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### Assessment and treatment of planning skills in adolescents with ADHD

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# **Chapter 1**

## **General introduction**

## ATTENTION DEFICIT HYPERACTIVITY DISORDER

Attention Deficit Hyperactivity Disorder (ADHD) is one of the most common psychiatric disorders among children: it affects approximately 5–10% of children and adolescents worldwide (Faraone, Sergeant, Gillberg, & Biederman, 2003; Polanczyk, Silva de Lima, Lessa Horta, Biederman, & Rohde, 2007; Willcutt, 2012) and impacts cognitive and behavioral functioning in academic, family and social contexts. Individuals with ADHD have symptoms of inattention, symptoms of hyperactivity/impulsivity or both. According to the fourth edition of the Diagnostic and Statistical Manual of Mental Disorders (DSM), there are three subtypes of the disorder: the inattentive subtype with mainly symptoms of inattention, the hyperactive/impulsive subtype with mainly symptoms of hyperactivity/impulsivity and the combined subtype with symptoms of both inattention and hyperactivity/impulsivity (American Psychiatric Association, 2000)\*.

ADHD is a neurodevelopmental disorder and symptoms change as a function of age: in adolescence, symptoms of hyperactivity/impulsivity decrease as compared to childhood (Fischer, Barkley, Smallish, & Fletcher, 2005), whilst problems with inattention often remain stable (Biederman, Mick, & Faraone, 2000) or even become more prominent in daily life (Barkley, 2004). As a result, in childhood the combined subtype is most prevalent, while in adolescence the inattentive subtype is more prevalent (Willcutt, 2012). In clinical samples 'pure' ADHD without comorbid disorders is rare: approximately 62% of children with ADHD below 19 years have at least one comorbid disorder, whereas 34% even have two or more comorbid conditions (Yoshimasu et al., 2012). The most prevalent comorbid disorders are oppositional defiant disorder (ODD), conduct disorder (CD), anxiety- and mood-disorders, adjustment disorder and substance use disorder (Yoshimasu et al., 2012). Also, more boys than girls have ADHD (3:1 respectively; Szatmari, Offord, & Boyle, 1989; Tuithof, Ten Have, Dorsselaer, & De Graaf, 2014) and girls appear to have less severe inattention, hyperactivity and impulsivity, but greater intellectual impairments than boys with ADHD (Gershon, 2002). In addition, girls tend to have more internalizing comorbid disorders than boys, whilst boys with ADHD are at higher risk for externalizing psychiatric comorbidities than girls (Yoshimasu et al., 2012). Taken together, the group of individuals with ADHD is a heterogeneous one.

Because the expression of ADHD is highly heterogeneous within the di-

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\* In this thesis subtypes of ADHD are described using DSM-IV criteria.

agnosed population and across time, a problem with using the DSM-IV subtypes was that they are often unstable or changing over time (Tannock, 2013). Therefore, in the most recent edition of the DSM, the DSM-5, the three subtypes of ADHD are replaced with presentations of the disorder. These presentations are used as 'specifiers' to classify the current manifestation at the time of assessment instead of presenting the subtypes as stable entities (Tannock, 2013), and map directly to the prior subtypes: the inattentive presentation with mainly symptoms of inattention, the hyperactive/impulsive presentation with mainly symptoms of hyperactivity/impulsivity and the combined presentation with symptoms of both inattention and hyperactivity/impulsivity (American Psychiatric Association, 2013).

## UNDERLYING PATHWAYS OF ADHD

In the past, several models have been proposed to explain the cause of ADHD as a simple, core deficit, for example the inhibition model or the motivational model (e.g., Barkley, 1997; Sagvolden, Aase, Zeiner, & Berger, 1998). As ADHD is such a heterogeneous disorder, none of these models can fully explain the disorder and models based on multiple pathways to ADHD have gained popularity, for example executive functioning, motivation, and temporal processing as underlying deficits towards ADHD symptomatology (e.g., Nigg, Goldsmith, & Sachek, 2004; Sonuga-Barke, 2003; Sonuga-Barke, 2005; Sonuga-Barke, Bitsakou, & Thompson 2010). In this thesis we will focus on two of these dissociable developmental patho-physiological pathways that have been proposed to be associated with ADHD behavior (Sonuga-Barke, 2003; Sonuga-Barke et al., 2010).

The first pathway associated with ADHD behavior, is the executive functioning pathway (Barkley, 1997; Sonuga-Barke, 2003). Executive Functions (EFs) are commonly described as neuro-cognitive processes that enable self-control (Lezak, 1995) and are necessary to maintain an appropriate problem solving set to attain a future goal (Welsh & Pennington, 1988). Examples of EFs are inhibition, working memory, cognitive flexibility and planning, which are best regarded as a set of inter-related but distinguishable domains (Miyake, Friedman, Emerson, Witzki, & Howerter, 2000). It has been shown that individuals with ADHD, compared to typically developing (TD) individuals, have problems with the EFs inhibition (Barkley, 1997), working memory and planning (Willcutt, Doyle, Nigg, Faraone, & Pennington, 2005). Planning is generally defined as the process of formulating a sequence of oper-

ations intended for achieving a goal (e.g., Hayes-Roth & Hayes-Roth, 1979). In general, planning skills develop later in life than for example inhibition, working memory and cognitive flexibility (Best, Miller, & Jones, 2009). Moreover, for adequate planning other EFs are needed, like working memory (remembering the goal to attain and the steps to get there), inhibition (not letting impulses distract from the goal to attain) and cognitive flexibility (letting go of one step of the plan and start with the next step towards the goal), which are therefore considered more basic EFs (Best et al., 2009).

The second pathway associated with ADHD-behavior, is the motivational pathway. Individuals with ADHD appear to have an aberrant motivational style (Barkley, 1997; Haenlein & Caul, 1987; Luman, Oosterlaan, & Sergeant, 2005; Sagvolden, Aase, Johansen, & Russell, 2005; Sonuga-Barke, 2003). For example, children with ADHD appear less sensitive to reinforcement as compared to controls, prefer immediate over delayed reward and need a higher intensity of reward than controls to show an optimal performance (Luman et al., 2005). Most individuals with ADHD show aberrant functioning in one of the two described pathways, and some individuals in both pathways. However, not all individuals with ADHD show problems with EF or motivation (Sonuga-Barke et al., 2010).

These underlying pathways go through important changes during adolescence (Blakemore & Choudhury, 2006; Fareri, Martin, & Delgado, 2008; McCormack, Brown, Maylor, Darby, & Green, 1999). For example, the risk-taking and reward seeking behavior that is typical for adolescence stems from different motivational and cognitive control systems than during childhood (Fareri et al., 2008). Because some individuals with ADHD already show aberrant motivational functioning during childhood, one can argue that these motivational problems will still be altered during adolescence (Toplak, Jain, & Tannock, 2005). Further, as the prefrontal cortex develops exceedingly in adolescence, EFs of TD adolescents improve markedly (e.g., Best et al., 2009; Huizinga, Dolan, & Van der Molen, 2006). In contrast, in daily life adolescents with ADHD are described to show prominent problems with perseverance, time-management and planning (Barkley, 2004; Wolraich et al., 2005). Also these EF problems have been confirmed on parent-rated questionnaires of EF deficits in daily life (Langberg, Dvorsky, & Evans, 2013; Toplak, Bucciarelli, Jain, & Tannock, 2009). However, the few studies using neuropsychological measures of planning skills in adolescents with ADHD show mixed results: some find deficits whereas others don't (Gau &

Shang, 2010; Gau, Chiu, Shang, Cheng, & Soong, 2009; Qian Shuai, Chan, Qian, & Wang, 2013; Toplak et al., 2009).

Whether a neuropsychological task to assess planning skills measures the same construct as parent-rated questionnaires is debatable: neuropsychological tasks involve standardized procedures that are administered by an examiner and usually assess accuracy and/or response time, whereas these rating measures of EF involve a parent reporting on difficulties with carrying out everyday tasks (Toplak, West, & Stanovich, 2013). In other words, neuropsychological tasks assess the processing efficiency of cognitive abilities, whereas ratings of executive function assess the extent to which the individual is achieving goals in daily life. So even though measuring the same construct, neuropsychological tasks and ratings of executive function assess different aspects of functioning (Toplak et al., 2013). Therefore, to find out on what aspect of planning skills adolescents with ADHD show deficits, it may be important to use neuropsychological tests and complement these with questionnaires. Also, little attention has been paid to differences in planning skills between subtypes of ADHD (but Qian et al., 2013), whilst the additional impulsivity and hyperactivity problems in the combined subtype may cause different problems when planning than only the inattention problems in the inattentive subtype. All in all, more research is needed to know how many adolescents with ADHD actually have planning problems, if there are subtype differences in planning problems and on what aspects of planning they show deficits (**chapter 2**).

## CONSEQUENCES OF ADHD IN ADOLESCENCE

In about 65% of children with ADHD, the disorder persists into adolescence (Biederman et al., 2000, Hill & Schoener, 1996; Tuithof et al., 2014). But also the majority of individuals in remission continues to struggle with a substantial number of ADHD symptoms and high levels of dysfunction by the age of 20 (Biederman et al., 2000). During adolescence, and especially in secondary school, various environmental changes take place that in particular appeal to the EF and motivation pathways. The control and help of parents and teachers diminishes as compared to childhood, while the transition to secondary school increases the need for EF (Evans et al., 2009). For example, in secondary school adolescents have to change class and teacher every hour, are expected to be there on time and bring necessary materials. Adolescents are expected to independently manage planning for long-term

projects, study for tests, and complete and turn-in assignments for multiple classes and teachers on time (Evans et al., 2009). At the same time cognitive demands and the number of homework assignments increase as compared to elementary school. Also, one could imagine that another challenge for adolescents with ADHD who have a preference for immediate- instead of delayed reinforcement, is to pay attention in class or during homework (delayed reinforcement), as it is relatively boring compared to for example, interacting with classmates/friends, the internet or television (immediate reinforcement). It has been shown that planning aspects of EF are highly predictive of school grades in adolescents with ADHD (Langberg et al., 2013) and self-rated motivation is highly predictive of overall impairment in students with ADHD (Dvorsky & Langberg, 2014). As a result, adolescents with ADHD are more likely to fail a grade or drop out of school, have higher rates of suspension, are less likely to attend college, show lower academic and occupational achievement, have lower rates of high school graduation and demonstrate more disruptive classroom behavior than their TD peers (Barkley, Fischer, Edelbrock, & Smallish, 1990; Fisher, Barkley, Edelbrock, & Smallish, 1990).

Next to impairments in school, the adolescent age is a phase in which individuals are at risk for developing psychiatric disorders (Merikangas et al., 2010). Even though this is true for all adolescents, long-term follow-up studies of children diagnosed with ADHD, have found that adolescents with ADHD show increased levels of anxiety, anti-social personality disorder and substance use disorders over and above rates seen in normal controls (Barkley et al., 1990; Biederman et al., 2006; Wolraich et al., 2005). Symptoms of comorbid depression in particular, increase during adolescence to rates that are comparable to adults with ADHD (Bird, Gould, & Staghezza, 1993; Wolraich et al., 2005). In sum, there is an increase in daily life impairments and risk for development of comorbidity in adolescents with ADHD, which emphasizes the need for evidence-based treatments for adolescents with ADHD.

## TREATMENTS FOR ADOLESCENTS WITH ADHD

Based on child studies, in several guidelines for treatment of ADHD in children and youth (e.g., Dutch Multidisciplinary Guidelines for ADHD, National Institute for Health and Care Excellence, and European clinical guidelines for hyperkinetic disorder), stimulant medication or behavioral treatment (mediation treatment aimed at changing the behavior of the child

through training parents or teachers) are recommended, or a combination of both (National Institute for Health and Care Excellence, 2008; Taylor et al., 2004; Richtlijnontwikkeling Trimbos Instituut, 2005). Parents of adolescents prefer psychosocial intervention but have better access to medication (Bukstein, 2004; MTA-cooperative group, 1999a).

Stimulant medication (e.g., methylphenidate) increases the availability of synaptic dopamine, and shows robust short- and middle-term effects reducing the hyperactivity, impulsivity, and inattention characteristic of individuals with ADHD (Volkow et al., 2001). Moreover, psycho-stimulants have direct beneficial effects on cognitive functions of children and adolescents with ADHD, for example on outcomes like memory, reaction time, reaction time variability and response inhibition (Coghill et al., 2014). In the largest treatment study to date in children with ADHD, the Multimodal Treatment of ADHD Study (MTA-study;  $n=579$  children of 9-11 years), 4 treatment groups were compared: community care (i.e. treatment as usual), behavioral treatment, medication management, and a combination of medication management and behavioral treatment. Primary analyses of short-term effects showed that all treatments resulted in reduced core ADHD-symptoms and related problems directly after treatment. Moreover, children that were in one of the two groups receiving medication showed less ADHD-symptoms directly after treatment than the groups receiving community care or behavioral treatment alone (MTA cooperative group, 1999a), with the combined group showing greater effect sizes than the medication only group (Conners et al., 2001).

However, stimulant medication is not effective for every child or adolescent with ADHD as only 70-80% of youth with ADHD benefits from it (Faraone & Buitelaar, 2010). And, even though the direct effects may be beneficial, on the longer term the effects of medication are less clear (Jensen et al., 2007; Molina et al., 2009). Follow-up studies of the MTA-study showed treatment differences dissipated 36 months after pretest, implying that benefits of medicated groups over non-medicated groups had disappeared (Jensen et al., 2007; Molina et al., 2009). Moreover, there may be adverse effects of medication on appetite, sleep and growth (Richtlijnontwikkeling Trimbos Instituut, 2005) and adolescents in particular may want to avoid the associated stigmatization, have feelings of shame about their medication, or can easily forget to take their medication, causing problems in medication adherence (Antshel & Olszewski, 2014). This may be why in adolescence adherence to



stimulants shows a decline as a function of age, such that older children are less likely to be continuing with their medication (Antshel & Olszewski, 2014; Wolraich et al., 2005). Finally, even though medication has positive effects on symptoms of ADHD, the effects on related impairments, like academic and social functioning or comorbid anxiety/depression are less clear (Abikoff et al., 2009; Antshel & Olszewski, 2014; Molina et al., 2009). Especially in adolescence it is therefore important to explore other nonpharmacological treatments instead of, or combined with medication, given their risks in development and non-adherence to pharmacological treatments.

The recommended nonpharmacological alternative is behavioral treatment (Richtlijnontwikkeling Trimbos Instituut, 2005). Behavioral treatment is defined as 'all interventions that employ learning principles to improve ADHD- or ADHD-related behavior, focusing on the child itself or on his/her parent or teacher' (Sonuga-Barke et al., 2013). The Dutch guideline for treatment of children and youth with ADHD, recommend behavioral treatment aiming at parents or teachers to mediate their child's behavior, using basic operant learning principles like contingency management (Richtlijnontwikkeling Trimbos Instituut, 2005). Meta-analyses show that although there are limited effects of behavioral treatments on core ADHD-symptoms of children with ADHD, considerably larger beneficial effects are found on related impairments and comorbid conditions (Sonuga-Barke et al., 2013; Daley et al., 2014).

Barkley and colleagues have investigated effects of behavioral treatment aiming at parents to mediate behavior of their adolescent child with ADHD (Barkley, 2004). They compared behavior training of parents to two types of family therapy that *did* involve the adolescent (one problem solving and communication training, one structural family therapy) in 64 families of adolescents with ADHD. Results showed that after treatment there was significant improvement on parent-adolescent communication, number of conflicts, school adjustment and internalizing and externalizing measures, but without treatment differences between three treatment modalities. Moreover, only 5 to 30% of adolescents showed clinical improvement (using a reliable change index; Barkley, Guevremont, Anastopoulos, & Fletcher, 1992). This is supported by another study in 97 families of adolescents with ADHD and comorbid ODD, comparing two family therapies (problem-solving communication training alone, or combined with parental behavior training). Results showed significant improvement over time, but again no treatment

differences and only 23% of families showed reliable change (Barkley, Edwards, Laneri, Fletcher, & Metevia, 2001). In addition to these sobering results, cessation of treatment was high as 28.1% of the families dropped out of treatment (Barkley et al., 2001). Taken together, even though behavioral treatments through parental mediation can cause beneficial effects in children with ADHD, these treatments appear not as effective in adolescents with ADHD as they are not more effective than family therapy involving the adolescent, few families show reliable change and drop-out rates are substantial (Antshel & Olszewski, 2014; Barkley, 2004; Pfiffner, DuPaul, & Barkley, 2005).

When developing treatments for adolescents, drop-out or cessation of treatment is an important factor to take into account. This is especially the case for adolescents with ADHD, as having an ADHD-diagnosis is an important predictor of treatment drop-out (Johnson, Mellor, & Brann, 2008). A possible explanation for this lack of adherence is the potential absence of intrinsic motivation for treatment. Individuals with ADHD are rather reinforced sooner than later (Luman et al., 2005), whereas treatment is -in most cases- a delayed reward: often treatment itself is not rewarding but the (long-term) effects thereof are. Moreover, adolescents are often sent to treatment by their parents, which can cause onset of resistance (Baruch, Gerber, & Fearon, 1998). Another possible reason for high drop-out rates might be that, in clinical practice, adolescents with ADHD often receive treatments that are developed for children with the disorder (Chronis, Jones, & Raggi, 2006), that mainly aim at mediation treatment through parents or teachers or reward systems using tangible rewards. As explained above these treatments are generally not effective for this age-range (Barkley et al., 1992, 2001). Therefore, for adolescents it is important to develop a nonpharmacological treatment alternative, tailored to this specific developmental phase.

Four issues are salient to adolescence and deserve consideration with respect to treatment development (Smith, Waschbusch, Willoughby, & Evans, 2000). First, adolescents have greater cognitive capacities than children, allowing them to be more self-conscious, better able to analyze their own performance, better able to predict their own behavior ahead of time, and better able to evaluate critically possible available strategies to achieve a goal (Smith et al., 2000). This means that they may have sufficient cognitive capabilities to cooperate with their therapist, set their own treatment goals and make their own choices regarding treatment strategies. Second, adolescents

have a focus on identity formation and the establishment of greater independence and autonomy (Antshel & Olszewski, 2014). Therefore, aiming treatment at their parents or teachers may be less effective (Antshel & Olszewski, 2014; Barkley, 2004; Barkley et al., 1992; Pfiffner et al., 2005). Third, adolescents have a greater reliance on peers as intimate partners rather than simply friends with similar interests. And finally, the transition to middle and high school results in a different daily routine than primary school (Smith et al., 2000). All in all, to develop a treatment for adolescents, it appears best to aim treatment on the adolescent him/herself, his/her own treatment goals to enhance independence/autonomy, and make use of their increased cognitive abilities. Furthermore treatment should be accorded with their daily routine, for example by focusing on problems regarding school- and homework (Antshel & Olszewski, 2014; Smith et al., 2000). We have developed two age-appropriate CBTs for adolescents with ADHD that met these criteria (Boyer, Kuin, Oberink, & Van der Oord, 2014; Kuin, Boyer, & Van der Oord, 2013).

## STUDIES ON TREATMENTS FOR ADOLESCENTS WITH ADHD

As compared to treatment studies in children with ADHD, randomized clinical trials (RCTs) investigating nonpharmacological treatments for adolescents are underrepresented (Daley et al., 2014; Sonuga-Barke et al., 2013); only six RCTs have been published for this age group (Barkley et al., 1992; Evans, Schultz, DeMars, & Davis, 2011; Langberg, Epstein, Becker, Girio-Herrera, & Vaughn, 2012; Molina et al., 2008; Sibley et al., 2013; Vidal et al., 2015). In treatment studies in adolescents with ADHD a dichotomy exists between treatments that are school-based (Evans et al., 2011; Langberg et al., 2012; Molina et al., 2008) and treatments that are clinic-based (Antshel & Olszewski, 2014; Barkley et al., 1992; Sibley et al., 2013; Vidal et al., 2015).

Especially in the United States, school-based approaches are more common (Evans et al., 2011; Langberg et al., 2012; Molina et al., 2008). For example, a school-based treatment called the Homework, Organization, and Planning Skills (HOPS) intervention, consists of 16 sessions, is delivered during the school day, and focuses on improving participants' physical organization (i.e., book bag, binder, and locker) and homework management (i.e., accurate homework and test recording and planning). Initial sessions occurred twice weekly (20 minutes each) and then moved to once a week for the last 6

sessions. HOPS showed superior effectiveness on planning skills and homework completion compared to a waitlist control group, with maintenance of effects to three-month follow-up ( $n=47$ ; Langberg et al., 2012). Another school-based intervention is the Challenging Horizon Program (CHP); an intensive after school program, focusing on planning- and organizational skills but also on social functioning for adolescents in middle school, that includes 2 meetings per week for 2:15 hours per meeting for a period of five months (Evans et al., 2011). Five months of CHP was more effective than community care on reduction of parent-rated hyperactivity/impulsivity and teacher-rated academic impairment (Evans et al., 2011). Molina et al. (2008) also investigated CHP in 10 weeks for 2 days a week (2 hours per session) and randomly assigned adolescents to either CHP or community care (total  $n=23$ ). Results showed few statistically significant group or time effects or group by time interactions; only internalizing problems improved over time and improved more in the CHP group (Molina et al., 2008). Improvements shown by Evans et al. (2011) and Molina et al. (2008) were less than when the CHP program was administered during the full academic year (Evans et al., 2005; using a quasi-experimental design), suggesting that a longer duration of treatment may be necessary for optimal improvement (Evans et al., 2011).

In the current European mental health care system, funding mental health care in schools is challenging and mainly takes place in mental health care institutes (World Health Organization, 2008). Further, because these school-based treatment programs are quite intensive, the feasibility of implementation and dissemination of these programs in schools using the existing infrastructure is currently limited (Kataoka, Rowan, & Hoagwood, 2009). Therefore, to make implementation and dissemination of treatments for adolescents with ADHD possible in the current European infrastructure, as for now clinic-based treatments are a first step toward treatment development for adolescents with ADHD. Notwithstanding that school-based programs may be developed in the future.

Clinic-based treatments are however less studied in adolescents with ADHD (Antshel & Olszewski, 2014), and are limited to two studies in the US and one in Spain. An RCT investigating the effects of clinic-based group CBT in comparison to a waitlist control group ( $n=119$ ) showed that ADHD symptoms and functional impairment significantly decreased with large effects sizes for adolescents receiving group CBT as compared to the waitlist control group (15–21 years of age; Vidal et al., 2015). The group CBT consisted of 12

manualized sessions and used components of motivational interviewing. It started with psycho-education, followed by an impulsivity module and a planning strategies and attention module (Vidal et al., 2015). Another pilot-study RCT investigated the effects of the clinic-based Supporting Teens' Academic Needs Daily (STAND) program, of which the goal is to train parents to implement behavior interventions that are typically administered at school, for example time management, homework completion and note-taking (Sibley et al., 2013). The pilot study ( $n=36$ ) took place in a clinic and results showed that adolescents (11–15 years) who received STAND showed greater improvements in parent-rated and observed academic and ADHD- and ODD-indices than a treatment as usual (TAU) group (Sibley et al., 2013). In addition, one non-randomized study ( $n=68$ ) investigated pre- to post-treatment effects for a clinic-based cognitive behavioral therapy (CBT) in a sample of adolescents with ADHD (14–18 years). This CBT included components of motivational interviewing, practice, review, and repetition of learned skills, and six modules regarding psycho-education, reducing distractibility and procrastination, cognitive restructuring strategies, improving communication skills and anger management (Antshel, Faraone, & Gordon, 2012). The largest effect sizes were observed for number of weekly missed classes, school tardiness, stimulant medication doses, parent-reported externalizing behaviors, parent-reported inattention symptoms, and teacher-reported inattention symptoms (Antshel et al., 2012).

Even though the results of these clinic-based treatments are promising, studies had small sample sizes (Sibley et al., 2013), designs lacked a control group (Antshel et al., 2012) or comparisons were made to a waitlist (Vidal et al., 2015), or to a TAU group (Sibley et al., 2013). Without an active control group, effect sizes may be high but could also be the result of non-specific treatment effects, like for example attending treatment sessions, visiting a mental health care institute or therapist, engaging in procedures directed at behavior change. Given the available research and evidence in this area, what is needed is an RCT comparing a developmentally appropriate treatment for adolescents with ADHD to a control treatment to control for non-specific treatment effects in a large sample. Also, by comparing to a control treatment without the proposed 'active' element, inferences can be made about the treatment mechanisms causing change. And to make implementation and dissemination possible within the current existing mental health-care in the Netherlands, these treatments need to be short term (because of

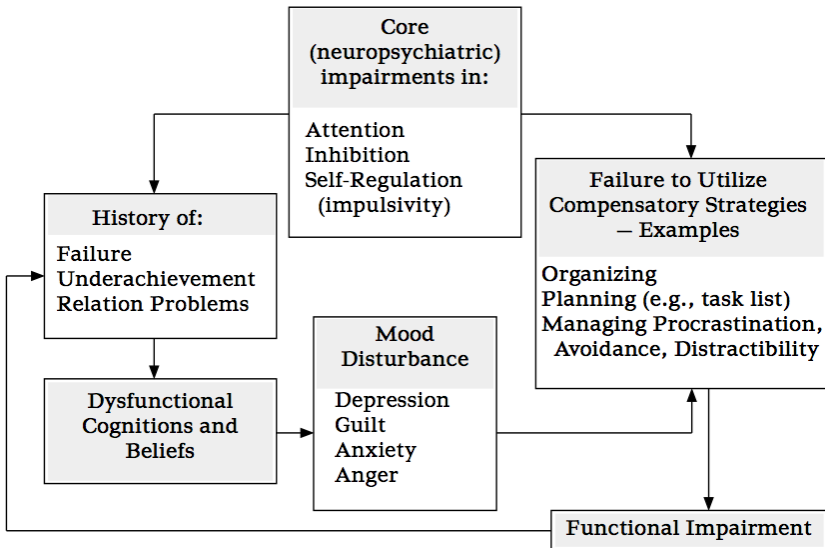
limited funds for mental health care and reimbursement for clinics for care for adolescents with a diagnosis of ADHD) and clinic-based.

## TWO NEW CBTs FOR ADOLESCENTS WITH ADHD

The growing autonomy and independence in adolescents and the demands inherent to secondary school increase the need for EF, causing inevitable planning deficits in the daily lives of adolescents with ADHD. Therefore, treatment aiming at enhancing EF and in particular planning skills may be beneficial (Evans et al., 2009). This idea is an extension of the *Cognitive-behavioral model of impairment* by Safren (2006), in which the functional problems (for example regarding school- and homework) that arise from the neuro-developmental symptoms of ADHD (e.g., inattention, impulsivity), are mediated by compensatory strategies like planning and organizing (see Figure 1.1). Following Safren's model, one might argue that enhancing planning and organizing skills might not diminish the disorder itself, but could decrease the functional problems arising from it (Safren, 2006). This is supported by studies in children and adults that showed significant improvement of ADHD-symptoms (in adults; Safren et al., 2005; Safren et al., 2010; Solanto et al., 2014) and academic- and organizational functioning (in children; Abikoff et al., 2013) after receiving treatment aiming at planning and organization skills. Moreover, the children and adults who received this treatment improved more than controls on a waitlist, controls receiving medication and controls receiving another nonpharmacological treatment (e.g., relaxation treatment; Abikoff et al., 2013; Safren et al., 2005; Safren et al., 2010; Solanto et al., 2014). However, no RCTs have been conducted, investigating planning skills-based CBT in adolescents with ADHD (Antshel & Olszewski, 2014).

Studies that *have* been conducted on the effects of treatment in adolescents with ADHD all acknowledge the importance of enhancement of planning skills in treatment (Antshel et al., 2012; Evans et al., 2011; Langberg et al., 2012; Molina et al., 2008; Sibley et al., 2013; Vidal et al., 2015). And clinic-based treatments have incorporated modules aiming at planning skills (Antshel et al., 2012; Sibley et al., 2013; Vidal et al., 2015), but none of them focused the entire treatment on enhancement of planning skills and therefore it is yet unknown whether this is an effective treatment mechanism. Plan My Life (PML; or Zelf Plannen in Dutch; Kuin et al., 2013) is a clinic-based CBT for adolescents with ADHD focusing on enhancing planning and

Figure 1.1: Cognitive-Behavioral Model of Impairment in ADHD (Safren, 2006)



organizing skills, with special attention for school and homework.

PML consists of 8 individual sessions for the adolescent and 2 for his/her parents. Because PML is a short-term treatment, consisting of only 10 sessions, it is easy to implement in the existing Dutch mental health care. PML is a CBT in which every session a fixed, planning skills focused, subject is discussed. Planning- and organization strategies (such as a to-do list) are presented of which the adolescent can compose the strategies he/she wants to try the following week. Whenever needed, negative thoughts about the new strategy are challenged and replaced by positive thoughts. Every session strategies that have been tried during the past week are discussed, including successes, possible room for improvement, and associated (negative/positive) cognitions. The two parental sessions aim at finding a balance between keeping control of their adolescent and letting go, by discussing which parenting goals are worth the battle. Finally, parents are taught how to formulate and implement rules in the household and how to facilitate positive communication with the adolescent (for a more extensive description of Plan My Life in Dutch, see **appendix**, p. 151).

To investigate the effects of PML on the ADHD- and comorbid symptoms

and associated problems of adolescents with ADHD over time, an RCT is conducted. To control for non-specific treatment effects, and investigate enhancement of planning skills as a treatment mechanism, the effects of PML are compared to a CBT without such an aim: a solution focused treatment (SFT; Boyer et al., 2014). In many ways the CBTs resemble each other, for example both PML and SFT are individual treatments consisting of eight adolescent sessions and two parental sessions of 45-60 minutes, in both CBTs the adolescent and therapist use a workbook and the first session is identical including psycho-education on ADHD. In SFT every session the adolescent discusses a problem he/she encounters and following fixed questions, the adolescent is guided towards a solution for the posited problem. The most important difference between both treatments is that whereas in PML every week planning skills are actively learned by discussing a fixed subject, in SFT the adolescent/parent has to choose the subjects themselves and is guided towards his/her own solution (Boyer et al., 2014; Kuin et al., 2013). To prevent treatment drop-out, in *both* PML and SFT, Motivational Interviewing (MI) techniques were used throughout both treatments to augment treatment motivation, as MI has been shown to have a positive effect on treatment adherence in adolescents (Antshel et al., 2012; Erickson, Gerstle, & Feldstein, 2005; Wolraich et al., 2005). In this thesis a large multi-site RCT comparing these two CBTs will be described in a sample of adolescents with ADHD. Not only will this allow us to see whether these adolescents improve during treatment, also will the comparison of both treatments make it possible to investigate the specific contribution of incorporating planning skills enhancement into CBT (**chapter 3**).

## LONG-TERM EFFECTS OF TREATMENTS FOR ADHD

When investigating treatments, the ultimate goal is to reach improvements on symptom level as well as on the level of comorbid and functional impairments after treatment -but maybe even more important- that these improvements are maintained on the longer term. Even though the literature on long-term treatment effects in ADHD is very limited especially in adolescents (including pharmacological treatment), reviews of the existing literature generally show that without treatment, individuals with ADHD have poorer long-term outcomes compared with individuals without ADHD (Parker, Wales, Chalhoub, & Harpin, 2013; Shaw et al., 2012). The MTA-study is probably the most important follow-up study, because of their large



sample size ( $n=579$ ) and four distinctive treatment groups. Results showed that treatment for ADHD (especially treatment with medication or combined medication and behavioral treatment) improved outcomes to one year after treatment as compared to untreated ADHD (or to community care), although usually the outcomes do not improve to normal levels (Molina et al., 2009; Parker et al., 2013; Shaw et al., 2012). However at the longer term, naturalistic follow-ups show that from three years after treatment these initial treatment differences dissipate (Molina et al., 2009; Parker et al., 2013).

RCTs specifically on the effects of planning- and organization-based treatments in children and adults with ADHD indeed show positive results of planning-focused treatments as compared to waitlist- and active control groups on short term (Abikoff et al., 2013; Safren et al., 2010). On the longer term these treatments showed maintenance of initial improvements or resulted in more gains than control treatments to at least nine months after treatment in children and adults with ADHD (Abikoff et al., 2013; Safren et al., 2010). However, no systematic RCTs have been conducted on long-term improvements of behavior following CBT aiming at planning skills in adolescents with ADHD. To know whether possible direct effects of both CBTs and potential differences between both CBTs are lasting, it is important to investigate their long-term effectiveness (**chapter 4**).

## MODERATING EFFECTS IN TREATMENTS FOR ADHD

ADHD is a heterogeneous disorder, which makes it is unlikely for one treatment to fit all and increases the need for personalized treatment. For some subgroups of individuals with ADHD, treatments may be more or less effective than for other subgroups, a phenomenon that is called moderation of treatment effects (Maric, Prins, & Ollendick, 2015). Moderators are pre-treatment variables for which treatment has a differential effect at different values of the moderating variable. Knowledge on such variables will inform us under what conditions treatments work and are therefore an important complement to main effects of RCTs (Maric et al., 2015).

To date, only two research groups have investigated a broad range of treatment moderators (child and family characteristics) in younger children with ADHD (up to 12 years of age; Jensen et al., 2001; MTA cooperative group, 1999b; Van den Hoofdakker et al., 2010, 2012, 2014). Although solely based on behavioral (parent) treatment of children with ADHD as compared to other types of treatment (i.e., medication, combined treatment or regular

care), the following characteristics appeared to positively influence the effects of behavioral (parent) treatment on core ADHD-symptoms: having no or one single comorbid disorder (Van den Hoofdakker et al., 2010), in particular anxiety (Jensen et al., 2001; MTA cooperative group, 1999b), being older of age, having a mother with high parenting self-efficacy (Van den Hoofdakker et al., 2010), or having particular genetic sensitivity due to having no or one single dopamine transporter gene DAT1 10-repeat allele (Van den Hoofdakker et al., 2012).

To date, no treatment moderation studies have been conducted in RCTs on the effects of CBT in samples of adolescents with ADHD. One non-randomized study ( $n=68$ ) on the effects of a planning focused CBT explored the pre-post treatment effects in different subgroups of adolescents with ADHD (Antshel et al., 2012). It showed that adolescents with comorbid anxiety and with comorbid depression improved more in comparison to adolescents with ADHD only. In contrast, adolescents with combined ADHD and ODD improved less from CBT when compared to adolescents with ADHD only (Antshel et al., 2012). Although no theoretical models of moderation for CBT in adolescents with ADHD exist, one can presume one of our CBTs to be a better fit for a certain subgroup of adolescents than the other. For example, one could argue that older adolescents, adolescents with higher IQs, or less ADHD symptoms or impairment or adolescents with parents who have been educated more, have more cognitive capacity or support at home, and therefore do better in a more 'open' treatment without a specific treatment aim like SFT. While younger adolescents, or with lower IQs, more severe ADHD or impairment and less educated parents fare better with a more structured treatment with a specific aim like PML. To know for which subgroups of adolescents with ADHD what CBT would be the most beneficial, it is important to investigate moderators of treatment effects (**chapter 5**).

## OUTLINE AND AIMS OF THIS THESIS

In sum, even though ADHD is highly prevalent in adolescents, and the risk for developing comorbid problems and functional impairments is substantial, effective nonpharmacological treatments are lacking or primarily school-based (Sonuga-Barke et al., 2013). Enhancing planning and organizing skills can possibly compensate for the executive dysfunctions that are associated with the disorder. However, to date not much attention has been paid to the assessment and treatment of planning skills of individuals with

ADHD within this age-range. Therefore, the aims of this thesis are the following:

1. Whereas planning skills of their TD peers go through major developments to secure an independent lifestyle, little is known about the planning skills of adolescents with ADHD. The first aim is to investigate how many adolescents with ADHD have planning problems using multiple measures of planning and exploring DSM-IV subtype differences (**chapter 2**, p. 29).
2. Clinic-based nonpharmacological treatments are sparse in the European context. We developed two CBTs with elements of Motivational Interviewing to treat adolescents with ADHD; one focusing on enhancement of planning skills (Plan My Life, in Dutch *Zelf Plannen*: see **appendix**, and one that does not (Solution Focused Treatment, in Dutch *Zelf Oplossen en Bedenken*). The second aim of this thesis is to investigate the effectiveness of these two new treatments for adolescents with ADHD on the short term to three months after treatment. Also, the comparison of both treatments enables us to make inferences about the specific contribution of incorporating planning skills into CBT (**chapter 3**, p. 49).
3. To know whether possible direct effects of both CBTs and differences between both CBTs are stable, the third aim of this thesis to investigate the effectiveness of these two new treatments for adolescents with ADHD on the longer term to one year after treatment (**chapter 4**, p. 83).
4. Because ADHD is a heterogeneous disorder, nonpharmacological treatment protocols cannot simply be used with every individual. Tailor-made or fitted treatments are therefore desirable. The fourth aim of this thesis is to investigate whether there are pre-treatment characteristics that moderate treatment-effects of PML and SFT in adolescents with ADHD (**chapter 5**, p. 95). When indeed moderators are found, this would be highly relevant for personalized treatment assignment, as it would make allocation of adolescents with ADHD to specific treatments possible, resulting in higher treatment efficacy.

In **chapters 3 to 5**, studies are described that are all based on the same sample of adolescents with ADHD, using the same inclusion criteria and all measures are calculated in the same way across studies. **Chapter 2** is also based on the same sample of adolescents with ADHD, however, a sample of TD adolescents was added to the design and different inclusion criteria were used as compared to the other three chapters: in **chapter 2** an ADHD diagnosis was confirmed by the Dutch Disruptive Behavior Disorder rating scale parent version (DBD; Oosterlaan, Scheres, Antrop, Roeyers, & Sergeant, 2000; Pelham, Gnagy, Greenslade, & Milich, 1992) and the Diagnostic Interview Schedule for Children for DSM-IV (DISC-IV; Shaffer, Fisher, Lucas, Dulcan, & Schwab-Stone, 2000). In chapters 3 to 5 only the DISC-IV was used to confirm ADHD diagnosis.

Further, scores of the *Behavioral Assessment of the Dysexecutive Syndrome* (BADS; Wilson, Alderman, Burgess, Emslie, & Evans, 1996) were calculated differently in **chapter 2** as compared to the other chapters. In **chapter 2** the strategy score and completion time of the BADS Key Search were reported separately, whereas in other chapters a strategy score-completion time ratio was used. Moreover, in **chapter 2** the order score and completion time of both conditions of the BADS Zoo Map were reported, whereas in other chapters the difference between the order score-time ratio of both conditions was calculated. The main reason for using ratio scores in the RCT studies (**chapter 3 to 5**) was to reduce the number of outcome variables to lower Type I error in the analyses, whereas in **chapter 2** a more detailed approach was appropriate to investigate planning skills.

The study was approved by the ethics committee of the University of Amsterdam (2010-KP-1079) and registered in the Clinical Trials Register (NTR2142).