior problems who had impairing disruptive symptoms after routine clinical pharmacotherapy and/or clinic-based behavioral parent training had been tried or offered. Our results suggest that home-based behavioral parent training is effective for these difficult-to-treat families: compared to a waiting list, the treatment significantly reduced children's disruptive behaviors, ADHD symptoms, ODD symptoms, and internalizing problems, with moderate (disruptive behaviors, ODD symptoms, and internalizing problems) to large (ADHD symptoms) effect sizes. The severity of disruptive behaviors of the children in the home-based behavioral parent training group almost dropped below the clinical range directly after treatment. Prior RCTs that investigated the effectiveness of behavioral parent training for school-aged children with ADHD found positive results regarding the reduction of ADHD symptoms, disruptive behaviors, and internalizing problems as well

(e.g., Anastopoulos, Shelton, DuPaul, & Guevremont, 1993; Chacko et al., 2009; Chronis-Tuscano et al., 2013; Fabiano et al., 2012; Katzmann et al., 2017; Rajwan, Chacko, Wymbs, & Wymbs, 2014; Van Den Hoofdakker et al., 2007; Xie et al., 2013). However, we are the first who showed this effect for a difficult-to-treat group by offering it as a follow-up intervention after routine treatments have been tried or, at least, offered. Noteworthy, we found higher effect sizes on ADHD symptoms (ES .89) and disruptive behavioral problems (ES .65-.88) compared to prior meta-analyses (ADHD symptoms ES .35-.68; disruptive behaviors ES .26-.59; Coates, Taylor, & Sayal 2015; Daley et al., 2014; Sonuga-Barke et al., 2013).

The home-based behavioral parent training was also more effective than our care-as-usual home-based treatment in reducing disruptive behaviors (moderate effect size), ADHD symptoms, and ODD symptoms (large effect sizes). As the evaluation of both active treatments may have been affected by parent-expectation bias (which we tried to minimize by not informing families about the nature of the allocated treatment), this finding suggests that the effectiveness of home-based behavioral treatment was not solely a result of parent-expectation bias. Between the home-based parent training group and the care-as-usual group no differences in reductions of internalizing problems and improvements in experiencing the disruptive behaviors as troublesome by the parents were present. However, the care-as-usual treatment lasted on average twice as long as the home-based parent training (13.9 versus 5.3 months respectively) and involved twice as many home visits (29.8 versus 13.1 visits).

While our study did not investigate which elements of the home-based parent training contributed specifically to its effectiveness, it is unlikely that only the treatment setting (at home) was crucial for the effectiveness of our program, given that the care-as-usual treatment was home-based as well, but less effective than the home-based parent training. It could be hypothesized that the use of live modeling and video feedback during the home sessions were important components as well (Webster-Stratton, 1990, 1996). Also, the opportunity for parents to directly practice their parenting skills with their child during the session may have been a crucial component (Kaminski et al., 2008). However, also other treatment elements may have played a role in the effectiveness of the home-based treatment, such as the length and number of sessions, and the fact that the program was individually tailored for each family. Furthermore, it cannot be ruled out

that the differences in education level, working experience, and degree of supervision between the therapists of the two treatments have partially driven our results. Possibly, the treatment of this difficult-to-treat-subgroup of children with disruptive behaviors requires higher educated staff with expert supervision.

We also found preliminary evidence for the effectiveness of home-based parent training in the long run. Six months after the end of treatment, children allocated to the home-based parent training group remained on average below the clinical range regarding the severity of disruptive behaviors. Also, parents in this group experienced the disruptive behaviors as significantly less troublesome compared to the care-as-usual group. The latter result was not found directly after treatment. Possibly, first the behavior of the child needed to improve, before the parents could adjust their cognitions about the behavior of their child in a more positive way.

High drop-out rates are common in behavioral parent training and limit their effectiveness (Boggs et al., 2004; Chacko et al., 2016; Patterson & Chamberlain, 1994). With respect to our home-based behavioral parent training program 26.4% of the eligible participants did not start the treatment or dropped out prematurely. This percentage is markedly lower than the 51% reported in a recent systematic review on parental engagement in behavioral parent training (Chacko et al., 2016). It is plausible that this lower drop-out can be attributed to offering the treatment at home; another home-based behavioral parent training for preschool children with ADHD also showed a relatively low drop out (Sonuga-Barke et al., 2018).

Limitations

This study should be interpreted in light of certain limitations. First, our small sample size, which was lower than planned, should be acknowledged. Having three different treatment arms may have been less suitable given our modest sample size. Another consequence of the small sample size was that we did not take into account changes in medication during treatment, nor the reasons why the families did not profit from routinely offered treatments. However, even with a smaller sample size we still had sufficient power and found significant results in favor of the home-based parent training. Second, we were unable to collect post-treatment assessments in one third of the participants who had received the care-as-usual treatment. It is conceivable that those

missings were from families who may have responded less well to the treatment. Thus, we may have somewhat underestimated the effectiveness of home-based parent training compared to care-as-usual. Third, the care-as-usual home-based treatment had a much longer duration than our home-based behavioral parent training. However, controlling for treatment duration and the number of sessions did not alter our findings. Finally, we fully relied on parent-self reports as outcome measure. Recent meta-analyses recommended the use of raters who are blinded to treatment allocation (e.g., clinicians, teachers) in order to control for the potential bias of parent-reported measures (Daley et al., 2014; Sonuga-Barke et al., 2013). While blinded raters may be desirable when the objective of the study is generalizability across situations, such a measure may be too strict when the goal of the treatment is to improve behavioral problems in a specific context (e.g., at home) that falls outside the scope of the blinded raters. The current study took into account possible parent-expectation bias by including an active control group in the study design and not revealing to participating families the type of the assigned home-based treatment. This approach enabled the usage of parent-reported measures in a more reliable way. However, we acknowledge that we did not check whether we were successful in keeping parents unaware of the differences between the two home-based treatments.

Future research

Apart from replicating our findings in a larger sample, future studies should try to identify essential program components and moderators of treatment effectiveness. In particular, assessing the added value of the different treatment modules and the extended sessions would be important. Furthermore, it would be worthwhile to investigate whether the home-based treatment is also effective when delivered by social workers rather than psychologists. Delivering the program by social workers would obviously make the treatment less expensive and easier to implement. Finally, we investigated home-based parent training as a follow-up treatment in a stepped-care ADHD treatment program. Unfortunately, we lack baseline scores at the time of initial treatment. We only systematically collected ratings after regular treatment had been offered. Future stepped-care studies may want to also collect response to initial treatment and/or determine its effectiveness as first- or second-line treatment.

Appendix Behavioral Parent Training Groningen for children with behavioral problems at home (BPTG@HOME)

A brief overview of the manual (Nobel & Van der Veen – Mulders, 2012).

General description	
Treatment goals	<ul style="list-style-type: none">- To decrease children's externalizing behaviors;- To decrease children's internalizing problems;- To increase parental knowledge of ADHD and comorbid problems;- To enhance parenting skills and competence;- To enhance parental problem-solving skills;- To change parental dysfunctional beliefs about their child;- To enhance positive communication between family members.
Treatment characteristics	<ul style="list-style-type: none">- Home-based and consisting of fourteen to sixteen weekly home visits (90-120 minutes per session) and two telephone or email contacts after every home visit (first contact within 24 hours after each home visit);- Manualized and including standard basic modules (consisting of two to six sessions), as well as additional modules (consisting of a maximum of two sessions) which can be used individualized: the therapist determines which additional treatment modules and how many sessions per module will be given;- The basic modules are based on the parent training treatment manuals of Barkley (1987), Forehand & McMahon (1981), and Eyberg and Funderburk (2011); the additional modules consist of basic cognitive behavioral therapy methods as well as family and marital therapy techniques;- Including the following main elements: psycho-education, stimulus control and contingency management techniques, relationship enhancing strategies, therapist guided video-feedback, homework assignments. The therapist needs to have a directive therapeutic approach.

Treatment structure and content		
Basic modules	Sessions	Content
Assessment 2 sessions	1. Assessment	1. Establishing treatment goals and assessing family functioning based on rating scales and observation/video of play time Between sessions: determine which treatment modules and how many sessions per module shall be given
	2. Treatment plan	2. Discussing treatment plan with parents
Following 3-6 sessions	1. Play time	1. Education on relationship enhancing techniques; use of therapist modeling during play time; parents practice with techniques during play time, using video feedback; homework: daily practice of learned techniques during play time
	2. Parental cognitions	2. Identify and change dysfunctional beliefs of parents about relationship enhancing techniques; parents practice techniques during play time with video feedback; homework: daily practice of learned techniques during play time
	3. Practicing play time	3. Parents practice techniques during play time, using video feedback; homework: daily practice of learned techniques during play time
	4-6. Practicing play time extended ¹	4-6. Parents practice techniques during play time, using video feedback; homework: daily practice of learned techniques during play time
Leading 3-6 sessions	1a. Providing structure	1a. Education on the need for a structured environment for children with ADHD and disruptive behaviors; structuring the environment in space and/or time

	1b. Providing structure extended ¹	1b. Structuring the environment in space and/or time
	2. Praise	2. Education on increasing the child's self-esteem through praise; practicing reinforcing positive behaviors through praise, using video feedback
	3a. Communication skills	3a. Education on the need for clear communication/commands for children with ADHD and disruptive behaviors; practicing commands/preparing child for specific troublesome situations, using video feedback
	3b. Communications skills extended ¹	3b. Practicing commands/preparing child for specific troublesome situations, using video feedback
	4. Providing structure/ Praise / Communication skills extended ¹	4. Structuring the environment in space and/or time / practicing praise with video feedback / practicing commands/preparing child for specific context, using video feedback
Compliance 3-5 sessions	1. Assessment of noncompliant behavior	1. Education on the relationship between ADHD and oppositional behaviors; filming oppositional behavior of the child at a challenging moment of the day (for parents) Between sessions: therapist conducts functional analysis (Antecedents-Behavior-Consequences) of child's and parents filmed behaviors
	2a. House rules	2a. Video feedback on previous session; education on the need for clear house rules for children with ADHD and disruptive behaviors; practicing installing (new) house rules, using video feedback

	2b. House rules extended ¹	2b. Practicing installing (new) house rules, using video feedback
	3a. Reward and punishment	3a. Education on reinforcement techniques and punishment; designing token system; implementing token system, using video feedback
	3b. Reward and punishment extended ¹	3b. Designing token system; implementing token system, using video feedback
Additional modules	Sessions	Content
Disagreement between parents regarding handling the child	1. Dealing with conflicts	1. Education on effects of parental conflicts on the child; assessment of parental conflicts; practicing to prevent parental conflicts
0-2 sessions	2. Communication with spouse	2. Filming parental discussion – psycho-education: communication rules – Practicing communication rules with video feedback
Aggressive parental behaviors towards the child	1. Dealing with parental frustration	1. Education on effects of aggressive parental behaviors on the child; assessing aggressive behaviors of the parent(s); practicing prevention and dealing with frustrations
0-2 sessions	2. Dealing with parental frustration extended ¹	2. Practicing prevention of parental aggression and dealing with parental frustrations
Anxious or depressed child	1. Dealing with an anxious child	1. Education on the relationship between ADHD and anxiety; assessing anxiety of the child; practicing parental skills for helping the child with anxiety
0-2 sessions		

	2. Dealing with a child with depressive symptoms	2. Psycho-education relationship ADHD and depressive symptoms; assessing depressive symptoms of the child; practicing parental skills for helping the child with depressive symptoms
Other problem behaviors of the child 0-x sessions	1a. Dealing with specific problem behavior 1	1a. Filming and assessing specific problem behavior of the child; designing intervention plan based on functional analyses of behavior; implementing intervention plan
	1b. Dealing with specific problem behavior 1 extended ¹	1b. Evaluating intervention plan of specific problem behavior; adjusting intervention plan as appropriate
	2a. Dealing with specific problem behavior 2	2a. Filming and assessing second specific problem behavior of the child; designing intervention plan based on functional analyses of behavior; implementing plan of action
	2b. Dealing with specific problem behavior 2 extended ¹	2b. Evaluating plan of action of specific problematic behavior; adjusting intervention plan as appropriate
		
Final basic module	Sessions	Content
Maintenance training 3 sessions	1. Evaluation	1. Evaluation of the treatment with the parents; evaluation of the treatment with the family; anticipating future problems
	2. Follow-up 1	2. Rehearsal and maintenance training; anticipating future problems
	3. Follow-up 2	3. Rehearsal and maintenance training; anticipating future problems

¹ Extended means repeating the particular home visit with same content.

General structure home visits

Presentation of the agenda of current home visit

Discussion of homework assignments

Introduction of new topic through education

Practicing new skills (using therapist video feedback)

Identify and change possible dysfunctional beliefs of the parents

Preparation of new homework assignment

Chapter 3

Effects on parenting parameters of a home-based parent training for children with attention-deficit/hyperactivity disorder with remaining disruptive behaviors after routine treatment: a randomized controlled trial

E. Nobel

B.J. van den Hoofdakker

P.J. Hoekstra

Abstract

Introduction: We recently showed that manualized home-based behavioral parent training was effective in reducing disruptive behavioral problems in school-aged children with attention-deficit/hyperactivity disorder (ADHD) who had responded insufficiently to routine clinical care. This study investigated the effects of this training on parenting outcomes.

Methods: In a randomized controlled study ($N=73$), manualized home-based behavioral parent training ($n=26$) was compared to a waiting list condition ($n=23$), and, to rule out possible non-specific treatment effects, also to a care-as-usual home-based treatment ($n=24$). It was not revealed to families which of the two home-based treatments they received. Our outcome measures included parenting sense of competence, parenting stress, and parenting skills and were analyzed with a mixed model for repeated measures.

Results: Compared to the waiting list, parents who received manualized home-based behavioral parent training improved significantly more in their sense of competence, regarding how capable they felt in their parent role (parenting efficacy). Parents also rated less parenting stress regarding their interactions with their child and regarding experiencing their child as less difficult to handle, and reported (concerning parenting skills) to use less harsh and inconsistent parenting. Compared to the care-as-usual treatment, harsh and inconsistent parenting reduced significantly more than in the home-based behavioral parent training group. When taking into account the difference in duration and intensity of the treatments, home-based behavioral parent training also had significantly better results in reducing overall parenting stress and improving parenting skills regarding positive verbal discipline, the use of praise and incentives, and monitoring of the child. Long-term effects on parenting sense of competence were not found.

Conclusion: Manualized home-based behavioral parent training is a promising second-line treatment for school-aged children with ADHD and remaining behavioral problems in reducing parenting stress, improving parenting efficacy, and specifically (compared to a non-specific home-based treatment) in reducing harsh and inconsistent parenting.

Introduction

The aim of behavioral parent training for children with attention-deficit/hyperactivity disorder (ADHD) and disruptive symptoms is to improve children's behavior and functioning by improving parenting skills. In school-aged children with ADHD, different randomized controlled trials confirmed the effectiveness of behavioral parent training on parenting parameters. Improvements included more frequent positive parenting, such as using praise, showing positive affect, and using positive physical communication. Also decreases in negative parenting, such as the use of negative commands and critical statements, and negative physical parenting behaviors, as shown by observations or parent-reported questionnaires, have been reported (Fabiano et al., 2012; Chacko et al., 2009; Xie et al., 2013; Hoath & Sanders, 2002; Chronis-Tuscano et al., 2013; Dose et al., 2017). Furthermore, positive effects of behavioral parent training in school-aged children with ADHD on parent-rated parenting sense of competence were found (Hoath & Sanders, 2002). Reductions of parent-rated parenting stress have been demonstrated (Chacko et al., 2009), albeit not all studies found this (e.g., Van den Hoofdakker et al., 2007).

Nevertheless, for a sizable proportion of children with ADHD and their parents, clinic-based parent training is not sufficient. This can be due to difficulties that parents may have to apply the learned techniques in daily life and/or low attendance to the training (Chacko et al., 2016; Koerting et al., 2013; Assenany & McIntosh, 2002). We therefore developed a manualized home-based behavioral parent training for school-aged children with ADHD who still have impairing disruptive symptoms after clinic-based parent training was tried or, at least, offered. A key feature of the new program was that the parent training was offered at home which optimizes attendance (Cunningham, Bremner, & Boyle, 1995; Sonuga-Barke et al., 2018), and enables a flexible and individually tailored program which was frequently recommended by parents and professionals (see for review Koerting et al., 2013). Further, the home setting provides parents multiple opportunities to practice with their child during treatment sessions, which may enhance treatment effectiveness (Kaminski, Valle, Filene, & Boyle, 2008). The new program consisted of a combination of regular behavior management techniques and relationship enhancing strategies, which was demonstrated to be more effective than behavioral

management techniques alone (Leijten et al., 2018). In a previous article, we already showed effects of this manualized home-based behavioral parent training on ADHD symptoms and disruptive behaviors of school-aged children with ADHD who had impairing disruptive symptoms after routine clinical pharmacotherapy and/or clinic-based parent training (Nobel et al., 2019; chapter 2).

The current study investigated the short- and long-term effects of our manualized home-based behavioral parent training program on several secondary parenting outcomes, covering parenting sense of competence, stress, and skills, using parent-report questionnaires. We fully embedded our pragmatic effectiveness trial in routine clinical practice and used broad inclusion criteria, including children with and without medication, families who were not able to start or complete clinic-based parent training, as well as families who finished parent training but of whom the child did not improve sufficiently. To investigate the effects of the home-based parent training, we compared the treatment to a waiting list and, to rule out non-specific treatment effects of the home-based behavioral parent training, also to a non-specific home-based treatment (care-as-usual). We expected to find effectiveness of the manualized home-based behavioral training on the parenting outcomes compared to the waiting list and the care-as-usual treatment.

Methods

Participants and procedures

Participants could be included if they had a diagnosis of ADHD at time of referral, as obtained from medical records (all comorbid disorders allowed), and if they previously had been offered and/or received routine treatments including ADHD medication and/or clinic-based behavioral parent training. Their current Global Assessment of Functioning score had to be < 55 , according to the DSM-IV-TR (American Psychiatric Association, 2000), and current Eyberg Child Behavior Inventory (ECBI) ratings had to be in the clinical range (i.e., Intensity scale > 131 and Problem scale > 3 ; Eyberg & Pincus, 1999). Furthermore, the children had to attend primary school and to be aged six to thirteen at time of inclusion, and to have a full scale, verbal and performance IQ above 70, as

established within the previous two years (in 94.5% based on the Wechsler Intelligence Scale for Children-III-NL [Wechsler, 2005]). Participants were excluded from the study if the family had received home-based treatment in the previous year, when the child had a medical condition that prohibited participation in the study, or when parents were unable to understand or follow instructions. Table 1 contains sample characteristics and information about previous and ongoing treatments.

This was a balanced (1:1:1), multi-arm parallel, superiority, randomized controlled trial. We started as a single-center study, but due to slow recruitment changed to a multicenter study, in which participants ($N = 73$) were recruited from three child mental health care organizations (Accare $n=60$, 82.2%; Jonx $n=4$, 5.5%; GGZ In de Bres $n=9$, 12.3%). Participants who had given informed consent were randomly assigned to either the home-based behavioral parent training ($n = 26$), a care-as-usual home-based treatment ($n = 24$), or a waiting list of four months ($n = 23$). An assistant conducted the simple balanced randomization with a computerized random number generator. After the pre-treatment assessment, parents received a sealed envelope containing a letter stating the outcome of randomization (i.e., starting treatment or waiting list). Parents who were randomized to one of the treatment conditions, were not told which of the two active treatments they received. Moreover, parents were not informed about the differences between the two treatments. Furthermore, no information about the differences between the treatments was published on a website, and the therapists who performed the care-as-usual treatment were not informed about the content of the other home-based treatment to prevent that this information about the differences between the treatments could leak to participating families.

The Medical Ethical Committee of the University Medical Center in Groningen had assigned ethical approval for the study (METC nr 2010.289). The trial has been registered at <http://www.trialregister.nl>: Home-based behavioral treatment for ADHD; NTR3021.

Treatments

We developed a manualized, cognitive-behavioral, home-based parent training program for children with ADHD (called "Behavioral Parent Training Groningen at Home [BPTG@HOME]"), based on the principles of Barkley's Defiant Children (1987), the program of Forehand & McMahon (Helping the Noncompliant Child; 1981), and Parent-

Table 1. Child and family characteristics ($N = 73$)

Child characteristics			
Age child (mean in years (<i>SD</i>) range)	8.8	(1.5)	6.01-12.05
Total IQ child (mean in years (<i>SD</i>) range) ^a	94.5	(12.3)	71-119
Male child (<i>No (%)</i>)	52	(71.2)	
Caucasian (<i>No (%)</i>)	68	(93.2)	
ADHD diagnoses assessed by clinician at time of referral (<i>No (%)</i>)			
ADHD combined type	48	(65.8)	
ADHD inattentive type	4	(5.5)	
ADHD hyperactive/impulsive type	11	(15.1)	
ADHD Not otherwise specified	10	(13.7)	
Family characteristics			
Family composition (<i>No (%)</i>)			
Two biological parents	34	(46.6)	
One biological, one stepparent	11	(15.0)	
Single parent	28	(38.4)	
Education level mothers (<i>No (%)</i>) ^b			
Low	27	(37.0)	
Middle	38	(52.1)	
High	8	(11.0)	
Unknown	0	(0.0)	
Education level fathers (<i>No (%)</i>) ^b			
Low	18	(37.5)	
Middle	24	(50.0)	
High	5	(10.4)	
Unknown	1	(2.1)	
Siblings with identified psychopathology (<i>No (%)</i>) ^c	21	(28.8)	
Treatment history			
Pharmacotherapy for ADHD (<i>No (%)</i>)	68	(93.2)	
Received parent training ^d (<i>No (%)</i>)	61	(83.5)	
Finished parent training (<i>No (%)</i>)			
Finished training	36	(49.3)	
Dropped out of parent training	30	(41.1)	
Unknown	7	(9.6)	
Years between parent training and inclusion in trial ($n=50$); mean (<i>Sd</i>) range)	1.8	(1.6)	0.04-6.54
Other psychological treatment for the child	38	(52.1)	
Child characteristics and received treatment during trial			
Comorbidity – externalizing problems (<i>No (%)</i>) ^e			
ODD	34	(46.6)	
CD	6	(8.2)	
Comorbidity – internalizing problems			
CBCL Internalizing T-score (mean (<i>SD</i>) range)	63.5	(8.4)	39-82
CBCL Internalizing			
Normal range (<i>No (%)</i>)	19	(26.0)	
Threshold range (<i>No (%)</i>)	12	(16.4)	

Clinical range (<i>No (%)</i>)	40	(54.8)	
Current medication (<i>No (%)</i>)	63	(86.3)	
Change in medication during trial (<i>No (%)</i>)	20	(27.3)	
Other current psychosocial care at T1 (mean frequency of sessions per month (<i>SD</i>) range)	2.9	(4.7)	0-24
Care focused on parents	1.9	(4.3)	0-24
Care focused on child	.9	(1.9)	0-8

Note: *SD* = standard deviation; *no* = number of cases; ADHD = attention-deficit/hyperactivity disorder, ODD = oppositional defiant disorder; CD = conduct disorder; CBCL = Child Behavior Checklist.

^a Eull scale IQ. ^b Classification according to a Dutch education classification system (Centraal Bureau voor de Statistiek, 2015) which is related to the International Standard Classification of Education (Unesco Institute for Statistics, 2014). ^c Established by asking parents if other siblings were diagnosed with psychopathology. ^d All remaining parents had only attempted to start behavioral parent training. ^e Established with Parent Interview of Child Symptoms (Schachar, Ickowicz, & Wachsmuth, 1995).

Child Interaction Therapy (Eyberg & Funderburk, 2011). Treatment goals were decreasing the child's externalizing and internalizing problems and parental dysfunctional beliefs about their child, increasing parental knowledge of ADHD and comorbid problems, and enhancing parenting skills, sense of competence, parental problem-solving skills, and positive communication between family members. The treatment consisted of fourteen to sixteen weekly home visits (90-120 minutes per session), spread over four months and was divided in different treatment modules. The first module was an assessment module in which the therapist determined which treatment modules, and how many sessions per module, should be given. The subsequent three basic modules Following, Leading, and Compliance were provided to all families, focusing on improving the parent-child relationship, learning to prevent disruptive behavior of the child, and teaching how to cope with noncompliant behavior. Examples of skills that parents learned during these modules were communicating positively with their child, giving clear instructions, providing more structure at home, installing clear house rules, reinforcing positive behaviors through praise and rewards, and using effective punishment techniques. After the basic modules, the therapists could select additional treatment modules, which focused on specific parental problems and/or comorbid problems of the child. In most sessions, parents received video feedback on the skill they had been practicing that week. Furthermore, psychoeducation and cognitive interventions to alter dysfunctional parental attributions were provided (see for a more detailed description of the treatment Nobel et al., 2019; chapter 2).

Psychologists who were experienced in clinic-based behavioral parent training (years of experience $M=3.5$, $Sd=1.5$, range 2-6) delivered the manualized home-based treatment. They all had received training in the treatment and received weekly expert-supervision from an experienced cognitive-behavioral therapist. Therapists completed a treatment integrity checklist after each session. Parts of the protocol that had not been covered were scheduled for the next session. Adherence to the protocol was high: the mean coverage percentage within treatment sessions was 94.3%. Non-adherence was mainly a result of the clinician's judgment that a small part of a treatment session required adjustment or was not applicable to the family at hand.

The care-as-usual treatment was also home-based, but uncontrolled, eclectic and without restrictions in duration, number of sessions, or content. The main goals of the treatment were to (1) reduce behavioral problems of the child, (2) reduce parenting stress, (3) improve parenting skills, and (4) build a social network. Therapists were experienced social workers (years of experience $M=4.4$, $Sd=2.0$, range 1.5-8.0), who received weekly peer-supervision. The treatment started with a four-to-six-week period in which observations at home were carried out. This period was also used to build a therapeutic relationship and to determine treatment goals. This was followed by a treatment phase, during which different techniques could be used, such as psychoeducation, techniques from solution-focused therapy, behavioral management techniques, and techniques from family therapy. Normally, the therapist visited the family once to twice a week for 1.5-2 hours. Continuation or discontinuation of the treatment was decided jointly by parents and therapist every three months. In the Netherlands, this care-as-usual treatment is commonly used for families with children with behavior problems. However, its effectiveness has not yet been investigated.

During the four-month waiting period, the families did not receive any form of home-based treatment. Other on-going treatment such as medication or social skills training for the child were allowed in all three conditions.

Measures

We measured parenting sense of competence with the subscales of the Parenting Sense of Competence Scale (PSOC; Gibaud-Wallston & Wandersmann, 1978; Johnston & Mash, 1989). The PSOC is a 17-item questionnaire on which parents rate their agreement

with statements (e.g., “Being a parent makes me tense and anxious” or “Being a parent is manageable, and any problems are easily solved”) on a six-point scale (1 = strongly disagree to 6 = strongly agree). On the subscale Efficacy parents evaluate their own capability levels and problem-solving abilities regarding their parental role. The PSOC subscale Satisfaction measures parental anxiety, motivation, and frustration. For mothers, good internal consistency has been found for both subscales (Johnston & Mash, 1989).

We used the total and subscale scores of the Parenting Stress Index-Short Form (PSI-SF; Abidin, 1995) to measure parenting stress. The PSI-SF is a 36-item self-report measure, on which parents indicate their agreement with statements (e.g., “Expected to have closer feelings for my child” or “Child does some things that bother me”) on a five-point scale. The total score expresses the overall level of stress that parents experience in their role as a parent. The PSI-SF subscale Parental distress rates whether parents feel competent, conflicted, and/or depressed in their parenting; the subscale Parent-child dysfunctional interaction indicates the extent to which parents feel contented with their child and the parent-child interactions; and the subscale Difficult child indicates the degree to which parents perceive their child as difficult to handle. Good convergent and discriminant validity for the PSI-SF have been demonstrated (Haskett, Ahern, Ward, & Allaire, 2006).

We rated parenting skills with the seven subscales of the Parenting Practices Inventory (PPI; Webster-Stratton, 1998; Webster-Stratton, Reid, & Hammond, 2001), a 73-item questionnaire on which parents indicate their reaction to misbehaviors of their child and their involvement in parental supervision. The PPI subscales include Appropriate discipline, Harsh & inconsistent discipline, Positive verbal discipline, Monitoring, Physical punishment, Praise and incentives, and Clear expectations.

We administered all outcome measures before randomization (T₁; baseline) and directly after the treatment or waiting list period (T₂). T₂ was around four months for the waiting list and our home-based treatment group, but variable for the care-as-usual treatment group ($M=13.4$ months; $Sd=3.6$), as the duration of this treatment was not fixed. The PSOC was also administered six months after both treatment conditions (T₃). As we could not keep parents in the waitlist condition for another six months, this

assessment was not done in that study arm. The primary caretaker filled in the questionnaires, in most cases (97.3%) the mother.

Sample size

We aimed to detect between-group differences on the primary outcome measure (ECBI) with an effect size (Cohen's d) of .4 with 90% power. Therefore, we needed to include $n=40$ children in each treatment arm. Due to lower referral to the study than expected, the recruitment was much more problematic than we had thought, which resulted in $N=73$ included families (March 2011-May 2014). Still, with this sample size the power for all outcome measures reported in this paper was above 80%, as calculated with GLIMMPSE (Kreidler et al., 2013).

Statistical analysis

Handling of missing data and outliers

If participants had more than 10% missing values on a questionnaire, or 20% missing values on a subscale, the total scores were not calculated but regarded as missing. In case of fewer missing values, the missing values were replaced by the average rating-per-item (sub)scale score. Using Cook's distance, we assessed potential outliers on all outcome measures.

Differences between study arms

With one-way analyses of variance (ANOVA; $p < .05$) we determined if there were significant differences between study arms with regard to characteristics of the treatments (treatment duration and number of sessions), medication changes during the trial, and received additional care during the trial.

Analyses of effects

* Home-based behavioral parent training versus waiting list

Effects of the manualized home-based treatment on the parenting outcome measures were analyzed with a linear mixed model for repeated measures (MMRM), following the intent-to-treat paradigm. We investigated the short-term effectiveness by comparing the

mean changes of the parenting outcome measures from baseline to T2 of BPTG@HOME with the waiting list. For the long-term effects, we only compared the mean changes on the PSOC subscales from baseline to T3 of the manualized home-based behavioral parent training with the care-as-usual treatment. Fixed effects in the models were treatment, time, and the interaction between time and treatment (time*treatment). The model intercepts were specified as random effects, using an unstructured covariance matrix for within-patient correlations ($p < .05$). The effects of the treatment were assessed by interpreting the time*treatment interaction effect, reflecting the short- or long-term change in outcome measure due to a specific treatment. The random intercept controlled for the variety present in the baseline scores on the outcome measures, ensuring that we did not confound patient heterogeneity with treatment effects.

We considered p values $< .05$ as indicating statistical significance. We did not adjust for multiple comparisons because, given the modest sample size, we were concerned to minimize Type II error. Effect sizes were computed by dividing the mean differences between BPTG@HOME and the waiting list/care-as-usual groups at T1 and T2/T3 by the pooled sample standard deviation (Taylor, 2015).

* Home-based behavioral parent training versus care-as-usual treatment

The same MMRM-analyses were performed to investigate differences between the mean changes of the parenting parameters from baseline to T2 in BPTH@HOME and in the care-as-usual treatment. When significant differences were found in the duration of the treatment, the number of sessions, medication changes during the trial, and/or amount of received extra care between study arms, MMRM analyses were repeated by extending the model with entering these variables. When more than two variables (given the “one in ten rule”; Peduzzi, Concato, Kemper, Holford, & Feinstein, 1996) showed a significant difference between study arms, we prioritized differences in treatment characteristics (duration of treatment and number of sessions). When we also found significant results at baseline on the other variables (medication changes during the trial and/or amount of received extra care), these variables were included in additional analyses combined with one of the two treatment characteristics.

Results

Outliers

For all outcome measures, no evidence for outliers was found, as Cook's distances varied between .00 and .35. This is well below the threshold of 1 (Cook & Weisberg, 1982).

Differences between study arms

No differences in medication use ($X^2(6, N = 53) = 10.0, p = .123$) and extra care during the trial (at T1: $X^2(38, N = 73) = 32.9, p = .706$; at T2: $X^2(28, N = 62) = 33.0, p = .235$; at T3: $X^2(13, N = 33) = 15.4, p = .285$) across study arms were found. The care-as-usual treatment lasted significantly longer (i.e., $M=13.9, SD=5.4$, range 5.7-20.8 vs. $M=5.3, SD=1.7$, range 1.3-8.8; $p=.006$) and consisted of significantly more home visits (i.e., $M=29.8, SD=18.0$, range 1-87 vs. $M=13.1, SD=5.1$, range 1-17; $p=0.001$) than BPTG@HOME.

Analyses of effects

Home-based behavioral parent training versus waiting list

Table 2 shows the results from the MMRM analyses. Compared to the waiting list, BPTG@HOME showed a significantly larger decline on PSOC Efficacy, PSI-SF Dysfunctional interaction, PSI-SF Difficult child, and PPI Harsh and inconsistent discipline between T1 and T2.

Home-based behavioral parent training versus care-us-usual treatment

Compared to the care-as-usual treatment, we found larger reductions in the BPTG@HOME group on the PPI Harsh and inconsistent discipline. Between T1 and T3, no significant differences on PSOC Efficacy and Satisfaction were found between BPTG@HOME and the care-as-usual treatment.

As the care-as-usual treatment lasted significantly longer and consisted of significantly more sessions than the manualized home-based treatment, we repeated the MMRM comparisons of BPTG@HOME versus the care-as-usual treatment at T1 and T2, while controlling for treatment duration and number of sessions. At T2, we now also found significantly larger reductions in the BPTG@HOME group in parenting stress (PSI-SF

total score; $\beta = 6.6$; $p = .019$; 95% CI = 1.1-12.1), harsh and inconsistent discipline (PPI Harsh and inconsistent discipline; $\beta = 7.3$; $p = .049$; 95% CI = .0-14.7), as well as larger increases in positive verbal discipline (PPI Positive verbal discipline $\beta = -4.2$; $p = .002$; 95% CI = -6.7 - -1.6), praise and incentives (PPI praise and incentives $\beta = -6.3$; $p = .001$; 95% CI = -9.3 - -3.4), and monitoring (PPI Monitoring $\beta = -1.8$; $p = .030$; 95% CI = -3.3 - -.2). The findings for T₃ remained nonsignificant.

Discussion

In this study, we investigated the effects of a manualized home-based behavioral parent training program as second-line non-pharmacological treatment for school-aged children with ADHD who had responded insufficiently to ADHD medication and clinic-based behavioral parent training on a range of secondary outcome measures related to parental functioning, i.e., parenting sense of competence, parenting stress, and a number of parenting skills (i.e., appropriate discipline, harsh and inconsistent discipline, positive verbal discipline, physical punishment, praise and incentives, clear expectations, and monitoring). Our results indicate effects of the treatment on several parenting parameters in a group of difficult to treat families. Concerning parenting skills, parents in the manualized home-based treatment showed significantly less harsh and inconsistent parenting compared to both the waiting list group and the care-as-usual treatment. This means that this improvement in parenting holds over and above non-specific effects of home-based treatment. Furthermore, compared with the waiting list, but not with the care-as-usual treatment, parents who received the manualized home-based behavioral parent training improved significantly more in their parenting efficacy (i.e., their perception of how capable they feel in their parental role) and concerning parenting stress (i.e., they experienced their child as less difficult to manage and evaluated the interactions with their child as less stressful). As these last results were not found compared with the care-as-usual home-based treatment, these improvements may be non-specific treatment effects of home-based treatment in general. In sum, while home-based treatment in general leads to improvements on a range of parenting parameters, the manualized home-based treatment additionally appears to lead to less harsh and

Table 2. Comparison of BPTG@HOME versus waiting list and care-as-usual treatment with linear mixed model for repeated measures.

	BPTG@HOME	Care-as-usual	Waiting list	BPTG@HOME vs. Waiting list				BPTG@HOME vs. Care-as-usual			
				β	p	95% CI	ES	β	p	95% CI	ES
Baseline (<i>M</i> (<i>SD</i>) [range])											
*PSOC											
Satisfaction	27.2 (1.2) [24.8-29.6]	27.3 (1.2) [24.8-29.7]	26.1 (1.3) [23.7-28.8]								
Efficacy	25.0 (.9) [23.2-26.9]	26.0 (.9) [24.1-27.8]	25.2 (1.0) [23.2-27.2]								
*PSI-SF											
Total score	100.0 (3.5) [93.1-106.9]	99.9 (3.5) [92.9-106.9]	90.6 (3.6) [83.3-97.8]								
Parental distress	28.9 (1.5) [25.9-32.0]	31.9 (1.6) [28.8-35.0]	26.6 (1.6) [23.4-29.8]								
Dysfunctional interaction	30.8 (1.4) [28.0-33.6]	27.8 (1.4) [25.0-30.7]	25.9 (1.5) [22.9-28.8]								
Difficult Child	40.1 (1.4) [37.4-43.0]	40.2 (1.4) [37.4-43.0]	38.2 (1.5) [35.3-41.1]								
*PPI											
Appropriate discipline	47.3 (1.6) [44.1-50.5]	53.4 (1.6) [50.1-56.6]	52.8 (1.7) [49.4-56.1]								
Harsh & inconsistent discipline	49.6 (2.3) [45.0-54.3]	53.5 (2.4) [48.8-58.3]	47.2 (2.5) [42.3-52.1]								
Positive verbal discipline	41.4 (1.4) [38.6-44.1]	44.4 (1.4) [41.4-46.9]	44.9 (1.4) [42.0-47.7]								
Physical punishment	9.1 (.6) [7.8-10.4]	10.0 (.7) [8.7-11.3]	8.3 (.7) [7.0-9.7]								
Praise and incentives	47.0 (1.7) [43.6-50.5]	47.6 (1.7) [44.2-51.1]	47.9 (1.8) [44.4-51.4]								
Clear expectations	21.3 (.8) [19.6-22.9]	23.1 (.8) [21.4-24.8]	22.3 (.9) [20.6-24.1]								
Monitoring	24.3 (1.0) [22.3-26.3]	24.8 (1.0) [22.7-26.8]	24.3 (1.1) [22.2-26.3]								
After treatment (<i>M</i> (<i>SD</i>) [range])											
*PSOC											
Satisfaction	22.5 (1.4) [19.8-25.2]	25.1 (1.6) [22.0-28.3]	23.1 (1.4) [20.2-25.9]	1.5	.419	-2.1-5.1	-	2.6	.183	-1.2-6.4	-
Efficacy	20.9 (1.0) [19.0-22.9]	22.6 (1.1) [20.4-24.8]	23.4 (1.0) [21.4-25.5]	2.5	.041*	-.1-4.9	.51	.8	.515	-1.7-3.3	-

*PSI-SF												
Total score	84.8 (3.9) [77.0-92.6]	92.1 (4.5) [83.2-101.1]	83.3 (4.2) [75.0-91.6]	7.6	.111	-1.8-16.9	-	7.7	.129	-2.1-17.6	-	
Parental distress	26.9 (1.5) [24.0-29.9]	30.6 (1.7) [27.2-34.0]	24.6 (1.6) [21.4-27.8]	.0	.991	-3.7-3.7	-	.7	.725	-3.2-4.6	-	
Dysfunctional interaction	24.7 (1.5) [21.8-27.7]	25.5 (1.7) [22.1-28.9]	23.8 (1.6) [20.7-27.0]	4.0	.049*	.1-8.1	.54	3.7	.086	-.5-8.0	-	
Difficult Child	33.2 (1.7) [29.8-36.6]	36.3 (1.9) [32.5-40.2]	35.1 (1.8) [31.5-38.7]	3.9	.040*	.2-7.6	.49	3.2	.123	-.8-7.1	-	
*PPI												
Appropriate discipline	47.4 (1.8) [44.2-51.3]	53.9 (2.1) [49.7-58.1]	52.0 (1.9) [48.2-55.8]	-1.2	.642	-6.2-3.8	-	.4	.888	-4.9-5.6	-	
Harsh & inconsistent discipline	40.8 (1.8) [37.2-44.4]	50.9 (2.1) [46.7-55.2]	44.6 (1.9) [40.7-48.5]	6.4	.038*	0.4-12.5	.58	6.5	.046*	.1-13.0	.58	
Positive verbal discipline	47.3 (1.1) [45.1-49.5]	47.7 (1.3) [45.1-50.3]	47.2 (1.2) [44.9-49.6]	-3.6	.094	-7.9-.6	-	-2.6	.251	-7.0-1.8	-	
Physical punishment	8.0 (.5) [6.9-9.1]	8.0 (.6) [6.7-9.3]	7.6 (.6) [6.4-8.8]	.5	.597	-1.3-2.2	-	-.8	.388	-2.7-1.3	-	
Praise and incentives	51.5 (1.5) [48.5-54.4]	47.2 (1.8) [43.7-50.7]	49.2 (1.6) [46.0-52.3]	-3.1	.216	-7.9-1.8	-	-4.9	.061	-10.0-.2	-	
Clear expectations	22.3 (.9) [20.7-24.3]	24.1 (1.0) [22.0-26.1]	24.0 (1.0) [22.1-26.0]	.5	.644	-1.6-2.6	-	.3	.784	-2.6-1.9	-	
Monitoring	26.2 (.9) [24.3-28.1]	24.8 (1.1) [22.6-27.0]	23.5 (1.0) [21.5-25.5]	-2.7	.068	-5.6-.2	-	-1.8	.243	-4.9-1.2	-	
6 months follow-up												
<i>(M (SD) [range])</i>												
*PSOC												
Satisfaction	23.1 (1.4) [20.2-26.0]	25.7 (1.6) [22.5-28.8]	-	-	-	-	-	2.7	.172	-1.1-6.3	-	
Efficacy	21.6 (1.0) [19.6-23.5]	22.5 (1.1) [20.3-24.7]	-	-	-	-	-	.0	.990	-2.7-2.7	-	

Note: *M* = mean; *Sd* = standard deviation; 95% CI = 95% confidence interval; ES = effect size; PSOC = Parenting Sense of Competence; PSI-SF = Parenting Stress Index-Short Form; PPI = Parenting Practices Inventory

* $p < .05$.

inconsistent parenting. This is in addition to our previous work (Nobel et al., 2019), where we showed that the manualized home-based treatment was more effective on several child parameters compared with the waiting list and the care-as-usual treatment (see Chapter 2). It should be noted though, that the comparison of the manualized home-based parent training with a waiting list was our primary comparison, and that comparison with the care-as-usual home-based treatment was done to investigate whether differences compared to waiting list would hold over and above non-specific effects of a care-as-usual home-based treatment.

Next to our main analyses described above, we also compared the manualized home-based treatment with the care-as-usual home-based treatment, while taking into account that the manualized treatment was on average twice as short and took on average half as many home visits. In that comparison, parents who received the manualized treatment experienced larger reductions in overall parenting stress and evaluated various parenting skills as more improved. In particular, they showed larger reductions in using harsh and inconsistent discipline, and more gains in using positive verbal discipline, praise and incentives, and monitoring their child. Apparently, the manualized home-based treatment reached in half the time and half the amount of sessions, at least the same results on these parameters as the care-as-usual treatment, which provides further support for the added value of the manualized treatment with regard to parenting outcomes. However, these effects on overall parenting stress, positive verbal discipline, praise and incentives, and monitoring were not found in comparison to the waiting list condition and should therefore be interpreted with caution.

Regarding parenting efficacy, our findings are in line with a meta-analysis of the effects of behavioral parent training that also showed a positive effect on parenting self-esteem. In seven of the eleven included studies in this meta-analysis behavioral parent training was compared to a waiting list (Coates, Talyor, & Sayal, 2015). Also, a study with preschool children with ADHD showed that parenting self-efficacy improved after a home-based parent training, compared to a psychoeducation control group (Lange et al., 2018). However, we did not find stronger improvement in parenting efficacy in the manualized home-based treatment group compared to those receiving care-as-usual treatment. This difference in findings between the two studies was possibly caused by the difference in control groups. Our care-as-usual treatment included not only

psychoeducation, as was the case in the study of Lange and colleagues (2018), but also support to parents and training in their parenting. This may have led to an increased sense of competence of parents in our control group, as was possibly not the case in the psycho-education group of Lange and colleagues. However, in contrast to our findings and those of Lange and colleagues (2018), and Sonuga-Barke, Daley, Thompson, Laver-Bradbury, and Weeks (2001) found a positive effect on parenting sense of competence in a behavioral parent training group compared with an active control group (a parent counseling and support group). To our knowledge, that study did not incorporate outcome ratings from parents who were blinded to treatment allocation, which we attempted to do in our study. This may have resulted in an overestimation of the effects on parenting sense of competence in the study of Sonuga-Barke and colleagues (2001). Other differences between our study and that of Sonuga-Barke were that the latter study offered the behavioral parent training as a first-line non-pharmacological treatment to parents of preschool children, while we conducted our study with a sample of school-aged children and with families who were not treatment naïve. Compared to parents of treatment naïve preschool children, parents of school-aged children who have already received extensive treatment (as in our group) may have experienced behavioral problems of their child for a longer period and may have had more failures and disappointments regarding their parenting role. This may have led to a parenting efficacy that was more resistant to change.

In randomized controlled trials of clinic-based behavioral parent training for school-aged children with ADHD, reductions of parenting stress were inconsistent. Chacko and colleagues (2009) found improvement in parenting stress, as opposed to Van den Hoofdakker and colleagues (2007) who did not find such improvement. In our study, we also did not find an effect on the total stress score of the PSI in the comparison between the home-based parent training and the waitlist. However, we found significant benefits of the home-based behavioral parent training group compared to the waiting list on two subscales of the PSI; parents evaluated the relationship with their child as more positive and experienced their child as less demanding. All studies measured parenting skills with the parent-report questionnaire Parenting Stress Index. However, they used different versions and (sub)scales, which makes comparison of the results difficult, and may explain the inconsistent findings.

The rationale of a behavioral parent training is changing the child's behavior by improving parenting skills. A meta-analysis in which most included studies had a behavioral parent training component (Daley et al., 2014), found significant effects of parent training on enhancing positive and reducing negative parenting. However, analyses of the separate parenting skills that make up "positive" and "negative" parenting were not done in most of these studies. Both our study and the study of Webster-Stratton, Reid, and Beauchaine (2011), which also used the PPI to investigate parenting behaviors, found that behavioral parent training had a significant effect on the reduction of harsh and inconsistent parenting. However, Webster-Stratton and colleagues (2011) also demonstrated that behavioral parent training significantly reduced the use of physical punishment by parents, which we did not find. Regarding positive parenting, our results suggest some effects on positive verbal discipline and praise and incentives, whereas Webster-Stratton and colleagues (2011) only found a significant result on appropriate discipline. It is unclear to us how these differences could be explained. However, it should be remembered that our study focused on school-aged children with parents receiving a second-line treatment (instead of preschool children and a first-line treatment).

The results of our study should be interpreted in light of its strengths and limitations. Important strengths included the focus on a difficult-to-treat group, the reliance on semi-blinded ratings, and the inclusion of two control groups so we could differentiate between non-specific and specific treatment effects. As limitations, first the modest sample size should be acknowledged. Second, regarding the assessment of parenting behaviors, we relied on a parent-based questionnaire rather than on direct observation of parenting behaviors. Finally, the relatively large number of statistical tests that we applied may have resulted in type I errors.

There are some clear avenues for future research. First, mediation analyses are necessary to directly relate improvements in parenting skills to reductions in children's disruptive behaviors. Second, to avoid expectation bias, it is important to investigate the association between parent ratings of parenting skills and observation of parenting skills at home by a blinded rater. Third, as an important difference between the two home-based treatments in this trial was the type of therapist who performed the treatment, future research should account for this.

In conclusion, the investigated manualized home-based behavioral parent training was more effective on multiple parenting parameters compared with a waiting list. Some of these changes in parenting may have been non-specific treatment effects (improvement in parenting efficacy and reductions in experiencing the child as difficult to manage and evaluating the interactions with the child as stressful), but specifically reductions in harsh and inconsistent parenting were also significant when the manualized home-based behavioral parent training was compared with a non-specific home-based treatment. As our previous work has shown that the manualized home-based parent training was also effective on several child outcomes, it should be considered as (second-line) treatment for children with ADHD who have responded insufficiently to regular outpatient treatments

Chapter 4

Which improvements in parenting mediate effects of home-based behavioral parent training for school-aged children with attention-deficit/hyperactivity disorder?

E. Nobel

P.J. Hoekstra

B.J. van den Hoofdakker

Abstract

Introduction: We aimed to investigate whether improvements in parenting would mediate changes in child behavior after home-based behavioral parent training in school-aged children with attention-deficit/hyperactivity disorder (ADHD) and disruptive behaviors (i.e., symptoms of oppositional defiant disorder [ODD]).

Method: In a multicenter randomized controlled trial ($N = 43$) comparing home-based parent training ($n = 22$) with a waiting list control group ($n = 21$), we investigated by using a pre-post treatment design the mediating effect of improvements of parenting parameters on improvements of the child's symptoms of ADHD and ODD. Parenting parameters were negative parenting (i.e., harsh and inconsistent parenting), parenting stress (i.e., stress concerning the manageability of the behavioral problems of the child and concerning the experience of the parent-child interaction), and parenting self-efficacy. Mediation effects were calculated with bias-corrected bootstrap confidence intervals using structural equation modeling, following the products-of-coefficients approach.

Results: Reductions in harsh and inconsistent parenting and parenting stress concerning the manageability of the behavioral problems of the child mediated improvements in ADHD and ODD symptoms, with 18.8-26.4% and 21.3-30.0%, respectively. No mediation effect was found for the reduction of the parents experiencing the interaction with their child as stressful. Parenting self-efficacy only showed a mediation effect on ODD symptoms (22.2-23.7%, depending on the outcome measure).

Conclusion: Our findings highlight that behavioral parent training should aim to reduce negative parenting and parenting stress and to increase parenting confidence.

Introduction

Behavioral parent training aims to improve children's behavior through training of parenting skills. In children with attention-deficit/hyperactivity disorder (ADHD) behavioral parent training has been shown to decrease children's ADHD symptoms and conduct problems (Coates, Taylor, & Sayal, 2015; Daley et al., 2014), and also to improve parenting sense of competence and quality of parenting behaviors (Coates et al., 2015; Daley et al., 2014). However, there is a lack of studies that have investigated to what extent changes in parenting behaviors mediate changes in children's behaviors, particularly in children with ADHD. Existing mediation studies have mostly been limited to the broad group of children with disruptive behavior problems, showing that approximately 45% of trials that investigated the effect of behavioral parent training also investigated mediation effects and in only 45% of these studies a mediation effect was found (Forehand, Lafko, Parent, & Burt, 2014).

Findings of the few behavioral parent training studies that investigated mediation effects of parenting in relation to child outcomes in school-aged children with ADHD have been inconsistent (Katzmann et al., 2017; Van der Stoep et al., 2017; Heath, Curtis, Fan, & McPherson, 2015; Chronis-Tuscano et al., 2011). Chronis-Tuscano and colleagues (2011), who investigated mediation effects in children with ADHD (six to twelve years old) whose mothers also had ADHD, found that reduced negative parenting mediated reductions of behavioral problems (i.e., symptoms of ADHD and/or oppositional defiant disorder [ODD]), whereas Katzmann and colleagues (2017) did not find this in a sample of children with ADHD and disruptive disorders aged four to eleven years old. The study of Heath and colleagues (2015) showed a mediating effect of reduced parenting stress and improved parenting self-efficacy on ADHD-symptoms in a sample of school-aged children (seven to twelve years old) with ADHD. However, Van der Stoep and colleagues (2017) found a reverse mediation effect in children with ADHD aged five to twelve years; reduced ADHD and disruptive symptoms mediated reductions in parenting stress. Katzmann and colleagues (2017) did not find a mediating effect of parenting self-efficacy on externalizing behaviors, but a mediating effect of reduced negative cognitions of the parent about their child on improvements in externalizing behaviors.

These inconsistent findings should not only be understood in light of the variety of samples that were examined in the studies, but also considering the different parent training programs. Regarding type of parent training programs, two of the studies examined mediation effects in guided self-help parent training (Katzmann et al., 2017; Van der Stoep et al., 2017). Another study (Chronis-Tuscano et al., 2011) examined a parent training program for children with ADHD who had a mother with ADHD. Only one study (Heath et al., 2015) used a standard group-based behavioral parent training program for children with ADHD. Apparently, mediation effects vary across samples and programs.

In this study, we investigated the mediation effects of reductions in negative parenting and parenting stress and improvement of parenting self-efficacy on child outcomes, during a manualized, home-based behavioral parent training program for school-aged children with ADHD and disruptive behaviors. Previously, we showed this parent training program to be more effective than a waiting list on reducing parent-rated child disruptive behaviors (i.e., symptoms of oppositional defiant disorder [ODD]) and ADHD symptoms (Nobel et al., 2019, see Chapter 2), as well as on improving a number of parenting factors. These included a reduction of negative parenting (i.e., harsh and inconsistent parenting), a decrease in parents' stress regarding the interaction with their child and regarding the manageability of the behavioral problems of their child, and improvement of parenting self-efficacy (see Chapter 3). In the present study we hypothesized that improvements on these parenting outcomes would mediate reductions of child disruptive behaviors and ADHD symptoms.

Methods

Participants and procedure

We used data of a multicenter randomized controlled trial in which participants were randomly assigned to either a home-based treatment (called Behavioral Parent Training Groningen at HOME [BPTG@HOME]; $n = 26$), a care-as-usual home-based family treatment ($n=24$), or a waiting list of four months ($n = 23$; for a detailed description of the trial design and procedures see Nobel et al., 2019, see Chapter 2). For the current study,

we only used data of the children in the BPTG@HOME and the waiting list arm ($N = 49$). All children met the following inclusion criteria: aged between six and thirteen years; attending primary school; having a diagnosis of ADHD (as obtained from medical records); and a full scale, verbal and performance IQ > 70 . Furthermore, they had responded insufficiently to previous treatments, which included at least attempts of ADHD medication and outpatient clinic-based parent training. After these treatments, Eyberg Child Behavior Inventory (ECBI) ratings still had to be in the clinical range (i.e., Intensity scale > 131 and Problem scale > 3 ; Eyberg & Pincus, 1999). Children could not be included when their families had received home-based treatment in the previous year.

The 49 children, 67.3% of whom were male ($n=33$) and 91.8% of Caucasian ethnicity, had a mean age of 8.77 years ($Sd=1.57$, range 6.01-12.1) and a mean full scale IQ of 94.4 ($Sd=11.5$, range 71-119). They all met DSM-IV-TR criteria of ADHD combined type (65.3%; $n=32$), ADHD predominantly hyperactive-impulsive type (14.3%; $n=7$), ADHD predominantly inattentive type (4.1% $n=2$), or ADHD not otherwise specified (16.3%; $n=8$); 51% also met the DSM-IV-TR diagnostic criteria of ODD and 10.2% of conduct disorder (CD; both disorders determined with the Parent Interview of Child Symptoms; Schachar, Ickowicz, & Wachsmuth, 1995). Furthermore, 51% of the children scored in the clinical range on the subscale Internalizing of the Child Behavior Checklist (Achenbach, 1991), as reported by the primary caregiver. Another 16.3% scored in the threshold range of this scale.

Treatment

We used a manualized, cognitive-behavioral home-based treatment for children with ADHD (Nobel & Van der Veen-Mulders, 2012), which was based on principles of Barkley's Defiant Children (1987), the program of Forehand & McMahon (Helping the Noncompliant Child; 1981), and Parent-Child Interaction Therapy (Eyberg & Funderburk, 2011). The treatment consisted of fourteen to sixteen weekly home visits (90-120 minutes per session), spread over four months, and was divided in different treatment modules. Basic treatment modules that were provided to all families were Assessment (selecting treatment modules and planning the number of sessions per module), Following (aimed at improving the parent-child relationship), Leading (aimed at preventing behavioral problems), and Compliance (aimed at coping with noncompliant behavior). Optional

treatment modules focused on specific parental problems and/or comorbid problems of the child. For a more detailed description of the treatment, see Nobel and colleagues (2019, Chapter 2). During the four-month waiting period, families did not receive any form of behavioral parent training.

Measures

We measured children's behavior problems with the parent-rated Intensity scale (36 items) of the ECBI, which assesses the frequency of behavioral problems on a seven-point scale (Eyberg & Pincus, 1999). The psychometric properties of the ECBI are generally good (Funderburk, Eyberg, Rich, & Behar, 2003; Silvan, Ridge, Gross, Richardson, & Cowell, 2008). Symptoms of ADHD and ODD were assessed with the Swanson, Nolan, and Pelham Questionnaire (SNAP-IV; Swanson, 1992), on which parents rated the severity of ADHD (18 items) and ODD symptoms (8 items) with a four-point scale. Reliability of this scale is high (Bussing et al., 2008).

Negative parenting was measured with the Harsh & Inconsistent Discipline subscale (15 items) of the Parenting Practices Inventory (PPI; Webster-Stratton, 1998; Webster-Stratton, Reid, & Hammond, 2001). The internal consistency for this subscale is good and the PPI has been shown to be sensitive for treatment effects (Webster-Stratton et al., 2001). The subscales Parent-Child Dysfunctional Interaction (12 items) and Difficult Child (12 items) of the Parenting Stress Index-Short Form (PSI-SF; Abidin, 1995) measured the degree to which parents experience the interaction with their child as stressful, and parenting stress concerning the manageability of the behavioral problems of their child, respectively. Good convergent and discriminant validity of the PSI-SF has been demonstrated (Haskett, Ahern, Ward, & Allaire, 2006). Finally, we assessed parenting self-efficacy with the Efficacy subscale of the Parenting Sense of Competence Scale (PSOC; Gibaud-Wallston & Wandersman, 1978; Johnston & Mash, 1989). For the Efficacy subscale a good internal consistency has been found (Johnston & Mash, 1989).

We administered all outcome measures before randomization (T₁; baseline) and directly after the treatment or waiting list period (T₂), which was around four months after T₁. All measures were filled in by the primary caretaker (in 97.3% of the cases the mother).

Statistical analysis

With independent sample t-tests we investigated possible differences between the two study arms on mean baseline scores of all measures. To examine mediation of change we followed the products-of-coefficients approach, which is recommended over the causal steps approach (e.g., Baron & Kenny, 1986) due to larger statistical power and lower risk of Type 1 error (MacKinnon, Lockwood, Hoffman, West, & Sheets, 2002). This approach investigates the presence of mediation by the statistical significance of the product $a*b$, where a is the coefficient relating the predictor variable to the mediator variable, and b is the coefficient relating the mediator variable to the outcome variable (figure 1). With c we denote the coefficient relating the predictor variable to the outcome variable, while c' represents the same coefficient, but controlling for change in the mediator (figure 1). We tested a pre-post treatment design using the change in variables from T1 to T2, in comparison to the waiting list control group.

Following Maric, Wiers, and Prins (2012), treatment status (BPTG@HOME or waiting list) was used as predictor variable, while the three child outcome measures were used as outcome variable. Parenting variables served as mediators. Mediation analyses were performed using structural equation modeling (SEM) in the R package *lavaan* (Rosseel, 2012). The SEM methodology is particularly useful to test mediation as it allows for simultaneous modeling of the direct (path c and path c' in Figure 1) and indirect effects (path a and path b in Figure 1; Blood, Cabral, Heeren, & Cheng, 2010). Bias-corrected

Figure 1. Mediation model for the effect of treatment condition (BPTG@HOME versus waiting list) on treatment outcome (i.e., child outcome), mediated by a mediator variable (i.e., parenting parameter)

bootstrap confidence intervals using 10,000 bootstrap iterations were obtained for all estimates, including the total indirect effect (path $a * \text{path } b$, Figure 1) (e.g., Sobel, 1982; Moscovitch, Hofmann, Suvak, & In-Albon, 2005). The indirect effect ($a*b$) represents the extent to which the change in parent outcome affects the relation between the treatment and child outcome (Preacher & Kelley, 2011). Statistical significance of the indirect effect evidences mediation. While this bootstrap analysis is particularly suitable for small samples (MacKinnon, Lockwood, & Williams, 2004), participants with missing data had to be excluded from the analyses in order to obtain valid estimates. As an additional measure of the strength of the mediation effect, we also computed the ratio of the indirect effect to the direct effect ($[(a*b)/c]*100$). Values closer to 100% indicating that a larger extent of the treatment effect on the child outcome was mediated by a change in the parent outcome (Moscovitch et al., 2005; Preacher & Kelley, 2011). P-values < 0.05 were considered as indicating statistical significance. Because we did not implement multiple measurements over time, we reversed the outcome and mediator variables as an additional test, to establish the direction of the mediation effect (Maric et al., 2012).

Results

Missing data

In six of the 49 children (12%; four in the BPTG@HOME group and two in the waiting list group) all T2 data were missing and they were therefore excluded from the mediation analyses (resulting in a final n of 43). Moreover, one child in the waiting list group had missing data on the SNAP ADHD symptoms, PSI Difficult Child, and PSOC Efficacy. This child was only excluded from analyses in which these measures were used.

Baseline differences

Table 1 shows that there were no significant differences between the BPTG@HOME and waiting list group in mean baseline scores on all measures, except a higher mean score on the PSI-SF Dysfunctional Interaction in the BPTG@HOME group compared to the waiting list group.

Table 1. Baseline differences between BPTG@HOME group and waiting list group

Measure	BPTG@HOME			Waiting list			<i>T</i>	<i>p</i>	95% CI
	<i>n</i>	<i>M</i>	<i>Sd</i>	<i>n</i>	<i>M</i>	<i>Sd</i>			
ECBI Intensity scale	22	160.8	22.2	21	149.7	19.1	1.767	.60	-1.60-23.95
SNAP ADHD scale	21	35.9	8.3	21	30.6	10.1	1.866	.91	-.44-11.07
SNAP ODD scale	22	13.9	4.8	21	11.9	5.3	1.331	.78	-1.06-5.16
PPI Harsh & inconsistent scale	22	50.3	13.3	21	46.1	10.5	1.129	.64	-3.27-11.55
PSI-SF Difficult child	22	41.4	5.9	20	37.1	7.4	2.091	.49	.14-8.49
PSI-SF Dysfunctional Interaction	22	31.6	6.6	20	26.0	7.8	2.561	.01	1.2-10.17
PSOC Efficacy scale	22	25.5	5.2	20	25.2	5.5	.228	.70	-2.96-3.71

Note: *n* = number of cases; *M* = mean; *Sd* = standard deviation; *T* = result of independent sample t-tests; *p* = significance level; 95% CI = 95% confidence interval; ECBI = Eyberg Child Behavior Inventory; SNAP = Swanson, Nolan, and Pelham Questionnaire; ADHD = attention deficit/hyperactivity disorder; ODD = oppositional defiant disorder; PPI = Parenting Practices Inventory; PSI-SF = Parenting Stress Index-Short Form; PSOC = Parenting Sense of Competence Scale.

Mediation of parental harsh and inconsistent discipline on changes in child behaviors

Table 2 presents the results of the analyses that tested whether changes in parental harsh and inconsistent discipline mediated changes in children's behavior problems, SNAP ADHD symptoms, and SNAP ODD symptoms. When comparing BPTG@HOME to the waiting list group, mediation of parental harsh and inconsistent discipline was found for the ECBI Intensity scale, the SNAP ADHD symptoms, and the SNAP ODD symptoms, as all three indirect effects ($a*b$) were found to be negative and significant ($p = .033$, $p = .049$ and $p = .040$, respectively). The mediator explained 24.3%, 18.8%, and 26.4% of the relation between the treatment and the respective treatment outcomes, indicating moderate effect sizes. Hence, part of the reductions of disruptive behaviors and ADHD and ODD symptoms can be attributed to reductions in harsh and inconsistent discipline when comparing BPTG@HOME to the waiting list condition.

Mediation of parenting stress on changes in child behaviors

Regarding the subscale Difficult Child of the PSI-SF, we found a mediation effect on improvements on the ECBI intensity scale and the SNAP ADHD and ODD symptoms, when comparing BPTG@HOME to the waiting list, as the indirect effects were negative and significant ($p = .025$, $p = .023$, and $p = .026$ respectively, Table 2). This mediator explained 30.0%, 21.3%, and 23.7% of the relation between the treatment and the ECBI Intensity scale, ADHD symptoms, and ODD symptoms, respectively (moderate effect

Table 2. Overview of mediation results for four mediator and three outcome variable combinations in trial comparing BPTG@HOME and a waiting list

	<i>a</i> (p-value)	<i>b</i> (p-value)	<i>a*b</i> (p-value) Indirect effect	<i>c'</i> (p-value)	<i>c</i> (p-value) Direct effect	$((a*b)/c)*100$ Percentage mediated (%)
<i>Outcome</i>	<i>Mediator: PPI Harsh and inconsistent discipline</i>					
ECBI Intensity scale	-.30 (.048*)	.33 (.016*)	-.10 (.033*)	-.31 (.040*)	-.41 (.007**)	24.3
SNAP ADHD scale	-.30 (.043*)	.28 (.039*)	-.09 (.049*)	-.40 (.011*)	-.48 (.001***)	18.8
SNAP ODD scale	-.30 (.043*)	.33 (.037*)	-.10 (.040*)	-.31 (.031*)	-.38 (.024*)	26.4
	<i>Mediator: PSI-SF Difficult Child</i>					
ECBI Intensity scale	-.29 (.061)	.38 (.005**)	-.11 (.025*)	-.30 (.043*)	-.41 (.007**)	30.0
SNAP ADHD scale	-.30 (.048*)	.35 (.005**)	-.10 (.023*)	-.37 (.010**)	-.47 (.001***)	21.3
SNAP ODD scale	-.29 (.061)	.31 (.018*)	-.09 (.026*)	-.29 (.067)	-.38 (.015*)	23.7
	<i>Mediator: PSI-SF Dysfunctional Interaction</i>					
ECBI Intensity scale	-.02 (.911)	.31 (.013*)	-.01 (.844)	-.40 (.004**)	-.41 (.007**)	-. ^a
SNAP ADHD scale	-.02 (.911)	.43 (.001***)	-.01 (.887)	-.47 (.001***)	-.47 (.001***)	-. ^a
SNAP ODD scale	-.02 (.911)	.30 (.044*)	-.004 (.826)	-.37 (.011*)	-.38 (.015*)	-. ^a
	<i>Mediator: PSOC Efficacy</i>					
ECBI Intensity scale	-.31 (.046*)	.30 (.036*)	-.09 (.043*)	-.31 (.046*)	-.41 (.007**)	22.0
SNAP ADHD scale	-.27 (.101)	.57 (.001*)	-.16 (.058)	-.32 (.012*)	-.48 (.001***)	-. ^a
SNAP ODD scale	-.29 (.061)	.31 (.018*)	-.09 (.026*)	-.29 (.067)	-.38 (.015*)	23.7

Notes: a = coefficient relating predictor to mediator; b = coefficient relating mediator to outcome; c' = coefficient relating predictor to outcome controlled for change in mediator; c = coefficient relating predictor to outcome; ECBI = Eyberg Child Behavior Inventory; SNAP = Swanson, Nolan, and Pelham Questionnaire; ADHD = attention deficit/hyperactivity disorder; ODD = oppositional defiant disorder; PPI = Parenting Practices Inventory; PSI-SF = Parenting Stress Index-Short Form; PSOC = Parenting Sense of Competence Scale. ^a As the indirect effect (a*b) was not significant, mediation was not established. Therefore this statistic was omitted.

* p < .05; ** p < .01; *** p < .001.

sizes). Therefore, part of reduction of disruptive behaviors, ADHD and ODD symptoms could be attributed to reductions in parents experiencing their child as difficult to manage when comparing BPTG@HOME to the waiting list condition. When considering the subscale Dysfunctional Interaction of the PSI-SF, no mediation was established for all three outcome measures, as the indirect effects were not significant (Table 2).

Mediation of parenting self-efficacy on changes in child behavior

The PSOC Efficacy scale mediated reductions on the ECBI Intensity scale and SNAP ODD symptoms when comparing BPTG@HOME to the waiting list as the indirect effects were significant and negative ($p = .043$, $p = .026$), but no mediation for the SNAP ADHD symptoms was found as the effect was not significant (Table 2). The mediator explained 22.2% and 23.7% of the relation between the treatment and the ECBI intensity scale and the SNAP ODD symptoms, respectively (moderate effect sizes). In conclusion, part of the reduction of disruptive behaviors and ODD symptoms could be attributed to an increase in parenting self-efficacy when comparing BPTG@HOME to the waiting list condition.

Reverse causality

Testing for reverse causality by reversing the outcome and mediator in the models did not result in significant mediation effects. Hence, we conclude that mediation runs in the hypothesized direction (i.e., from parent to child measure).

Discussion

We investigated whether improvements of ADHD symptoms and disruptive behaviors of school-aged children with ADHD through home-based behavioral parent training were mediated by improvements in parenting variables. We found that reductions of children's ADHD and ODD symptoms were mediated by reductions in harsh and inconsistent parental discipline and parents' stress concerning the manageability of the behavioral problems of their child. However, parenting self-efficacy only mediated ODD symptoms and not ADHD symptoms. Therefore, to change children's ADHD symptoms as well as ODD behaviors it appears to be particularly important to teach parents to use less harsh

and inconsistent discipline. With regard to changing ODD symptoms it may also be helpful to focus the parent training on improving parents' feelings of competence in their role as a parent. The improvement of parents in experiencing the interaction with their child as stressful did not mediate the reduction in the child's ADHD or ODD symptoms. Also, reverse mediation effects were not found for this variable. As the child's behavior may play a pivotal role in the parental judgement of the parent-child relationship it may thus be that the improvement in the quality of the parent-child relationship occurred simultaneously with the decline of the behavioral problems of the child.

Although we did not investigate which treatment element was associated with what specific improvement on parenting outcomes, we hypothesize that the treatment modules that focused on preventing disruptive behaviors and coping with noncompliant behavior (i.e., "Leading", "Compliance", and "Aggressive parental behavior towards the child") were important in reducing harsh and inconsistent parenting. These modules were specifically designed to teach parents parenting skills that are incompatible with harsh and inconsistent parenting. Furthermore, we tailored the treatment to individual families by taking parents' learning speed and confidence into account, included the module "Following" (which focuses on establishing a positive parent-child relationship) and performed cognitive interventions directed at altering negative parental cognitions at every home visit. Possibly, these treatment components were associated with the decrease of parenting stress concerning the manageability of the behavioral problems of the child and the improvement of parenting self-efficacy.

Our findings regarding the mediation effect of reductions of harsh and inconsistent parenting on the improvement of children's ODD symptoms are in line with the study of Chronis-Tuscano and colleagues (2011), who also found a mediating role of negative parenting on disruptive behavioral problems in an ADHD sample. Also, studies of behavioral parent training in children with disruptive behavior disorders showed that reductions of harsh and inconsistent parenting mediate improvements in children's disruptive behavior problems (Forehand et al., 2014; Sandler, Schoenfelder, Wolchik, & MacKinnon, 2011). However, our study was the first that showed that this mediation effect was not restricted to ODD symptoms, but were present in the reduction of ADHD symptoms as well.

Two previous studies contradict each other concerning the direction of the mediation effect between reduced parenting stress and disruptive behaviors of school-aged children with ADHD after behavioral parent training (Van der Stoep et al., 2017; Heath et al., 2015). These studies investigated parenting stress with the total score of the PSI-SF. Our study points to different outcomes for two different subscales of the PSI-SF. Our subscale findings suggest that the relation between improvements in parenting stress and improvements in children's behavior is complex. We could hypothesize that reductions of certain parental stressors (i.e., helping parents in experiencing the manageability of the behavioral problems of their child as less stressful) could lead to improvements in the child's behavior, while reductions in other parental stressors (i.e., parents experiencing the parent-child interaction as less stressful) perhaps develop more simultaneously with the child's behavior. Therefore, it is advisable to include the subscales of the PSI-SF in future mediation analyses of treatment studies, in order to get a better understanding of the interaction between different parental stressors and the child's behavior.

Our findings concerning the mediation effect of parenting self-efficacy on ODD symptoms were in line with the study of Katzmann and colleagues (2017). However, we did not confirm the findings of Heath and colleagues (2015), who found that change in ADHD symptoms was also mediated by parenting self-efficacy. This difference in findings may be due to the variety in child outcome measures or differences in the way parenting self-efficacy was assessed. We measured child symptoms with the SNAP, while the Behavior Assessment System of Children-Second Edition (Reynolds & Kamphaus, 2004) was used in the study of Heath and colleagues (2015). In our study, parenting self-efficacy was assessed with the PSOC Efficacy subscale, whereas the Self-Efficacy Parenting Tasks Index (Coleman & Karraker, 2000) was used by Heath and colleagues (2015). Also sample differences may have led to different findings compared to Heath and colleagues (2015), as we included children who were offered a second line, non-pharmacological, individualized home-based treatment, while in the study of Heath and colleagues (2015) children were treatment naïve and received group-based parent training. Possibly, these treatment naïve children and parents had a greater room for improvement on ADHD symptoms and parenting self-efficacy, which made detecting mediation effects statistically easier.

Limitations of our study were the small sample size and the lack of multiple

assessments over time, which makes causality harder to establish (Maric et al., 2012). To alleviate reverse causality concerns, reverse mediation analyses were performed.

Nevertheless, this study should be seen as an exploratory study that generates hypotheses for future research. Furthermore, there was a significant baseline difference between the BPTG@HOME group and the control group on the degree that parents experienced the interaction with their child as stressful. This baseline difference implies we should interpret the results (i.e., absence of mediation effects) for this variable with caution.

In conclusion, we found support for the notion that improvements in children's behavior through behavioral parent training are (partly) mediated by reductions in harsh and inconsistent parenting and parents' stress concerning the manageability of the behavioral problems of their child and improvement in parenting self-efficacy. This highlights the importance of aiming behavioral parent training programs at reducing negative parenting and parenting stress and improving parenting confidence.

Chapter 5

Parent-clinician agreement in rating presence and severity of attention-deficit/hyperactivity disorder symptoms

E. Nobel

J.A. Brunnekreef

R.J. Schachar

B.J. van den Hoofdakker

P.J. Hoekstra

*This chapter is published in ADHD Attention Deficit and Hyperactivity Disorders, 2019; 11; 21-29;
<https://doi.org/10.1007/s12402-018-0267-8>*

Abstract

We determined the validity of a parent-report questionnaire as a research tool for rating attention-deficit/hyperactivity disorder (ADHD) symptoms in children. Using Cohen's kappa and Pearson correlation, we examined the agreement between parent reports of ADHD symptoms (using the Swanson, Nolan, and Pelham Questionnaire-IV; SNAP-IV) and clinical judgment (using a semi-structured parent interview). Also, we explored factors that may be associated with the level of agreement, using regression analyses. We found moderate levels of agreement for severity of overall ADHD ($r = .43$) and for hyperactive impulsive symptoms ($r = .54$), but no significant agreement for inattentive symptoms. On individual symptom level (range kappa = $-.05$ – $.22$) and for the presence/absence of ADHD (kappa = $.14$), agreement was poor. Therefore, we conclude that parent-report questionnaires may be acceptable to rate the overall severity of ADHD symptoms in treatment effect studies, but not to detect the presence of ADHD in epidemiological studies.

Introduction

There is clear consensus on how to diagnose attention-deficit/hyperactivity disorder (ADHD) in school-aged children; this should be done on the basis of interviews with multiple informants such as parents and teachers who provide information on the child's behavior in different settings, like at home, school, or with peers (e.g., The National Institute for Health and Care Excellence [NICE], National Collaborating Centre for Mental Health & Royal College of Psychiatrists' Research and Training Unit, 2009). Moreover, the impairment of the symptoms is considered as crucial in establishing the presence and the severity of ADHD (American Psychiatric Association, 2013). However, such an extensive assessment is not always feasible in epidemiological or treatment studies, e.g., due to time and financial constraints. Parent-rated ADHD questionnaires are then frequently used. An important and unresolved question is how valid the use of these questionnaires is for determining the presence (as in epidemiological studies) and severity (as in treatment effect studies) of ADHD symptoms, in comparison with clinician ratings based on a semi-structured interview.

A well-known parent-based questionnaire is the Swanson, Nolan, and Pelham -IV (SNAP-IV; Swanson, 1992), which is frequently used as primary outcome measure in treatment studies (e.g., McCann et al., 2014; The MTA Cooperative Group, 1999; Yang, Gao, Li, & Zhao, 2015; Wagner, Vallerand, & McLennan, 2014) and for assessing ADHD in epidemiological studies (e.g., Tsai, Chen, Lin, & Gau, 2017; Yamada et al., 2013; Ullebø, Posserund, Heiervang, Obel, & Gillberg, 2012). The SNAP-IV determines the severity of ADHD symptoms by asking parents to rate the frequency of symptoms albeit without taking impairment into account. The validity of the SNAP-IV as a measure to determine the severity of ADHD and especially the presence of ADHD symptoms is uncertain. Some studies (Alda & Serrano-Troncoso, 2013; Bussing et al., 2008) concluded that the SNAP-IV is a reliable and valid *screening* instrument to identify children with ADHD concerns, but no studies have examined the psychometric properties of the SNAP-IV as a *diagnostic* tool. While another well-known rating scale, the Conners' parent and teacher scales (Conners, 2001) had good sensitivity (83.5%), but a disappointing level of specificity (35.7%) as a diagnostic tool for ADHD, clinical ratings with semi-structured interviews

with parents and teachers scored higher on both criteria (sensitivity: 91.8%; specificity: 70.7%; Conners, 2001).

One possible way to examine the value of a parent ADHD questionnaire for clinical trials and epidemiological studies is to examine the agreement between the parent-rated ADHD symptoms and those rated by clinicians with a semi-structured clinical interview. However, besides studies investigating agreement in children with externalizing disorders in general (Bird, Gould, & Staghezza, 1992; De Los Reyes & Kazdin, 2005; Jensen & Weisz, 2002; Klein, Lavigne, & Seshadri, 2010), no studies have focused on a specific diagnosis such as ADHD, nor did they compare a parent questionnaire and a clinician judgment based on a semi-structured interview.

This study aimed to provide a first exploratory step in examining the suitability of the SNAP-IV for two types of studies: epidemiological studies and treatment effect studies, by determining the agreement with regard to the presence and severity of ADHD symptoms between the SNAP-IV and clinicians' ratings based on a semi-structured parent interview. For this we chose the Parent Interview for Child Symptoms (PICS-IV; Schachar, Ickowicz, & Wachsmuth, 1995), as it closely follows the ADHD criteria of the diagnostic and statistical manual of mental disorders, fourth edition (DSM-IV; American Psychiatric Association, 2000) of which it provides concrete operationalizations. With the PICS-IV, it is the clinician who judges the presence and severity of each symptom based on parent reports of the frequency and the associated impairment of the behavior. While the PICS-IV is also dependent on parental input, the involvement of clinician's weighing is a major difference with a parent questionnaire in which ratings are being done directly by the parents. It may be questioned whether parents are able to assess the presence of DSM-IV based ADHD criteria, as the textual descriptions of the DSM are meant to be employed by individuals with appropriate clinical training and experience in diagnosis, which parents typically lack.

First, we examined agreement on two different levels, i.e., (1) meeting/not meeting overall DSM-IV symptom criteria for the presence of ADHD and (2) total severity of ADHD symptoms. The former is mostly used in epidemiological studies, and the latter in treatment effect studies. Additionally, to explore potential differences between agreement of parents and clinicians on specific symptoms, we also examined agreement on (3) the presence/absence of each of the 18 ADHD symptoms and (4) severity ratings of

each of the 18 ADHD symptoms. We expected to find low levels of agreement on symptom level and higher agreement for meeting overall DSM-IV symptom criteria (American Psychiatric Association, 2000) and total severity of ADHD symptoms as used in epidemiological and treatment effect studies, respectively. In addition, we explored whether certain child variables (i.e., age, sex, number of ADHD symptoms, severity of oppositional defiant behaviors, and medication status) and parental factors (i.e., education level, parenting stress, depressive symptoms, and ADHD symptoms of the primary caretaker) would be associated with the level of agreement. In children with externalizing disorders, better agreement between clinicians and parents has been demonstrated for males, younger children, more impaired patients, and in those with greater symptom severity (Bird et al., 1992; Jensen & Weisz, 2002; Klein et al., 2010). Some findings have indicated that higher parenting stress, more parental anxiety, and depression symptoms are associated with less agreement between parents' ratings and those of other informants (De Los Reyes & Kazdin, 2005).

Methods

Participants

Patients were recruited from three different mental health care centers in the North of the Netherlands. Data were obtained from a baseline assessment of a sample of 70 children who participated in a randomized controlled trial in which the effect of a home-based behavioral parent training for school-aged children with ADHD and behavior problems was being evaluated. All patients had previously been offered routine treatments (i.e., pharmacological treatment and/or parent training) without sufficient response, often due to non-completion of the treatment.

Children with ADHD, aged six to thirteen years, were eligible to participate in the study. At the time of referral to the study, all children met diagnostic DSM-IV-TR criteria for ADHD as confirmed by clinical interviews with parents and teacher. Comorbidity was not an exclusion criterion. The child had to have a full scale, verbal and performance IQ above 70. Table 1 contains the main sample characteristics.

The Medical Ethical Committee of the University Medical Center in Groningen has assigned ethical approval for the study (METC nr 2010.289). The trial is registered at <http://www.trialregister.nl>, home-based behavioral treatment for ADHD; NTR3021. All persons gave their informed consent prior to their inclusion in the study.

Table 1. Sample characteristics ($N=70$)

Number of boys (%)	50 (71.4%)
Mean age of child in years (SD; range)	8.9 (1.5; 6.0-12.1)
Mean intelligence of child ^a (SD; range)	94.0 (11.3; 71-119)
Number using ADHD medication (%)	61 (87.1%)
Number of highest finished educational level (%)	
Primary school	4 (5.7%)
High school	23 (32.9%)
Undergraduates programs	35 (50%)
College	8 (8%)
Mean parenting stress ^b (SD; range)	96.9 (17.2; 56-130)
Mean parental depression ^c (SD; range)	11.2 (8.5; 0-14)
Mean parental ADHD ^d (SD; range)	14.1 (9.6; 0-40)
Mean child ODD ^e (SD; range)	10.8 (4.3; 0-20)

Notes: SD = standard deviation, ADHD = attention-deficit/hyperactivity disorder, ODD = oppositional defiant disorder.

^a Full scale IQ. ^b Total score on the Parenting Stress Index- Short Form. ^c Total score on the Beck Depression Inventory-II.

^d Total score on the Adult ADHD rating scale. ^e Severity of ODD symptoms as rated by the Parent Interview of Child Symptoms.

Measures

The ADHD section of the SNAP-IV is an 18-item ADHD symptom scale (nine inattentive and nine hyperactive/impulsive DSM-IV symptoms) that is rated on a four-point scale from not at all (0) to very much (3) with regard to the previous week (Swanson, 1992), as rated by the primary caretakers of the children (i.e., mothers in 97% of all subjects). Reliability of this scale has been found to be high (coefficient alpha of .90 for inattentive symptoms and of .79 for hyperactive-impulsive symptoms; Bussing et al., 2008).

The PICS-IV is a semi-structured clinical interview with the parents that covers the full range of DSM-IV symptoms of ADHD and symptoms of oppositional defiant disorder (ODD), based on information restricted to direct observations in naturalistic contexts (daily situations) made by the parents. The clinician conducting the interview decides whether each symptom is present and rates its severity based on the frequency, the impairment that is associated with a particular symptom, and on the appropriateness of the behavior in relation to the child's age. Each symptom is rated by the clinician on a

four-point scale over the previous half year, ranging from symptom is absent (0), to marked abnormality (3). The clinician has to score blank when he/she does not get enough detailed information to score the symptom (De Los Reyes & Kazdin, 2005). The inter-rater reliability of the PICS-IV for an ADHD diagnosis has been shown to be good ($\kappa=.73$). Kappas for individual ADHD symptoms have been shown to range from .50 to .96. Intra-class correlation coefficients for overall symptom scores were also excellent (inattentive symptoms: .93; hyperactivity/impulsivity symptoms: .97; Ickowics et al., 2006). Clinicians in the study were psychologists or nearly graduated psychologist students (seven in total) who had been intensively trained in the PICS-IV and supervised by an experienced clinician.

Education level was defined as the highest level of completed education of the primary caretaker. Parenting stress by the primary caretaker was assessed with the Parenting Stress Index-Short Form (PSI-SF). This is a 36-item self-report measure where parents use a five-point scale to indicate the degree to which they agree with each statement (Abidin, 1995). Depressive and ADHD symptoms of the primary caretaker were rated with, respectively, the Beck Depression Inventory-II (BDI-II; Beck, Steer, & Brown, 1996; a 21-item self-report form with a four-point scale) and the Adult ADHD Rating Scale (AARS; Barkley & Murphy, 1998; an 18-item self-report form with a four-point scale). Table 1 provides means, standard deviations, and ranges of all measurements.

The primary caretaker filled in all questionnaires at the same time; the PICS-IV was always done after completion of the SNAP-IV, thus avoiding potential confounding of the parent ratings through knowledge of the clinician's questions. The mean time between completion of the questionnaires and the PICS-IV was .74 weeks ($SD=1.45$; range 0-8 weeks); for the majority of the participants, all data were obtained within two weeks ($n=63$; 90%). With both the SNAP-IV and PICS-IV, the behavior of the child was evaluated as how the child is currently seen by the parents in the home setting, regardless of the possible use of ADHD medication when prescribed.

Data analyses

Agreement

The agreement between parents and clinicians in severity of inattention, hyperactivity-impulsivity, and overall ADHD symptoms was determined with the Pearson product-moment correlation. The severity of ADHD symptoms according to the SNAP-IV and the PICS-IV was the sum of all individual symptom ratings.

Cohen's kappas (Cohen, 1960) were determined for parent-clinician agreement with respect to the ordinal severity ratings on each of the 18 ADHD symptoms and for parent-clinician agreement on the presence/absence of each ADHD symptom (ratings 0 or 1 versus ratings 2 or 3 on the SNAP-IV and PICS-IV). Furthermore, the agreement between parents and clinicians with respect to criterion A of the DSM-IV description of ADHD (i.e., the child has six or more inattentive symptoms and/or hyperactive-impulsive symptoms) was determined. Based on the number of symptoms on the SNAP-IV (six or more symptoms per domain, following the DSM-IV), the children were divided into four categories (ADHD combined type, ADHD predominantly inattentive type, ADHD predominantly hyperactive-impulsive type, and not fulfilling full ADHD criteria). The same was done regarding the number of symptoms on the PICS-IV. Agreement in ADHD types was again calculated with Cohen's kappas. Kappa scores were interpreted using Altman's (1994) conventions: <.20 as poor, .21-.40 as fair, .41-.60 as moderate, .61-.80 as good, and .81-1.00 as very good level of agreement.

Participants who had more than 10% missing values per measurement were excluded from the analyses (loss of 3 participants). In case of less missing values (SNAP $n=4$; PICS $n=12$), these were replaced by the average rating-per-item subscale score.

Factors influencing agreement

First, we explored which child characteristics (i.e., number of ADHD symptoms and severity of oppositional defiant behavior) and parental variables (i.e., education level, parenting stress, depressive symptoms, and ADHD symptoms of the primary caretaker) might have been related to the level of agreement by determining Pearson product-moment correlations of all continuous variables with the difference scores between the ADHD total, ADHD inattentive, and ADHD hyperactive-impulsive scores on the SNAP-IV

and PICS-IV. We first used t-tests to assess difference scores between the SNAP-IV and the PICS-IV between categories (i.e., sex of the child and the presence/absence of current ADHD medication of the child). Factors with a p value below .10 were subsequently analyzed in a multiple regression analysis. When running these analyses, we checked for multicollinearity using variance inflation factors.

Parenting stress, depressive symptoms, and ADHD symptoms of the primary caretaker were determined by calculating the total scores on the PSI-SF, BDI-II, and AARS, respectively. Missing values (less than 10% per questionnaire) were replaced by the average rating-per-item subscale score (PSI-SF $n=8$; BDI $n=5$; AARS $n=5$). The presence of ADHD-symptoms of the child was based on ratings of 2 or 3 ratings on PICS-IV ADHD items. The level of the child's ODD was based on the sum of all PICS-IV ODD symptoms each rated on a 0-3 scale.

We also performed a number of sensitivity analyses where missing values were analyzed differently. First, when establishing the level of agreement between the severity and the presence of single symptoms and meeting criteria A of the DSM-IV, blank scores on the PICS-IV were treated as zero scores (instead of as missing values). Second, when establishing total scores on the different questionnaires, we replaced missing values in a sensitivity analysis with a zero score (instead of the average rating-per-item).

We considered p values $<.05$ as indicating nominal statistical significance. However, as we performed 49 independent tests, we controlled for multiple-comparison using the Benjamini-Hochberg procedure (false discovery rate). This procedure ranks the p values in ascending order (the smallest has a rank of 1), computes a critical value based on the rank and the number of tests, and considers results as significant of which the p value is smaller than the critical value. This procedure suggested considering a test as significant if $p < .009$. Significance in tables and text has been determined with respect to this value.

Results

Parent clinician agreement

With regard to overall symptom severity (the sum of all individual symptom ratings; $n=67$), the SNAP-IV severity of hyperactivity/impulsivity ($M=15.9$; $SD=5.6$; range 3-27) and

total ADHD symptoms ($M=32.1$; $SD=9.6$; range 5-51) were significantly related to the PICS-IV severity of hyperactivity/impulsivity ($M=14.2$; $SD=5.0$; range 4.5-27) and total ADHD symptoms ($M=25.8$; $SD=6.9$; range 9-46). For the severity of the hyperactivity/impulsivity symptoms ($r=.54$, $p<.009$, 95% CI .35-.69) and for the total ADHD symptoms ($r=.42$, $p<.009$, 95% CI .20-.60), moderate effects were found. No statistically significant effect for the severity of inattention ($r=.33$, $p=.01$, 95% CI .01-.53) was found. Also, the SNAP-IV severity of inattention ($M=16.2$; $SD=5.2$; range 2-27) was not significantly related to the PICS-IV severity of inattention ($M=11.5$; $SD=3.9$; range 3-22). A sensitivity analysis (where missing values were replaced with a zero-score instead of average rating-per-item score) yielded similar results.

Furthermore, we determined Cohen's kappas at the level of meeting criteria A of the DSM-IV classification. We found no significant kappas with regard to agreement on types of ADHD ($\kappa=.14$; $p=.067$; $n=52$, 95% CI -.05-.34) and on the presence or absence of any ADHD type ($\kappa=.19$; $p=.139$; $n=52$, 95% CI -.15-.53). Table 2 presents Cohen's kappas for agreement between the SNAP-IV and PICS-IV regarding severity and the presence/absence of single ADHD symptoms. In contrast to the moderate agreement between parents and clinicians on the overall severity of ADHD symptoms, the levels of agreement between parents and clinicians on the severity of single ADHD symptoms and on the presence/absence of single symptoms were mostly poor. For one of the 18 ADHD symptoms ("talks excessively"), significant agreement with regard to severity of ADHD symptoms was found, and for five symptoms ("loses things"; "fidgeting/squirming"; "runs about/climbs excessively"; "talks excessively"; and "blurts out answers before questions were completed") concerning the presence/absence of symptoms. For all other ADHD symptoms, significant agreement was absent. A sensitivity analysis (where a blank score on the PICS-IV was transformed into a zero score instead of a missing value) resulted in similar outcomes.

Factors influencing agreement

In explorative analyses (Pearson product-moment correlations for continuous variables and t -tests for categorical variables) the child's current medication use ($t=1.81$; $p=.076$), parenting stress (PSI-SF; $r=.21$, $p=.090$), and the total number of ADHD symptoms according to the PICS-IV ($r=-.24$; $p=.053$) were identified as candidate factors for

agreement on total ADHD severity. For agreement on ADHD inattentive severity, candidate factors were parenting stress (PSI-SF; $r=.20$; $p=.098$) and number of inattentive ADHD symptoms according to the PICS-IV ($r=-.31$; $p=.012$). Current medication use of the child ($t = 1.774$; $p = .081$), number of hyperactive-impulsive symptoms based on the PICS-IV ($r = -.32$; $p = .010$), and total number of ADHD symptoms according to the PICS-IV (r

Table 2. Level of agreement between the SNAP-IV and PICS-IV in the severity and presence of single ADHD symptoms

ADHD symptom	<i>n</i>	Severity	<i>p</i>	95% CI	Presence	<i>p</i>	95%CI
Fails to pay attention to details	68	-.004	.958	-.14-.13	.29	.014	.06-.52
Cannot sustain attention	70	-.05	.339	-.15-.15	-.07	.409	-.35-.11
Does not seem to listen when spoken to	69	.15	.038	.00-.30	.17	.147	-.05-.39
Does not follow through on instructions	67	.04	.586	-.11-.19	.11	.341	-.13-.35
Disorganizing tasks and activities	66	.08	.180	-.05-.21	.09	.410	-.13-.31
Avoids tasks that require sustained mental effort	66	-.01	.926	-.14-.12	.04	.744	-.22-.30
Loses things	70	.08	.235	-.06-.22	.46	<.001*	.25-.67
Distractibility	70	.06	.303	-.05-.17	.06	.357	-.05-.17
Forgetful	69	.04	.567	-.11-.19	.14	.214	-.07-.35
Fidgeting/squirming	70	.06	.428	-.10-.22	.34	.005*	.10-.58
Difficulty staying seated	65	-.003	.964	-.14-.14	.22	.066	-.01-.43
Runs about/climb excessively	70	.10	.150	-.04-.24	.38	.001*	.17-.59
Not playing quietly	70	-.003	.968	-.14-.14	.16	.170	-.06-.38
On the go/driven by a motor	70	.15	.031	.00-.30	.26	.027	.02-.50
Talks excessively	70	.22	.002*	.07-.37	.42	<.001*	.20-.64
Blurts out answers before questions were completed	70	.17	.017	.01-.34	.30	.008*	.09-.51
Does not wait for his/her turn	65	.11	.078	-.02-.24	.17	.084	-.02-.36
Interrupts/intrudes on others	67	.09	.107	-.04-.21	.16	.052	-.01-.30

Notes: SNAP-IV= Swanson, Nolan, and Pelham Questionnaire-IV; PICS-IV= Parent Interview of Child Symptoms; ADHD= attention-deficit/hyperactivity disorder; *n*= number of participants; Severity= kappa of agreement on symptom severity; Presence= kappa of agreement on presence/absence of symptom; 95% CI= 95% confidence interval.

* $p < .009$ (according to Benjamini-Hochberg correction for multiple testing)

$= -.24$; $p = .051$) were the factors for agreement on ADHD hyperactive-impulsive score. Severity of comorbid oppositional defiant behavior, parental educational level, and parental psychopathology (depression and ADHD symptoms) were not related to agreement on total, hyperactive, or inattentive ADHD severity (p values $> .10$).

Parents rated ADHD severity higher than did clinicians, with the difference between the SNAP-IV-based total ADHD severity and the PICS-IV based total ADHD severity being on average 6.3 points ($SD=9.1$; range $-11.4--27.0$); only 20.9% of the primary caretakers scored lower on the SNAP-IV than did the clinician on the PICS-IV. The difference between the severity score on the inattentive ADHD symptoms according to the SNAP-IV- and the PICS-IV-based severity was on average 4.7 points ($SD=5.4$; range $-7.4-16.0$); only 16.4% of the parent scores were lower than the clinician scores. For the total severity on hyperactive-impulsive symptoms, the average difference between the two measurements was 1.7 points ($SD= 5.1$; range $-9.0-13.0$). Here, 38.8% of the parents scored lower than the clinicians.

Subsequent regression analysis established that the more parenting stress the parents experienced (PSI-SF; $\beta=.13$, $p=.038$, 95% CI $.01-.26$) and the less ADHD symptoms were scored by the clinician (PICS-IV; $\beta=-.80$, $p=.025$, 95% CI $-1.50- -.104$), the larger was the difference between the parents and the clinician, with the primary caretakers scoring the ADHD severity higher than the clinicians. The child's medication status ($\beta=-4.57$, $p=.145$, 95% CI $-10.77-1.62$) did not significantly influence parent-clinician differences. The regression analysis for the difference between SNAP-IV and PICS-IV on inattentive severity revealed that the lower the number of ADHD inattentive symptoms was scored by the clinician (PICS-IV; ($\beta=-.99$, $p=.008$, 95% CI -1.72 to $-.26$), the bigger was the difference between the parents and the clinicians in severity, with the parents scoring higher than the clinicians. No statistically significant results were found for the factor parenting stress ($\beta=-.07$, $p=.063$, 95% CI $-.01-.14$).

Furthermore, the factors child's current medication use of the child ($\beta=-2.64$, $p=.140$, 95% CI $-6.17-.89$), number of hyperactive-impulsive symptoms based on the PICS-IV ($\beta=-.84$, $p=.105$, 95% CI $-1.86-.18$), and the total number of ADHD symptoms according to the PICS-IV ($\beta=.14$, $p=.700$, 95% CI $-.59-.87$) were not significantly associated with the difference score between the SNAP-IV and the PICS-IV on the hyperactive-impulsive

severity. Multicollinearity was found not be an issue, as the variance inflation factors remained well below the accepted boundary of 10 (Hair, Black, Babin, & Anderson, 2010), with all variance inflation factors values being below 4.

A sensitivity analysis (where missing values were replaced with a zero-score instead of average rating-per-item score) for the total ADHD severity and the hyperactive-impulsive severity yielded similar results. However, in the sensitivity analysis the inattentive severity of the number of inattentive symptoms according to the PICS-IV ($\beta = -.99$, $p = .009$) fell just short of significance in association with the level of agreement with regard to inattentive severity.

Discussion

We investigated the agreement between parent reports of ADHD symptoms as measured by a parent questionnaire (the SNAP-IV) and clinicians' ratings of ADHD symptoms based on a semi-structured parent interview (the PICS-IV), to evaluate the suitability of parent questionnaires for two types of studies: epidemiological studies and treatment effect studies. First, we found no significant agreement between the SNAP-IV and the PICS-IV on ADHD type and on the presence or absence of ADHD in school-aged children. Based on these results, it is not recommended to use parent questionnaires to determine the presence of ADHD, as is commonly done in epidemiological studies (e.g., Tsai et al., 2017; Yamada et al., 2013; Ullebø et al., 2012). While there are a few changes in ADHD diagnostic criteria between the DSM-IV-TR and the current (fifth) edition of the DSM (DSM 5; American Psychiatric Association, 2013), these are relatively minor and there is no reason why DSM 5-based parent questionnaires would compare differently to clinicians' interviews than those based on the DSM-IV-TR.

Second, in treatment effect studies, parent questionnaires such as the SNAP-IV are frequently used to determine the change over time in severity of ADHD symptoms (e.g., McCann et al., 2014; The MTA Cooperative Group, 1999; Yang et al., 2015; Wagner et al., 2014). We found moderate levels of agreement with respect to total ADHD symptom severity, and therefore, these questionnaires seem to be suitable as severity rating in

treatment effect studies. Most parents (79.1%) rated ADHD severity of their child higher compared with the judgment of clinicians.

Parents appeared to be in line with clinicians in evaluating the presence and severity of the total severity of hyperactive and impulsive symptoms but not so in assessing the total severity of inattentive symptoms of their child. The same pattern was found for the agreement between parents and clinicians on single ADHD symptoms; the symptom with a significant level of agreement in severity was a hyperactive symptom, and also, four of the five symptoms with a significant level of agreement in the presence/absence were hyperactive-impulsive symptoms. Hyperactive-impulsive symptoms are generally more readily externally observable behaviors, while inattentive symptoms are less recognizable; this may explain that the agreement between parents and clinicians was better for hyperactive-impulsive symptoms than for inattentive symptoms. This assumption is confirmed by the significant agreement level on the inattentive symptom that is relatively concrete and observable: "often loses things". This symptom is not only more concrete, but it also shows less overlap with other inattentive symptoms, in contrast to the overlap between "cannot sustain attention" and "distractibility" and the overlap between "does not follow through on instruction", "forgetful", and "disorganizing tasks and activities".

These findings may implicate that in order to make parent questionnaires suitable for treatment effect studies, especially the inattentive symptoms of ADHD should be described in a more concrete, observable way instead of following exact DSM descriptions, which have never been designed to be rated by parents. In addition, not only a frequency rating of the behavior but also an impairment rating should preferably be given by the parent. Future studies might investigate whether such adjustments in a parent questionnaire would lead to better levels of agreement between a parent rating and a clinician's judgment. Some parent questionnaires do already contain some global form of impairment rating, e.g., the National Institute for Children's Health Quality ADHD rating scale (NICHQ Vanderbilt Assessment Scales, 2002) asks parents to rate their child's performance in a variety of domains, yet do not break-down whether the presence of each ADHD symptom criterion is justified by it being associated with impairment. However, an additional advantage of a semi-structured interview over parent-report questionnaires in treatment effect studies is the possibility to establish blind

measurements (Daley et al., 2014; Sonuga-Barke et al., 2013). It would also be worthwhile to investigate whether teachers show better agreement with clinicians with regard to inattentive symptoms, as these may perhaps be easier observed in a classroom setting, where sustained attention and engagement with tasks are critically important.

The number of child ADHD symptoms appeared to influence the level of agreement on the severity of inattentive symptoms of ADHD between parents and clinicians. Fewer child ADHD symptoms were associated with more disagreement between parents and clinicians, whereas parents tend to estimate the severity of the inattentive symptoms higher than the clinician. Notably, we did not find factors that were related to the agreement on the severity of the more concrete, observable hyperactive-impulsive symptoms. Apparently, child factors have no significant influence on agreement when ADHD symptoms are described in concrete, objective, and observable terms such as most hyperactive and impulsive symptoms, in contrast to the less distinctive and observable inattentive symptoms. This may point to the necessity of clinical expertise to assess such symptoms, and again therefore, semi-structured interviews should be considered in treatment effect studies. Furthermore, we found no relation between level of agreement and parental educational level, parental psychopathology (depression and ADHD symptoms), and severity of comorbid oppositional defiant behavior.

While not the main focus of the study, our findings confirm existing recommendations to use semi-structured interviews to diagnose children suspected to have ADHD in clinical practice rather than to rely on parent-report questionnaires, preferably by collecting data from multiple settings. Indeed, the NICE-guideline states that a diagnosis of ADHD should not be made on the basis of rating scales alone (National Collaborating Centre for Mental Health & Royal of Psychiatrists' Research and Training Unit, 2009). This recommendation also aligns with the finding that questionnaires alone fail to systematically assess all relevant clinical features of a disorder (Parker & Corkum, 2016; Raiker et al., 2017).

Limitations

Some limitations of this study need to be acknowledged. The most important limitation is the relatively modest sample size and the presence of missing data. Moreover, we possibly included a diagnostically more complex and mostly medicated group. This

selection bias may have resulted in over-estimation of ADHD symptom severity by parents. Another limitation of the study is that the SNAP-IV and PICS-IV give different instructions in judging the severity of an ADHD symptom and with regard to setting. That is, the SNAP-IV asks parents to assess how frequent a specific symptom is present at home or at school in the previous week, whereas in the PICS-IV the clinician is asked to rate the severity of a specific symptom in specific situations at home in the previous half year, taking into account the frequency, impairment, and age appropriateness of the behavior. To improve the comparability of the scorings on a parent questionnaire and a semi-structured interview in future studies, it would be preferable to give parents and clinicians the same timescale and context. This difference in timescale can also influence the agreement with regard to the presence/absence of ADHD, and therefore, our conclusion that the SNAP-IV is less acceptable for epidemiological studies needs to be interpreted carefully.

Furthermore, it should be remembered that the reliability and validity estimates from the SNAP-IV and PICS-IV stem from English versions and may differ from the Dutch versions that we used. However, the PICS-IV is a semi-structured interview in which clinicians do not use literal wordings, whereas translation of the SNAP-IV was in line with the official DSM-IV-TR Dutch translation. Thus, we are confident that the influence of translation is negligible. Finally, when interpreting the level of agreement based on correlation coefficients, one should keep in mind that these are insensitive against linear differences in severity between both methods. Furthermore, while the PICS-IV has been previously shown to have good inter-rater reliability (Ickowics et al., 2006), we did not assess inter-rater reliability for the PICS-IV assessors of the current study, as individual children were only rated by one clinician instead of multiple.

Conclusions and future directions

In conclusion, a DSM-based parent questionnaire appears to be less valid to establish whether an ADHD diagnosis is present or absent, as is commonly done in epidemiological studies. Therefore, the use of parent questionnaires for diagnostic purpose should be reconsidered, and we recommend the use of continuous ratings of ADHD severity rather than categorical measures of ADHD when parent ratings are being used in epidemiological samples. Moreover, parent questionnaires appear to be appropriate

instruments to rate the overall severity of hyperactive-impulsive ADHD symptoms in children in treatment effect studies, but less suitable to rate the severity of individual symptoms. This is especially true for children with fewer ADHD symptoms. Future research should examine the test-retest reliability of parent questionnaires, as the consistency of measurement across time is a critical indicator of a measure's utility as a treatment outcome index. A semi-structured interview in which a clinician evaluates the presence and severity of the ADHD behavior may be a better alternative, because clinicians are better at rating the more internal inattention symptoms and, moreover, because these instruments can be used in a blinded way. Parent questionnaires can, however, be improved by describing the inattention symptoms in more concrete, observable behaviors of a child. Future research should also focus on the influence of the use of different clinicians in scoring ADHD symptoms. Unfortunately, this could not be done reliably in the current study, given the small sample size.

Chapter 6

Summary and general discussion

Aims of this thesis

The primary aim of this thesis was to investigate the short- and long-term effectiveness on child and parent outcomes of a home-based behavioral parent training as a second-line treatment for school-aged children with attention-deficit/hyperactivity disorder (ADHD) and disruptive behaviors (i.e., symptoms of oppositional defiant disorder [ODD]) who had responded insufficiently to pharmacotherapy and/or clinic-based behavioral parent training. Both medication and behavioral parent training are well established and evidence-based therapies for children with ADHD (Cortese et al., 2018; The MTA Cooperative Group, 1999; Coates, Taylor, & Sayal, 2015; Evans, Owens, Wymbs, & Ray, 2018). Several families need more or other treatment after these two interventions, due to insufficient response, discontinuation of pharmacological treatment, or low attendance in parent training (Banaschewski et al., 2006; Riera et al., 2017; Van de Loo-Neus, Rommele, & Buitenlaar, 2011; Graham et al., 2011; Assenary & McIntosh, 2002).

A secondary aim of this thesis was to examine whether changes in parenting factors mediated improvements in child behaviors. Finally, whether the severity of a child's ADHD symptoms can be validly assessed by using parent-rated questionnaires as trial outcome measures is disputed in the literature (Daley et al., 2014; Sonuga-Barke et al., 2013). Therefore, a third aim was to explore the suitability of the parent-rated questionnaires for determining ADHD presence and severity that were used in this study.

Home-based behavioral parent training: theoretical underpinnings

For this study, we developed a home-based behavioral parent training specifically designed for families with a school-aged child with ADHD and disruptive behaviors who had not benefited sufficiently from routine treatments. This home-based treatment, called Behavioral Parent Training Groningen at HOME (BPTG@HOME), is a manualized, cognitive-behavioral parent training. The content of the treatment was based on the principles of evidence-based behavioral parent programs (e.g., Barkley's Defiant Children

[1987], *Helping the Noncompliant Child* [Forehand & McMahon, 1981], and *Parent-Child Interaction Therapy* [Eyberg & Funderburk, 2011]).

In this new parent training program we incorporated several elements, based on prior studies, to make it suitable for families who were difficult to treat. First, we offered the treatment at home, as studies have shown that attendance rates of parent training were higher when treatment was located closer to home (i.e., in neighborhood schools rather than in local clinics; Cunningham, Bremner, & Boyle, 1995) or at home (Sonuga-Barke et al., 2018). Also, Moran and Gbate (2005) advised to include home visits in parenting programs. A meta-analysis of Kaminski, Valle, Filene, and Boyle (2008) showed that letting parents practice with their child during treatment sessions (using modelling and video-feedback) was associated with better outcome of parent training. These practices can be more easily organized in a home-based treatment. Second, the new treatment offers a combination of behavioral management techniques and relationship enhancing strategies. A recent meta-analysis showed the superior effectiveness of parenting programs for reducing disruptive child behavior that integrate relationship enhancement with behavior management techniques compared to behavior management alone (Leijten et al., 2018). Third, the treatment has a modular format, because research on barriers to parent training programs has shown that parents highly appreciate flexible and individually tailored programs (see for review Koerting et al., 2013). Fourth, BPTG@HOME includes treatment modules with which family problems can be addressed, as Smith and colleagues (2015) recommended to address not only the child's needs in parent training, but also the parents' own needs. Fifth, in each home visit the therapist focused on the dysfunctional parental cognitions regarding their parenting and the behavioral problems (i.e., symptoms of ADHD and/or ODD) of their child. Studies showed that altering dysfunctional cognitions of the parents was associated with better treatment effects (Chronis, Jones, & Raggi, 2006; Cunningham, 2007; Daley et al., 2018).

Study design

All studies in this thesis were based on data of a three-armed, parallel, multicenter, superiority, randomized controlled trial with balanced randomization. This was a

pragmatic effectiveness trial, fully embedded in out-patient mental health clinics in the Netherlands. The trial included children with ADHD who still had disruptive behaviors after receiving, or at least considering pharmacotherapy and/or clinic-based behavioral parent training, irrespective of the cause of the remaining impairments.

We included two control groups. First, BPTG@HOME was compared with a waiting list group to investigate the effectiveness of BPTG@HOME. Second, BPTG@HOME was compared with a home-based care-as-usual group to control for non-specific home-based treatment effects. Parents were not told which treatment they received (BPTH@HOME or home-based care-as-usual treatment). The effectiveness was investigated on multiple parent-reported parameters of the child (e.g., ADHD symptoms, comorbid disruptive behaviors, and internalizing symptoms; Chapter 2) and the parent (e.g., parenting skills, parenting sense of competence, and parenting stress; Chapter 3). Measurements took place before treatment, directly after treatment and when families received one of the two treatments six months after finishing treatment. To examine the mediation effects of parenting factors on child behaviors, BPTG@HOME was compared with the waiting list control group, using a pre-post treatment design following the products-of-coefficients approach (chapter 4). To investigate the validity of a parent-report questionnaire as outcome measure for rating attention-deficit/hyperactivity disorder (ADHD) symptoms in children, the agreement between parent reports of ADHD symptoms and clinical judgment was explored, using the pre-treatment data of the children who were included in the trial (chapter 5).

Clinical relevance: main findings

Effectiveness of BPTG@HOME as second-line treatment

In chapter 2, we reported that children who received BPTG@HOME (compared to the waiting list group and the home-based care-as-usual group) improved significantly more regarding their behavioral problems: both ADHD symptoms and oppositional defiant disorder (ODD) symptoms significantly reduced directly after treatment. The frequency of the remaining behavioral problems of children in the BPTG@HOME group almost reached below the clinical range, as opposed to children in the waiting list group and the

home-based care-as-usual group. Internalizing problems of children in the BPTG@HOME condition significantly diminished compared to the waiting list group, but not compared to the home-based care-as-usual treatment group.

Furthermore, in chapter 3, significant improvements on several parenting parameters after BPTG@HOME were reported. Compared to the waiting list group, parents who had received BPTG@HOME showed significantly more reductions in using harsh and inconsistent discipline, experiencing the behavior of their child as less difficult to manage, and in finding the relationship with their child as less stressful. Also, they improved significantly more in their parenting efficacy (i.e., their perception of how capable they feel in their parental role). Compared to the home-based care-as-usual group, parents who received BPTG@HOME used significantly less harsh and inconsistent discipline strategies. When controlling for the frequency of home visits and length of the treatments, parents in the BPTG@HOME condition improved significantly more on overall parenting stress compared to the parents in the home-based care-as-usual treatment group. Moreover, parents learned to use more positive verbal discipline, praise and incentives, and to monitor their child. Apparently, both treatments showed positive results on all these parenting parameters, but BPTG@HOME achieved these positive results in less time and by using fewer home visits.

Six months after treatment parents who received BPTG@HOME experienced the behavioral problems of their child as less troublesome than parents who had received home-based care-as-usual treatment, although the frequency of the behaviors was not significantly reduced compared to immediately after treatment (chapter 2). Also, parenting self-efficacy did not change between post-treatment and follow-up (chapter 3).

For clinicians these findings can be of importance. Up to now, there were no studies that could guide clinicians in their treatment choice for school-aged children with ADHD who were not sufficiently benefitting from evidence-based first-line treatments. This study indicated that home-based behavioral parent training is an effective treatment for this group. Children showed behavioral improvements and parents improved in their parenting. Also, the treatment drop-out compared to what was reported in studies involving clinic-based behavioral parent training (Chacko et al., 2016) was low. Noteworthy, it is unlikely that the effects of BPTG@HOME were only caused by providing treatment at home, as BPTG@HOME was also more effective in reducing

children's behavioral problems and parental harsh and inconsistent discipline strategies than the home-based care-as-usual treatment. Apparently, home-based parent training for this population does not have to be lengthy or involve a large number of home visits. Rather, the type of therapist and the content of the treatment (for example, using live modeling, video-feedback during practicing with the child, and individual tailored treatment) may be important factors for its effectiveness.

The mediating role of parenting parameters on child outcome

Our mediation analyses, described in chapter 4, showed that reductions in harsh and inconsistent parental discipline and parents' stress concerning the manageability of the behavioral problems of their child mediated improvements of children's behavioral problems in the BPTG@HOME group. Furthermore, improving the confidence of parents in their parenting role had a positive effect on the severity of children's ODD symptoms after treatment. In contrast, that parents experienced the relationship with the child as less stressful did not mediate improvements in children's behaviors.

For clinicians it may be important to not only implement the well-known effective behavioral management techniques that teach parents how to deal with behavioral problems of their child and become less harsh and inconsistent in their parenting; for those parents who had to deal with persistent disruptive behaviors of their child despite multiple treatment efforts, investing in parents' feelings of parenting self-efficacy may be important as well. Although we cannot be sure which treatment element was associated with what specific improvement, we can hypothesize that implementation of the module Following (which focuses on establishing a positive parent-child relationship), and cognitive interventions directed at altering negative cognitions of the parents' parenting capacities were beneficial in improving parenting self-efficacy. Also, the tailored treatment modules and home visits that take parents' learning speed and confidence into account could have contributed to this effect.

The suitability of parent-rated questionnaires for ADHD in clinical practice

In chapter 5 we reported on the agreement between parent reports of ADHD symptoms (using the Swanson, Nolan, and Pelham Questionnaire-IV; SNAP-IV [Swanson, 1992]) and clinical judgement (using the semi-structured parent interview Parent Interview for Child

Symptoms-IV, PICS-IV [Schachar, Ickowicz, & Wachsmuth, 1996]). We found moderate levels of agreement for severity of overall ADHD and hyperactive-impulsive symptoms, but no significant agreement for inattentive symptoms. On individual symptom level and with regard to the presence/absence of the diagnosis ADHD agreement was poor.

From a clinical perspective, our findings suggest that parent-rated questionnaires are not suitable for establishing an ADHD diagnosis, as parents are not able to discriminate between the presence or absence of ADHD. Clinical interviews that not only focus on the frequency of ADHD symptoms, but also on impairment and age-appropriateness of behaviors are preferred over parent-rated questionnaires. However, parent-rated questionnaires can be used to rate the severity of ADHD symptoms, which can be helpful in evaluating treatment effect. Clinicians have to keep in mind that parents are particularly accurate in rating concrete and observable hyperactive and impulsive behaviors, but less competent in recognizing the internal, cognitive inattentive symptoms. For the evaluation of inattention, forgetfulness and planning problems (i.e., the attention deficit symptoms as described in the DSM-V [American Psychiatric Association, 2013]) and other cognitive functions, clinicians should weigh the performance of the child in the context that parents describe, ask teachers or perform a neuropsychological assessment.

Scientific relevance: considerations

Tension between internal and external validity in randomized controlled trials

In health sciences, a randomized controlled trial is an important tool to test the effect of a treatment. Mostly, researchers start to investigate the efficacy of a newly developed treatment in an explanatory trial (under ideal controlled conditions). Ideally, this is followed by an investigation of the effectiveness of a treatment in routine clinical practice, using a pragmatic trial. Both types of trials are required to establish the relevance of a given treatment, the former aiming to test efficacy of a treatments under strict conditions, while the latter in the effectiveness of the treatment in real-life, everyday care. Particularly, pragmatic trials are of importance for clinicians and policy makers, as they allow for better generalizability and clinical implementation of study

results. However, pragmatic trials are underrepresented in the literature (Patsopoulos, 2011). In other words, in the scientific field the primary focus is to establish internal validity, while only infrequently followed by studies focusing on external validity.

It is not always feasible to create the controlled conditions required for an explanatory trial. In particular, when treatments are developed for specific target groups (e.g., non-responders to treatments, patients with rare disorders, or severe disorders requiring immediate treatment), the demands placed on explanatory trials, such as strict inclusion criteria, randomization to inactive control groups, and sufficiently large sample sizes, can be hindering. However, the objective to develop effective treatments remains, and even more so given the - often difficult to treat patients. While explanatory trials for these specific groups could be developed using expensive, multi-center studies, it is more efficient and less costly to start with pragmatic trials that relatively quickly determine the clinical relevance of a treatment. When successful, follow-up explanatory trials can be performed to explore the efficacy of the treatment, knowing that the investment is worthwhile and socially responsible, given the already established clinical relevance of the treatment.

The current study followed the approach of first performing a pragmatic trial before an explanatory trial had been carried out. Our target group is hard to include in an explanatory trial: the number of children is small and heterogeneous in terms of reasons of not benefitting from routine clinical treatment, making it hard to reach the required sample size of a homogeneous group for an explanatory trial. The severity of behavioral problems experienced by the parents is high, however, and therefore requiring short-term, effective treatment. The use of a pragmatic trial in our study offered the benefit that most children in the target group could be admitted to the trial by not requiring strict inclusion criteria, thus allowing to establish the ecological validity of the treatment for a wide range of children. At the same time, this study illustrates that a pragmatic trial does not have to be pragmatic at all points compared to an explanatory trial. In particular, the current study attempted to control for many of the biases that can arise in pragmatic trials, by applying randomization, having an active and inactive control group, blinding of participants where possible, and maintaining treatment integrity of BPTG@HOME. Thus, while the trial was pragmatic regarding its inclusion criteria given the broad target group (also recommended by Parmar, Sydes, & Morris, 2016), it adhered to many of the

requirements put forth for explanatory trials. Consequently, despite the lack of insight on efficacy, the current study pointed to effectiveness of a new treatment modality for this hard-to-treat group of children, equipping the clinician with a new treatment option for a group for which routine treatments had been ineffective.

Challenges in a study with non-responders

The target group of the current study were school-aged children with ADHD and disruptive behaviors *who had not responded sufficiently to routine clinical treatment*; we refer to this group as non-responders. It is imaginable that families with a “difficult-to-treat” child and various complex family characteristics, are under great pressure, especially after disappointing prior treatments. This leads to some unique challenges when it comes to participation in a scientific study.

Based on our experience, the motivation of these families to participate in a trial is limited, due to various reasons. For example, parents may fear to be randomized to a non-active control group (i.e., not receiving treatment), divorced parents may differ in their willingness to participate, or parents may be reluctant to perform study assessments. In the current study, ten percent of the parents who were invited to participate in the study did not want to participate or did not give informed consent. As the study progressed, the percentages of absent or incomplete data rose from fifteen to twenty-five percent (see flow chart chapter 2). Such numbers could give rise to biases.

To increase the participation in studies, it is advisable to reduce the length of the waiting list periods, to allow other interventions during the waiting period, and to lower the probability to be randomized to the waiting list. The current study incorporated this principle by offering a chance of only 1:3 of being allocated to the waiting list, which only lasted four-months. Participation can also be increased by adapting the study design. For example, reducing the number of assessment moments, limiting the amount of outcome measures, and filling in questionnaires at home or during treatment, can all contribute to higher and prolonged participation rates. In the current study this was implemented by having only three assessment moments, using a limited number of primary outcome measures, and allowing respondents to fill in questionnaires at home. Employing statistical methods suited to accommodate potential biases arising from small samples and missing data, which are likely to remain even after the recommendations above, is

also recommended. The current study followed the intent-to-treat paradigm. Moreover, if this is complemented with statistical methods that can account for multiple measurements and the presence of missing data, such as the mixed models (Chapters 2 and 3; Bliese, Maltarich, & Hendricks, 2018) and bootstrap (Chapter 4; Enders, 2002) approaches used here, statistical power is further increased.

Beyond difficulties to recruit families willing to participate from the target group of non-responders to previous treatments, a second challenge lies in the nature of the target group. Given the focus on non-responders to prior treatments, one of the inclusion criteria of the current study was that prior treatments had to be finished unsuccessfully. Naturally, this reduces the amount of potential participants (not every child with ADHD can participate). One solution to address this issue is to create a multi-center study. Such a study requires, however, that multiple centers use the same approaches as prior treatments (i.e., for the current study pharmacotherapy or clinic-based behavioral parent training) to have a uniform participant group. Given the various treatment options used in clinical practice (not always in line with clinical guidelines), ensuring that similar prior treatments were used across sites was (and in general is) not an easily fulfilled requirement. Therefore, in the current study 82% of participants came from one center, while other centers had more difficulty to find children who fulfilled the study criteria. Thus, multi-center studies are not always a panacea to increase population size, in particular when focusing on subgroups which have to fulfill specific criteria concerning prior treatments to be considered for inclusion. When engaging in a multi-center study, we recommend to ensure that committed site investigators are available at every center and to ask consent for inclusion in the study early in the treatment process. We believe that having an investigator (and preferably multiple) involved in the study who also fulfills clinical duties is an added benefit in that respect.

A third challenge in studies with difficult to treat families may be to perform the manualized treatment to the included families, as crisis situations may interfere with the planned treatment. In the treatment that was subject of our study, every session started with the clinician probing for current difficulties in the family not directly related to the treatment goals. Moreover, the treatment protocol allowed to postpone a scheduled session should an acute crisis demand this.

Considering suitable instruments

Studies assessing the impact of behavioral parent training on ADHD and disruptive behaviors have employed a variety of measurement instruments for child and parenting outcomes. With respect to child behavioral outcomes, measurement of the severity of behavioral problems is often of interest to evaluate treatment effectiveness. The current study relied on parent-report measures (i.e., the Eyberg Child Behavior Inventory [ECBI; Eyberg & Pincus, 1999] and Swanson, Nolan, and Pelham Questionnaire [SNAP-IV; Swanson, 1992]). Recent meta-analyses recommend, to control for potential bias in parent-report measures, the use of raters who are blinded to treatment allocation such as clinicians and teachers (Daley et al., 2014; Sonuga-Barke et al., 2013). While such blinded raters may be preferred if the objective of the treatment is generalizability across situations, this approach may be too strict when the goal of the treatment is to improve behavioral problems in a specific context (i.e., at home) that falls outside the scope of the blinded raters. The current study showed that when it comes to measuring the severity of behavioral problems, there is sufficient agreement between parent-report and clinician-report measures, especially when the behaviors are concrete and observable (Chapter 5). This limits the potential bias of these measures. Thus, future studies can reliably use parent-report measures in a situation where blind raters are hard to apply; especially when parents are blinded for randomization outcome.

This study also provided insights in the validity of parent-report outcome measures. First, discussions on the suitability of parent-report compared to observational measures have arisen. A recent meta-analysis showed a small, but significant association between the two types of measures (Hendriks, Van der Giessen, Stams, & Overbeek, 2018). While the current study relied on parent-report measures, the inclusion of a second active treatment arm relying on the same parent-report measures reduced bias concerns, because any systematic bias arising due to parent-report measures should be of similar size in the active treatment groups. Results indicated that BPTG@HOME was not only more effective than the waiting list control group, but also, regarding some measures, compared to the other active treatment. This suggests that the bias of using parent-report measures is not of such an extent that it explained the findings in the current study entirely.

Second, the current study measured parenting stress (Chapters 3, 4, and 5) using the Parenting Stress Index-Short Form (PSI; Abidin, 1995). While prior studies found diverging results on the influence of parenting stress in relation to behavioral parent-training (Van den Hoofdakker et al., 2007; Chacko et al., 2009), the current study showed that these diverging findings probably occur due to only reporting outcomes on the aggregate full scale, instead of reporting the outcomes on the subscales (Chapter 3). In particular, in the case where various subscales show results in opposite directions, these might be averaged out when aggregating to the full scale, obscuring the impact of the domains that were assessed with the subscales. This becomes particularly salient in our study that focused on the mediating effects of improvements in parenting on changes in children's behaviors (chapter 4). While changes on some subscales of the PSI mediate improvements in the child's behavior, scores on other PSI subscales might have decreased more simultaneously with the child's behavior. Not accounting for differences in treatment effects on subscales of the PSI might lead to incorrectly conclude a treatment is ineffective. Based on these results, we therefore recommend using the various subscales, instead of using the aggregate full scale.

Limitations

This study had some limitations that should be acknowledged. First, the modest sample size was a concern, especially as we had two control groups. Fortunately, post-power analyses performed on the outcome measures showed sufficient power for the effectiveness analyses (chapter 2 and 3). However, the modest sample size made it impossible to control for various factors that could have been of influence on treatment effects (for example use of, and changes in medication use) or to investigate moderators of treatment outcome. Second, we could not check whether we were successful in keeping parents unaware of the differences between the two home-based treatments (BPTG@HOME and home-based care-as-usual treatment). Unfortunately, we therefore are not completely sure that the reported effectiveness of BPTG@HOME was due to a treatment effect, or to an expectation bias. Third, both treatments were performed by different types of therapists. Psychologists who were experienced in offering clinic-based

behavioral parent training, but who had no experience in behavioral home-based treatments delivered BPTG@HOME. Social workers, who had many years of experience with eclectic, non-manualized home-based treatments offered the home-based care-as-usual treatment. Therefore, it cannot be ruled out that the differences between the therapists have played a role in our findings.

Future directions

An important aim for future research should be to investigate the timing of a home-based parent training for school-aged children with ADHD. In this study, it was used as a treatment after clinic-based behavioral parent training was shown insufficient. However, perhaps it is more efficient or effective to offer home-based behavioral parent training (for certain subgroups) as a first-line non-pharmacological treatment. Such a comparison has not been done for school-aged children with ADHD. From studies with preschool children with ADHD, there is some indication that home-based parent training is effective in treating children with ADHD when offered as first-line treatment. However, it appears to be no more beneficial than clinic-based parent training (Sonuga-Barke et al., 2018; Lange et al., 2018). Therefore, it is worthwhile to compare the effectiveness of clinic-based and home-based behavioral parent training as first-line non-pharmacological treatment. Furthermore, it is important to investigate whether home-based parent training was effective because parents received repetition and coaching in their parenting (which can also be established by repeating previous received clinic-based parent training), or whether the fact that the treatment took place at home - with the therapeutic benefits outlined before - was a crucial factor in its success as a second-line treatment. In addition to this, it is worthwhile to investigate the effective elements of the treatment. The current study showed that BPTG@HOME is more effective than the home-based care-as-usual treatment. Therefore, it would be interesting to investigate how the various treatment modules of BPTG@HOME contribute to the effectiveness of the treatment, i.e., the use of video-feedback, and the implementation of cognitive interventions to change negative thoughts of the parents about their child and their self-efficacy as a parent.

Another direction for future research could be to investigate the characteristics of non-responders, and whether these characteristics are predictors and/or moderators for treatment effectiveness of home-based behavioral parent training. Due to the small sample size of this study, we could not perform such comparisons. However, it is important to understand which child and family characteristics predict disappointing results of pharmacotherapy and clinic-based parent training, and which of these subgroups particularly benefit from a second-line home-based parent training and which subgroups should be offered another type of treatment.

At last, currently there are just a few mediation studies that investigated whether changes in parenting mediated changes in child behavior after behavioral parent training. All of these studies had limitations in terms of statistical power (Katzmann et al., 2017; VanderStoep et al., 2017; Heath, Curtis, Fan, & McPherson, 2015; Chronis-Tuscano et al., 2011). Therefore, it would be worthwhile to conduct a meta-analysis including multiple mediation studies in order to establish clearer evidence regarding the effective elements contributing to the effectiveness of the treatment.

Overall conclusion

Up to now, there had been no studies that could guide clinicians in their treatment choice for school-aged children with ADHD who were not sufficiently benefitting from evidence-based first-line treatments (i.e., pharmacotherapy and clinic-based parent training). The current study showed, from a clinical perspective, the effectiveness of a newly developed home-based behavioral parent training (BPTG@HOME) for this population. Children showed behavioral improvements and parents improved in their parenting.

Furthermore, from a scientific perspective, this study is a good example of a study design for a population that is difficult to include in a trial. It showed how a pragmatic trial can be designed in such a way that biases and inclusion barriers for a difficult to treat or reach population can be reduced as much as possible. Our choices can be used in future studies with similar population characteristics.

References

- Abidin, R.R. (1995). *Parenting Stress Index – Professional manual* (3rd edn.). Psychological Assessment Resources: Odessa.
- Achenbach, T.M. (1991). *Interactive guide for the 1991 CBCL 14-18, YSR, and TRF Profiles*. Burlington: University of Vermont, Department of Psychiatry.
- Alda, J. A., & Serrano-Troncoso, E. (2013). Attention-deficit hyperactivity disorder: agreement between clinical impression and the SNAP-IV screening tool. *Actas Espanolas de Psiquiatria*, 41(2), 76-83.
- Altman, D.G. (1994). *Practical statistics for medical research*. London: Chapman and Hall.
- American Academy of Pediatrics. (2011). ADHD: clinical practice guideline for the diagnosis, evaluation, and treatment of attention-deficit/hyperactivity disorder in children and adolescents. *Pediatrics*, 128, 1007–1022.
- American Psychiatric Association. (2000). *Diagnostic and statistical manual of mental disorders* (4th ed.). Washington DC.
- American Psychiatric Association. (2013). *The diagnostic and statistical manual of mental disorders* (5th ed.). Washington DC.
- Anastopoulos, A.D., Guevremont, D.C., Shelton, T.L., & DuPaul, G. (1992). Parenting stress among families of children with attention deficit hyperactivity disorder. *Journal of Abnormal Child Psychology*, 20 (5), 503- 520.
- Anastopoulos, A.D., Shelton, T.L., DuPaul, G.J., & Guevremont, D.C. (1993). Parent training for attention-deficit hyperactivity disorder: its impact on parent functioning. *Journal of Abnormal Child Psychology*, 21(5), 581-596.
- Assenany, A.E., & McIntosh, D.E. (2002). Negative treatment outcomes of behavioral parent training programs. *Psychology in the Schools*, 39(2), 209-219.
- Banaschewski, T., Coghill, D., Santosh, P., Zuddas, A., Asherson, P., Buitelaar, J., ..., Sergeant, J. (2006). Long-acting medications for the hyperkinetic disorders. *European child & adolescent psychiatry*, 15(8), 476-495.
- Barbarese, W. J., Katusic, S. K., Colligan, R. C., Weaver, A. L., & Jacobsen, S. J. (2007). Long-term school outcomes for children with attention-deficit/hyperactivity disorder: a population-based perspective. *Journal of Developmental & Behavioral Pediatrics*, 28(4), 265-273.
- Barkley, R.A. (1987). *Defiant children: A clinician's manual for parent training*. New York: Guilford Press.
- Barkley, R.A., & Murphy, K.R. (1998). *Attention deficit hyperactivity disorder: A clinical workbook* (3rd ed.). New York: Guilford Press.
- Baron, R. M., & Kenny, D. A. (1986). The moderator-mediator variable distinction in social psychological research: Conceptual, strategic, and statistical considerations. *Journal of Personality and Social Psychology*, 51, 1173–1182.
- Beck, A.T., Steer, R.A., & Brown, G.K. (1996). *Beck Depression Inventory – Second Edition manual*. San Antonio, TX: The Psychological Corporation.
- Bendiksen, B., Svensson, E., Aase, H., Reichborn-Kjennerud, T., Friis, S., Myhre, A.M., & Zeiner, P. (2014). Co-occurrence of ODD and CD in preschool children with symptoms of ADHD. *Journal of Attention Disorders*, 21(9), 741-752.
- Berger, I., Dor, T., Nevo, Y., & Goldzweig, G. (2008). Attitudes toward attention-deficit hyperactivity disorder (ADHD) treatment: parent and children's perspectives. *Journal of Child Neurology*, 23(9), 1036-1042.
- Biederman, J., Newcorn, J., & Sprich, S. (1991). Comorbidity of attention deficit hyperactivity disorder. *American Journal of Psychiatry*, 148(5), 564-577.
- Bird, H. R., Gould, M. S., & Staghezza, B. (1992). Aggregating data from multiple informants in child psychiatry epidemiological research. *Journal of the American Academy of Child & Adolescent Psychiatry*, 31(1), 78-85.
- Bliese, P. D., Maltarich, M. A., & Hendricks, J. L. (2018). Back to basics with mixed-effects models: nine take-away points. *Journal of Business and Psychology*, 33, 1–23.
- Blood, E. A., Cabral, H., Heeren, T., & Cheng, D. M. (2010). Performance of mixed effects models in the analysis of mediated longitudinal data. *BMC Medical Research Methodology*, 10(1), 16.
- Boggs, S. R., Eyberg, S. M., Edwards, D. L., Rayfield, A., Jacobs, J., Bagner, D., & Hood, K. K. (2004). Outcomes of parent-child interaction therapy: A comparison of treatment completers and study dropouts one to three years later. *Child & Family Behavior Therapy*, 26(4), 1–22.
- Briscoe-Smith, A. M., & Hinshaw, S. P. (2006). Linkages between child abuse and attention-deficit/hyperactivity disorder in girls: Behavioral and social correlates. *Child Abuse & Neglect*, 30(11), 1239-1255.
- Bussing, R., Fernandez, M., Harwood, M., Hou, W., Garvan, C. W., Eyberg, S. M., & Swanson, J. M. (2008). Parent and teacher SNAP-IV ratings of attention deficit hyperactivity disorder symptoms: psychometric properties and normative ratings from a school district sample. *Assessment*, 15(3), 317-328.
- Bussing, R., Mason, D. M., Bell, L., Porter, P., & Garvan, C. (2010). Adolescent outcomes of childhood

- attention-deficit/hyperactivity disorder in a diverse community sample. *Journal of the American Academy of Child & Adolescent Psychiatry*, 49(6), 595-605.
- CADDRA Canadian ADHD Resource Alliance. (2011). *Canadian ADHD practice guidelines 2011*. Retrieved from <http://www.caddra.ca/practice-guidelines/download>.
- Catalá-López, F., Hutton, B., Núñez-Beltrán, A., Page, M.J., Ridao, M., Macías Saint-Gerons, ... Moher, D. (2017). The pharmacological and non-pharmacological treatment of attention deficit hyperactivity disorder in children and adolescents: a systematic review with network meta-analyses of randomized trials. *PLoS One*, 12(7).
- Cengel-Kultur, E., Cuhadaroglu-Cetin, F., & Gokler, B. (2007). Demographic and clinical features of child abuse and neglect cases. *Turkish Journal of Pediatrics*, 49(3), 256.
- Centraal Bureau voor de Statistiek (2015). *Standaard onderwijsindeling 2006*, editie 2014/'15. Den Haag/Heerlen.
- Chacko, A., Jensen, S., Lowry, L.S., Cornwell, M., Chimiklis, A., Chan, E., ... Pulgarin, B. (2016). Engagement in behavioral parent training: Review of the literature and implications for practice. *Clinical Child and Family Psychology Review*, 19, 204-215.
- Chacko, A., Wymbs, B.T., Arnold, F.W., Pelham, W.E., Swanger-Gagne, M., Girio, E.L., ... O'Connor, B. (2009). Enhancing traditional behavioral parent training for single-mothers of children with ADHD. *Journal of Clinical Child and Adolescent Psychology*, 38, 206-218.
- Chronis, A. M., Jones, H. A., & Raggi, V. L. (2006). Evidence-based psychosocial treatments for children and adolescents with attention-deficit/hyperactivity disorder. *Clinical Psychology Review*, 26, 486-502.
- Chronis-Tuscano, A., Clarke, T. L., O'Brien, K. A., Raggi, V. L., Diaz, Y., Mintz, A. D., ... Lewinsohn, P. (2013). Development and preliminary evaluation of an integrated treatment targeting parenting and depressive symptoms in mothers of children with attention-deficit/hyperactivity disorder. *Journal of Consulting and Clinical Psychology*, 81(5), 918-925.
- Chronis-Tuscano, A., O'Brien, K.A., Johnston, C. Jones, H.A., Clarke, T.A., Raggi, V.L., ... Seymour, K.E. (2011). The relation between maternal ADHD symptoms & improvement in child behavior following brief behavioral parent training is mediated by change in negative parenting. *Journal of Abnormal Child Psychology*, 39(7), 1047-1057.
- Coates, J., Taylor, J. A., & Sayal, K. (2015). Parenting interventions for ADHD: A systematic literature review and meta-analysis. *Journal of Attention Disorders*, 19(10), 831-843.
- Cohen, J. (1960). A coefficient of agreement for nominal scales. *Educational and Psychological Measurement*, 20, 121-153.
- Coleman, P. K., & Karraker, K. H. (2000). Parenting self-efficacy among mothers of school-aged children: conceptualization, measurement, and correlates. *Family Relations*, 49, 13-24.
- Conners, C. K. (2001). *Conners' Rating Scales: Revised technical manual*. Toronto, Ontario, Canada: Multi-Health Systems.
- Cook, R. D., & Weisberg, S. (1982). *Residuals and influence in regression*. New York: Chapman and Hall.
- Corkum, P. V., McKinnon, M. M., & Mullane, J. C. (2005). The effect of involving classroom teachers in a parent training program for families of children with ADHD. *Child & Family Behavior Therapy*, 27(4), 29-49.
- Cortese, S., Adamo, N., Del Giovane, C., Mohr-Jensen, C., Hayes, A.J., Carucci, S., ... Cipriani, A. (2018). Comparative efficacy and tolerability of medications for attention-deficit hyperactivity disorder in children, adolescents, and adults: a systematic review and network meta-analysis. *Lancet Psychiatry*, 5, 727-38.
- Cortese, S., Ferrin, M., Brandeis, D., Holtmann, M., Aggensteiner, P., Daley, D., ... Sonuga-Barke, E. J. (2016). Neurofeedback for attention-deficit/hyperactivity disorder: meta-analysis of clinical and neuropsychological outcomes from randomized controlled trials. *Journal of the American Academy of Child & Adolescent Psychiatry*, 55(6), 444-455.
- Craig, F., Operto, F.F., De Giacomo, A., Margari, L., Frolli, A., Conson, ... Margari, F. (2016). Parenting stress among parents of children with neurodevelopmental disorders. *Psychiatry Research*, 242, 121-129.
- Cunningham, C. E. (2007). A family-centered approach to planning and measuring the outcome of interventions for children with attention-deficit/hyperactivity disorder. *Journal of Pediatric Psychology*, 32(6), 676-694.
- Cunningham, C.E., Boyle, M., Offord, D., Racine, Y., Hundert, J., & Secord, M. (2000). Triministry study: Correlates of school-based parent course utilization. *Journal of Consulting and Clinical Psychology*, 68, 928-933.
- Cunningham, C. E., Bremner, R. B., & Boyle, M. (1995). Large group community-based parenting programs for families of preschoolers at risk for disruptive parent behavior disorders: Utilization, cost effectiveness, and outcome. *Journal of Child Psychology and Psychiatry*, 36, 1141-1159.

- Daley, D., Van Der Oord, S., Ferrin, M., Cortese, S., Danckaerts, M., Doepfner, M., ... Sonuga-Barke, E. J. (2018). Practitioner review: current best practice in the use of parent training and other behavioural interventions in the treatment of children and adolescents with attention deficit hyperactivity disorder. *Journal of Child Psychology and Psychiatry*, 59(9), 932–947.
- Daley, D., Van der Oord, S., Ferrin, M., Danckaerts, M., Doepfner, M., Cortese, S., & Sonuga-Barke, E. J. (European ADHD Guidelines Group). (2014). Behavioral interventions in attention-deficit/hyperactivity disorder: A meta-analysis of randomized controlled trials across multiple outcome domains. *Journal of the American Academy of Child & Adolescent Psychiatry*, 53(8), 835–847.
- Danckaerts, M., Sonuga-Barke, E. J., Banaschewski, T., Buitelaar, J., Döpfner, M., Hollis, C., ... Taylor, E. (2010). The quality of life of children with attention deficit/hyperactivity disorder: a systematic review. *European Child & Adolescent Psychiatry*, 19(2), 83–105.
- Deault, L. C. (2010). A systematic review of parenting in relation to the development of comorbidities and functional impairments in children with attention-deficit/hyperactivity disorder (ADHD). *Child Psychiatry & Human Development*, 41(2), 168–192.
- De Los Reyes, A., & Kazdin, A. E. (2005). Informant discrepancies in the assessment of childhood psychopathology: a critical review, theoretical framework, and recommendations for further study. *Psychological Bulletin*, 131(4), 483.
- Dose, C., Hautmann, C., Buerger, M., Schuermann, S., Woitecki, K., & Doepfner, M. (2017). Telephone-assisted self-help for parents of children with attention-deficit/hyperactivity disorder who have residual functional impairment despite methylphenidate treatment: a randomized controlled trial. *Journal of Child Psychology and Psychiatry*, 58(6), 682–690.
- DuPaul, G. J., McGoe, K. E., Eckert, T. L., & VanBrakle, J. (2001). Preschool children with attention-deficit/hyperactivity disorder: Impairments in behavioral, social, and school functioning. *Journal of the American Academy of Child & Adolescent Psychiatry*, 40(5), 508–515.
- Eisenstadt, T. H., Eyberg, S., McNeil, C. B., Newcomb, K., & Funderburk, B. (1993). Parent-child interaction therapy with behavior problem children: Relative effectiveness of two stages and overall treatment outcome. *Journal of Clinical Child Psychology*, 22(1), 42–51.
- Enders, C. K. (2002). Applying the bollen-stine bootstrap for goodness-of-fit measures to structural equation models with missing data. *Multivariate Behavioral Research*, 37(3), 359–377.
- Erskine, H. E., Ferrari, A. J., Nelson, P., Polanczyk, G. V., Flaxman, A. D., Vos, T., ... Scott, J. G. (2013). Research Review: Epidemiological modelling of attention-deficit/hyperactivity disorder and conduct disorder for the Global Burden of Disease Study 2010. *Journal of Child Psychology and Psychiatry*, 54(12), 1263–1274.
- Erskine, H. E., Ferrari, A. J., Polanczyk, G. V., Moffitt, T. E., Murray, C. J. L., Vos, T., ... Scott, J. G. (2014). The global burden of conduct disorder and attention-deficit/hyperactivity disorder in 2010. *Journal of Child Psychology and Psychiatry*, 55, 328–336.
- Erskine, H. E., Norman, R. E., Ferrari, A. J., Chan, G. C., Copeland, W. E., Whiteford, H. A., & Scott, J. G. (2016). Long-term outcomes of attention-deficit/hyperactivity disorder and conduct disorder: a systematic review and meta-analysis. *Journal of the American Academy of Child & Adolescent Psychiatry*, 55(10), 841–850.
- Ertan, C., Özcan, Ö. Ö., & Pepele, M. S. (2012). Paediatric trauma patients and attention deficit hyperactivity disorder: correlation and significance. *Emergency Medicine Journal*, 29(11), 911–914.
- Evans, S., Ling, M., Hill, B., Rinehart, N., Austin, D., & Sciberras, E. (2018). Systematic review of meditation-based interventions for children with ADHD. *European Child & Adolescent Psychiatry*, 27(1), 9–27.
- Evans, S. W., Owens, J. S., Wymbs, B. T., & Ray, A. R. (2018). Evidence-based psychosocial treatments for children and adolescents with attention deficit/hyperactivity disorder. *Journal of Clinical Child & Adolescent Psychology*, 47(2), 157–198.
- Eyberg, S.M., & Funderburk, B.W. (2011). *Parent-Child Interaction Therapy protocol*. Gainesville, FL: PCIT International.
- Eyberg, S.M., & Pincus, D.B. (1999). *Eyberg child behavior inventory and Sutter-Eyberg behavior inventory-revised: Professional manual*. Odessa: Psychological Assessment Resources.
- Fabiano, G. A., Pelham, W. E., Cunningham, C. E., Yu, J., Gangloff, B., Buck, M., ... Gera, S. (2012). A waitlist-controlled trial of behavioral parent training for fathers of children with ADHD. *Journal of Clinical Child & Adolescent Psychology*, 41(3), 337–345.
- Faraone, S. V., Asherson, P., Banaschewski, T., Biederman, J., Buitelaar, J. K., Ramos-Quiroga, J. A., & Franke, B. (2015). Attention-deficit/hyperactivity disorder. *Nature Reviews: Disease Primers*, 1, 15020.
- Faraone, S. V., Biederman, J., & Mick, E. (2006). The age-dependent decline of attention deficit hyperactivity disorder: a meta-analysis of follow-up studies. *Psychological Medicine*, 36, 159–65.
- Forehand, R., Lafko, N., Parent, J., & Burt, K.B. (2014). Is parenting the mediator of change in behavioral parent

- training for externalizing problems of youth? *Clinical Psychology Review*, 34, 608–619.
- Forehand, R.L., & McMahon, R.J. (1981). *Helping the noncompliant child: a clinician's guide to parent training*. New York: Guilford Press.
- Fridman, M., Banaschewski, T., Sikirica, V., Quintero, J., Erder, M.H., & Chen, K.S. (2017). Factors associated with caregiver burden among pharmacotherapy-treated children/adolescents with ADHD in the caregiver perspective on pediatric ADHD survey in Europe. *Neuropsychiatric Disease and Treatment*, 13, 373–386.
- Funderburk, B. W., Eyberg, S. M., Rich, B. A., & Behar, L. (2003). Further psychometric evaluation of the Eyberg and Behar rating scales for parents and teachers of preschoolers. *Early Education and Development*, 14(1), 67–82.
- Gibaud-Wallston, J., & Wandersmann, L. P. (1978). *Development and utility of the Parenting Sense of Competence Scale*. John F. Kennedy center for research on education and human development.
- Graham, J., Banaschewski, T., Buitelaar, J., Coghill, D., Danckaerts, M., Dittmann, R. W., ... Hulpke-Wette, M. (2011). European guidelines on managing adverse effects of medication for ADHD. *European Child & Adolescent Psychiatry*, 20(1), 17–37.
- Graziano, P. A., McNamara, J. P., Geffken, G. R., & Reid, A. (2011). Severity of children's ADHD symptoms and parenting stress: A multiple mediation model of self-regulation. *Journal of Abnormal Child Psychology*, 39(7), 1073.
- Haack, L.M., Villodas, M., McBurnett, K., Hinshaw, S., & Pfiffner, L.J. (2017). Parenting as a mechanism of change in psychosocial treatment for youth with ADHD, predominantly inattentive presentation. *Journal of Abnormal Child Psychology*, 45, 841–855.
- Hair, J.F., Black, W.C., Babin, B.J., & Anderson, R.E. (2010), *Multivariate Data Analysis* (7th ed.). Upper Saddle River, NY: Pearson Prentice Hall.
- Haskett, M. E., Ahern, L. S., Ward, C. S., & Allaire, J. C. (2006). Factor structure and validity of the parenting stress index-short form. *Journal of Clinical Child & Adolescent Psychology*, 35(2), 302–312.
- Heath, C. L., Curtis, D. F., Fan, W., & McPherson, R. (2015). The association between parenting stress, parenting self-efficacy, and the clinical significance of child ADHD symptom change following behavior therapy. *Child Psychiatry & Human Development*, 46(1), 118–129.
- Hendriks, A. M., Van der Giessen, D., Stams, G. J. J. M., & Overbeek, G. (2018). The association between parent-reported and observed parenting: a multi-level meta-analysis. *Psychological Assessment*, 30(5), 621–633.
- Hinshaw, S. P. (2002). Preadolescent girls with attention-deficit/hyperactivity disorder: I. Background characteristics, comorbidity, cognitive and social functioning, and parenting practices. *Journal of Consulting and Clinical Psychology*, 70(5), 1086.
- Hoath, F. E., & Sanders, M. R. (2002). A feasibility study of enhanced group triple P - positive parenting program for parents of children with attention-deficit/hyperactivity disorder. *Behaviour Change*, 19(4), 191–206.
- Holmberg, K., & Hjern, A. (2008). Bullying and attention-deficit–hyperactivity disorder in 10-year-olds in a Swedish community. *Developmental Medicine & Child Neurology*, 50(2), 134–138.
- Ickowicz, A., Schachar, R. J., Sugarman, R., Chen, S. X., Millette, C., & Cook, L. (2006). The parent interview for child symptoms: a situation-specific clinical research interview for attention-deficit hyperactivity and related disorders. *The Canadian Journal of Psychiatry*, 51(5), 325–328.
- Jensen, P.S., Hinshaw, S.P., Swanson, J.M., Greenhill, L.L., Conners, C.K., & Arnold, L.E. (2001). Findings from the NIMH multimodal treatment study of ADHD (MTA): Implications and applications for primary care providers. *Journal of Developmental and Behavioral Pediatrics*, 22, 60–73.
- Jensen, C. M., & Steinhausen, H. C. (2015). Comorbid mental disorders in children and adolescents with attention-deficit/hyperactivity disorder in a large nationwide study. *ADHD Attention Deficit and Hyperactivity Disorders*, 7(1), 27–38.
- Jensen, A. L., & Weisz, J. R. (2002). Assessing match and mismatch between practitioner-generated and standardized interview-generated diagnoses for clinic-referred children and adolescents. *Journal of Consulting and Clinical Psychology*, 70(1), 158.
- Johnston, C., & Mash, E. J. (1989). A measure of parenting satisfaction and efficacy. *Journal of Clinical Child Psychology*, 18(2), 167–175.
- Johnston, C., & Mash, E. J. (2001). Families of children with attention-deficit/hyperactivity disorder: review and recommendations for future research. *Clinical Child and Family Psychology Review*, 4(3), 183–207.
- Kaiser, N.M., McBurnett, K., & Pfiffner, L.J. (2011). Child ADHD severity and positive and negative parenting as predictors of child social functioning: evaluation of three theoretical models. *Journal of Attention Disorders*, 15(3), 193–203.

- Kaminski, J.W., Valle, L.A., Filene, J.H., & Boyle, C.L. (2008). A meta-analytic review of components associated with parent training program effectiveness. *Journal of Abnormal Child Psychology*, 36, 567-589.
- Katzmann, J., Hautmann, C., Greimel, L., Imort, S., Pinior, J., Scholz, K., & Döpfner, M. (2017). Behavioral and nondirective guided self-help for parents of children with externalizing behavior: Mediating mechanisms in a head-to-head comparison. *Journal of Abnormal Child Psychology*, 45(4), 719-730.
- Keen, D., & Hadjikoumi, I. (2011). ADHD in children and adolescents. *BMJ Clinical Evidence*.
- Klein, J. B., Lavigne, J. V., & Seshadri, R. (2010). Clinician-assigned and parent-report questionnaire-derived child psychiatric diagnoses: correlates and consequences of disagreement. *American Journal of Orthopsychiatry*, 80(3), 375-385.
- Koerting, J., Smith, E., Knowles, M.M., Latter, S., Elsey, H., Mccann, D.C., . . . Sonuga-Barke, E.J. (2013). Barriers to, and facilitators of, parenting programmes for childhood behaviour problems: A qualitative synthesis of studies of parents' and professionals' perceptions. *European Child and Adolescent Psychiatry*, 22, 653-670.
- Kreidler, S. M., Muller, K. E., Grunwald, G. K., Ringham, B. M., Coker-Dukowitz, Z. T., Sakhadeo, U. R., ... Glueck, D. H. (2013). GLIMMPSE: online power computation for linear models with and without a baseline covariate. *Journal of Statistical Software*, 54(10).
- Lange, A. M., Daley, D., Frydenberg, M., Houmann, T., Kristensen, L. J., Rask, C., ... Thomsen, P. H. (2018). Parent training for preschool ADHD in routine, specialist care: a randomized controlled trial. *Journal of the American Academy of Child & Adolescent Psychiatry*, 57(8), 593-602.
- Lara, C., Fayyad, J., de Graaf, R., Kessler, R. C., Aguilar-Gaxiola, S., Angermeyer, M., ... Sampson, N. (2009). Childhood predictors of adult attention-deficit/hyperactivity disorder: results from the World Health Organization World Mental Health Survey Initiative. *Biological Psychiatry*, 65, 46-54.
- Le, H. H., Hodgkins, P., Postma, M. J., Kahle, J., Sikirica, V., Setyawan, J., ... Doshi, J. A. (2014). Economic impact of childhood/adolescent ADHD in a European setting: the Netherlands as a reference case. *European Child & Adolescent Psychiatry*, 23(7), 587-598.
- Leibson, C. L., Barbaresi, W. J., Ransom, J., Colligan, R. C., Kemner, J., Weaver, A. L., & Katusic, S. K. (2006). Emergency department use and costs for youth with attention-deficit/hyperactivity disorder: associations with stimulant treatment. *Ambulatory Pediatrics*, 6(1), 45-53.
- Leijten, P., Melendez-Torres, G. J., Gardner, F., Van Aar, J., Schulz, S., & Overbeek, G. (2018). Are Relationship Enhancement and Behavior Management "The Golden Couple" for Disruptive Child Behavior? Two Meta-analyses. *Child Development*, 89(6), 1970-1982.
- MacKinnon, D. P., Lockwood, C. M., Hoffman, J. M., West, S. G., & Sheets, V. (2002). A comparison of methods to test mediation and other intervening variable effects. *Psychological methods*, 7(1), 83.
- MacKinnon, D. P., Lockwood, C. M., & Williams, J. (2004). Confidence limits for the indirect effect: Distribution of the product and resampling methods. *Multivariate Behavioral Research*, 39(1), 99-128.
- Maric, M., Wiers, R. W., & Prins, P. J. (2012). Ten ways to improve the use of statistical mediation analysis in the practice of child and adolescent treatment research. *Clinical Child and Family Psychology Review*, 15(3), 177-191.
- McCann, D. C., Thompson, M., Daley, D., Barton, J., Laver-Bradbury, C., Hutchings, J., ... Caddy, J. (2014). Study protocol for a randomized controlled trial comparing the efficacy of a specialist and a generic parenting programme for the treatment of preschool ADHD. *Trials*, 15(1), 142.
- McGee, R., Prior, M., Williams, S., Smart, D., & Sanson, A. (2002). The long-term significance of teacher-rated hyperactivity and reading ability in childhood: Findings from two longitudinal studies. *Journal of Child Psychology and Psychiatry*, 43(8), 1004-1017.
- Moran, P., & Ghate, D. (2005). The effectiveness of parenting support. *Children and Society*, 19, 329-336.
- Moscovitch, D. A., Hofmann, S. G., Suvak, M. K., & In-Albon, T. (2005). Mediation of changes in anxiety and depression during treatment of social phobia. *Journal of Consulting and Clinical Psychology*, 73(5), 945.
- Mrug, S., Hoza, B., & Gerdes, A. C. (2001). Children with attention-deficit/hyperactivity disorder: Peer relationships and peer-oriented interventions. In D. Nangle & C. Erdley (Eds.), *New directions for child and adolescent development: Friendship and psychological adjustment* (pp. 51-77). San Francisco, CA: Jossey-Bass.
- Mrug, S., Hoza, B., Gerdes, A. C., Hinshaw, S., Arnold, L. E., Hechtman, L., & Pelham, W. E. (2009). Discriminating between children with ADHD and classmates using peer variables. *Journal of Attention Disorders*, 12(4), 372-380.
- National Collaborating Centre for Mental Health & Royal College of Psychiatrists' Research and Training Unit. (2009). *Attention deficit hyperactivity disorder: The NICE guideline on diagnosis and management of ADHD in children, young people and adults*. Leicester/London: The British Psychological Society and The Royal

College of Psychiatrists.

- NHS Quality Improvement Scotland. (2009). *Management of attention deficit and hyperkinetic disorders in children and young people: a national clinical guideline*. 112. Scottish Intercollegiate Guidelines Network: Edinburgh. Retrieved from <http://www.sign.ac.uk/guidelines/fulltext/112/>.
- NICE. (2008). *Attention deficit hyperactivity disorder: diagnosis and management of ADHD in children, young people and adults*. Clinical Guideline 72. London: National Institute for Health and Care Excellence.
- NICHQ Vanderbilt Assessment Scales (2002). Retrieved from nichq.org/~media/files/resources/nichqvanderbiltassessment-full.ashx.
- Nixon, R. D., Sweeney, L., Erickson, D. B., & Touyz, S. W. (2004). Parent-child interaction therapy: One-and two-year follow-up of standard and abbreviated treatments for oppositional preschoolers. *Journal of Abnormal Child Psychology*, 32(3), 263-271.
- Nobel, E., Hoekstra, P. J., Brunnekreef, J. A., Messink-de Vries, D. E., Fischer, B., Emmelkamp, P. M., & Van den Hoofdakker, B. J. (2019). Home-based parent training for school-aged children with attention-deficit/hyperactivity disorder and behavior problems with remaining impairing disruptive behaviors after routine treatment: a randomized controlled trial. *European Child & Adolescent Psychiatry*, 1-14.
- Nobel, E., & Van der Veen-Mulders, L. (2012). *BPTG@HOME: Behavioral Parent Training Groningen for children with behavioral problems – at home*. Groningen: Accare University Center of Child and Adolescent Psychiatry, unpublished manual (available upon request).
- Parker, A., & Corkum, P. (2016). ADHD diagnosis: as simple as administering a questionnaire or a complex diagnostic process?. *Journal of Attention Disorders*, 20(6), 478-486.
- Parmar, M. K. B., Sydes, M. R., & Morris, T. P., (2016). How do you design randomised trials for smaller populations? A framework. *BMC Medicine*, 14, 183.
- Patsopoulos, N. A. (2011). A pragmatic view on pragmatic trials. *Dialogues in Clinical Neuroscience*, 13(2), 217
- Patterson, G. R., & Chamberlain, P. (1994). A functional analysis of resistance during parent training therapy. *Clinical Psychology: Science & Practice*, 1(1), 53-70.
- Peduzzi, P., Concato, J., Kemper, E., Holford, T. R., & Feinstein, A. R. (1996). A simulation study of the number of events per variable in logistic regression analysis. *Journal of Clinical Epidemiology*, 49(12), 1373-1379.
- Pelham, Jr. W. E., & Fabiano, G. A. (2008). Evidence-based psychosocial treatments for attention-deficit/hyperactivity disorder. *Journal of Clinical Child & Adolescent Psychology*, 37(1), 184-214.
- Pimentel, M. J., Vieira-Santos, S., Santos, V., & Vale, M. C. (2011). Mothers of children with attention deficit/hyperactivity disorder: relationship among parenting stress, parental practices and child behaviour. *ADHD Attention Deficit and Hyperactivity Disorders*, 3(1), 61-68.
- Pliszka, S., & AACAP Work Group on Quality Issues. (2007). Practice parameter for the assessment and treatment of children and adolescents with attention-deficit/hyperactivity disorder. *Journal of the American Academy of Child & Adolescent Psychiatry*, 46(7), 894-921.
- Polanczyk, G., De Lima, M. S., Horta, B. L., Biederman, J., & Rohde, L. A. (2007). The worldwide prevalence of ADHD: a systematic review and metaregression analysis. *American Journal of Psychiatry*, 164(6), 942-948.
- Polanczyk, G. V., Salum, G. A., Sugaya, L. S., Caye, A., & Rohde, L. A. (2015). Annual research review: A meta-analysis of the worldwide prevalence of mental disorders in children and adolescents. *Journal of Child Psychology and Psychiatry*, 56(3), 345-365.
- Polanczyk, G. V., Willcutt, E. G., Salum, G. A., Kieling, C., & Rohde, L. A. (2014). ADHD prevalence estimates across three decades: an updated systematic review and meta-regression analysis. *International Journal of Epidemiology*, 43(2), 434-442.
- Preacher, K. J., & Kelley, K. (2011). Effect size measures for mediation models: quantitative strategies for communicating indirect effects. *Psychological methods*, 16(2), 93.
- Raiker, J. S., Freeman, A. J., Perez-Algorta, G., Frazier, T. W., Findling, R. L., & Youngstrom, E. A. (2017). Accuracy of Achenbach scales in the screening of attention-deficit/hyperactivity disorder in a community mental health clinic. *Journal of the American Academy of Child & Adolescent Psychiatry*, 56(5), 401-409.
- Rajwan, E., Chacko, A., Wymbs, B. T., & Wymbs, F. A. (2014). Evaluating clinically significant change in mother and child functioning: Comparison of traditional and enhanced behavioral parent training. *Journal of Abnormal Child Psychology*, 42(8), 1407-1412.
- Reale, L., Bartoli, B., Cartabia, M., Zanetti, M., Costantino, M. A., Canevini, M. P., ... Lombardy ADHD Group. (2017). Comorbidity prevalence and treatment outcome in children and adolescents with ADHD. *European Child & Adolescent Psychiatry*, 26(12), 1443-1457.
- Reynolds, C. R., & Kamphaus, R. W. (2004). *BASC-2: behavioral assessment for children manual*, 2nd edn. AGS Publishing, Circle Pines.

- Riera, M., Castells, X., Tobias, A., Cunill, R., Blanco, L., & Capella, D. (2017). Discontinuation of pharmacological treatment of children and adolescents with attention deficit hyperactivity disorder: meta-analysis of 63 studies enrolling 11,788 patients. *Psychopharmacology (Berl)*, 234(17), 2657-2671.
- Rosseel, Y. (2012). "lavaan: An R Package for Structural Equation Modeling." *Journal of Statistical Software*, 48(2), 1-36.
- Sandler, I. N., Schoenfelder, E. N., Wolchik, S. A., & MacKinnon, D. P. (2011). Long-term impact of prevention programs to promote effective parenting: Lasting effects but uncertain processes. *Annual review of psychology*, 62, 299-329.
- Schachar, R.J., Ickowicz, A., & Wachsmuth, R. (1995). *Parent Interview for Child Symptoms (P.I.C.S.-4)*. Unpublished document: The Hospital for Sick Children, Toronto, ON Canada.
- Schachter, H.M., Pham, B., King, J., Langford, S., & Moher, D. (2001). How efficacious and safe is short-acting methylphenidate for the treatment of attention-deficit disorder in children and adolescents? A meta-analysis. *Canadian Medical Association Journal*, 165(11), 1475-1488.
- Schatz, N.K., Fabiano, G. A., Cunningham, C. E., dosReis, S., Waschbusch, D. A., Jerome, S., ... Morris, K. L., (2015). Systematic review of patients' and parents' preferences for ADHD treatment options and processes of care. *Patient*, 8, 483-497.
- Shaw, M., Hodgkins, P., Caci, H., Young, S., Kahle, J., Woods, A. G., & Arnold, L. E. (2012) A systematic review and analysis of long-term outcomes in attention deficit hyperactivity disorder: effects of treatment and non-treatment. *BMC Medicine*, 10, 99.
- Shilon, Y., Pollak, Y., Aran, A., Shaked, S., & Gross-Tsur, V. (2012). Accidental injuries are more common in children with attention deficit hyperactivity disorder compared with their non-affected siblings. *Child: Care, Health and Development*, 38(3), 366-370.
- Sivan, A. B., Ridge, A., Gross, D., Richardson, R., & Cowell, J. (2008). Analysis of two measures of child behavior problems by African American, Latino, and non-Hispanic Caucasian parents of young children: a focus group study. *Journal of Pediatric Nursing*, 23(1), 20-27.
- Smith, E., Koerting, J., Latter, S., Knowles, M.M., Mccann, D.C., Thompson, M., & Sonuga-Barke, E.J. (2015). Overcoming barriers to effective early parenting interventions for attention- deficit hyperactivity disorder (ADHD): Parent and practitioner views. *Child Care Health and Development*, 41, 93-102.
- Sobanski, E., Banaschewski, T., Asherson, P., Buitelaar, J., Chen, W., Franke, B., ... Stringaris, A. (2010). Emotional lability in children and adolescents with attention deficit/hyperactivity disorder (ADHD): clinical correlates and familial prevalence. *Journal of Child Psychology and Psychiatry*, 51(8), 915-923.
- Sobel, M. E. (1982). Asymptotic confidence intervals for indirect effects in structural equation models. *Sociological Methodology*, 13, 290-312.
- Sonuga-Barke, E. J., Barton, J., Daley, D., Hutchings, J., Maishman, T., Raftery, J., ... Little, L. (2018). A comparison of the clinical effectiveness and cost of specialised individually delivered parent training for preschool attention-deficit/hyperactivity disorder and a generic, group-based programme: a multi-centre, randomised controlled trial of the New Forest Parenting Programme versus Incredible Years. *European Child & Adolescent Psychiatry*, 27(6), 797-809.
- Sonuga-Barke, E.J., Brandeis, D., Cortese, S., Daley, D., Ferrin, M., Holtmann, M., ... Sergeant, J. (European ADHD Guidelines Group). (2013). Nonpharmacological interventions for ADHD: Systematic review and meta Analyses of randomized controlled trials of dietary and psychological treatments. *The American Journal of Psychiatry*, 170(3), 275-289.
- Sonuga-Barke, E. J., Daley, D., Thompson, M., Laver-Bradbury, C., & Weeks, A. (2001). Parent-based therapies for preschool attention-deficit/hyperactivity disorder: a randomized, controlled trial with a community sample. *Journal of the American Academy of Child & Adolescent Psychiatry*, 40(4), 402-408.
- Steinhausen, H. C., Nøvik, T. S., Baldursson, G., Curatolo, P., Lorenzo, M. J., Pereira, R. R., ... Rothenberger, A. (ADORE Study Group). (2006). Co-existing psychiatric problems in ADHD in the ADORE cohort. *European Child & Adolescent Psychiatry*, 15(1), i25-i29.
- Swanson, J. (1992). *School-based assessments and interventions for ADD students*. Irvine, CA: K.C. Publications.
- Szobot, C.M., Rohde, L.A., Bukstein, O., Molina, B.S., Martins, C., Ruaro, P., et al. (2007). Is attention-deficit/hyperactivity disorder associated with illicit substance use disorders in male adolescents? A community-based case-control study. *Addiction*, 102, 1122-1130.
- Taurines, R., Schmitt, J., Renner, T., Conner, A. C., Warnke, A., & Romanos, M. (2010). Developmental comorbidity in attention-deficit/hyperactivity disorder. *ADHD Attention Deficit and Hyperactivity Disorders*, 2(4), 267-289.
- Taylor, A. (2015). Standardised Effect Size in a Mixed/Multilevel Model. Retrieved from http://www.psy.mq.edu.au/psystat/documents/standardised_effect_size_in_mixed_ML_models.pdf.

- Taylor, E., Döpfner, M., Sergeant, J., Asherson, P., Banaschewski, T., Buitelaar, J., ... Steinhausen, H. C. (2004). European clinical guidelines for hyperkinetic disorder—first upgrade. *European Child & Adolescent Psychiatry*, 13(1), i7-i30.
- The MTA Cooperative Group. (1999). A 14-month randomized clinical trial of treatment strategies for attention-deficit/hyperactivity disorder. *Archives of General Psychiatry*, 56, 1073-1086.
- Thomas, R., Sanders, S., Doust, J., Beller, E., & Glasziou, P. (2015). Prevalence of attention-deficit/hyperactivity disorder: a systematic review and meta-analysis. *Pediatrics*, 135, 994-1001.
- Tsai, C. J., Chen, Y. L., Lin, H. Y., & Gau, S. S. F. (2017). One-year trajectory analysis for ADHD symptoms and its associated factors in community-based children and adolescents in Taiwan. *Child and Adolescent Psychiatry and Mental Health*, 11(1), 28.
- Ullebø, A. K., Posserud, M. B., Heiervang, E., Obel, C., & Gillberg, C. (2012). Prevalence of the ADHD phenotype in 7-to 9-year-old children: effects of informant, gender and non-participation. *Social Psychiatry and Psychiatric Epidemiology*, 47(5), 763-769.
- Unesco Institute for Statistics. (2014). International Standard Classification of Education. *ISCED-F 2013*. Montreal, Canada.
- Van de Loo-Neus, G. H., Rommelse, N., & Buitelaar, J. K. (2011). To stop or not to stop? How long should medication treatment of attention-deficit hyperactivity disorder be extended? *European Neuropsychopharmacology*, 21(8), 584-599.
- Van Den Hoofdakker, B. J., Van der Veen-Mulders, L., Sytema, S., Emmelkamp, P. M., Minderaa, R. B., & Nauta, M. H. (2007). Effectiveness of behavioral parent training for children with ADHD in routine clinical practice: A randomized controlled study. *Journal of the American Academy of Child and Adolescent Psychiatry*, 46, 1263-1271.
- Vander Stoep, A., McCarty, C.A., Zhou, C., Rockhill, C.M., Schoenfelder, E.N., & Myers, K. (2017). The children's attention-deficit hyperactivity disorder telemental health treatment study: caregiver outcomes. *Journal of Abnormal Child Psychology*, 45, 27-43.
- Vreugdenhil, C., Doreleijers, T. A., Vermeiren, R., Wouters, L. F., & Van Den Brink, W. (2004). Psychiatric disorders in a representative sample of incarcerated boys in the Netherlands. *Journal of the American Academy of Child & Adolescent Psychiatry*, 43(1), 97-104.
- Wagner, D. J., Vallerand, I. A., & McLennan, J. D. (2014). Treatment receipt and outcomes from a clinic employing the attention-deficit/hyperactivity disorder treatment guideline of the children's medication algorithm project. *Journal of Child and Adolescent Psychopharmacology*, 24(9), 472-480.
- Webster-Stratton, C. (1990). Predictors of treatment outcome in parent training for families with conduct problem children. *Behavior Therapy*, 21, 319-337.
- Webster-Stratton, C. (1996). Early Intervention with videotape modeling: Programs for families of children with oppositional defiant disorder or conduct disorder. In Hibbs E, Jensen P (Eds.) *Psychosocial treatments for child and adolescent disorders: Empirically based strategies for clinical practice*, 435- 474. New York: APA.
- Webster-Stratton, C. (1998). Preventing conduct problems in Head Start children: strengthening parenting competencies. *Journal of Consulting and Clinical Psychology*, 66(5), 715.
- Webster-Stratton, C. H., Reid, M. J., & Beauchaine, T. (2011). Combining parent and child training for young children with ADHD. *Journal of Clinical Child & Adolescent Psychology*, 40(2), 191-203.
- Webster-Stratton, C., Reid, M. J., & Hammond, M. (2001). Preventing conduct problems, promoting social competence: A parent and teacher training partnership in Head Start. *Journal of Clinical Child Psychology*, 30(3), 283-302.
- Wechsler, D. A. (1991). *The Wechsler Intelligence Scale for Children-III*. London: Pearson Assessment.
- Wells, K. C., Chi, T. C., Hinshaw, S. P., Epstein, J. N., Pfiffner, L., Nebel-Schwalm, M., ... Elliott, G. R. (2006). Treatment-related changes in objectively measured parenting behaviors in the multimodal treatment study of children with attention-deficit/hyperactivity disorder. *Journal of Consulting and Clinical Psychology*, 74(4), 649.
- Willcutt, E.G. (2012). The prevalence of DSM-IV attention-deficit/ hyperactivity disorder: a meta-analytic review. *Neurotherapeutics*, 9, 490-499.
- Wittchen, H. U., Jacobi, F., Rehm, J., Gustavsson, A., Svensson, M., Jönsson, B., ... Fratiglioni, L. (2011). The size and burden of mental disorders and other disorders of the brain in Europe 2010. *European Neuropsychopharmacology*, 21(9), 655-679.
- Xie, Y., Dixon, J. F., Yee, O. M., Zhang, J., Chen, Y. A., DeAngelo, S., ... Schweitzer, J. B. (2013). A study on the effectiveness of videoconferencing on teaching parent training skills to parents of children with ADHD. *Telemedicine and E-Health*, 19(3), 192-199.

- Yamada, D. C., Porter, A. A., Conway, J. L., LeBlanc, J. C., Shea, S. E., Hancock-Friesen, C. L., & Warren, A. E. (2013). Early repair of congenital heart disease associated with increased rate of attention deficit hyperactivity disorder symptoms. *Canadian Journal of Cardiology*, 29(12), 1623-1628.
- Yang, R., Gao, W., Li, R., & Zhao, Z. (2015). Effect of atomoxetine on the cognitive functions in treatment of attention deficit hyperactivity disorder in children with congenital hypothyroidism: a pilot study. *International Journal of Neuropsychopharmacology*, 18(8).

Samenvatting in het Nederlands

Inleiding

Kinderen met een aandachtsdeficiëntie-/hyperactiviteitsstoornis (ADHD) hebben persisterende concentratieproblemen en/of hyperactief en impulsief gedrag in verschillende situaties in het dagelijks leven. Deze gedragingen zijn van dien aard dat ze niet passen bij het ontwikkelingsniveau van het kind en het functioneren in het dagelijks leven bemoeilijken. Kinderen met ADHD hebben vaak problemen op school (zoals met leren) en in het contact met leeftijdsgenoten. Dit alles met een verminderde kwaliteit van leven tot gevolg. ADHD is een van de meest voorkomende psychiatrische diagnoses onder kinderen en adolescenten en ongeveer 75% van de kinderen met ADHD heeft nog een andere bijkomende stoornis, zoals een gedragsstoornis of een angst- of stemmingsstoornis. Ouders van kinderen met ADHD ervaren vaak stress rondom het ouderschap. Deze stress en de opvoedvaardigheden van ouders hangen samen met de ernst van de ADHD en de aanwezigheid van bijkomende problematiek van het kind. Al met al is het belangrijk dat kinderen met ADHD effectief worden behandeld.

Inmiddels zijn er twee behandelvormen voor kinderen met ADHD die binnen meerdere wetenschappelijke studies effectief zijn gebleken en wereldwijd worden ingezet: psychofarmaca (met name psychostimulantia) en gedragstherapeutische oudertraining. Oudertraining heeft als doel de gedragsproblemen van de kinderen met ADHD te verminderen door ouders gedragstherapeutische vaardigheden aan te leren die ze thuis kunnen inzetten. Dergelijke trainingen zijn van origine geprotocolleerd, worden in instellingen vaak in een groep gegeven en bestaan meestal uit acht tot twaalf wekelijkse sessies. Zowel medicatie als oudertraining of de combinatie ervan, zijn echter niet voor alle kinderen met ADHD een toereikende behandeling. Een deel van de kinderen hebben na (een poging tot) deze behandelingen meer hulp nodig, bijvoorbeeld als er niet voldoende verbetering optreedt. Daarnaast is medicatie soms geen optie door vervelende bijwerkingen of omdat ouders een dergelijke behandeling niet acceptabel vinden. Oudertraining wordt bovendien vaak (in ongeveer 50% van de gevallen) niet volledig gevolgd, bijvoorbeeld omdat ouders het niet georganiseerd krijgen om wekelijks naar een instelling te komen. In al deze gevallen is vervolgbehandeling nodig. Meerdere behandelopties voor ADHD zijn beschikbaar, maar niet of nauwelijks (voor zover goed onderzocht) effectief gebleken, zoals behandelingen met het kind (bijvoorbeeld

socialevaardigheidstraining of neurofeedback), meditatie gerichte behandelingen en dieetbehandelingen. Wat zou dan wel een effectieve vervolgbehandeling kunnen zijn voor deze gezinnen?

Thuisbehandeling als vervolgbehandeling voor kinderen met ADHD

In *hoofdstuk 1* heb ik de behoefte aan vervolgbehandeling voor kinderen met ADHD uiteengezet en een in Groningen ontwikkelde thuisbehandeling beschreven, die mogelijk een effectieve vervolgbehandeling kan zijn. Deze thuisbehandeling (genaamd *Behavioral Parent Training Groningen @HOME* [BPTG@HOME]) is een gedragstherapeutische oudertraining die barrières van een poliklinische oudertraining mogelijk wegneemt, waardoor ouders toch van een dergelijke behandeling kunnen profiteren. Het is een geprotocolleerde behandeling, bestaande uit veertien tot zestien wekelijkse huisbezoeken. Tijdens de huisbezoeken leren ouders opvoedingsvaardigheden met behulp van video-feedback. Ook ontvangen ze psycho-educatie en worden cognitieve interventies ingezet om hun negatieve gedachten over hun kind of hun ouderschap te bewerken. Ondanks dat de behandeling geprotocolleerd is, bestaat deze uit verschillende behandelmodules, waarbij de therapeut bekijkt welke behandelmodules noodzakelijk zijn voor een gezin en bepaalt hoeveel sessies hij/zij besteedt aan een bepaalde behandelmodule. Hierdoor kan er zorg op maat worden geleverd.

De effectiviteit van thuisbehandeling als vervolgbehandeling voor kinderen met ADHD

In *hoofdstuk 2* beschrijf ik een gerandomiseerde gecontroleerde studie, waar de effectiviteit van BPTG@HOME als vervolgbehandeling op verschillende kind-uitkomstmaten is onderzocht. In de studie hebben we 73 kinderen met ADHD van de basisschoolleeftijd geïnccludeerd, die niet (voldoende) hadden kunnen profiteren van medicatie en/of poliklinische oudertraining. Ouders van deze kinderen rapporteerden

ondanks eerdere behandel pogingen nog een verhoogd niveau van gedragsproblemen (symptomen van ADHD en/of gedragsstoornissen). Kinderen en hun ouders hebben we gerandomiseerd naar BPTG@HOME ($n=26$), een wachtlijst van 4 maanden ($n=23$) of een thuisbehandeling met eclectische opzet zoals die gebruikelijk werd gegeven ($n=24$). Ouders die thuisbehandeling kregen, wisten niet of ze de experimentele of de gebruikelijke thuisbehandeling ontvingen. De onderzoeksgegevens hebben we geanalyseerd met *mixed models of repeated measures*, waaruit bleek dat ouders die BPTG@HOME ontvingen ten opzichte van de wachtlijst significant minder gedragsproblemen in brede zin (combinatie van druk, ongeconcentreerd en opstandig gedrag), ADHD-symptomen, oppositioneel-opstandige gedragsstoornis (ODD)-symptomen en internaliserende klachten rapporteerden. Vergeleken met de gebruikelijke thuisbehandeling vonden we dezelfde resultaten, met uitzondering van het effect op de internaliserende klachten. Omdat de meeste resultaten gevonden werden in vergelijking met zowel de wachtlijst als met de gebruikelijke thuisbehandeling, kan geconcludeerd worden dat niet enkel het feit dat de behandeling thuis werd aangeboden het effect bepaalde. Tot slot bleek dat zes maanden na afronding van de thuisbehandelingen ouders die BPTG@HOME ontvingen de gedragsproblemen van hun kind als minder problematisch ervoeren dan de ouders die de gebruikelijke thuisbehandeling kregen. Kortom, BPTG@HOME bleek een effectieve vervolgbehandeling om de gedragsproblemen te verminderen van kinderen met ADHD die van eerdere behandeling niet voldoende waren opgeknapt.

In *hoofdstuk 3* heb ik op gelijke wijze de effectiviteit van BPTG@HOME onderzocht rondom oudermaten. We onderzochten of ouders na thuisbehandeling verbetering lieten zien in hun gevoel van ouderlijke competentie, ouderlijke stress en opvoedingsvaardigheden. Vergeleken met de wachtlijstgroep rapporteerden ouders die BPTG@HOME ontvingen significante verbeteringen aangaande hun gevoel van ouderlijke competentie, ervoeren ze minder stress als het ging om interacteren met hun kind en het omgaan met de gedragsproblemen van hun kind. Als we keken naar opvoedingsvaardigheden, dan gebruikten de ouders die BPTG@HOME ontvingen significant minder harde en inconsistente disciplineren. Wanneer we de ouders die BPTG@HOME ontvingen vergeleken met ouders die de gebruikelijke thuisbehandeling volgden zagen we dat de ouders die BPTG@HOME hadden gevolgd significant minder

harde en inconsistente disciplineren gebruikten. De effecten rondom gevoel van ouderlijke competentie en ouderlijke stress zoals vergeleken met de wachtlijstgroep vonden we nu niet. Echter, BPTG@HOME duurde de helft korter en bevatte de helft minder huisbezoeken ten opzichte van de gebruikelijke thuisbehandeling. Wanneer we met dit verschil rekening hielden, was BPTG@HOME op meer maten effectiever dan de gebruikelijke thuisbehandeling: naast het gebruik van minder harde en inconsistente disciplineren gebruikten ouders die BPTG@HOME kregen ook meer positieve verbale disciplineren, complimentjes en beloningen en hielden ze hun kind beter in de gaten. Ook nam de ouderlijke stress in brede zin bij ouders die BPTG@HOME ontvingen significant meer af ten opzichte van de ouders die de gebruikelijke thuisbehandeling ontvingen. Kortom, ook in deze studie werd de meerwaarde van BPTG@HOME als vervolgbehandeling zichtbaar. Langetermijneffecten (zes maanden na afronding van behandeling) op de oudermaten vonden we echter niet.

De mogelijke werking van thuisbehandeling

De gedachte achter een oudertraining is dat door de opvoedingsvaardigheden van ouders te verbeteren, de gedragsproblemen van het kind afnemen. In *hoofdstuk 4* beschrijf ik onderzoek naar dergelijke mediatie-effecten voor BPTG@HOME. Dit hebben we gedaan door aan de hand van *structural equation modeling* te kijken of er bij de BPTG@HOME-groep ($n=22$) ten opzichte van de wachtlijst groep ($n=21$) een mediërend effect was van de gevonden effectieve oudermaten op de gevonden effectieve kindmaten. In deze studie bleek dat vermindering van harde en inconsistente disciplineren en ouderlijke stress aangaande het omgaan met de gedragsproblemen van het kind leidde tot afname van zowel ADHD- als ODD-symptomen bij het kind. De verbetering van ouders in hun gevoel van ouderlijke competentie leidde met name tot vermindering van ODD-symptomen bij het kind, maar niet tot vermindering van ADHD-symptomen. We vonden geen mediërend effect van vermindering van door ouders ervaren stressvolle ouder-kind interacties op het gedrag van het kind. Hieruit konden we concluderen dat binnen een thuisbehandeling voor kinderen met ADHD die van eerdere behandelingen onvoldoende hebben geprofiteerd, het belangrijk lijkt dat er gerichte interventies worden gedaan op

het verminderen van harde en inconsistente disciplineren door de ouders, het verminderen van stress bij ouders aangaande het omgaan met de gedragsproblemen van hun kind en het verbeteren van hun gevoel van ouderlijke competentie.

Het meten van de aanwezigheid en ernst van ADHD

In veel studies naar de effectiviteit van oudertrainingen worden door ouders gerapporteerde uitkomstmaten gebruikt, terwijl zij vaak niet geblindeerd zijn voor de ontvangen behandeling. Zodoende hebben recente studies geadviseerd om geblindeerde beoordelaars zoals klinici en leerkrachten in te zetten om bijvoorbeeld de ernst van de ADHD te beoordelen voor en na de behandeling in effectiviteitsstudies. De vraag is echter of deze beoordelaars een goed beeld kunnen geven van het functioneren in de thuissituatie als zij het kind in een instelling of op school observeren en beoordelen. Wij hebben zodoende bij de hierboven beschreven studies toch gekozen voor door ouders gerapporteerde uitkomstmaten, waaronder de ernst van de ADHD-symptomen. In *hoofdstuk 5* heb ik gekeken of dat een acceptabele keuze is geweest, door te onderzoeken of er voldoende overeenstemming was tussen ouders en klinici aangaande de ernst van de ADHD-symptomen van het kind voorafgaand aan de behandeling. Met regressieanalyse is de overeenstemming aangaande ADHD (symptomen) bepaald tussen ouderrapportage met de *Swanson, Nolan and Pelham Questionnaire-IV* (SNAP-IV) en het oordeel van een clinicus op basis van het semigestructureerde interview *Parent Interview for Child Symptoms* (PICS-IV; $N=70$). Er bleek voldoende overeenstemming tussen ouders en klinici te zijn voor de ernst van (totale) ADHD-symptomen en de ernst van impulsieve en hyperactieve symptomen. We vonden echter onvoldoende overeenstemming voor de ernst van aandachtsdeficiëntie-symptomen. Verder vonden we slechts een lage overeenstemming tussen ouders en klinici over de ernst en aanwezigheid van specifieke ADHD-symptomen en de aanwezigheid van een diagnose ADHD. We concludeerden dat het gebruik van ouderrapportage om de ernst van de ADHD-symptomen te bepalen in effectiviteitsstudies acceptabel is, maar niet om een diagnose ADHD vast te stellen in bijvoorbeeld epidemiologische studies. Daarbij kunnen vragenlijsten waar ouders de mate van ADHD rapporteren mogelijk verbeterd worden door de aandachtsdeficiëntie-

symptomen in meer observeerbaar gedrag te beschrijven (zoals hyperactief en impulsief gedrag) in plaats van als moeilijk observeerbare cognitieve functies.

Conclusie

In *hoofdstuk 6* staat een korte samenvatting van de vier hierboven beschreven studies. Ik ga specifiek in op de klinische implicaties van de gevonden resultaten. Tevens heb ik relevante wetenschappelijke inzichten die al werkend aan het promotieonderzoek zijn opgedaan op een rij gezet.

De belangrijkste klinische implicatie is dat BPTG@HOME een effectieve vervolgbehandeling is voor kinderen met ADHD die onvoldoende geprofiteerd hebben van medicatie en poliklinische oudertraining. Na behandeling met BPTG@HOME zijn kinderen haast niet meer te onderscheiden van kinderen uit een normgroep wat betreft het aanwezig zijn van gedragsproblemen. Met de studies in dit proefschrift laat ik enerzijds zien dat het gedrag van de kinderen verbetert en anderzijds dat ouders in hun ouderlijke rol vooruitgang boeken. Tot nu was er geen duidelijke vervolgbehandeling voor kinderen die onvoldoende baat hebben bij de eerste keus behandelingen (medicatie en oudertraining). Daarbij hoeft vervolgbehandeling niet noodzakelijk langdurig en intensief te zijn om effectief te zijn. Een behandeling van vier maanden met wekelijkse huisbezoeken blijkt al effect te hebben. Ook al zijn er geen studies gedaan naar de mogelijk effectieve elementen van BPTG@HOME, in de hoofdstukken heb ik wel hypothesen geformuleerd over wat belangrijk elementen in deze vervolgbehandeling zouden kunnen zijn. Mogelijk relevant is het inzetten van academisch geschoolde psychologen als therapeuten. Maar ook elementen in de behandeling zijn wellicht belangrijk, zoals het gebruik maken van directe modeling en video-feedback en het tempo van de behandeling laten aansluiten bij de leersnelheid van ouders. Ook lijkt het belangrijk dat de behandeling zich niet alleen richt op ouders leren verschillende disciplineringsmethodes effectief toe te passen, maar ook op het versterken van het zelfvertrouwen van ouders aangaande hun ouderlijke vaardigheden. Dit wordt binnen BPTG@HOME mogelijk bereikt door het uitdagen van negatieve gedachten van ouders over hun pedagogische rol. Tot slot heb ik kort stil gestaan op welke wijze ADHD-

vragenlijsten die door ouders worden ingevuld gebruikt kunnen worden in de klinische praktijk. Onze studie liet zien dat ze niet geschikt zijn om een ADHD diagnose te stellen. Semigestructureerd klinische interviews lijken daarvoor geschikter. De vragenlijsten zijn wel geschikt om ouders aan te laten geven hoe ernstig de aanwezige ADHD-symptomen zijn, wat bijvoorbeeld praktisch is bij de evaluatie van behandelingen.

Gedurende het werken aan dit promotieonderzoek zijn een aantal wetenschappelijke vraagstukken aan de orde gekomen. Zo wordt voor het onderzoeken van de effectiviteit van behandelingen meestal eerst binnen een *explanatory trial*, zoals een gerandomiseerde gecontroleerde studie, de effectiviteit van een behandeling onderzocht. In een dergelijke trial wordt geprobeerd de effectiviteit van een behandeling vast te stellen onder ideale, gecontroleerde situaties. Vervolgens zou de klinische relevantie getoetst moeten worden binnen een *pragmatic trial* door de effecten van een behandeling vast te stellen in de alledaagse klinische praktijk, wat vaak niet wordt gedaan. In studies zoals deze, gericht op kinderen die niet geprofiteerd hebben van eerdere behandelingen, is het in de praktijk lastig om een explanatory trial uit te voeren en zijn we dus gestart met een pragmatic trial. Ook barrières bij het opzetten van een studie met deelnemers die niet op eerdere behandeling hebben gereageerd benoem ik, zoals inclusieproblemen, keuze van passende statistische methodes en problemen rondom het opzetten van een multicenter studie. Tot slot heb ik gereflecteerd over het wel of niet valide kunnen inzetten van ouderrapportage-vragenlijsten als uitkomstmaten binnen effectiviteitsstudies.

Dankwoord

Ik heb in mijn leven mogen opgroeien in een warm, veilig gezin, waar ik me ten volle kon ontwikkelen en ontplooiën en leren hoe je gelukkig kan zijn in een leven vol uitdagingen. Het heeft me een basis gegeven, waarop ik nog altijd terug kan vallen, wanneer het moeilijk is, ik word geconfronteerd met mijn beperkingen en zoekend ben in hoe er voor een ander te zijn. En die basis kreeg onder andere een klank in een liedje van troost, wat ik in de buik van mijn moeder en bij verdriet in mijn leven elke keer weer gezongen hoorde.

*O, give me a home where the buffalo roam,
Where the dear and the antelope play,
Where seldom is heard a discouraging word,
And the skies are not cloudy all day.*

(Dr. Brewster Higley)

Toen een onderzoek naar de effecten van *thuis*behandeling bij kinderen met ADHD op mijn pad kwam, hoefde ik dus niet lang na te denken. Een rol mogen spelen in de ontwikkeling van een thuisbehandeling en nagaan of dat gezinnen kan helpen, die door het leven flink worden uitgedaagd, was een bijzonder voorrecht. Elk mens gun ik een jeugd *where seldom is heard a discouraging word* en die leidt naar een leven waar *the skies are not cloudy all day*. Daar me voor inzetten was fantastisch. En wat ben ik onder de indruk van deze gezinnen. In donkere tijden, waar ze het zwaar hebben, niet meer weten de cirkel te doorbreken, durven vragen naar hulp bij de opvoeding van je kinderen en dan ook nog je blootgeven voor wetenschappelijk onderzoek in het belang van een ander, verdient niet meer dan respect en een heel diepe buiging. Mijn dankwoord begint dus ook met het uitspreken van mijn bijzondere dankbaarheid naar alle ouders en hun kinderen die mij, het onderzoeksteam en de behandelaren in hun thuis hebben toegelaten. Zonder hen geen onderzoek, geen promotie, geen kostbare persoonlijke levenslessen, geen vastgestelde effectiviteit van thuisbehandeling bij een moeilijk te behandelen groep en geen duidelijkheid voor de clinicus wat te doen bij andere gezinnen zoals zij. Wauw!

De gezinnen waren de basis van mijn promotieonderzoek. Terzake, er was werk aan de winkel en ik ging aan de slag met een compleet onderzoeksteam. Ook hen ben ik

dankbaar. Pieter Hoekstra en Barbara van den Hoofdakker ben ik dankbaar, dat zij mij de gelegenheid en hun tijd en energie hebben gegeven om onder hun begeleiding te promoveren. Het is een lange weg geweest, van nabijheid en afstand, waarvan we nu tegen elkaar kunnen zeggen: het is gelukt, er ligt een eindresultaat, een tevreden afronding. Bedankt.

De dataverzameling was een grote klus in deze studie, doordat we elke meting bij de gezinnen op bezoek gingen om de interviews en vragenlijsten af te nemen. Dit werd haalbaar met een mooie club jonge psychologen, die met veel energie en gezelligheid richting de gezinnen stoven. Nancy Anbergen, Mafalda de Baets, Pim de Geus, Michell Ramaker, Anne Marie Scholte, Jennie Viersen en Annemaaïke van der Wal bedankt voor jullie inzet, gezelligheid en altijd weer het aanbod of jullie nog wat anders voor me konden doen dan alleen de metingen. Ook Anita Schelling wil ik bedanken voor het zorgen voor de geblindeerde lotingen. Een speciale plek moet Mascha van den Akker krijgen. Mascha kwam, zag en overwon. Wat heerlijk, dat ik met jouw komst mij opeens, naast de taken die ik als clinicus had, vooral op de inhoud van het onderzoek en de werving van proefpersonen kon focussen. Je nam de hele logistieke organisatie van het onderzoek van me over, vroeg in het begin of je in het proces wat mocht veranderen (wat altijd een verbetering was) en daarna deed je dat volledig op eigen inzicht. Wat zalig dat ik daarop blind kon vertrouwen. Terwijl je ook een luisterend oor had, wanneer ik even niet meer wist of dit onderzoek wel een einde ging krijgen. Bedankt lieve Mascha en ik geniet van onze telefoontjes zo op z'n tijd, waar we even bijkletsen over onze gezinnen.

Om een klinische studie tot een succes te maken is niet alleen een onderzoeksteam vereist. Ook is er een klinische praktijk nodig, die de studie met open armen ontvangt. Accare ben ik dankbaar, dat het doen van wetenschappelijk onderzoek hoog in hun vaandel staat. De behandelaren van Groningen en toen de inclusie tegen viel ook de behandelaren van Friesland en Drenthe gingen vier jaar lang aan de slag met het uitnodigen van gezinnen voor het onderzoek. Ook GGZ In de Bres en Lentis Jonx kwamen helpen met de inclusie, wat voor hen betekende eigen zorgprocessen her-evalueren, professionals informeren, onderzoeker toelaten bij MDO's en behandelaren trainen voor BPTG@HOME. Super! En dan natuurlijk alle behandelaren, die de thuisbehandelingen hebben verzorgd. Dames en die ene heer, bedankt voor jullie inzet. En dan hebben we het over de BPTG@HOME behandelaren Bregitte Pruim, Nienke Hoekstra, Lotte Houwing,

Marijn Nijboer, Petra Nijland, Marjolein Pieper, Nienke Straatman en Lotte Taminiau. We hebben het over de IPG-behandelaren Piet Alkema, Annette Bakker, Gertrude Bouma, Miranda van Donderen, Marga Hertsenberg, Grystje Hiddema, Carla van Kalsbeek, Rommie Kranenbrug, Nynke Kroes, Joske Prummel en Marjan Sikkema. En we hebben het over Jetty Kappelhof, die naar de gezinnen ging nadat hun wachtlijst-tijd voorbij was. Ook wil ik Lianne van der Veen bedanken voor het samen schrijven aan en aanscherpen van de behandeling BPTG@HOME, het samen trainen van alle BPTG@HOME behandelaren en hen superviseren tijdens het onderzoek.

Tot slot ben ik de mensen dankbaar, die een cruciale rol hebben gespeeld in de afronding van mijn proefschrift. Op een speciale plek wil ik neerzetten Stella Noach en Robert Schoevers. Stella, jij hebt mij hernieuwd *vertrouwen* gegeven dat het goed ging komen en hebt me geholpen om op het universitaire systeem te vertrouwen. Zonder dit had ik het misschien wel opgegeven. En man, wat kon ik erop *vertrouwen*. Robert, mijn dank is groot voor het valideren van mijn gevoel en daarna je doortastende en open aanpak, waarmee er met een oogwenk een klaar proefschrift lag. Een onwerkelijk moment. En wat heerlijk, wanneer er in mei en juni vlot achter elkaar driemaal een goedkeuring van de beoordelingscommissie komt: prof. dr. J.K. Buitelaar, prof. dr. S.A. Reijneveld en prof. dr. R.A. Schoevers hartelijk dank daarvoor.

Promoveren bleek zoveel meer dan een baan. Het vroeg een grote investering, ook de vijf jaar nadat de baan was afgelopen. Ik had het niet kunnen doen zonder de mensen in mijn leven, die me trouw zijn, me liefde geven en me elke keer weer een *bemoedigend woord* boden. Het gaf een basis van *zonneschijn*, wat me op de been hield.

Op elke plek, waar ik heb mogen leren, was daar iemand, die zich met me verbond. Stuk voor stuk vriendinnen, die ik niet kan missen. Naast de verjaardag-high-tea's en -dinertjes waren jullie er al die jaren voor me, allemaal op jullie eigen manier. Krista, vanaf de middelbare school zijn we ieder met een heel andere weg, al vallend en opstaand, *spelend als onschuldige herten*, samen volwassen geworden. Zo fijn om bij iemand te zijn, die het werkelijk niet boeit of ik promoveer of niet, als ik maar doe waar ik gelukkig van word. Wilja, ik heb je mogen leren kennen tijdens de studie psychologie en sindsdien hebben we elkaars life events mogen vieren. Jouw no-nonsens mentaliteit, directe eerlijkheid en mij af en toe een goede spiegel voorhouden, is zo kostbaar gebleken om

niet in *ontmoediging* te verdrinken. Aaltsje, tijdens het *buffelen* in de GZ-opleiding hebben wij onze band gesmeed. Toen ik ging promoveren was daar op het werk altijd het vriendelijke gezicht van Anne Marleen: ruimte gevend en zo een andere kijk op het leven. Bedankt voor je rust en het me vertragen om niet te vergeten, *welke zon er al door de wolken kwam*. Samen lesgeven aan aankomend talent in de GZ-opleiding was meer dan top; met jou is werken ontspannend. En daarna kwam Lisanne, terwijl we samen onze promoties aan het afronden waren en het niet konden laten om nog een opleiding te beginnen. Meiden, for life, wat mij betreft!

En dan de belangrijkste app-groepen in mijn telefoon in de afgelopen jaren. De naam "Waanzin-proef" app groep zegt alles wat het is. Een zo belangrijke plek; samen de waanzin die ons overkwam geridiculiseerd en gevierd dat we elkaar mochten hebben om zelf niet ook door te draaien. Edo, José, Lisanne en Marjolein, we have survived en dat was mij niet gelukt zonder jullie humor, jullie zorg voor mij en het mogen zorgen voor jullie. Kwetsbaarheid ontvangen om zo te mogen helpen, is een belangrijk geschenk. Thanks lui! Tja, en dan de app groep die eigenlijk niet met goed fatsoen in een dankwoord kan worden genoemd. Maar ik doe drie meiden te kort, als ik het niet doe. Dus daar gaan we: Anke, Charlotte en Esther, de "Onderkantjes" app-groep kan geen betere naam hebben dan deze. Het is zo dichtbij, zo veilig, zo ontwapend wat ik met jullie mag delen. Jullie hebben mij een enorm welkom gevoel gegeven, een ruimte waar ik in alle vrijheid heb mogen helen en weer heb durven leren. Door jullie ben ik klaar om mijn vleugels met jullie uit te slaan. We gaan zulke mooie dingen doen, in een *speeltuin* op het werk en daarbuiten; ik weet het zeker. Dank je wel lieverds!

Wie wil ik daar straks achter me hebben staan voor morele steun? Natuurlijk zijn twee bijzondere vriendinnen mijn paranimfen. Lieve Aaltsje, jij bent mijn voorbeeld van en vakidioot zijn en het hebben van een gebalanceerd leven tussen werk en privé, waarbij er tussen beide ook een verbinding mag en kan zijn. We werken met zulke verschillende doelgroepen, maar onze basishouding in het werk is gelijk. Niet alleen evidence-based werken en een bijdrage willen leveren aan de ontwikkeling van het vak telt, maar ook dat dat met een bepaalde, mogelijk christelijk gemotiveerde, integriteit dient te gebeuren. Hiervoor was jij er altijd als klankbord. Dank je wel daarvoor. Gekke Lisanne, mijn prettig gestoorde vriendinnetje, je hebt me geraakt met jouw puurheid, open blik voor echt alles en dan ook nog hoe gekker hoe liever. Ik kan niet anders dan bij jou kwetsbaar zijn en

nooit heb je daarbij de gelijkwaardigheid doorbroken. Wat ben jij loyaal zonder je kritische houding tegenover mij te verliezen en wat kan jij streng zijn. In mijn hoofd *zingt* het maar door: "Ellen, jij zult dit vieren"! Ik ben zo blij, dat jij dit afrondingsritueel ten volle hebt bewaakt. Bedankt lieve schat.

Lieve pappa en mamma, wat een kracht hebben jullie mij voor het leven meegegeven. Het rotsvaste vertrouwen, dat ik altijd heb mogen ontvangen, staat als een *huis*. Laat het een *troost* zijn: bescherming heb ik niet meer nodig, door jullie vertrouw ik op mezelf en kan ik me waar nodig zelf beschermen. Wat ben ik gezegend met jullie. Liefde is een machtig wapen. Eeuwig dankbaar ben ik voor dat cadeau. Johan en Anneke, jullie zijn een top broer en top zus. Zonder oordeel werd er elke keer met interesse gevraagd naar hoe mijn promotie ging. We hadden het erover wanneer ik het nodig had, we lachten wanneer ik wat te vieren had en we zwegen wanneer ik even afstand wilde. Meer had ik niet nodig. Dank jullie wel.

Lieve, lieve Niels, jij was de laatste tijd ook druk bezig met mijn dankwoord. Gekscherend opperde jij de volgende woorden: "Dank je wel Niels, love of my life, ster aan mijn horizon". Tuurlijk schat; staat genoteerd. Maar even serieus. Jij biedt zo veel meer dan troost, zoveel meer dan zwerven door het leven, zoveel meer dan spelend herinneringen maken, zoveel meer dan een aanmoediging, zoveel meer dan een zorgeloos bestaan. Niels, ja, jij bent de liefde van mijn leven, jij bent mijn alles, jij bent mijn thuis. Ik hou van je!

Het is klaar, het is af, het is verwerkt, het kan gevierd worden. Thomas, voor jou een thuis mogen zijn, is mijn feestje. Vanaf nu alleen voor jou:

*O, give me a home where the buffalo roam,
Where the dear and the antelope play,
Where seldom is heard a discouraging word,
And the skies are not cloudy all day.*

Curriculum vitae

Ellen Nobel werd op 30 oktober 1983 geboren in Groningen. Ze groeide op in Blokzijl en vervolgens in Coevorden. In 2011 behaalde ze aan de Scholengemeenschap de Nieuwe Veste in Coevorden haar Vwo-diploma en rondde in 2006 haar studie psychologie af aan de Rijksuniversiteit Groningen met zowel het hoofddoctoraal klinische psychologie als het hoofddoctoraal ontwikkelingspsychologie. Ze schreef een onderzoeksscriptie over gedragsregulatie van kinderen met ADHD. Tijdens haar studie was ze verder voorzitter van de Faculteitsraad en werkte ze als testleider mee aan het TRAILS Clinical Cohort onderzoek van Accare.

Ellen ging in 2006 als psycholoog werken bij Accare Universitair Centrum voor Kinder- en Jeugdpsychiatrie in Groningen. Ze startte binnen het team psychodiagnostiek en werkte daarna binnen het zorgprogramma ADHD en gedragsstoornissen. In 2011 kreeg ze haar registratie Gezondheidszorg(GZ)-psycholoog en startte ze haar promotie-onderzoek, terwijl ze bij Accare werkzaam bleef als clinicus. In 2014 startte ze als docent en hoofddocent bij Opleidingsinstituut PPO (Groningen), waar ze zowel bij de GZ-opleiding Volwassenen en Ouderen als bij de GZ-opleiding Kinderen en Jeugd meerdere blokken ontwikkelde en doceerde over ontwikkelingsstoornissen (ADHD en gedragsstoornissen in het bijzonder) en over neuropsychologische diagnostiek. Vanuit een gevoel van maatschappelijke verantwoordelijkheid met betrekking tot de beeldvorming van ADHD heeft Ellen geregeld bij kerken, scholen en ouderverenigingen presentaties gegeven over ADHD en de gevolgen daarvan in het dagelijks leven van een kind en zijn of haar ouders.

In 2017 is Ellen de opleiding tot Klinische Neuropsycholoog (KNP) bij RINO Utrecht gestart. Ten behoeve van deze opleiding heeft ze gewerkt bij het specialistische ziekenhuis Kempenhaeghe (Heeze) met kinderen en volwassenen met epilepsie (2018-2019) en bij GGZ Eindhoven (GGzE) binnen de afdeling NESTOR neuropsychiatrie ambulant (2019-2021). Verder is ze hoofdauteur van het hoofdstuk "Neuropsychologisch onderzoek" van de Richtlijn Epilepsie vanuit de Nederlandse Vereniging voor Neurologie en heeft ze meegeschreven aan semiwetenschappelijke publicaties over epilepsie. Ellen heeft haar KNP-opleiding in augustus 2021 afgerond en werkt nog altijd als klinisch neuropsycholoog bij GGzE afdeling NESTOR neuropsychiatrie ambulant. Naast haar klinische werk zet zij zich daar specifiek in voor het implementeren van wetenschap en innovaties in de klinische praktijk. Tevens is Ellen haar opleidingstaken bij PPO Groningen

aan het afbouwen en is als docent actief geworden binnen RINO Utrecht. Daar doceert ze over neuropsychologie bij kinderen en jeugdigen en over hoe neuropsychologisch onderzoek een vertaalslag kan krijgen in een behandelplan.

Ellen is verloofd met Niels Holtrop, zij wonen met hun zoontje Thomas (2020) in Maastricht.