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TEMPERAMENTAL CHARACTERISTICS OF CHILDREN WITH DEVELOPMENTAL STUTTERING

FROM PARENT QUESTIONNAIRE TO NEUROPSYCHOLOGICAL PARADIGMS

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Doctoral thesis in Biomedical Sciences
Leuven 2012
Doctoral thesis in Developmental Psychology
Tilburg 2012

Temperamental characteristics of children with developmental stuttering: From parent questionnaire to neuropsychological paradigms.

ISBN 9789090271859

For my dad

In paradisum deducant te angeli.

Life and death are one.

Acknowledgements

How to start a writing that heralds the end of a yearlong process? The study described in this book, has determined to a large degree the last years of my life and consequently also that of my family, but, and that must also be said, has offered me numerous invigorating opportunities. Writing a thesis is a challenge that not only presumes personal commitment but also the support and cooperation of many people. Let me therefore start by expressing my feelings of gratitude to the latter.

Thank you Luc, for allowing me to walk the path of my choice but nonetheless to guide me steadily in the right direction and keeping me focused. Our discussions and Skype sessions, or the more exotically flavored meetings from Philadelphia, to Rio de Janeiro and Miami, always gave me plenty of food for thought and ideas on how to solve emerging problems. Thank you Bea, for your openness and willingness in helping me to apply the concepts of reactivity and self-regulation to the area of developmental stuttering, for your encouragements, and for sometimes pointing out the obvious, or at other times advising me the less obvious. For this area of research, I could not have wished for a better team of promoters.

I would like to thank the members of my doctoral guidance and/or exam committee, prof. Patricia Bijttebier, prof. Edward Conture, prof. Jaap Denissen, prof. Mark Jorissen, prof. Joost van den Oord, and prof. Inge Zink, for guiding my work and making constructive suggestions, for reading and evaluating my thesis, and for being present at my defense.

Thank you to the many people who inspired me to specialize in fluency disorders: René, behavioral therapist at heart, I have always felt privileged to follow in your footsteps, and am very grateful for making me pursue this PhD-research; Sonja, you were the ideal clinical supervisor during my fluency specialization training, quiet, considerate, but above all a very competent therapist; my clients, Rob and Nick, the happy twins, prototypical examples of individual differences, Stijn and Wai-Kin, both in the prime of your lives but profoundly impeded by stuttering, your individual stories have been pivotal for my work as a fluency specialist. Also a special thank you to Jane Fraser, president of the Stuttering Foundation, for giving me the opportunity to participate in the long tradition of intensive SFA workshops, the first that was held at Iowa University, and to workshop leaders prof. Tricia Zebrowski and Toni Cilek, for your insights, enthusiasm, and hospitality (not to mention the Belgian beer you served me in your homes).

A number of people I want to thank because they contributed to a large extent to the feasibility of our research: prof. Flora Carrijn, Vice-chancellor of Lessius University College, for unhesitatingly answering my request to make research an important part of my job description; Kris Lambers, former head of the SLT department of Lessius, for tuning my other tasks to my research and for giving me the opportunity to participate in various international conferences; students Karolien Gebruers, Julie Germanes, Sofie Mangelschots, Karen Remans, Sophie Sambre, and Hanne Vermeire for their assistance with processing some of the data; and the participating children and parents, speech and language therapists, fluency therapists, school principals, teachers and centers for student guidance, without whom these had remained merely ideas.

And then there are the supporters on the sidelines: Rik, thank you for listening and your advice during the difficult times; Chris and Magd, Lieve and Jan, Eric, thank you for the little encouragements from time to time; my colleagues at Lessius, who, since the last two years, I can proudly call my team; my European colleagues at the ECSF consortium, and especially Margaret and Carl, now we have even more time for our steering committee meetings; my colleagues at the private practice; …

Mum and dad, I can only hope you realize how grateful I am, just sad that I couldn’t share the end of the journey with the both of you. Floris, finally the time is here to resume our golf sessions ... Fore! Marthe, no more work on Sundays or solitary confinement in my upstairs office, that’s a promise.
Lastly I want to thank Els, my support through thick and thin. You fitted your schedules to my work schedule, took care of a large part of the family tasks, and made sure everything ran on wheels, … forever indebted. Your support has been more important than you will ever suspect.

Although I would rather consider this as a beginning and not an end, no better words to end with then these lines out of one of Strauss’s Four Last Songs, ‘Beim Schlafengehen’, written by Hermann Hesse (1911): “*Nun der Tag mich müd gemacht, soll mein sehnliches Verlangen, freundlich die gestirnte Nacht, wie ein müdes Kind empfangen. Hände, laßt von allem Tun, Stirn, vergiß du alles Denken, alle meine Sinne nun, wollen sich in Schlummer senken.”*

Kurt Eggers
October 2012

Abstract

Developmental stuttering is a speech fluency disorder characterized by frequent, involuntary repetitions, prolongations, and/or blocks of sounds, syllables, or words. Often these disruptions are accompanied by secondary behaviors such as struggle, postponement, and avoidance behaviors and frequent indications of an emotional impact. The incidence and prevalence are respectively about 5 and 1%. Stuttering usually starts between 2-to-5 years of age and recovers spontaneously in about 80% of the cases. Current views on the etiology of stuttering emphasize a multifactorial model, combining genetic, neurobiological, behavioral, emotional, and environmental factors. During the last decades several authors emphasized the possible importance of temperamental characteristics for the onset and development of stuttering. Due to the limited amount of studies in this area and the fact they were primarily parent questionnaire-based, several questions with regard to the association between developmental stuttering and temperamental characteristics still remained unanswered. Temperament can be defined as constitutionally based individual differences in reactivity and self-regulation. Reactivity refers to the speed and intensity of motor, cognitive, affective, and autonomic responses. Self-regulation refers to those processes that can modulate reactivity, i.e. facilitate or inhibit.

The general aim of this thesis was to gain in depth insight in the relation between temperament and developmental stuttering, including temperamental components of attention and inhibitory control. This general aim was further specified in three main research objectives, as reflected in the different studies. For the first research objective a temperament questionnaire was administered to large groups of children who stutter (CWS) and children who not stutter (CWNS) to detect possible differences. For the second and third research objective, neuropsychological computer tasks were employed to detect possible differences between CWS and CWNS on attentional networks and inhibitory control.

The Children's Behavior Questionnaire was administered to the parents of CWS, CWNS, and children with vocal nodules. Analysis of the underlying factor structure revealed, apart from some minor differences, a congruent factor structure for the three

participant groups. Consequent analyses of the factors and scales showed CWS to be higher in negative reactivity and lower in self-regulation, partly determined by higher scale scores for anger/frustration, motor activation, and approach and lower scale scores for inhibitory control and attentional shifting.

The following studies always included a group of CWS matched to a group of CWNS. For the second research objective, the Attention Network Test, a combination of a cued reaction time task and a flanker task, was used. Findings revealed a lower efficiency of the orienting network for CWS and a trend towards a lower efficiency of the executive control network.

For the third and final research objective, the efficiency of inhibitory control was evaluated by using a Go/NoGo task, a stop signal task, and a sustained attention task. CWS were found to be less able to suppress prepotent response tendencies in those tasks where response inhibition had to be generated endogenously while they performed comparable to CWNS in a task where response control was externally triggered.

In conclusion, current studies have shown that CWS, as a group, differ from CWNS on temperamental constellation, more in specific they are higher in negative reactivity and lower in self-regulation. With regard to self-regulation, differences emerged on attentional regulation and inhibitory control. These findings have led to an increased insight in the relation between temperament and developmental stuttering and have laid the foundations for future longitudinal studies. The latter will be necessary to unveil the way of interaction between temperament and developmental stuttering.

Korte inhoud

Ontwikkelingsstotteren is een stoornis in de spreekvloeiendheid die gekenmerkt wordt door het voorkomen van frequente, onvrijwillige herhalingen, verlengingen, en/of blokkeringen van klanken, syllaben, of woorden. Vaak gaat dit gepaard met bijkomende gedragingen zoals duw- en uitstel- en vermijdingsgedragingen en zijn er ook frequente indicaties van een emotionele impact. De incidentie en prevalentie bedragen respectievelijk ongeveer 5 en 1%. Stotteren ontstaat meestal tussen de leeftijd van 2 tot 5 jaar en in ongeveer 80% van de gevallen treedt er spontaan herstel op. Hedendaagse visies benadrukken een multifactorieel verklaringsmodel voor het ontstaan van stotteren waarbij zowel genetische, neurobiologische, gedragsmatige, emotionele en omgevingsfactoren een rol vervullen. In de voorbije decennia werd door een aantal auteurs het mogelijke belang van temperamentfactoren in het ontstaan en de ontwikkeling van stotteren benadrukt doch gezien het beperkt aantal studies binnen dit domein en het feit dat deze voornamelijk waren gebaseerd op oudervragenlijsten, bleven heel wat vragen omtrent de associatie tussen ontwikkelingsstotteren en temperamentskenmerken tot op heden onbeantwoord. Onder temperament verstaat men constitutioneel bepaalde individuele verschillen op vlak van reactiviteit en zelfregulatie. Reactiviteit verwijst naar de snelheid en de intensiteit van motorische, cognitieve, affectieve en autonome responsen. Zelfregulatie verwijst naar processen die deze reactiviteit kunnen moduleren, zij het faciliteren of inhiberen.

Het doel van deze studie was het verkrijgen van een grondiger inzicht in de relatie tussen temperament en ontwikkelingsstotteren, met inbegrip van de temperamentcomponenten aandacht en inhibitie. Hiertoe werden drie onderzoeksluiken uitgewerkt, zoals weerspiegeld in de verschillende studies, waarbij in het eerste onderzoeksluik aan de hand van een temperamentsvragenlijst, afgenomen bij een grote groep van kinderen die stotteren (KDS), werd nagegaan of deze verschillend scoorden dan kinderen die niet stotteren (KDNS). Bij het tweede en derde onderzoeksluik werden neuropsychologische computertaken gehanteerd om eventuele verschillen op gebied van aandacht en inhibitie tussen KDS en KDNS vast te stellen.

De Children's Behavior Questionnaire werd ingevuld door de ouders van KDS, KDNS, en kinderen met stembandknobbels. Analyse van de onderliggende factorstructuur toonde aan dat, buiten een paar mineure verschillen, deze in grote mate gelijk was voor de 3 groepen. Bij daaropvolgende factor- en schaalanalyses vertoonden KDS een hogere negatieve reactiviteit en een lagere zelfregulatie, mede bepaald door respectievelijk een hogere schaalscore voor woede/frustratie, motorische activatie en toenadering en een lagere score voor inhibitie en het verdelen van aandacht.

De volgende onderzoeksluiken werden telkenmale uitgevoerd bij een groep van KDS en een gematchte groep van KDNS. In het tweede onderzoeksluik toonde de afname van de Attention Network Test, een combinatie van reactietijdtaak met cueing en een flankertaak, aan dat KDS een lagere efficiëntie hadden van het oriënteringsnetwerk en een trend vertoonden naar een lagere efficiëntie van het executieve aandachtsnetwerk.

Tijdens het laatste onderzoeksluik onderzochten we, op basis van een Go/NoGo-taak, een stop signal taak en een volgehouden aandachtstaak, de efficiëntie van inhibitie. KDS waren minder in staat hun eerste responstendensen te onderdrukken in taken waar deze responsinhibitie endogeen diende te worden gegenereerd terwijl extern getriggerde responsinhibitie ongeveer gelijkaardig verliep.

Samenvattend kunnen we stellen dat huidig onderzoek heeft aangetoond dat KDS, als groep, wel degelijk verschillen op vlak van temperament van KDNS, met name vertonen zij een verhoogde reactiviteit en een verlaagde zelfregulatie. Met betrekking tot deze laatste werden zowel verschillen aangetoond op vlak van aandachtsregulatie en inhibitie. Deze bevindingen hebben geleid tot een beter begrip van hoe temperament en ontwikkelingsstotteren gerelateerd zijn en hebben de basis gelegd voor eventuele verdere longitudinale onderzoeken. Deze laatste zijn noodzakelijk om de manier van beïnvloeding tussen temperament en ontwikkelingsstotteren verder bloot te leggen.

Acronyms and abbreviations

ADHD	attention deficit and hyperactivity disorder
alert.	alerting
ANOVA	analysis of variance
ANCOVA	analysis of covariance
ANT	Attention Network Test
ASIA	Antwerp Screening Instrument for Articulation
ATQ	Adult Temperament Questionnaire
AWS	adults who stutter
BAS	behavioral approach system
BSQ	Behavioral Style Questionnaire
CBQ	Children's Behavior Questionnaire
CBQ-D	Children's Behavior Questionnaire - Dutch
CWS	children who stutter
CWNS	children who do not stutter (also referred to as typically developing children)
CWVN	children with vocal nodules
DBS	deep brain stimulation
DIS	Detection Instrument Stuttering
EAS	emotionality, activity, and sociability
e.g.	exempli gratia: for example
ENT	ear, nose, and throat
et al.	et alia: and others
exec.	executive attention
fig.	figure

IBQ	Infant Behavior Questionnaire
IC	inhibitory control
i.e.	id est: that is to say
IFC	inferior frontal cortex
KU Leuven	Katholieke Universiteit Leuven
KDS	kinderen die stotteren
KDNS	kinderen die niet stotteren
MANCOVA	multiple analysis of covariance
ms	millisecond
n	number
NYLS	New York Longitudinal Study
orient.	orienting
PASW	Predictive Analytics SoftWare
RT	reaction time
SD	standard deviation
SLP	speech language pathologist
SMA	supplementary motor area
SPSS	statistical package for the social sciences
SSD	stop signal delay
SSI	Stuttering Severity Instrument
SSRT	stop signal reaction time
STN	subthalamic nucleus
TDC	typically developing children
TvS-L/NL	Test voor Stotterernst – Lezers/ Niet-Lezers: Test for Stuttering Severity-Readers/Non-Readers
U	university
WISC-III	Wechsler Intelligence Scale for Children-Third Edition

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CHAPTER 1

General introduction

1.1 Motivation for the present research

At the time I was working clinically, I often experienced individual differences in children who stutter (CWS) with regard to how and the way in which they reacted to their moments of stuttering, to listener reactions, to stuttering-evoking situations, and more in general to both internal and external stimuli. Questions with regard to these reaction patterns became part of our standard parent interview during the initial assessment procedure and quite often, similar patterns seemed to emerge. Some CWS appeared to react somewhat stronger to certain situations and/or appeared less able to cope with certain (stressful) situations. Guitar (2006) formulated it by stating: "*Many of us who work with children who stutter have often heard parents describe their child as particularly sensitive. Upon questioning, these parents frequently say that, even before stuttering began, the child was more easily upset by changes in routine or was shyer with strangers than his or her siblings These emotional and behavioral characteristics may be a part of the child's inherited physiology"* (pp. 63).

Several authors have alluded to the possible importance of considering these individual differences in reactivity and self-regulation, i.e. temperament, to gain a better understanding in the nature of developmental stuttering (e.g., Bloodstein & Bernstein Ratner, 2006; Conture, 2001; Guitar, 2006; Yairi & Ambrose, 2005). Zebrowski and Conture (1998) wrote "*Presently, relatively little is know about the specific temperamental characteristics of children who stutter and how they compare to those expressed by nonstuttering children. Even less is known about the influence of temperament on the child's performance in stuttering therapy*" (pp. 302).

More than a decade later, we have gained some additional insights in the temperamental characteristics of CWS but still many questions remain unanswered. The general trend in the studies published meanwhile seems to confirm the rather subjective clinical impression of higher reactivity and lower self-regulation in CWS as a group (Anderson, Pellowski, Conture, & Kelly, 2003; Embrechts, Ebben, Franke, & van de Poel, 2000; Karrass, et al., 2006; Lewis & Goldberg, 1997; Wakaba, 1998), although not all findings pointed in the same direction. Moreover, these findings are based on a limited number of studies, some of which with a small number of participants, and all through the use of caregiver-questionnaires.

Therefore, systematic research on a larger population is essential to evaluate if temperamental characteristics observed in all or some CWS may be related to stuttering, stuttering development, and possibly stuttering symptomatology (Seery, Watkins, Mangelsdorg, and Shigeto, 2007; Yairi, 2007). Ideally, these questionnaire-based findings are to be corroborated by other ways of measuring temperament characteristics, e.g. neuropsychological measures, since some authors emphasized the susceptibility to parental bias of these questionnaire-based methods (Strelau, 1998; Vaughn, Taraldson, Cuchton, & Egeland, 2002).

In this chapter, we will provide a general introduction to developmental stuttering (1.2), the history of psychological research in stuttering (1.3), the concept of temperament (1.4) temperament and developmental stuttering research (1.5). We will conclude this chapter by stating the different research objectives (1.7) and providing a short description of the included studies (1.7).

1.2 Developmental stuttering

1.2.1 Definition

Stuttering has been described by the World Health Organization (1994) as "*speech that is characterized by frequent repetition or prolongation of sounds or syllables or words, or by frequent hesitations or pauses that disrupt the rhythmic flow of speech. Minor dysrhythmias of this type are quite common as a transient phase in early childhood, or as a minor but persistent speech feature in later childhood and adult life. They should be classified as a disorder only if their severity is such as markedly to disturb the fluency of speech. There may be associated movements of the face and/or other parts of the body that coincide in time with the repetitions, prolongations, or pauses in speech flow"* (p. 227).

A widely used and accepted definition of stuttering, albeit more a description of the symptoms, is by Wingate (1964) who has defined stuttering as *" 1. (a) Disruption in the fluency of verbal expression, which is (b) characterized by involuntary, audible or silent, repetitions or prolongations in the utterance of syllables, and words of one syllable. These disruptions (c) usually occur frequently or marked in character and (d) are not readily controllable. 2. Sometimes the disruptions are (e) accompanied by accessory activities involving the speech apparatus, related or unrelated body structures, or stereotyped speech utterances. These activities give the appearance of being speech-related struggle. 3. Also, there are not infrequently (f) indications or* *report of the presence of an emotional state, ranging from a general condition of 'excitement' or 'tension' to more specific emotions of a negative nature such as fear, embarrassment, irritation, or the like. (g) The immediate source of stuttering is some incoordination expressed in the peripheral speech mechanism; the ultimate cause is presently unknown and may be complex or compound"* (p. 488).

1.2.2 Onset of stuttering

1.2.2.1 Genetics

Numerous studies have shown that stuttering runs in families, suggesting a possible genetic base for the disorder. Data on genetics and stuttering have come from four major research methods, twin studies (e.g., Ooki, 2005), adoption studies (e.g., Felsenfeld, 1995), family studies (e.g., Kidd, 1984), and more recently, biological genetics (for an overview see Kraft & Yairi, 2012). Family studies revealed that between 23 and 68 percent of the people who stutter also have family members who stutter, while this is only 1 to 18 percent for people who do not stutter (Conture, 2001). Twin studies, comparing the appearance of stuttering in identical versus non-identical twins, have shown heritability estimates of about 70 percent (Howell, 2011). Recent advances in the field of genetics have introduced the use of genetic markers to analyze variations within a family or population, resulting in several candidate genes for stuttering (e.g., Shugart et al., 2004; Wittke-Thompson et al., 2007).

In spite of remarkable progress in uncovering the genetics of stuttering, the exact nature of the genetic components and the role and interactions with environmental factors remains unclear. Some authors have indicated a polygenic model (Cox, Kramer, Kidd, & Rao, 1984), whereas others found evidence for a single major locus model (Ambrose, Yairi, & Cox, 1993). Most likely, the liability for stuttering might depend on the interaction of several factors, including environmental, linguistic, neurological factors, alongside genetic components (Ward, 2006).

1.2.2.2 Age and pattern at onset

It has been established for a long time that developmental stuttering can begin within a wide age range, but usually starts during early childhood. The youngest age for onset being reported consistently in these studies was 18 months (e.g., Milisen & Johnson, 1936) and the oldest age reported was 13 years (Meltzer, 1935), although most studies did not report an onset beyond 9 years. A more recent study by Yairi and Ambrose (2005) showed a somewhat narrower age range for onset, between 16 to 69 months, with a mean age of 33 months. This average age of onset complied with other findings, such as those by Månsson (2000). Yairi and Ambrose also reported that over 85% of onsets had occurred before 42 months of age, increasing to 95% by age 4 years. So although stuttering may start at any childhood age, stuttering is much more likely to develop at the youngest age groups.

Some studies have also examined the pattern of onset, a gradual versus sudden onset, pointing to the way in which parents perceived the stuttering symptoms to appear. Yairi and Ambrose (2005) reported that over 40% of the children exhibited a sudden onset (1-3 days), 32% an onset between 1-2 weeks, and the smallest group, 27%, showed a gradual onset, i.e. 3 weeks or above.

1.2.2.3 Prevalence and incidence

The prevalence of a disorder is an estimate of how many people have a disorder at a given time. The incidence reflects the chance of occurrence of the disorder, in other words, how many people have stuttered in their lives. Bloodstein and Bernstein Ratner (2006) reviewed and summarized the results of over 40 studies. In general population estimates point to a prevalence of about 1% and an incidence of approximately 5%. It needs to be added that the prevalence of stuttering varies considerably with age, and that this 1% refers to the school-aged group while the younger ages have shown a higher prevalence and the older ages a lower prevalence (Guitar, 2006). Ambrose, Cox, and Yairi (1997) point to the fact that the difference between prevalence and incidence, approximately 4%, are those people that have recovered from stuttering, spontaneously or through treatment.

1.2.2.4 Spontaneous recovery versus chronic stuttering

A considerable group of the children that have started stuttering have been shown to recover spontaneously, i.e. without treatment (e.g., Andrews & Harris, 1964; Curlee & Yairi, 1997). Percentages of recovery cited in the literature, as reported by Bloodstein & Bernstein Ratner (2006), were as high as 78.9%. Most of these findings were based on retroactive research and parental reports. A more recent longitudinal study by Yairi's group (Paden, Yairi, & Ambrose, 1999; Watkins, Yairi, & Ambrose, 1999; Yairi & Ambrose, 1999) confirmed this high percentage of recovery and has provided more insight in the predictors of recovery and persistence in developmental stuttering. Most of the children that recovered did so within the first 2-to-3 years after stuttering onset. Factors such as gender (girls had a higher recovery rate compared to boys), family history (children who had relatives with persistent stuttering were more likely to demonstrate persistent stuttering), time since onset (the longer a child had been stuttering, the less likely spontaneous recovery was) were found to be associated with spontaneous recovery. Zebrowski and Conture (1998) have hypothesized that also temperamental constellation might be one of these associated factors.

1.2.2.5 Male-to-female ratio

Studies have consistently been showing a higher prevalence of stuttering in boys compared to girls and the ratio that often has been cited is about 3 males to 1 female (Guitar, 2006). There is a strong evidence though that this male-to-female ratio increases as children grow older. For example, Kloth, Kraaimaat, Janssen, and Brutten (1999) found a male-to-female ratio of 1.1:1 near stuttering onset, rising to 2.5:1 six years later, a similar pattern as was found by Månsson (2000).

This gradual increase in proportion of boys who stutter might be linked to an increase in the amount of boys that start to stutter with growing older or to a higher proportion of girls recovering spontaneously; the latter seemed to be corroborated by findings by Yairi and Ambrose (2005). Several hypotheses have been proposed to explain this difference, from environmental (Johnson et al., 1959) to hormonal influences (Geschwind & Galaburda, 1985). Kidd, Kidd, and Records (1978) have suggested a model in which both genetic and environmental factors contributed to the liability for stuttering; in this model males were hypothesized to have a lower susceptibility threshold than females.

1.2.3 Stuttering development

What seems to be a consistent feature of developmental stuttering is that it gradually changes after onset. Stuttering commonly starts with monosyllabic word or part-word repetitions without struggle, avoidance behaviors, or speech-related anxiety and often tends to evolve towards prolongations and blocks, coinciding in many children with additional secondary struggle behaviors or speech-anxiety (Guitar, 2006). In some children this transition from effortless and relaxed repetitions to more effortful prolongations and blocks seems to be very gradual and can take several months while in other children this can develop in just a couple of days or weeks. Some authors believe that this is linked with the ease in which classical and operant conditioning processes take place, processes that are being influenced by environmental factors (e.g., listener reactions) or child-specific factors (e.g., low frustration tolerance) (Starkweather, 2002). At onset children will not always be aware of their stuttering symptoms but the longer the stuttering has continued, the higher the likelihood of increased awareness in their stuttering. Caregivers might try not to draw attention to these disfluencies whereas others might actively try to change the disfluencies by giving advice to the child, such as asking the child to slow down or to take a deep breath. These listener reactions, in combination with the child’s own reactions, might lead to negative conditioning of the moments of stuttering or stuttering-evoking situations, resulting in increased anxiety and secondary behaviors. In turn this might lead to avoidance behaviors, increased fear and anxiety and a higher expectancy to stutter, resulting in a vicious cycle (Ward, 2006).

1.2.4 Etiological theories

Over the last decades, several theoretical conceptualizations about the etiology of stuttering have been put forward, ranging from neuropsychological (e.g., Orton & Travis's cerebral dominance theory, 1931), psycholinguistic (e.g., Postma & Kolk's covert repair hypothesis, 1993), speech motor control (e.g., Webster, 1988), to learned behavior approaches (e.g., Sheehan's approach-avoidance conflict, 1970, and Brutten & Shoemaker's two-factor model, 1967). All were influenced by the zeitgeist of that specific era (for an overview: see Packman & Attanasio, 2004).

Current views on the etiology of stuttering emphasize a multifactorial model, combining genetic, neurobiological, behavioral, emotional, and environmental factors (Bloodstein & Ratner, 2008). These multifactorial models (e.g., Smith & Kelly's dynamic multifactorial model, 1997, 1999) argue that the cause of stuttering is the result of a combination of a range of innate and environmental factors, possibly different in each person, and that on their own are not sufficient for a child to start stuttering.

1.2.5 Anatomical correlates of developmental stuttering

Brain imaging studies have revealed both structural as well as functional differences between stuttering and nonstuttering adults in areas related with speech motor and language production (e.g., De Nil, Kroll, Lafaille, & Houle, 2003; Ingham, 2001; Neumann et al., 2003; Sommer et al., 2002; Watkins, Smith, Davis, & Howell, 2008). Cortical areas that have repeatedly been implicated in these kinds of studies are the anterior cingulate and the inferior frontal cortex, respectively associated with motor initiation and planning, and the superior and middle temporal areas, associated with language processing (Ward, 2006). Different activation patterns also occurred in the cerebellum and in the subcortical structure of the basal ganglia (e.g., Watkins, Smith, Davis, & Howell, 2008). Since all of these studies have been done in an adult population, some caution against misinterpretation is warranted since these findings do not necessarily point to the underlying cause for stuttering but might also be the functional result of having stuttered for a long period of time (Howell, 2011).

1.3 History of psychological research in stuttering

Initial research into the psychological contributors to the etiology of stuttering pertained to an old and tainted history (e.g., Sheehan & Zussman, 1951), primarily psycho-dynamically oriented or employing standardized psychiatric or psychological testing instruments, looking for 'abnormality' in the stuttering population. Theories suggesting stuttering to be the result of a disordered personality development or neurotic parental personalities used to be quite popular. The rationale for this personality related research in the stuttering population was basically to determine if the average person who stuttered should be regarded as neurotic or severely maladjusted. Influenced by Freud's psychoanalytical viewpoints, in the first half of the 20th century, but also lingering on in the following decades, some considered stuttering to be "*the result of a neurotic disorder in which personality disturbance is in part reflected in disturbances of speech*" (Glauber, 1958, p.73). Results of several studies (for an overview see Bloodstein & Bernstein Ratner, 2006) however have shown that people who stutter were not distinctly neurotic or severely maladjusted individuals; moreover, most people who stuttered performed well within the range of normality.

Yairi and Ambrose (2006) stated that currently there seems to be a renewed interest in this domain and referred to recent models of stuttering etiology in which personality and temperamental concepts were hypothesized to influence the developmental course of stuttering. Important to add though is that these recent hypotheses did not suggest underlying abnormalities in these areas but rather pointed to the possibility of subtle differences in psychological profiles, all falling within normality.

1.4 The concept of temperament

1.4.1 Different perspectives on temperament

1.4.1.1 History of temperament

The word temperament is derived from the Latin *temperare*, meaning 'to mix, to combine in a proper proportion', referring to the various components working together to create behavior (Kristal, 2005). The ancient Greeks already used a temperament model to explain behavioral characteristics. Behavior was considered to be the result of biological differences in bodily fluids (i.e. blood, yellow and black bile, and phlegm), also labeled as the four humors. Individual behavioral differences were the result of the balance or misbalance of these humors. A fourfold typology, i.e. melancholic, choleric, sanguine, and phlegmatic, was used to explain the different behavioral types. A melancholic person, linked to a predominance of black bile, was considered moody, with a tendency to fear and sadness. A choleric individual was active, touchy, and aggressive, with a predominance of yellow bile. The sanguine person was sociable, easygoing, and was linked to a predominance of blood. Finally, the phlegmatic person, with a predominance of phlegm, was considered calm and slow to emotion. The merit of this Greek typology is the postulation that individual behavioral differences were linked to underlying physiological mechanisms (Strelau, 1998) and more recent studies on the biological basis of temperament, showing the relation between emotions and the endocrine system (Kagan, 1998; Strelau, 2001), have provided support for this ancient conceptualization. This typology remained to be used, albeit in a slightly changed form, until the late 1800s.

In the 20th century a renewed interest in temperament was apparent and several conceptualizations were put forward, spearheaded by scholars from three different research centers: Thomas and Chess, Teplov and Nebylitsyn, and Eysenck (Strelau, 1998). Eysenck (1947), who considered temperament and personality as synonymous (but primarily used the term personality to refer to his dimensions of Psychoticism, Extraversion, and Neuroticism), tried to explain individual differences in terms of physiological constructs, based on a wide range of empirical studies. Teplov and Nebylitsin (Bodunov, 1993) applied Pavlov's typology to the human central nervous system to study the physiological mechanisms of temperament. Thomas and Chess (1977), widely considered as the promoters of contemporary temperament research, launched a longitudinal study on the role of environmental (parenting) factors and temperament in child behavioral disorders.

Towards the midst of 20th century, differences in emotional expressions, including strength and speed of emotional responses, were being explored as part of personality theories (e.g., Eysenck, 1947) and child psychologists were putting more focus on the biological and maturational aspects of psychological development and supported the role of heredity, separate from environmental influences (Gesell & Ames, 1937). The concept of individual differences triggered the nature-versus-nurture debate in the second half of the 20th century, with arguments that genetics and biology accounted for the individual differences from the nature proponents, while the nurture followers stressed the importance of early environmental influences, such as the caregiver's role (Kristal, 2005). The New York Longitudinal Study (NYLS), started in 1956 by Thomas and Chess (1977), studying individual differences in a large cohort of children, changed the emphasis to the nature component.

1.4.1.2 Temperament models

During the last few decades, different theoretical conceptualizations on temperament have been developed. Some of these have focused mainly or even exclusively on emotion-oriented traits (e.g., Goldsmith & Campos, 1990) while others saw temperament more as a behavioral style (e.g., Thomas & Chess, 1977). In spite of differences in defining temperament, most temperament researchers seem to agree that temperament a) refers to behavioral characteristics in which individuals differ (several terms are used, e.g. traits, dispositions, dimensions), b) is relatively stable throughout the child's development and is characterized by considerable cross-situational consistency, c) has a biological basis, and d) refers to formal characteristics of behavior or reactions, such as intensity and speed (Strelau, 1998).

Moreover, temperament theories can also be categorized into child-oriented (Buss & Plomin, 1984; Goldsmith & Campos, 1990; Kagan, 1998; Rothbart & Derryberry, 1989; Thomas & Chess, 1977) or adult-oriented (e.g., Eysenck, 1947; Gray, 1991; Strelau, 1998) theories of temperament. Seen the topic of the current thesis (developmental stuttering) and the age range of the participants (3;01-10;11), we have limited this overview to the well-known, child-oriented theories Table 1.1 provides an overview of the different temperament characteristics used in these theories.

Table 1.1: Overview of the temperament characteristics used by Thomas and Chess, Buss and Plomin, Goldsmith and Campos, Kagan, and Rothbart.

Thomas & Chess	Buss & Plomin	Goldsmith & Campos	Kagan	Rothbart
Activity level	Emotionality	Activity level	Inhibited	Positive anticipation
Rhythmicity	Activity	Pleasure	Uninhibited	High intensity pleasure
Approach/withdrawal	Sociability	Social fearfulness		Smiling/Laughter
Adaptability		Anger proneness		Activity level
Threshold		Interest/Persistence		Impulsivity
Intensity				Shyness
Quality of mood				Discomfort
Distractibility				Fear
Attention span/Persistence				Anger/Frustration
				Sadness
				Soothability
				Inhibitory control
				Attentional focusing
				Low intensity pleasure
				Perceptual sensitivity

Thomas and Chess (1977; Chess & Thomas, 1986) studied a group of children, and their families - initially 22 but later over 100 families were added – from infancy to adulthood in the NYLS. Based on extensive interviews with the parents they gathered information on the concrete behaviors of the child. They differentiated between three behavioral components, namely the ‘what’, ‘why’, and ‘how’. The ‘what’ referred to how well a child performed a specific task, the ‘why’ referred to the underlying motivational aspect and the ‘how’ referred to the specific behavioral style children were using. The ‘how’ was described by Thomas and Chess as the temperamental component. All of these collected data, pointing to behavioral styles across different situations, were clustered together to find common descriptors. Nine temperament characteristics were identified: a) *activity level*, defined as the level and frequency of motor activity, b) *rhythmicity*, the predictability of bodily functions such as sleep and hunger, c) *approach versus withdrawal*, the child’s first response to novel persons, objects or situations, d) *adaptability*, how easily a child can modify responses to changes in the direction desired by the caregiver, e) *threshold*, the intensity of stimulation required to evoke a response, f) *intensity*, the intensity of a child’s reaction, whether happy, sad, or angry, g) *quality of mood*, the amount of pleasant behavior versus the amount of unpleasant behavior, h) *distractibility*, how easily a child can be distracted by external stimuli, and i) *attention span/persistence*, the ability to continue an activity even when the child is getting frustrated. Based on the constellation of these nine traits, children were categorized as ‘difficult’, ‘easy’, or ‘slow-to-warm-up’. Difficult children (about 10% of the children) were characterized by having a negative quality of mood, withdrawing, being unadaptable, high intensity and low in regularity. Children who were at the opposite poles of these characteristics, i.e. positive quality of mood, high in approach, very adaptable, low intensity, and high rhythmicity, were labeled as ‘easy’. This was the largest group, about 40%. The ‘slow-to-warm-up’ group (approximately 15%) was low in activity level and intensity, withdrawing at first, but showing adaptability over time. The remaining 35% could not clearly be categorized in either of these categories. There has been a reasonable amount of critique to this classification because of its qualitative labeling and even more, what is considered difficult behavior in a certain situation might be ideal in other situations. Chess and Thomas (1986) also developed the concept of ‘goodness or poorness of fit’ to explain the interaction between the child and the environment. ‘Goodness of fit’ was defined as the compatibility between the child’s capacities and temperament and the environmental demands and expectations (especially parents, teachers, peers). A long lasting ‘poorness of fit’ was considered to possibly lead to a maladaptive functioning. Thomas and Chess assessed temperament based on structured caregiver’s interviews, especially in the youngest age ranges, for the older children, parental or teacher questionnaires were developed.

Buss and Plomin (1984) defined temperament as stable, early detectable inherited traits. They considered two criteria crucial for classifying behavioural characteristics as temperamental traits, namely the presence from early childhood on and the contribution of a genetic factor to differences in this characteristic. They distinguished three independent temperament traits, namely Emotionality, Activity, and Sociability, which provide the basis for later personality development. Emotionality is defined as the tendency to be aroused quickly and intensely and is expressed in the emotion identified as distress. Distress is observable (crying) starting at the first days of life and develops during infancy into fear and/or anger. Positive emotions were not included in their model. Activity refers to the level of activity or the total energy output, consisting of tempo and intensity of moving and speaking. Sociability is the tendency to prefer the presence of being with others rather than being alone. This trait has a clear directional component and can be measured by e.g. the attempts to make social contacts. Buss and Plomin developed several EAS-based inventories to measure temperament in children, adolescents and adults. This theory also makes predictions with regard to the match or mismatch between the child and its environment (e.g., if both mother and child are both high in emotionality, the child’s fears may be intensified by the mother’s emotionality, leading to neurotic behaviors). Buss and Plomin also state that temperament may exert an influence on the environment through selecting environments and modifying environmental impact.

Goldsmith and Campos (Goldsmith et al., 1987, Goldsmith & Campos, 1990) defined temperament from an exclusively emotion-oriented viewpoint, namely as individual differences in experiencing and expressing primary emotions, such as distress, fear, anger, sadness, pleasure, and surprise. These dimensions are expressed behaviorally through different modalities such as facial, vocal, and gestural expressive systems. In their theoretical model, limited to infants, emotionality is synonymous to temperament, in that they viewed emotionality as the regulator of internal psychological processes as well as of social behaviors. They acknowledged some degree of temperamental stability over time and saw it as a precursor for the later development of personality. They advocated a multi-modal assessment of temperament including questionnaires, caregiver interviews, and laboratory assessments. They also developed the Toddler Behavior Assessment Questionnaire for assessing temperament in infants.

Kagan's temperamental concept (1998) referred to inherited behavioral and biological profiles that mediate reaction patterns. In his view temperament is present from infancy and is influenced by childhood experiences. His theoretical conceptualization is based on one continuous dimension, namely inhibited versus uninhibited temperament, referring to the way in which a child reacts when confronted with unfamiliar persons, objects or situations. An inhibited temperament is seen in children that are predominantly shy, reserved and timid, while children that are predominantly social, spontaneous, and minimally fearful in similar conditions, are labeled as uninhibited. These categories are comparable to approach/withdrawal characteristics or extraversion-introversion dimensions, used in other temperament models. Kagan did not develop any temperament questionnaires because of his critique towards the susceptibility to bias of questionnaires; his preferred way of assessing children's temperament was observing children's behaviors under standardized conditions (e.g., measuring a child's reactions to unfamiliar objects, or to separation from the mother). These observations across different conditions were clustered and led to an aggregate index of inhibition.

1.4.2 Rothbart's temperament model

Rothbart's temperament model, described as a developmental (child-oriented), constitutional-psychobiological (causal), and multidimensional approach with the focus on the child's whole behavior (Strelau, 1998), has gained much popularity among child-oriented temperament researchers. Rothbart and Derryberry (1981) defined temperament as constitutionally based individual differences in reactivity and self-regulation. *Constitutional* is referring the person's relatively enduring biological make-up, influenced over time by genetics, maturation, and experience. *Reactivity* refers to the arousability of motor, autonomic, cognitive, affective, and endocrine response systems. In this view, reactivity can be assessed through parameters like reaction threshold, latency, intensity, time to peak intensity, and recovery time. *Self-regulation* refers to those processes that can modulate (facilitate or inhibit) reactivity, such as attention shifting, behavioral inhibition, and withdrawal.

Rothbart (1989a) developed an overall framework specifying the particular role and interactional patterns between the different reactivity- or self-regulation-related processes (see Figure 1.1). When a child is confronted with a stimulus, depending on several stimulus characteristics or child-specific factors, this stimulus might lead to either positive or negative reactivity within the child. With growing older, a child will be able to consciously (effort) modulate this reactivity by employing self-regulatory processes in order for the positive/negative reactivity to increase or decrease, respectively.

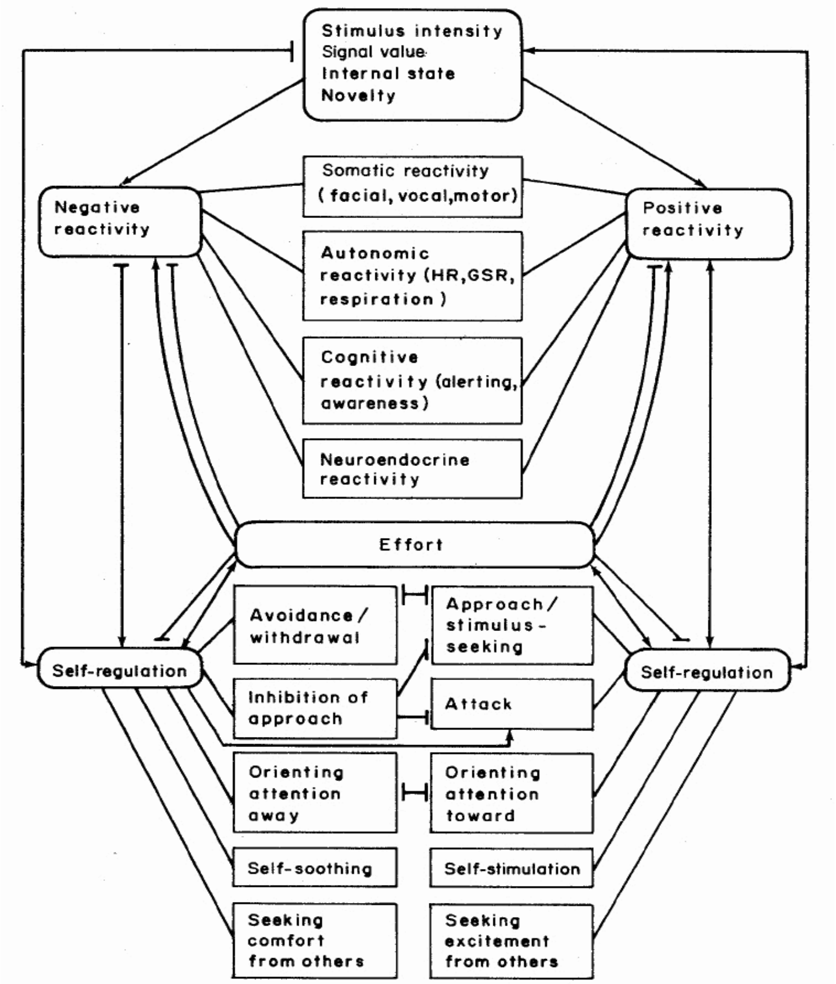


Figure 1.1: A general framework for temperament (adapted from Rothbart, 1989a). Direct lines indicate more specific instances (square-edged boxes) of a more general category (round-edged boxes). Arrows illustrate facilitation while straight lines at the end of a connecting line illustrate inhibition.

1.4.2.1 Positive and negative reactivity

As depicted in Figure 1.1, stimulus characteristics influencing positive either negative reaction patterns include the intensity of the stimulus, the previous coupling of a positive or negative value to the stimulus, and the novelty aspect. In general, low *stimulus intensity* leads to positive reactivity while high intensity results mostly in negative reactions. Nevertheless this is also individual-specific in that some children have a low stimulus threshold whereas others have a high stimulus threshold. In case of a low threshold the ideal stimulus intensity (generating positive reactions such as smiling and laughter) is reached at lower intensity levels compared to children with a high threshold, who are in need of more intense stimulation in order to experience and express pleasure. *Signal value* is referring to the previous experiences with and evaluation of the stimulus (intrinsic meaning) that have resulted in either a positive or negative conditioned stimulus. Anticipation of possible rewards will trigger positive reactivity (e.g., approach) while anticipation of possible punishments (fear) will lead to negative reactions (e.g., avoidance). The child's *internal state*, such as fatigue and hunger, will generate negative reactivity unless their needs are being fulfilled (e.g., feeding), resulting in positive reactivity. Finally, *novelty* is expected to trigger primarily negative reactions although depending on the environment in which the new stimulus is presented, this might also result in positive reactivity (e.g., in some cases of humor).

Positive and negative reactivity are both expressed via somatic (e.g., facial expressions), autonomic (e.g., increase in heart rate, or galvanic skin response) cognitive (e.g., alerting), and neuroendocrine (e.g., cortisol release) reactions and can be respectively experienced as pleasure or distress. In situations with high stimulus intensities, children with a high susceptibility to negative reactions will experience more feelings of distress whereas children with a low susceptibility to negative reactions will experience more pleasure-related feelings in similar circumstances. These specific reaction patterns do not all follow a similar time course (Rothbart & Derryberry, 1981): attentional alerting is initiated in a few milliseconds whilst activation of the neuroendocrine system takes a few minutes to possibly a few hours (Rothbart, 1989a)

1.4.2.2 Self-regulation

Children differ in reactive temperamental components but Rothbart's model (1989a; 2011) also emphasizes that children also differ in the ease with which self-regulation, i.e. attentional and motor control processes, are initiated. Some of these motor self-regulatory processes are aimed at self-soothing (e.g., thumb-sucking) in situations of distress. Attentional self-regulatory components include attentional orienting, i.e. orienting one's attention towards (attentional focusing) or away (attentional shifting) from the exciting or distressing environment. This attentional orienting can also be applied to internal representations, e.g. thinking of a pleasurable activity when confronted with a stress-evoking situation. Self-regulation is triggered by the positive or negative reactions experienced by a child (e.g., negative reactivity elicits withdrawal, positive reactivity elicits approach) and by the signal value of the stimulus. As a consequence, these self-regulatory processes will in turn also modulate (facilitate or inhibit) the reactivity, showing a bidirectional pattern between reactive and self-regulatory components.

Also within the self-regulatory system of this model, interactions are apparent: e.g. behavioral inhibition opposes approach, and approach inhibits avoidance.
Effortful (conscious) control allows children, when becoming older, to dissociate behavioral patterns from purely reactivity-driven behaviors. For instance, a person can approach a specific situation, with effort, even when this situation is triggering negative reactivity. Based on this framework, caregivers can also exert an influence on the child's reactivity and self-regulation by e.g. stimulating a child in a boring environment or by soothing a child, or distracting the child's attention away from a stressful situation.

1.4.2.3 Children's Behavior Questionnaire

Based on the above-described model, Rothbart developed specific temperament questionnaires for different age groups, such as the Infant Behavior Questionnaire (IBQ; Gartstein & Rothbart, 2003), the Children's Behavior Questionnaire (CBQ; Rothbart, Ahadi, Hershey, & Fisher, 2001), and the Adult Temperament Questionnaire (ATQ; Evans & Rothbart, 2007). The CBQ, aimed at 3-to-7-year-old children, the age-range addressed in this thesis, provides a highly differentiated assessment of the different temperamental characteristics of children. While most of the previously developed questionnaires were based on the nine dimensions identified in the NYLS, the development of the current questionnaire was based on a rational, theory-driven approach. The central constructs of Rothbart's model, positive/negative reactivity and self-regulation were first decomposed into subconstructs (scales). Subsequently items were rationally written, based on the conceptual definition for each scale, to reflect these subconstructs (Rothbart et al., 2001), administered to a group of parents, and items with low item-total correlations were eliminated. The finalized CBQ consisted of 15 temperament characteristics, clustering under the three broad dimensions of Rothbart's temperament model, labeled as Extraversion/Surgency (or positive reactivity), Negative Affectivity (or negative reactivity), and Effortful Control (or self-regulation). The different scales and sample items are described in Table 1.2.

Table 1.2: CBQ scale (Rothbart, Ahadi, Hershey, & Fisher, 2001) descriptions and sample items.

CBQ-scale	Definition
Activity Level	The level of gross motor activity including rate and extent of locomotion. *Sample item*: Moves about actively (runs, climbs, jumps) when playing in the house.
Anger/Frustration	The amount of negative affect related to interruption of ongoing tasks or goal blocking. *Sample item*: Gets quite frustrated when prevented from doing something s/he wants to do.
Approach	The amount of excitement and positive anticipation for expected pleasurable activities. *Sample item*: Becomes very excited while planning for trips.
Attentional focusing	The tendency to maintain attentional focus upon task-related channels. *Sample item*: When picking up toys or other jobs, usually keeps at the task until it's done.
Discomfort	The amount of negative affect related to sensory qualities of stimulation, including intensity, rate or complexity of light, movement, sound or texture. *Sample item*: Is quite upset by a little cut or bruise.
Falling reactivity/ Soothability	The rate of recovery from peak distress, excitement or general arousal. *Sample item*: Calms down quickly following an exciting event.
Fear	The amount of negative affect, including unease, worry or nervousness related to anticipated pain or distress and/or potentially threatening situations. *Sample item*: Is afraid of loud noises.
High intensity pleasure	The amount of pleasure or enjoyment related to situations involving high stimulus intensity, rate, complexity, novelty and incongruity. *Sample item*: Likes to play so wild and recklessly that s/he might get hurt.
Impulsivity	The speed of response initiation. *Sample item*: Usually rushes into an activity without thinking about it.
Inhibitory control	The capacity to plan and to suppress inappropriate approach responses under instructions or in novel or uncertain situations. *Sample item*: Can easily stop an activity when s/he is told "no".
Low intensity pleasure	The amount of pleasure or enjoyment related to situations involving low stimulus intensity, rate, complexity, novelty and incongruity. *Sample item*: Enjoys "snuggling up" next to a parent.
Perceptual sensitivity	The amount of detection of slight, low intensity stimuli from the external environment. *Sample item*: Is quickly aware of some new items in the living room.
Sadness	The amount of negative affect and lowered mood and energy related to exposure to suffering, disappointment and object loss. *Sample item*: Becomes upset when loved relatives or friends are getting ready to leave following a visit.
Shyness	Slow or inhibited approach in situations involving novelty or uncertainty. *Sample item*: Sometimes prefers to watch rather than join other children playing.
Smiling/Laughter	The amount of positive affect in response to changes in stimulus intensity, rate, complexity and incongruity. *Sample item*: Laughs a lot at jokes and silly happenings.
Motor activation	The amount of excess repetitive small-motor movement, such as finger tapping. *Sample item*: Fidgets during quiet activities, such as hearing a story, looking at pictures.
Excitatory control	The capacity to perform an action when there is a strong tendency to avoid it. *Sample item*: Forces her/himself to complete projects, even when tired.
Attentional shifting	The ability to transfer attentional focus from one activity/task to another. *Sample item*: Can easily shift from one activity to another.

1.4.3 Measurement of temperament

Since there is no absolute consensus among researchers as to how temperament should be defined, there is no commonly accepted view on how temperament should be measured (Strelau, 1998). Several approaches have been used to measure temperament in children, ranging from caregiver questionnaires, such as the above-described CBQ, (e.g., Gartstein & Rothbart, 2003), self-reports for older children, (semi-) structured interviews (e.g., Garrison, Biggs, & Williams, 1990), home observational measures and laboratory measures (e.g., Goldsmith & Rothbart, 1991); more recently psychophysical and psychophysiological measures have also been introduced (e.g., McManis, Kagan, Snidman, & Woodward, 2002).

Rothbart and Bates (1998) state that every method has relative advantages, as well as disadvantages. While parent questionnaires have the advantage of tapping into the vast knowledge base of parents who have seen the child in many different situations over a long period of time, including infrequently occurring behaviors, home observations might have a higher degree of objectivity, and in laboratory procedures researchers can precisely control the context of the child's behavior. On the other hand, parent questionnaires may be biased by subjective factors, such as perceptual bias in the informant (e.g., Kagan, 2001), home observation measures are often not long enough to gain an adequate sample of all relevant behaviors, and laboratory measures on the other hand might have limitations with regard to the particular kinds of behavior that can be elicited.

1.4.4 Temperament and development

1.4.4.1 Normal development

Temperament, i.e. reactivity and self-regulation, develops in the early years of life and the time of emergence of the different temperamental characteristics is depicted in Figure 1.2. There is a balance between temperament traits and some traits can activate or inhibit others. Effortful control develops around the 10 months and serves several motivational purposes including anger, fear, and approach. In other words, the temperament structure changes over time, from a predominantly reactivity-driven concept in infants to a structure with more emphasis on self-regulatory processes in older children (Putnam, Ellis & Rothbart, 2001).

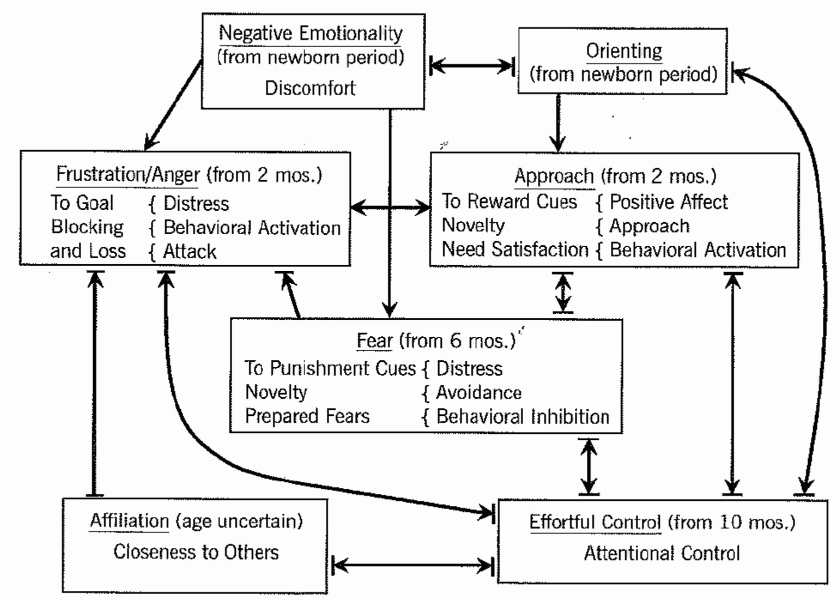


Figure 1.2: A model for the development of temperament systems (adapted from Rothbart, 2011). Systems depicted at the top appear earlier in life compared to systems placed towards the bottom. Lines indicate relations between systems. Arrows illustrate facilitation while arrows with a straight line illustrate inhibition.

While temperament itself develops over time, it also exerts an important influence on the child's social-emotional development, personality development, and the family system (Goldsmith, 1996; Rothbart, Derryberry & Hershey, 2000) because it influences learning processes and learning experiences. One of the most significant ways that temperament shapes development is by causing children to have different experiences of the environment (Shiner & Caspi, 2012). First, by the fact that temperament shapes the responses evoked by others. Children high in negative emotions are inclined to evoke more negative attempts from parental to exert control, whereas extraverted, self-controlled children tend to receive more parental warmth (Caspi & Shiner, 2006). Second, environmental experiences are interpreted differently depending on children's temperaments. For example, children high in anger/frustration tend to perceive negative events in their lives as more threatening compared to children with lower levels of negative emotions. Temperament traits also make some life outcomes more or less likely to occur (Shiner, 2012). For example, more inhibited children have a somewhat greater chance of developing social anxiety or depression and children high in effortful control tend to develop a stronger social competence and have a lower likelihood of developing behavior problems (Kagan, 2001).

1.4.4.2 Temperament and behavioral disorders

During the last years, research has focused on how temperamental characteristics are related to disorder development. Several studies (Johnson, Turner, & Iwata, 2003; Lonigan & Phillips, 2001; Lonigan, Vasey, Phillips, & Hazen, 2004; McFarland, Shankman, Tenke, Bruder, & Klein, 2006) have provided evidence for an association between extreme levels, both very high and very low, of positive or negative reactivity and psychopathology. For example, children with high levels of fear were found to be more prone to internalizing disorders (e.g., anxiety disorders), whereas children high in anger/frustration had a higher chance on developing externalizing disorders (Rothbart & Bates, 2006). This vulnerability for psychopathology is not merely limited to the reactive components but also self-control plays a crucial role in this mechanism (Bijttebier & Roeyers, 2009). Normal reactivity levels combined with very low levels of self-regulation have been associated with ADHD. Moreover, self-regulation supports children in using more positive alternatives (e.g., compliance, empathy, social competence) to problematic behaviors and inhibit tendencies toward anger or avoidance. The child's self-regulatory capacities thus seem to help protect against the effects of high levels of positive or negative reactivity at an early age (Rothbart, 2011). In other words, susceptibility to psychopathology is seen in highly reactive temperaments in combination with low levels of self-regulatory capacities (Lonigan & Phillips, 2001).

1.4.4.3 Temperament and communication disorders

A possible role of temperament has also been studied in other communication disorders, although this is limited to a few studies only. With regard to language development, the different studies seem to point to the fact that temperamental characteristics such as emotionality, approach/withdrawal, and attentional factors may impact language development (Dixon & Shore, 1997; Dixon & Smith, 2000; Paul & Kellogg, 1997; Sajaniemi, Hakamies-Blomqvist, Makela, Avellan, Rita, von Wendt, 2001; Slomkowski, Nelson, Dunn, & Plomin, 1992) but Karrass and Braungart-Rieker (2003) showed that this interaction was also influenced by parenting style.

In the area of voice disorders, children with vocal nodules were shown to be more distractible and acting out (Green, 1989); studies in adults suggested that specific temperamental traits contributed to the dispositional base for vocal nodules and functional dysphonia (Roy & Bless, 2000; Roy, Bless, & Heisey, 2000; Roy, McGrory, Tasko, Bless, Heisey, & Ford, 1997).

1.5 Temperament and developmental stuttering research

1.5.1 Questionnaire-based findings

Most of the temperament studies in stuttering have been done using caregiver questionnaires in children. In what follows we will give a brief overview of the main findings of these studies. Although Fowlie and Cooper (1978) did not actually use a standardized temperament questionnaire but a bipolar adjective checklist, the traits evaluated compare favourably to temperamental traits. Participants were 34 CWS and 34 matched controls (mean age = 8;04 years; range = 6;012-11;02). Mothers of CWS described their children as more anxious, introverted, fearful, sensitive, withdrawn, and insecure.

Lewis and Goldberg (1997) used the Parent Childhood Temperament Questionnaire in a group of 11 preschool-aged children ‘at risk’ for stuttering (mean age = 3;10 years; range = 3;01-4;10) and 11 controls (mean age = 3;09 years; range = 2;11-4;09). Their results showed, contradictory to both earlier and later research findings, CWS to be more adaptable, and have a better quality of mood and higher rhythmicity.

Embrechts, Ebben, Franke, and van de Poel (2000) administered the CBQ to the parents of 30 CWS and a matched control group (mean age = 5;01 years; range = 3;00-7;08). The stuttering group was found to be more active (the level of gross motor activity) and impulsive (the speed of response initiation), and lower in inhibitory control (the capacity to plan/suppress inappropriate approach responses under instructions or in novel or uncertain situations), attentional focusing (the tendency to maintain attentional focus upon task-related channels) and perceptual sensitivity (the detection of low intensity stimuli from the external environment).

Anderson, Pellowski, Conture, and Kelly (2003) used the Behavioral Style Questionnaire (BSQ; McDevitt & Carey, 1978), a parent temperament questionnaire with the parents of 31 CWS and 31 matched controls (mean age = 4;00 years; range = 3;00-5;04). They found CWS to be less distractible (defined as the effectiveness of extraneous stimuli in drawing attention away from ongoing behaviors), lower in adaptability (defined as the ease/difficulty with which behaviors can be changed in a desired way), and lower in biological rhythmicity (defined as the regularity or irregularity of physiologic functions).

Karrass, et al. (2006) administered the BSQ, similar to Anderson et al. (2003), to the parents of 65 CWS and 56 controls (mean age = 4;01 years; range = 3;00-5;11). BSQ-items were clustered to specifically assess reactivity, emotion regulation, and attention regulation. The derived factor of emotional reactivity was defined as the degree of emotional responses. Emotional and attention regulation were respectively defined as the management of emotional responses to situations and using attentional components (e.g., attentional shifting) in order to limit the emotional effects of a stimulus. Results showed that CWS were more emotionally reactive, less able to regulate their emotions, and had poorer attention regulation capacities.

1.5.2 Psychophysical and psychophysiological findings

At the beginning of our study, only a limited amount of studies had been published employing temperament measures in stuttering other than caregiver questionnaires. Guitar (2003) used the acoustic startle response in 14 adults who stuttered (AWS; mean age = 45 years; range = 17-58) and 14 controls (mean age = 47 years; range = 18-62) in combination with the Taylor–Johnson Temperament Analysis. The acoustic startle response measures the amplitude of the eye blink response to a brief pulse of white noise and is considered a physiological measure of reactivity. The eye blink magnitude was found to be significantly higher in the stuttering group, pointing to a higher reactivity. Although no differences were apparent on the overall score of the temperament questionnaire, AWS scored higher on the nervousness-scale.

Alm and Risburg (2007) used a similar acoustic startle paradigm as Guitar's (2003) study but did not find any between group differences in reactivity measured. Participants were 22 AWS (mean age = 39 years; range = 19-58) and 22 controls (mean age = 39 years; range = 24-60).

Finally, Schwenk, Conture, & Walden (2007) employed behavioral observation to study temperament-related behaviors in a laboratory setting. Eighteen CWS (mean age = 3;11 years; range = 3;00-5;04) and 18 CWNS (mean age = 4;03 years; range = 3;02-5;09) were evaluated with regard to attention maintenance and reaction to background stimuli. During a conversation with a caregiver, the child's shifts of attention including duration and latency of shifts to a background camera movement were measured. CWS had more attentional shifts and shorter reaction times. Findings were taken to suggest that CWS, compared to CWNS, were more reactive to, distracted by, and slower to adapt and habituate to environmental stimuli.

1.5.3 Communication-Emotional model of stuttering

Conture and his colleagues (2006) developed a conceptual framework (Figure 1.3) for instances of developmental stuttering, in which previous research findings in the area of stuttering together with findings in the area of temperament and stuttering are incorporated to account for the onset and development of stuttering. The framework includes speech, language, and learning processes, and also temperamental components. Two kinds of contributing components are distinguished, namely distal and proximal contributors. The distal contributors, referring to the conditions that lay the foundations for the variables triggering moments of stuttering, are genetic predisposition and environment. The interaction between genetics (e.g., a slow and/or inefficient lexical retrieval and encoding) and both shared and non-shared, both internal and external environmental variables (e.g., requirements to speak very rapidly) might influence speech-language planning and production (e.g., a system that plans speech/language at rates beyond its abilities to do so efficiently). Several research findings have shown that this speech-language planning and production, including motor speech variables, are atypical or less efficient in CWS. Conture referred to these as the proximal contributors because this level is assumed to be directly responsible for the moments of stuttering. The way in which the child reacts and copes with these speech disruptions might be influenced by his/her emotional reactivity and regulation, resulting in stronger reactions in children with a higher reactivity and lower regulation. Finally, in the model it is speculated that through experience the path of influence between proximal and exacerbating contributors might be strengthened; temperamental factors might increase the amount of speech disruptions and these in turn might trigger more reactivity. The latter process is hypothesized to take place especially in those children where stuttering becomes chronic.

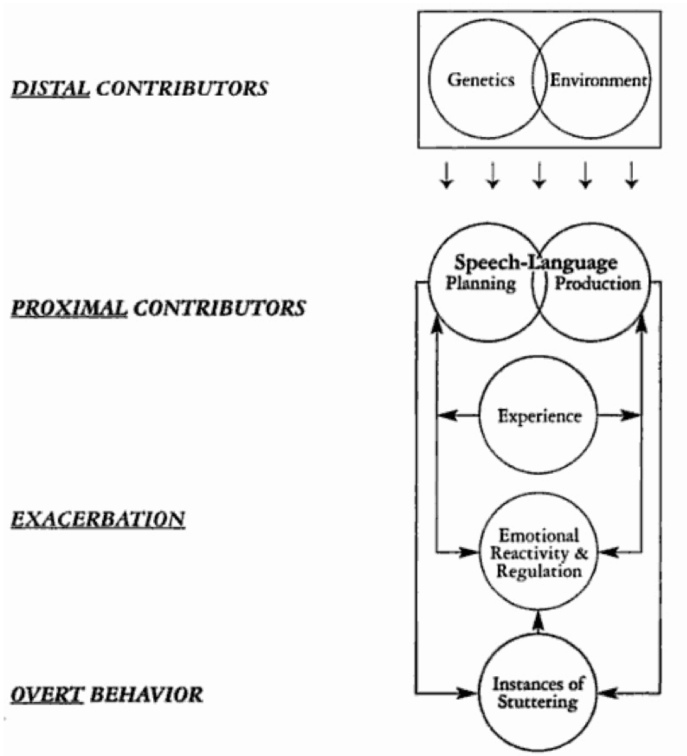


Figure 1.3: Communication-Emotional model of stuttering. (Adapted from Conture et al., 2006)

1.6 Research objectives

In recent years there has been an increased interest in the relationship between temperament and developmental stuttering, as documented by the growing number of studies in this area. Based on Rothbart's taxonomy of positive and negative reactivity versus self-regulation, many of the previously discussed findings in CWS can be understood as pointing toward increased reactivity or reduced self-regulation or a combination of both. Evidence for an increase in positive reactivity includes findings that show CWS, compared to typically developing children (TDC), to be (a) more sensitive and/or reactive to environmental changes and (b) more active and impulsive. Higher ratings on scales for nervousness, anxiousness, fearfulness, and emotional reactivity point to an increase in negative reactivity. Evidence for a reduced self-regulation include data suggesting that CWS are (a) less distractible, (b) lower in adaptability, (c) lower in inhibitory control, (d) less efficient in emotional and attentional regulation, and (e) lower in biological rhythmicity.

Although some of the observed differences between the stuttering and non-stuttering groups in the various studies discussed in the general introduction point to trait differences that can be replicated, results are far from unequivocal, especially since data have been confounded by differences in age, gender, sample size, and measurement tools that were used. Therefore the need for a large-scale, questionnaire-based study on temperament in CWS and an age- and gender-matched control group became apparent; ideally this study also included a third group of children with another communication disorder, e.g. voice disorders, since it was not clear to what extent any temperamental differences are specific to stuttering or are shared with children who experience other forms of communication disorders. Moreover, since most studies that have been carried out in this domain have been questionnaire-based, it is unclear to what extent these preliminary findings are reflected in actual behavioral patterns or can be tested experimentally. Therefore, there was also a scientific need to evaluate if these findings could be corroborated experimentally using neuropsychological measures.

Some of these temperament-based studies have also yielded differences on attentional and inhibitory control-related processes between stuttering and non-stuttering groups, and seem to suggest these processes may be involved in the development and/or maintenance of developmental stuttering. Findings from studies on attentional functioning in stuttering (e.g., Felsenfeld, van Beijsterveldt, & Boomsma, 2010; Karrass et al., 2006; Schwenk et al., 2007) generally have shown that people who stutter tend to be a) less efficient in attention regulation, b) more or less distractible depending on the used methodology, c) less able to allocate attentional resources under dual task conditions, and d) more prone to exhibit attention disorders. Therefore, the study of attentional skills of CWS is potentially important to understanding the nature of the disorder.

While several authors have alluded to a possible role for self-regulatory processes and attentional control processes in the development of stuttering, almost no studies have reported on the more specific concept of inhibitory control in the domain of developmental stuttering. Inhibitory control is the ability to suppress, interrupt or delay an inappropriate response under instructions or in novel or uncertain situations (Clark, 1996; Rothbart, 1989) or to ignore irrelevant information (Dagenbach & Carr, 1994; Dempster & Brainerds, 1995; Rothbart & Posner, 1985). Inhibitory control is essential for the performance of everyday tasks and has been implicated in cognitive development, executive functioning, and the conscious use of attention or attentional control. It is strongly related to the coordination and integration of mental processes in successful task performance and plays an important role in the self-regulation of emotional states (Kochanska, Murray, Jacques, Koenig, & Vandegeest, 1996). There is support for a distinction between endogenous (i.e. autonomously, from within oneself and without external sources) and exogenous response control since different brain regions are involved depending upon whether response control is externally or internally generated (Fernandez-Duque & Posner, 2001; Robertson et al., 1997; Van Den Bergh, 2005). While one study, employing a parent-report questionnaire, reported CWS to be lower in inhibitory control, others found no difference between CWS and TDC (Anderson & Wagovich, 2010). Thus, experimentally testing inhibitory control processes, by using neurocognitive computer paradigms, might also contribute to the insight in the etiology of developmental stuttering.

The general aim of this thesis was to gain in depth insight in the relation between temperament and developmental stuttering, including with the temperamental components of attention and inhibitory control. This general aim was further specified in three main research objectives, as reflected in the different studies.

The first objective was to test in a large population whether CWS, compared to TDC, differed in reactivity and self-regulation, as measured by a parental temperament questionnaire. Based on the available research findings we hypothesized that CWS would have a higher positive and/or negative reactivity and a lower self-regulation. Before we could test this hypothesis, it was important to evaluate the similarity in terms of the underlying temperamental construct for CWS and TDC since the equivalence of temperament structure provided a basis for further comparison of mean group scores on the individual temperament scales. The second objective was to evaluate the underlying attentional component of self-regulation by testing the efficiency of the attentional processes in CWS, through the use of a cognitive computer task. We expected some attentional processes would be less efficient in CWS, compared to TDC. Our final objective was to examine inhibitory control in both populations and we assumed CWS to be less efficient in inhibitory compared to the TDC.

1.7 Thesis outline

Chapter two presents the results of the study comparing the similarity in temperamental construct between CWS and TDC. Hereto, the Dutch Children's Behavior Questionnaire (CBQ-D; Van den Bergh & Ackx, 2003) was administered to the parents of 149 TDC and 69 CWS. The CBQ-D is a 233-item parent-report temperament questionnaire for 3- to 8-year-old children, based on Rothbart's temperament model (Rothbart, Ahadi, Hershey & Fisher, 2001). An additional parent group of 41 children with voice disorders, i.e. vocal nodules, was recruited to evaluate possible differences and/or similarities in underlying temperamental construct between different communication disorders. A principle axis factor analysis was performed on the questionnaire data to extract the factor structure for each participant group. Consequent congruence analyses compared the similarities between the different factor structures. The content of this chapter has been published in Eggers, De Nil, & Van den Bergh (2009).

In **Chapter three** the differences between CWS and TDC on the individual CBQ-D-scales and the overall composite temperament factor scores of Extraversion/Surgency, Negative Affectivity, and Effortful Control, were reported. The 116 children (3;04-8;11 years) who participated in this study, 58 CWS and 58 age- and gender-mached TDC, were part of the larger study described in chapter two. The material presented in this chapter has been published in Eggers, De Nil, & Van den Bergh (2010).

Chapter four concerns our second research objective, namely to evaluate the underlying attentional component of self-regulation. Forty-one CWS and 41 age- and gender-matched controls, ranging from 4;00 to 9;00 years, were administered the Attention Network Test (Fan, McCandliss, Sommer, Raz, & Posner, 2002). This computer task is a combination of a cued reaction time task and a flanker task, and measures the efficiency of 3 distinct attentional networks, i.e. alerting, orienting, and executive attention. This chapter has been published in Eggers, De Nil, & Van den Bergh (2012a).

Chapter five provides the result of the study investigating whether previously reported questionnaire-based differences in inhibitory control would be supported by direct computerized measurement of inhibitory control. Sixty children between 4;10 and 10;00 years of age, 30 CWS and 30 age- and gender-matched controls, completed the Go/NoGo-task of the Amsterdam Neuropsychological Tasks (De Sonneville, 2009). This task measures the capacity to inhibit a preplanned or prepotent response tendency. The material presented in this chapter has been recently accepted for publication (Eggers, De Nil, & Van den Bergh, 2012b).

Chapter six focuses more in depth on two forms of response inhibition namely exogenously triggered and endogenously generated (from within oneself) response inhibition. Participants were 18 CWS and 18 age- and gender-matched controls (age range: 7;04 - 10;11). Endogenous and exogenous response inhibition was assessed by a sustained attention task (Amsterdam Neuropsychological Tasks; De Sonneville, 2009) and a stop signal task (Verbruggen, Logan, & Stevens, 2008) respectively. This chapter has been submitted recently for publication (Eggers, De Nil, & Van den Bergh, 2012c).

In conclusion, **Chapter 7** presents a general discussion on the different results obtained in current research project and discusses future research objectives.

Table 1.3: Chapter overview including main research questions, participant variables, and instruments used.

Chapters	Purpose	Participants					Instruments
		n	*Age mean (SD)*	*Stuttering Severity (SSI-3)*	*Language (TvK) mean Pc (range)*	*Intelligence (WISC-3) mean (range)*	
Chapter 2	The purpose of this study was examining the equivalence of the factorial temperament structure across CWS, TDC, and CWVN.	69 CWS (55♂ & 14♀)	5;10 (1;10)	Mild: 12 Moderate: 45 Severe: 11 Very severe: 1	No reported problems	/	Children's Behavior Questionnaire – Dutch (Van den Bergh & Ackx, 2003)
		146 TDC (83♂ & 63♀)	6;04 (1;10)	/	No reported problems	/	
		41 CWVN (36♂ & 5♀)	7;01 (1;03)	/	No reported problems	/	
Chapter 3	The purpose of this study was to determine whether CWS and CWNS differ from each other on composite temperament factors or on individual temperament scales.	58 CWS (45♂ & 13♀)	5;11 (1;09)	Mild: 8 Moderate: 35 Severe: 14 Very severe: 1	No reported problems	/	Children's Behavior Questionnaire – Dutch (Van den Bergh & Ackx, 2003)
		58 CWNS (45♂ & 13♀)	5;11 (1;10)	/	No reported problems	/	
Chapter 4	The purpose of this study was to evaluate whether previously reported questionnaire-based differences in self-regulatory behaviors between CWS and CWNS would also be reflected in their underlying attentional networks.	41 CWS (31♂ & 10♀)	6;09 (1;05)	Mild: 14 Moderate: 24 Severe: 2 Very severe: 1	Vocab: 63 (23-99) Sentence: 54 (31-96)	Vocab: 19.34 (7-41) Block: 24.73 (6-59)	Baseline speed Task (De Sonneville, 2009) Attention Network Test (Fan, McCandliss, Sommer, Raz, & Posner, 2002)
		41 CWNS (31♂ & 10♀)	6;09 (1;05)	/	Vocab: 72 (23-96) Sentence: 63 (23-94)	Vocab: 19.95 (5-41) Block: 27.98 (4–52)	
Chapter 5	The purpose of this study was to investigate whether previously reported questionnaire-based differences in inhibitory control between CWS and CWNS would be supported by direct measurement of IC using a computer task.	30 CWS (24♂ & 6♀)	7;05 (1;05)	Mild: 13 Moderate: 15 Severe: 1 Very severe: 1	Vocab: 62 (28 - 99) Sentence: 54 (28-99)	Vocab: 19.20 (7-34) Block: 23.77 (6-58)	Baseline speed Task (De Sonneville, 2009) Go/NoGo task (De Sonneville, 2009)
		30 CWNS (24♂ & 6♀)	7;05 (1;05)	/	Vocab: 71 (27 - 97) Sentence: 62 (28-94)	Vocab: 19.73 (7-41) Block: 28.60 (4-52)	
Chapter 6	The purpose of this study was examining relations between children's exogenously triggered and endogenously generated response inhibition and stuttering.	18 CWS (15♂ & 3♀)	9;01 (0;11)	Mild: 9 Moderate: 8 Severe: 1	Vocab: 61 (20-93) Sentence: 56 (20-97)	Vocab: 28.28 (9-39) Block: 42.56 (10-63)	Baseline speed Task (De Sonneville, 2009) Sustained attention task (De Sonneville, 2009) Stop signal task (Verbruggen, Logan, & Stevens, 2008)
		18 CWNS (15♂ & 3♀)	9;01 (1;00)	/	Vocab: 66 (20-96) Sentence: 61 (20-99)	Vocab:29.56 (15-41) Block: 44.50 (19-62)	

CHAPTER 2

Factorial temperament structure in stuttering, voice disordered, and normal speaking children.

The content of this chapter has been published as:
Eggers, K., De Nil, L., & Van den Bergh, B. (2009). Factorial Temperament Structure in Stuttering, Voice Disordered, and Normal Speaking Children. *Journal of Speech, Language, and Hearing Research*, 52, 1610-1622.

Parts of the content of this chapter have been presented as:
Eggers, K. (2007, August). *Temperament structure in stuttering, voice disordered, and typically developing children.* Seminar presented at the Convention of the International Association of Logopedics and Phoniatrics, Copenhagen.

Abstract
Purpose: The purpose of this study was to determine whether the underlying temperamental structure of the Dutch Children's Behavior Questionnaire (CBQ; B. Van den Bergh & M. Ackx, 2003) was identical for children who stutter (CWS), typically developing children (TDC), and children with vocal nodules (CWVN).
Method: A principal axis factor analysis was performed on data obtained with the Dutch CBQ from 69 CWS, 149 TDC, and 41 CWVN. All children were between the ages of 3;0 and 8;11.
Results: Results indicated a 3-factor solution, identified as Extraversion/Surgency, Negative Affect, and Effortful Control, for each of the participant groups, showing considerable similarity to previously published U.S., Chinese, Japanese, and Dutch samples. Congruence coefficients were highest for CWS and TDC and somewhat more modest when comparing CWVN and TDC. The Effortful Control factor consistently yielded the lowest congruence coefficients.
Conclusion: These data confirm that although stuttering, voice-disordered, and typically developing children may differ quantitatively with regard to mean scores on temperament scales, they are similar in terms of their overall underlying temperament structure. The equivalence of temperament structure provides a basis for further comparison of mean group scores on the individual temperament scales.

2.1 Introduction

The temperament model by Rothbart (Derryberry & Rothbart, 1984; Rothbart & Derryberry, 1981), developed in the beginning of 1980s, has received widespread attention in the scientific literature on child temperament. Rothbart (1989a&b) has defined temperament as constitutional differences in both reactivity and self-regulation. As part of her definition, reactivity refers to somatic, autonomic, cognitive, and neuro-endocrine responses to internal and external stimuli. Self-regulation, in turn, are processes serving to modulate this reactivity, such as approach, withdrawal, inhibitory control, and effortful control of attention (Rothbart, 1989a&b).

Within this model it is supposed that there is a continuous interaction between reactivity and self-regulation with the latter one increasing and becoming more under volitional control with increasing age (Eisenberg & Spinrad, 2004; Rothbart & Posner, 1985). Reactivity is expressed through somatic (facial, vocal, and motor), autonomic (e.g., heart rate), cognitive, and neuro-endocrine channels, and is reflected in response parameters like threshold, intensity, and rise time. Reactivity can be either positive or negative depending on stimulus intensity, signal value (e.g., intrinsic meaning), the internal psychological state of the individual, and novelty (Rothbart, 1991). Individuals differ in their threshold for and intensity of positive and negative reactions and the rise and recovery time of these reactions (Rothbart, 1989b). There is evidence for at least two temperament-related control systems, namely fear or behavioral inhibition and attentional (self-regulative) control, with the first emerging earlier in the child's development than the second (Eisenberg, Fabes, Murphy, Maszk, Smith, & Karbon, 1995). Through processes like avoidance and withdrawal (fear), or shifting the attention away (attentional) these control systems play an active role in modulating reactivity.

Most current temperament theories assume a biological basis, including genetic predisposition, for individual differences, an assumption that has gained considerable support from twin and adoption studies (Goldsmith, Buss & Lemery, 1997; Loehlin, 1992; McCrae et al., 2000). Based on heritability estimates, 20 to 60% of individual variation in temperament is generally assumed to be attributed to genetic factors, although some authors provide even higher estimates (Riemann, Angleitner & Strelau, 1997). Thus, 80 to 40% of heterogeneity in the population can be considered environmentally determined, primarily through nonshared environmental influences (e.g., peers, differential parental treatment, accidents) (Saudino, 2005), starting before birth, and individual-specific experiences within the family.

Although temperament refers to relatively stable dispositions (Buss, & Plomin, 1984; Costa & McCrae, 2001), expressions of temperamental traits are susceptible to change given the interaction between temperament and environment (Caspi, 1998) as well as the inherent developmental processes involved in its biological underpinnings

(Goldsmith, 1996; Rothbart, Derryberry & Hershey, 2000). It has been argued that some of these changes can be attributed to a change from predominantly reactivity-driven temperament in infants to more self-regulatory processes in older children (Putnam, Ellis & Rothbart, 2001). In addition, there is evidence that both the stability of temperament across childhood development as well as the developmental changes in temperament are to some extent genetically determined (Saudino, 2005).

A number of age-specific parent-report or self-report measurement instruments have been developed within the theoretical framework proposed by Rothbart (e.g., Capaldi & Rothbart, 1992; Gartstein & Rothbart, 2003). One of the most well-known among these is the Children's Behavior Questionnaire (CBQ; Rothbart, Ahadi, Hershey & Fisher, 2001), a caregiver questionnaire for children between 3 and 7 years of age. This instrument consists of 15 scales. Factor analytic studies have shown that the scales can be grouped under three superfactors. The first factor, labelled as *Extraversion/Surgency*, loads positively on the scales of impulsivity, high intensity pleasure, activity level, approach, smiling/laughter, and negatively on shyness. The second factor, labelled as *Negative Affectivity*, loads positively for the scales of sadness, discomfort, anger/frustration, fear, and negatively for falling reactivity/soothability. The third factor, *Effortful Control*, is positively defined by the scales of low intensity pleasure, inhibitory control, perceptual sensitivity, and attentional focusing (Rothbart & Bates, 1998).

In recent years, there have been a growing number of studies investigating the relationship between temperament and stuttering. Using Rothbart's triad of positive and negative reactivity versus self-regulation, many of the findings from these studies can be understood as pointing towards increased reactivity or reduced self-regulation, or a combination of both in people who stutter. Evidence for an increase in positive reactivity are findings that show CWS to be (a) more sensitive and/or reactive to environmental changes (Fowlie & Cooper, 1978; Glasner, 1949; Guitar, 2003; Oyler (1988) in Zebrowski & Conture; Schwenk, Conture, & Walden, 2007; Wakaba, 1998), and (b) more active and impulsive (Embrechts, Ebben, Franke, & van de Poel, 2000). Higher ratings on scales for nervousness, anxiousness, fearfulness, and emotional reactivity point to an increase in negative reactivity (Fowlie & Cooper, 1978; Guitar, 2003; Karass, et al. 2006). Evidence for a reduced self-regulation are data suggesting that CWS are (a) less distractible (Anderson, Pellowski, Conture, & Kelly, 2003), (b) lower in adaptability (Anderson, Pellowski, Conture, & Kelly, 2003), (c) lower in inhibitory control (Embrechts, Ebben, Franke, & van de Poel, 2000), (d) less efficient in emotional and attentional regulation (Karass, et al., 2006; Schwenk, Conture, & Walden, 2007), and (e) lower in biological rhythmicity (Anderson, Pellowski, Conture, & Kelly, 2003). Although some of the observed differences between the stuttering and the non-stuttering participant groups in the various studies discussed so far may be taken to suggest some common traits, the results are far from unequivocal, especially since data were confounded by differences in age, gender, and test instruments used. Moreover it is not clear to what extent any temperamental differences are specific to stuttering or are shared with children who experience other forms of communication disorders. For instance, language development (Paul & Kellogg, 1997; Sajaniemi, Hakamies-Blomqvist, Makela, Avellan, Rita, von Wendt, 2001), and voice disorders (Green, 1989; Roy & Bless, 2000; Roy, Bless, & Heisey, 2000; Roy, McGrory, Tasko, Bless, Heisey, & Ford, 1997) are other speech-language disorders where possible associations with temperament are being explored. Specifically, the results of the Roy & Bless (2000) study indicated that people with vocal nodules scored higher on the extraversion trait, which is consistent with the higher scores on scales for acting out and distractibility found by Green (1989), which may make this a suitable comparison group for a study on stuttering.

When testing for mean group differences in temperament dimensions between CWS and other participant groups, it is implicitly assumed in most questionnaire-based studies that the underlying temperament structure is the same for the different groups. Byrne, Shavelson, and Marsh (1993) however, argued that this kind of multi-group analysis should ideally be preceded by testing if the structure of the underlying construct being measured is identical for the different groups. If this condition is not met, results of mean group score comparison will be difficult to interpret since possible differences in underlying construct could be confounding the results (Van den Bergh & Van Ranst, 1998). With regard to the CBQ this assumption implies that the pattern and size of the relations between the different CBQ scales are the same across groups. This is an important assumption to be tested as it would determine whether temperamental differences observed between these groups of children constitute either real mean group differences or have possibly more to do with differences in the underlying temperamental make-up.

Therefore the purpose of this study was to examine the equivalence of the factorial temperament structure across CWS, TDC, and children with vocal nodules (CWVN), and by doing so, laying the basis for further comparison of mean group scores on the individual temperament scales (Eggers, De Nil, & Van den Bergh, 2008). This was preceded by a reliability analysis of the Dutch version of the CBQ for 3-8 year olds since previous analysis by Van den Bergh & Ackx (2003) was only performed for a group of 8- and 9-year olds.

2.2 Method

2.2.1 Participants

Data were collected on 256 participants: 69 CWS (14 girls and 55 boys), 146 normal speaking children (controls, 63 girls and 83 boys), and 41 CWVN (5 girls and 36 boys). Table 2.1 provides participants' age ranges and mean ages per group. Because children under the age of four are only rarely referred to an ear, nose, and throat (ENT) specialist for voice problems resulting from vocal nodules (Dobres, Lee, Stemple, Klummer & Kretschmer, 1990) no CWVN were included in the youngest age group.

All of the participants were native Dutch speakers and had neither reported speech, language & hearing problems - except for stuttering in the CWS, and voice disorder in the CWVN - nor neurological, or psychological disorders. All CWS produced at least three or more within-word disfluencies (sound/syllable repetition, prolongation or blocks) and/or monosyllabic word repetition in 100 words of spontaneous speech (Conture, 2001), and scored at least 'mild' on a standardised stuttering severity test. Stuttering severity was assessed by the child's own speech-language therapist, based on the Stuttering Severity Instrument-3 (SSI-3; Riley, 1994) or the Test for Stutter Severity-Readers/Non-Readers (TvS-L/NL; Boey, 2000). All therapists were qualified stuttering treatment experts who had completed a 2-year specialization training program. Seventeen percent of the stuttering children were classified as mild (7 boys and 5 girls), 65% were classified as moderate (38 boys and 7 girls), 16% were rated severe (9 boys and 2 girls), and 1% was found to be very severe (1 boy). Five of the CWS had not received fluency therapy prior to the data collection. The other 64 CWS had received fluency therapy for a period of time ranging from 1 month to 26 months (mean = 8.6 months; SD = 7.1). The CWVN were all diagnosed by an ENT-specialist during laryngoscopic examination as having vocal nodules. Eleven of the CWVN had not yet received voice therapy. The other 30 CWVN were receiving therapy at the time of testing for a period ranging from 1 month to 17 months (mean = 5.2 months; SD = 4.1).

Table 2.1: Age Ranges and Mean Ages of CWS (n = 69), Controls (n = 146) and CWVN (n = 41) for 3- & 4-, 5- & 6-, and 7- & 8-Year Olds.

	CWS			Controls			CWVN		
Age group	*n*	*Range*	*M*	*n*	*Range*	*M*	*n*	*Range*	*M*
Age 3 & 4	26	3;02 - 4;11	4;01	40	3;01 - 4;11	3;10	0	-	-
Age 5 & 6	22	5;00 - 6;11	5;10	38	5;00 - 6;11	6;01	20	5;00 - 6;11	5;11
Age 7 & 8	21	7;02 - 8;11	8;03	68	7;00 - 8;11	8;00	21	7;01 - 8;11	8;01

2.2.2 Detection Instrument for Stuttering

The 'Detection Instrument for Stuttering' (DIS; Stes & Boey, 1997), a parental questionnaire, was completed by all parents. The DIS is a Dutch, validated screening instrument for stuttering. It consists of 6 questions exploring core and secondary behaviors, frequency of disfluencies, attitude of the child towards speaking, listener reactions, and time since onset of the disfluencies (e.g., 'What kind of disfluencies does your child produce', 'How does your child react during these disfluencies'). Each question is scored, weighed and a total score is calculated. Total scores range between 6 and 21. A score below 8 indicates the absence stuttering while scores above 12 are an indication for stuttering. Scores between 8 and 12 are in need of further testing. The instrument has a sensitivity of 0.92, a specificity of 0.88 and a predictive value of 1.05 (based on a sample of 513 children, 77 CWS and 436 controls). In order to assure that the normally fluent children as well as the voice disordered children did not experience any stuttering difficulties, all children in the two control groups who scored higher than a '6', the lowest score possible, were excluded from the study.

2.2.3 Temperament Questionnaire

Temperament of the participants was evaluated by making use of the Dutch version of 'Children's Behavior Questionnaire' (CBQ; Van den Bergh & Ackx (2003). The CBQ is a parent-report questionnaire aimed at children between the age of 3 to 7 years (up to 9 year olds for the Dutch version), and thus incorporates traits that are present in early childhood as well as later developing traits. It originally consisted of 15 subscales. Three scales (motor activation, excitatory control, and attentional shifting) were later added by Rothbart and colleagues, as documented in Van den Bergh and Ackx (2003), resulting in a total of 234 questions (233 in the Dutch version). In order to complete the questionnaire, parents rate each item using a 7-point Likert scale (1 = extremely untrue of your child, 2 = quite untrue of your child, 3 = slightly untrue of your child, 4 = neither true nor false of your child, 5 = slightly true of your child, 6 = quite true of your child, and 7 = extremely true of your child). When the child has not been observed in a situation as described in one of the items, a "not applicable" response option is provided. The CBQ scale descriptions and sample items are presented in Table 1.2.

2.2.4 Procedure

All of the participating CWS and CWVN were contacted through their speech-language therapist. Absence of any other speech, language, and hearing problems, and neurological or psychological disorders was checked by the SLP. Children with reported concomitant problems, or children who had ever had concomitant problems in the past, were not selected as participants. The instructions for completing the questionnaires were given by the speech-language pathologist to the parents of the participants. The parents were asked to fill out the questionnaire at home and to return it at the following treatment session. The CBQ, DIS-form, together with the scores on the standardised stuttering severity test - which was administered by the speech-language therapist -were returned in a self-addressed and stamped envelope. Since the vast majority of the questionnaires were completed by the mothers, only these were used; 7 questionnaires, completed by the father or by the father and the mother together, were discarded.

The healthy control participants were recruited from three elementary schools. The questionnaires, with the written instructions, and the DIS-forms were distributed through the schools. The questionnaires were returned in a closed envelope to the school teacher where they were collected. Absence of speech, language, and hearing problems, as well as neurological or psychological disorders was determined by a qualified speech-language pathologist (first author) and based on parental and teacher reports combined with reports from Centers for Student Guidance who evaluate each school-age child once during each academic year for speech, language, and hearing problems. Reasons for exclusion from the control group included reported language and articulation difficulties, possible ADHD, and other reported problems, which could have affected the outcome of our results.

2.2.5 Data analysis

Internal consistency (Cronbach α) of the CBQ scales was determined. Reliability differences between scales of different participant groups were tested using the program ‘Alphatest’ (Hox, 2006).

An exploratory factor analysis, using principle axis factoring, based on the matrix of intercorrelations of the scores on the original 15 CBQ scales was used to investigate the factor structure. Factor analysis is a multivariate statistical technique used to reduce a large number of variables into a smaller number of latent variables or ‘factors’. In this study we wanted to uncover the underlying structure for the 15 CBQ scales. Figure 2.1 gives a schematic overview of the exploratory factor analysis. The extent to which a factor plays a role in the score on an individual variable is expressed in a weighing factor ranging from 0 (no role) to 1 (fully determined), also called ‘factor loading’. Mathematically this is the correlation between the variable and the factor. Therefore the squared factor loading indicates the proportion of variance in that variable explained for by the factor (common variance). The total variance for all variables explained by a factor is called the ‘eigenvalue’. To facilitate the interpretation of a factor structure it is feasible that a) every variable has a high factor loading on just one factor, b) every factor consists of more then one variable, and c) most factor loadings are either high or low. Since this is usually not the case, it is often necessary to redefine the factors using a rotation method (Figure 2.2). Oblique rotations allow for the factors to be correlated, while orthogonal methods do not. Analogous to Rothbart et al. (2001), we used an oblique rotation method to obtain a clear pattern of loadings. The extracted factors were rotated using a Promax rotation.
Kaiser’s criterion, stating that only factors with eigenvalues greater than or equal to 1 should be retained, was used (Bryant & Yarnold, 1995; Maris, 2003).

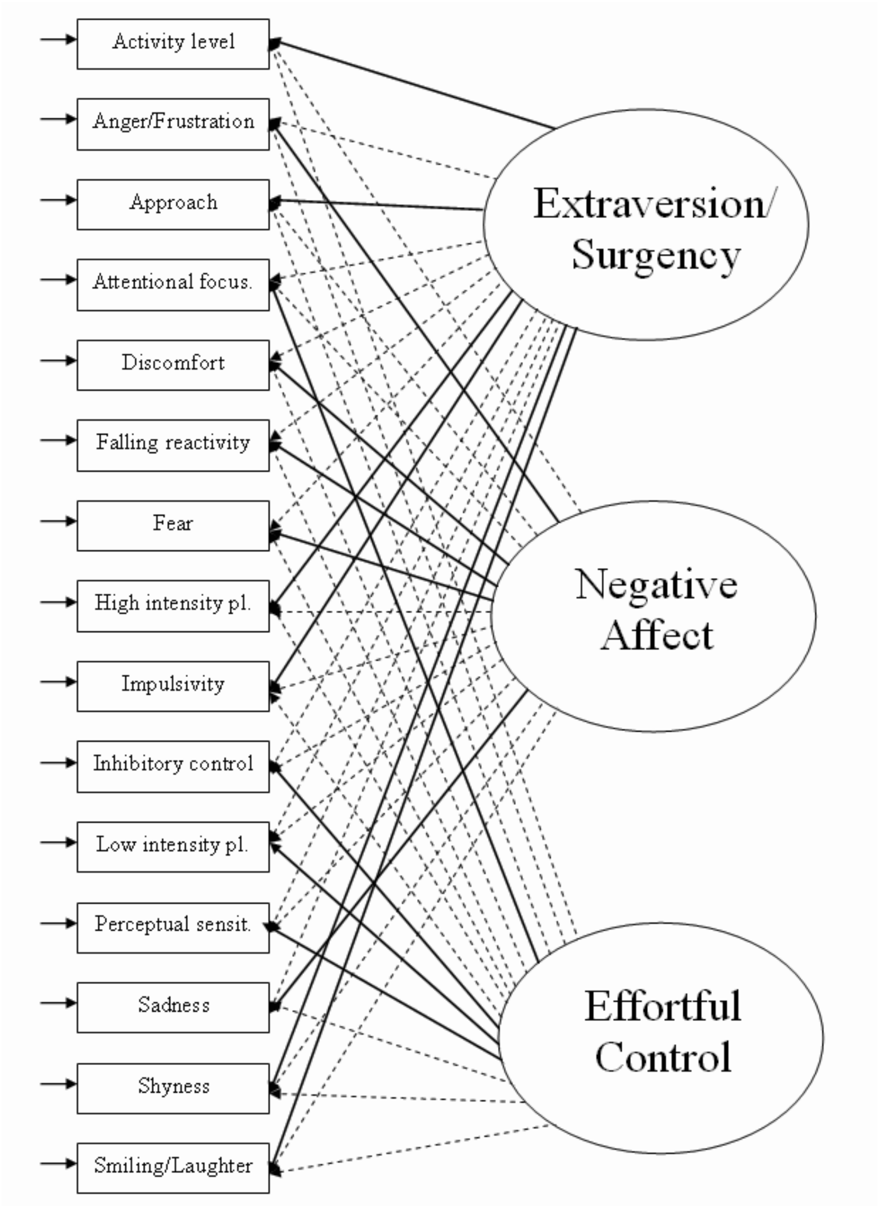


Figure 2.1: Schematic overview of the exploratory factor analysis.

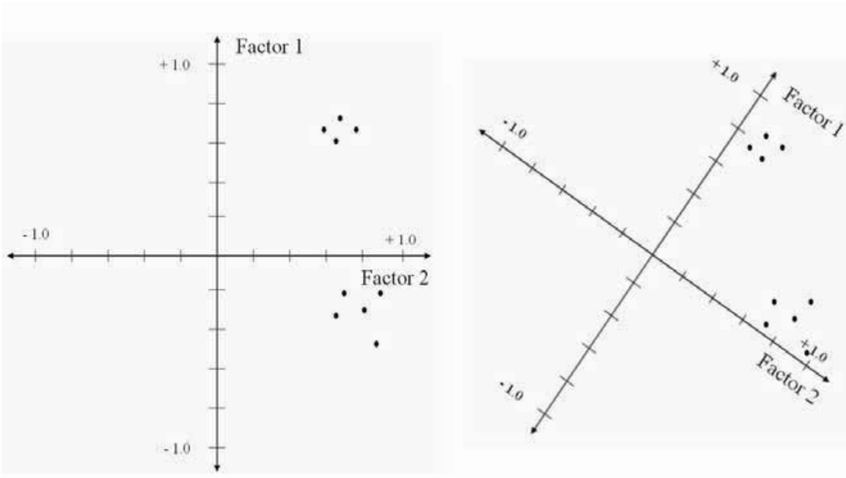


Figure 2.2: Illustration of rotation of a 2-factor solution after initial factor analysis.

Factor congruence coefficients were computed to asses the similarity of the mutual structure for the TDC, CWS, and CWVN. The use of congruence coefficients is quite common in literature (Korth & Tucker, 1975; Koschat & Swayne, 1991; Rolland, Parker, & Stumpf, 1998; Sakamoto, Kijima, Tomoda, & Kambara, 1998; Shek, 1988; Voeten & van den Bercken, 2003) and examines the extent to which the scales have similar loadings on two compared factors. In this analysis the scales are treated as cases and the factors as variables. The correlation is computed between the loadings that the scales have on the 3 factors in each sample. The coefficient takes values from 1.00 (perfect positive similarity) through 0 (total dissimilarity) to -1.00 (perfect negative similarity).

Different authors use different reference systems for the amount of congruence. Sakamoto et al. labeled coefficient values above .80 as 'high' and above .90 as 'very high'. Koschat and Swayne claimed factors were virtually equal above coefficient value of .85.

2.3 Results

2.3.1 Dutch CBQ scale reliabilities

Table 2.2 presents the internal reliability coefficients (Cronbach α) for the data reported in this paper and previous studies. The internal reliability coefficients of the current samples ranged from .58 (low intensity pleasure) to .91 (shyness) and all of the scales, except for the scale of low intensity pleasure in one participant group, have coefficients above .60. There was an average internal consistency of .75 for the 15 scales. These data compare favorably with previous Dutch and US data as reported by Van den Bergh and Ackx (2003) and Rothbart et al. (2001), which were also added in Table 2.2.

Comparison of reliabilities shows that the scales of activity level and attentional focusing are more reliable in the Dutch version whereas the scale of smiling/laughter is more reliable in the English version. The scales of low intensity pleasure and shyness were less reliable for some of the participant groups in the Dutch version. Cronbach α's did not differ significantly for the other scales.

Table 2.2: CBQ scales internal reliability coefficients (Cronbach α) for the current study, the Dutch samples (Van den Bergh & Ackx, 2003), and the US samples (Rothbart, Ahadi, Hershey, & Fisher, 2001).

	Current study			**Previous studies**		
Scales	**TDC**[a] (3;01 - 8;11)	**CWS**[b] (3;02 - 8;11)	**CWVN**[c] (5;00 - 8;11)	**Dutch**[d] (8;10 – 9;02)	**US**[e] (4;00 – 5;11)	**US**[f] (6;00 – 7;11)
Activity level **	.79	.87	.81	.83	.75	.75
Anger/frustration	.80	.80	.83	.84	.80	.81
Approach	.71	.80	.75	.81	.74	.77
Attentional focusing***	.83	.80	.80	.87	.67	.69
Discomfort	.72	.73	.82	.66	.73	.67
Falling reactivity/soothability	.69	.67	.60	.47	.66	.67
Fear	.77	.75	.65	.73	.70	.70
High intensity pleasure	.76	.83	.79	.77	.79	.77
Impulsivity	.74	.78	.74	.72	.74	.78
Inhibitory control	.78	.75	.73	.82	.76	.78
Low intensity pleasure *	.75	.58	.69	.55	.64	.73
Perceptual sensitivity	.77	.67	.79	.71	.64	.71
Sadness	.68	.61	.79	.56	.69	.71
Shyness **	.88	.90	.91	.84	.92	.92
Smiling/laughter **	.64	.70	.68	.67	.75	.80

[a] *N* = 146, [b] *N* = 69, [c] *N* = 41, [d] *N* = 71, [e] *N* = 228, [f] *N* = 183
Results overall p-value Alphatest: *p < .05, **p < .01, ***p < .0001

2.3.2 Structure of Dutch CBQ scales

2.3.2.1 Factor structure of typically developing children

Table 2.3 illustrates the factor pattern matrix for the 3 participant groups. For the TDC three factors were found with eigenvalues greater than 1, in total accounting for 49.77% of the explained variance. The first factor (eigenvalue = 3.06; 20.11% explained variance) was defined by the scales impulsivity, activity level, high intensity pleasure, approach, smiling & laughter, and a negative loading on shyness. The size of the loadings were equivalent to those reported by Rothbart et al. (2001) but higher than those reported by Van den Bergh and Ackx (2003), except for a slightly lower loading for smiling/laughter and shyness. The scales approach and shyness also loaded on factor 2, consistent with the findings by Rothbart et al. and Van den Bergh and Ackx. In contrast to the study by Van den Bergh and Ackx, smiling/laughter also loaded on factor 3, analogous to Rothbart, et al.

Table 2.3: Factor pattern based on 15 subscales of the Children's Behavior Questionnaire (CBQ; Rothbart, Ahadi, Hershey, & Fisher, 2001) of the TDC (n = 146), CWS (n = 69), and CWVN (n = 41).

Scales	Factor loadings								
	Factor 1			Factor 2			Factor 3		
	TDC	CWS	CWV	TDC	CWS	CWV	TDC	CWS	CWV
Extraversion/Surgency									
Impulsivity	.858	.917	.824	-.292	-.096	-.109	.014	-.020	-.067
Activity level	.750	.850	.721	-.004	-.055	.272	-.181	-.059	-.262
High intensity pleasure	.754	.708	.662	-.144	-.118	-.066	-.103	.052	-.054
Approach	.517	.489	.478	.346	.377	.615	.253	.110	.361
Shyness	-.510	-.555	-.472	.415	.271	.479	-.047	-.049	-.070
Smiling/laughter	.498	.399	.493	-.042	-.068	-.153	.623	.636	.373
Negative Affect									
Discomfort	-.074	-.196	-.466	.737	.750	.646	.125	.126	.069
Sadness	-.046	.044	-.081	.695	.832	.766	.068	.069	-.099
Anger/frustration	.394	.247	.191	.647	.751	.815	-.101	-.013	.038
Fear	-.201	-.283	-.257	.544	.467	.661	.141	-.063	-.190
Falling	.213	.209	.078	-.455	-.729	-.735	.230	.026	-.103
Effortful Control									
Low intensity pleasure	-.061	-.188	-.191	.160	.071	-.104	.733	.651	.827
Perceptual sensitivity	-.009	.046	-.128	.191	.191	.411	.622	.695	.507
Inhibitory control	-.332	-.565	-.351	-.292	-.377	-.351	.529	.279	.196
Attentional focusing	-.225	-.093	.087	-.142	-.092	-.087	.430	.543	.660

Note. Factor loadings > /.30/ are highlighted.

The second factor (eigenvalue = 2.51; 16.55% explained variance) was defined by positive loadings for the scales discomfort, sadness, anger/frustration, fear, and a negative loading on the scale of falling reactivity/soothability. The size of the loadings were comparable or even higher as those in the study by Rothbart et al. (2001) except for a slight lower loading on falling reactivity/soothability. In contrast to the results in the study by Van den Bergh and Ackx (2003), anger/frustration also loaded on factor 1 although this is in line with findings by Rothbart et al. (2001) in 6 and 7 year olds.

The third factor (eigenvalue = 1.99; 13.10% explained variance) had high loadings on low intensity pleasure, perceptual sensitivity, inhibitory control, and attentional focusing. Inhibitory control also loads, although negatively, on factor 1, conform Rothbart et al. (2001) and Van den Bergh and Ackx (2003). The strength of the loadings is comparable to Rothbart et al.

2.3.2.2 Factor structure of children who stutter

The principal factor analysis of the CBQ scores obtained for the CWS identified three factors with eigenvalues greater than 1. The three factors together explained 53.53% of the variance. The first factor (eigenvalue = 3.36; 22.22% explained variance) loaded on the same scales as those observed for the control participants, except for anger/frustration. The scales impulsivity, activity level, shyness, and inhibitory control had a higher loading whereas the loading for all the other scales was lower than that observed for the control group.

Factor 2 (eigenvalue = 3.01; 19.87% explained variance), included the same scales as the control group, except for shyness, plus the inhibitory control scale. All scales, except the fear scale, had stronger loadings to those in the control group.

The final factor (eigenvalue = 1.73; 11.44% explained variance), was composed of the same scales as those for the control group except for inhibitory control. All scales except low intensity pleasure loaded higher then the TDC.

2.3.2.3 Factor structure of children with vocal nodules

Three factors were recovered and together they accounted for 56.79% of the explained variance. In contrast to the other 2 participant groups, the strongest factor (eigenvalue = 3.68; 25.00% explained variance) for the CWVN contained the scales of anger/frustration, sadness, falling reactivity/soothability, fear, and discomfort, which complies with the second factor in the other two participant groups. Similar to the CWS but contrary to the TDC, inhibitory control also loaded negatively on this factor. The fact that perceptual sensitivity also loaded was different from Rothbart et al. (2001) but consistent with Van den Bergh and Ackx (2003). All the scales, except for discomfort loaded higher compared to the TDC.

The second extracted factor (eigenvalue = 2.85; 19.36% explained variance), had similar components as the first extracted factor in the control group, with the exception of the anger/frustration scale which did not load on this factor for the children in this group, similar to those in the stuttering group. In contrast, the discomfort scale did load on the second factor. The scales of discomfort and inhibitory control had higher loadings then for the TDC; while loadings on the other scales were lower or almost identical.

In contrast to the other participant groups, the third factor (eigenvalue = 1.83; 12.43% explained variance) comprised the approach scale. Similar to the CWS this factor did not include inhibitory control. Low intensity pleasure, attentional focusing, and approach loaded higher whereas perceptual sensitivity and smiling/laughter had a lower loading.

2.3.3 Factor congruence coefficients

Congruence analysis results based on the rotated factor matrices of the three participant groups are shown in Table 2.4. The resulting congruence coefficients ranged from .86 to .97. The highest congruence in factor structure was found between the CWS and the TDC with coefficients ranging from .94 (Effortful Control) to .97 (Extraversion/Surgency and Negative Affect). The comparable congruence coefficients for the CWVN were somewhat more modest, ranging from .86 (Effortful Control) to .94 (Extraversion/Surgency and Negative Affect). The congruence was found to be the lowest for the superfactor of Effortful Control.

Table 2.4: Factor congruence coefficients based on the comparison of the TDC (n = 146; mean age = 6;4), CWS (n = 69; mean age = 5;1), and CWVN (n = 41; mean age = 7;1).

	Factor		
	1	**2**	**3**
Groups	**Extraversion/Surgency**	**Negative Affect**	**Effortful Control**
TDC versus CWS	.975	.974	.938
CWS versus CWVN	.961	.951	.905
TDC versus CWVN	.940	.940	.858

2.4 Discussion

Earlier studies have already reported mean group differences in temperament dimensions between stuttering, voice disordered, and normal speaking individuals (Anderson, Pellowski, Conture, & Kelly, 2003; Embrechts, Ebben, Franke, & van de Poel, 2000; Fowlie & Cooper, 1978; Glasner, 1949; Guitar, 2003; Karass, et al., 2006; Lewis & Goldberg, 1997; Oyler (1988) in Zebrowski & Conture; Roy & Bless, 2000; Schwenk, Conture, & Walden, 2007; Wakaba, 1998) but it is unclear to what extent the underlying structure of the temperamental construct is uniform. The present study used factor analysis to investigate the presence or absence of such structural differences in the factorial temperament matrix.

2.4.1 Dutch CBQ scale reliabilities

Van den Bergh and Ackx (2003) translated the CBQ (Rothbart, Ahadi, Hershey & Fisher, 2001) into Dutch and their internal consistency data have already shown the Dutch version of the CBQ to be a reliable temperament questionnaire for 8- and 9-year-olds. Reliability analysis in this study shows that this also applies for 3- to 8-year-olds. Average reliability ($\alpha = .75$) was identical to Rothbart et al.'s findings in 6- and 7-year-olds and somewhat higher compared to her 4- and 5-year-old group ($\alpha = .73$). We compared scale reliabilities using Hox's Alphatest and reported on significant global differences between the Dutch and the English version. Although this test also allows for post hoc pairwise group comparison, these data would not have added significantly to our study.

2.4.2 Structure of temperament

The principle axis factor analysis performed on the CBQ data of the control group, revealed a three-factor solution. Because the loading on these factors was almost identical to earlier reported data based on American (Rothbart, Ahadi, Hershey & Fisher, 2001), Chinese (Ahadi, Rothbart, & Ye, 1993), Japanese (Kusanagi, 1993), and Dutch samples (Van den Bergh & Ackx, 2003), it seems justifiable to label these factors with the descriptors used in these previous studies. Therefore, we will refer to these factors as Extraversion/Surgency, Negative Affectivity, and Effortful Control. In addition, given the parallel between our results and those reported by Van den Bergh and Ackx, the obtained reliability coefficients and the factor structure confirmed both the validity of the Dutch questionnaire and the validity of our control group.

Similar to the control group, the factor analysis for the CWS and CWVN derived three superfactors. With regard to the factor *Extraversion/Surgency*, CWS had higher loadings on impulsivity, activity level, and shyness, although the last one had a negative loading. The speed in which responses are generated (impulsivity) and gross motor activity seem to be the most determining scales in this superfactor for the CWS. Absence of inhibitory control for the CWS was more associated with Extraversion/Surgency then the control group. For the CWVN, on the other hand, the loadings for all scales were typically lower than for the other two groups, except for the loading on smiling/laughter, which was equivalent to the TDC; contrary to the other groups the absence of discomfort was also associated with this superfactor.

All of the scales related to *Negative Affect*, except for discomfort and fear, loaded higher for the CWS and the CWVN compared to the TDC. The discomfort scale loaded higher for the CWS, and fear loaded higher for the CWVN. Higher loadings compared to the control subjects was especially noticeable for the falling reactivity/soothability scale. These findings may be taken to suggest that in the two groups of children with communication disorders, this trait plays a more important role in the make-up of the three temperamental factors, compared to the typical developing children. When confronted with distressing situations, the lack of soothability will more rapidly evoke feelings of negativity such as discomfort, sadness, or frustration.
Opposite to the TDC, the absence of inhibitory control was associated with Negative Affect in CWS and to a lesser degree in CWVN. In CWVN, perceptual sensitivity was also associated with Negative Affect. It may be the case that perceptual sensitivity in this group will lead to an increase in Negative Affect.

With regard to the superfactor of *Effortful Control* the loadings on the attentional focusing scale were higher for CWS, and even more for CWVN. The attentional skills addressed in inhibitory control tasks have been related to the development of the executive attention system (Gartstein & Rothbart, 2003) and higher loadings on attentional focusing were linked to lower loadings on inhibitory control. This is consistent with the lower loadings found for the CWS and CWVN on the scale of inhibitory control. In both of these groups the capacity to plan and to suppress inappropriate responses seems less present as a contributing factor in self-control. The fact that the loadings for low intensity pleasure gradually increase when we compared CWS, TDC, and CWVN may be explained by the increasing mean age of these participant groups. For the same reason an increase in the loadings for perceptual sensitivity could have been expected although a different pattern emerged. In the group of CWS we saw that the perceptual sensitivity scale had higher loadings than the control group. The fact that in the CWVN group, which had the oldest mean age, this scale contributed the least to the factor of Effortful Control, probably demonstrates that this group is least able to use the detection of low intensity stimuli as a self-regulative tool. Finally, in CWVN the amount of positive anticipation for pleasurable activities (approach) seems more related to the factor of Effortful Control compared to other groups; probably an effect that can be partly attributed to the lowest loading of all participant groups on inhibitory control.

2.4.3 Congruence of temperament structures

Congruence coefficients show that the similarity in the factorial matrix is least strong between the TDC and the CWVN. One could assume that this is partly due to mean age differences of the participant groups, with the CWVN having the highest mean age. This is also reflected in the fact that of the three superfactors, Effortful Control seemed to yield the lowest similarity between all groups, which is consistent with the idea that the self-regulative system becomes more influential with increasing age (Rothbart & Posner, 1985).

Based on the high congruence coefficients between the TDC, the CWS, and the CWVN, we can conclude that the factor structure of these three groups is highly similar. Appropriate methods, using Jöreskog and Sörbom's (2006) LISREL confirmatory factor analyses, could be applied to gain more insight in the factorial invariance, including equivalence of measurement and equivalence of structure. Van den Bergh and De Rycke (2003) describe how after establishing well-fitting baseline models for each group, equality of factor loadings, error variances per item, and correlations between factors can be tested in constrained and less restrictive models.

2.5 Conclusion

While we have discussed some congruence and scale loading differences between the groups in the previous section, it is important to emphasize that these differences were all very subtle differences and as such should not be overinterpreted. Given the high similarity in temperament structure between the three groups, we can tentatively conclude that possible group differences on the individual temperament scales between CWS, CWVN, and TDC reflect real differences and are not a consequence of differences in underlying temperamental construct.

What remains to be seen is whether these similarities in underlying temperament structure are specific to the model suggested by Rothbart et al. (2001) or can be confirmed in other theoretical models as well. It also remains to be investigated whether differences on temperamental scales result in measurable differences in observable behavior between these groups of children. Further research, based on direct observations, behavioral experiments, psychophysical, and/or psycho physiological measures is therefore needed for a multidimensional and more in depth view of possible differences and relationships. This would also overcome possible limitations of parental questionnaires, as addressed by some authors (e.g., Kagan, 2001; Strelau, 1998; Vaughn, Taraldson, Cuchton, & Egeland, 2002), i.e. susceptibility to parental bias and low interparental agreement, but contested by others (e.g., Rothbart & Bates, 1998; Slabach, Morrow, & Wachs, 1991), who would argue for the satisfactory test-retest reliability, cross-time stability, as well as a moderate to strong degree of validity of these questionnaires. Finally, it remains to be seen whether temperamental differences can be translated into disorder specific, diagnostic, and therapeutic intervention strategies.

CHAPTER 3

Temperament dimensions in stuttering and typically developing children.

The content of this chapter has been published as:
Eggers, K., De Nil, L., & Van den Bergh, B. (2010). Temperament dimensions in stuttering and typically developing children. *Journal of Fluency Disorders*, 35, 355-372.

Parts of the content of this chapter have been presented as:
Eggers, K. (2010, October). *Stuttering, temperament, and attention.* Seminar presented at the Kolloquium zu Störungen der Kommunikation und Kognition, RWTH-Aachen University, Aachen.

Eggers, K., De Nil, L, & Van den Bergh, B. (2010, August). *Attention set shifting in children who stutter.* Poster presented at the Convention of the International Association of Logopedics and Phoniatrics, Athens.

Abstract
Purpose: The purpose of this study was to determine whether children who stutter (CWS) and typically developing children (TDC) differ from each other on composite temperament factors or on individual temperament scales.
Methods: Participants consisted of 116 age and gender-matched CWS and TDC (3.04–8.11). Temperament was assessed with a Dutch version of the Children's Behavior Questionnaire (CBQ-D; Van den Bergh and Ackx, 2003), a caregiver rating scale.
Results: Results indicated significant differences between participant groups on the composite temperament factors of Negative Affectivity, and Effortful Control. Analysis of the individual temperament scales showed that CWS, compared to the TDC, scored significantly lower on the scales of 'Inhibitory Control' and 'Attentional Shifting' and higher on the scales of 'Anger/Frustration', 'Approach' and 'Motor Activation'. Stuttering severity and months of therapy were not associated with either of the temperament dimensions.
Conclusions: The present study provides data that support the hypothesis that CWS and TDC differ on both composite temperament factors and temperament scales. The findings were interpreted within existing frameworks of temperament development, as well as with regard to previous studies of temperament in CWS.

3.1 Introduction

The purpose of this study was to investigate the relationship between temperament and developmental stuttering, using the Children's Behavior Questionnaire – Dutch (CBQ-D; B. Van den Bergh & M. Ackx, 2003), a parental temperament questionnaire. Because temperament has been defined and interpreted differently in the last decades, we start the introduction by addressing the concept of temperament. This is followed by a brief review of the role of temperament in the development of behavioral disorders, followed by a review of current research on temperament and developmental stuttering.

3.1.1 The concept of temperament

At present, most theorists agree that temperament refers to biologically based individual differences that are relatively stable over time, and appear early in the child's development (e.g., Goldsmith et al., 1987). Early approaches stressed the importance of stability of these traits (Buss, & Plomin, 1984; Costa & McCrae, 2001) and saw it as a behavioral (Thomas & Chess, 1977) or primarily emotion-oriented style (Goldsmith & Campos, 1982). More recent models acknowledge that temperament itself develops over time (Goldsmith, 1996; Plomin & Dunn, 1986; Rothbart, 1989b), incorporates motivational and self-regulatory systems (Posner & Rothbart, 1998) and is influenced by environmental interactions (Arcus, 2001; Halverson & Deal, 2001; Saudino, 2005).

Rothbart defines temperament as 'constitutionally based individual differences in reactivity and self-regulation' (Rothbart, Ahadi, Hershey, & Fisher, 2001). In her definition, 'reactivity' refers to the arousability of physiological and sensory response systems, and 'self-regulation' are those processes that can modulate (facilitate or inhibit) one's reactivity. 'Constitutional', in turn, is referring to the individual's biological basis, influenced over time by genetics, maturation, and experience. In other words, the temperament structure changes over time, from a predominantly reactivity-driven concept in infants to a structure with more emphasis on self-regulatory processes in older children (Putnam, Ellis & Rothbart, 2001). In order to assess temperamental characteristics, Rothbart developed a number of questionnaires aimed at different age ranges. The Children's Behavior Questionnaire (CBQ; Rothbart, Ahadi, Hershey, & Fisher, 2001) assesses temperament in early to middle childhood and consists of 15 temperament scales. Factor-analyses of these scales repeatedly have revealed 3 composite temperament factors, namely positive reactivity (i.e., the tendency to actively and energetically approach new experiences in an emotionally positive way), negative reactivity (i.e., the tendency to be sad, fearful, easily frustrated, and irritable), and effortful control (i.e., the ability to sustain attention, control one's behavior, and regulate one's emotions) (Ahadi, Rohbart, & Ye, 1993; Eggers, De Nil, & Van den Bergh, 2009; Kusanagi, 1993; Rothbart, Ahadi, Hershey,

& Fisher, 2001; Van den Bergh & Ackx, 2003). In the CBQ, Positive Reactivity (or Extraversion/Surgency) comprises the scales Impulsivity, Activity Level, High Intensity Pleasure, Motor Activation, Shyness, Approach, and Smiling/Laughter. Negative Reactivity (or Negative Affectivity) comprises the scales Anger/Frustration, Discomfort, Sadness, Fear, and Falling Reactivity/Soothability. Low Intensity Pleasure, Inhibitory Control, Perceptual Sensitivity, Attentional Focusing, Attentional Shifting, and Excitatory Control cluster under Effortful Control (Van den Bergh & Ackx, 2003; for scale definitions and questionnaire sample items see Table 3.1).

3.1.2 Temperament as a moderator in the development of behavioral disorders

Child temperament researchers recognize how both innate individual differences and the environmental context shape children's behavior. In particular, temperamental concepts are being used to explain behavioral and physiological patterns, and responses that are evoked under conditions of stress (e.g., novelty situations, interaction with unfamiliar persons, intense stimuli), and conditioned responses to certain stimuli (Gray, 1987; Strelau, 2001). Moreover, the idea of temperament predisposing the susceptibility for or moderating the development of certain disorders (e.g., anxiety disorders) has received widespread attention in health psychology literature (e.g., Kubzansky, Martin, & Buka, 2009; Puttonen, et al., 2008; Smith, & Williams, 1992; Williams, Wiebe, & Smith, 1992). Recent integration of temperamental research and childhood psychopathology (Frick, 2004; Nigg, & Goldsmith, 1998; Rettew & McKee, 2005) has created new insights in possible ways of temperament interaction. For instance, there is mounting empirical evidence that both reactive temperamental factors (Extraversion/Surgency & Negative Affectivity) as well as regulative processes (Effortful Control) play an important role in the onset, development and maintenance of disorders such as anxiety disorders (Bijttebier & Roeyers, 2009; Lonigan & Vasey, 2009). Temperament dimensions have also been identified as important individual characteristics influencing the child's reaction to specific types of treatment (Mash, 2006) and moderating or mediating treatment outcome in various disorders such as anxiety disorders (Rapee & Jacobs, 2002) and attention deficit and hyperactivity disorder (Purper-Ouakil, et al., 2010).

3.1.3 Temperament and developmental stuttering

Recently, several researchers have considered the potential role of temperament in the onset and development of stuttering. For instance, Conture and colleagues (Conture et al., 2006) have proposed the 'Communication-Emotional model of stuttering'. In this model, distal (i.e. genetics and environment) and proximal contributors (i.e. speech-language planning and production) are linked with exacerbating factors (i.e. experience, emotional reactivity and regulation) and overt stuttering behaviors. They hypothesized that children begin to stutter as a result of deficiencies in speech-language planning and production. The presence of emotional reactivity may lead some children, after continued experience with stuttering, to react stronger to these disfluencies. Trying to cope with these disfluencies (regulation) may interact directly with linguistic planning and execution. Furthermore, Conture et al. suggested that this emotional reactivity/regulation consequently may lead to changes in disfluency types, duration, and/or physical tension. However some recent findings do not seem to support this extension of their model (Mulcahy, Hennessey, Beilby, & Byrnes, 2008).

Other authors have speculated about the possible significance of temperament and/or temperament-related concepts (such as sensitivity towards stuttering, perfectionism, frustration tolerance, anxiety) for understanding the onset, development, and even treatment efficacy of stuttering (Anderson, Pellowski, Conture, & Kelly, 2003; Amster, 1995; Conture, 1991; Conture, 2001; Conture & Melnick, 1999; Embrechts, Ebben, Franke, & van de Poel, 2000; Felsenfeld, 1997; Gregory, 2003; Guitar, 1976, 1998, 2003; Karrass, et al., 2006; Lewis & Goldberg, 1997; Messenger, Onslow, Packman, & Menzies, 2004; Oyler (1998) in Zebrowski & Conture; Wakaba, 1998). Guitar (1998) for example speculates that some CWS might be born with a heightened emotional sensitivity (hypersensitivity) making them "*especially reactive to their early disfluencies*" (pp. 83).

Temperament also may be a useful concept to understand the influence of stress and specific stressors on stuttering. It has been demonstrated that stuttering can be influenced by emotional reactions as a result of situational stress (e.g., Alm, 2004a; Blood, Blood, Bennett, Simpson, & Sussman, 1994; Ezrati-Vinacour & Levin, 2004; Menzies, Onslow, & Packman, 1999; Peters & Hulstijn, 1984). Temperament differences may affect the susceptibility of individuals to learning processes and experiences. For instance, specific temperament traits, such as extraversion, make some individuals more susceptible to particular classical and operant conditioning processes (Gray, 1991), which are known to have an important role in the development of compensatory and other behaviours in stuttering (Bloodstein, 1995; Brutten & Shoemaker, 1967; Kamhi, 2003).

Previous studies, employing parental temperament questionnaires, have reported temperament differences between CWS and typically developing children (TDC) (Anderson, Pellowski, Conture, & Kelly, 2003; Embrechts, Ebben, Franke, & van de Poel, 2000; Karrass, et al., 2006; Lewis & Goldberg, 1997; Oyler (1998) in Zebrowski & Conture; Wakaba, 1998) and provided support for the interactional patterns of temperament and stuttering described above. The results of these studies, in general, showed that CWS scored significantly lower on scales related to self-regulation (e.g., inhibitory control, adaptability), and higher on reactivity related scales (e.g., activity, impulsivity) compared to children in the control group (for a more detailed overview: see Eggers, De Nil, & Van den Bergh, 2009).

In the present study, the Children's Behavior Questionnaire (CBQ; Rothbart, Ahadi, Hershey, & Fisher, 2001) was used to study temperament dimensions of stuttering and nonstuttering children. Several approaches can be used for assessing temperament, including behavioral observations in natural (home) or in more structured semi-natural (laboratory) settings (e.g., Goldsmith & Rothbart, 1991), interviews (e.g., Garrison, Biggs, & Williams, 1990; Thomas & Chess, 1977), temperament questionnaires (e.g., Carey & McDevitt, 1978; Duijsens, Spinhoven, Verschuur, & Eurelings-Bontekoe, 1999; Gartstein & Rothbart, 2003), or psychophysical and psychophysiological indicators (Kagan, 1998; McManis, Kagan, Snidman, & Woodward, 2002), as well as a combination of two or more of these methods. Although temperament questionnaires can be susceptible to parental bias and inaccuracy (Vaughn, Taraldson, Cuchton, & Egeland, 2002), they tap into the vast knowledge of caregivers who have experienced the child's reactions in different situations and over a long period of time. Also, several studies have shown satisfactory test-retest reliability (Slabach, Morrow, & Wachs, 1991), as well as a moderate to strong degree of validity for parental reports (Rothbart & Bates, 1998). Our choice for using the CBQ was based on three elements: (a) the theoretical basis of the questionnaire (e.g., multidimensionality), (b) the availability of a reliable and valid Dutch translation (Van den Bergh & Ackx, 2003), and (c) the age range for usability of the CBQ. Moreover, one of the greatest advantages of this theory-derived instrument, compared to other questionnaires, is that it includes scales that measure traits developing past infancy, resulting in a more varied and detailed view on temperament at that age. In addition, the CBQ not only focuses on behavioral style characteristics but also includes affective qualities, such as strength and speed of responses to emotional stimulation.

Prior to the current study, we investigated whether the structure of the underlying construct being measured was identical for both participant groups. Although there were some minor scale loading differences, the factor-analyses of the CBQ-data obtained from CWS, TDC, and children with vocal nodules in this preceding study (Eggers, De Nil, & Van den Bergh, 2009), revealed, a similar and highly congruent three-factor temperament structure. Given this similarity in temperament structure between the participant groups we can conclude that if group differences between CWS and TDC on the composite temperament factors (e.g., Effortful Control) or individual temperament scales (e.g., Inhibitory Control) would emerge, they reflect real differences and are not confounded by differences in underlying temperamental make-up between the two groups (Byrne, Shavelson, and Marsh, 1993).

Based on our overview of the literature, we know that high levels of negative reactivity and low levels of effortful control have an impact on disorder onset (e.g., Bijttebier & Roeyers, 2009), and that temperament plays a role in stress responses, conditioning processes (e.g., Gray, 1987), and treatment outcome (e.g., Purper-Ouakil, et al., 2010). With regard to stuttering, studies in CWS have revealed elevated scores on reactivity-related scales and lowered scores on self-regulatory scales (for an overview: see Eggers, De Nil, & Van den Bergh, 2009). However, while many of the findings from these previous studies in stuttering can be understood as pointing toward increased reactivity or reduced self-regulation, with the exception of a study by Embrechts, Ebben, Franke, & van de Poel (2000), who used a preliminary version of the CBQ, no studies employed a questionnaire specifically conceptualized to measure the triad of positive/negative reactivity and self-regulation in a highly integrated manner. Moreover, previous researchers did not validate their results by evaluating if the structure of the underlying construct being measured was similar for all groups. Therefore, the current study builds on the results reported in our previous paper in which we showed the underlying temperamental construct was similar for both speaker groups. Specifically, in this study, we used the CBQ, a questionnaire that includes the three composite temperament factors, to investigate the following research questions: (a) do CWS, compared to TDC, have a heightened score on the composite temperament factors of Negative Affectivity and/or Extraversion/Surgency and on some of the individual reactivity-related scales; (b) do CWS, compared to TDC, have a lower score on the composite temperament factor of Effortful Control and on some of the individual scales related to self-regulation; (c) is temperament in CWS associated with length of therapy and/or stuttering severity.

3.2 Method

3.2.1 Participants

The 116 children who participated in this study were part of a larger study which was described in more detail in Eggers, De Nil, and Van den Bergh (2009). TDC were matched for age (± 2 months) and gender (13 girls and 45 boys) to the CWS. If more than one typically developing child could be matched to a child who stuttered, selection occurred at random. The children in the current study ranged in age from 3;04 to 8;11 years. There were 58 CWS (mean age = 5;11 years; SD = 1;09 years) and 58 TDC (mean age= 5;11 years; SD = 1;10 years). All were native Dutch speakers, with no reported speech, language, hearing, neurological, or psychological problems, except for stuttering in the CWS. The participating CWS produced three or more within-word disfluencies (sound/syllable repetition, prolongation or blocks) and/or monosyllabic word repetition per 100 words of spontaneous speech and scored at least 'mild' on either the Stuttering Severity Instrument-3 (Riley, 1994) or the Test for Stutter Severity-Readers/Non-Readers, a Dutch severity scale which has been shown to correlate very high ($r=.94$) with the SSI (Boey, 2000). Eight CWS were classified as mild, 35 as moderate, 14 were rated severe, and 1 was rated very severe. Four of the CWS had not received fluency therapy prior to the data collection. The other 54 CWS had received fluency therapy for a period of time ranging from 1 month to 24 months (mean = 8.9 months; SD = 6.5).

3.2.2 Temperament questionnaire

The Dutch version of the Children's Behavior Questionnaire (CBQ-D; Van den Bergh & Ackx, 2003) was used to assess the temperamental profiles of the participants. As previously stated, the CBQ was developed by Rothbart, Ahadi, Hershey & Fisher (2001), and is a caregiver report measure that provides detailed assessment of temperament in children conforming to Rothbart's reactive and self-regulative model of temperament (Rothbart, 1981, 1989). Although the original English version of the CBQ only included normative data up to age 7, Van den Bergh & Ackx (2003) have demonstrated good reliability and validity for 8-and 9-year-olds, based on a study of 71 children in this age range (average internal consistency of the 18 scales: Cronbach's alpha [α] = .71).

The CBQ-D consists of 18 subscales, 15 original scales and 3 (Motor Activation, Excitatory Control, and Attention Shifting) that were added as part of the Dutch adaptation (Van den Bergh and Ackx, 2003). The subscales, clustering under the 3 composite temperament factors of Extraversion/Surgency, Negative Affectivity, and Effortful Control, are defined in Table 3.1. The CBQ-D consists in a total of 233 items. Each item is rated by the parents using a 7-point Likert scale ranging from "extremely untrue of your child" to "extremely true of your child". When the child has not been observed in a situation as described in an item, a "not applicable" response option is provided. Factor analysis of the 18-scale instrument was completed in order to compute composite temperament factor scores. Although the matching requirements for the current study resulted in a somewhat smaller sample then our 2009 study, the factor analysis was performed on the larger subject sample of our 2009 study (146 TDC and 69 CWS) in order to reduce sampling error and increase factorial structure stability and reliability (MacCallum, Widaman, Zhang, & Hong, 1999). The factor pattern matrix, which is similar to the one reported in 2009, is depicted in Table 3.2. Cronbach's alpha coefficients across the 18 scales averaged .74, indicating a high level of internal consistency (Eggers, De Nil, & Van den Bergh, 2009).

3.2.3 Procedure

CWS were recruited with the assistance of speech-language pathologists practicing in the Dutch speaking part of Belgium. Children who participated in the control group were recruited through their school system. The parents were asked to fill out the CBQ-D questionnaires. All questionnaires included in the current study were completed by the children's mothers. A qualified speech-language pathologist determined absence of other speech, language, and hearing problems; no neurological or psychological disorders were reported. More detailed information on participant recruitment and procedure can be found in the paper by Eggers, De Nil, & Van den Bergh (2009).

Table 3.1: Scale definitions of the Children's Behavior Questionnaire-Dutch (CBQ-D) and sample items (Van den Bergh and Ackx, 2003).

Scale	Definition
Extraversion/Surgency	
1. Impulsivity	The speed of response initiation. *Sample item: Usually rushes into an activity without thinking about it.*
2. Activity level	The level of gross motor activity including rate and extent of locomotion. *Sample item: Moves about actively (runs, climbs, jumps) when playing in the house.*
3. High intensity pleasure	The amount of pleasure or enjoyment related to situations involving high stimulus intensity, rate, complexity, novelty, and incongruity. *Sample item: Likes to play so wild and recklessly that s/he might get hurt.*
4. Motor activation	The amount of excess repetitive small-motor movement, such as finger tapping. *Sample item: Fidgets during quiet activities, such as hearing a story, looking at pictures.*
5. Shyness	Slow or inhibited approach in situations involving novelty or uncertainty. *Sample item: Sometimes prefers to watch rather than join other children playing.*
6. Approach	The amount of excitement and positive anticipation for expected pleasurable activities. *Sample item: Becomes very excited while planning for trips.*
7. Smiling/laughter	The amount of positive affect in response to changes in stimulus intensity, rate, complexity, and incongruity. *Sample item: Laughs a lot at jokes and silly happenings.*
Negative Affect	
8. Anger/frustration	The amount of negative affect related to interruption of ongoing tasks or goal blocking. *Sample item: Gets quite frustrated when prevented from doing something s/he wants to do.*
9. Discomfort	The amount of negative affect related to sensory qualities of stimulation, including intensity, rate or complexity of light, movement, sound or texture. *Sample item: Is quite upset by a little cut or bruise.*
10. Sadness	The amount of negative affect and lowered mood and energy related to exposure to suffering, disappointment, and object loss. *Sample item: Becomes upset when loved relatives or friends are getting ready to leave following a visit.*
11. Fear	The amount of negative affect, including unease, worry or nervousness related to anticipated pain or distress, and/or potentially threatening situations. *Sample item: Is afraid of loud noises.*
12. Falling reactivity/soothability	The rate of recovery from peak distress, excitement or general arousal. *Sample item: Calms down quickly following an exciting event.*
Effortful Control	
13. Low intensity pleasure	The amount of pleasure or enjoyment related to situations involving low stimulus intensity, rate, complexity, novelty, and incongruity. *Sample item: Enjoys "snuggling up" next to a parent.*
14. Inhibitory control	The capacity to plan and to suppress inappropriate approach responses under instructions or in novel or uncertain situations. *Sample item: Can easily stop an activity when s/he is told "no".*
15. Perceptual sensitivity	The amount of detection of slight, low intensity stimuli from the external environment. *Sample item: Is quickly aware of some new items in the living room.*
16. Attentional focusing	The tendency to maintain attentional focus upon task-related channels. *Sample item: When picking up toys or other jobs, usually keeps at the task until it's done.*
17. Attentional shifting	The ability to transfer attentional focus from one activity/task to another. *Sample item: Can easily shift from one activity to another.*
18. Excitatory control	The capacity to perform an action when there is a strong tendency to avoid it. *Sample item: Forces her/himself to complete projects, even when tired.*

Table 3.2: Factor pattern based on the 18 subscales of the Dutch version of the Children's Behavior Questionnaire (CBQ) of the TDC (n = 146) and CWS (n = 69).

	Factor loading					
	Factor 1		Factor 2		Factor 3	
Scale	TDC	CWS	TDC	CWS	TDC	CWS
Extraversion/Surgency						
Impulsivity	.831	.891	.089	.089	-.278	-.124
Activity level	.796	.867	.233	.154	.482	-.160
High intensity pleasure	.780	.708	.004	.036	-.368	-.043
Motor activation	.516	.427	.306	.529	-.583	-.348
Shyness	-.485	-.524	.267	.172	.078	-.013
Approach	.481	.498	.446	.457	-.001	.019
Smiling/laughter	.400	.283	.024	-.137	.411	.552
Negative Affect						
Anger/frustration	.464	.292	.782	.827	-.345	-.129
Discomfort	-.043	-.190	.681	.668	.037	.031
Sadness	.007	.057	.672	.795	-.022	-.082
Fear	-.183	-.264	.447	.401	.156	-.151
Falling reactivity/soothability	.146	.184	-.420	-.666	.204	.115
Effortful Control						
Low intensity pleasure	-.151	-.289	.055	-.130	.716	.603
Inhibitory control	-.409	-.636	-.467	-.577	.711	.434
Perceptual sensitivity	-.060	-.089	.127	-.012	.554	.586
Attentional focusing	-.271	-.182	-.231	-.235	.539	.685
Attentional shifting	-.279	-.478	-.528	-.636	.368	.151
Excitatory control	.377	.167	-.002	.244	-.022	.461

Note. Factor loadings > /.30/ are highlighted.

3.2.4 Data analysis

The data were analyzed using PASW Statistics version 17 for Windows (IBM Company, 2009, Chicago, IL). Based on the CBQ-D 18-scale factor structure three normalized composite factor scores were computed for each participant using the regression method (Distefano, Zhu, & Mîndrilă, 2009). A factor score is a composite variable providing information on the participant's placement on the factor. Several procedures can be used to compute composite factor scores, ranging from non-refined methods (e.g., summing raw scores corresponding to all items loading on a factor) to more refined methods (e.g., regression method), resulting in more exact and valid composite factor scores. The regression method uses standardized information to create composite factor scores, producing standardized scores similar to a z-score metric. The group of 146 TDC was taken as the norm group in which the computed composite factor scores were standardized to a mean of zero. A subsequent analysis of variance (ANOVA) was performed on the composite temperament factor scores of the 58 CWS and 58 TDC of the current study to examine whether significant differences existed between the participant groups on the 3 composite factors. Possible differences between TDC and CWS on the 18 individual temperament scales were also investigated using an ANOVA, with participant group as the independent variable; and composite factor scores and temperament scales as the dependent variables. The dependent variables were tested simultaneously in order to control for Type 1 errors.

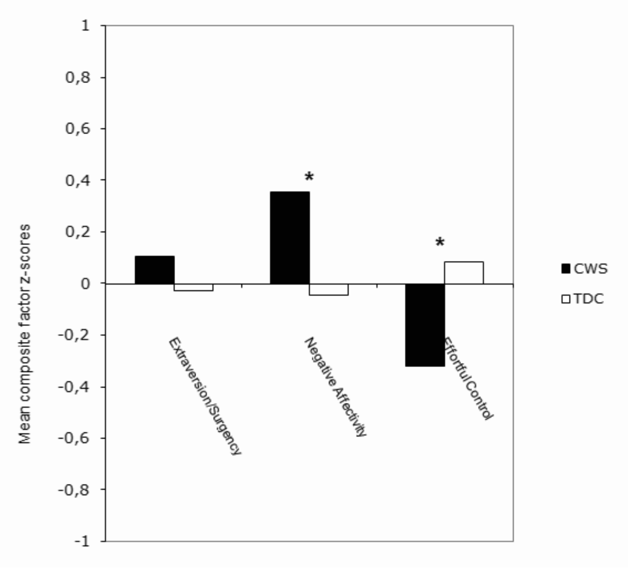
Spearman's rank correlations were calculated to examine the relationships between duration of therapy (in months), stuttering severity (overall severity ratings were used to create four groups of severity: mild, moderate, severe, and very severe), and respectively composite temperament factor scores and individual temperament scales.

3.3 Results

3.3.1 Overall group differences

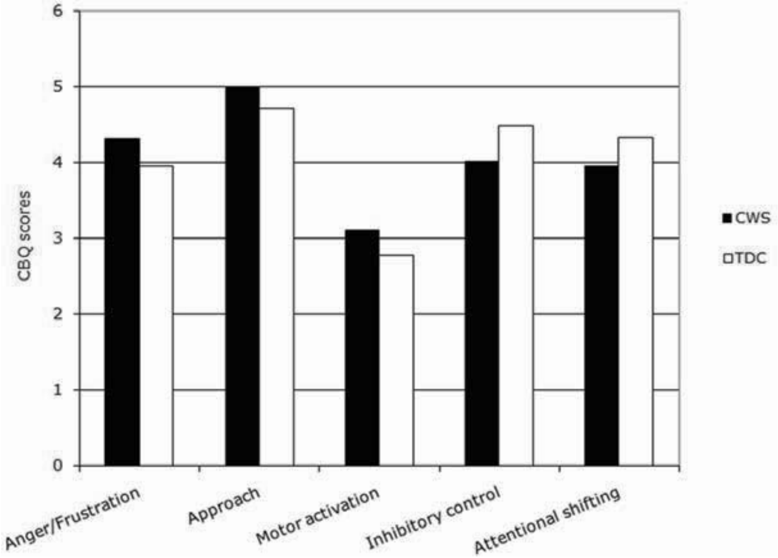
Figure 3.1 gives an overview of the normalized factor scores for the three composite temperament factors for each of the participant groups. There were significant between-group differences found for the composite factor scores Negative Affectivity, $F(1, 114) = 4.49$, $p = .04$ and Effortful Control $F(1, 114) = 5.43$, $p = .02$ (Table 3.3). Extraversion/Surgency did not significantly differ between both groups, $F(1, 114) = .48$, $p = .49$.

The mean scores on the temperament scales and their standard deviations for the CWS and TDC are represented in Table 3.4. Significant between-group differences were found for the scales Anger/Frustration, $F(1, 114) = 4.72$, $p = .03$; Approach, $F(1, 114) = 3.89$, $p = .05$; Motor Activation, $F(1, 114) = 4.34$, $p = .04$; Inhibitory Control $F(1, 114) = 7.49$, $p = .01$; and Attentional Shifting $F(1, 114) = 6.85$, $p = .01$ (Figure 3.2).



* p ≤ .05

Figure 3.1: Mean composite temperament factor z-scores for CWS and TDC on the Children's Behavior Questionnaire (CBQ; Rothbart, Ahadi, Hershey, & Fisher, 2001).



Note. Nonsignificant between-group differences are not included.

Figure 3.2: Mean Scores on the CBQ Scales with Significant Between-Group Differences for CWS and TDC.

Table 3.3: Means, Standard Deviations, and between-group effect analysis of the composite factor z-scores for CWS and TDC on the CBQ.

	CWS		TDC			
Composite factor	***Mean***	***SD***	***Mean***	***SD***	***F***	***p***
Extraversion/Surgency	0.10	1.02	- 0.03	0.99	.483	.49
Negative Affectivity	0.36[a]	1.15	- 0.05[a]	0.88	4.490*	.04*
Effortful Control	- 0.32[a]	0.88	0.09[a]	0.99	5.435*	.02*

[a] Significantly different mean scores

* p ≤ .05

Table 3.4: Means, Standard Deviations, and between-group effect analysis of the CBQ scale scores for CWS and TDC.

	CWS		TDC			
Scale	***M***	***SD***	***M***	***SD***	***F***	***p***
Impulsivity	4.67	0.80	4.56	0.70	.685	.41
Activity level	4.87	0.91	4.59	0.87	2.946	.09
High intensity pleasure	4.30	1.51	4.50	1.11	.711	.40
Motor activation	3.11[a]	0.83	2.77[a]	0.92	4.340*	.04*
Shyness	3.56	1.16	3.48	1.09	.120	.73
Approach	4.99[a]	0.85	4.71[a]	0.65	3.893*	.05*
Smiling/laughter	5.32	0.70	5.50	0.67	2.080	.15
Anger/Frustration	4.32[a]	1.02	3.94[a]	0.84	4.718*	.03*
Discomfort	3.89	1.00	3.72	0.93	.922	.34
Sadness	4.05	0.76	3.95	0.66	.597	.44
Fear	3.92	1.04	3.72	0.86	1.299	.26
Falling reactivity/Soothability	4.73	0.77	4.91	0.69	1.777	.19
Low intensity pleasure	5.06	0.72	5.20	0.77	1.025	.31
Inhibitory control	4.01[a]	0.97	4.47[a]	0.82	7.489*	.01*
Perceptual sensitivity	4.00	0.78	5.07	0.85	.181	.67
Attentional focusing	4.84	0.88	4.92	0.84	.232	.63
Attentional shifting	3.95[a]	0.82	4.32[a]	0.70	6.854*	.01*
Excitatory control	4.61	0.65	4.50	0.66	.928	.34

[a] Significantly different mean scores

* p ≤ .05

3.3.2 Relationship with therapy duration

Two-tailed Spearman's Rank correlations revealed no significant correlations between the duration of therapy (in months) for CWS and the composite factor scores or individual scales that differentiated both participant groups: Negative Affectivity (r_s = .05, p =.70), Effortful Control (r_s = -.02, p =.88), Anger/Frustration (r_s = .06, p = .66), Approach (r_s = -.01, p = .96), Motor activation (r_s = -.25, p = .06), Inhibitory Control (r_s = -.00, p =.99), and Attentional Shifting (r_s = -.10, p = .44). Thus, therapy duration does not seem to be related to any of the differences found in these temperament dimensions. Similarly, no significant correlations were found for any of the other temperament dimensions (Table 3.5 and Table 3.6).

3.3.3 Relationship with stuttering severity

Two-tailed Spearman's Rank correlations indicated that there was no significant correlation between the stuttering severity ratings and the composite temperament factor scores or the temperament scales that differentiated between the CWS and the control group: Negative Affectivity (r_s = .19, p =.15), Effortful Control (r_s = -.16, p =.23), Anger/Frustration (r_s = .11, p = .42), Approach (r_s = .11, p = .43), Motor Activation (r_s = .09, p = .49), Inhibitory Control (r_s = -.15, p =.28), and Attentional Shifting (r_s = -.19, p = .16). In other words, stuttering severity was not found to be related to any of the between group temperamental differences. Similarly no differences were found for the remaining composite temperament factor (Table 3.5) or any of the other scales (Table 3.6).

Table 3.5: Spearman' Rank correlations between composite temperament factor scores and respectively duration of therapy and stuttering severity for CWS.

	Therapy duration		Stuttering severity	
Composite factor	r_s	**p**	r_s	**p**
Extraversion/Surgency	- .078	.56	- .035	.79
Negative Affectivity	.053	.70	.189	.15
Effortful control	- .020	.88	- .160	.23

Table 3.6: *Spearman' Rank correlations between temperament scale scores and respectively duration of therapy and stuttering severity for CWS.*

	Therapy duration		Stuttering severity	
Scale	r_s	***p***	r_s	***p***
Impulsivity	- .160	.23	.079	.56
Activity level	- .031	.82	- .074	.59
High intensity pleasure	.033	.81	- .075	.58
Motor activation	- .247	.06	.094	.49
Shyness	.023	.86	.068	.62
Approach	- .006	.96	.107	.43
Smiling/laughter	.016	.91	- .143	.29
Anger/Frustration	.059	.66	.110	.42
Discomfort	.167	.21	.069	.61
Sadness	- .030	.82	.140	.30
Fear	.111	.41	.253	.06
Falling reactivity/Soothability	- .091	.50	- .219	.10
Low intensity pleasure	.077	.57	- .230	.09
Inhibitory control	- .002	.99	- .148	.28
Perceptual sensitivity	- .233	.08	- .043	.75
Attentional focusing	.015	.91	- .020	.88
Attentional shifting	- .104	.44	- .189	.16
Excitatory control	.060	.65	.010	.94

3.4 Discussion

The primary purpose of our study was to examine whether significant differences could be found in the three composite temperament factors - Extraversion/Surgency, Negative Affect, and Effortful Control - and the individual temperament scales between CWS and TDC. In addition we examined whether both of these temperament dimensions were associated with the stuttering severity and/or treatment duration. The findings of the present parent questionnaire study have confirmed our hypothesis of increased Negative Affectivity and lowered Effortful Control in CWS. In addition, on the individual temperament scales CWS scored lower than TDC in Inhibitory Control and Attentional Shifting, and higher in Motor Activation and Approach (Figure 3.2). These results are consistent with the previous literature describing CWS as more sensitive and/or reactive (Embrechts, Ebben, Franke, & van de Poel, 2000; Fowlie & Cooper, 1978; Glasner, 1949; Guitar, 2003; Karrass, et al., 2006; Oyler (1998) in

Zebrowski & Conture; Wakaba, 1998) and lower in self-regulatory processes (Anderson, Pellowski, Conture, & Kelly, 2003; Embrechts, Ebben, Franke, & van de Poel, 2000; Karrass, et al., 2006).

Byrne, Shavelson, and Marsh (1993) have argued that multigroup analysis should be preceded by testing if the structure of the underlying construct being measured is similar for all groups because results of mean group score comparisons might be confounded by possible differences in underlying construct. Based on our previous (Eggers, De Nil, & Van den Bergh, 2009) and current study in which a high similarity in temperament structure for CWS and TDC was reported, we can state that these found differences reflect real mean group differences and are not a reflection of differences in the underlying temperament structure.
No significant correlation was found between temperament and duration of therapy or stuttering severity for the stuttering group. In the remainder of this paper, we will provide a more in-depth discussion of our findings.

3.4.1 Group differences in composite temperament factor scores

Compared to TDC, CWS scored higher on the composite factor score of Negative Affect, due primarily to the observed between-group difference on Anger/Frustration, and lower on Effortful Control, due to the differential score on Inhibitory Control and Attentional Focusing. There are no other studies in CWS where clustering in these higher order factors was performed, so we cannot directly compare to other studies. However, Karass et al. (2006), using McDevitt & Carey's Behavior Style Questionnaire (BSQ), found significant higher scores for 3-to-5 year old CWS compared to a group of matched TDC on a scale measuring emotional reactivity, and significantly lower scores on scales measuring emotion and attention regulation. Their reactivity and regulation scales were self-constructed by clustering different BSQ-items which rated emotional reactivity and emotion and attention regulation. While their index for emotional reactivity correlated moderately with the CBQ's composite temperament factor of Negative Affect ($r = .42$; $p < .05$; in Karass, et al., 2006), thus corroborating our current findings, no information was provided on the correlation between emotion/attention regulation indices and the CBQ's composite factor Effortful Control.

High levels of positive and/or negative reactivity (i.e., Negative Affect) combined with low levels of Effortful Control were found to play an important role in the onset, development and maintenance of a number of behavioral disorders (Bijttebier & Roeyers, 2009; Lonigan, 2004; Lonigan & Vasey, 2009). Specific, high levels of negative reactivity and low levels of Effortful Control are considered to predispose the onset of these disorders or to influence their development and symptomatology over time (Muris & Ollendick, 2005; Nigg, 2006). Researchers have only recently begon to study the underlying mechanisms and they are still not fully understood.

Lonigan and colleagues (2004) found evidence for an increased attentional bias (increased vigilance and orienting) towards threat-relevant and negatively valenced stimuli in individuals with high Negative Affect. This increase in attentional bias was only present when they also scored lower on Effortful Control. Individuals high in Negative Affect and Effortful Control were able to compensate for their reactive attentional bias through their high capacity for attentional control (Lonigan & Vasey, 2009). Given that research in other developmental disorders has provided evidence of a link between temperament and disorder onset, development, and maintenance, one could easily extend this observed link to developmental stuttering. This hypothesis is in line with earlier suggestions made by Seery et al. (2007) and Yairi (2007), that the temperamental dimensions observed in all or some CWS may be related to stuttering development and symptomatology. Furthermore, the speculation by Lonigan & Vasey that high efficiency of Effortful Control could act as a protective factor for the development of behavioral disorders also deserves further investigation with regard to stuttering.

The advantage of composite temperament factors, the highest order traits, is their enormous bandwidth, making them ideal for detecting general distinctions in temperament between participant groups, although they are not as valuable for predicting specific behavioral patterns; the disadvantage is that in any hierarchical representation, one always loses information as one moves up the hierarchical levels (John & Srivasta, 2001). In other words, item information is inevitably lost when aggregated into scales, and scale information is lost when aggregated into composite factors (John, Hampson, & Goldberg, 1991). Comparing participant groups on the three composite temperament factors will therefore give a rather broad overview of possible differences, but is not specific enough to provide adequate insight in the more fine-grained temperament dimensions. For that reason, we also compared our participant groups at the lower level of the hierarchy, i.e. the 18 temperament scales, which provides a clearer, more accurate view and offers information that is otherwise masked at a higher level (Briggs, 1989).

3.4.2 Group differences in individual temperament scales

3.4.2.1 CWS scored higher on Anger/Frustration, Approach, & Motor Activation

The analysis of the lower levels of the temperamental hierarchy revealed that CWS, when compared to normally fluent peers, scored higher on Anger/Frustration, Approach, and Motor Activation. Rothbart et al. (2001) defined Anger/Frustration as the amount of negative affect related to interruption of ongoing tasks or goal blocking. Others have also described low frustration tolerance, a concept similar to Anger/Frustration, as a component with the potential to influence the development of stuttering (e.g., Hill, 1999; Riley & Riley, 1979). Starkweather (2002) even speculates on the possibility of low tolerance for frustration as one of the genetically transmitted traits influencing the probability that a child will develop stuttering.

The higher ratings on Approach, which is defined as the amount of excitement and positive anticipation for expected pleasurable activities, might result in an increase in situational stress, especially when combined with the increased scores on Anger/Frustration. Approach behaviors are believed to be activated through the Behavioral Approach System (BAS), which has been associated with the basal ganglia and its projections, and is moderated by the neurotransmitter dopamine (Gray, 1991). It could be that the lower scores on Inhibitory Control, a trait that can regulate approach tendencies, have an impact on Approach, which in turn would resulting in an increase of the latter.

Motor Activation is defined as the amount of excess motor movement such as eye blinking, finger tapping, muscle twitching, fidgeting, and chewing fingernails. While at first glance some of these excessive motor movements could be related to secondary stuttering behaviors, the items in the Children's Behavior Questionnaire are formulated in such a way that these movements are not related to the speech act itself. Instead, within the context of temperament research, motor activity is seen as an indication of positive reactivity. This is thought to suggest that in stressful situations people with a higher motor reactivity are easier aroused which in turn can be reflected in an overall increased muscular tension (Kagan, 1998). Therefore, this finding might suggest that CWS increase overall muscular tension more easily in demanding situations. Building on Bloodstein's (1995) suggestion that the more localized increased facial and/or glottal muscle tension during moments of stuttering in CWS might reflect the emerging extra muscular effort when anticipating difficulties in their speech, one could speculate that CWS are more prone to reacting with increased muscular tension during speech disruptions compared to TDC. This could also map onto Guitar's (1998) speculation that increased muscular tension may act as a precipitator of speech disruptions.

3.4.2.2 CWS scored lower on Inhibitory Control & Attentional Shifting

CWS compared to the control participants scored lower on Inhibitory Control and Attentional Shifting, indicating that they are less able to suppress inappropriate approach responses under instructions or in new or uncertain situations or to shift attention from one activity to another. Our finding is in line with the results of the study by Embrechts, Ebben, Franke, and van de Poel (2000), and similar results have been reported in a number of other studies that have used the BSQ. Although the CBQ scales of Attentional Focusing and Shifting are not represented as such in the BSQ, they are related to the BSQ-scales of Attention Span/Persistence, and Distractibility (McDevitt & Carey, 1978; Thomas & Chess, 1977), for which differences between stuttering and nonstuttering children have been reported (Anderson, Pellowski, Conture, & Kelly, 2003; Karrass, et al. 2006).

It has been argued that children are able to regulate their behavior through the use of two different systems, namely an emotional system (fear) and a later developing more attention based system (e.g., effortful control) (Rothbart, Ahadi, & Evans, 2000). Children that are able to volitionally use their attention, by focusing on or shifting away, can easier inhibit dominant inappropriate responses (Kochanska, 1997). Neuroimaging data (e.g., Casey et al. 1995) have provided evidence for the presence of three attentional networks related to alehypothrting, orienting and executive control. The main focus of the executive attention network, also named the anterior attention system, is monitoring and handling conflict situations and is involved in the regulation of emotional reactivity as well as cognitive processing. It has been associated with the anterior cingulate gyrus and the basal ganglia and is assumed to be modulated through the neurotransmitter dopamine (Posner, Rothbart, & Rueda, 2003). Inhibitory control and attentional shifting (sometimes included in the attentional focusing scale), which according to some authors is the primary index for effortful control, are the scales most related to tasks requiring the anterior attentional network (Davis, Bruce, & Gunnar, 2002). Because this network is linked to the activation of the Anterior Cingulate and the basal ganglia and these cortical and subcortical structures are implicated in some neural models of developmental stuttering (e.g., Alm, 2004b; Smits-Bandstra & De Nil, 2007), one might therefore speculate that the lower scores on Inhibitory Control and Attentional Shifting are the result of a lower efficiency in the anterior attentional network.

3.4.3 Relationship with therapy duration and stuttering severity

Our results did not point to a significant correlation between treatment duration and composite factor scores or temperament scales. This might suggest that temperament characteristics are robust against therapy interventions lasting a few weeks to a few months; or alternatively, that treatment approaches would need to address temperament directly in order to result in significant changes. On the other hand this might suggest that duration of direct speech treatment and counseling are not necessarily associated with intersubject variability of temperament scale scores. However, treatment or the type of treatment was not directly controlled in this study and as such any conclusions regarding the predictive value of temperament dimensions for treatment duration are tentative at best.

We also found no association between any of the temperament dimensions and overall stuttering severity. This does not mean however that temperament cannot be associated with specific aspects of stuttering severity, such as duration of stuttering moments and secondary behaviors. Gray (1991) already hypothesized, based on his classification of three emotional response systems (BIS, behavioral inhibition system; BAS, behavioral activation system; and the fight/flight system), that one individual with a specific temperamental constellation may be more prone to struggle or escape behaviors while another is more prone to a ‘freezing’ reaction. This was also mentioned by Guitar (1998). Unfortunately, our data did not allow for a differential correlation of these various elements that may make up severity, but such more detailed correlational analyses should be attempted in future studies.

3.4.4 Implications for the role of temperament in developmental stuttering

Although our methodology does not allow for a direct causal linkage between temperament and stuttering, one could speculate about the role temperament may play in the onset and/or development of stuttering and treatment outcome through one or more of several interaction pathways, namely a) as a moderator in stress-related situations, b) as a moderator in conditioning processes, and/or c) as suggested by Conture et al. (2006), as a moderator in linguistic processing. First, temperament could act as a moderator in stress-related situations. Our present findings, pointing in the direction of an increased reactivity combined with a limited self-regulation for CWS, could mean that when confronted with moments of stuttering and/or abnormal disfluencies in their speech, children react stronger to these interruptions in combination with a feeling of loss of control emerging faster due to the lack of adequate self-regulation.

Alternatively, there could also be an indirect pathway by which children evaluate a novel situation more easily as stressful (i.e., reactivity), resulting from difficulty in shifting their attention away from such situation (i.e., regulation) and having trouble selecting the proper reaction. This in turn may lead to increased negative emotional arousal, which has been associated with conditioning processes (LeDoux, 1994). In addition, increased excitement for certain activities in CWS could further complicate their behavior.

Finally, our findings might also be interpreted by the possibility of temperament serving as a moderator in linguistic processing. Wolfe and Bell (2002) found evidence that parental ratings of Inhibitory Control and Attentional Focusing (including shifting) were related to working memory. Low scores on these temperament scales led to lowered scores on working memory tasks, such as yes-no and Stroop-like tasks (e.g., MacLeod, 1991). In addition, performance on these tasks (i.e. yes-no and Stroop-like tasks) was also negatively correlated to scores on the approach scale. Both findings were also apparent in our group of stuttering children. Working memory, responsible for storing and processing incoming information, plays a crucial role in language processing. Baddely's model (2003) of working memory is comprised of three subsystems, including a phonological working memory (i.e. phonological loop), which consists of a limited capacity phonological storage and an articulatory rehearsal system. This phonological loop plays an important role in speech-language acquisition, comprehension, and production (Adams & Gathercole, 1995; Montgomery & Windsor, 2007). Based on Levelt's model, speech-language production can be divided in three distinct phases: conceptualization, formulation, and articulation (Postma & Kolk, 1993). During the formulation stage grammatical and phonological encoding, i.e. selecting the appropriate words and phonemes, take place. Working memory is essential for storing these encoded sequences before they are being articulated. Central to this language production model is the role of several monitoring loops and various authors have suggested stuttering to be a result of aberrant monitoring (Bernstein Ratner & Wijnen, 2006; Postma & Kolk, 1993). While still speculative, our findings could map onto this hypothesis because central attentional control functions are key to the monitoring loops of this system.

3.4.5 Caveats and suggestions for future research

While we have discussed primarily the scales that showed between-group differences, some might argue that an equally important finding is that most scales did not yield significant differences. However, one would not expect to find differences on the majority of the temperament scales since it would be most unlikely that they all have a role in the onset and development of stuttering. Furthermore, it may be important to emphasize that high or low scores on temperamental traits do not point to clinical disorders but are merely a reflection of the distribution of scores on a bipolar continuum in a normal population. For instance, a lower group mean score on inhibitory control is not indicative of any disorder but only reflects a lower efficiency in suppressing inappropriate approach responses.

Every method used for assessing temperament has some limitations, including (parental) questionnaires (cf. supra). While most of the temperament research is based on questionnaire assessment procedures for reasons discussed in the introduction (Strelau, 1998), our current findings need to be replicated using experimental methods that allow us to directly observe and measure behavioral patterns. Such experimental paradigms also will allow us to analyze the existence of a direct link, if any, between temperament and stuttering.

3.5 Conclusion

The present study provides further data supporting the hypothesis that CWS and typically developing children differ in temperamental characteristics. Evidence was found for heightened reactivity (higher in Negative Affect, and in individual scales of Anger/Frustration, Motor Activation, and Approach) and limited processes of self-control (lower in Effortful Control, and in individual scales of Inhibitory Control and Attentional Shifting).

While communication disorders may worsen or improve over time, temperament is supposed to be relatively stable (Rothbart, Derryberry, & Hershey, 2000). So the study of temperament may offer a way to capture pre-onset causal, trigger or contributing factors as opposed to reactive effects of the communication disorder.

Trying to formulate clinical considerations is quite premature since further research, based on direct observations, behavioral experiments, psychophysical, and/or psycho physiological measures is still needed for a multidimensional and more detailed view of possible differences and relationships. However, such research may provide additional information that would allow clinicians to match treatment approach with specific temperamental patterns observed in individual CWS. It also may shed further light on issue of spontaneous recovery and treatment outcome. If the current results are confirmed in follow-up studies, they could validate the frequently described use of desensitization training for the moments of stuttering but also for specific stress inducing stimuli or environments (e.g., Gregory, 2003; Shapiro, 1999; Van Riper, 1973) as this will reduce the reactivity of a child towards certain stimuli. It would also illustrate the importance of parental guidance (e.g., Rustin, Boterill, & Kelman, 1996; Shapiro, 1999) in young CWS, training parents to react adequately to certain behaviors and situations. Children with heightened Negative Reactivity may benefit from a less protective parenting style, allowing them to acquire essential coping strategies, while children with lowered Effortful Control may experience more difficulties with an authoritarian or permissive parenting style (Kristal, 2005; Kochanska, 1993).

CHAPTER 4

The efficiency of attentional networks in children who stutter.

The content of this chapter has been published as:
Eggers, K., De Nil, L., & Van den Bergh, B. (2012a). The Efficiency of Attentional Networks in Children who Stutter. *Journal of Speech, Language, and Hearing Research,* 55, 946-959.

Parts of the content of this chapter have been presented as:
Eggers, K. (2011, November). *The role of attention in developmental stuttering.* Keynote lecture presented at the annual symposium of the Panhellenic Association of Logopedics.

Eggers, K. (2011, January). *Attentional networks and inhibitory processes in developmental stuttering.* Lecture presented at the Escola Superior de Saude do Alcoitão, Portugal.

Eggers, K., De Nil, L, & Van den Bergh, B. (2010, August). *Attention set shifting in children who stutter.* Poster presented at the Convention of the International Association of Logopedics and Phoniatrics, Athens.

Eggers, K., De Nil, L, & Van den Bergh, B. (2009, August). *Attention processes in children who stutter.* Seminar presented at the 6th Congress of the International Fluency Association, Rio de Janeiro.

Eggers, K. (2008, April). *Attentional processes in children who stutter*. Lecture presented at the First European Symposium on Fluency Disorders, Antwerp.

Abstract
Purpose: The purpose of this study was to evaluate whether previously reported questionnaire-based differences in selfregulatory behaviors (Eggers, De Nil, & Van den Bergh, 2009, 2010) between children who stutter (CWS) and children who do not stutter (CWNS) would also be reflected in their underlying attentional networks.

Method: Participants consisted of 41 CWS (mean age = 6;09) and 41 CWNS (mean age = 6;09) ranging in age from 4;00 to 9;00. Participants were matched on age and gender. The efficiency of the attentional networks was assessed by using the computerized Attention Network Test (Fan, McCandliss, Sommer, Raz, & Posner, 2002).
Results: Primary results indicated that CWS had a significantly lower efficiency of the orienting network compared with CWNS, whereas no differences were found on the alerting or executive control network.
Conclusion: Current findings corroborate previously found differences in self-regulatory behavior and were taken to suggest a possible role for attentional processes in developmental stuttering.

4.1 Introduction

Several researchers have found evidence suggesting that attentional processes may be involved in the development and/or maintenance of developmental stuttering (Bajaj, 2007; Bernstein Ratner & Wijnen, 2006; Bosshardt, 1999, 2002, 2006; Bosshardt, Ballmer, & De Nil, 2002; Felsenfeld, van Beijsterveldt, & Boomsma, 2010; Foundas, Corey, Hurley, & Heilman, 2004; Karrass, et al., 2006; Oomen & Postma, 2001; Schwenk, Conture, & Walden, 2007; Smits-Bandstra & De Nil, 2007; Van Lieshout, Hulstijn, & Peters, 1996; Vasic & Wijnen, 2005; Webster, 1990). Findings from these studies generally have shown that people who stutter tend to be a) less efficient in attention regulation (Felsenfeld et al., 2010; Karass et al., 2006), b) more (Embrechts, Ebben, Franke, & van de Poel, 2000; Schwenk et al., 2007) or less distractible (Anderson, Pellowski, Conture, & Kelly, 2003), c) less able to allocate attentional resources under dual task conditions (Bosshardt, 1999, 2002, 2006; Bosshardt et al., 2002; Smits-Bandstra & De Nil, 2007; Vasic & Wijnen, 2005), and d) more prone to exhibit attention disorders (Alm & Risberg, 2007; Felsenfeld et al., 2010; Monfrais-Pfauwadel & Lacombe, 2002). Therefore, the study of attentional skills of CWS is potentially important to understanding the nature of the disorder.

In a recent series of studies (Eggers, De Nil, & Van den Bergh, 2009, 2010), we examined aspects of attention in CWS using a parent-report temperament questionnaire, the Children's Behavior Questionnaire (CBQ; Rothbart, Ahadi, Hershey, & Fisher, 2001). The CBQ was developed by Rothbart and is based on her process-oriented and multidimensional model of temperament. She defines temperament in terms of individual differences in reactivity and self-regulation (Rothbart, 2011). Reactivity refers to the arousability of physiological and sensory response systems, and can be measured through response threshold, latency and intensity. It is operationalized in the CBQ in positive (e.g., Approach, Activity Level) and negative (e.g., Anger/Frustration, Fear) reactivity scales. Self-regulation, measured by scales such as Inhibitory Control and Attentional Focusing/Shifting, are processes that facilitate or inhibit the modulation of one's behavioral reactivity. With increasing age, these self-regulatory processes have a stronger impact on the reactivity temperament dimensions as a result of underlying developing cognitive functions, including memory, language (Kopp, 1982) and attentional processes (Rothbart, Derryberry, & Posner, 1994). Factor analysis of the CBQ scales (e.g., Rothbart et al., 2001) revealed 3 superfactors, namely Positive Reactivity (or Extraversion/Surgency; i.e., the tendency to actively and energetically approach new experiences in an emotionally positive way), Negative Reactivity (or Negative Affectivity; i.e., the tendency to be fearful, sad, irritable and easily frustrated) and Effortful Control (i.e., the ability to suppress a dominant response in order to activate a subdominant response). In a previously published study (Eggers et al., 2010) with 3-to-8 year-old CWS, we found that the CWS scored significantly lower on the CBQ's superfactor of Effortful Control and higher on the superfactor of Negative Affectivity. On Inhibitory Control (capacity to plan and suppress inappropriate approach responses under certain instructions or in novel or uncertain situations) and Attentional Shifting (ability to transfer attentional focus from one activity to another), which are two scales that cluster under Effortful Control, CWS scored significantly lower compared to CWNS. Finally, CWS scored significantly higher on three reactivity-related scales: Anger/Frustration (amount of negative affect related to interruption of ongoing tasks or goal blocking), Approach (amount of excitement and positive anticipation for expected pleasurable activities) and Motor Activation (amount of excess repetitive small-motor movement). The observed differences between CWS and CWNS in Effortful Control, as reported by parents on the CBQ, led to the current study. In particular, the study was designed to test experimentally one component of Effortful Control, namely attention.

Sheridan (2007) described attention as the process of focusing sensory, motor, and/or mental resources to environmental aspects in order to acquire knowledge. Several authors have discussed the role of attention in the regulation of reactivity (Beauregard, Lévesque, & Bourgouin, 2001; Ellis, Rothbart, & Posner, 2004; Ochsner & Gross, 2005; Rueda, Posner, & Rothbart, 2004) and some even view self-regulation as the outcome of executive attention development (Rothbart, Ellis, Rueda, & Posner, 2003). Already at a young age, children can increase their positive reactivity in response to enjoyable stimuli by focusing their visual attention towards those stimuli. In contrast, negative reactivity as a result of stress-evoking stimuli can be attenuated by shifting attention away from that specific stimulus towards other stimuli (e.g., when a child spontaneously engages in a new activity on the playground when he/she realizes the current activity is too difficult for him/her). The ability to orient or to shift visual attention is therefore an important factor in controlling distress (Derryberry & Reed, 1994; Ruff & Rothbart, 1996). Harman, Rothbart, and Posner (1997) found evidence that even in 3- to 6-months old infants negative reactivity could be reduced, albeit only momentarily, by providing a new visual stimulus and drawing their attention to it. With maturation, children develop the means to achieve this regulation on their own without external intervention (Posner & Rothbart, 2000a; Rueda et al., 2004), although the degree to which children are able to disengage and move on is individually determined (Colombo, 2004; Fox, 1998).

Posner and Peterson (1990) proposed that the attentional system could be divided into three separate subsystems or networks: (1) the alerting or vigilance network, which is responsible for achieving and maintaining an alert state, in other words readiness to react to environmental stimuli; (2) the orienting or selection network, responsible for the selection of information from sensory input; and (3) the executive control or conflict network, responsible for resolving conflict among responses. These three networks were reliably observed across different studies, including clinical, experimental, and neuroimaging studies (Callejas, Lupianez, & Tudela, 2004; Fan, McCandliss, Fossella, Flombaum, Posner, 2005; Raz & Buhle, 2006) and were associated with specific underlying anatomical areas (Botvinick, Nystrom, Fissell, Carter, & Cohen, 1999; Corbetta & Shulman, 2002; Laberge, 1995; Rothbart & Posner, 2001; Rueda et al., 2004). Posner and his colleagues (Fan, McCandliss, Sommer, Raz, & Posner, 2002) developed the Attention Network Test (ANT), a useful, reliable, and broadly used instrument designed to evaluate alerting, orienting, and executive control in adults as well as in children (e.g., Adolfsdottir, Sorensen, & Lundervold, 2008; Costa, Hernández, & Sebastián-Gallés, 2008; Dennis, Chen, & McCandliss, 2007; Greene, et al., 2008; Wang, Fan, Dong, Wang, Lee, & Posner, 2005). The ANT takes approximately 30-minutes to administer, and was found to have reasonable test-retest reliability (Fan et al., 2002) and good internal validity (Sobin, Kiley-Brabeck, Daniels, Blundell, Anyane-Yeboa, & Karayiorgou, 2004). The ANT was also shown to be valid as a phenotypic assay for genetic studies on attention (Fosella, et al. 2002).

The current study was set up to investigate differences in attentional network performance between CWS and CWNS, as measured by the Attention Network Test. From our previous findings on the CBQ (Eggers et al., 2010), where the CWS scored lower on the Attentional Shifting scale and the Inhibitory Control scale, we hypothesized that the orienting network as well as the executive control network will be less efficient in CWS. Since no significant differences were found on the CBQ-scales related to the alerting network (Attentional Focusing and Perceptual Sensitivity[1]), we hypothesized that no significant differences in the efficiency of the alerting network will emerge.

4.2 Method

4.2.1 Participants

Participants were 82 monolingual Dutch speaking children (62 boys and 20 girls) who were between 4;00 and 9;00 years old. Forty-one children (CWS) were diagnosed with developmental stuttering, and 41 children (CWNS) were nonstuttering, typically developing children, matched by age (± 3 months) and gender to the CWS. The mean

[1] These scales are defined as the tendency to maintain attentional focus upon task-related channels and the amount of detection of slight, low intensity stimuli from the external environment respectively.

age was 6;09 years (*SD* = 1;05 years) for the CWS and 6;09 years (*SD* = 1;05 years) for the CWNS. Participants had no known or parent-reported neurological, psychological, developmental or hearing problems. All participants had normal or corrected to normal vision, and successfully passed an articulation, language, cognitive and hearing screening (see below). Participants were all paid volunteers recruited by their speech-language pathologist (for the CWS) or through the schools (for the CWNS).

All participants passed a screening for articulation using the Antwerp Screening Instrument for Articulation (ASIA-5, Stes & Elen, 1992), a picture-based standardized test used to evaluate children's ability to produce age-specific phonemes in different word positions. Hearing screening was done using Accuscreen (Wood, 2003), a handheld hearing-screening device, employing transient-evoked otoacoustic emissions. In order to screen language ability, participants were administered two subtests (Vocabulary Production and Sentence Production) of the Language Test for Children (van Bon & Hoekstra, 1982). In the Vocabulary Production subtest, participants are presented with pictures and have to complete a phrase with the target-word. The Sentence Production subtest provides participants with phrases that contain syntactical errors, which they have to correct. The cutoff for participants was 1SD below the mean (percentile 16) on both subtests. Mean percentiles on the Vocabulary subtest of the Language Test for Children were 63 for the CWS (range 23-99) and 72 for the CWNS (range 23-96). For the Sentence Production subtest, the CWS had a mean percentile of 54 (range 31-96) compared to 63 for the CWNS (range 23-94). The group differences in scores for the two subtests were not statistically significant (Vocabulary: $t = 80, p = .76;$ Sentence Production scores: $t = 80, p = .23$).

To exclude group differences in general cognitive skills, the Vocabulary and Block Design subtests of the Wechsler Intelligence Scale for Children-Third Edition Dutch (WISC-III; Wechsler, 2005) were administered. The Vocabulary subtest requires participants to explain the meaning of single words, while in the Block Design subtest participants have to rebuild as quickly as possible a visually presented geometrical pattern, using red and white cubes. These two subtests were chosen because they have been shown to correlate highly with the WISC-III overall score (Groth-Marnat, 1997). The mean scores for the Vocabulary subtests were 19.34 for the CWS (range 7 - 41) and 19.95 (range 5 - 41) for the CWNS. On the Block Design subtest, the CWS scored on average 24.73 (range 6 - 59) compared to 27.98 for the CWNS (range 4 - 52). No significant between-group differences were found for either subtest (Vocabulary: $t = 80, p = .74;$ Block Design: $t = 80, p = .29$).

In order to compare parental socio-economic status between the two subject groups (Hackman & Farah, 2009), the highest educational level (1=primary education, 2=high school, 3=college degree, 4=university degree) of each parent was obtained; for each child, the parents' educational levels were added to arrive at a composite score. No significant between-group differences were found between the two groups (CWS: mean 5.63 (range 3-8); CWNS: mean 5.79 (range 4-8); *t (80) = -.71, p = .48.)* Spontaneous speech samples were collected from all participants during two free play interactions on different days with the examiner. Scores on the Stuttering Severity Instrument-3 (SSI-3; Riley, 1994) were calculated based on a minimum sample of 300 words across both free play situations. The participating CWS produced at least three percent within-word disfluencies (sound/syllable repetition, including monosyllabic word repetitions, prolongation or blocks) (Conture, 2001). The average word-based percentage of within-word disfluencies for CWS was 9.89 (*SD* = 5.76). Fourteen CWS were classified on the SSI as mild, 24 as moderate, 2 were rated severe, and 1 was rated very severe. CWNS had an average percentage of within-word disfluencies of .88 (*SD* = .72).

4.2.2 Materials

During the experiment, each participant was presented with two tasks, The Baseline speed subtask from the Amsterdam Neuropsychological Tasks (De Sonneville, 2009) and the Attention Network Test (Fan et al., 2002). Each task is described below.

4.2.2.1 Baseline Speed Task

The baseline speed subtask of the Amsterdam Neuropsychological Tasks (De Sonneville, 2009) is a simple computer-based reaction time (RT) task. This allowed us to evaluate if between-group RT differences existed, which could have confounded ANT results. Each trial began with a white fixation cross on a black background in the center of a computer monitor. As soon as the fixation cross was replaced by the target stimulus, a white centralized square, participants had to press a response button as quickly as possible. Ten practice trials were followed by two experimental blocks of 32 trials each, one for the right index finger and one for the left index finger, as is standard for the subtask. The square target stimulus stayed until a response was received. Valid responses fell between 150 ms and 4000 ms after stimulus onset. The interstimulus interval between trials varied randomly between 500 ms and 2500 ms. Baseline speed measured simple RT averaged 478 ms (*SD* = 151) for CWS and 489 ms (*SD* = 161) for CWNS. Between group comparisons using the Student’s t-test showed no significant between-group differences on simple RT: *t (80) = .32, p = .75.*

4.2.2.2 Attention Network Test

The Attention Network Test (Fan et al., 2002) is a computer task that measures the efficiency of 3 distinct attentional networks, i.e. alerting, orienting, and executive attention. Rueda et al. (2004) developed a child-specific version of this task, a combination of a cued RT task and a flanker task, for children 4 years of age and above. Since the participant group in Rueda et al.’s study consisted of children between 6 and 10 years of age, we tested the suitability of the task for younger children by conducting a pilot study with a 4- and a 5-year old child, respectively, prior to the experiment proper reported below. Moreover, the minimal overall accuracy of participants in the current study was 60% and over 80% of the youngest participants, i.e. 4- and 5-year-olds, had an overall accuracy rate above 70%, which is consistent with the values reported in literature (Rueda, Rothbart, McCandliss, Saccomanno, & Posner, 2005). In the child version of the task, the stimuli, which in the original ANT consisted of 5 black lines with or without arrowheads pointing in a specific direction, were replaced with 5 yellow fish on a blue-green background. Children are asked to feed the middle fish by pressing a button corresponding to the direction in which the fish is looking. In contrast to the adult version, children get visual and auditory feedback, which increases the attractiveness of the task. The rationale for providing feedback in the child version was that children were found to work best when there is clear feedback on their performance and when the task is embedded in a story (see ‘procedure’ below; Berger, Jones, Rothbart, & Posner, 2000). Feedback for correct responses consists of the fish moving, blowing air bubbles, and hearing a child saying “Woohoo!” In case of an incorrect answer, the fish is not animated and the child hears a simple single tone.

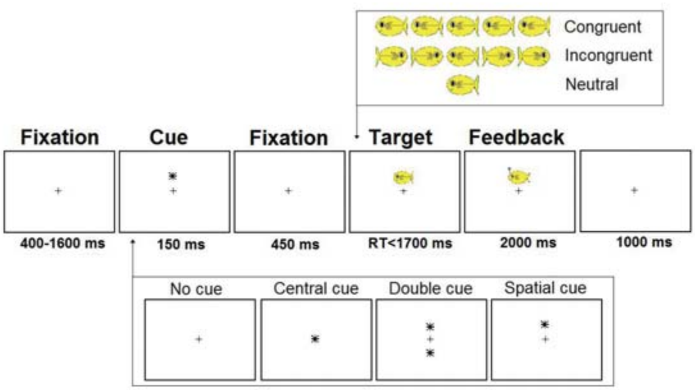


Figure 4.1: Schematic overview of the Children’s Attention Network Test.

An outline of the ANT procedure is depicted in Figure 4.1. Each trial consists of a fixation task and a target task. A trial always begins with a fixation period during which the child has to focus on a central cross. The fixation period has a randomly distributed duration between 400 and 1600 ms. This is followed by presentation of a cue for 150 ms. There are 4 different cue conditions: no cue, a central cue, a double cue, and a spatial cue. In the no-cue condition, the central fixation cross remained on the screen without an asterisk stimulus until the target was presented (see below). In the central cue condition, an asterisk appeared over the central fixation cross. In the double cue condition, 2 asterisks appeared simultaneously, one above and another below the central fixation cross. Finally, in the spatial cue condition an asterisk appeared either above or below the fixation cross. In each of the cue conditions, there was a short fixation period of 450 ms when the cue disappeared before the target (fish) was presented. While all cues provided information on the forthcoming appearance of the target, only spatial cues provided information on target location. The target was a drawing of a yellow fish, or a group of 5 yellow fish, positioned horizontally either above or below the fixation cross location. There were 3 different target types: neutral, congruent, and incongruent. The neutral target consisted of only one fish, looking left or right. The other two target types consisted of a fish with four flanker fish (two on each side), which were either all looking in the same direction of the central fish (congruent) or in the opposite direction (incongruent). Each target remained on the screen, for a maximum of 1700 ms, or until the child responded. After a response was recorded, automated computer feedback (see earlier description) was provided for 2000 ms.

The ANT comprises of a practice session of 24 trials, followed by 3 experimental blocks of 48 trials each. The 12 conditions (4 cues x 3 target types) were represented equally and randomly in each experimental block. Between each of the three blocks a pause of 5 minutes was provided during which the child received some pieces of a puzzle of a cartoon figure. At the end of the experimental session the child had obtained all the pieces and could complete the puzzle. Recording performance during each of the 3 experimental blocks offers the possibility of measuring how persistent participants are in sustaining their attention over a period of time. For each trial, RT and accuracy were recorded automatically and transferred to a linked Excel file.

As described by Fan et al. (2002) and Rueda et al. (2004), we calculated three subtractions to assess the efficiency of the different attentional networks; these were based on measuring how RTs were influenced by cue type and flanker conditions. The efficiency of the alerting network was calculated by subtracting the median RT in the double cue condition from the median RT in the no cue condition ($\text{Eff.}_{\text{alert.}} = \text{median RT}_{\text{double cue}} - \text{median RT}_{\text{no cue}}$). Orienting efficiency was calculated by subtracting the median RT obtained in the spatial cue condition from the median RT in the central cue condition ($\text{Eff.}_{\text{orient.}} = \text{median RT}_{\text{central cue}} - \text{median RT}_{\text{spatial cue}}$). And finally, to evaluate the executive control network, the median RT for congruent flanker conditions were subtracted from the median RT for incongruent flanker conditions ($\text{Eff.}_{\text{exec.}} = \text{median RT}_{\text{incongruent flanker}} - \text{median RT}_{\text{congruent flanker}}$). The higher the score on alerting and orienting the higher the efficiency in both networks as a result of the presence of a cue and the presence of a directional cue respectively. In contrast, the higher the score on executive control, the less efficient the executive attention network is since there is more interference from incongruent flankers.

4.2.3 Procedure

All tests were conducted in a quiet setting at the participant's home during two visits by the first author, a qualified speech-language pathologist with expertise in stuttering.

These two test sessions (A & B) took approximately one hour each, and were structured in such a way that the most attention-dependent tasks were administered at the beginning of each session. During test session A the following tests were administered in the order mentioned: a) the Attention Network Test (Fan et al., 2002), b) the Vocabulary and Block Design subtest of WISC-III (Wechsler, 2005), c) the speech sample, and d) Accuscreen (Wood, 2003). Test session B consisted of the following tests: a) the Baseline Speed subtask of the Amsterdam Neuropsychological Tasks (De Sonneville, 2009), b) the Vocabulary Production and Sentence Production of the Language Test for Children (van Bon & Hoekstra, 1982), c) the speech sample, and d) the ASIA-5 (Stes & Elen, 1992). To avoid a possible confound of testing order, half of the participants in each group were presented with test session A during the initial visit while the other half started with test session B.

For the Attention Network Test, children were told that they would need to feed hungry fish that appeared on a computer screen. They could do this by pressing the left or right mouse button that corresponded with the direction the fish was swimming in. To make sure they understood the task they were shown several printouts of fish looking left or right and were asked to press the correct mouse button. After completing these initial practice trials, they were told that sometimes there would just be one fish while at other times there would be several fish swimming alongside. These fish could be swimming in the same or the opposite directions as the fish in the middle. Children again were told that their job was to feed only the middle fish. Again they were able to practice using printouts of a fish flanked by four fish, two on either side, sometimes all looking in the same direction, and other times looking in opposite directions. When it was clear that the children comprehended the task, the formal practice session was started on the computer. Children were instructed to maintain their focus on the fixation cross at the center of the screen and to press the correct mouse button to feed the fish as soon as it appeared. Stimuli were presented on a laptop computer with a 15-inch screen. The optimal distance between child and computer screen is calculated automatically by the ANT program, and was 18 inches for all children. A large black pliable cardboard was positioned around the laptop to avoid distracting visual stimuli in the surrounding environment. The child wore headphones to listen to the auditory feedback and to reduce possible distracting environmental sounds.

4.3 Results

4.3.1 Between-group differences in RT and error percentages

As part of the standard and automatic calculations done by the ANT program, trials with a RT greater than 2 standard deviations from the child's mean for each cue and flanker combination were excluded. Similar to the procedure followed by Rueda et al. (2004), group RT data were based on the mean of each child's median RT. The use of

median RTs, often results in a more accurate representation of the ‘average value’ when data are not symmetrically distributed.
To evaluate differences in RTs for each of the cue and flanker type combinations, a Multivariate Analysis of Covariance (MANCOVA) was used with group as independent variable, RT data in each of the cue and flanker conditions as dependent variables, and chronological age as a covariate. The omnibus MANCOVA test, using Pillai’s trace, showed no significant main effect for group, $V = .19$, $F(12, 67) = 1.33$, $p = .22$, and no significant interaction effect between age and group, $V = .17$, $F(12, 67) = 1.13$, $p = .35$; a significant main effect was found for age $V = .64$, $F(12, 67) = 9.89$, $p < .001$. B-values of the covariate were negative (ranged between -5.95 and -9.02) which shows a significant decrease in RTs with increasing age for all cue and flanker combinations ($p < .001$ for all combinations).

Differences in error percentages for each of the cue and flanker type combinations were also evaluated using a Multivariate Analysis of Covariance (MANCOVA) with group as independent variable, error percentages as dependent variables, and chronological age as a covariate. The omnibus MANCOVA test, using Pillai’s trace, showed no significant main effect for group, $V = .23$, $F(12, 67) = 1.67$, $p = .09$, and no significant interaction effect between age and group, $V = .19$, $F(12, 67) = 1.31$, $p = .24$; a significant main effect was found for age $V = .52$, $F(12, 67) = 5.86$, $p < .001$. Analyses of the B-value of the covariate (ranged between -.002 and -.005) showed a significant decrease in error percentages with increasing age for all cue and flanker combinations ($p < .05$ for all combinations).

Separate univariate between-group Analyses of Covariance (ANCOVA) were performed for overall RT and overall accuracy (collapsed across cue and flanker conditions). No significant between-group effects were revealed for overall RT, $F(1, 79) = .96$, $p = .33$, or overall accuracy $F(1, 79) = 2.66$, $p = .107$ (Table 4.1). A significant age effect was found for overall RT, $F(1, 79) = 84.55$, $p < .001$, and for overall accuracy, $F(1, 79) = 52.00$, $p < .001$. Again, analyses of the B-value of the covariate showed significantly faster overall RTs ($B = -6.69$, $p < .001$) and overall accuracy ($B = .37$, $p < .001$) with increasing age.

Table 4.1: Attention Network Test overall RT in ms and overall accuracy for CWS and CWNS.

	Overall RT		**Overall accuracy**	
Group	***M***	***SD***	***M***	***SD***
CWS	921	160	91.8	7.9
CWNS	945	154	89.2	11.7

In order to evaluate interactions between cue type and flanker conditions, a series of 4 (cue: no, double, central, spatial) x 3 (flanker: congruent, incongruent, neutral) ANCOVAs were calculated, with chronological age as covariate. This was done in both participant groups separately, for RTs as well as error rates. The nature of interaction between cue type and flanker conditions is shown in Figures 4.2 and 4.3. For the CWS significant RT main effects of cue type, $F(3, 480) = 4.31, p < .005$, and flanker condition, $F(2, 480) = 25.7, p < .001$ (Figure 4.2a) were observed; Tukey post hoc comparisons indicated that the average RT in the no cue condition ($M = 969$, $SD = 178$) was significantly higher than in the double ($M = 907$, $SD = 182$, $p < .05$), and the spatial cue condition ($M = 891$, $SD = 199$, $p < .005$); RTs in incongruent flanker conditions ($M = 1005$, $SD = 177$) were significantly higher than those in the congruent ($M = 887$, $SD = 182$, $p < .001$), and the neutral flanker conditions ($M = 874$, $SD = 185$, $p < .001$).

Error rate only showed a significant main effect for flanker condition, $F(2, 480) = 12.0, p < .001$ and not for cue type $F(2, 480) = .375, p = .77$ (Figure 4.2b). Tukey post hoc comparisons showed error rates in incongruent flanker conditions ($M = .11$, $SD = .13$) were significantly higher than both congruent ($M = .05$, $SD = .09$, $p < .001$) and neutral flanker conditions ($M = .06$, $SD = .08$, $p < .001$). No significant interaction effect for cue type and flanker condition were found for RT, $F(6, 480) = .32, p = .92$, or error rate, $F(6, 480) = .43, p = .86$.

The analyses of variance for the CWNS showed a similar pattern: significant RT main effects were found for cue type, $F(3, 480) = 6.82, p < .001$, and flanker condition, $F(2, 480) = 19.23, p < .001$ (Figure 4.3a). Tukey post hoc comparisons also indicated that the average RTs in the no cue condition ($M = 996$, $SD = 181$) were significantly higher than in the double cue ($M = 933$, $SD = 188$, $p < .05$), as well as the spatial cue condition ($M = 894$, $SD = 169$, $p < .001$); and that RTs in incongruent flanker conditions ($M = 1014$, $SD = 180$) were significantly higher than both congruent ($M = 911$, $SD = 167$, $p < .001$), and neutral flanker conditions ($M = 906$, $SD = 188$, $p < .001$). For error rate a main affect was only found for the flanker condition $F(2, 479) = 7.93, p < .001$ and not for cue type $F(2, 479) = .244, p = .865$ (Figure 4.3b). Tukey post hoc comparisons showed error rates in incongruent flanker conditions ($M = .14$, $SD = .18$) were significantly higher than congruent ($M = .08$, $SD = .12$, $p < .001$), as well as neutral flanker conditions ($M = .09$, $SD = .12$, $p < .005$). No significant interaction effect for cue type and flanker condition emerged for RT, $F(6, 480) = .81, p = .56$, or error rate, $F(6, 479) = .36, p = .90$.

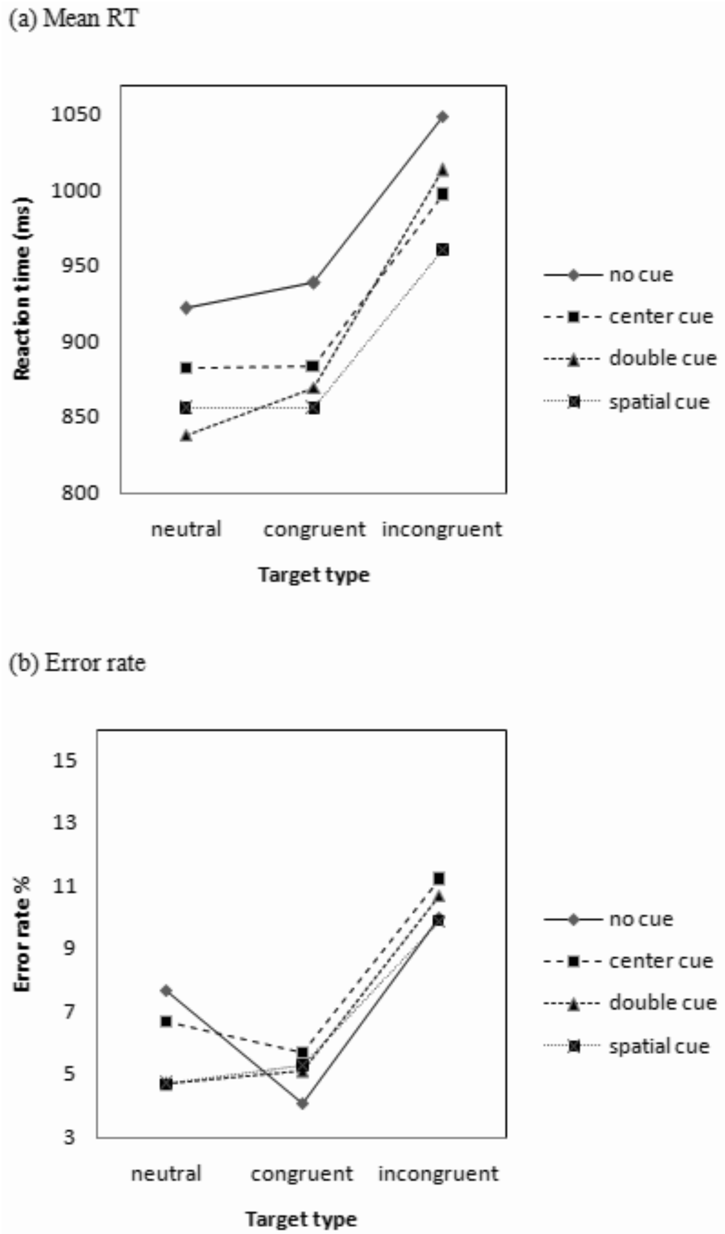


Figure 4.2: Mean RT (a) and error rate (b) for the correct trials as function of cue and flanker conditions for the CWS.

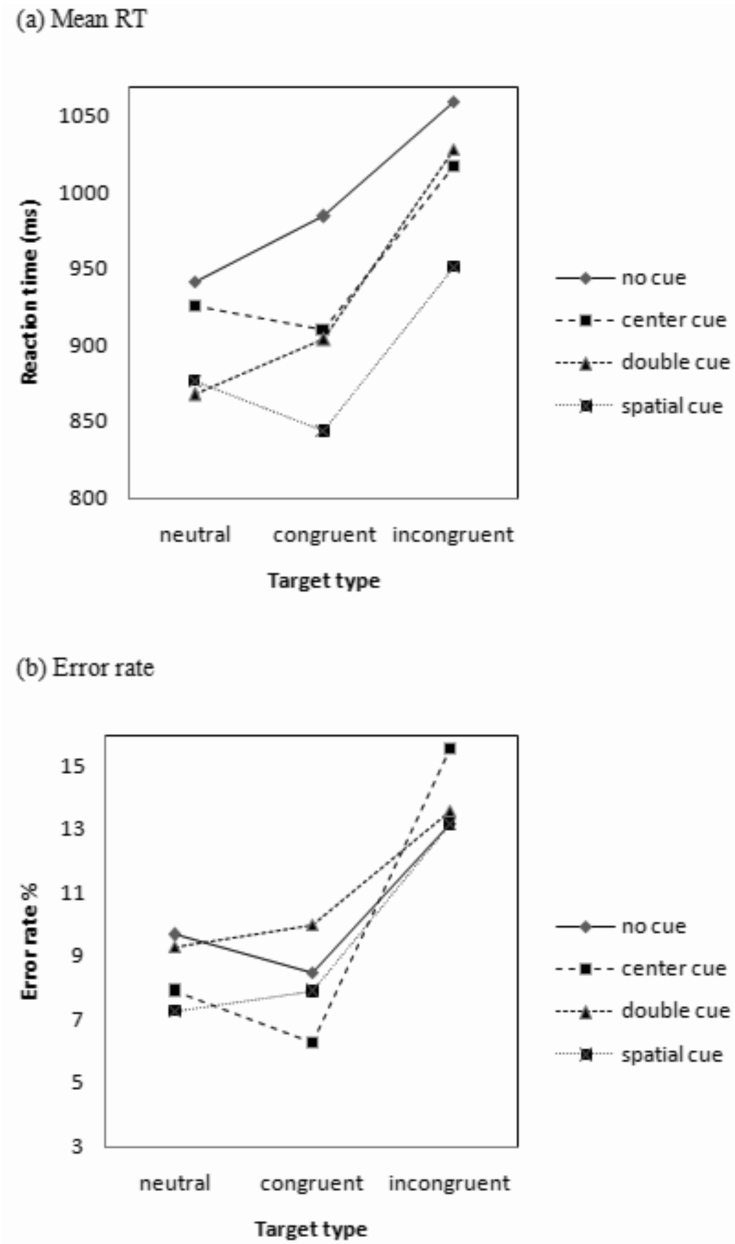


Figure 4.3: Mean RT (a) and error rate (b) for the correct trials as function of cue and flanker conditions for the CWNS

4.3.2 Changes in mean RT and error rates over the three experimental blocks

Mean RTs and error percentages over the three experimental blocks (collapsed across cue and flanker conditions), were compared for CWS and CWNS (see Figure 4.4). The effect of group classification on changes in mean RTs was analyzed with a mixed ANCOVA with participant group as between-subjects variable and mean RT for each of the three experimental blocks as within-subjects variable or factor; chronological age was set as a covariate. The main effect for block was significant $F(2, 78) = 17.68$, $p < .001$, as was the block X age interaction, $F(2, 78) = 8.24$, $p < .005$. The group X block interaction was not significant, $F(2, 78) = .81$, $p = .45$. Tests of between-subjects effects showed no significant group differences, $F(1, 79) = .32$, $p = .58$, while tests of within-subjects effects showed a significant effect for block, $F(2, 158) = 21.23$, $p < .001$, and block X age interaction, $F(2, 158) = 9.90$, $p < .001$. Pairwise comparisons showed a significant decrease in mean RT over the three experimental blocks, with the mean RT for experimental block 1 ($M = 1068$, $SD = 227$) being significantly higher than the mean RT for experimental block 2 ($M = 1011$, $SD = 203$), $p < .001$. Block 2, in turn, was significantly higher than the mean RT for experimental block 3 ($M = 955$, $SD = 192$), $p < .001$. A similar mixed ANCOVA with changes in error rates as within-subjects variable (or factor) did not yield any main effects for block, $F(2, 78) = 2.02$, $p = .14$, block X age interaction, $F(2, 78) = 2.04$, $p = .14$, or group X block interaction, $F(2, 78) = .16$, $p = .85$. Tests of between-subjects effects showed no significant group differences, $F(1, 79) = 2.66$, $p = .11$, and also tests of within-subjects effects showed no significant effects for block, $F(2, 158) = 2.04$, $p = .13$, and block X age interaction, $F(2, 158) = 2.07$, $p = .13$.

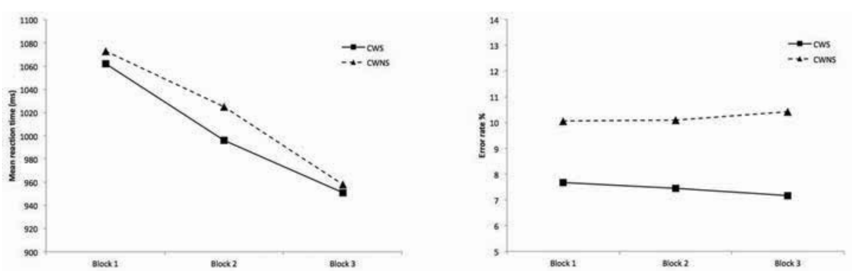


Figure 4.4: Performance change in RT and error rate over the three experimental blocks for the CWS and CWNS.

4.3.3 Between-group differences in attentional networks

Between group differences in attentional network scores were analyzed using a MANCOVA. Participant group was set as the independent variable, with alerting, orienting, and conflict scores as dependent variables, and chronological age as covariate. The omnibus MANCOVA test, using Pillai's trace, showed a significant main effect for group, $V = .10$, $F(3, 76) = 2.91$, $p < .05$, and no significant interaction effect between age and group, $V = .09$, $F(1, 76) = 2.61$, $p = .058$. Tests of between subject effects (see Figure 4.5) showed that CWS scored significantly lower on the orienting network compared to the CWNS, $F(1, 78) = 5.04$, $p < .05$. No significant differences were found for the alerting ($F(1, 76) = 0.49$, $p = .825$) or conflict scores ($F(1, 76) = 3.47$, $p = .066$).

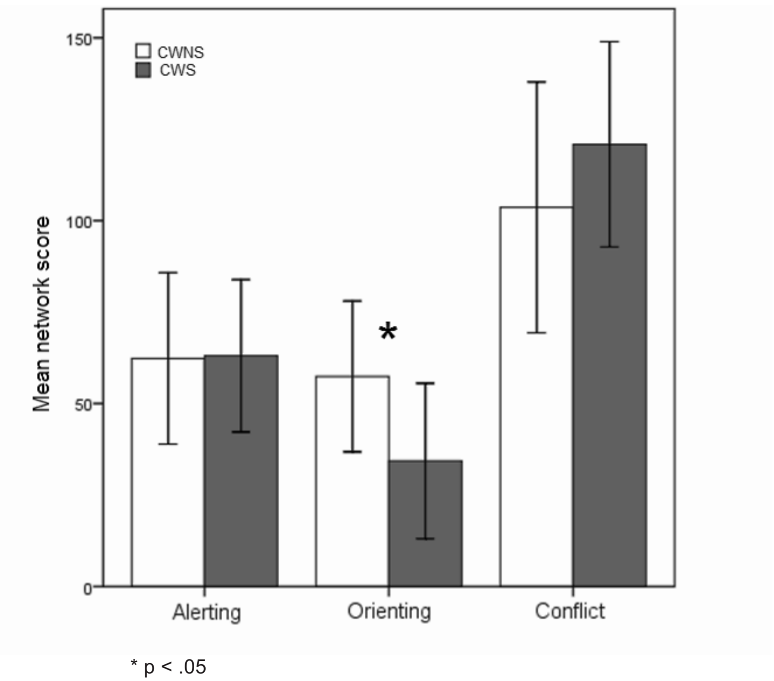


Figure 4.5: Attention network scores for the CWS and CWNS.

4.4 Discussion

In previous studies (Eggers et al., 2009, 2010) we reported on differences in self-regulatory behaviors, i.e. inhibitory control and attentional shifting, between CWS and CWNS as reported by parents on the CBQ. In the current study the Attention Network Test was used to evaluate experimentally whether differences between CWS and CWNS participant groups were present in the attentional networks that form the basis for these self-regulatory behaviors.

4.4.1 Between-group differences in RT and error percentages

No significant between-group differences were found for RT or error percentages for each of the cue and flanker type combinations or for overall RT and overall accuracy. The current findings for RT and error percentages for the CWNS are similar to the averages for 6- and 7-year old typically developing children reported in the study by Rueda et al. (2004), thereby confirming the validity of the current testing procedures as well as the representativeness of our CWNS group. Similar to Rueda et al.'s findings, RTs and error rates, for all cue and flanker combinations, gradually improved in the nonstuttering subjects with increasing age, pointing towards maturation of cognitive and motor response systems. A similar result was found for the stuttering children in the current study.

Although our findings with children were contrary to those reported by Fan et al. (2002) who found a significant interaction between cue type and flanker condition in adult nonstuttering participants, they were consistent with the findings by Rueda et al. (2004). These investigators observed significant main effects, but no interaction, for cue type and flanker condition for the RT measure, while the error measure only yielded a main effect for flanker conditions. The reasons for the contradictory findings between adults and children are not yet understood, but could be related to developmental processes, as well as to methodological factors such as size and color of targets and flankers, and using more concrete stimuli (fish) compared to more abstract arrows (Moore & Egeth, 1998; Rueda, et al., 2004).

In the current study, the presence of incongruent flankers in both participant groups led to an increase in RTs and error percentages. This effect, also known as the noise-compatibility effect, was first described by Eriksen and Eriksen (1974, 1979), who showed that the discriminative response to a target was influenced by flankers (noise). Flankers associated with a similar response as the target (congruent flanker condition) resulted in reduced RTs and error percentages compared to flankers associated with a response different from the one to the target (incongruent flanker condition). During incongruent trials both correct and incorrect competing responses are activated, requiring attention-based inhibition of the incorrect response (Eriksen & Schultz, 1979). Because this response inhibition occurs with a certain delay, correct responses on incongruent trials are slower. In other words, responses given before the response inhibition occurs are primarily determined by the flankers since they outnumber the target; this results in more incorrect responses during incongruent flanker conditions and more correct responses during congruent flanker conditions (Gratton, Coles, & Donchin, 1992). Cueing prior to the presentation of the target also reduced RTs for both CWS and CWNS, and this effect was most noticeable when cues included spatial information. Posner and colleagues (Posner, 1980; Posner, Snyder, & Davidson, 1980) were the first to describe this spatial cue-induced reduction in response latencies. They attributed the improved efficiency to a focusing of the attentional resources to the appropriate visual field area (Posner & Peterson, 1990). Later studies (Coull & Nobre, 1998; Nobre, 2001) showed that this cueing effect is not only limited to spatial information but is also present during temporal allocation of attention (see more below).

4.4.2 Changes in mean RT and error rates over the three experimental blocks

While the RT in both groups decreased significantly over the three experimental blocks, no differences were found between CWS and CWNS regarding changes in performance over the three blocks. The similarity between the two participant groups in their improvement of RT towards the end of the task as well as reduction in the amount of errors, suggests that CWS are as persistent in sustaining their attention over a period of time during an attention-demanding task, as are the CWNS. This observation is in line with earlier findings (Anderson & Wagovich, 2010; Eggers et al., 2010) that the scores of CWS on the CBQ scale of Attentional Focusing are comparable to those of CWNS, reflecting their tendency to maintain task-related attentional focus. Anderson et al. (2005) reported similar results in a study using the Behavioral Style Questionnaire (McDevitt & Carey, 1978). In their study, the scale measuring attention span/persistence, which is defined as "*the ability to continue the activity in the face of distractions*" (p.1225), did not yield any significant differences (p = .07) between participant groups.

4.4.3 Between-group differences in attentional networks

Both the magnitude of and the relationships between the network scores of our CWNS group are in accordance with the average scores for similar age groups in the study of Rueda et al. (2004). Furthermore, the large standard deviations for the network scores, pointing towards a high individual variability, is analogous to observations in other studies (Adolfsdottir et al., 2008; Rueda et al., 2004), especially those done with children.

Difference scores in RT between stimuli with congruent and incongruent flanker conditions, i.e. conflict effect, or with no cue and double cue types, i.e. alerting effect, did not significantly differentiate between our two participant groups. This leads us to propose that earlier CBQ findings of differences in inhibitory control (Eggers et al., 2010; Embrechts et al., 2000) are not associated with a lower efficiency of the underlying executive attentional network. Although no statistical between group differences were found for conflict, there was a clear trend (p = .066) for the CWS to show a tendency in line with our original hypothesis, namely a higher score and thus a lower efficiency on the executive control network. However, because post hoc test results showed that the power to detect differences on conflict was very low (power = .056) in our study, and indeed considerably lower than for both alerting (.54) and orienting (.61), caution against misinterpretations is warranted. Based on our findings it may be worthwhile to explore inhibitory control using tasks aimed at testing in specific that cognitive process (e.g., Logan, 1994), such as go-nogo tasks (tasks where participants are instructed to respond to certain stimuli but not to respond to others) or stop-signal tasks (tasks where participants are instructed to respond to stimuli but to refrain from responding if a stop-signal signal appears after stimulus onset). Also in studies of children with ADHD, differences in inhibitory control emerged by using a stop-signal paradigm (Pliszka, Liotti, Woldorff, 2000), whereas no differences were found for the broader underlying executive attentional network (Adolfsdottir et al., 2008; Booth, 2003).

RT differences for the orienting effect, i.e. between spatial and central cue types, did reveal a significant between-group difference. CWS, as a group, had a lower efficiency of the orienting network compared to CWNS. These findings are in agreement with our initial hypothesis that the orienting network in CWS would be less efficient and corroborates our earlier finding of a significant lower score for CWS on the Attentional Shifting scale of the CBQ (Eggers et al., 2010). Furthermore, this finding confirms previous results which suggest that attentional processes may be involved in the development and/or maintenance of developmental stuttering (Bajaj, 2007; Bernstein Ratner & Wijnen, 2006; Bosshardt, 1999, 2002, 2006; Bosshardt et al., 2002; Felsenfeld et al., 2010; Foundas et al., 2004; Karrass, et al., 2006; Oomen & Postma, 2001; Schwenk et al., 2007; Smits-Bandstra & De Nil, 2007; Van Lieshout et al., 1996; Vasic & Wijnen, 2005; Webster, 1990).

The orienting network encompasses the ability to select information from numerous sensory inputs. This top-down process is responsible for optimizing our perception and action through selectively biasing information processing based on changing motivation or expectation (Lepsien & Nobre, 2006). Specifically for the visual modality, this orienting process relates to spatial expectations of where task-relevant events are bound to occur. In a classical Posner cue paradigm, the attention can be drawn to a specific location by a centrally placed arrow pointing towards the location where the target will appear (endogenous cueing) or by presenting a cue in the peripheral location (exogenous cueing), as is the case in the ANT. The subsequent visual orienting can be achieved by either directing the eyes to the location of interest (overt orienting) or by giving priority to a specific area in the visual field without overtly moving the eyes (covert orienting) (Posner, 1980). The ANT is primarily based on covert orienting (because children are asked to focus on the fixation cross), and thus explores primarily internal mechanisms of attention.

Both overt and covert visual spatial orienting (Corbetta, 1998), as well as exogenous and endogenous cues (Corbetta, Miezin, Shulman, & Petersen, 1993; Nobre, et al., 1997) have been found to activate similar anatomical regions. Studies in stroke patients revealed that the different components of orienting, i.e. disengagement from the current attended location, attentional movement, and engagement at the target location, have a distinct anatomy and that lesions in the neural structures underlying each component can result in different behavioral orienting-related deficits, such as perseverative errors resulting from a disengagement impairment (Fernandez-Duque & Posner, 2001).

Nobre et al. (2004) demonstrated that it is possible to orient spatial attention internally to locations held within the working memory. The activation of this orienting neural network is not just limited to spatial related attention tasks but is also active during tasks where participants had to shift their attention between other stimulus dimensions such as shape and color (Le, Pardo, & Hu, 1998) or during tasks where participants were receiving cues not on ‘where’ to expect a stimulus to appear but on ‘when’ a stimulus would appear (temporal orienting), directing their attention to specific points in time (Coull & Nobre, 1998). A considerable overlap was found for networks for spatial and temporal orienting (Coull & Nobre, 1998; Nobre, 2001).

Although we did not examine temporal orienting in CWS in the current study, based on findings from adult studies there is ample evidence for a high degree of overlap between spatial and temporal orienting networks (Coull & Nobre, 1998; Nobre, 2001) as well as for a cross-modal interaction between spatial orienting and temporal order judgments (Santangelo & Spence, 2009), allowing us to speculate about possible interrelationships. Coull and Nobre state that while complex movements require exquisite temporal coordination, temporal orienting may affect motor preparation, motor coordination or motor sequencing/timing. Therefore, it can be speculated that a decreased efficiency of the orienting network might be linked to reports of disturbed timing of activation in speech-relevant brain areas in adults who stutter (Sommer, Koch, Paulus, Weiller, & Büchel, 2002) or temporal programming deficits (e.g., Kent, 1984). In addition, the interplay between attentional orienting (both spatial and temporal) and working memory may lead to speculation on the possible role of attentional orienting in a number of other etiological models of stuttering that imply involvement of working memory, such as the psycholinguistic (e.g., Bernstein Ratner & Wijnen, 2006; Vasic & Wijnen, 2005), speech motor (see e.g., Smits-Bandstra & De Nil, 2007), and resource allocation/competition based models (see e.g., Bosshardt, 2006).

The results from our study are in line with robust findings from dual task experiments (e.g., Arends, Povel, & Kolk, 1988; Bajaj, 2007; Bosshardt, 1999, 2002, 2006; Bosshardt et al., 2002; Smits-Bandstra & De Nil, 2007; Vasic & Wijnen, 2005) of less proficient performance in people who stutter. Due to the lower efficiency of the orienting network, PWS might be less able to orient their attentional resources between concurrent tasks, including speech planning and execution. This interpretation appears consistent with the observation that stuttering is often exacerbated in both linguistically (Bernstein Ratner & Sih, 1987) and environmentally (Ezrati-Vinacour & Levin, 2004) situations that demand increased attentional demands, such as increased syntactic complexity and perceived situational stress.

Finally, it is well known that the severity of stuttering symptoms is prone to anxiety or emotional reactions resulting from situational stress (Alm, 2004a; Ezrati-Vinacour & Levin, 2004; Menzies, Onslow, & Packman, 1999). It has been shown that attentional orienting plays a significant role in regulating the emotional reactivity in young children, thus controlling distress (Rueda et al., 2004; Harman et al., 1997). By orienting their attention away from stress-evoking situations, children are able to decrease their level of arousal. Similar processes may also be present in adults, because those who report a better ability to focus and shift attention also report experiencing less negative affect (Derryberry & Rothbart, 1988). CWS, due to their lower ability to orient attention, may be less able to regulate their emotional reactivity in stressful situations (see also Conture, et al., 2006). This finding suggests that CWS, in anticipation of both positive (e.g., counting down to the day of a birthday party) and negative (e.g., giving an oral presentation in the classroom) stressful situations, have difficulty stopping focusing on these situations and change their mindsets, resulting in increased arousal. Similarly, during stressful situations, CWS may be less capable of disengaging their attention from the emotion-triggering stimuli, resulting in maintenance or even increase in already high arousal.

4.4.4 Additional considerations

It should be noted that current findings represent group tendencies and may not necessarily apply to individual children. In other words, some CWS may have a high efficiency of the orienting network while some CWNS may have a low efficiency of the orienting network. Therefore the lower efficiency of this attentional network may be a factor in developmental stuttering for some CWS but not for others. As a result, whether there is a direct link between our findings of differences in the attentional network and the manifestation of stuttering in individual children remains speculative at best and needs further investigation.

A further limitation of the current study is that our findings are only based on visual stimuli, and more research is needed into the effect of stimulus and cue characteristics. Future studies on the relationship between attention and stuttering should therefore investigate the effect of, for instance, auditory as well as emotional cues, especially given the role of emotional stressors in stuttering (Ezrati-Vinacour & Levin, 2004). Similarly, studies investigating not only spatial but also temporal orienting are needed.

Finally, some parallels can be drawn between our findings relating to congruent and incongruent flanker conditions as well as spatial information cues, and the results from recent studies on priming, in which CWS were found to react differently to priming compared to CWNS (Anderson & Conture, 2004; Hartfield & Conture, 2006; Melnick, Conture, & Ohde, 2003). Both paradigms modulate the focusing of attention to the target albeit using different cues and expected responses: in the priming studies auditory primes (e.g., phonological primes, word primes, sentence-structure primes) were used and the output was verbal (target word), while the current study used visual cues and the output was a motor response. It might be interesting to investigate if individuals who score higher or lower on specific networks also perform better or worse on specific priming tasks.

4.5 Conclusions

Our results, based on a visual spatial orienting task, revealed that CWS, as a group, were less able to select information from sensory input. In other words, the orienting network, which has an important role in allocating attentional resources to both external perceptual stimuli as well as to working memory representations, appears to be less efficient in CWS. No significant differences were found on the alerting or executive control network, although the latter showed a trend towards lower efficiency in CWS. These findings were taken to suggest a possible role for attentional processes in developmental stuttering.

CHAPTER 5

Inhibitory control in childhood stuttering.

The content of this chapter has been accepted for publication as:
Eggers, K., De Nil, L., & Van den Bergh, B. (2012b). Inhibitory control in childhood stuttering. Accepted for publication in *Journal of Fluency Disorders*.

Parts of the content of this chapter have been presented as:
Eggers, K. (2011, January). *Attentional networks and inhibitory processes in developmental stuttering.* Lecture presented at the Escola Superior de Saude do Alcoitão, Portugal.

Eggers, K. (2010, April). *Inhibitory control in CWS.* Poster presented at the Second European Symposium on Fluency Disorders, Antwerp.

Eggers, K., De Nil, L., & Van den Bergh, B. (2009, November). *Inhibitory control in children who stutter.* Poster presented at the annual convention of the American Speech-Language-Hearing Association, New Orleans.

Abstract
Purpose: The purpose of this study was to investigate whether previously reported parental questionnaire-based differences in inhibitory control (IC; Eggers, De Nil, & Van den Bergh, 2010) would be supported by direct measurement of IC using a computer task.
Method: Participants were 30 children who stutter (CWS; mean age = 7;05) and 30 children who not stutter (CWNS; mean age = 7;05). Participants were matched on age and gender (± 3 months). IC was assessed by the Go/NoGo task of the Amsterdam Neuropsychological Tasks (De Sonneville, 2009).
Results: Results indicated that CWS, compared to CWNS, a) exhibited more false alarms and premature responses, b) showed lower reaction times for false alarms, and c) were less able to adapt their response style after experiencing response errors.
Conclusions: Our findings provide further support for the hypothesis that CWS and CWNS differ on IC. CWS, as a group, were lower in IC pointing towards a lowered ability to inhibit prepotent response tendencies. The findings were linked to previous IC-related studies and to emerging theoretical frameworks of stuttering development.

5.1 Introduction

Inhibitory control (IC) is the ability to suppress, interrupt or delay an inappropriate response under instructions or in novel or uncertain situations (Clark, 1996; Rothbart, 1989b) or to ignore irrelevant information (Dagenbach & Carr, 1994; Dempster & Brainerds, 1995; Rothbart & Posner, 1985). IC is essential for the performance of everyday tasks (Simpson & Riggs, 2009) and has been implicated in cognitive development (Harnishfeger & Bjorklund, 1994), executive functioning (Friedman & Miyake, 2004), and the conscious use of attention or attentional control (Desimone & Duncan, 1995; Kochanska, 1997). It is strongly related to the coordination and integration of mental processes in successful task performance (Dowesett & Livesey, 1999) and plays an important role in the self-regulation of emotional states (Kochanska, Murray, Jacques, Koenig, & Vandegeest, 1996; Kopp, 1982).

Several authors have alluded to a possible role for self-regulatory processes, attentional control processes, and more specifically inhibitory control in the development of stuttering. Evidence for possible reduced self-regulation has come from observations that CWS are (a) lower in adaptability (Anderson, Pellowski, Conture, & Kelly, 2003), (b) lower in biological rhythmicity (Anderson, et al., 2003) and (c) less efficient in emotional regulation (Karrass et al., 2006), although the latter finding was not confirmed in a recent study from the same research group (Arnold, Conture, Key, & Walden, 2011). With regard to attentional control, studies have reported CWS to be (a) more or less distractible, depending on the measurement method used (Embrechts, Ebben, Franke, & van de Poel, 2000; Schwenk, Conture, & Walden, 2007; Anderson et al., 2003), (b) less efficient in attention regulation (Felsenfeld, van Beijsterveldt, & Boomsma, 2010; Karrass et al., 2006; Schwenk et al., 2007), and (c) less efficient in attentional orienting (Eggers, De Nil, & Van den Bergh, 2010, 2012a); also studies in adults who stutter pointed to a lowered efficiency in allocating attentional resources under dual task conditions (Bosshardt, 1999, 2002, 2006; Bosshardt, Ballmer, & De Nil, 2002; Smits-Bandstra & De Nil, 2007; Vasic & Wijnen, 2005). Finally, some studies reported that CWS were lower in inhibitory control (Eggers et al., 2010; Embrechts et al., 2000), while others found no difference (Anderson & Wagovich, 2010).

Further study of IC in stuttering may be particularly interesting because of its role in speech motor planning and production (e.g., Alm, 2004b; Smits-Bandstra & De Nil, 2007; Xue, Aron, & Poldrack, 2008); moreover, imaging studies in stuttering (for an overview: see Watkins, Smith, Davis, & Howell, 2008) have revealed aberrant activity in the underlying cortical and subcortical structures of IC, namely the right prefrontal cortex (e.g., Casey, et al., 1997; Aron, Fletcher, Bullmore, Sahakian, & Robbins, 2003) and the fronto-basal ganglia circuit (Aron, Durston, et al., 2007; Chambers, Garavan, & Bellgrove, 2009; Congdon et al., 2010).

In a number of recent studies, we have found evidence for a possible role of IC in developmental stuttering (Eggers et al., 2009, 2010). These studies were done using the Children's Behavior Questionnaire (CBQ; Rothbart, Ahadi, Hershey, & Fisher, 2001), a parent-report temperament questionnaire for young children based on Rothbart's temperament model. Rothbart defines temperament as constitutionally based individual differences in reactivity and self-regulation (Rothbart, 1989a&b, 2011; Rothbart & Derryberry, 1981). Reactivity refers to motor, emotional, and attentional responses to internal and external stimuli and is operationalized in CBQ-scales such as Approach and Anger/Frustration. Self-regulation are those processes serving to modulate - i.e., facilitate or inhibit - the aforementioned reactivity, and is measured in the CBQ by scales such as Inhibitory Control and Attentional Focusing/Shifting. In a recent study of 3-to-8 year-old children (Eggers et al., 2010) we found that CWS scored significantly lower on the self-regulation-related scales of IC and Attentional Shifting and their overarching superfactor of Effortful Control, a finding that was consistent with other questionnaire-based studies in CWS (Embrechts et al., 2000; Karrass, et al., 2006).

In a subsequent study (Eggers, et al., 2012a), we examined whether the parent-reported lower self-regulation and inhibitory control in CWS could be corroborated experimentally using measures of attentional processes, which are central to these self-regulatory behaviors (Rothbart, Ellis, Rueda & Posner, 2003). Using the child version of the Attention Network Test (ANT; Fan, McCandliss, Sommer, Raz, & Posner, 2002; Rueda, et al., 2004), a computer task measuring the efficiency of the 3 attentional networks, we found CWS to be significantly lower in the efficiency of their orienting network, which is linked to the Attentional Shifting scale of the CBQ. However, for the executive control network, the network underlying IC, only a non-significant trend ($p = .066$) towards a lower efficiency for CWS was found. This led us to propose that our earlier reported CBQ-based IC findings were either not associated with a lower efficiency of the executive control network or, that the paradigm used to test the executive attentional network in the previous study lacked the necessary power to detect significant between-group differences. One reason for the need of more specific measures is the fact that executive attention consists of three integrated, measurable mechanisms, namely error detection and correction, conflict resolution, and inhibition of automatic responses (e.g., Norman & Shallice, 1986; Posner & Raichle, 1994; Rothbart & Posner, 2001). As such, attempts to measure a complex network such as executive attention using one global measure may be less likely to be successful. Some indirect support for this comes from the observation that in similar studies with ADHD children, differences in IC emerged by using a stop-signal paradigm (Pliszka, Liotti, Woldorff, 2000), while no differences were found for the broader underlying executive attentional network (Adolfsdottir, Sorensen, Lundervold, 2008; Booth, 2003). Therefore, the current study was designed to examine specifically IC in CWS by using a more targeted experimental measurement.

There is a considerable variability in the paradigms used to measure IC and several experimental measures have been developed to assess IC across different age-ranges, e.g. Go/NoGo or stop-signal tasks, Stroop-like or card sorting paradigms, and Mistaken Gift or Gift Delay Tasks (Baron, 2004; Carlson & Moses, 2001; Christ, White, Mandernach, & Keys, 2001). According to Barkley's model of response inhibition (1997), these measures are directed at evaluating three interrelated processes: a) inhibition of an initial prepotent response, which can be measured using a Go/NoGo task (Casey et al., 1997) or a gift delay task (Kochanska et al., 1996); b) stopping of an ongoing response, as measured for instance using a stop-signal task (Aron & Poldrack, 2005; Pliszka, Borcherding, Spratley, Leon, Irick, 1997); and c) protection of self-initiated responses from disruption by conflicting events or interference, for instance as measured by a Stroop-like task (Gerstadt, Hong, Diamond, 1994).

The purpose of this study was to test experimentally previous findings of parent-reported (CBQ) differences in IC between CWS and CWNS, in particular the inhibition of prepotent responses, using a Go/Nogo task. Based on these previous findings, we hypothesized that CWS, as a group, would be lower in IC compared to CWNS.

5.2 Method

5.2.1 Participants

Participants consisted of 30 children (24 boys and 6 girls) diagnosed with developmental stuttering and 30 typically developing nonstuttering children, matched by age (± 3 months) and gender to the CWS. The mean age was 7;05 years (*SD* = 1;05 years; range = 4;10-10;00) for the CWS and 7;05 years (*SD* = 1;05 years; range = 4;10-9;11) for the CWNS. All children were monolingual Dutch speaking. All participants had normal or corrected to normal vision and normal speech and language development (except for stuttering in the experimental group), based on the criteria described below. Participants had no known or reported neurological, psychological, developmental or hearing problems. CWS were recruited through their speech-language therapists, all specialized in fluency disorders, while the CWNS were recruited through the schools. All participants were paid for their involvement. The study was approved by the Research Ethics committee of Leuven University Hospitals and all parents signed an informed consent form. The Antwerp Screening Instrument for Articulation (ASIA-5, Stes & Elen, 1992), in which children are tested on their ability to produce age-appropriate phonemes in different word positions, was used to screen participants for articulation disorders. Children who did not pass this test were excluded from the study. Hearing function of all participants was evaluated as within normal limits as measured using the Accuscreen (Wood, 2003), a handheld hearing-screening device. To assess their language skills, participants were administered two subtests (Vocabulary Production and Sentence Production) of the

Language Test for Children (van Bon & Hoekstra, 1982). In the Vocabulary Production subtest participants needed to complete a sentence with the target-word, based on a presented picture. The Sentence Production subtest provides participants with phrases that have syntactical errors, which they have to correct. Participants had to score above percentile Pc16 (mean - 1SD) on both subtests in order to show normal language function. The mean percentiles on the Vocabulary Production subtest were 62 (range 28 - 99) for CWS and 71 (range 27 - 97) for CWNS. On the Sentence Production subtest, the mean percentiles were 54 (range 28 - 99) for CWS and 62 (range 28 - 94) for the CWNS. The differences between both participant groups on both Vocabulary Production, $t(58) = -1.71, p = .10$, and Sentence Production scores, $t(58) = -1.75, p = .09$, were not statistically different.

Two subtests of the Wechsler Intelligence Scale for Children-Third Edition Dutch (WISC-III; Vander Steene, et al., 1986; Wechsler, 2005), Vocabulary and Block Design, were administered to exclude cognitive group differences. The verbal subtest Vocabulary requires participants to explain the meaning of single words. In the nonverbal Block Design subtest, a visual reconstruction task, participants have to rebuild as quickly as possible a geometrical pattern, by using red and white cubes. Both subtests correlate highly with the WISC-III overall score (Groth-Marnat, 2009). The mean scores for the Vocabulary subtests were 19.20 for the CWS (range 7 - 34) and 19.73 (range 7 - 41) for the CWNS. On the Block Design subtest, the CWS scored on average 23.77 (range 6 - 58) compared to 28.60 for the CWNS (range 4 - 52). No significant between-group differences were found for either Vocabulary, $t(58) = -.26, p = .80$, or Block Design, $t(58) = -1.34, p = .19$.

Parental socio-economic status was determined based on the combined scores of the highest educational level (1=primary education, 2=high school, 3=college degree, 4=university degree) for each parent. The average score for parents of CWS was 5.67 (range 3-8), and for parents of CWNS it was 5.93 (range 4-8). The between-group difference in socio-economic status was not significant, $t(58) = -.68, p = .50$.

In order to get a better and more valid understanding of overall stuttering severity, spontaneous speech samples were obtained from all participants during two free play situations recorded on different days with the first author. A minimum of 300 words per participant was used to calculate scores on the Stuttering Severity Instrument-3 (SSI-3; Riley, 1994). The CWS who participated in the study produced at least three within-word disfluencies (sound/syllable repetitions, including monosyllabic word repetitions, prolongations or blocks) per 100 words of spontaneous speech (Conture, 2001) and scored at least mild on the SSI-3 (Riley, 1994). The average percentage stuttered words was 8.54 (*SD* = 6.77) for CWS and .98 (*SD* = .88) for CWNS. Thirteen of the CWS were classified as mild, 15 as moderate, 1 as severe, and 1 was rated very severe.

5.2.2 Materials

5.2.2.1 Baseline Speed Task

To avoid the possible confound of between-group reaction time differences on the experimental Go/NoGo task, all participants were administered the baseline speed subtask of the Amsterdam Neuropsychological Tasks (De Sonneville, 2009) prior to the experimental task (see below). Completing this task first also allowed for greater familiarization with the computer equipment to be used during the experiment. Each trial began with a white fixation cross on a black computer screen background. As soon as a white square in the middle of the screen (the target signal) replaced the fixation cross, participants had to press a response button as quickly as possible. Practice sessions of 10 trials were followed by two experimental blocks of 32 trials each, one for the right index finger and one for the left index finger, as is standard for this task. Target signal duration was variable and lasted until a response was recorded. Valid responses fell between 150 ms and 4000 ms after stimulus onset. Inter-trial intervals varied randomly from 500 ms to 2500 ms.

5.2.2.2 Experimental task: Go/NoGo Task

The Go/NoGo task of the Amsterdam Neuropsychological Tasks (De Sonneville, 2009) is a computer task that measures the inhibition of prepotent responses. An overview of the paradigm is shown in Figure 5.1. The Go/NoGo task consists of a practice session of 8 trials, followed by an experimental block of 48 trials (24 Go- and 24 NoGo-trials). Each trial began with a fixation period during which the child focused on a central white cross on a black computer screen background. This fixation period had a fixed duration of 500 ms and was followed by the presentation of the target stimulus. Two different stimuli were possible: a) the Go-stimulus (a green walking man), and b) the NoGo-stimulus (a red standing man). Both target stimuli were presented randomly but with equal frequency during the experimental block. Prior to the practice session, children were shown pictures of the Go- and NoGo-stimuli and were explained they respectively had to press the response button or refrain from pressing. When they understood these task requirements, the practice session was started. The goal of this practice session was to get the children acquainted with the task. Children were instructed to press the response button as quickly as possible in response to the Go-stimulus, and to refrain from pressing the response button when the NoGo-stimulus appeared (i.e., the measure for efficiency of IC). They were told to make as few mistakes as possible. The response button was the right or left mouse button below the track pad of the laptop, depending on whether the child responded with the left or right-hand. A target stimulus remained on the screen until either the child responded or the maximum stimulus duration of 800 ms had been reached. No feedback was given after the response. As part of the standard procedure of this Go/NoGo task, valid responses fell between 200 ms and 2300 ms after stimulus onset. The total trial duration was fixed at 2800 ms. The overall duration of the task was about 4 minutes. Normative data are available for children between the ages of 4 and 13 years (De Sonneville, 2009).

For each participant, the frequency of the following variables was automatically recorded and stored: a) 'hits' (when a Go-stimulus was followed by a response falling between 200 and 2300 ms after stimulus onset), b) 'misses' (when a Go-stimulus was not followed by a response), c) 'false alarms' (when a NoGo-stimulus was followed by pressing the response button between 200 and 2300 ms after stimulus onset), and d) 'premature responses' (when the response button was pressed between 0 and 200 ms after stimulus onset). In case a child exhibited a false alarm or premature response on two or more trials out of the 48 trials, this was defined as exhibiting 'multiple false alarms' or 'multiple premature responses'. In addition, for the variables 'hits' and 'false alarms' mean RTs were also recorded.

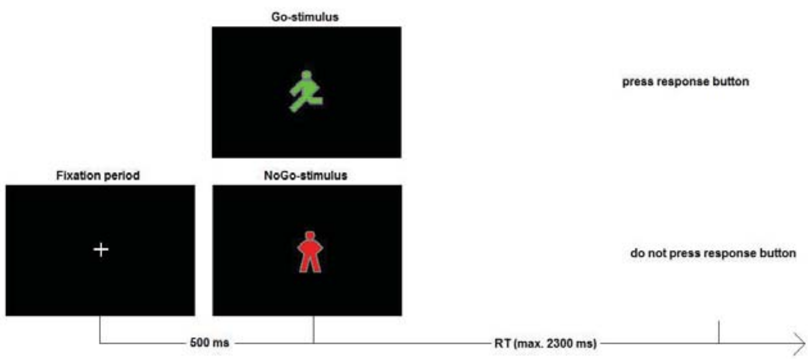


Figure 5.1: Overview of the Go/Nogo task

5.2.3 Procedure

Tests were conducted in a quiet setting at the home of the participant during two separate visits (test sessions A and B) of approximately 45 minutes each. All test sessions were conducted by the first author, a qualified fluency specialist. During test session A participants were administered the speech and language tests, and the hearing screening. The first spontaneous speech sample during play also was collected. During test session B, participants completed the simple reaction time (RT) task and the Go/NoGo task. In addition, the intelligence subtests, and the second spontaneous speech sample was collected. In order to minimize a possible test order confound, half of the participants completed test session A during the first visit while the other half completed test session B first.

The stimuli were presented on a 15-inch screen of a laptop computer, placed on a table. The distance between participant, seated on a chair, and computer screen was approximately 18 inches. To avoid distracting visual stimuli, a large black pliable cardboard was positioned around the laptop, and participants wore noise-reducing headphones to minimize possible distracting environmental sounds.

5.3 Results

Differences in baseline speed RT were evaluated using a t-test. The mean RT for CWS (414 ms, *SD* = 91) and CWNS (423 ms, *SD* = 115) was not significantly different: $t\ (58) = -.95,\ p = .72$.

Table 5.1: Mean percentage of misses, false alarms, and premature responses for CWS and CWNS.

	Misses		**False alarms**		**Premature responses**	
	M	***SD***	***M***	***SD***	***M***	***SD***
CWS	.56	1.81	8.47	6.79	2.80	4.55
CWNS	.69	1.92	4.72	4.61	.27	1.06
U-value	435		308*		296**	

*p < .05, **p < .005

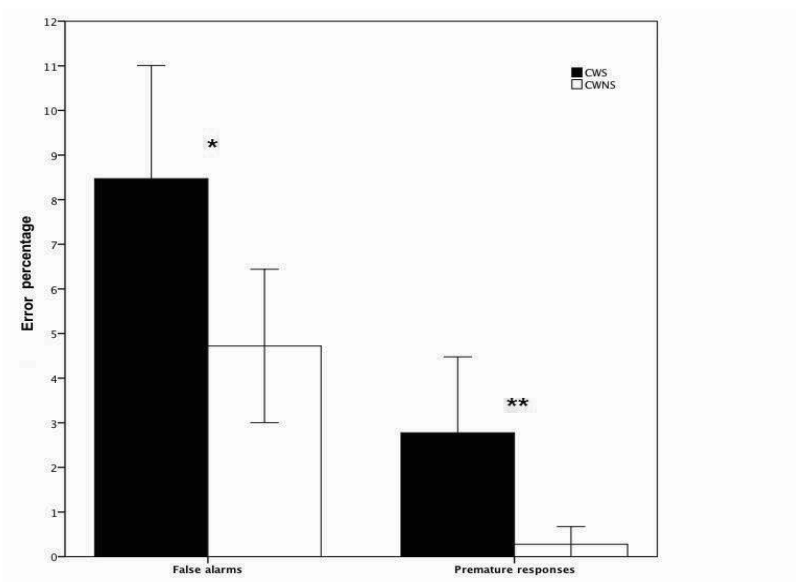
Table 5.2: Correlations between the Go/NoGo measures (misses, false alarms, and premature responses) and chronological age for CWS, CWNS, and both groups combined.

Measures		**Misses**		**False alarms**		**Premature responses**	
	Group	r_s	***p***	r_s	***p***	r_s	***p***
Age	CWS	-.35*	.05	.15	.41	.00	.99
	CWNS	-.49*	.01	.20	.29	-.28	.14
	Combined	-.43**	.00	.18	.17	-.09	.51
False alarms	CWS	.04	.84	1		-.01	.97
	CWNS	.00	.99	1		-.16	.39
	Combined	-.01	.96	1		.07	.59
Premature responses	CWS	-.01	.96	-.01	.97	1	
	CWNS	.33	.08	-.16	.39	1	
	Combined	.06	.66	.07	.59	1	

*p < .05, **p < .005

Table 5.1 provides an overview of the mean error percentages of Go/NoGo task variables (misses, false alarms, and premature responses) for both participant groups. Spearman's rank correlations were calculated to examine the relationships between these variables and chronological age (Table 5.2). No significant correlations were found between the Go/NoGo task variables, pointing to the independence of these measures; the significant negative correlation between chronological age and misses, for both CWS and CWNS, reveals that less misses occured with increasing age.

Between-group differences in error percentages of Go/NoGo task variables were evaluated using nonparametric Mann-Whitney U tests for 2 samples with participant group as the independent variable and error percentages of misses, false alarms and premature responses as dependent variables respectively. Between-group differences emerged for false alarms, U (58) = 308, Z = -2.17, p < .05, and premature responses, U (58) = 296, Z = -3.08, p < .005, showing that the mean number of false alarms and premature responses was higher in the stuttering group than in the nonstuttering group (Figure 5.2). No significant differences were found for misses, U (58) = 435, Z = -.385, p = .70.



* p < .05, ** p < .005

Figure 5.2: Error percentages for CWS and CWNS with significant between group differences.

In order to explore the between group differences in more detail, the data for false alarms and premature responses were analyzed further. The total number of false alarms ranged from 0 to 5 for the CWS and from 0 to 3 for the CWNS. Although the likelihood of having false alarms was the same for the children in the stuttering as for the children in the nonstuttering group (χ^2 = 1.49; *df* = 1; *p* = .22), the likelihood of having multiple (≥2) false alarms on the other hand did differ significantly between both groups, χ^2 = 10; *df* = 1; *p* < .005, suggesting that more CWS (60%) exhibited multiple false alarms compared to CWNS (20%). In other words, the number of children with false alarms was not different between the two groups, but among those children who had false alarms, CWS had a higher frequency than did CWNS (higher mean percentage of false alarms for CWS according to the Mann-Whitney). This is consistent with the reported finding of more multiple false alarms among the CWS.

A similar analysis was done for premature responses. The total number of premature responses ranged from 0 to 4 for the CWS and from 0 to 1 for the CWNS. More CWS (47%) exhibited premature responses than CWNS (7%), a difference that was statistically significant (χ^2 = 12.27; *df* = 1; *p* < .001). This was also the case for multiple (≥2) premature responses (χ^2 = 9.23; *df* = 1; *p* < .005). Only CWS showed multiple premature responses (27%), while none were seen in the CWNS (Table 5.3).

Differences in mean RTs for hits and false alarms were analyzed using separate univariate ANCOVAs with participant group as factor and mean RT of hits and mean RT of false alarms as dependent variables, respectively; chronological age was set as a covariate. Mean RT of hits was similar for both participant groups, *F* (1, 57) = 1.67, *p* = .20, while mean RT of false alarms was significantly shorter for the CWS, *F* (1, 43) = 5.65, *p* < .05. Table 5.4 gives an overview of the mean RTs for both participant groups.

Table 5.3: Number of CWS and CWNS exhibiting false alarms or premature responses.

	False alarms		**Premature responses**	
Number of responses	***CWS***	***CWNS***	***CWS***	***CWNS***
1	7	15	6	2
2	7	2	7	0
3	4	4	0	0
4	4	0	1	0
5	3	0	0	0

Table 5.4: Mean reaction times in ms for CWS and CWNS.

	Hits		False alarms	
Group	***M***	***SD***	***M***	***SD***
CWS	509	132	382*	111
CWNS	534	104	457*	145

*p < .05

5.4 Discussion

Previous studies based on parent reports already reported mean group differences in IC between CWS and CWNS, with the CWS scoring lower than the control participants (Eggers et al., 2009, 2010; Embrechts et al., 2000), although this finding was not confirmed in all studies (Anderson & Wagovich, 2010). The present study used a computer-based Go/NoGo-paradigm to investigate the presence or absence of group differences in IC.

5.4.1 CWS, as a group, exhibited a less controlled response style

The most common error type in both participant groups was false alarms, resulting from a failure to inhibit a response to the NoGo-signal; misses, i.e., a failure to respond to the Go-signal, on the other hand were much less frequent. Classically, false alarms have been called the most important measures in Go/NoGo-tasks (Christ et al., 2001) and have been linked to a less controlled, more impulsive response style. Misses, on the other hand, might reflect attention and concentration factors (Baron, 2001; Trommer, Hoeppner, Lorber, & Armstrong, 1988). Our results showed that CWS, as a group, had a higher mean number of false alarms compared to CWNS, with no significant differences in mean percentage of misses. In other words, CWS were less able to inhibit their responses to non-targets. More in depth analyses revealed that false alarms occurred as often in CWS as in CWNS but of those children who had false alarms, CWS had more false alarms compared to the CWNS. This seems to imply that at least some CWS were less able to adapt their response style, e.g. slowing down their reactions, after experiencing response errors. This also appears to be corroborated by the fact that RTs for false alarms in CWS were significantly faster compared to those in the control group. This latter observation might be explained by a speed-accuracy trade-off (also known as Fitts' law; Förster, Higgins, Bianco, 2003; Magill, 2011) which suggests that CWS, compared to CWNS, do not slow down their RTs (after experiencing response errors) but try to maintain high RTs, resulting in a higher occurrence of false alarms. This interpretation appears to find support in other studies, using paradigms in which attentional shifting/switching processes have been known to play a role (Eggers et al., 2012a;

Subramanian & Yairi, 2006), that have reported a tendency for shorter RTs among CWS. This observation is remarkable because most previous studies (for an overview see Bloodstein & Ratner, 2008) have shown that persons who stutter (PWS) have longer RTs compared to persons who not stutter (PWS).

CWS also made significantly more premature responses compared to CWNS. Compared to false alarms, premature responses were much more infrequent in both participant groups, but especially in the CWNS. Only a few CWNS exhibited a single premature response during the paradigm whereas almost half of the CWS group exhibited these kinds of errors and over a quarter exhibited multiple premature responses. Premature responses, defined as responses falling between 0 and 200 ms after stimulus onset, were not a direct reaction to the presented stimulus but rather could be considered resulting from impulsivity during the experimental task; in other words, this finding points to a more impulsive response style (e.g., Ballanger et al., 2009) and/or to a higher anticipatory load/expectation of the upcoming signal. The observation that more CWS have difficulties with adjusting their response style after experience response errors parallels our finding on false alarms. These results (more false alarms and more premature responses) are consistent with earlier CBQ-based findings of lower IC in CWS (Eggers et al., 2010; Embrechts et al., 2001). They also challenge the finding by Anderson & Wagovich (2010) who reported no between-group differences on IC between CWS and CWNS. It is possible that the nonsignificant finding in the Anderson et al. study was due to the considerably low sample size (9 CWS and 14 CWNS). The fact that CWS were found to be lower in IC although they were not inattentive (i.e., no differences emerged on the frequency of hits/misses) also seems to confirm previous parent-questionnaire based findings of CWS scoring similar to CWNS on the CBQ-scale of Attentional Focusing (i.e., the tendency to maintain attentional focus upon task-related channels; Eggers et al., 2010) and on the Behavioral Style Questionnaire (BSQ; McDevitt &Carey, 1978) scale of Attention Span/Persistence (i.e., the ability to continue the activity in the face of distractions; Anderson et al., 2003). Finally, our finding that CWS seemed less able to adapt their response style after experiencing response errors, as indicated by more multiple false alarms and premature responses, seems to correspond to Anderson et al.'s (2003) finding that CWS scored lower on the BSQ-scale of Adaptability (i.e., the ease or difficulty with which behaviors can be changed in a desired way), compared to CWNS.

In one of our previous studies, examing the underlying attentional network of IC (Eggers et al., 2012a), only a non-significant trend towards a lower efficiency of the executive control/attention network emerged. Based on that observation, we already hypothesized that a more specific testing paradigm might be needed to fully evaluate IC. This suggestion seems to be confirmed by our current data and demonstrates that the underlying executive attention network encompasses other components besides the inhibition of automatic responses (e.g., Norman & Shallice, 1986; Posner & Raichle, 1994).

5.4.2 Theoretical implications for the development of stuttering

While response execution, as measured by response speed and accuracy, generally was found to improve with increased age (Kail, 1991), not all results with regard to the development of IC are consistent. While some have argued for a developmental effect for IC between the age of 4 and 12 years or later (e.g., Bedard et al., 2002; Carver, Livesey, & Charles, 2001; Durston et al., 2002; Williams et al., 1999), others have suggested that IC primarily develops during early childhood with marked improvements between 3 and 6 and only limited development after the age of 7 (Christ et al., 2001; Diamond & Taylor, 1996; Frye et al., 1995; Gerstadt et al., 1994; Johnstone et al., 2007; Kochanska et al., 1996; Schachar & Logan, 1990). Our data seem to support the latter findings since no correlations were found between chronological age and the most relevant measure of IC, namely false alarms. While, descriptively, both CWNS and CWS showed less missed responses as age increased, it is important to note that over 60% of our participants were over the age of 7 while only 10% were younger than 6 years. It is likely, thus, that the children who participated in our study were too old to show the typical developmental pattern for IC.

Data from imaging (e.g., Casey, et al., 1997), ERP (e.g., Bokura, Yamaguchi, Kobayashi, 2001; Johnstone et al., 2007), and lesion studies (e.g., Aron, Fletcher, Bullmore, Sahakian, & Robbins, 2003) have provided converging evidence for the right prefrontal cortex (inferior frontal gyrus and medial frontal areas, especially pre-SMA) as one of the core anatomical correlates of inhibitory control. In addition to these frontal areas, the basal ganglia, including the subthalamic nucleus, also play a crucial role in this predominantly right-hemispheric network for motor response inhibition (Boehler, Appelbaum, Krebs, Hopf, & Woldorff, 2010), and this for both manual and spoken responses (Xue, Aron, & Poldrack, 2008). Recently, more support was found for this network of cortical and subcortical regions, typically identified as the fronto-basal ganglia circuit (Aron, Durston, et al., 2007; Chambers, Garavan, & Bellgrove, 2009; Congdon et al., 2010). The basal ganglia, a conglomerate of subcortical nuclei, are the core components of extensive circuits linking the cortex to the frontal lobe cortex (pre-SMA). There is evidence that this linking takes place via a direct (via the striatum and internal globus pallidus) and an indirect pathway (via the striatum, external globus pallidus, subthalamic nucleus, and internal globus pallidus) (Mink & Thach, 1993; Mink, 1996). More recently, evidence was found also for a so-called 'hyperdirect' pathway, which does not pass through the striatum (Ballanger et al., 2009; Nambu, Tokuno, & Takada, 2002). Both indirect and hyperdirect routes are responsible for inhibition of the motor program while the direct route activates the desired motor program. It is suggested that upon presentation of a Go-signal, the response is released via the direct pathway, after all motor programs were suppressed via the hyperdirect pathway; No/Go signals are believed to be mediated by the indirect or hyperdirect routes (Boehler et al., 2010; Chambers et al., 2009).

Interestingly, it was shown that in patients with Parkinson's disease both ventral and dorsal subthalamic nucleus stimulation improved motor symptoms but only ventral stimulation affected the Go/NoGo performance resulting in decreased hits and increased false alarms (Hershey et al., 2010).

Imaging studies in stuttering have revealed aberrant activity in these cortical and subcortical structures (for an overview: see Watkins, Smith, Davis, & Howell, 2008). The structures that form part of the cortical-basal ganglia network have been implicated in several emerging (theoretical) conceptualizations about underlying processes of developmental stuttering (e.g., Alm, 2004b; Smits-Bandstra & De Nil, 2007). Alm hypothesized that this fronto-basal ganglia network plays an important role in the etiology stuttering. He claims that the core dysfunction of stuttering might lie in the "*impaired ability of the basal ganglia to produce timing cues*" (pp. 359). As described by Alm, at the end of one component in a movement sequence, the globus pallidus generates an internal cue triggering the SMA to switch to the next component of the sequence; failing to generate these cues might explain some of the core features of stuttering, namely disruptions in the speech motor act. Smits-Bandstra and De Nil have proposed that stuttering might be associated with deficits in motor sequence skill learning and automaticity development, processes for which the involvement of the basal ganglia and the fronto-basal ganglia circuit is well established (Saint-Cyr, 2003). The current findings that more CWS are having difficulties with adjusting their response style after experience response errors might provide further evidence for this hypothesis.

Several authors (Bernstein Ratner & Wijnen, 2006; Postma & Kolk, 1993) have suggested stuttering to be the result of aberrant monitoring during linguistic processing. Most of these conceptualizations are based directly or indirectly on Levelt's model (1983) of language production. According to this model, language production is comprised of 3 stages, namely the conceptualization, formulation and articulation phase. This model also assumes the existence of several monitoring loops, checking the output at different stages of the production process. According to some, IC plays an important role in this monitoring process. That is, reduced IC might affect linguistic processing and have an impact on error-detection or error-processing (Engelhardt, Ferreira, & Nigg, 2009; Meyer, Wheeldon, & Krott, 2007). In our earlier work we already hypothesized a possible link between aberrant monitoring and findings in CWS of lower IC (Eggers et al. 2010) and lower efficiency of attentional orienting (Eggers et al., 2012a). Recently, Engelhardt, Corley, Nigg, and Ferreira (2010), studying children with ADHD, found evidence for a relation between IC and the ability to repair speech and language disfluencies. Although speculative, we might therefore link our findings of lower IC to possible difficulties in monitoring of speech production.

Finally, our findings could also be interpreted from a more temperament-oriented point of view. Children with a lowered IC are less able to regulate successfully their emotions (Carlson & Wang, 2007; Kochanska et al., 1996; Walcot & Landau, 2004), which, in turn, may lead to an increased emotional arousal response in stressful situations. Furthermore, it might increase the amount of stress-related situations that CWS encounter because lower IC impedes their ability to withhold their responses long enough to consider the complexities of a specific situation and to engage appropriate social skills. Several studies have shown that emotional arousal and anxiety have the potential to exacerbate stuttering symptoms (Alm, 2004a; Ezrati-Vinacour & Levin, 2004; Menzies, Onslow, & Packman, 1999). Consequently, lower IC might increase the amount of emotional arousal some CWS experience in stressful situations, impacting their stuttering symptoms. This is in line with the questionnaire-based finding by Karrass et al. (2006) of lowered emotion regulation in CWS, providing support for their 'Emotional Reactivity, Regulation and Stuttering (EERS) Model'. In this model, emotion regulation and reactivity are considered exacerbating or maintaining factors for childhood stuttering.

5.4.3 Possible clinical implications

Although clinical implications based on the results from the current study may be considered premature, lowered IC has been found repeatedly in studies of CWS, both using parent-questionnaires and experimental paradigms. This may at least allow us to make some speculative clinical suggestions. CWS, who exhibit a lowered IC, would most likely exhibit difficulties in suppressing prepotent responses across a variety of settings (e.g., school setting, playing with a friend). Therefore it might be important to council parents that these children may have more difficulties dealing with everyday situations requiring response inhibition (e.g., following instructions, waiting for something, ending an activity because he/she is asked to), resulting in increased emotional arousal. Both parental guidance techniques (e.g., "wait time technique", giving the child more time to comply) as well as helping children to acquire more self-regulatory behaviors might be appropriate (Kristal, 2005; Wodka et al., 2007). Working on increasing self-regulatory behaviors also may include identifying difficult situations, discussing expected behaviors, consequences of reacting in a certain way, helping the child to use self-directed speech, and using reminders (Kristal, 2005). The above-mentioned approaches in CWS, similar to the frequently used problem-solving strategies in cognitive-based stuttering treatment programs (see e.g. Shapiro, 1999), are aimed at decreasing the emotional arousal, and thus reducing its possible impact on the exacerbation of stuttering symptoms.

Our findings also seem to imply that some CWS are less efficient in altering their response style, e.g. slowing down, after experiencing response errors. Possibly, this could lead to longer sound prolongations or repetitions, or even the observed tendency to cluster disfluencies (Sawyer & Yairi, 2010). Findings like those reported here might validate stuttering treatment components that allow clients to monitor and conscientiously change their speech patterns such as increased monitoring of one's own speech, providing proprioceptive feedback, altering speech rate, providing positive feedback for fluent rather than disfluent speech (e.g., Guitar & McCauley, 2010).

5.4.4 Caveats, limitations and suggestions for future research

While the findings reported here are intriguing, it should be noted that the differences represent group tendencies and do not reflect individual performances. However, compared to the control group, a significantly larger proportion of CWS showed differences on the occurrence of multiple commission errors (60% of the CWS versus only 20% of the CWNS). Observations such as this suggest that problems with IC might impact stuttering development and/or maintenance for at least a considerable subgroup of CWS. Seery et al. (2007) already highlighted the need for delineating stuttering subtypes in order to study possible different developmental pathways of early stuttering (see also e.g., Tumanova, Zebrowski, Throneburg, & Kulak Kayikci, 2011). The finding of multiple false alarms in a large proportion of CWS might be used as a potential subtyping feature in future studies.

Moreover, although this study was not designed to evaluate the role of IC in the continuation and/or development of stuttering, given the age distribution of the sample it is possible for the reduced IC to be related to a tendency to develop persistent stuttering. Therefore, it might be interesting for future studies to evaluate if similar findings are also apparent in adults who stutter since this might yield additional insights in a potential role for IC in the persistence or recovery in stuttering.

While the current investigation was motivated by findings from a previous parent questionnaire study (Eggers, et al. 2010), no questionnaire was administered in the present study. Follow-up studies might consider combining multiple independent measures of IC, such as computer tasks and parental questionnaires, within the same design. In studying temperamental components in CWS, such as IC, it is also important to include language testing of the participants because of the possible interaction between temperament and language abilities (e.g., Bird, Reese, & Tripp, 2006). In the current study, there were no significant between-group language differences.

CWS showed more false alarms in combination with significantly faster RTs for false alarms. While we interpreted these findings by stating that CWS were less able to adapt their response style by e.g. slowing down their reactions, we acknowledge that another, although in our view less likely interpretation is possible, namely that CWS are simply less skilled in inhibiting the prepotent response rather than being less able to adapt their response style.

A difference between the current study and a number of previous studies is that the overall frequency of Go- and No/Go-signals in our study was held equal. Other investigators (e.g, Casey et al., 1997) used a proportionally higher frequency of the Go-signals, thereby increasing the prepotent tendency towards response execution and thus increasing the inhibitory control demands during NoGo-signals. It would be interesting to examine whether increasing the proportion of Go-signals, and thus increasing inhibitory control demands, would result in larger group differences between CWS and CWNS.

Finally, although Go/NoGo and stop-signal paradigms both assess inhibitory control and thus also share some underlying neural substrates, the inhibition takes place at different stages of the response execution process: in a Go/NoGo paradigm the inhibition occurs when response execution is at preparational or early-activational level while in a stop-signal paradigm execution of the response is already initiated (Johnstone et al., 2007). Therefore, it would be interesting to find out if lower efficiency in IC would also be observed in CWS when executing a stop-signal task.

5.5 Conclusions

Our results, based on a computer based Go/NoGo paradigm, provide further support for the hypothesis that CWS and CWNS differ in IC. CWS, as a group, were lower in IC, which suggests a lowered ability to inhibit prepotent response tendencies. The findings were linked to previous IC-related studies and to emerging theoretical frameworks of stuttering development.

CHAPTER 6

Exogenously and endogenously triggered response inhibition in developmental stuttering

The content of this chapter has been submitted for publication as:
Eggers, K., De Nil, L., & Van den Bergh, B. (2012c). Exogenously and endogenously triggered response inhibition in developmental stuttering. *Manuscript submitted for publication.*

Parts of the content of this chapter have been presented as:
Eggers, K., De Nil, L., & Van den Bergh, B. (2012, March). *Endogenous and exogenous response control in children who stutter.* Poster presented at the Third European Symposium on Fluency Disorders, Antwerp.

Eggers, K. (2011, October). *Responscontrole in kinderen die stotteren. [Response control in children who stutter].* Lecture presented at the 12th Symposium Logopedie & Audiologie U Ghent.

Eggers, K., De Nil, L., & Van den Bergh, B. (2010, December). *Endogene en exogene responscontrole in stotterende kinderen. [Endogenous and exogenous response control in children who stutter].* Poster presented at the annual convention of the Vlaamse Vereniging voor Logopedisten *[*Flemish SLT federation*]*.

Eggers, K., De Nil, L., & Van den Bergh, B. (2010, November). *Endogenous and exogenous response control in children who stutter.* Poster presented at the annual convention of the American Speech-Language-Hearing Association, Philadelphia.

Abstract
Purpose: The purpose of the present study was to examine relations between children's exogenously triggered and endogenously generated (from within oneself) response inhibition and stuttering.
Method: Participants were 18 children who stutter (CWS; mean age = 9;01) and 18 children who not stutter (CWNS; mean age = 9;01). Participants were matched on age and gender (± 3 months). Endogenous and exogenous response inhibition was assessed by a sustained attention task (Amsterdam Neuropsychological Tasks; De Sonneville, 2009) and a stop signal task (Verbruggen, Logan, & Stevens, 2008) respectively.

Results: Results suggest that CWS, compared to CWNS, perform worse in tasks where response control has to be generated endogenously and perform comparable to CWNS in tasks where response control is externally triggered.
Conclusions: Our findings corroborate previous questionnaire-based findings (Eggers, De Nil, & Van den Bergh, 2010) of a decreased efficiency of response inhibition.

6.1 Introduction

Several authors have hypothesized about a possible role of the basal ganglia or the cortical and/or subcortical structures of the fronto-basal ganglia circuit in the pathophysiology of developmental stuttering (e.g., Alm, 2004b; Caruso, 1991; Smits-Bandstra & De Nil, 2007; Toyomura & Omori, 2004). Alm suggests that a dysfunction of this circuit may have various causes, such as focal lesions or aberrant neurotransmitter release, but implies that the core dysfunction lies in the "*impaired ability of the basal ganglia to produce timing cues*" (pp. 359). In a same line of reasoning, Smits-Bandstra and De Nil proposed that dysfunctions in this circuit might result in deficits in motor sequence skill learning and reduced automaticity development.

The fronto-basal ganglia circuit plays a crucial role in response inhibition. Response inhibition is the ability to suppress a preplanned (Eagle, Bari, & Robbins, 2008), a habitual or a prepotent response (Congdon et al., 2010) or behaviors that are inappropriate or no longer required (Chambers, Garavan, & Bellgrove, 2009). It refers to the suppression of both motor actions as well as higher order responses, such as thoughts and emotions (Verbruggen & Logan, 2009); it is critical to deliberately or unconsciously (Eimer & Schlaghecken, 2003) stopping automatic behaviors in response to goals or environmental contingencies; and, it is also a key component of executive control (Cools, 2008). Although response inhibition is sometimes used as a term for one specific process, namely stopping a motor response, it generally reflects a set of related response control processes such as attending to and interpreting stimuli, decision making based on these stimuli and related internal or external cues, response selection, and successfully executing the appropriate motor response (Eagle et al., 2008; Nigg, 2000).

Previous studies in stuttering have yielded results pointing in the direction of reduced response inhibition. Using the Children's Behavior Questionnaire (CBQ; Rothbart, Ahadi, Hershey, & Fisher, 2001), a parent-report temperament questionnaire, Eggers, De Nil, and Van den Bergh (2009, 2010) found a lowered score for 3-to-8-year-old CWS on the 'Inhibitory Control' scale, a finding in line with an earlier questionnaire-based study by Embrechts, Ebben, Franke, and van de Poel (2000). Since response inhibition can be directly linked to the broader concept of self-control (Aron, Durston, et al., 2007), the lowered scores for CWS on the CBQ-superfactor of Effortful Control (Eggers et al., 2010) and on the Behavioral Style Questionnaire (McDevitt & Carey, 1978) scales of emotional and attentional self-regulation (Karrass et al., 2006) corroborate these findings.

Several behavioral paradigms have been developed to investigate response inhibition across different age ranges and currently a variety of measures are being employed for which it is often assumed they all evaluate a common or at least closely related inhibitory mechanism (Baron, 2004; Chambers et al., 2009). Frequently used well-defined paradigms of response inhibition are stop signal tasks (e.g., Logan, 1994), gonogo tasks (e.g., Bokura, Yamaguchi, & Kobayashi, 2001), sustained attention to response tasks (e.g., Robertson, Manly, Andrade, Baddeley, & Yiend, 1997), continuous performance tasks (e.g., Klee & Garfinkel, 1983), and anti-saccadic tasks (e.g., Anderson, Husain, & Sumner, 2008). The common feature of these tasks is that they require participants to respond to one set of stimuli (Go trials) and inhibit their response to another set of stimuli (Stop trials). Response inhibition is also thought to be involved in other but related forms of 'effortful' inhibition, such as response interference control and response switching (e.g., Dimoska-Di Marco, McDonald, Kelly, Tate, Johnstone, 2011), which are generally assessed by using paradigms like the Stroop color word tasks (e.g., Milham et al., 2002) or flanker tasks (e.g., Eriksen, 1995). During these tasks participants need to maintain their goal-oriented behavior when confronted with distractors or strongly activated but misleading representations (Friedman & Miyake, 2004). Many of these paradigms have been used in studying response inhibition deficits in different clinical populations such as attention deficit and hyperactivity disorder (Schachar et al., 2007), autism (Schmitz et al., 2007), Parkinson's disease (Hershey et al., 2010), and Tourette's syndrome (Li et al., 2006). Traditionally these tasks have been used in children 6 or 7 years of age or older (Williams, Ponesse, Schachar, Logan, & Tannock, 1999) but Carver, Livesey, and Charles (2001) showed that modifications to popular tasks, such as the stop signal paradigm (e.g., the use of simple shapes as stimuli, longer stimulus presentation times, fewer trials), make them also appropriate for use in preschool and kindergarten children, and thus making them ideal tools to study aging and developmental effects across a wide age range.

Many authors have adhered to the idea that a single mechanism underlies the ability to inhibit responses by generalizing the results obtained from different paradigms previously mentioned and data point in the direction of at least partly overlapping circuits, especially with respect to the involvement of the pre-SMA and IFC (Aron, Durston, et al., 2007). Others have argued that these tasks assess slightly different but related response inhibition processes or even that it is not clear that all of these tasks isolate response inhibition processes rather than related control processes such as response selection, conflict resolution, sustained attention, and working memory (Nigg, 2000). At the least, there seems a reasonable amount of support for the existence of different forms of response inhibition such as endogenously and exogenously triggered response inhibition since different brain regions are involved depending upon whether response control is externally triggered (e.g., by an auditory signal indicating the response has to be inhibited) or internally generated, i.e., from within the person (Van den Bergh et al., 2005, 2006). The latter refers to tasks with no external stop signals and in which the person needs to self-sustain conscious processing of repetitive stimuli that would otherwise lead to habituation or distraction to other stimuli (Robertson et al., 1997). Support for the distinction between these two forms of response control can be found in the fact that several functions involving the prefrontal cortex (e.g., vigilance, directing of attention, generating motor patterns) have been found to implicate different areas depending upon whether they were externally or internally generated (Fernandez-Duque & Posner, 2001; Van den Bergh, 2005); moreover, both clinical (Robertson et al., 1997) and normal (Van den Bergh et al., 2005, 2006) populations have shown different proficiencies in externally versus internally generated response control. For example, findings in patients with Parkinson's disease point towards an impaired ability to self-initiate movements versus a better-preserved ability for externally cued movements (Georgiou et al., 1994; Hanakawa, Fukuyama, Katsumi, Honda, Shibasaki, 1999).

Although there are some studies in the area of stuttering that have evaluated response inhibition (see above), the nature and strength of this evidence might be considered limited. Especially since it only concerns three parent questionnaire-based studies, critiqued by some authors (e.g., Kagan, 2001, Vaughn et al., 2002) for parental bias susceptibility and low interparental agreement. Therefore, these findings would need to be confirmed by more objective measures of response inhibition, such as behavioral paradigms. Moreover, to our knowledge, no studies have specifically investigated differences in externally (exogenous) versus internally (endogenous) generated response inhibition. Thus the goal of the current study was to evaluate differences in both exogenous and endogenous forms of response inhibition between CWS and typically developing, nonstuttering children (CWNS), employing behavioral paradigms. For this reason, two paradigms were selected, a stop signal task (Verbruggen, Logan, & Stevens, 2008) and a sustained attention task (De Sonneville, 2009), of which it is assumed they respectively assess exogenous and endogenous response inhibition. The stop signal task consisted of a primary reaction time (RT) task where the participant had to press the left or right response button depending on the presented stimulus; on a limited number of trials an auditory stop signal, was presented after the primary stimulus, indicating the response had to be inhibited. The sustained attention task was a choice RT task where the participant also had to press the left or right response button depending on the presented stimulus, but this second task was especially chosen because it was a rather long and somewhat boring task with no pauses and longer interstimulus intervals where the participant had to self-sustain his/her attention despite of the numerous repetitive stimuli and the monotonous character of the task (Robertson, Manly, Andrade, Baddeley, & Yiend, 1996). Based on the earlier formulated hypothesis by Alm (2004b), and the findings on the impact of exogenous feedback on stuttering symptoms (Saltuklaroglu et al., 2003, 2004), our hypothesis was that CWS, compared to CWNS, would have a similar efficiency of exogenously triggered response inhibition but a lower efficiency of endogenously generated (from within themselves) response inhibition.

6.2 Method

6.2.1 Participants

Participants in the study were 30 boys and 6 girls between the ages of 7;04 and 10;11 years. Eighteen of the children (15 boys and 3 girls) were diagnosed with developmental stuttering. The other 18 children were nonstuttering, typically developing children, matched by age (± 3 months) and gender to the CWS. The mean age was 9;01 years (*SD* = 0;11 years) for the CWS and 9;01 years (*SD* = 1;00 years) for the CWNS. All children were monolingual Dutch speaking. Participants had no known or parent-reported neurological, psychological, developmental or hearing problems. All participants had normal or corrected to normal vision and normal speech and language development (except for stuttering in the CWS), based on the criteria described below. Participants and parents were all paid volunteers recruited through their fluency specialists (for the CWS) or through their school systems (for the CWNS). The Research Ethics committee of Leuven University Hospitals approved the study; informed consent forms were signed by all parents.

All participants were administered a verbal and nonverbal subtest of the Wechsler Intelligence Scale for Children-Third Edition Dutch (WISC-III; Wechsler, 2005). In the Vocabulary subtest participants had to explain the meaning of single words. The Block Design subtest, a visual reconstruction task, required participants to rebuild as quickly as possible a geometrical pattern, by using red and white cubes. Both subtests correlate highly with the WISC-III overall score (Groth-Marnat, 2009). To avoid a possible influence of socio-economic status (Hackman & Farah, 2009), parental socio-economic status was assessed based on the highest educational level (1=primary education, 2=high school, 3=college degree, 4=university degree) of both parents, resulting in a composite score. On the WISC-III, the CWS had a mean Vocabulary score of 28.28 (range 9 – 39) and a Block Design score of 42.56 (range 10 – 63) while the CWNS scored respectively 29.56 (range 15 – 41) and 44.50 (range 19 – 62). In terms of socioeconomic status, the average for CWS was 5.61 (range 4 – 8), and 6.22 (range 3 – 8) for the CWNS. No significant between-group differences were found for either Vocabulary ($t = .55$, $p = .59$), Block Design ($t = .47$, $p = .64$) or parents' socio-economic status ($t = 1.32$, $p = .20$).

Participants were also administered two subtests of the Language Test for Children (van Bon & Hoekstra, 1982). In the Vocabulary Production subtest participants had to complete a phrase with a target-word linked to the presented picture. The Sentence Production subtest required participants to correct syntactically incorrect sentences. Participants had to score above percentile 16 (mean – 1SD) on both subtests in order to show normal language function. All participants passed a screening for articulation and hearing disorders, using the Antwerp Screening Instrument for Articulation (ASIA-5, Stes & Elen, 1992), a picture based test used to evaluate if children are able to produce age-specific phonemes in different word positions, and Accuscreen (Wood, 2003), a handheld hearing-screening device, employing transient-evoked otoacoustic emissions. Mean percentiles on Vocabulary Production were 61 for the CWS (range 20 – 93) and 66 for the CWNS (range 20 – 96). For Sentence Production, CWS had a mean percentile of 56 (range 20 – 97) compared to 61 for the CWNS (range 20 – 99). No significant differences were found for Vocabulary Production ($t = .75, p = .46$) or Sentence Production ($t = .68, p = .50$).

Spontaneous speech samples were collected during two free play situations. For each participant, a minimum of 300 words was used to calculate severity scores on the Stuttering Severity Instrument-3 (SSI-3; Riley, 1994). CWS produced a minimum of three within-word disfluencies (sound/syllable repetitions, including monosyllabic word repetitions, prolongations or blocks) per 100 words of spontaneous speech (Conture, 2001) and scored at least mild on the SSI-3. Average word-based percentages of within-word disfluencies were 10.25 (*SD* = 5.22) for CWS and .91 (*SD* = .72) for CWNS. Nine CWS were classified as mild, 8 as moderate, and 1 as severe.

All participants were part of an ongoing series of studies on the relationship between temperament and attentional processes in developmental stuttering (see Eggers, De Nil, & Van den Bergh, 2012a, 2012b).

6.2.2 Materials

6.2.2.1 Baseline Speed Task

The baseline speed task of the Amsterdam Neuropsychological Tasks (De Sonneville, 2009), a simple RT task, was administered to all participants prior to the other tasks. In addition to serving as a preparation for the sustained attention and stop signal tasks, it provided simple RT measures that could be used as a baseline, preventing an influence of possible between-group differences in simple RT on the later measurements. The task consisted of two experimental blocks of 32 trials each, one for the right index finger and one for the left index finger (standard for this task), preceded by a 10-trial practice session. All trials started with a white fixation cross on a black background, followed by the stimulus, a white centralized square, disappearing with the response. Participants were instructed to press the response button as quickly as possible after the appearance of the target; valid responses fell between 150 ms and 4000 ms after stimulus onset. Inter-trial intervals varied randomly between 500 ms and 2500 ms.

6.2.2.2 Measure of exogenously triggered response inhibition: Stop Signal Task

The stop signal task (Verbruggen, Logan, Stevens, 2008) consisted of a primary choice RT task where on a random selection of trials an auditory stop signal appeared,

indicating participants to withhold their response. The primary task was a shape judgment task requiring participants to discriminate between a square and a circle. When the square was presented, they had to press to right mouse button below the track pad of the laptop and when a circle appeared the left mouse button needed to be pressed (see Figure 6.1). To make sure they understood this primary task they were shown several printouts on paper of stimuli and they had to demonstrate that they understood the task by pressing the correct mouse button. Each trial began with a white centralized fixation cross on a black background. The fixation period had a fixed duration of 250 ms and was followed by the presentation of the primary-task stimulus, which remained on the screen until the end of the trial. Participants had to respond as quick and accurately as possible. Maximum RT was set at 2500 ms. On 25% of the trials, the primary-task stimulus was followed by a stop signal (a 750 Hz tone, 75 ms in duration), presented through earphones, during which participants had to refrain from responding (stop trials). Interstimulus interval was 500 ms.
The speed of the inhibition process, the stop signal RT (SSRT), cannot be observed because the response to the stop signal is a covert response. Logan (1994) used a race model to estimate SSRT. In this model, two independent processes race against each other: the go process (triggered by the primary-task stimulus and resulting in a button press) and the stop process (triggered by the stop signal and resulting in response inhibition). Depending on which process finishes first, the response will be executed or inhibited. To estimate the SSRT, a specific tracking procedure was used in which the delay between the primary-task stimulus and the stop signal, the stop signal delay (SSD), was varied. When the SSD increased, the chances of responding on a stop signal trial also increased; in other words, the longer the interval between primary-task stimulus and stop signal, the more difficult it became to withhold the response. Initial SSD was set at 250 ms and was adjusted continuously as follows: if the child inhibited successfully, the SSD was increased with 50 ms (thus making it more difficult to inhibit the response on the next stop signal trial); if the child failed to inhibit, the SSD was decreased with 50 ms (thus making it easier to inhibit one's response on the next stop signal trial). The goal of this so-called 'staircase tracking procedure' was to allow children to inhibit their responses on about 50% of the stop signal trials (see Figure 6.2). SSRT was calculated by subtracting the mean SSD from the mean primary-task RT.

The task consisted of a practice session of 32 trials, followed by three experimental blocks of 64 trials each. Between each experimental block a pause of 5 minutes was inserted. During these intervals children received, as a reward, some pieces of a puzzle of a cartoon figure. At the end of the task they could complete the puzzle with all the pieces they had obtained. Total task duration was approximately 20 minutes, including 2 breaks of each 5 minutes.

The primary performance measure for the stop signal task was SSRT. In addition, mean SSD, mean RT for go trials, mean RT for stop trials, percentage of correct go trials, and percentage of missed go trials were calculated.

Figure 6.1: Overview of the stop signal task

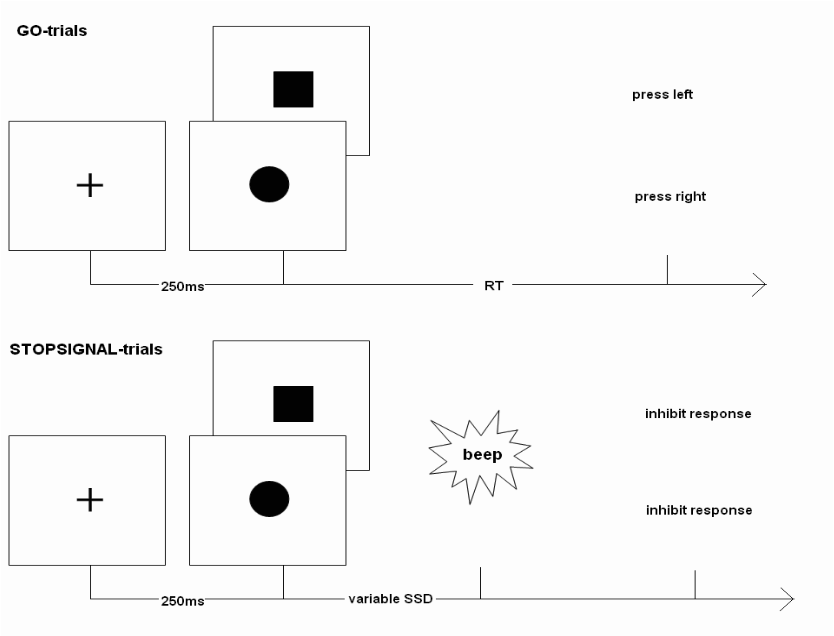
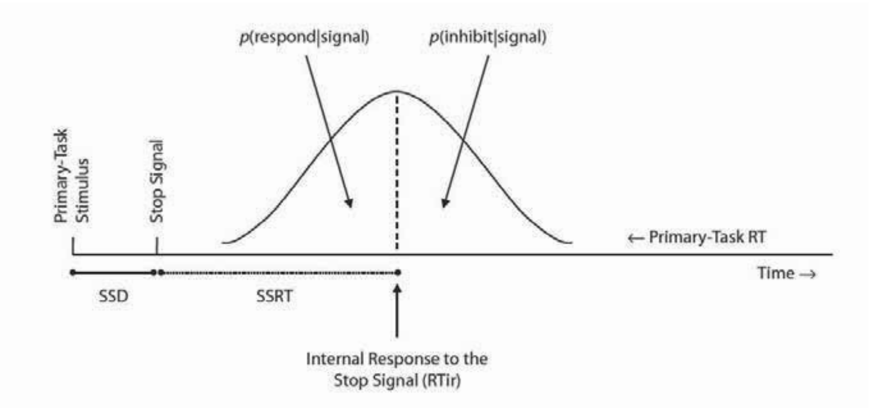


Figure 6.2: Illustration of the stop signal task probabilities of responding [p(respond|signal)] based on the horserace model. Adapted from Verbruggen, Logan, and Stevens (2008)

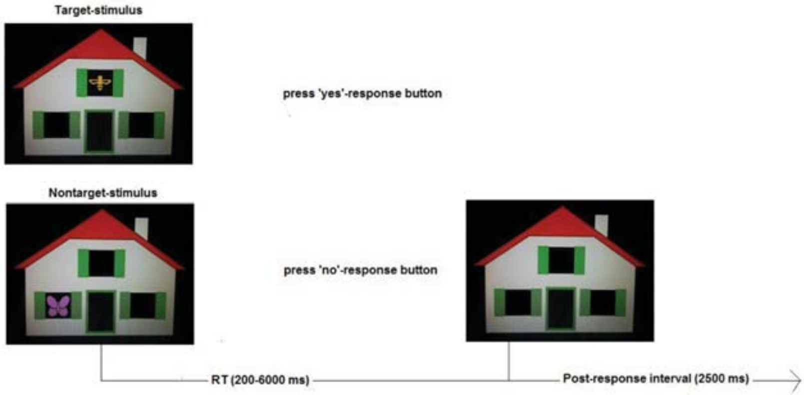


6.2.2.3 Measure of endogenously generated response inhibition: Sustained Attention Task

The sustained attention task of the Amsterdam Neuropsychological Tasks (De Sonneville, 2009) is a visual choice RT task. During the entire task a white house with 3 windows and a door is presented on a black computer screen (see Figure 6.3). Trials started with the presentation of the stimulus, one of 3 animals – a bee, a bird, and a butterfly – that appeared separately in one of the windows. If the target stimulus, i.e., the bee, appeared (in 50% of the trials), participants had to press as quickly as possible the 'yes' response button. If the target stimulus was absent (i.e., presentation of a bird or a butterfly; in 50% of the trials), they had to press the 'no' response button. For right-handed participants, the 'yes' and 'no' response button were respectively the right and left mouse button below the track pad of the laptop, while this was reversed for left-handed participants. If the wrong response button was pressed, an auditory feedback signal was generated. Before the task was started, participants were shown several printouts of the stimuli in order for them to demonstrate they understood the task. The signal duration was variable and dependent on response time. Valid responses fell between 200 and 6000 ms. The post-response interval had a fixed duration of 2500 ms. The task consisted of a practice session of 24 trials followed by one experimental block of 240 trials. Total task duration was approximately 15 minutes. The task was developed for children aged 5 and above (De Sonneville, 2009).

The frequency of the following variables were automatically calculated for each participant: a) 'misses' (when the target-stimulus was followed by a 'no' response), b) 'false alarms' (when a nontarget-stimulus was followed by a 'yes' response), c) 'premature responses' (when the response button was pressed between 0 and 200 ms after stimulus onset), and d) 'omissions' (when a stimulus was not followed by a response). Overall mean RT was taken as a measure of processing speed and SD of the RT as a measure of consistency in responding and ability to sustain attention.

Figure 6.3: Overview of the Sustained Attention Task



6.2.3 Procedure

Participants were tested in a quiet setting during two home visits (test session A & B) by the first author, a qualified speech-language therapist with expertise in fluency disorders. For the computer paradigms, stimuli were presented on a 15-inch screen of an Asus laptop computer. A black pliable cardboard screen was positioned around the computer to avoid distracting visual stimuli; headphones reduced distracting environmental sounds. Test session A involved the following tests: a) the Baseline Speed subtask of the Amsterdam Neuropsychological Tasks (De Sonneville, 2009), b) the Sustained attention task of the Amsterdam Neuropsychological Tasks (De Sonneville, 2009), c) the Vocabulary and Block Design subtest of WISC-III (Wechsler, 2005), d) the collection of a speech sample. During test session B the following tests were administered: a) the Stop signal task (Verbruggen, Logan, Stevens, 2008), b) the Vocabulary and Sentence Production subtests of the Language Test for Children (van Bon & Hoekstra, 1982), and c) the collection of a speech sample, and d) the ASIA-5 (Stes & Elen, 1992), and e) Accuscreen (Wood, 2003). To avoid an influence of testing order, half of the CWS and their matched controls, were presented with test session A during the first visit while the rest of the participants started with test session B.

6.3 Results

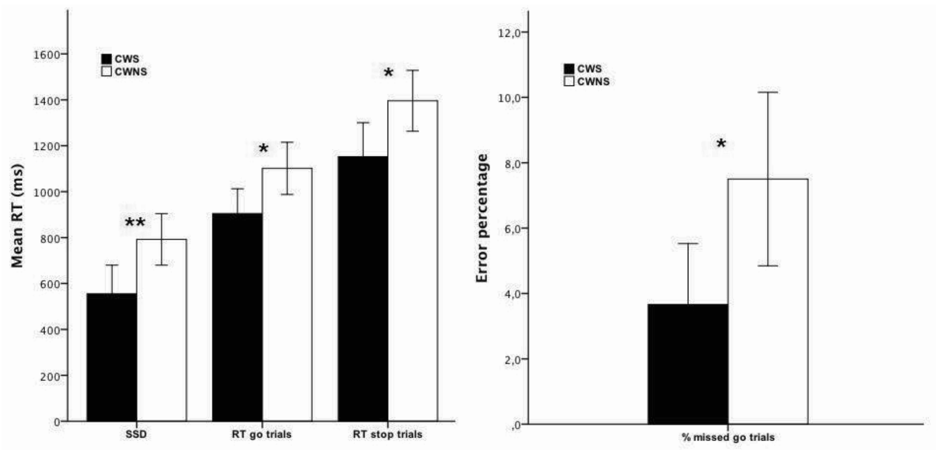
The average baseline speed RT was 366 ms (*SD* = 57) for CWS and 368 ms (*SD* = 49) for CWNS; both groups did not differ significantly: t (34) = -.11, p = .92.

Table 6.1 gives an overview of the performance measures of the Stop signal task for both participant groups. One-way Analyses of Covariance (ANCOVAs) were conducted to investigate the effect of participant group on each of these variables (SSRT, SSD, RT go trials, RT stop trials, % correct go trials, and % missed go trials); chronological age was set as a covariate. No differences were found for SSRT, F (1, 33) = .09, p = .77, and percentage of correct go trials, F (1, 33) = 2.75, p = .11. Between-group differences were found for RT go trials, F (1, 33) = 6.80, $p < .05$, RT stop trials, F (1, 33) = 6.50, $p < .05$, and SSD, F (1, 33) = 8.61, p = .005, showing that the RTs and SSDs were lower in the stuttering group than in the nonstuttering group. Also a significant lower percentage of missed go trials was found for the CWS, F (1, 33) = 6.23, $p < .05$ (Figure 6.4).

Table 6.2 provides an overview of the Sustained attention task variables for both participant groups. Between-group differences were evaluated using one-way ANCOVAs with percentages of misses, false alarms, premature responses, and omissions, overall mean RT and SD of overall mean RT as dependent variables and chronological age as covariate. Significant between-group differences emerged for percentage of misses, F (1, 33) = 9.22, p = .005, and percentage of false alarms, F (1,

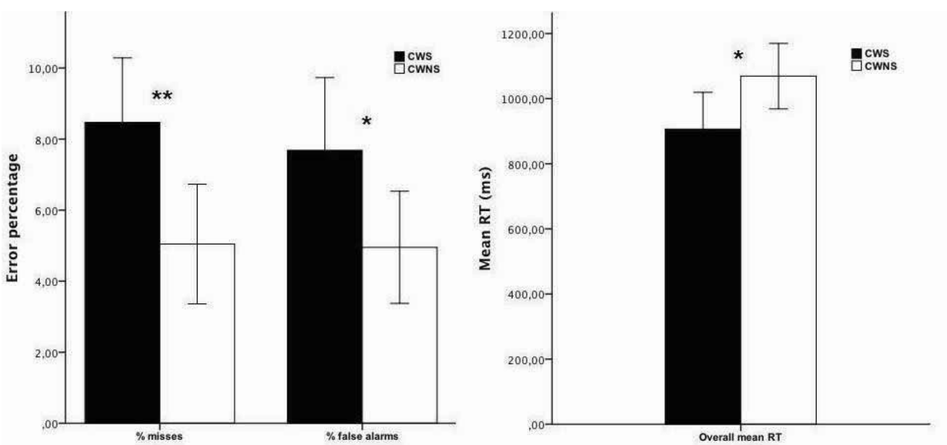
33) = 5.25, *p* < .05, showing that the mean number of misses and false alarms was higher in the stuttering group. CWS also had a significant lower overall mean RT, *F* (1, 33) = 6.29, *p* < .05 (Figure 6.5). No differences were found for percentage of premature responses, *F* (1, 33) = .00, *p* = .98, percentage of omissions, *F* (1, 33) = .00, *p* = .99, and SD of overall mean RT, *F* (1, 33) = 2.61, *p* = .12.

Figure 6.4: Mean stop signal task performance measures for CWS and CWNS with significant between group differences.



* p < .05, ** p < .005

Figure 6.5: Mean Sustained Attention Task measures for CWS and CWNS with significant between group differences.



* p < .05, ** p < .005

Table 6.1: Mean Stop signal task performance measures for CWS and CWNS

	SSRT		SSD		RT go trials		RT stop trials		% correct go trials		% missed go trials	
Group	*M*	*SD*	*M*	*SD*	*M*	*SD*	*M*	*SD*	*M*	*SD*	*M*	*SD*
CWS	594	114	557**	253	905*	217	1152*	298	94	4,55	3,66*	3,75
CWNS	604	123	792**	225	1101*	229	1396*	266	89	11,23	7,50*	5,34

*p < .05, **p = .005

Table 6.2: Mean Sustained attention task performance measures for CWS and CWNS

	% misses		% false alarms		% premature responses		% omissions		Overall mean RT		SD of overall mean RT	
Group	*M*	*SD*	*M*	*SD*	*M*	*SD*	*M*	*SD*	*M*	*SD*	*M*	*SD*
CWS	8,47**	3,65	7,68*	4,12	0,16	0,32	0,07	0,29	906*	227	329	153
CWNS	5,04**	3,39	4,85*	3,18	0,16	0,25	0,07	0,16	1069*	202	411	177

*p < .05, **p = .005

In order to test for a possible between-group difference in decline in performance over time (time-on-Task), the first 24 trials of the task were compared to the last 24 task trials, providing an index of the maximal decline over time. The effect of participant group on changes in respectively percentage of misses, percentage of false alarms, and overall mean RT was analyzed with separate repeated measures ANCOVAs with participant group as between-subjects variable and selected performance measures as within-subjects variables; chronological age was set as covariate. The group X time-on-Task interactions were not significant: percentage of misses, F (1, 33) = .02, p = .88, percentage of false alarms, F (1, 33) = .19, p = .66, overall mean RT, F (1, 33) = .28, p = .60.

6.4 Discussion

Previous questionnaire-based studies revealed differences between CWS and CWNS on response inhibition or on inhibition-related concepts, such as emotional regulation (Eggers et al., 2010, Embrechts et al., 2001; Karrass et al., 2006) pointing towards a lowered response inhibition in CWS. The current study employed two paradigms, a stop signal task (Verbruggen et al., 2008) and a sustained attention task (de Sonneville, 2009) to specifically examine two distinct forms of response inhibition, namely exogenously triggered response inhibition and endogenously generated response inhibition respectively.

6.4.1 CWS and CWNS were equally efficient in exogenously triggered response inhibition

With regard to the stop signal task, no differences were found on the most important measure of inhibition, namely SSRT, the speed of response inhibition. This is a parameter that cannot be directly measured but has to be estimated because if the inhibition process is successful, the response to the primary-task stimulus is suppressed. Our results showed that CWS were as fast in generating this response suppression as CWNS. In other words, if an external sound signals that a motor response needs to be interrupted, CWS are as fast as CWNS to react to this signal; furthermore, during this task, overall they were even significantly faster because of lower RTs both in go and stop trials. These shorter RTs were also found in previous computer-based RT paradigms (Eggers et al., 2011; Subramanian &Yairi, 2006) although remarkable at first sight because most studies (for an overview see Bloodstein & Ratner, 2008) showed longer RTs in people who stutter. The shorter RTs automatically resulted in shorter SSDs because due to the staircase-tracking procedure of the task, participants were able to inhibit their responses in about 50% of the trials; if participants respond faster to the primary stimulus, then also the time between the presentation of the primary stimulus and the stop signal needs to be shorter since otherwise participants will not be able to inhibit their responses in 50% of the trials. What is remarkable is that, despite the faster RTs in CWS, the percentage of correct go trials, i.e. trials with no stop signal where the response to the primary stimulus was correct, showed no between-group differences. Combined with the lower percentage of missed go trials, this leads us to conclude that CWS were as efficient in exogenously triggered response inhibition, even more, in these circumstances (during a task with external auditory signals) they appear to react faster (lower RTs) and seem more able to maintain their concentration and attentional focus (less missed go trials). The latter findings seem to be in line with Anderson, Pellowski, Conture, and Kelly's (2003) findings of lower scores for CWS on the distractibility scale (defined as "*the effectiveness of extraneous stimuli in drawing attention away from ongoing behaviors";* pp.1225) of the Behavioral Style Questionnaire. Overall, these findings confirm our original hypothesis of a similar efficiency of exogenously triggered response inhibition in CWS.

6.4.2 CWS were less efficient in endogenously generated response inhibition

The results of the sustained attention task showed a different pattern compared to the results of the stop signal task. CWS showed more misses and false alarms, or respectively more 'no' responses to target stimuli and 'yes' responses to nontargets. Classically, this kind of commission errors have been linked to a less controlled and more impulsive response style (Christ et al., 2001). This more impulsive response style might also be underscored by the shorter RTs, a result that was also found in the stop signal task. So for the sustained attention task shorter RTs coincide with higher error percentages, which can be interpreted in the light of a speed-accuracy trade-off, namely, that the CWS maintained their low RTs (instead of e.g., slowing down) despite of the errors they were making. Interestingly, this explanation though is not applicable to the stop signal task because faster RTs did not coincide with more errors (CWS even had a nonsignificant higher percentage of correct ‘go’ trials with smaller SDs), so without any apparent speed-accuracy-trade-off. Analyses of the differences between the two tasks might yield an explanation for this apparent performance difference between the two tasks. Possibly the higher number of trials in the sustained attention task (one block of 240 trials) versus the stop signal task (3 blocks of 64 trials each interspersed with a 5-minute pause between each block) might explain the previous difference. If this would have been the case, then comparing the first 24 trials with the last 24 trials (time-on-Task) should also have yielded a significant between-group difference, but a similar performance decline was found in both groups, so this aspect could not have contributed to the performance difference between the sustained attention and stop signal task. The performance difference could also not be attributed to a lowered attentional focus in CWS during the sustained attention task because no between-group differences were found in percentage of omissions, typical for concentration ‘slips’ (Baron, 2001; Trommer, Hoeppner, Lorber, & Armstrong, 1988).

A plausible explanation might be found in the different characteristics of the ‘stop’ trials (stop signal task) and ‘nontarget’ trials (sustained attention task) of both tasks. The most obvious difference between these two conditions is that in the stop signal task an external, auditory signal was provided, indicating that the participant had to refrain from responding. This external signal might exogenously trigger response inhibition and facilitate the inhibition process. Since no external, auditory signals were available during the ‘nontarget’ trials of the sustained attention task, response inhibition was not triggered externally and had to be generated endogenously, i.e. from within the child. Under these circumstances, CWS seemed to be less able to withhold their prepotent response long enough (i.e., responding with their dominant) in order to be able to give a correct answer (i.e., pressing the response button with their nondominant finger) in a ‘nontarget’ trial.

Another difference between the two conditions was that the sustained attention task contained a decision-making component, which was absent in the stop signal task. In the sustained attention task used, children had to select a response strategy (‘yes’-response or ‘no’-response) before response execution, depending on the presence of a target or nontarget stimulus. Each trial of the stop signal task always started out as a ‘go’-trial, and in case an auditory signal appeared, this always meant that the response had to be inhibited, so the child did not have to make an interpretative decision.

In line with our hypothesis, our findings seem to confirm that CWS have a lower efficiency of endogenously generated (from within themselves) response inhibition.

6.4.3 Possible implication for the role of the basal ganglia in developmental stuttering

Current findings seem to be in line with previous imaging studies (Brown, Ingham, Ingham, Laird, Fox, 2005; Ludlow & Loucks, 2003; Watkins, Smith, Davis, & Howell, 2008; Wu et al., 1995) and seem to provide additional validation for the hypotheses linking the fronto-basal ganglia network to the etiology of developmental stuttering (Alm, 2004b; Smits-Bandstra & De Nil, 2007), be it only indirectly.

The prefrontal cortex has been viewed as playing an important role in the ability to inhibit stimulus-evoked responses (Miller & Cohen, 2001) and neuroimaging studies have unveiled an integrated network of cortical and subcortical regions crucial for cancelling responses (Chambers, Garavan, & Bellgrove, 2009; Chikazoe, 2009). This fronto-basal ganglia network interconnects the inferior frontal cortex (IFC) and pre-supplementary motor area (pre-SMA) through the basal ganglia structures and the thalamus to the cortex (Aron, Behrens, Smith, Frank, & Poldrack, 2007; Aron & Poldrack, 2006; Mink, 1996, 2003). There is evidence (e.g., Ballanger et al., 2009; Jahfari et al., 2011; Mink, 1996, 2003; Nambu, 2004) for the existence of 3 different loops, namely a direct pathway (via the striatum and internal globus pallidus), an indirect pathway (via the striatum, external globus pallidus, subthalamic nucleus, and internal globus pallidus) and a hyperdirect pathway, which does not pass through the striatum (directly via the subthalamic nucleus, and internal globus pallidus). The hyperdirect pathway is responsible for suppressing all motor programs before response execution. This initial ‘reset’ signal is followed by releasing the selected motor response via the direct pathway. Finally, responses are terminated again via the slower indirect pathway. On stop trials in a stop signal paradigm, it is believed that the faster hyperdirect pathway is reactivated to inhibit responses (Boehler et al., 2010; Chambers et al., 2009; Jahfari et al., 2011).

Imaging studies in people who stutter have provided evidence for atypical activations in cortical and/or subcortical structures of this circuit, such as overactivity in the midbrain at the level of the basal ganglia nuclei (e.g., Brown, Ingham, Ingham, Laird, Fox, 2005; Ludlow & Loucks, 2003; Watkins, Smith, Davis, & Howell, 2008). Studies in stroke patients revealed that perseveration errors, both continuous (abnormal prolongation of a specific activity) and recurrent perseverations (repetition of a previous response to a subsequent stimulus), were strongly associated with lesions involving the nucleus caudatus (Nys, van Zandvoort, van der Worp, Kapelle, & de Haan, 2006). A more recent study on neurogenic stuttering by Theys, De Nil, Thijs, van Wieringen, and Sunaert (2012) revealed that one or several lesions throughout the fronto-basal ganglia circuit could result in neurogenic stuttering as opposed to a dysfunction limited to one specific brain area. Also in other motor movement-related disorders sometimes being linked to stuttering, such as Parkinson’s disease (Seiss & Praamstra, 2004; Shahed & Jankovic, 2001), Tourette’s syndrome

(Li, Chang, Hsu, Wang, & Ko, 2006; Serrien, Orth, Evans, Lees, & Brown, 2005), and tic disorders (Mulligan, Anderson, Jones, Williams, & Donaldson, 2003), the implication of the basal ganglia has been extensively studied. Moreover, the basal ganglia also play a key role in several other functions where differences have emerged between stuttering and nonstuttering populations, such as set-shifting, attention, and movement initiation (Brown, Schneider, & Lidsky, 1997; Hauber, 1998)

Although it would be too speculative, based on current findings, to suggest possible dysfunctional loci within this fronto-basal ganglia network in stuttering, recent studies showed that deep brain stimulation (DBS) of the subthalamic nucleus or the ventral intermediate nucleus of the thalamus affected movement inhibition in patients with Parkinson's disease (Ray et al., 2009; Van den Wildenberg et al., 2006) but could also lead to increased impulsivity (Frank, Samanta, Moustafa, Sherman, 2007). Furthermore, Hershey et al. (2010) showed that DBS of both ventral and dorsal subthalamic nuclei increased false alarms and decreased hits in a response inhibition task, similar to our sustained attention task results. Based on the effects of DBS in Parkinson's disease and the hypothesized etiological role of these structures in stuttering, one would also expect an impact on stuttering symptomatology linked with DBS in people who stutter. This seems to be confirmed by a series of recent studies showing that DBS of the subthalamic nucleus in Parkinson's disease (Burghaus et al., 2006; Toft & Dietrichs, 2011; Walker et al., 2009) or of the globus pallidus in dystonia (Kupsch et al., 2006; Nebel, Reese, Deuschl, Mehdorn, & Volkmann, 2009) can exacerbate the symptomatology of neurogenic stuttering. Based on these findings in patients with Parkinson's disease and neurogenic stuttering, it seems reasonable to hypothesize about a similar role in developmental stuttering.

6.4.4 Additional considerations and suggestions for future research

Some caveats of the study must be mentioned. First of all, the findings discussed here represent group means and must therefore also be interpreted as such. Although CWS, as a group, were less efficient in endogenously inhibiting motor responses, individual variations are possible and the findings may therefore not necessarily apply to each individual CWS. Secondly, endogenous and exogenous response inhibition were operationalized by using only two specifically chosen computer paradigms; in order to reach more definite conclusions on these different kinds of response inhibition in CWS, additional studies employing other paradigms would be needed. This might also yield additional insights in the impact of various stimulus characteristics on task performances. Future studies might also combine different ways of evaluating response inhibition, such as questionnaires, computer paradigms, and laboratory testing. Since the majority of our participants had a stuttering severity score in the range mild-to-moderate and only one participant scored in the severe-to-very severe range, results might be sample specific and our dataset did not allow for evaluating a correlation with stuttering severity, although our previous findings do not point towards a direct correlation with stuttering severity (Eggers et al., 2010).

6.5 Conclusion

In previous studies (Eggers et al., 2010, Embrechts et al., 2001; Karrass et al., 2006) differences were reported between CWS and CWNS on response inhibition or on inhibition-related concepts, such as emotional regulation. In the current study two paradigms, a stop signal task (Verbruggen et al., 2008) and a sustained attention task (de Sonneville, 2009) were used to evaluate two distinct forms of response inhibition, namely exogenously triggered response inhibition and endogenously generated response inhibition respectively. CWS, compared to CWNS, seemed to perform worse in tasks where response control had to be generated endogenously and performed comparable to CWNS in tasks where response control was externally triggered. These findings were taken to suggest a possible role of the fronto-basal ganglia circuit in the etiology of developmental stuttering.

CHAPTER 7

General conclusions

7.1 General discussion of research findings

This research project aimed at gaining additional insight in the relation between temperament and developmental stuttering, including temperamental components of attention and inhibitory control. In order to achieve this goal, several consecutive studies, both questionnaire- and cognitive paradigm-based, in which stuttering children were compared to nonstuttering children were stepwise executed. Chapters 2 and 3 described a questionnaire-based (CBQ) study investigating differences in temperament characteristics and its underlying temperament structure, as conceptualized by Rothbart (Rothbart et al., 2001). Differences in efficiency of attentional networks, as conceptualized by Posner and Peterson (1990), were evaluated in Chapter 4. Chapter 5 and 6 focused on possible differences in inhibitory control and reported the findings of a study using a Go/NoGo task, and a study employing a stop signal and sustained attention task to specifically evaluate differences in exogenously triggered versus endogenously generated response inhibition.

In this concluding chapter the results of our studies will be summarized and discussed with regard to the three main research objectives, namely the differences in reactivity and self-regulation as measured by a parental temperament questionnaire (7.1.1), the evaluation of the underlying attentional component of self-regulation by testing the efficiency of attentional processes through the use of a cognitive computer task (7.1.2), and finally the evaluation of the inhibition efficiency in both populations also measured by a cognitive computer task (7.1.3). We will shortly discuss two models, the vulnerability and disability model, which can be used to interpret our findings (7.1.4), speculate on how temperament characteristics might impact early childhood stuttering onset and development (7.1.5), and provide some additional considerations (7.1.6). We will conclude this chapter by making some future research directions with regard to multimodal measurements of temperament (7.2.1), prospective longitudinal research (7.2.2), and treatment related issues (7.2.3).

7.1.1 CWS are higher in negative reactivity and lower in self-regulation, as measured by a parental temperament questionnaire

Previously published studies in CWS could be interpreted as pointing towards an increase in positive (Embrechts et al., 2000; Fowlie & Cooper, 1978; Glasner, 1949; Guitar, 2003; Oyler (1988) in Zebrowski & Conture; Schwenk et al., 2007; Wakaba, 1998) or negative reactivity (Fowlie & Cooper, 1978; Guitar, 2003; Karass, et al. 2006) and a decrease in self-regulation (Anderson et al., 2003; Embrechts et al., 2000; Karass, et al. 2006; Schwenk et al., 2007). Nevertheless results from these studies were far from unequivocal and were confounded by differences in age, gender, and test instruments used. Moreover, none of these studies used a questionnaire specifically conceptualized to measure reactivity and self-regulation in a highly integrated manner nor were these results validated by evaluating if the structure of the underlying construct being measured was similar for all groups. Therefore, a questionnaire-based study on a large population was required to compare temperamental characteristics between CWS and CWNS. Prior to this study it was necessary to evaluate if the underlying temperamental structure was identical in all participant groups because otherwise emerging scale differences might be due to differences in underlying temperamental construct (Byrne et al., 1993).

Based on a large sample of CWS, CWNS, and children with vocal nodules, the underlying factor structure of the CBQ-D was determined by performing a principle axis factor analysis and a subsequent congruence analysis (Chapter 2). A similar three-factor structure, labeled as Extraversion/Surgency (positive reactivity), Negative Affect (negative reactivity), and Effortful Control (self-regulation), emerged for all participant groups. The highest congruence was found between CWS and CWNS and for the factors of extraversion/Surgency and Negative Affect. These findings formed the basis for further between-group comparison of the individual temperament scales and factors.

An age- and gender-matched subgroup was selected (Chapter 3) from the larger participant group from previous study. Factor comparison revealed that CWS scored higher on Negative Affect and lower on Effortful Control. When the individual scales were compared, CWS scored higher on the scales of Anger/Frustration, Approach, and Motor activation, and lower on the scales of Inhibitory Control and Attentional Shifting. No correlations were found with therapy duration or stuttering severity.

In line with Lonigan's (2004; Lonigan & Vasey, 2009) and Bijttebier and Roeyers' (2009) findings that higher scores on Negative Affect and lower scores on Effortful control could play a role in the onset, development, and maintenance of certain behavioral disorders, these results were taken to suggest that this temperamental constellation might play a role in the onset and/or exacerbation of developmental stuttering. The latter also complies with Conture et al.'s (2006) Communication-Emotional model of stuttering and with recent multifactorial models in stuttering (Bloodstein & Ratner, 2008). Moreover, in combination with the individual scale differences, we speculated these specific temperamental characteristics could play a role as a moderator in stress-related situations and in conditioning processes. For example, when confronted with abnormal disfluencies CWS might react stronger (e.g., higher in Anger/Frustration) to these interruptions combined with a feeling of loss of control emerging faster because of the lack of an adequate self-regulation. Alternatively, (novel) situations might be evaluated more easily as stressful due to difficulties in shifting the focus of their attention away from stressful situations. In conclusion we can say that these findings confirmed our original stated hypothesis.

7.1.2 Attentional processes, as measured by cognitive computer tasks, are less efficient in CWS

Our second objective was to evaluate the underlying attentional component of self-regulation by testing the efficiency of the attentional processes in CWS. Based on the available literature suggesting less efficient attentional skills in CWS (Alm & Risberg, 2007; Anderson et al., 2003; Bosshardt, 1999, 2002, 2006; Bosshardt et al., 2002; Embrechts et al., 2000; Felsenfeld et al., 2010; Karass et al., 2006; Monfrais-Pfauwadel & Lacombe, 2002; Schwenk et al., 2007; Smits-Bandstra & De Nil, 2007; Vasic & Wijnen, 2005) and our own previous findings of lower scores on Attentional Shifting (Eggers et al., 2010), we expected attentional processes to be less efficient in CWS.

We administered the Attention Network Test to 41 4-to-9-year old CWS and 41 age- and gender-matched controls (Chapter 4). This test is a combination of a cued RT task and a flanker task, measuring the efficiency of 3 distinct attentional networks, i.e. alerting, orienting, and executive attention. Findings showed that CWS scored significantly lower on the orienting network, meaning they were less able to make use of visual attentional cues, and also a trend towards a lower score on the executive attention network was apparent (p = .066). Since attentional orienting has been found to play a significant role in regulating the emotional reactivity and controlling distress by orienting the attention away from stress-evoking situations (Rueda et al., 2004; Harman et al., 1997), we interpreted these findings as CWS perhaps being less capable of disengaging their attention from the emotion-triggering stimuli, resulting in more frequent emotional reactions and a higher arousal linked to certain stimuli, of which it is known this can influence stuttering symptoms (e.g., Alm, 2004a; Ezrati-Vinacour & Levin, 2004; Menzies, Onslow, & Packman, 1999). Because of the high degree of overlap between spatial and temporal orienting networks (e.g., Nobre, 2001), and the importance of temporal orienting for motor preparation, motor coordination and motor sequencing/timing (Coull & Nobre, 1998), these findings were also linked to reports of disturbed timing of activation in speech-relevant brain areas in adults who stutter (Sommer, Koch, Paulus, Weiller, & Büchel, 2002) and temporal programming deficits (e.g., Kent, 1984).

A recently published study by Johnson, Conture, and Walden (2012) using a traditional and an affect cued RT time task in CWS seems to contradict our finding of differences in the orienting network, since no between-group differences were found for the traditional cued RT task. In our study only valid cues were employed while in the Johnson et al. study both valid and invalid cues were used. In order to give a correct answer upon the presenting of an invalid cue, attentional resources have to be disengaged from the cued location, shifted from the cued to another location, and finally engaged to this new location. Since several findings point towards a decreased efficiency in attentional shifting (Eggers et al., 2010; Embrechts et al., 2000) one would expect also to find differences in this task. The nonsignificant findings in their study might therefore be attributed to a too small sample size for these kind of studies or to other methodological issues, such as stimuli used, as stated by the authors themselves: "*if the present study included a sample similar to that used in the Eggers et al. (2012a) study findings may have been more consisten*t" (p.274) or "*it may be the case that between-group differences in RT were not observed because the task was more concrete, overly easy and/or appealing…; it could be the case that the preparatory signal implied an accurate response for each response instead of merely a preparatory signal.*" (p.275).

The trend towards a lower efficiency of the executive attention network was further explored by neurocognitive experimental tasks employing inhibition-related paradigms (see 7.1.3) and by a very recently presented study, not included in the previous chapters, specifically aimed at measuring auditory attentional set-shifting (Jansson-Verkasalo, Eggers, Aro, De Nil, & Van den Bergh, 2012). The auditory set-shifting task of the Amsterdam Neuropsychological Tasks (de Sonneville, 2009) consisted of three experimental blocks. During the first block, children were presented with one or two low tones and had to press the response button respectively once and twice. During the second block, consisting of incompatible mapping, high tones were used and participants had to press once when two tones were presented and twice when one tone was presented. During the final block of the task, a random mix of compatible and incompatible stimuli was presented, forcing the child to switch his/her attentional set continuously. This task is a prototypical example of a task measuring the executive attention network. Preliminary results on a group of 10 6-to-10-year old CWS and 18 6-to-10-year old CWNS revealed no significant differences in experimental block 1 and 2 but in the final block CWS made significantly more errors compared to the control group, $F(1, 26) = 4.47$, $p < .05$. Concluding we can say that these overall findings were in line with our original stated hypothesis of a lower efficiency of the orienting and executive attention network.

7.1.3 Inhibition, as measured by cognitive computer tasks, is less efficient in CWS

Our third and final objective was to examine IC in CWS by direct measurement using neurocognitive computer paradigms. Based on both findings by other groups (Embrechts et al., 2000) as well as our own (Eggers et al., 2010) we assumed CWS to be less efficient in IC, although we need to add that not all questionnaire-based findings were unequivocal (Anderson & Wagovich, 2010). In a first study (Chapter 5) a Go/NoGo task was administered to a group of 30 4-to-10-year old CWS and a matched control group. A subsequent study (Chapter 6) was specifically aimed at evaluating differences in exogenously and endogenously generated inhibition because literature findings suggested possible differences in impact from exogenous versus endogenous cueing in stuttering (Alm, 2004b; Saltuklaroglu et al., 2003, 2004). Hereto we administered to a group of 18 7-to-10-year old CWS and a matched control group a stop signal task and a sustained attention task, tasks assumed to respectively test exogenously and endogenously generated inhibition.

CWS were found to be lower in IC as measured by the Go/NoGo-paradigm because they were less able to inhibit their prepotent response tendencies during non-targets and also made premature responses. These findings suggested that CWS exhibited a less controlled response style and were less able to adapt this response style after experiencing response errors. Lower IC results in difficulties in successfully regulating ones emotions (e.g., Carlson & Wang, 2007; Kochanska et al., 1996), of which it is known to impact stuttering symptomatology (e.g., Alm, 2004a; Ezrati-Vinacour & Levin, 2004; Menzies, Onslow, & Packman, 1999). Moreover, reduced IC might also have an impact on error-detection and error-processing during the linguistic stages of speech-language planning and production (Engelhardt, Ferreira, & Nigg, 2009; Meyer, Wheeldon, & Krott, 2007) and might thus also be linked to psycholinguistic models of stuttering etiology (e.g., Bernstein Ratner & Wijnen, 2006; Postma & Kolk, 1993) and to the Communication-Emotional model of stuttering (Conture et al., 2006).

CWS performed comparable to CWNS in the stop signal task, a task where inhibition was triggered externally; on the sustained attention task, a task where inhibition is assumed to be generated endogenously, CWS performed worse. An important difference between the stop signal task and both the sustained attention and the Go/NoGo task was that in the latter two tasks no external auditory signal was provided indicating inhibition was required but the decision to inhibit from responding had to based on a decision-making component, which was absent in the stop signal task. Based on these findings we concluded that endogenously generated inhibition was impaired in CWS while exogenously triggered inhibition was comparable for both groups.

Since these inhibition-related tasks clearly have been linked to the underlying anatomical correlates of the fronto-basal ganglia circuits, (Aron, Durston, et al., 2007; Boehler et al., 2010; Chambers et al., 2009; Congdon et al., 2010), a network frequently implicated in emerging conceptualizations about processes underlying developmental stuttering (e.g., Alm, 2004b; Smits-Bandstra & De Nil, 2007), our findings could also be interpreted as corroborative data for these models.

Finally it also needs to be mentioned that an alternative, although in our view less likely, interpretation of our findings might be possible. With regard to response inhibition, Schachar et al. (2007) made the distinction between action restraint and action cancellation, with restraint referring to the inhibition of a motor response before the actual response has been started, and cancellation referring to the inhibition of a motor response during its execution. In this view, the stop signal task we employed could be labeled as an action cancellation task and the sustained attention task and Go/NoGo task as action restraint tasks. Consequently, our findings could be interpreted as CWS having no difficulties with action cancellation but only with action restraint. Eagle et al. (2008) hypothesized that action restraint tasks would measure whether a person can initiate and maintain response inhibition whereas action cancellation tasks would evaluate the actual time required for the response inhibition. If we would apply this to our findings, it would seem odd that CWS would have no difficulties with inhibiting responses after the execution of the motor response has started while they would have difficulties with inhibiting the response at the earlier stages of motor planning and execution (restraint) unless, it was due to difficulties in the decision making process of the 'yes'- and 'no'-response in which case it overlaps with our earlier given rationale for the lower efficiency of endogenous response inhibition.

In conclusion we can state that our findings corroborate our previous CBQ-based results and our original hypothesis stating that CWS would be less efficient in IC compared to CWNS.

7.1.4 Vulnerability or disability hypothesis

While we have discussed temperamental differences between CWS and CWNS and have suggested possible pathways by which temperament might exude its influence on the onset and development of stuttering (e.g., via a role in generating emotional reactions, in stress-related situations, in conditioning processes, in error-detection and error-processing), it is less evident to make any strong statements about the exact nature of the underlying relation, be it causal, correlational or consequential in nature. Roy & Bless (2000) already highlighted a similar problem with regard to the exact nature of interaction between temperament and voice disorders, stating "*methodological inadequacies make it very difficult to generalize the results and to evaluate the specific nature of the voice disorder–personality relationship*" (p. 738).

Roy and Bless proposed two possible models, namely the disability model and the vulnerability model. The disability model (also referred to as the scar model) assumes that certain temperamental characteristics are the consequence of certain disorders, in other words that the repeated experience with the disorder results in temperament changes. The vulnerability model (also referred to as the predisposition model) proclaims temperamental characteristics cause the development of the disorders or (indirectly) modify the development or the expression of the disorder. Based on previous and our current findings, the suggestion of a disability model with regard to temperament and stuttering seems unlikely because several of the studies referred to in previous chapters, including our own, have focused specifically on young CWS and one would not expect relatively stable temperament traits to change over the course of a couple of months as a result of exposure to stuttering. Therefore the vulnerability model seems more plausible for the temperament-stuttering interaction, although additional research in this domain is needed to make a more valid statement. This is a comparable model to the one suggested by Bijttebier and Roeyers (2009) and Lonigan (2004; Lonigan & Vasey, 2009) for the influence of high negative reactivity and low self-regulation scores on the development of behavioral disorders.

In this context, Strelau (1998) uses the term 'temperament risk factor', which he defined as "*any temperament trait, or configuration of traits, that in interaction with other factors acting excessively, persistently, or recurrently (e.g., physical and social environment, educational treatment, situations, the individual's characteristics) increases the risk of developing behavior disorders*" (p. 376; see also Strelau, 1995). This description maps well upon the multifactorial views (see 1.2.4) considering developmental stuttering to be the result of both child-specific and environmental factors, possibly different in each person, and on their own not sufficient to start stuttering.

7.1.5 Temperament characteristics and early childhood stuttering onset and development

Combining the findings of all in the current research project described studies and in line with the previously depicted vulnerability model, we speculate that temperament may affect developmental stuttering in a number of ways. Recent multifactorial models (e.g., Conture et al., 2006; De Nil, 1999; Smith & Kelly, 1999) have considered a combination of genetic, neurobiological, behavioral, emotional, and environmental components as predisposing, precipitating and persisting factors in the etiology of stuttering. Our findings have provided a solid base to speculate that temperament, or certain temperamental components, can be linked to all three of these factors.

Stuttering usually starts between 2-to-5 years of age with a gradual or more sudden onset. This onset of stuttering has been attributed to a combination of genetic and environmental factors (e.g., Brutten & Shoemaker, 1967; Conture et al., 2006; Howell, 2011). Besides the fact that temperament might be one of several inherited traits (e.g., Subramanian & Yairi, 2006) that predispose some children to start stuttering, it might also act as a precipitating factor, triggering the occurrence of moments of stuttering in children with a genetic predisposition for stuttering. Furthermore, some of our findings in the self-regulatory temperamental components (attentional orienting & response inhibition) seem to point directly to a possible underlying causal speech motor mechanism, namely dysfunctional cortical-basal ganglia circuits (see Chapter 5 & 6), while other findings can be linked more indirectly to existing theoretical conceptualizations about stuttering etiology, such as psycholinguistic models (see Chapter 3 & 4). Therefore, we assume that moments of stuttering might emerge as a result of aberrant monitoring and/or error detection during linguistic programming or as a result of a failure to execute complex speech motor movements due to problems with motor preparation, motor coordination and/or motor sequencing/timing.

In those children that have started to stutter, temperamental characteristics most likely also play a role in the further development of the disorder. First, CWS, as a group, were found to be more easily frustrated when ongoing tasks were interrupted or when goals were being blocked. In combination with higher approach tendencies, a lowered ability to suppress inappropriate response tendencies and a lowered ability to shift one's attention away from stress-evoking situations, these temperamental characteristics result in an increase in the amount of encountered stress-related situations, of which it is known they exacerbate stuttering symptomatology (e.g., Ezrati-Vinacour & Levin, 2004); possibly these result also in an increased emotional reactivity related with their own disfluencies and moments of stuttering. Second, an increased experience with moments of stuttering combined with increased emotional reactions automatically leads to a faster and stronger negative conditioning of both moments of stuttering as well as stuttering evoking situations (Ledoux, 1994), resulting in more secondary behaviors (Brutten & Shoemaker, 1967). Third, the higher motor reactivity seen in CWS, which is often reflected in increased muscular tension in stressful situations (Kagan, 1998), in turn, might also act as a precipitator for speech disruptions (e.g., Bloodstein, 1995; Guitar, 1998). Finally, in some CWS the number of iterations and the duration of prolongations or blocks might gradually increase over time due to their lowered ability to alter their response style after experiencing these moments of stuttering, similar to what was found in some of the experimental paradigms. To summarize, when CWS grow older, temperament might contribute to more negatively valenced situations and moments of stuttering, resulting in more stuttering, more secondary behaviors, and an increase in overall stuttering severity.

A considerable percentage of the children that start to stutter, recovers spontaneously within the first 2-to-3 years after stuttering onset. We assume that temperamental characteristics, more in specific high negative reactivity and low effortful control might also be one of the factors contributing to chronicity, as already suggested by

Zebrowski and Conture (1998). Not just because of the previously discussed temperament-conditioning pathway, but also because of the fact that this temperamental constellation was found to contribute to the maintenance of other developmental disorders (Lonigan et al., 2004; Lonigan & Vasey, 2009; Muris & Ollendick, 2005)

Finally, we also consider temperament to be a factor possibly related to treatment outcome in stuttering since temperament characteristics have been found to influence children's reactions to specific types of treatment (Mash, 2006) or to moderate or mediate treatment outcome in other disorders, such as anxiety disorders or ADHD (Purper-Ouakil, et al., 2010; Rapee & Jacobs, 2002). Directly addressing temperamental components, such as increased negative reactivity, reduced self-regulation or lowered IC, might therefore impact stuttering symptomatology. This could be done through both indirect parent-counseling techniques as well as direct treatment strategies focusing on identification and coping strategies for difficult situations (e.g., Kristal, 2005; Wodka et al, 2007). Examples of these have been described in cognitive-based and family system-based stuttering treatment programs (e.g., Kelman & Nicholas, 2008; Shapiro, 1999). The finding that some CWS were less able to change their response style after experiencing errors, might also validate treatment strategies such as increased monitoring of one's own speech, altering speech rate, and providing feedback (e.g., Guitar & McCauley, 2010). With regard to the latter, the frequently described effects of parental - and thus exogenous - contingencies to the child's fluent or disfluent utterances (e.g., Jones et al., 2005) might be confirmed by our finding that CWS were equally efficient in exogenously triggered response inhibition but less efficient in endogenously generated inhibition.

7.1.6 Additional considerations

A possible limitation of the studies presented in the previous chapters that needs to be considered is the fact that many of the stuttering participants were in treatment at the time of the measurements. The duration of treatment for the CWS included in the different studies (Chapter 2 – Chapter 6) ranged between 0-26 months (mean = 9.5 months; SD = 7.2). One might argue that ideally no participant would have received treatment prior to the testing but this would have resulted in a considerably comprised participant group and consequently a much lower power to detect possible between-group differences. Therefore, at the beginning of our research project, during one of our first studies (Chapter 3) we investigated the correlation between treatment duration and both the composite temperament factors and the individual temperament scales. No significant correlations were found for any of these characteristics. At that point we tentatively concluded temperament characteristics to be robust against therapy interventions lasting a few weeks to a few months or that treatment approaches would need to address temperament directly in order to result in significant changes, which was most likely not the case since treatment approaches used were primarily an eclectic combination of both direct and indirect approaches combining both stuttering modification and fluency shaping based principles. However, treatment or the type of treatment was not directly controlled in this study.

Furthermore, in our opinion it is important to emphasize that high or low scores on the different dependent variables used in this study, i.e., temperamental scale or factor scores, attentional network efficiencies, IC-related variables, are not a representation of clinical disorders but are merely a reflection of the distribution of scores on a bipolar continuum in a normal population. For instance, a lower group mean score on inhibitory control is not indicative of any disorder but only reflects a lower efficiency in suppressing inappropriate approach responses.

All of the findings discussed in previous chapters represent group means and must therefore also be interpreted as such. For example, although CWS, as a group, were less efficient in endogenously inhibiting motor responses, individual variations are possible and the findings may therefore not necessarily apply to each individual CWS.

Finally, we would like to acknowledge that in the statistical analyses of the different studies, also other statistical techniques could have been used (e.g., confirmatory factor analyses in Chapter 2 or testing the interactions between cue type and flanker conditions jointly in one single analysis in Chapter 4).

7.2 Future research directions

7.2.1 Multimethod approach

Although our studies of attentional networks and IC were originally motivated by previous findings of parental questionnaire-based differences on IC between CWS and CWNS (Embrechts et al., 2000; Eggers et al., 2010; Karrass et al., 2006), these studies did not include questionnaire-based data. Future studies pursuing this line of research might consider combining multiple independent measures of temperament characteristics, such as cognitive computer tasks, parental questionnaires, and psychophysiological measurements within the same study. Although this would result in more elaborate testing sessions and an increased working load for participants, it would allow for comparison of IC across different measures.

As previously discussed in Chapter 1, every method of measuring temperament has some kind of limitations (Rothbart & Bates, 1998; Strelau, 1998): while temperament questionnaires might be biased by subjective factors, laboratory measurements are limited in the amount of situations they can assess and might be prone to some kind of artificiality, and behavioral observation in natural settings is very time-consuming and does not allow for full control of behaviors. By combining several of these methods, the individual limitations of each method might be overcome. This is in line with

Kagan's approach that combined structured observations, parental interviews, and physiological measures as the basic strategy for assessing temperament in children (Kagan, 1994; Kagan, Reznick, & Gibbons, 1989). Nevertheless, one of the problems that might emerge is a lack of consistency between some of the measures. While, for instance, positive correlations have been found between e.g. the CBQ-measure of inhibitory control and the performance on a stroop-like task (Gerardi, Rothbart, Posner, Kepler, 1996), Strelau (1998) states that "*comparisons between different methods show that only rarely does agreement reach a correlation of .50*" (p.320).

Based on the findings of our questionnaire-based studies, we further explored some of the self-regulatory components of temperament in CWS by using cognitive computer paradigms. Current project did not further examine the reactivity-related findings in CWS by using other than questionnaire-based methods. Therefore, future studies would also need to focus on our emotional reactivity-related findings. Different methodologies might be used, ranging from behavioral observation in laboratory settings (e.g., Ntourou, 2012) to electrophysiological studies (e.g., Arnold, 2012; Arnold et al., 2011).

7.2.2 Temperament interactions

Our different studies in the previous chapters have primarily focused on the possible associations between temperament and stuttering. Temperament, although its expressions are relatively stable over time, is primarily reactivity-driven at a young age and becomes more governed by self-regulatory processes when children grow older. This change is both influenced by maturation as well as by learning experiences. With regard to the latter it is widely accepted (e.g., Rothbart, 2011) that the influential pattern is bidirectional, in other words children's temperament has an influence on the environment and is being influenced by environmental factors (see 1.4.4.1 for more detail); therefore, future research would also need to explore these bidirectional interaction patterns in developmental stuttering. In this context also the concept 'goodness or poorness of fit' (see 1.4.1.2), i.e., the compatibility between the capacities and temperamental constellation of the child and environmental demands and expectations, especially from parents, including child-rearing styles (Bates, 2001; Halverson & Deal, 2001), might be also a potential avenue for future research. With regard to the studies described here, this might mean that in older CWS the impact of environmental factors is higher compared to preschoolers since they have been more exposed to these environmental influences (e.g., a dominant parental style might have had a stronger, more long-lasting impact in older children). Another important environmental factor to be considered is treatment. We acknowledge that we were not able to control all of these factors and that these should be studied more in depth in the future.

Besides environmental interactions, different temperamental components within a child's temperamental constellation might interact, sometimes referred to as temperament-by-temperament interactions (see e.g., Strelau, 1998). For instance, a child with low adaptability to novelty, who is also relatively non-distractible may find it particularly difficult to "change the topic" when confronted by novelty. Hence, this child might maintain initial non-adaptibility/reactivity for longer than is beneficial. Although we have briefly discussed the importance of this type of interaction at the higher hierarchical level of superfactors (see Chapter 3), suggesting that high levels of positive and/or negative reactivity in combination with low levels of Effortful Control might play a role in the onset, development and/or maintenance of developmental stuttering, further study of these kinds of interactional patterns at the lower hierarchical level, i.e. the different temperamental components, and the role this could play in diverting the child's attentional, cognitive and linguistic resources away from ongoing speech-language planning, might be interesting.

Finally, our research has shown that CWS differ from CWNS in temperament characteristics. While temperament characteristics are relatively stable, stuttering varies considerably within CWS. Therefore some other relatively changeable variable must mediate/moderate the temperament-stuttering association. The above-discussed temperament-environment or temperament-temperament interactions might be candidate areas to explore in future research.

7.2.3 Prospective longitudinal research

While current research project was aimed at studying temperamental characteristics in young CWS, it remains to be seen if similar patterns emerge in older persons who stutter. This is an interesting line of research because it might yield additional insights in the role of these characteristics in stuttering development and spontaneous recovery. In a first attempt to address this issue we administered the same Go/NoGo paradigm as used in Chapter 5 to a group of 24 adults who stuttered and 24 age-and gender-matched controls (Eggers, 2012). The results of this study (not discussed in previous chapters) showed, similar to our findings in CWS, that adults who stuttered also had a significantly higher amount of false alarms (F (1, 45) = 4.89, $p < .05$); in other words, adults who stutter, as a group, also seem to have a less efficient IC when compared to matched controls.

Whilst employing similar paradigms in an older population is one possibility for gaining more insight in factors related to chronicity versus spontaneous recovery, perhaps a better way to approach this question might be the use of a prospective longitudinal research design versus a cross sectional design as was currently used in our study. If such a prospective cohort study would start with children at a considerably young age, this could also yield more insight in the exact nature of the underlying relation (causal, correlational, consequential), as discussed in 7.1.4. At the time we started this research project, the understanding of the possible role of temperament in developmental stuttering and of the temperamental differences between CWS and CWNS was too limited to start a well-founded longitudinal study; hence our rationale for opting for a cross sectional design. Based on the findings of the current research project, a new study might be set up in which the currently found differences are specifically targeted in a prospective longitudinal design. Alternatively, also some of our current participants might be followed up to evaluate a possible influence of e.g. temperamental characteristics such as efficiency of attentional orienting and IC on spontaneous recovery.

Future studies might also evaluate whether children high in negative affect and low in effortful control have a higher likelihood of developing stuttering symptomatology or, if already stuttering, have a higher chance on chronic stuttering; in other words, evaluating if high effortful control also acts as a protective factor for the further development of stuttering, such as was found to be the case in other disorders (e.g., Bijttebier & Roeyers, 2009).

In one of our initial studies (Chapter 3) we did not find any correlation with stuttering severity but at that point no detailed information was available for all components making up stuttering severity. Therefore, future studies might address more detailed correlational analyses between overt as well as covert components of stuttering severity. These studies have currently been set up as part of bachelor and master's theses at Lessius and KU Leuven.

While we already suggested that children lower in IC might benefit from parent counseling and strategies aimed at acquiring more self-regulatory behaviors, a prospective, possibly randomized control trial would yield more insight in this hypothesis. Such a longitudinal design would also best include a subgrouping, as suggested by Seery et al. (2007) and Subramanian and Yairi (2005) to avoid a possible confound of motor, linguistic, cognitive, and personality-related factors.

7.3 General conclusions

In the current research project, temperamental characteristics of CWS were assessed by a temperament questionnaire as well as through several computerized neurocognitive paradigms. Results showed that CWS were higher in negative reactivity and lower in self-regulation. Self-regulation was further examined by evaluating its underlying attentional processes and one of its key components, inhibitory control. CWS were found to be less efficient in both attentional orienting and inhibitory control, primarily endogenously generated inhibition. These findings have contributed to our knowledge base with regard to the relation between temperament and developmental stuttering and have laid the foundations for future longitudinal studies. The latter will be necessary to unveil the way of interaction between temperament and developmental stuttering.

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Curriculum vitae

Personal

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Born:	May 2nd 1972, Bonheiden
E-mail:	kurt.eggers@lessius.eu
Civil Status:	Married to Els Van Loock
Children:	Floris (°April 10th 1999) and Marthe (°April 10th 2001)

Education

2007-present	PhD program in Biomedical Sciences, KU Leuven, Belgium
2004	Lidcombe-stuttering therapist, U of Nijmegen, The Netherlands
2003	Intensive workshop for stuttering specialists, Stuttering Foundation of America, U of Iowa, USA
2001-2003	CIOOS stuttering specialization training, U of Antwerp, Belgium
1998-1999	Post academic training voice disorders, VVL - U of Antwerp, Belgium
1995-1997	Master in Speech-Language Therapy & Audiology, KU Leuven, Belgium
1990-1995	Bachelor in Medicine, KU Leuven, Belgium

Coordinating experience

2010-present	Head of Speech-Language Therapy and Audiology Department, Group Health and Well-Being, Lessius U College
2007-2010	Research coordinator Speech-Language Therapy and Audiology Department, Group Health and Well-Being, Lessius U College
2006-present	Coordinator European Consortium on Fluency Disorders (Lessius U College, U of Leuven, Artevelde U College, KHBO Bruges, Trinity College Dublin, U of Malta, U of Gothenburg, RWTH-U Aachen, U of Oulu, U of Applied Sciences Utrecht, U of Turku, Escola Superior de Saude do Alcoitão, Michael Palin Centre London, Stammering Support Center Leeds)

Lecturing experience

2010-present	Visiting lecturer Department of Logopedics, U of Turku, Finland
2006-present	Visiting lecturer Department of Logopedics, U of Oulu, Finland

2001-present Head Lecturer Speech-Language Therapy and Audiology Department Lessius U College
2006-2008 Guest lecturer Department of Logopedics, Fontys U College Eindhoven, The Netherlands

Clinical experience

2000-2010 Private practice for stuttering therapy

Financed projects

2012-2014 Promoter of 'Upgrading Speech Language Therapy (SLT) in Suriname through quality improvement of the educational SLT- curriculum and development of SLT-health care services' (VLIR-UOS University Development Cooperation, South Initiatives, VLIR-grant)
2010-2013 Lead partner of work package 2 in 'Network for Tuning Standards and Quality of Education programs in Speech and Language Therapy across Europe' (Erasmus Multilateral Network 177075-LLP-1-2010-1-FR-ERASMUSENWA, EU-grant)
2006-2008 Coordinator of 'European Clinical Specialization Course on Fluency and Fluency Disorders' (Module development project under Erasmus Socrates Program 28095-IC-1-2005-1-BE-ERASMUS-MODUC-1, EU-grant)

Other research projects

2011-2016 Speech fluency and disfluency in Finnish children (promoter: Prof. E. Jansson-Verkasalo, U of Turku; submitted for research grant)
2010-2012 Cross cultural study on spontaneous recovery in adults (promoter Prof. K. Neumann, U of Frankfurt)

Scientific committees

Review committee 7th World Congress on Fluency Disorders, July 2012, Tours, France.
Review committee Oxford Disfluency Conference, July 2011, United Kingdom.
Review committee Convention of the International Association of Logopedics and Phoniatrics, August 2010, Athens, Greece.
Invited discussant at 'Stuttering: a clinical symposium', Australian Stuttering Research Centre, May 2010, Cavtat, Croatia.
Organization committee Biennial European Symposium on Fluency Disorders Antwerp, Belgium.
Fluency committee member of the International Association of Logopedics and Phoniatrics

Reviewed for

International Journal of Language and Communication Disorders

Journal of Communication Disorders
Journal of Fluency Disorders
Journal of Speech, Language, and Hearing Research
Logopedics, Phoniatrics, & Vocology
Stem-, Spraak- en Taalpathologie
SIG, scientific projects

List of publications

Publications in international journals

Eggers, K., De Nil, L., & Van den Bergh, B. (2009). Factorial Temperament Structure in Stuttering, Voice Disordered, and Normal Speaking Children. *Journal of Speech, Language, and Hearing Research*, 52, 1610-1622.

Eggers, K., De Nil, L., & Van den Bergh, B. (2010). Temperament dimensions in stuttering and typically developing children. *Journal of Fluency Disorders*, 35, 355-372.

Eggers, K., De Nil, L., & Van den Bergh, B. (2012). The efficiency of attentional networks in children who stutter. *Journal of Speech, Language, and Hearing Research,* 55, 946-959.

Eggers, K., De Nil, L. F., & van Den Bergh, B. R. H. (2012). Inhibitory control in childhood stuttering. Accepted for publication in *Journal of Fluency Disorders*.

Eggers, K., De Nil, L., & Van den Bergh, B. (2012). Exogenously and endogenously triggered response inhibition in developmental stuttering. *Manuscript submitted for publication*.

Eggers, K., & Leahy, M. (2011). The European Clinical Specialization on Fluency Disorders (ECSF). *Journal of Fluency Disorders*, 36, 296-301.

Jansson-Verkasalo, E., Eggers, K., Järvenpää, A., Suominen, K., Van den Bergh, B., & De Nil, L. (2012). Central auditory processing in children who stutter as indexed by the mismatch negativity. *Manuscript submitted for publication.*

Invited publications

Eggers, K. (2012). Responsinhibitie bij volwassen personen die stotteren versus niet stotteren *[Response inhibtion in adults who stutter versus adults who do not stutter]*. *Logopedie [Logopedics]*, July-August Issue, 76-83.

Eggers, K. & Ebben, H. (2006). Therapie bij schoolgaande stotterende kinderen *[Therapy in school-aged stuttering children]*. In: H.F.M. Peeters, et al. (Eds.). Handboek Stem-, Spraak- en Taalpathologie *[Handbook on Voice, Speech, and Language Pathology]*. Houten: Bohn Stafleu van Loghum.

Eggers, K., Heylen, L., & Mertens, F. (1997). Follow-up studie van de subjectieve status van de stem. Een studie bij 46 vrouwen minstens 1 jaar na behandeling *[Follow-up study of subjective vocal status 1 year after treatment]*. Logopedie *[Logopedics]*, 10, 34-38.

Jansson-Verkasalo, E., & Eggers, K. (2010). Änkytys *[Stuttering]*. In P. Korpilahti, O. Aaltonen, & M. Laine (Eds.) Kieli ja aivot: Kommunikaation perusteet, häiriöt ja kuntoutus. *[Language and brain: The basis, disturbances and rehabilitation of communication]*. Turku: Kognitiivisen neurotieteen tutkimuskeskus.

Contributions to conferences

Eggers, K. (2012, December). *Stuttering: Assessment and intervention from a cognitive-behavioral perspective.* Two-day workshop to be presented for the Special Interest Group Fluency Disorders, Thessaloniki.

Eggers, K. (2012, September). *Principles of early intervention in stuttering.* Lecture presented at the 2nd symposium of the Leeds Stammering Support Center, Leeds.

Leahy, M., Agius, J., Eggers, K., & Hylebos, C. (2012, July). *The European Clinical Specialization on Fluency Disorders (ECSF): Review of the first four years.* Lecture presented at the 7th Congress of the International Fluency Association, Tours.

Pertijs, M., Eggers, K., Leahy, M., & De Nil, L. F. (2012, March). *Working towards minimal standards for EU fluency specialists.* Poster presented at the 3rd European Symposium on Fluency Disorders, Antwerp.

Jansson-Verkasalo, E., Eggers, K., Aro, K., De Nil, L. F., & Van Den Bergh, B. R. H. (2012, March). *Auditory attention shifting in children who stutter.* Poster presented at the 3rd European Symposium on Fluency Disorders, Antwerp.

Eggers, K., De Nil, L., & Van den Bergh, B. (2012, March). *Endogenous and exogenous response control in children who stutter.* Poster presented at the 3rd European Symposium on Fluency Disorders, Antwerp.

Eggers, K. & Alewaters, H. (2012, March). *Inhibitory control in adults who stutter.* Poster presented at the 3rd European Symposium on Fluency Disorders, Antwerp.

Eggers, K. (2011, November). *Intervention strategies for a cognitive behavioral treatment of stuttering.* Workshop presented for the Special Interest Group Fluency Disorders of the Panhellenic Association of Logopedics, Athens.

Eggers, K. (2011, November). *The role of attention in developmental stuttering.* Keynote lecture presented at the annual symposium of the Panhellenic Association of Logopedics.

Eggers, K. (2011, October). *Responscontrole in kinderen die stotteren. [Response control in children who stutter].* Lecture presented at the 12th Symposium Logopedics & Audiology U Ghent.

Eggers, K. (2011, January). *Assessment, evaluation and diagnosis in developmental stuttering.* Workshop presented at the Escola Superior de Saude do Alcoitão, Portugal.

Eggers, K. (2011, January*). Attentional networks and inhibitory processes in developmental stuttering.* Lecture presented at the Escola Superior de Saude do Alcoitão, Portugal.

Eggers, K., De Nil, L., & Van den Bergh, B. (2010, December). *Endogene en exogene responscontrole in stotterende kinderen. [Endogenous and exogenous response control in children who stutter].* Poster presented at the annual convention of the Vlaamse Vereniging voor Logopedisten *[*Flemish SLT federation*]*.

Eggers, K., De Nil, L., & Van den Bergh, B. (2010, November). *Endogenous and exogenous response control in children who stutter.* Poster presented at the annual convention of the American Speech-Language-Hearing Association, Philadelphia.

Eggers, K., Leahy, M., De Nil, L., & Pertijs, M. (2010, November). *Working towards minimal standards for European fluency specialists.* Poster presented at the annual convention of the American Speech-Language-Hearing Association, Philadelphia.

Eggers, K. (2010, October). *Stuttering, temperament, and attention.* Seminar presented at the Kolloquium zu Störungen der Kommunikation und Kognition *[*Seminars on disorders of communication and cognition*]*, RWTH-Aachen University, Aachen.

Eggers, K. (2010, September*). Stotteren: theorie en therapie. Verleden, heden en toekomst [*Stuttering: theory and treatment: Past, present, and future*].* Lecture presented at the 80 year Jubilee symposium of Logopedics Utrecht U College, Hogeschool Utrecht, Utrecht.

Eggers, K. (2010, August). *What is normal dysfluency and why measure it: Belgium.* Seminar presented at the Convention of the International Association of Logopedics and Phoniatrics, Athens

Eggers, K., De Nil, L, & Van den Bergh, B. (2010, August). *Attention set shifting in children who stutter.* Poster presented at the Convention of the International Association of Logopedics and Phoniatrics, Athens.

Eggers, K. & Leahy, M. (2010, August). *Educating fluency specialists in Europe: an innovative program.* Poster presented at the Convention of the International Association of Logopedics and Phoniatrics, Athens.

Eggers, K. & Leahy, M. (2010, August). *Rationale for treatment decision-making in older children and adults.* Seminar presented at the Convention of the International Association of Logopedics and Phoniatrics, Athens.

Eggers, K. & Leahy, M. (2010, August). *Importance of individual differences in treatment and outcomes.* Seminar presented at the Convention of the International Association of Logopedics and Phoniatrics, Athens.

Bosshardt, H.G., Packman, M., Neumann, K., Eggers, K., Fibiger, S., Leahy, M., Andrade, C., Blomgren, M., Boucand, V., & Cook, F.M. (2010, August*). Treatment decision-making: Future directions.* Seminar presented at the Convention of the International Association of Logopedics and Phoniatrics, Athens.

Blomgren, M., Bosshardt, H.G., Eggers, K., Packman, N., Cook, F., Leahy, M., Fibiger, S., & Boucand, V. (2010, August). *IALP Stuttering Assessment Survey.* Seminar presented at the Convention of the International Association of Logopedics and Phoniatrics, Athens.

Bosshardt, H.G., Blomgren, M., Andrade, C., Eggers, K., Cook, F.M., Boucand, V., Fibiger, S., Leahy, M., Neumann, K., & Packman, M. (2010, August). *IALP Assessment frameworks: Future directions.* Seminar presented at the Convention of the International Association of Logopedics and Phoniatrics, Athens.

Eggers, K. (2010, April). *Educating fluency specialists in Europe.* Poster presented at the 2nd European Symposium on Fluency Disorders, Antwerp.

Eggers, K. (2010, April). *Inhibitory control in CWS.* Poster presented at the Second European Symposium on Fluency Disorders, Antwerp.

Eggers, K. & De Bal, C. (2009, December). *Spraakonvloeiendheden bij personen met syndroom van Down [*Speech disfluencies in persons with Down's syndrome*].*

Lecture presented at the annual symposium of the Vlaamse Vereniging voor Logopedisten [Flemish SLT federation], University of Antwerp, Antwerp.

De Bal, C. & Eggers, K. (2009, December). *Prevalentie van stotteren bij personen met syndroom van Down* [Prevalene of stuttering in persons with Down's syndrome]. Poster presented at the annual symposium of the Vlaamse Vereniging voor Logopedisten [Flemish SLT federation], University of Antwerp, Antwerp.

Eggers, K. & Leahy, M. (2009, November). *Educating fluency specialists in Europe.* Poster presented at the annual convention of the American Speech-Language-Hearing Association, New Orleans.

Eggers, K., De Nil, L., & Van den Bergh, B. (2009, November). *Inhibitory control in children who stutter.* Poster presented at the annual convention of the American Speech-Language-Hearing Association, New Orleans.

Leahy, M., & Eggers, K. (2009, October). *The education of EU fluency specialists.* Lecture presented at the Annual Conference of the Irish Association of speech and language therapists, Gallway.

Eggers, K. (2009, August). *Intervention strategies for a cognitive behavioral treatment of stuttering.* Seminar presented at the 6th Congress of the International Fluency Association, Rio de Janeiro.

Eggers, K., De Nil, L, & Van den Bergh, B. (2009, August). *Attention processes in children who stutter.* Seminar presented at the 6th Congress of the International Fluency Association, Rio de Janeiro.

Eggers, K., & Leahy, M. (2009, August). *EU education of fluency specialists: an innovative program.* Seminar presented at the 6th Congress of the International Fluency Association, Rio de Janeiro.

Eggers, K. (2009, June). *Temperament en stotteren: hype of therapeutisch relevant [Temperament and stuttering: hype or therapeutically relevant].* Conference organized by the Nederlandse Vereniging voor Stottertherapie [Dutch federation for stuttering therapists], Utrecht.

De Bal, C, & Eggers, K. (2009, May). *The prevalence of stuttering in persons with Down Syndrome.* Poster presented at the Convention of CPLOL, Ljubljana.

Eggers, K (2008, December). *To treat or not to treat… meer dan een shakespeareaans dilemma? [To treat or not to treat… more than a shakespearean dilemma?].* Lecture presented at the annual symposium of the Vlaamse Vereniging voor Logopedisten [Flemish SLT federation], University of Antwerp, Antwerp.

Eggers, K (2008, November). *Een kind in mijn klas met spraak-, taal-, en gehoorproblemen [*A child in my class with speech-, language-, or hearing disorders*]*. Lecture presented at the in-service teacher conference of the Vlaamse Vereniging voor Logopedisten *[*Flemish SLT federation*]*, University of Antwerp, Antwerp.

Eggers, K. (2008, October). *Stotteren: recente theoretische en therapeutische inzichten [*Stuttering: Recent theoretical and therapeutical insights*]*. Lecture presented at the 50 year Jubilee conference of the Lessius Department of Speech-, Language Therapy and Audiology, Lessius U College, Antwerp.

Eggers, K. (2008, April). *Attentional processes in children who stutter.* Lecture presented at the First European Symposium on Fluency Disorders, Antwerp.

Eggers, K. & Geudens, A. (2007, August). *Phonological abilities in children who stutter*. Seminar presented at the Socrates Intensive Program, Fontys University College, Eindhoven.

Eggers, K. (2007, August). *Specialization in SLT: The only way forward?* Seminar presented at the Socrates Intensive Program, Fontys University College, Eindhoven.

De Bal, C., & Eggers, K. (2007, August). *Prevalence of stuttering in people with Down syndrome.* Seminar presented at the Convention of the International Association of Logopedics and Phoniatrics, Copenhagen.

Eggers, K. (2007, August). *Temperament structure in stuttering, voice disordered, and typically developing children.* Seminar presented at the Convention of the International Association of Logopedics and Phoniatrics, Copenhagen.

Eggers, K. (2007, August). *Specialization in SLT: The only way forward?* Seminar presented at the Convention of the International Association of Logopedics and Phoniatrics, Copenhagen.

Eggers, K. (2007, June). *Temperamentkenmerken van kinderen die stotteren [*Temperamental characteristics of children who stutter*]*. Lecture presented at the CIOOS stuttering conference, University of Antwerp, Antwerp.

Eggers, K. (2007, March). *Onvloeiendheden bij personen met Down syndroom [Disfluencies in persons with Down Syndrome]*. Lecture presented at the International Congress of the Down Syndrome Association Flanders, University of Ghent.

Eggers, K. (2006, December). *Stuttering: To treat or not to treat... a Shakespearean dilemma in early intervention.* Seminar presented at the University of Oulu, Finland.

Eggers, K. & De Bal, C. (2006, November). *Speech dysfluencies in people with Down's Syndrome: New insights.* Seminar presented at the annual convention of the American Speech-Language-Hearing Association, Miami, Fl.

Eggers, K., De Nil, L. F., & Van den Bergh, B. R. H. (2006, September). *The role of temperament in developmental stuttering.* Poster presented at the 40 year Jubilee symposium of the department of LAW, Leuven.

Eggers, K. (2006, August). *Stuttering: prevention and early intervention.* Key Lecture presented at the Socrates Intensive Program, Université Louis Pasteur, Strasbourg.

Eggers, K., & De Bal, C. (2006, August). *Stuttering and/or cluttering in people with Down's Syndrome.* Seminar presented at the Socrates Intensive Program, Université Louis Pasteur, Strasbourg.

Eggers, K. & De Bal, C. (2006, July). Speech dysfluencies in people with Down's Syndrome. Seminar presented at the 5th IFA-congress, Dublin.

Eggers, K., De Nil, L. F., & van Den Bergh, B. R. H. (2006, July). *Temperamental structure of children who stutter.* Seminar presented at the 5th IFA-congress, Dublin.

Eggers, K. (2006, April). *Preventie van stotteren en vroegbegeleiding [*Prevention and early treatment of stuttering*].* Workshop presented at the annual symposium of the Association of the University of Leuven, Bruges.

Eggers, K. (2006, April). *Stotteren: alarmsignalen en risicofactoren [*Stuttering: Alarm signs and risk factors*].* Lecture presented at the annual symposium of the Association of the University of Leuven, Bruges.

Eggers, K., De Nil, L. F., & van Den Bergh, B. R. H. (2005, November). *Temperament Structure of stuttering, Voice Disordered, and Normal Speaking Children.* Seminar presented at the annual convention of the American Speech-Language-Hearing Association, San Diego, CA.

Eggers, K. (2005, November). *Belang van temperament bij stotterende kinderen [*The importance of temperament in children who stutter*].* Keynote presentation at the World Stuttering Day Conference, University Ghent, Ghent.

Eggers, K. (2005, August). *Cluttering: recent insights.* Key Lecture presented at the Socrates Intensive Program, University of Oulu.

Eggers, K. (2005, August). *Temperament and stuttering.* Key Lecture presented at the Socrates Intensive Program, University of Oulu.

Eggers, K. (2004, June). *Manual facilitation in hyperfunctional dyshonia.* Seminar presented at the Socrates Intensive Program, University of Malta.

Eggers, K., Meersman, M., Peelman, M., Stes, S., Van Coillie, L. (2004, March). *Broddelen: nieuwe inzichten [*Cluttering: New insights*].* Seminar presented as part of the continued professional development lectures by Lessius University College, Antwerp.

Eggers, K. (2003, August). *The acoustical analysis of stuttered speech.* Seminar presented at the Socrates Intensive Program, University of Valladollid.

Eggers, K. (2003, August). *The rationale and implications of cognitive-behavioral treatment of stuttering.* Seminar presented at the Socrates Intensive Program, University of Valladollid.

Eggers, K. (2003, June). *The use of PRAAT for the acoustical analysis of stuttered speech.* Lecture presented at the Intensive workshop of the Stuttering Foundation, University of Iowa, Iowa.

Eggers, K. (2002, August). *Basic principles of manual therapy at hypertonic dysphonia.* Seminar presented at the Socrates Intensive Program, Trinity College, Dublin.

Eggers, K. (2002, April). *De logopedische behandeling van kinderen met heesheid [The logopedic treatment of children with hoarseness].* Seminar presented at the Post Academic Training Voice Disorders, University Hospital Antwerp, Antwerp.