Preschool children with ADHD symptoms and behavioral problems
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Chapter 6

Summary and general discussion
After a brief description of the aims and the design of my study, this chapter provides a summary of my main findings. Moreover, I discuss implications for clinical practice and future research, as well as study limitations.

**Aims**

The first aim of this thesis was to analyze whether parental ratings of child behavior correspond or discrepant, in order to improve the adequate use of these ratings in clinical practice and in research. Subsequently, we investigated whether behavioral parent training under routine care conditions reduces disruptive behaviors in preschool children and improves parenting skills, and whether improvements in child and parental behavior were related. Finally, we examined the effectiveness of pcIT in comparison with methylphenidate in those referred preschool children who had remaining behavior problems at home, after previous treatment with behavioral parent training.

**Study design**

We conducted our studies in the naturalistic setting of our outpatient mental health clinic in Groningen, The Netherlands. For the first study into correspondence and discrepancy of parental ratings on child behavior, data were collected in a clinical and a nonclinical sample. Participants in the clinical sample were referred preschool children aged between 2.7 and 6 years and their parents. The non-clinical sample consisted of 121 control children (aged between 2.6 and 6 years) and their parents, recruited from school and child care centers in the northern part of the Netherlands.

All children in the clinical sample received a regular diagnostic evaluation and, in addition, the parents were given psycho-education in which treatment options were discussed. Behavioral parent training was advised as the first treatment of choice. In case parents indeed started parent training, they were asked to participate in the second and third study, regarding changes in child behavior and parenting skills after parent training, and the relation between these. All parents’ and teachers’ assessments were collected as part of routine clinical care.

If children had residual behavior problems at home after behavioral parent training, parents were asked to participate in the fourth study, a randomized controlled trial, in which we compared the effectiveness of pcIT and methylphenidate. Also, children with remaining
disruptive behaviors after previous parent training outside our outpatient clinic could participate. Only children who had already received treatment with PCIT in the last year, ongoing treatment with psychotropic medication, or a major-medical condition that would interfere with involvement in the study were excluded. We included 35 preschool children in this study.

**Main findings and their clinical relevance**

**Predictors of discrepancies between fathers and mothers in rating behaviors of preschoolers**
In the clinical sample, mothers rated the severity of the child’s disruptive behaviors significantly higher than did fathers. Discrepancy between parents was not predicted by child factors or interparental differences in psychopathology, but was predicted by interparental differences in parenting stress. This effect was significantly larger in the nonclinical sample than in the clinical sample. High maternal parenting stress was related to larger differences between parental ratings of externalizing behavior problems, while high paternal parenting stress was associated with smaller differences. If parents experienced similar levels of parenting stress, they agreed more on ratings of their child’s disruptive behaviors.

Our findings highlight the necessity to consider levels of perceived parental stress in parents when using rating scales for clinical purposes, especially when parents differ in their ratings of their child’s behavior. For diagnostic procedures, the value of questionnaires may be limited in general, but especially when only the mother is used as informant. Clinicians should evaluate and interpret the outcomes of rating scales in the context of a multi-informant strategy, including a clinician-based severity rating.

**Children’s response to first-step behavioral parent training and parental predictors of outcome**
While parents reported no changes of disruptive behaviors in the period between initial assessment and start of the treatment, fathers and mothers reported significant and large improvements of children’s behaviors after parent training. Moreover, also teachers indicated improvement of child behavior. In addition, parents experienced fewer child behaviors as troublesome after parent training. Although treatment attendance was high, about one-third of the parents terminated the treatment before the last session, mostly because they could not manage to organize participation on a regular basis.
We found that the pre-treatment level of behavior problems was the most important predictor of treatment outcome, while parental factors only played a small role. Higher scores of disruptive behaviors before treatment were associated with an elevated level of remaining problems after parent training. Behavioral parent training was most effective in children of mothers rating themselves as less adequate in disciplining before treatment. Furthermore, mothers perceived more behavior problems after treatment in case fathers were currently more problematic alcohol users. We found no association between parental ADHD, parental antisocial behavior, nor maternal sense of parenting competence and treatment outcome. Based on our findings, clinicians are recommended to advise behavioral parent training as the first-step treatment for young children with disruptive behaviors in routine clinical care. Furthermore, clinicians are recommended to put additional effort in motivating and facilitating parents to actually participate in behavioral parent training. Former studies had shown that a brief motivational intervention enhanced parents’ motivation to start and attend behavioral parent training sessions (Chacko, Wymbs, Chimiklis, Wymbs, & Pelham, 2012; Nock & Kazdin, 2005). To temper unrealistic expectations, clinicians should discuss beforehand that behavioral parent training may decrease, but not necessarily completely eliminate disruptive behaviors of the child.

Changes in parenting after behavioral parent training

After behavioral parent training, mothers’ self-reports of discipline practices and parenting sense of competence significantly changed over time after behavioral parent training. Maternal ratings of their discipline practices in the clinical sample did not differ anymore from maternal ratings in the nonclinical sample. Fathers reported no improvements in the use of discipline practices. Despite this lack of effect on parenting behaviors, in both fathers and mothers, a reduction in authoritarian, emotional, and harsh parenting after behavioral parent training was associated with improvement of the child’s disruptive behaviors.

While both fathers and mothers showed significant improvements in parenting sense of competence after behavioral parent training, these were not associated with improvements in child behavior. Moreover, after the parent training parents still experienced a significantly lower parenting sense of competence than parents in the nonclinical sample. Although their child’s behavior problems had improved, the remaining problems apparently were still demanding for both parents.

Thus, inadequate disciplining practices in mothers may improve through behavioral parent training, resulting in fewer behavior problems in children. Furthermore, both fathers
and mothers may experience an increased sense of parenting competence regarding their troublesome child after parent training. They may feel more comfortable in their parenting role even when their children’s’ behavior problems remain challenging.

Our results regarding the benefits of behavioral parent training can be used to motivate parents to participate in this treatment as the advised first-step treatment. Knowledge on the content of the treatment and possible effects on disruptive child behavior may positively influence parental views of the treatment and enhance parental motivation if needed (Koerting et al., 2013).

Effectiveness of PCIT versus methylphenidate as a second step intervention
In preschool children who did not benefit sufficiently from behavioral parent training as a first-step intervention, our subsequent randomized controlled trial indicated that methylphenidate was superior over PCIT regarding the primary outcome of intensity of parent-rated disruptive behaviors. Children in both treatment arms improved significantly over time in intensity and number of behavior problems, while changes over time of children in the care as usual treatment were non-significant. No statistically significant interaction effect of time and treatment group was found for ADHD symptoms. However, these symptoms only reduced significantly over time after methylphenidate, but not after PCIT.

As a consequence of the small sample size, this study should be seen as a pilot study rather than as a hypothesis testing study. Moreover, our high dropout rates in both treatments, both in starting and continuing treatment, complicated the interpretation of the results. Further research with a larger sample is needed for more definitive conclusions and clinical recommendations. Based on the preliminary results of our small sample study with high dropout rates, methylphenidate may be more effective than PCIT in reducing preschool children’s disruptive behavior. However, treatment with methylphenidate in preschool children is off-label and should be carefully monitored with an individually tailored dose. Evaluation of the necessity of the treatment after a beforehand determined treatment period, and discontinuation of medication, should be attempted regularly.

Limitations

A strength of our studies on changes of child behavior and parenting parameters after behavioral parent training, was the embedding within clinical practice and the use of both mother and father ratings, as well as teacher ratings. However, because of the lack of a control
group, it is possible that factors other than the intervention contributed to the changes in child and parenting behaviors. However, some additional findings underscore our results. For instance, a comparison of our results to changes in child behavior in the period between initial assessment and start of behavioral parent training, demonstrated that during that period no changes were reported. Furthermore, the more independent teacher ratings also showed a positive change in child behaviors. In the studies on changes in parenting parameters, we evaluated our results, before and after behavioral parent training, to a nonclinical sample to measure whether changes came into normal ranges after treatment.

Maybe due to the modest sample size, we only found two out of eight possible parental predictors to be associated to changes in children’s disruptive behaviors after parent training. Also, the sample size, especially regarding fathers, may have affected study power and our negative findings regarding changes in parenting after behavioral parent training and regarding the predictors should also be seen in this light. Furthermore, we only used information from parent-report questionnaires and not from blinded observers. Blinded observational data could have led to less strong results of our behavioral parent training, especially on children’s disruptive behaviors, as has been reported in a recent meta-analysis (Rimestad, Lambek, Zacher Christiansen, & Hougaard, 2016). In this meta-analysis, a small to moderate significant effect was found on children’s conduct problems based on parent ratings (0.44), but not on independent raters (0.31). Effects on negative parenting were found based on both self-reported negative parental style (0.63) and on blinded assessments (0.33, in this meta-analysis (Rimestad et al., 2016). The effects on negative parenting in our study are in line with these results.

To the best of our knowledge, we conducted the first randomized controlled study directly comparing a behavioral and pharmacological treatment as second-step interventions for preschool children with disruptive behaviors. As far as we know, such clinically important direct comparisons have not yet been done in preschool children. Moreover, we had a comparison group of comparable children who were not involved in any of the two treatments of our trial. Despite these strengths, a major limitation of our randomized controlled study has been that, because of many barriers such as unwillingness in parents to participate in one of both treatment arms and aversion of clinicians to refer children to the trial, we only managed to include a number of 35 families in total. Therefore, our sample may not be representative of clinical cases in general, which limited the generalizability of our findings. More empirical support is needed from studies with larger and more representative samples.
General discussion

A randomized controlled trial under routine care conditions: obstacles and challenges
To constitute a randomized controlled trial under routine care conditions is a challenging task, especially when two completely different treatment options are involved. In our trial, before randomization, parents had to agree to (possibly) participate in a psychological intervention for themselves or a pharmacological treatment for their child. More than 72% of the eligible families declined because one or both treatment arms was not acceptable for them. Parents had a pronounced preference for treatment with medication or for non-medical treatment. Before randomization, we informed parents about the pros and cons of treatment with medication and PCIT to enhance parents’ knowledge in order to facilitate a balanced decision-making process. Most parents of eligible children were able to make such a decision. Moreover, even if families consented to randomization, but were not allocated to their preferred treatment, there was a considerable risk for treatment dropout.

The external validity of a randomized controlled trial may be limited when, so few families are willing to participate. Funding organizations and researchers should look beyond the qualities of only randomized trials to build on efficacious treatments to children with psychiatric disorders and their families (Hoekstra, 2017). A patience preference trial may have been a viable alternative, providing better opportunities to enroll enough eligible families, increase treatment attendance, and make results generalizable. In such a trial, participants with a strong preference for a specific treatment are allowed their desired treatment, and those with no strong preference are being randomized (Hoekstra, 2017).

Enrollment in treatment and treatment dropout
Not only in our randomized controlled trial but also in our studies into children’s response to behavioral parent training, about one third of the parents for whom parent training was indicated never started this treatment, and about one third of the parents who did engage in parent training dropped out of the treatment. This finding is in accordance with results of a recent review regarding engagement in behavioral parent training (Chacko et al., 2016), showing that at best half of the eligible parents completed treatment. At least 25% of families for whom parent training was indicated never started, and 26% started treatment but dropped out of treatment before the last treatment session (Chacko et al., 2016). In a study on barriers to effective early parenting interventions for parents of preschool children with disruptive behaviors, parents highlighted disengagement if they experienced the program as
unhelpful in relation to their expectations and perceptions of progress (Smith et al., 2015). A study on a parent training prevention program for conduct problems in preschoolers showed that parents with lower incomes and less social support were less likely to be enrolled (Baker, Arnold, & Meagher, 2011). Clinical experience suggests that suboptimal parental planning and organizational skills in parents with ADHD may also play a role in enrollment to and continuation of behavioral parent training. For these parents, it may be harder to organize to come to the treatment sessions.

Except negative expectations of progress, organizational deficits, and social-economic problems, lack of parental-confidence and self-efficacy may also contribute to less motivation and problems with starting and continuing parent training (Smith et al., 2015). In our study, we found parenting sense of competence to be significantly lower in parents who received parent training compared to parents of nonclinical children. Furthermore, parenting sense of competence increased after behavioral parent training, but did not reach levels of parents of nonclinical children. Therefore, in line with recommendations of Chacko (Chacko et al., 2014), part of psycho-education with parents should be a good explanation of how behavioral parent training works and a discussion of its potential evidence based benefits. Furthermore, effective behavioral parent training should start with a preparation phase in which possible practical and perceived obstacles into engaging in the parent training are identified and addressed, and expectations on the effectiveness of the treatment are discussed. Concrete goal setting and progress monitoring should also be part of this preparation phase (Chacko et al., 2014). As results of such an intervention are not yet available, further empirical research is needed to investigate the effects of removing barriers on treatment engagement and outcome (Daley et al., 2017).

Another way to enhance treatment attendance could be to decrease the amount of treatment sessions and focus on the probably most helpful components of the intervention. Behavioral parent training consists of various elements but until now we do not know which modules are most effective for whom (Daley et al., 2017). Therefore, therapists now typically offer complete treatments to parents instead of effective parts in isolation (Daley et al., 2017). Future studies should investigate which parts of treatment programs are most effective for behavior problems to strengthen treatment effects and enhance participation in effective treatments with tailored treatment programs.
Participation of fathers
Although most of the fathers participated in the behavioral parent training in our parent training studies, and attended on average only one session less than did mothers, their parenting practices hardly changed after parent training. One could therefore question whether participation of fathers is necessary. A meta-analysis of father involvement in parent training for disruptive child behaviors (Lundahl, Tollefson, Risser, & Lovejoy, 2008) showed that results on adequate parenting practices and on child behavior directly after treatment were superior in studies with fathers who were involved in parent training compared to studies not involving fathers. Also in that meta-analysis, mothers benefitted more ($d = 0.84$) regarding change in parenting behaviors directly after treatment than did fathers ($d = 0.53$) (Lundahl et al., 2008).

Perhaps participation of fathers plays a mediating role in the positive results regarding changes in maternal parenting practices and child behavior. One could imagine that mothers, most of the time the primary caretaker and spending most of the time with the child, feel supported when fathers also participate in parent training. Further research is needed on this subject and on how to enhance and consolidate positive changes in paternal negative parenting practices. A study on a parent training especially developed for fathers of school-age children with ADHD (Fabiano et al., 2012) showed promising results on fathers’ negative verbalizations and praise directly after treatment, but unfortunately these results did not generalize to the mothers who did not participate in parent training, nor did they remain one month after treatment.

Second step treatments in preschool children
It is not yet clear which treatment to advice as the preferred second step for young children with disruptive behaviors who have remaining behavior problems after parent training. Although methylphenidate appeared more effective for behavioral problems in preschool children who did not benefit enough from behavioral parent training, clinicians should be cautious in prescribing methylphenidate for this age-group as it is off-label and long-term effects especially in children with comorbid ODD symptoms have not been established (Riddle et al., 2013). Furthermore, not all parents might prefer pharmacological treatment as a second step, as was the case in 70% parents of young medication naïve children with ADHD.

Alternative behavioral second step treatments after weekly outpatient manualized parent training should certainly be considered, especially when disruptive behaviors are still the main problem after behavioral parent training, and not core ADHD symptoms. Clinicians
could think of parent training involving the child such as PCIT, home-based behavioral parent training, or behavioral treatment at school, and/or treatment of parental problems and family support. PCIT may be useful in case child noncompliance is the main residual problem, or when participation of the child enhances parental motivation to further participate in parent training, or for parents who profit more from in vivo-coaching than from practicing parenting skills at home. Home-based parent training could be a solution for parents who were not able to apply learned parent training techniques at home.

Finally, some studies have shown that pre- or additional treatment of parental psychopathology may be beneficial to treatment outcome on child behavior, but findings are mixed. For example, a study on treatment of maternal depression showed that externalizing behavior problems decreased in children of mothers in whom the depression remitted but increased if mothers who remained depressive (Wickramaratne et al., 2011). Also, an integrated treatment study targeting parenting and depressive symptoms in mothers of school-aged children with ADHD showed a small to moderate effect relative to behavioral parent training alone on observed negative parenting, observed child deviance, and maternal depressive symptoms (Chronis-Tuscano et al., 2013). However, a study on treatment of maternal ADHD in addition to parent training has found that treatment of maternal ADHD did not improve the effectiveness of parent training on children’s disruptive behavior problems (Jans et al., 2015), nor did a recently published pilot study (Chronis-Tuscano, Wang, Woods, Strickland, & Stein, 2017).

**Parental ADHD and behavioral parent training**

In our behavioral parent training study, elevated levels of parent-rated parental ADHD, compared to the general adult population (Barkley & Murphy, 1998), were measured in one third of the participating parents. However, no significant association was found between parental ADHD and outcome on child behavior after behavioral parent training. This finding is in line with a recent study on the influence of parental ADHD on effects of parent training for preschool children with ADHD (Forehand et al., 2017), but in contrast to two other studies which showed that pre-treatment maternal ADHD predicted less improved mother-reported disruptive behaviors in children after behavioral parent training (Chronis-Tuscano et al., 2011; Sonuga-Barke, Daley, & Thompson, 2002). In one of these two studies, the relation between maternal ADHD symptomatology and diminished response to behavioral parent training was mediated by less change in observed maternal negative parenting; mothers with
elevated ADHD symptoms were less able to reduce their use of negative parenting behaviors (Chronis-Tuscano et al., 2011).

We found no significant association was found between parental psychopathology and outcome on child behavior after behavioral parent training, but we did find that mother-reported authoritarian, emotional, and harsh parenting were reduced to normal levels, i.e. equal to mothers of non-clinical children. Furthermore, we found an association between reduction of negative parenting practices and improvements in child behavior, in line with a previous study suggesting that success of behavioral ADHD treatments is related to the degree of change in negative/ineffective parental discipline (Greenhill et al., 2003; Greenhill et al., 2001; Hinshaw et al., 2000). Perhaps the parents with elevated ADHD levels in our study increased their ability to inhibit and change negative reactions during parent training. Our protocol did contain specific treatment elements that may have been beneficial for those parents, such as skill-based practice between and within treatment sessions, instructions regarding home-work assignments adjusted to the specific needs of parents, highly structured treatment sessions and the stimulating and rewarding attitude of the therapist towards changes in parent behavior.

Further research is needed to expand our knowledge on the mechanisms underlying a possible relation between treatment components and parental characteristics on the one hand, and positive results on harsh parenting after treatment on the other hand, in order to improve parent training programs. More knowledge on how to decrease harsh parenting for parents with particular characteristics, i.e. ADHD, may result in less behavior problems in children after behavioral parent training.

**Potential risk of early labeling**

Another issue of discussion is the assessment and treatment of disruptive behaviors in preschool children versus potential risk of early labeling. There is some concern in society that early diagnoses may lead to stigmatization, fueled by concerns that this may indicate that something is wrong with the child rather than within the family or with the parental parenting practices resulting in children’s disruptive behaviors. If a child behaves normally much of the time but shows disruptive behavior only in particular situations, one may think that the child is deliberately acting out or that the parents do not have sufficient control (Hinshaw, 2008). According to reviews on stigmatization of ADHD, there is significant stigmatization of children, adolescents, and adults with ADHD: parents and teachers of children with ADHD experienced negative biases regarding the academic skills of children.
with ADHD and parents felt they were negatively evaluated themselves (Lebowitz, 2016; Mueller, 2012). In families of school-aged children with ADHD, high parental stigma was related to negative parenting (Mikami, 2015). Stigma may also be associated with decreased treatment adherence and efficacy (Mueller, 2012).

Therefore, clinicians should be critical to the need and purpose of an ADHD diagnosis. One could argue that parents of treatment naïve preschool children, referred because of disruptive behaviors, should be offered a behavioral parent training first. In most behavioral parent training programs, a formal diagnosis is not needed. In case the disruptive behaviors do not, or not sufficiently, change after behavioral parent training, diagnostic assessment might be the next step. t. Regular re-evaluation of diagnoses should also be part of clinical care as symptoms may change over years (Bufferd, 2011).

Parent ratings versus blinded outcome measures in research
A last point of discussion is the use of parent-rated outcomes in studies on treatment effectiveness. Blinded outcomes are the gold standard nowadays, as participants of treatment studies may be “biased” when using self-reports as they have invested in the treatment and may want it to be a success (Daley et al., 2017; Sonuga-Barke et al., 2013). Results, based on blinded raters, were less pronounced or absent regarding conduct problems and ADHD symptoms, compared to unblinded raters (Daley et al., 2014; Rimestad et al., 2016; Sonuga-Barke et al., 2013). But is it problematic that parents are biased in rating their child’s behavior after behavioral parent training? It is very well possible that changes in child behavior reported by parents merely reflect changes in parental perceptions rather than actual improvements in the behavior of the child (Daley et al., 2017). However, main targets of behavioral parent training are that parents consider the behaviors of their child as less challenging and feel more comfortable and competent in handling their child. Therefore, their opinion on changes in the child’s behavior could be considered as the most important one.

Main conclusions

We end with a brief overview of our main findings. Regarding discrepancy between parents’ ratings on the severity of the child’s disruptive behaviors, this appeared to be predicted by interparental differences in parenting stress. As a first-step intervention, behavioral parent training under routine care conditions was associated with improvements in
disruptive behaviors in preschool children, maternal parenting skills, and sense of parenting competence in both parents. Positive changes in parenting, particularly less over-reactive parenting behaviors, were related to a decrease of disruptive child behaviors. Finally, as second-step treatment, methylphenidate was more effective than PCIT in children with remaining disruptive behaviors after parent training.

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