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UNIVERSITY OF TURKU

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Maria Paphiti

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Cover Image: The half-open door on the cover, from the humble house of my late grandfather, Kyriakos Paphitis, symbolises the way research opens doors in life, inviting us to explore and discover beyond the familiar. Photo by Stavros Karantonis.

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To my three girls: Stephanie, Eleana, and Simone.
Your love and support give me the strength
to take risks and follow my passions.

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Developmental Stuttering
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ABSTRACT

Developmental stuttering is a multifactorial, neurodevelopmental disorder that usually appears between the ages of 2 and 4 years and persists in 20–40% of children who start stuttering. The lifespan prevalence is around 0.8% and the lifetime incidence in the general population is 8%. Living with stuttering may negatively affect not only an individual's communicative ability but also their overall quality of life. Recent theoretical models and research findings speculate that there is a link between inhibitory control (IC) and cognitive flexibility (CF) and the onset, development, and/or persistence of stuttering. Previous studies with children who stutter (CWS) and children who do not stutter (CWNS) have only focused on whether there are IC/CF differences between the two groups and have included either younger (3–6yrs) or older children (6–12yrs); these investigations were conducted with a variety of measures and the findings are inconsistent.

This thesis aimed to examine IC and CF in CWS and CWNS between the ages of 4 to 9 years and comparisons were made between younger and older subgroups. This study was the first to use visual computer tasks to examine IC and CF in a combined manner in these two groups. Additionally, the link between IC, CF, and speech disfluencies was studied by investigating possible associations between the results from the experimental paradigm and the number of disfluencies produced in speech samples based on story retelling and spontaneous conversation.

The results showed that under IC and CF task-conditions (a) CWS slow down more compared to age- and gender-matched CWNS, (b) lower IC and CF performance was associated with increased production of stuttering-like disfluencies (SLDs) in CWS, and (c) older CWS (7–9yrs) were slower and made more errors, while younger (4–6yrs) had comparable results to CWNS.

Our findings provide further support for previous claims of weaknesses in IC and CF in CWS as well as for a distinct role of IC and CF in the production of SLDs and the development and/or persistence of stuttering.

KEYWORDS: inhibitory control, cognitive flexibility, speech disfluencies, developmental stuttering.

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TIIVISTELMÄ

Kehityksellinen änkytys on monitekijäinen neurologinen häiriö, jolla on kehityksellinen tausta. Änkytys ilmaantuu yleensä 2–4 vuoden iässä ja on pysyvää 20–40 prosentilla lapsista. Änkytyksen elinikäinen esiintyvyys on noin 0.8% ja ilmaantuvuus väestössä 8%. Änkytys voi vaikuttaa negatiivisesti paitsi henkilön kommunikaatiokykyyn, myös yleiseen elämänlaatuun. Viimeaikaiset teoreettiset mallit ja tutkimustulokset esittävät, että inhibitiokontrollilla ja kognitiivisella joustavuudella on yhteyksiä änkytyksen alkamiseen, kehittymiseen ja/tai pysyvyyteen. Aiemmat tutkimukset ovat keskittyneet vain siihen, onko lasten, jotka änkyttävät ja lasten, jotka eivät änkytä, välillä eroja inhibitiokontrollissa ja kognitiivisessa joustavuudessa, ja ne ovat tarkastelleet joko nuorempia (3–6-v.) tai vanhempia lapsia (6–12-v.). Tutkimuksia on tehty erilaisilla menetelmillä, ja tulokset ovat olleet ristiriitaisia.

Tämä väitöskirja tarkasteli inhibitiokontrollia ja kognitiivista joustavuutta 4–9-vuotiailla lapsilla, jotka änkyttävät, ja lapsilla, jotka eivät änkytä. Vertailuja tehtiin nuorempien ja vanhempien lasten välillä. Tämä tutkimus oli ensimmäinen, jossa tutkittiin inhibitiokontrollia ja kognitiivista joustavuutta samanaikaisesti visuaalisissa tietokonetehtävissä näissä kahdessa ryhmässä. Lisäksi inhibitiokontrollin, kognitiivisen joustavuuden ja puheen sujumattomuuksien välistä yhteyttä tutkittiin tarkastelemalla mahdollisia yhteyksiä kokeellisen paradigman tulosten ja puheessa ilmenevien sujumattomuuksien määrän välillä. Puheen sujumattomuudet oli laskettu tarinan uudelleenkerronnasta ja spontaanista keskustelusta.

Tulokset osoittivat, että inhibitiokontrollia ja kognitiivista joustavuutta mittaavissa tehtävissä (a) lasten, jotka änkyttävät, suoriutuminen oli hitaampaa kuin ikä- ja sukupuolikontrolloiduilla verrokeilla, (b) heikompi suoriutuminen liittyi suurempaan määrään änkytyksen kaltaisia sujumattomuuksia lapsilla, jotka änkyttävät ja (c) vanhemmat lapset, jotka änkyttävät (7–9-v.) olivat hitaampia ja tekivät enemmän virheitä, kun taas nuorempien (4–6-v.) suoriutuminen oli samankaltaista kuin sujuvasti puhuvilla verrokeilla.

Tutkimuksen tulokset tukevat aiempia löydöksiä inhibitiokontrollin ja kognitiivisen joustavuuden heikkouksista lapsilla, jotka änkyttävät, sekä löydöksiä inhibitiokontrollin ja kognitiivisen joustavuuden roolista änkytyksen kaltaisissa sujumattomuuksien esiintymisessä ja änkytyksen kehittymisessä ja/tai jatkuvuudessa.

ASIASANAT: inhibitiokontrolli, kognitiivinen joustavuus, puheen sujumattomuus, kehityksellinen änkytys.

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November 2024
Maria Paphiti

Table of Contents

Tables

Figures

List of Original Publications

This dissertation is based on the following original publications, which are referred to in the text by their Roman numerals:

I Paphiti, M., Jansson-Verkasalo, E., & Eggers, K. (2022). Complex response inhibition and cognitive flexibility in school-aged Cypriot-Greek-speaking children who stutter. Frontiers in Psychology, 13. https://doi.org/10.3389/fpsyg.2022.991138

II Paphiti, M., & Eggers, K. (2022). Cognitive flexibility in younger and older children who stutter. *Frontiers in Psychology*, 13. https://doi.org/10.3389/fpsyg.2022.1017319

III Paphiti, M., Talias, M.A., & Eggers, K. Inhibitory control, cognitive flexibility, and the production of disfluencies in children who do and do not stutter. *American Journal of Speech-Language Pathology*. https://doi.org/10.1044/2024_AJSLP-23-00242

1 Introduction

In early childhood, between the ages of 2 and 6 years, 5–15% of children present with developmental stuttering for either a brief (a few weeks) or a prolonged (a few months or over one to two years) period of time (Bloodstein & Bernstein Ratner, 2008; Månsson, 2000). Developmental stuttering is a multifactorial disorder that usually first appears during a period of great overall development (i.e., motor, language, emotional). Symptoms may vary from sound-, syllable-, or monosyllabic-word repetitions, to severe blocks or extended sound prolongations. If symptoms are severe and/or persistent, it is possible for children to develop secondary behaviours (e.g., movements of facial parts to get the words out) (Guitar, 2014) and/or a negative attitude toward speech already starting at preschool-age (Vanryckeghem et al., 2005).

Over the years, several theoretical models have attempted to explain why stuttering appears. During the past decades, evidence has suggested a link between developmental stuttering and domain-specific processes associated with emotional, motor, sensory, and speech-language development (e.g., Forster & Webster, 2001; Karrass et al., 2006; Ntourou et al., 2011; Watkins et al., 2008). These findings have provided support for previously existing theoretical models and led to the creation of new ones. Two of the models that received partial empirical evidence are the covert repair hypothesis (Kolk, 1991; Postma & Kolk, 1993; Postma et al., 1990) and the vicious circle hypothesis (Vasić & Wijnen, 2005). Both suggest a link between disfluencies and language processes or the speech monitoring system (Anderson et al., 2022). A newly proposed theoretical model, the executive function model of developmental stuttering (Anderson & Ofoe, 2019), proposes a direct link between developmental stuttering and the core executive functions (EFs) of inhibitory control (IC) and cognitive flexibility (CF). EF is an umbrella term that refers to top-down neurocognitive processes that allow for the planning and execution of novel or complex tasks (Diamond, 2013; Miyake et al., 2000). These processes have been associated with self-regulation (Eichorn & Pirutinsky, 2021; Nigg, 2017) and the execution of goal-driven actions, such as speech and language production (Crosbie et al., 2009; Deák, 2004; Engelhardt et al., 2009). Findings from studies with non-stuttering populations suggested that IC weaknesses contribute to disfluency (Engelhardt et al., 2010, 2013) and recently, a study with adults who stutter (AWS) reported a link between IC weaknesses and stuttering severity (Bakhtiar & Eggers, 2023). Furthermore, there is evidence from studies with non-stuttering individuals that CF weaknesses also play a role in disfluency production (Dayalu et al., 2022; Hart, 2008).

Thus far, studies in developmental stuttering have investigated EFs with a variety of measures attempting to see if children who stutter (CWS) present with less-well-developed EFs when compared to children who do not stutter (CWNS). Some studies have used parental questionnaires (Eggers et al., 2010; Ntourou et al., 2018), and despite inconsistencies, in most cases CWS were reported to receive lower scores than the CWNS on measures of IC and CF. IC and CF have also been investigated with the use of experimental paradigms. Similar to questionnaire-based studies, findings on IC were not unequivocal (e.g., Eggers et al., 2013; Eggers & Jansson-Verkasalo, 2017; Piispala et al., 2016), but in the studies that investigated CF, the findings have been clearer (e.g., Anderson et al., 2020; Eggers & Jansson-Verkasalo, 2017; Eichorn et al., 2018; Eichorn & Pirutinsky, 2021).

Motivated by the above-mentioned theoretical models and research findings, this thesis aims to provide additional insight by a combined evaluation of IC and CF in CWS and CWNS. In addition, it aims to investigate whether IC and/or CF are related to the production of disfluencies in CWS. This research is timely because none of the previous studies investigated the possible associations between IC and/or CF and the production of disfluencies in CWS or conducted a cross-sectional CF study including both preschool and school-age CWS and CWNS. Lastly, this thesis makes a distinctive contribution by evaluating IC and CF in a combined manner via the visual domain and conducting a more in-depth examination of set-shifting through the comparison between trials with and without set-shifting.

While this thesis does not focus on intervention, it is important for speech and language pathologists to be aware of the potential contributing factors that may lead to the appearance and/or persistence of developmental stuttering when delivering services to children and their families. Therefore, this thesis may be valuable for clinicians because understanding the significance of these EFs in the appearance, development, and/or persistence of stuttering equips them with a more comprehensive insight into the nature of the disorder, enabling them to implement optimal therapeutic services.

2 Review of the Literature

2.1 Developmental stuttering

2.1.1 Speech fluency and disfluency

Speech fluency is recognized and characterized by a smooth ongoing flow of speech muscular movements while producing the different speech sounds. It requires precise and appropriate coordination of the respiratory, phonatory, and articulatory systems to move from one sound, syllable, and word to the next without any interruptions (Yairi & Seery, 2021, p. 8). On the other hand, speech disfluency refers to interruptions in the ongoing flow of speech (Guitar, 2014, p. 16). Over the years, several systems have been developed for classifying disfluencies. One of the first systems was the Iowa classification system (Johnson et al., 1959). This system distinguished between the following disfluencies: (a) interjections or repetitions of syllables, sounds, words, or phrases, (b) revisions (c) incomplete phrases, (d) broken words, and (e) prolonged sounds. The authors emphasised that an overlap existed between disfluencies produced by people who do and do not stutter (Yaruss, 1997). Several years later, different classification systems made a clearer distinction between two broad categories of disfluencies, namely disfluencies that were more likely to be produced by individuals who stutter and disfluencies that were more likely to be produced by individuals who do not stutter (Conture, 1990; Gregory, 1993; Meyers, 1986; Yairi, 1996; Yairi & Ambrose, 1992; Yairi et al., 1993; Yairi et al., 1996). The terms stutter-type and normal-type disfluencies were introduced by Meyers (1986), while Gregory (1993) introduced the terms less-typical and more-typical disfluencies. Conture (1990) suggested a somewhat different distinction between the two types of disfluencies: within-word disfluencies (monosyllabic word, sound/syllable repetitions, audible/inaudible prolongations, as indicators of moments of stuttering) and between-word disfluencies (phrase/polysyllabic word repetitions, interjections, and revisions, as indicators of normal disfluency). In the following years, the terms stuttering-like disfluencies (SLDs) and other disfluencies (ODs) were suggested by Yairi and colleagues (Yairi, 1996; Yairi & Ambrose, 1992; Yairi et al., 1993; Yairi et al., 1996). In the first category, part-word repetitions, prolongations, broken words, and tense pauses were included, while in the second category, whole-word/phrase repetitions, revisions, incomplete phrases, and interjections were included. This system was later finalised and is now known as the Illinois classification system (Ambrose & Yairi, 1999). This system was chosen for this thesis as it has been widely used over the last two decades in several studies (Eggers et al., 2020). Based on this system, SLDs are (a) part-word repetitions, (b) single-syllable word repetitions, (c) dysrhythmic phonations: i.e., prolongations, blocks, and broken words. ODs are (a) interjections, (b) revisions/abandoned utterances, and (c) multisyllable word/phrase repetitions.

2.1.2 Developmental stuttering defined

"*Stuttering is more than just stuttering*" (Reardon-Reeves & Yaruss, 2013, p. 8). According to the Diagnostic and statistical manual of mental disorders-5 (American Psychiatric Association, 2013), childhood-onset fluency disorder, also known as developmental stuttering, is a multifactorial speech disorder that is characterised by disturbances in the natural-sounding flow of speech. The diagnostic criteria are as follows: (a) the child presents with at least one type of the disfluencies such as monosyllabic word, part-word repetitions (sound or syllable repetitions), sound prolongations, broken words, blocks, circumlocutions (word substitutions to avoid stuttering), physical tension during speech production; (b) the disorder causes anxiety about speaking in different communicative situations or environments; (c) the time of onset is placed in the early years of development, and (d) the disorder is not caused by any other deficit or medical condition.

Most stuttering definitions are descriptions of the core and secondary behaviours (i.e., symptoms) (e.g., Wingate, 1964). The first core behaviours that young children present are usually repetitions and sound prolongations, while blocks are typically the last to appear (Guitar, 2014). As developmental stuttering persists and as children experience the core behaviours, they begin to react to escape from them or to avoid them. Such reactions are considered secondary behaviours, also known as *escape* or *avoidance behaviours*, expressed for example, as body-part movements to escape from the moment of stuttering, or as substitutions of words or phrases to avoid the occurrence of a moment of stuttering (Coleman & Yaruss, 2014). If developmental stuttering persists for several months or years, it is possible for even young children to develop negative feelings and attitudes towards verbal communication (e.g., Vanryckeghem et al., 2005).

2.1.3 Phenomenology of developmental stuttering

Close to onset, disfluencies usually occur at the beginning of an utterance and children exhibit mainly repetitions (part-word or monosyllabic/multisyllabic-word repetitions). Most children are unaware of these repetitions, especially if they are limited in number (one or two). In cases where the disfluencies persist and increase in number and frequency, children become aware of them. In their attempt to avoid repetitions, other types of disfluencies may appear, such as prolongations, blocks, or broken words. Given that in some cases CWS continue to be disfluent for several months or years, it is possible as time passes and as the stuttering severity increases, to also develop secondary behaviours. Secondary behaviours (escape or avoidance) are a long list of behaviours that may differ from child to child (Vanryckeghem & Mukati, 2006), ranging from avoidance of eye contact to back and forth movements of the torso. Both the primary and secondary behaviours of developmental stuttering are considered "*surface features*" (Manning, 1999, p. 123) which is what we see and hear when a person stutters. Findings suggest that CWS as early as three years old, may develop a negative attitude towards their speech when compared to their non-stuttering age-matched peers (Clark et al., 2012; Vanryckeghem et al., 2005) and that this attitude increases as they get older and as stuttering persists (Vanryckeghem & Brutten, 1997). These affective, cognitive, and behavioural aspects, i.e., the degree in which communication and life are affected or limited by developmental stuttering, have been labelled the "*deep structure*" of the pathology (Manning, 1999, p. 124). To add to this, school-age CWS are at a higher risk of being bullied (Blood & Blood, 2007; Langevin & Bortnick, 1998), something that has been associated in the general population with developing anxiety later in life (Hawker & Boulton, 2000; McCabe et al., 2010). In the case of AWS, the results of a metanalytic review showed that they present with both trait (to be anxious in several life domains) and social anxiety (to be anxious in various social situations or contexts) (Craig & Tran, 2014).

2.1.4 Age of onset and spontaneous recovery

Developmental stuttering is usually reported to first appear approximately between two and four years of age which is a period of rapid speech-language and motor development (Yairi & Ambrose, 2013). Reportedly, 60% of the CWS present stuttering prior to age 3 and 85% prior to the age of 4 years (Yairi & Seery, 2021). The onset may be sudden (within a few days) or gradual (Reilly et al., 2009). The persistence of stuttering appears to be related to multiple factors. Such factors are a positive family history of stuttering, poorer articulation and phonological test performances, high frequency of SLDs in spontaneous speech samples, lower attentional skills, and lower accuracy on nonword repetition tasks (Singer et al., 2020; Walsh et al., 2021). Regarding recovery rate, despite the different factors affecting its measurement, a summary of 23 longitudinal studies reported a mean recovery rate of 58.7% across studies (Einarsdóttir et al., 2020), something that is not in agreement with a previously suggested approximation of 80% (Yairi & Ambrose, 1999).

2.1.5 Prevalence, incidence, and male-to-female ratio

According to Yairi and Ambrose (2013), based on the findings of several studies, the lifespan prevalence rate seems to centre around 0.8%, while the lifetime incidence rate was found to be around 8%. Even though gender is not associated with stuttering persistence (Ambrose et al., 2015; Kefalianos et al., 2017), the male-to-female ratio is reported to be 1.5:1 by three years old (Reilly et al., 2009) and 5.3:1 by twelve years old (Howell et al., 2008), which is higher than the previously reported 4:1 in adolescence by Craig et al. (2002).

2.1.6 Aetiological theories

In the last decades, there have been several theoretical models attempting to explain the nature and aetiology of developmental stuttering (Bloodstein & Bernstein Ratner, 2008). Most recent theories are multifactorial in nature and include genetic, cognitive, linguistic, and emotional components. This thesis has been influenced by two psycholinguistic aetiological models: the covert repair hypothesis (Kolk, 1991; Postma & Kolk, 1993; Postma et al., 1990), and the vicious circle hypothesis (Vasić & Wijnen, 2005).

In the first model, it is speculated that stuttering occurs when the internal monitoring system (as proposed by Levelt, 1989) is challenged to detect and correct errors in the pre-articulatory phase (i.e., during speech planning). Real or perceived errors are detected in the output of the formulating system and are prevented by interrupting articulation and initiating a repair (Bloodstein & Bernstein Ratner, 2008). This occurs due to poor phonological encoding skills, that is, slower phonological processing systems of the CWS compared to the CWNS (Anderson et al., 2022). Therefore, phonetic plans are prone to errors, leading to covert self-repairs which impede fluent speech production. A repetition occurs if the restart is prior to the point of the interruption. A pause, a prolongation, and/or a block occur when the person who stutters fixates on the articulatory apparatus until a repair of the speech plan is in place (Bernstein Ratner & Wijnen, 2007).

The second model, the vicious circle hypothesis, also speculates an internal monitoring system responsible for speech production. This system becomes activated to respond to different phenomena, not necessarily errors. In the case of people who stutter (PWS), the system is hypervigilant and creates more problems; specifically, the three attention parameters of the monitor—effort, focus, and threshold—are not appropriately set in PWS. Effort refers to investing more energy than required for adequate speech production; focus refers to an excessive attention on temporal fluctuation and discontinuity; and threshold refers to setting the bar so high that any type of discontinuity in speech is perceived as disfluency (Vasić & Wijnen, 2005). In simpler terms, repairs are made during the articulatory phase, even when they are not needed. It is assumed that PWS are overly attentive to monitoring their speech, leading to interruptions in the ongoing speech which then requires further repairs (Bernstein Ratner & Wijnen, 2007).

Both IC and CF have been associated with speech-language planning and execution (Crosbie et al., 2009; Deák, 2004; Engelhardt et al., 2010, 2013). The covert repair hypothesis and the vicious circle hypothesis, although not explicitly mentioning IC and CF, indirectly support the idea that they are related to stuttering. IC enables the discontinuation of inappropriate responses, while CF allows for flexible shifting of attention—such as moving from one articulatory plan to another. The covert repair hypothesis posits that PWS have slower phonological processing, leading to more speech errors and an increased likelihood of covert repairs. Weaknesses in IC and CF could impair the ability to regulate cognitive and behavioural responses effectively, making it harder to detect and correct errors prior to articulation and redirect attention away from errors, ultimately contributing to speech disfluencies. The vicious circle hypothesis could also be linked to weaknesses in IC and CF contributing to disfluencies. The hypervigilant internal monitoring system in PWS leads to unnecessary repairs during the articulatory phase. In the case of IC weaknesses, it becomes difficult to suppress these inappropriate repairs, exacerbating disfluencies. Similarly, weaknesses in CF may limit the ability to adjust articulatory plans in a timely manner, further increasing disfluencies during speech production.

The research for this thesis was also influenced by a recently proposed theoretical model that attempts to explain the onset and persistence of developmental stuttering: the executive function model of developmental stuttering (Anderson & Ofoe, 2019). This model states that the domain-specific processes of motor, sensory, language, speech, and emotion are interrelated with the domain-general processes or EFs of working memory, IC, and CF. Any weaknesses in the EFs directly impact the domain-specific processes and any weaknesses or delays in the domain-specific processes directly impact the EFs. For example, any weaknesses in working memory and/or IC may contribute to less stable long-term phonological and/or lexical representations in the mental lexicon and any weaknesses in CF may pose limitations to flexibly adopting a new more accurate production pattern. The model receives validation from recent evidence that suggests that CWS score lower on language testing (but within normal limits) (Ntourou et al., 2011). It is therefore possible that this impacts the EFs, given their association with language. Lastly, fluency is considered as one of the domain-specific processes and it is viewed as being directly interrelated with the EFs and indirectly related with the language processes suggesting that it becomes affected when weaknesses are present (Figure 1).

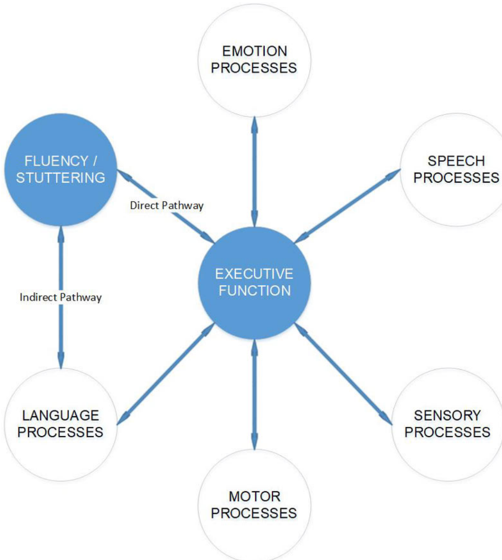

Figure 1. Executive function model of developmental stuttering (reproduction from Anderson and Ofoe, 2019).

2.2 Executive Functions

2.2.1 Definition and development

EFs are "a set of general-purpose control mechanisms, often linked to the pre-frontal cortex of the brain, that regulate the dynamics of human cognition and action" (Miyake & Friedman, 2012, p. 8). EF is usually delineated into three distinct core but interdependent components: working memory, IC, and CF (Diamond, 2013; Miyake et al., 2000) that allow "to effortfully guide behaviour toward a goal, especially in nonroutine situations" (Barkley, 2012, p. 7). The three core EFs share a common mechanism which is the foundation upon which they develop: effortful attentional control (Garon et al., 2008). Other researchers over the last decades have suggested different components for the EF. For example, Anderson (2002) suggested a somewhat different model which incorporates four inter-correlated but dissociable components of the EF: CF, goal setting, attentional control and information processing. In this model, working memory is a subcomponent of CF, while aspects of IC are placed under the attentional control components. The view adopted in this thesis is the one proposed by Diamond (2013), as it has been adopted widely in studies within the stuttering population (e.g., Anderson & Ofoe, 2019; Ntourou et al., 2018; Torrington Eaton & Ratner, 2016). Reportedly, the EFs emerge early in development with important changes occurring between the ages of 2- to 5-years. While they typically reach a level comparable to adult performance at about 12 years of age, some aspects of the EFs continue to develop thereafter (Zelazo & Müller, 2011). On one hand, EF has been associated with the overall child development (e.g., emotional, linguistic; Miyake et al., 2000), school readiness and performance (e.g., Blair, 2016), mental and physical health (e.g., Burton et al., 2022; Johnson, 2012) as it plays an important role in successful task completion in everyday life (e.g., Beer et al., 2011; Pisoni et al., 2010; Zelazo & Cunningham, 2007). On the other hand, executive dysfunction has been associated with different developmental disorders (e.g., Sparrow, 2012; Zelazo & Müller, 2011) such as speech sound disorders (e.g., Crosbie et al., 2009; Torrington Eaton & Ratner, 2016), language disorders (e.g., Kapa et al., 2017), and developmental stuttering (e.g., Anderson & Ofoe, 2019).

2.2.2 Working memory

Working memory is the ability to temporarily hold information in mind and manipulate it, that is, to generate action (Davidson et al., 2006; Zelazo et al., 2003). It is transient as it holds the information in hand for a short period of time, until a given task is completed (Logan, 2004). It begins to develop as early as 9 months of age (Diamond, 2013) and continues to develop throughout early childhood and adolescence (Best, 2010). Baddeley (1996) suggests a model for working memory in which the main component is the central executive system that has two subcomponents: the phonological loop and the visuospatial sketchpad (Sasisekaran & Basu, 2017) that both feed back into the central executive system (Alloway et al., 2004). According to this model, when a task involves speech and language processes, then the phonological loop is involved, but when it involves visual and spatial information then the visuospatial sketchpad is involved. When performing a task, the central executive system can coordinate and manipulate information from either of the two subcomponents (Zelazo et al., 2003). It is believed that this system is in place by the age of 4 to 6 years (De Luca & Leventer, 2008).

Working memory has been extensively investigated in developmental stuttering (Ofoe et al., 2018) and was not investigated in this thesis. Some of the tasks used involved nonword repetitions (e.g., Hakim & Ratner, 2004; Pelczarski & Yaruss, 2016) and repetitions of lists of digits, letters, or words—forward memory spans (e.g., Bakhtiar et al., 2007; Pelczarski & Yaruss, 2016; Sasisekaran & Byrd, 2013). Findings in most of the studies reported CWS compared to CWNS to score lower in both nonword repetition and forward span measures (Anderson & Ofoe, 2019).

2.2.3 Inhibitory control

IC is an umbrella term that encompasses interference control and response inhibition (Friedman & Miyake, 2004). Interference control refers to resistance to proactive interference and resistance to distractor interference. In the first case, it refers to suppressing thoughts and memories (cognitive inhibition), while in the second case, it refers to inhibiting distracting or irrelevant information, staying focused on what is important—selective attention (Diamond, 2013; Friedman & Miyake, 2004). Response inhibition refers to inhibiting a manual response when it is no longer appropriate (Garon et al., 2008). It is considered complex (complex response inhibition [CRI]) when the requirement is not to inhibit a response but rather to execute a conflicting one (Anderson & Wagovich, 2017). IC has been associated with speech-language development, formulation, and production (e.g., Engelhardt et al., 2009; Müller et al., 2009) and appears to have a reciprocal relationship with speech and language. Language allows the use of self-directed speech to inhibit inappropriate action plans (Müller et al., 2009) and IC allows withholding inappropriate motoric movements for speech production (Engelhardt et al., 2009, 2010).

IC can be evaluated with the use of parental questionnaires, such as the Children's Behaviour Questionnaire (Rothbart et al., 2001) which includes a scale for IC. It can also be evaluated with the use of behavioural observations, for example the gift delay procedure during which a child is asked not to peek while the experimenter noisily wraps a gift for the child (Kochanska et al., 1996). Lastly, IC can be evaluated with the use of experimental paradigms (both computerised and non-computerised). Specific types of experimental paradigms allow for the assessment of either interference control or response inhibition.

Interference control may be examined with the use of antisaccadic tasks such as the Stroop Effect (Stroop, 1992) or the Flanker task (Eriksen & Eriksen, 1974). In a colour/word Stroop Effect task, participants are asked to name the colour of the ink while ignoring the written word. In the Flanker task, participants are instructed to respond to a central object based on its location, ignoring the surrounding distractor images (Zhou et al., 2022). Briefly, in the Stroop Effect task, participants must inhibit the automatic tendency to read the word, while in the Flanker task, they must inhibit the influence of surrounding distractors and focus on the central stimulus. This is why some researchers (e.g., Miyake et al., 2000) view the Stroop Effect task as a measure of inhibition of prepotent responses. Newer versions, such as the Day/Night task are considered measures of CRI (Loe et al., 2015), as they require both inhibition and memory (Gerstadt et al., 1994). Nevertheless, the colour/word Stroop Effect task is widely used to investigate interference control (Van Mourik et al., 2005).

Response inhibition may be examined with the use of a Go/NoGo (Mesulam, 1985) or of a Stop-signal task (Logan & Cowan, 1984). During a Go/NoGo task participants are instructed to press a response key when the go signal is present but withhold the motor response when the nogo signal is present. This assesses the ability to suppress prepotent responses when faced with infrequent nogo cues. During a Stop-signal task, participants are required to inhibit the already-initiated response, which allows for the measuring of the ability to interrupt ongoing actions and therefore providing a dynamic assessment of the speed and efficacy of the inhibitory process. Therefore, the first task evaluates how well participants can suppress strong, automatic responses, while the second task evaluates how well participants can stop an already-initiated response, indicating that they are not identical in their inhibitory requirements (Verbruggen & Logan, 2008). Lastly, CRI which requires altering the motoric response, is usually assessed by comparing the performance in a block of trials in which the participants are instructed to respond in a specific way, while in the following block of trials they are instructed to respond in a conflicting manner. In this case, a mixed-block design task, as described by Gopher et al. (2000) may be used, similar to the one used in this thesis.

2.2.4 Cognitive flexibility

CF is also labelled in the literature as "*mental flexibility*", "*set-shifting*", or "*mental set-shifting*" (Diamond, 2013). Based on the existing definitions of CF, it has been conceptualised as a (a) set-shifting ability of the cognitive system, (b) higher-order ability of cognitive control, (c) property of cognition or mental states, and (d) measure of divergent thinking (Ionescu, 2012). It has been associated with speech-language development (Crosbie et al., 2009; Deák, 2004) and self-regulation (Eichorn & Pirutinsky, 2021; Nigg, 2017). There are no questionnaires that solely evaluate CF. However, the Behavioural Rating Inventory of Executive Function-Preschool (Gioia et al., 2003) includes one scale entitled 'Shift' which focuses on CF. CF may also be evaluated with behavioural observations; an example of this is an object sorting procedure as described by Ionescu (2012) where seven objects (coloured cups and toys) are used and the participant needs to place the target object in two distinct categories (i.e., a category with red items or a category with toys) based on either colour or object use. Lastly, CF may be evaluated with the use of experimental paradigms. An example of an experimental paradigm suitable for children is the Dimensional Change Card Sort (Frye et al., 1995) during which the participants are instructed to sort cards first based on one dimension (e.g., by colour), and then based on another dimension (e.g., shape); an activity that evaluates the ability to switch. The ability to set-shift is usually evaluated with the use of a Mixed-block design task, as was the case in this thesis. In these paradigms, set-shifting is conceptualised as randomly performing task switching within a block of trials. Both compatible and incompatible trials are presented in a fixed but random order requiring appropriate motor responding. CF may be measured by comparing the performance during a compatible block of trials, with the performance during the compatible trials of a mixed block. In the compatible block, participants repeat a motoric response (task repetition), while in the mixed block the motoric responses are either repetitive (trials of similar compatibility) or altered (trials of conflicting compatibility). Therefore, the use of a Mixed-block design task allows for a direct comparison of the ability to set-shift.

2.3 Developmental stuttering, inhibitory control, and cognitive flexibility

2.3.1 Inhibitory control and developmental stuttering

Studies examining IC in CWS were conducted either with the use of parental questionnaires (Anderson & Wagovich, 2010; Eggers et al., 2010; Ntourou et al., 2018; Rocha et al., 2019) or experimental paradigms investigating either interference control or response inhibition (Anderson & Wagovich, 2017; Eggers et al., 2012, 2013, 2018; Eggers & Jansson-Verkasalo, 2017; Harrewijn et al., 2017; Piispala et al., 2016, 2017, 2018). There are a small number of studies that used behavioural observations, but because their focus was on emotional regulation, they are not included in this literature review (e.g., Arnold et al., 2011; Johnson et al., 2010; Ntourou et al., 2013). Findings from parental questionnaire-based studies seem inconsistent (Table 1): two of the studies suggested comparable findings between CWS and CWNS (Anderson & Wagovich, 2010; Rocha et al., 2019), while the other two suggested lower scores in CWS (Eggers et al., 2010; Ntourou et al., 2018). These inconsistencies may be attributed to several factors. First, sample sizes and age ranges varied across the studies. For instance, the study by Anderson and Wagovich which reported comparable IC, had a much smaller sample size compared to the other three studies. Additionally, the two studies that reported lower IC in CWS included younger children, suggesting that IC deficits may be more pronounced in early childhood and could diminish with age. Lastly, given that the four studies were conducted in three different countries, cultural differences may also account for some of the variability.

The experimental-paradigm study investigating interference control reported comparable findings between CWS and CWNS (Eggers et al., 2012). Two of the studies investigating response inhibition suggested a lower IC for the CWS (Piispala et al., 2017, 2018), while the other three reported comparable results between CWS and CWNS (Eggers et al., 2018; Harrewijn et al., 2017; Piispala et al., 2016). Based on the findings, with the exception of the Piispala et al. (2016) study, the studies that used the Go/NoGo task reported lower IC performance for the CWS compared to the CWNS. In contrast, the studies that used the more challenging Stop-signal task, reported comparable results between the two groups (Table 2). The difference in findings may be attributed to the different stimulus mapping. In the Go/NoGo task, the participants are asked to respond in the go stimulus and not in the nogo stimulus, while in the Stop-signal task, they are asked to refrain from responding when the stop signal appears. The difference may also be attributed to variations in the age ranges across the studies. The Go/NoGo task studies included children as young as 4 years old, while the two studies that used the Stop-signal task included children aged 7 to 14 years old. In this thesis (in Studies I and III), given Diamond's (2013) argument that Go/NoGo and Stop-signal tasks may not be paradigmatic examples of IC as used in everyday life, a CRI task was chosen. While it also does not perfectly mirror real-world scenarios, it was considered a better choice for examining IC. Its heightened sensitivity and complexity require constant monitoring, decision-making, and adaptation—better reflecting the dynamic nature of IC demands in everyday settings. Notably, only two studies have used CRI tasks, both of which were auditory-based and reported lower performance for the CWS compared to the CWNS (Anderson & Wagovich, 2017; Eggers & Jansson-Verkasalo, 2017). Using a visual task was the most logical approach to clarify previous findings and ensure that the auditory processing difficulties often reported in CWS (Foundas et al., 2004; Hampton & Weber-Fox, 2008; Jansson-Verkasalo et al., 2014; Liotti et al., 2010) did not confound the results, thereby providing further evidence to elucidate previous conclusions. Overall, this remains an understudied area of research with inconsistencies in the findings that may be attributed to a variety of factors.

Table 1. Summary of studies that used questionnaires to examine IC in CWS and CWNS.

Study	n	Age range	Questionnaire used	Findings
Eggers et al., 2010	58 CWS 58 CWNS	3.00–9.00	Children's Behaviour Questionnaire	CWS, compared to CWNS, were rated lower on IC. No association was found between IC and overt stuttering symptoms.
Anderson and Wagovich, 2010	9 CWS 14 CWNS	3.00–6.00	Children's Behaviour Questionnaire (Short Form)	Comparable findings in the ratings of the two groups (CWS & CWNS).
Ntourou et al., 2018	75 CWS 75 CWNS	3.00–6.00	Behavioural Rating Inventory of Executive Function-Preschool	CWS, compared CWNS, were rated lower on IC. No association was found between IC and overt stuttering symptoms.
Rocha et al., 2019	50 CWS 50 CWNS	7.00–12.00	Temperament in Middle Childhood Questionnaire	Comparable findings in the ratings of the two groups (CWS & CWNS).

Table 2. Summary of studies that used experimental paradigms to examine IC in CWS and CWNS.

Study	n	Age range	Paradigm used	Domain and response	Type of paradigm	Findings
Eggers et al., 2012	41 CWS 41 CWNS	4.00–9.00	Attention Network Test	D: Visual R: Manual	Computerised/ behavioural	Comparable findings between CWS and CWNS (interference control).
Eggers et al., 2013	30 CWS 30 CWNS	4.00–10.00	Go/NoGo	D: Visual R: Manual	Computerised/ behavioural	CWS presented with reduced IC.
Piispala et al., 2016	11 CWS 19 CWNS	5.00–10.00	Go/NoGo	D: Visual R: Manual	Computerised/ behavioural	Comparable findings between CWS and CWNS.
Harrewijn et al., 2017	17 CWS 19 CWNS	9.00–14.00	Stop-signal	D: Visual R: Manual	Computerised/ behavioural & neurocognitive	Comparable findings between CWS and CWNS in the behavioural measure. CWS presented with increased IC (reflecting problems with action selection) in the neurocognitive measure which was associated with decreased overt stuttering symptoms.
Piispala et al., 2017	11 CWS 19 CWNS	5.00–10.00	Go/NoGo	D: Visual R: Manual	Computerised/ neurocognitive	CWS presented with reduced IC.
Anderson and Wagovich, 2017	41 CWS 41 CWNS	3.00–6.00	Grass-snow and Baa-meow	D: Auditory R: Manual	Computerised/ behavioural	CWS presented with reduced IC. Compared to CWNS, they (a) were slower and (b) less accurate in tasks measuring CRI.
Eggers and Jansson-Verkasalo, 2017	16 CWS 16 CWNS	6.00–10.00	Auditory Set-shifting	D: Auditory R: Manual	Computerised/ behavioural	CWS presented with reduced IC. Compared to CWNS, they were less accurate in tasks measuring CRI.
Piispala et al., 2018	11 CWS 19 CWNS	5.00–10.00	Go/NoGo	D: Visual R: Manual	Computerised/ neurocognitive	CWS presented with reduced IC.
Eggers et al., 2018	18 CWS 18 CWNS	7.00–11.00	Stop-signal	D: Auditory R: Manual	Computerised/ behavioural	Comparable findings between CWS and CWNS.

2.3.2 Cognitive flexibility and developmental stuttering

Similar to IC, studies that examined CF in CWS compared to CWNS have used either parental questionnaires (Eggers et al., 2010; Ntourou et al., 2018) or experimental paradigms (Anderson et al., 2020; Eggers & Jansson-Verkasalo, 2017; Eichorn et al., 2018; Eichorn & Pirutinsky, 2021; Ntourou et al., 2018; Rocha et al., 2019). In the two questionnaire-based studies, the participating children had overlapping ages, 3- to 8-year-olds (Eggers et al., 2010) and 3- to 6-year-olds (Ntourou et al., 2018). In both studies, weaknesses in shifting were reported for the CWS. Among the remaining studies that used experimental paradigms to examine CF in CWS and CWNS, two used noncomputerised paradigms (Ntourou et al., 2018; Rocha et al., 2019). Ntourou et al. (2018) reported lower performance for the 3-year-old CWS, while Rocha et al. (2019) reported lower performance for the 7- to 9-year-old and not for the 10- to 12-year-old subgroups of CWS. The four studies (see Table 3) that used computerised paradigms (Anderson et al., 2020; Eggers & Jansson-Verkasalo, 2017; Eichorn et al., 2018; Eichorn & Pirutinsky, 2021) reported lower performance for the CWS group (either in speed or accuracy). Given the limited number of studies investigating CF in CWS, further research is needed, implementing improved methodology. In this thesis, two visual computerised paradigms were used, focusing on comparisons of performance during set-shifting and no set-shifting trials. Such comparisons provide a more representative assessment of CF, reflecting the constant set-shifting required in real-world communication and functioning.

Table 3. Summary of studies that used experimental paradigms to examine CF in CWS and CWNS.

Study	n	Age range	Paradigm used	Domain and response	Type of paradigm	Findings
Eggers and Jansson-Verkasalo, 2017	16 CWS 16 CWNS	6.00–10.00	Auditory Set-shifting	D: Auditory R: Manual	Computerised	CWS presented with reduced CF (more errors, unable to slow down to reduce errors).
Eichorn et al., 2018	16 CWS 30 CWNS	3.00–7.00	Dimension Card Change Sort	D: Visual R: Manual	Computerised	CWS presented with reduced CF (greater slowing down after switching and greater concern about error).
Ntourou et al., 2018	75 CWS 75 CWNS	3.00–6.00	Head-toes-knees-shoulders	D: Auditory R: Manual	Noncomputerised	3-year-old CWS presented with reduced CF. All other age groups had comparable findings. No association was found between CF and overt stuttering symptoms.
Rocha et al., 2019	50 CWS 50 CWNS	7.00–12.00	Children's Colour Trail Test	D: Visual R: Manual	Noncomputerised	7- to 9-year-old CWS presented with reduced CF (were slower and made more errors).
Anderson et al., 2020	44 CWS 44 CWNS	3.00–6.00	Double Semantic and Perceptual Categorization	D: Visual R: Manual	Computerised	CWS presented with reduced CF (slower when switching in both the verbal and nonverbal domains).
Eichorn and Pirutinsky, 2021	15 CWS 18 CWNS	8.00–12.00	Dimension Card Change Sort	D: Visual R: Manual	Computerised	CWS presented with reduced CF (were slower in the mixed block).

2.3.3 Inhibitory control, cognitive flexibility, and speech disfluencies

Several studies have explored the role of IC/CF in speech-language production across various non-stuttering populations and age groups, with some suggesting an association between the two (e.g., Dayalu et al., 2022; Engelhardt et al., 2010, 2013; Lee et al., 2017). Engelhardt et al. (2010) reported that the speech-language production system relies on IC, while other researchers have suggested that weaknesses in IC contribute to disfluencies (Engelhardt et al., 2011; Lee et al., 2017; MacFarlane et al., 2017). Dayalu et al. (2022) found an association between lower CF scores and increased disfluency in a study involving adults with Parkinson's disease, a similar finding also being reported in an unpublished study by Hart (2008) with preschoolers. Overall, it has been claimed that children with EF weaknesses experience a higher frequency of SLDs and ODs (Engelhardt et al., 2013).

In the area of developmental stuttering research, only two questionnaire-based studies have explored a potential association between IC weaknesses and disfluency production (Eggers et al., 2010; Ntourou et al., 2018), and one study used the Stop-signal task to examine IC (Harrewijn et al., 2017). None of the questionnaire-based studies found an association between IC performance and SLD production, whereas Harrewijn et al. (2017) reported a link between increased IC and decreased SLDs. These same questionnaire-based studies also investigated whether CF is related to SLD production but reported similar results to those found for IC. Interestingly, none of the experimental-paradigm studies have explored the possibility of an association between CF weaknesses and disfluency production.

Study III was designed in the knowledge that there are limited number of studies directly investigating the association between EF weaknesses and disfluency, coupled with the executive function model's suggestion that such weaknesses may contribute to disfluencies. This study aimed to directly examine the relationship between IC/CF and SLDs/ODs. By addressing this gap in the literature, Study III attempted to provide a clearer understanding of how these EFs potentially influence disfluency production.

2.3.4 Existing literature gap in developmental stuttering, inhibitory control, and cognitive flexibility

Previous research on IC in CWS compared to CWNS has produced inconsistent results, with some behavioural studies on response inhibition (Eggers et al., 2018; Piispala et al., 2016) not fully aligning with parental-questionnaire studies that often reported lower IC in CWS. These inconsistencies may stem from methodological differences, such as varying age ranges among participants (e.g., studies including very young children like Anderson & Wagovich, 2017, versus those spanning childhood to adolescence like Harrewijn et al., 2017) and differences in the specific types of IC measured. Notably, studies focusing on CRI (Anderson & Wagovich, 2017; Eggers & Jansson-Verkasalo, 2017) have shown more consistent findings, suggesting that CRI measures might be more sensitive in detecting differences between stuttering and non-stuttering groups. However, these CRI studies exclusively used auditory tasks, which is significant given the documented auditory processing difficulties in CWS Therefore, conducting a CRI study using visual measures is warranted to extend previous findings and potentially resolve the inconsistencies observed in earlier research.

Regarding CF, although findings have been clearer compared to IC, there is still limited information on how CF develops in CWS and how it relates to disfluency production. Most studies have focused on either preschool children (Anderson et al., 2020; Eichorn et al., 2018) or school-age children (Eggers & Jansson-Verkasalo, 2017; Eichorn & Pirutinsky, 2021; Rocha et al., 2019). Rocha et al. (2019) conducted the only cross-sectional study to date, but it included school-age children, leaving a gap in our understanding of how CF develops between the ages of 4 and 6. Furthermore, while set-shifting—considered a robust measure of CF (Crone et al., 2006; Gajewski et al., 2010; Liu et al., 2016)—has been explored in just two studies (Eggers & Jansson-Verkasalo, 2017; Eichorn & Pirutinsky, 2021) these studies did not compare set-shifting and no set-shifting trials. Most previous research on CF has primarily focused on the concept of switching, but set-shifting, which examines how participants adapt to random shifts between compatible and incompatible trials, offers a more comprehensive understanding of how cognitive processes handle unpredictable rule changes. This approach better reflects real-life situations (Schmitter-Edgecombe & Langill, 2006) and justifies the need for a deeper investigation of CF, especially to understand its development between preschool and school-age CWS.

Lastly, research investigating the association between weaknesses in IC/CF and speech disfluencies is also limited, with only three studies conducted to date, and their results have been mixed (Eggers et al., 2010; Harrewijn et al., 2017; Ntourou et al., 2018). Additionally, none of these studies have directly examined both SLD and OD production, as they focused solely on SLDs.

In conclusion, the existing literature on EFs and developmental stuttering is both limited and inconclusive especially in the studies that focused on IC. Most studies have employed various methods, often investigating either IC or CF. This gap highlights the need for further research. To address this gap, in Study I, IC and CF were examined in a combined manner via the visual domain, including children of similar ages to those in a previous auditory-based study (i.e., Eggers & Jansson-Verkasalo, 2017) to determine if similar findings would emerge. In Study II, CF was explored in both preschool and school-age children allowing for comparisons between the two subgroups (focusing on CF development) with the use of improved methodology (comparing set-shifting trials to no set-shifting trials as in Study I). Lastly, in Study III, the association between IC/CF and SLD/OD production was investigated, an area that is also understudied with only three published studies with inconsistent findings (Eggers et al., 2010; Harrewijn et al., 2017; Ntourou et al., 2018).

3 Aims of the Study

This thesis focuses on examining IC and CF in CWS and CWNS and their role in speech fluency. This is the first time that IC and CF have been investigated through the visual domain using a single behavioural measure and whether the given performance was in any way associated with speech disfluencies produced by CWS. In addition, this is the first time that an in-depth examination of CF has been conducted comparing, set-shifting to no set-shifting trials and preschool to school-age CWS and CWNS, allowing, to a certain extent, an investigation of how CF develops in 4- to 9-year-old CWS.

Study I aimed to examine IC, more specifically CRI and CF, in a combined manner in terms of performance costs, in 6- to 9-year-old CWS and CWNS. CWS were hypothesised to exhibit slower response times and higher error percentages (higher performance costs in both cases) under IC and CF task-conditions when compared to age- and gender-matched CWNS.

Study II aimed to examine CF, within and across two classification groups (CWS versus CWNS) and two age groups (younger versus older). The assessment was conducted with 4- to 9-year-old age- and gender-matched CWS and CWNS. CWS (both age groups) were hypothesised to have slower response times (higher performance costs in speed), reflecting weaknesses in CF when compared to the corresponding age group of CWNS.

Study III aimed to investigate whether there were any associations between the speed/accuracy IC/CF performance costs calculated from the computer paradigm used in Study I and speech disfluencies produced by CWS in two speech samples combined: story retelling and casual conversation. It was hypothesised that there would be associations between the IC/CF performance costs and speech disfluencies produced by the CWS.

4 Materials and Methods

4.1 Research composition

4.1.1 Participants and ethics

For Studies I and III, data were collected in Cyprus by the PhD candidate, while for Study II, data were collected in Belgium by the PhD supervisor as part of a large series of ongoing studies. The study group for Studies I and III consisted of 19 CWS and 19 age- and gender-matched CWNS between the ages of 6–9 years; while for Study II, the study group consisted of 37 CWS and 37 age- and gender-matched CWNS between the ages of 4–9 years. Only Greek-speaking children who attended mainstream schools were invited for Studies I and III. The same applied for Study II but with Dutch-speaking children. All participants, when recruited were acquiring native competence in a single language which was the language spoken at home by both parents and the language being used at school. In the three studies, CWS were recruited after an open call for study participation, sent to speech-language pathologists working with CWS; CWNS were recruited through the school system. For all studies, participants were divided into two groups based on a diagnosis of stuttering (CWS, CWNS), while for Study II, participants were further divided based on their age (younger: CWS-Y, CWNS-Y, older: CWS-O, CWNS-O).

All studies in this thesis had roughly similar inclusion criteria. For all the participants in the CWS group, there was parental concern about stuttering and a diagnosis of stuttering by a speech-language pathologist. For Study I and III, participants had to receive a score of at least "*mild*" on the Stuttering Severity Instrument-4 (SSI-4; Riley, 2009). For Study II, they had to receive a score of at least "*very mild*" on the Stuttering Severity Instrument-3 (SSI-3; Riley, 1994) and have produced at least three monosyllabic word repetitions and/or within-word disfluencies (sound/syllable repetitions, prolongations, broken words, or blocks) in 100 words of spontaneous speech (Conture, 2001). Stuttering severity (for the CWS) is presented in Table 4. For the children in the CWNS group, for all studies, there had to be no parental concern regarding stuttering and their score on the stuttering severity test had to be less than "*mild*". In addition, all participants had to (a) have no known or questionnaire-reported psychological, neurological, or developmental problems, except for stuttering for the CWS group, (b) have normal or corrected-to-normal vision, and (c) pass a hearing screening at 500, 1,000, 2,000 and 4,000 Hz with signals presented at 20 db. To preclude any cognitive group differences, all participants were screened using the vocabulary and block design subtests of the Wechsler Intelligence Scale for Children-Third Edition (WISC-3; Wechsler, 1997, 2005). The two subtests were chosen as they correlate well with the overall score of the test (Groth-Marnat, 1997). To assess language abilities, in Study I and III, the Bus Story Test (Renfrew, 2010) was administered since there are no standardised language tests for 6–9-year-old Greek-speaking children. This test is widely used in its unstandardised form in studies with Greek-speaking children (e.g., Theodorou et al., 2016). In Study II, the Vocabulary Production and Sentence Production subtests of the Language Test for Children (Van Bon & Hoekstra, 1982), were administered. Data collection was conducted in two 35- to 45-minute sessions, approximately one week apart, in rooms where sounds and distractions were minimal at either the children's school or speech clinic. For the completion of the behavioural tasks, a 15-in. screen laptop was placed on a table in front of a plain wall approximately 18 in. from the participant.

All the families were volunteers. Prior to enrolling in the studies, written consent was obtained from the parents and/or the participants. The study design of Study I and III was approved by the National Bioethics Committee of Cyprus, while Study II was approved by the Research Ethics Committee of Leuven University Hospitals. All studies were conducted in accordance with the Declaration of Helsinki.

Table 4. Stuttering severity of CWS in Studies I, II, and III.

	Study I and III	**Study II**
Stuttering severity		
Very mild	0	5
Mild	7	17
Moderate	9	12
Severe	2	2
Very severe	1	1

4.1.2 Behavioural measures

Baseline speed task

The Baseline speed task from the Amsterdam Neuropsychological Tasks (ANT; de Sonneville, 2009) was administered in all the studies prior to undertaking any other behavioural measures. This was done to allow the participants to become familiarised with computerised testing and to eliminate the possibility of response time differences between the groups. The stimulus was a central white square that appeared on a centralised white fixation cross presented on a black screen. Participants were seated in front of the laptop, were instructed to place the index finger of their non-dominant hand on the keypad and to press the response key as soon as the white cross changed into a white square. Prior to initiating the experimental session of 32 trials for both index fingers, participants were asked to watch an instruction session of two trials and to complete a practice session of 10 trials. The signal duration was variable until participants responded. Valid responses had to fall between 150 ms and 4,000 ms after stimulus onset. Post-response intervals varied randomly from 500 ms to 2,500 ms.

Response organisation objects task

The Response organization objects task (ROO; de Sonneville, 2009) is a visual computer task appropriate for children aged 4 to 12 years old (See Figure 1 on page 6 of Article 1). It was used to assess CRI and CF in a combined manner. It measures both the speed and accuracy of manual responses by asking participants to place both index fingers on the corresponding response keys of the keypad and respond as soon and as correctly as possible when the stimulus appears on the screen. The task comprises of three blocks: Block 1 (hereafter compatible block), where a green ball appears on either the left or right side of the black screen and participants respond in a compatible manner; Block 2 (hereafter incompatible block), where a red ball appears on either the left or the right side of the screen and participants respond in an incompatible manner; and Block 3 (hereafter mixed block), where either a green or a red ball appears and participants respond accordingly. Signals are presented in a fixed randomised order in all blocks and remain on the screen until a valid response is given within 200 ms and 6,000 ms. The compatible and incompatible blocks have 30 trials each, while the mixed block has 60 trials. The results obtained by the ROO task were used for Studies I and III.

Shifting attentional set-visual task

The Shifting attentional set-visual task (SSV; de Sonneville, 2009) is a visual computer task (See Figure 1 on page 5 of Article 2). According to the manual, it is appropriate for children aged five years or older but it has been used in studies with children as young as 4 years old (e.g., Buizer et al., 2005).The task evaluates both CRI and CF but for the purposes of this thesis only CF data were analysed. Similar to ROO, SSV consists of three blocks, with Block 1 having a compatible, Block 2 an incompatible, and Block 3 a mixed stimulus-response mapping. The stimulus is either a green or red square that jumps randomly to the left or right of a horizontal bar of 10 blank squares, depending on the stimulus-response mapping. Participants were instructed to place both index fingers on the corresponding response keys and respond according to the stimulus colour, either in a compatible (green square) or incompatible manner (red square). Valid responses had to fall between 150 ms and 5,000 ms and signals in all blocks were presented in a fixed randomised order, remaining on the screen until a response was given. The compatible and incompatible blocks have 40 trials each, while the mixed block has 80 trials.

Data collection in Belgium occurred before data collection in Cyprus, but data from Cyprus were analysed first. In Belgium, the SSV task was selected due to a publication using the Auditory Set-shifting task of the ANT (Eggers & Jansson-Verkasalo, 2017), as it is its visual counterpart. In that study, using the Auditory Set-shifting task, weaknesses in IC and CF were highlighted within the CWS group. For the data collection in Cyprus (Studies I & III), the ROO task was chosen because in this thesis it was aimed to further study IC and CF through the visual domain given the limited and inconclusive research. Given the very limited research on set-shifting, (Eggers & Jansson-Verkasalo, 2017; Eichorn & Pirutinsky, 2021), a key component of CF, when the SSV data were analysed, it was also decided to focus exclusively on CF. It needs to be noted that the two tasks used are similar in that they are both visual, mixed-block design tasks, but they do differ to some extent. The SSV task is more challenging since the stimulus is dynamic, changing position on the horizontal bar, whereas in the ROO, it is static and in the mixed block of the SSV there are 80 trials, compared to 60 of the ROO task.

4.1.3 Speech disfluencies

Speech samples were collected and analysed for all studies for the administration of the SSI-3 (Study II) and SSI-4 (Study I & III) with the total combined speech sample for each participant being a minimum of 300 words. In Studies I and II, casual conversation speech samples were collected during Sessions 1 and 2, while in Study I, a story retelling speech sample was also collected during Session 1. In Study III, the casual conversation and story retelling speech samples collected in Study I (session 1) were used for a more in-depth analysis of the disfluencies.

For Study III, story retelling involved the administration of the Bus Story Test as per the test manual. Participants were instructed to listen carefully to the story while looking at the pictures and after the examiner finished narrating the story, they had to retell it while looking at the pictures. Casual conversation involved participants responding to standard questions about their family, school, friends, hobbies, or afternoon activities. Both speech samples were video recorded and orthographically transcribed. Disfluencies were identified and categorised in two general categories: SLDs and ODs. SLDs were considered (a) part-word repetitions (e.g., "b-but"), (b) single-syllable word repetitions (e.g., "you you you"), (c) dysrhythmic phonations: i.e., prolongations (e.g., "mmmmy" "cooookie"), blocks (e.g., "#toy"), and broken words (e.g., "o#pen"); ODs were considered (a) interjections (e.g., "um"), (b) revisions/abandoned utterances (e.g., "Mom ate/Mom fixed dinner," "I want/Hey look at that"), and (c) multisyllable word/phrase repetitions (e.g., "because because," "I want I want to go") (Ambrose & Yairi, 1999). Unintelligible utterances, isolated affirmatives or negatives were not counted and in cases where two or more SLDs occurred on the same syllable, only the first one was counted (Tumanova et al., 2014).

4.2 Data analysis

4.2.1 Inhibitory control

IC (more specifically CRI) was examined in Study I. The two terms, IC, and CRI will be used interchangeably. Two analyses of covariance (ANCOVA) were used. The mean values of response times and error percentages for each participant were set as the dependent variables. Group (CWS versus CWNS) was set as the between-subjects variable, block (compatible versus incompatible) as the within-subject repeated measures variable and age as a covariate. The mean difference in speed and/or accuracy between compatible and incompatible blocks were labelled as IC performance costs. To detect possible outliers, the distributions of the dependent variables (response times and error percentages) were checked with the criterion of ±3 standard deviations from the mean (Howell, 1998). To investigate the effect of age on the results, two-tailed correlations between age and performance costs were run as post hoc tests. Data analysis was conducted using SPSS (Version 25) and the significance level was set at $\alpha = 0.05$.

4.2.2 Cognitive flexibility

CF was examined in Studies I and II. In Study I, two pairs of ANCOVA were conducted. According to the task manual (de Sonneville, 2009), both in the ROO and the SSV task, CF is assessed by comparing mean response times and/or percentage of errors of the compatible block with the compatible trials of the mixed block. Expanding on the task manual's recommendations, for Study I, CF was assessed in two different sets of comparisons which allowed the set-shifting factor to be considered and to control for possible effects of IC. The comparisons were the following: (a) the compatible block versus the compatible trials of the mixed block with (i) no set-shifting (i.e., a compatible trial preceded a compatible trial) and (ii) set-shifting (i.e., an incompatible trial preceded a compatible trial), and (b) incompatible block versus the incompatible trials with (I) no set-shifting (i.e., an incompatible trial preceded an incompatible trial) and (II) set-shifting (i.e., a compatible trial preceded an incompatible trial). The comparisons of the mean difference in speed and/or accuracy between the blocks (or trials of the blocks) were labelled performance costs, in all cases. The dependent variables (mean value of response times and error percentages) and the between-subjects variable (group) were the same in all the analyses, similarly to those in IC. The within-subjects variable was block but with three levels: (1) compatible or incompatible block, (2) compatible or incompatible trials of the mixed block with no set-shifting, and (3) compatible or incompatible trials of the mixed block with set-shifting, depending on whether the analyses were run with the compatible or the incompatible trials.

In Study II, CF was investigated in a similar way to Study I, but a cross-sectional study was conducted in which younger CWS and CWNS were compared with older. CF was investigated by comparing speed and accuracy of the compatible block with the compatible trials of the mixed block (with no set-shifting and with set-shifting) and the incompatible block to the incompatible trials (with no set-shifting and with set-shifting) of the mixed block. A gamma generalised linear mixed model was used for investigating speed, which allows the investigation of the multilevel experimental design. In addition, it is suitable for positively skewed non-negative data, as in this case. The dependent variable was the response times (the actual and not the mean values). The model included the following: (1) fixed effects for blocks with three levels for both the analyses between the compatible and the incompatible comparisons, (2) all two- and three-way interactions between these variables, and random intercepts for each subject. A binomial generalised linear mixed model was used for investigating accuracy. The dependent dichotomous variable was error (correct versus incorrect response per trial). Similar fixed effects, interactions, and random intercepts were included in the speed analyses. The performance costs that occurred when comparing the compatible block to the mixed block (compatible trials with no set-shifting and with set-shifting) and the incompatible block to the mixed block (incompatible trials with no set-shifting and with set-shifting) were labelled mixing-costs, while the ones that occurred when comparing no set-shifting to set-shifting trials of the mixed blocks were labelled set-shifting-costs. In both studies, as with IC, to detect possible outliers, the distributions of the dependent variables (response times and error percentages) were checked with the criterion of ±3 standard deviations from the mean (Howell, 1998). Also, the data analyses were conducted using SPSS (Version 25) and the significance level was set at $\alpha = 0.05$.

4.2.3 Inhibitory control, cognitive flexibility, and speech disfluencies

While in Studies I and II speech disfluencies were analysed for determining group classification and/or stuttering severity, in Study III, the speech disfluencies produced in two combined speech samples (collected in Study I) were examined in relation to performance costs measured in Study I. To determine the coding reliability for disfluencies, 10 speech samples (five from the CWS and five from the CWNS) were evaluated by a second examiner (an ASHA-certified speech-language pathologist). The interjudge reliability score was high for all disfluencies, Krippendorff's α = .97, 95% CI [0.93, 0.99]. For IC, the performance costs were calculated as the difference of the mean values in speed and accuracy between the compatible block and the incompatible block as explained in 4.2.1. CF was operationalised based on the performance costs obtained when comparing the speed and accuracy between the compatible block and the compatible trials with set-shifting, as these were consistently the highest performance costs for both groups. The independent variables were the different IC/CF performance costs in speed and accuracy, and the dependent variables were the average percentages of SLDs and ODs. Gamma regression models with an identity link function were used to model disfluencies, as this is suitable for dependent variables with continuous, non-negative values. The model's goodness of fit was checked by examining the deviance of residuals. The data analysis was conducted with R programming language (Version 4.2.2) and the significance level was set at $\alpha = 0.05$.

5 Results

5.1 Inhibitory control (Study I)

IC was assessed by evaluating CRI in terms of response times (speed) and error percentages (accuracy). The speed analyses indicated a significant block × group interaction, $F(1, 35) = 4.53$, $p = 0.04$, partial $\eta^2 = 0.11$. The CWS exhibited higher performance costs compared to CWNS, $t(24.09) = -2.08$, $p = 0.049$, Cohen's $d = -0.67$, despite both groups being slower in the incompatible block. The effect of age was also significant $F(1, 35) = 10.75$, $p < 0.001$, partial $\eta^2 = 0.23$. Further investigation using a two-tailed Spearman's correlation revealed a negative correlation between the performance costs in speed and age for the CWS ($\rho = -0.49$, $p < 0.005$) but not for the CWNS ($\rho = -0.24$, $p = 0.30$). It is important to note though that there was substantial overlap in the 95% confidence intervals between CWS and CWNS [−0.772, −0.046] and [−0.626, 0.240], respectively. Lastly, the accuracy analyses did not reveal any significant differences between the two groups.

5.2 Cognitive flexibility (Study I and II)

In Study I, the speed comparison between the compatible block and the compatible trials of the mixed block revealed a significant block × group interaction $F(1.54, 54.02) = 5.16$, $p = 0.01$, partial $\eta^2 = 0.13$. In the comparison of the compatible block versus the compatible trials with no set-shifting, CWS demonstrated higher performance costs, i.e., they slowed down more in the mixed block compared to the CWNS, $t(21.71) = -2.23$, $p = 0.04$, Cohen's $d = -0.08$. Similarly, in the comparison between the compatible block and the compatible trials with set-shifting of the mixed block, CWS demonstrated higher performance costs compared to CWNS, $t(23.71) = -2.44$, $p = 0.02$, Cohen's $d = 0.47$. The effect of age was significant, and the two-tailed Pearson's correlation analyses revealed a negative correlation between the different performance costs and age for CWS: compatible block versus mixed block (compatible trials) $r = -0.676$, $p < 0.005$, when there was no set-shifting, and $r = -0.623$, $p = 0.01$, when there was set-shifting. No significant correlations with age were found for the CWNS group, $r = -0.232$, $p = 0.68$, when there was no set-shifting, and $\rho = -0.077$, $p = 1.00$, when there was set-shifting. There was substantial overlap of the 95% confidence intervals of the two groups for both no set-shifting, $\rho = -.077$ (CWS: [−0.87, −0.32]; CWNS: [−0.621, −0.25]) and set-shifting trials (CWS: [−0.84, −0.24]; CWNS: [−0.51, −0.39]). The accuracy analyses did not reveal any significant differences between the two groups.

In the comparison of the incompatible trials, between the incompatible block and the incompatible trials of the mixed block, there were significant group differences $F(1, 35) = 6.23$, $p = 0.02$, partial $\eta^2 = 0.15$, with CWS being slower. The block × group interaction was also significant, $F(2, 70) = 3.69$, $p = 0.03$, partial $\eta^2 = 0.10$. Further investigation of this interaction revealed that CWS were slower than CWNS in the mixed block (compatible part) for both the no set-shifting ($p = 0.01$) and the set-shifting trials ($p = 0.02$). Furthermore, there were higher performance costs for the CWS group, as they slowed down more in the mixed block (incompatible part) $t(24.18) = -2.18$, $p = 0.04$, Cohen's $d = -0.71$ when there was no set-shifting. The effect of age was significant, $F(1, 35) = 14.16$, $p < 0.001$, and the two-tailed Pearson's correlation analysis revealed a significant correlation between performance costs and age only for the CWS group, $r = -0.549$, $p = 0.03$, when there was set-shifting. There was substantial overlap of the 95% confidence intervals of the two groups for both the no set-shifting (CWS: [−0.73, −0.05]; CWNS: [−0.58, 0.30]), and the set-shifting trials (CWS: [−0.80, −0.13]; CWNS: [−0.76, 0.03]). The accuracy analyses did not reveal any significant differences between the two groups. Lastly, there were no significant results in the investigation of the correlation between stuttering severity and the various performance costs in either speed or accuracy (p values between 0.24 and 1.00).

In Study II, in the speed analyses of the compatible trials, the between-subjects effect of the classification group factor was not significant $F(1, 5{,}908) = 2.37$, $p = 0.12$ (for CWS: $M = 1{,}150$ ms, $SE = 41$ ms; for CWNS: $M = 1{,}242$ ms, $SE = 44$ ms), but the effect of age group was significant, $F(1, 5{,}908) = 13.10$, $p < 0.001$ (for younger participants: $M = 1{,}309$ ms, $SE = 45$ ms; for older participants: $M = 1{,}091$ ms, $SE = 40$ ms). Only one significant interaction occurred, between age group and block, $F(2, 5{,}908) = 34.92$, $p < 0.001$. Investigating this interaction revealed that both age groups had an increase in mixing-costs (longer response times), for both the no set-shifting and the set-shifting trials ($p < 0.001$ in all cases). Comparing also set-shifting to no set-shifting trials revealed that both age groups had longer response times in the set-shifting trials (younger participants: $t(5908) = 10.08$, $p < 0.001$; older participants: $t(5908) = 9.29$, $p < 0.001$) indicative of an increase in set-shifting-cost. Furthermore, the two age groups differed in the compatible block, $t(5908) = -6.45$, $p < 0.001$, but had comparable performance in the mixed block for both the no set-shifting and the set-shifting trials: $t(5908) = -1.70$, $p = 0.09$, $t(5908) = -1.85$, $p = 0.06$), respectively. The block × classification group × age group interaction was also investigated. It was found that CWS-O were faster in the no set-shifting trials $t(5908)$

= −2.09, $p = 0.04$, but had a similar speed to CWS-Y in the shifting trials, $t(5908) = -1.54$, $p = 0.12$. There were no differences in speed between the CWNS-O and the CWNS-Y in either the no set-shifting, $t(5908) = -0.31$, $p = 0.76$, or the set-shifting trials $t(5908) = -1.08$, $p = 0.28$. Lastly, there were significant mixing-costs for all combinations of the age group and classification group factors ($p < 0.001$ in all cases). Two-tailed unpaired *t*-test analyses revealed that the difference in performance costs between any two subgroups was not significant.

In the accuracy analyses of the compatible trials, a significant interaction was found between age group and block $F(2, 5{,}908) = 8.18$, $p < 0.001$. Further investigation of the interaction revealed that both age groups performed less accurately in the mixed block compared to the compatible block for both the no set-shifting and the set-shifting trials. In the no set-shifting trials the younger participants scored: $M = 23.2\%$, $SE = 3.4\%$, $t(5908) = -3.77$, $p < 0.001$; the older participants scored: $M = 22.2\%$, $SE = 3.5\%$, $t(5908) = -3.88$, $p < 0.001$), while in the set-shifting trials, the younger participants scored: $M = 23.7\%$, $SE = 3.4\%$, $t(5908) = -3.34$, $p < 0.005$; the older participants: $M = 20.3\%$, $SE = 3.2\%$, $t(5908) = -4.52$, $p < 0.001$), compared to the compatible block in which there was no difference between the no set-shifting and the set-shifting trials of the mixed block for either the younger participants: $M = 11.2\%$, $SE = 1.9\%$, or the older participants: $M = 5.4\%$, $SE = 1.1\%$. Additionally, the two age groups differed significantly in the compatible block $t(5908) = -2.64$, $p = 0.01$.

Investigation of the non-significant classification group × age group × block interaction revealed that all subgroups made more errors in the mixed block for both the no set-shifting and the set-shifting trials ($p < 0.001$ in all cases). CWS-O made significantly more errors in set-shifting trials compared to no set-shifting trials, $t(5908) = 2.05$, $p = 0.04$ and had a larger set-shifting-cost compared to CWNS-O $t(35) = 2.15$, $p = 0.02$. CWS-O also had a significant increase in errors when shifting from set-shifting to no set-shifting trials compared to CWS-Y, $t(35) = 1.98$, $p = 0.03$.

In the speed analyses of the incompatible trials, the classification group factor was not significant $F(1, 5{,}908) = 1.20$, $p = 0.27$ (CWS: $M = 1{,}373$ ms, $SE = 62$ ms; CWNS: $M = 1{,}473$ ms, $SE = 66$ ms), whereas the age group effect was significant $F(1, 5{,}908) = 4.47$, $p = 0.03$ (younger participants: $M = 1{,}521$ ms, $SE = 67$ ms; older participants: $M = 1{,}329$ ms, $SE = 62$ ms). Furthermore, the age group × block interaction was significant, $F(2, 5{,}908) = 24.88$, $p < 0.001$, revealing that both age groups had higher mixing-costs from the incompatible to the mixed block (incompatible trials) for both the no set-shifting and set-shifting trials ($p < 0.001$ in all cases, except for the case of comparing the incompatible block versus the mixed block no set-shifting trials for the younger participants, $p = 0.84$). Set-shifting trials compared to no set-shifting trials required longer response times for both age groups (for younger participants: $t(5908) = 7.96$, $p < 0.001$; for older participants: $t(5908) =$

7.75, $p < 0.001$). The two age groups differed in the incompatible block, $t(5908) = -4.11$, $p < 0.001$, but did not differ in the mixed block (set-shifting trials: $t(5908) = -0.91$, $p = 0.36$; no set-shifting trials: $t(5908) = -1.04$, $p = 0.30$). No other interactions were significant, except for the block × classification group × age group interaction. This investigation revealed significant mixing-costs for all combinations of the age group and classification group factors ($p < 0.001$ in all cases), except for the difference in response times between the incompatible block and the incompatible no set-shifting trials of the mixed block for the CWS-Y and the CWNS-Y subgroups. Two-tailed unpaired *t*-test analyses revealed that CWS-O had a larger set-shifting-cost than CWNS-O (slowed down more) from no set-shifting to set-shifting trials, $t(35) = 1.83$, $p = 0.04$. Similarly, the CWNS-O had larger performance costs than CWS-O from the incompatible block to the mixed block (no set-shifting trials), $t(35) = 3.17$, $p < 0.005$. All other performance costs between any subgroups were not significant.

In the accuracy analyses of comparing the incompatible trials, the only significant interaction was that between age group and block, $t(35) = 1.98$, $p = 0.03$. Both groups made more errors moving from the incompatible block to the mixed block (i.e., higher mixing-costs), in both the no set-shifting and the set-shifting trials ($p < 0.001$ in all cases, except for the mixing-cost for the CWS-Y in the no set-shifting trials, $p = 0.16$). Both age groups were less accurate in the set-shifting trials compared to the no set-shifting trials (younger participants: $t(5908) = 6.44$, $p < 0.001$; older participants: $t(5908) = 5.76$, $p < 0.001$). The two age groups differed in the incompatible block, $t(5908) = -3.67$, $p < 0.001$, but had comparable performances in the mixed block for both the no set-shifting and the set-shifting trials. Investigating the non-significant interaction (group × age group × block), revealed significant performance costs for all combinations of the age group and classification group factors ($p = 0.001$ in all cases except for the performance costs between the incompatible block and the incompatible no set-shifting trials of the mixed block for the CWS-Y, CWNS-Y, and the CWS-O subgroups). Two-tailed unpaired *t*-test analyses revealed no significant performance cost differences between any subgroups.

5.3 Association between inhibitory control, cognitive flexibility, and speech disfluencies (Study III)

In the combined speech sample used for Study III, CWS, compared to CWNS, produced significantly more SLDs ($Z = 3.823$, $p < .001$), but had a similar OD production ($Z = 1.087$, $p = .277$). Investigation of the associations between the different performance costs in IC (speed and accuracy) and disfluencies produced

(SLDs and ODs), revealed one significant interaction: group × IC performance costs in speed for the SLDs (p = .031). Additionally, in terms of CF, there was one significant interaction: group × CF performance costs in accuracy for the SLDs (p = .003). These results suggest that the effect of IC/CF performance costs on SLD production was significantly higher in the CWS compared to the CWNS group.

6 Discussion

6.1 Inhibitory control in children who stutter

IC was investigated in Study I and III. The CRI task performance measured in Study I was also used in Study III to investigate possible associations with disfluency production in the collected speech samples. CRI was elicited by comparing the average speed and accuracy results of the compatible to the incompatible block of the ROO computer task. In Study I, it was hypothesised that the CWS, compared to the CWNS, would have higher performance costs (lower performance in the incompatible block) in terms of both speed and accuracy. This hypothesis was partially supported. The results indicated higher performance costs for the CWS, but only in terms of speed and not in terms of accuracy.

The finding of reduced IC for CWS in Study I is in agreement with the two earlier studies that also investigated CRI, one in younger (3- to 6-year-old) (Anderson & Wagovich, 2017) and one in older children (6- to 9-year-old) (Eggers & Jansson-Verkasalo, 2017), both via the auditory domain. This finding is also in agreement with other earlier studies that focused on motor response inhibition in CWS and CWNS (Piispala et al., 2017, 2018), as well as with studies that used parental questionnaires to assess IC (Eggers et al., 2010; Ntourou et al., 2018). Study I is of importance not only because it is the only study to investigate CRI via the visual domain, but also because, of the inclusion of older participants, similar to the study of Eggers and Jansson-Verkasalo (2017), where the probability of natural recovery is markedly decreased (Howell & Davis, 2011). Overall, Study I provides additional evidence to the claim that CWS compared to CWNS tend to present with lower IC.

The performance costs measured in Study I were further studied in Study III and related to the different types of produced disfluencies (SLDs and ODs) in a combined speech sample of retelling a story and casual conversation. It was hypothesised that the effect of the performance costs in speed and accuracy would be higher on disfluency production (of both types), for the CWS group. This was partially supported as one significant group × IC performance costs (in speed) interaction was revealed for SLDs. This result suggests that the effect of the IC performance costs (speed) on the production of SLDs was significantly higher for the CWS group compared to the CWNS group. In other words, the increased slowing down of the

CWS group in the incompatible block was associated with the increased production of SLDs, but not ODs.

This finding contrasts with the results of the two questionnaire-based studies that found no association between reduced IC and overt stuttering symptoms (Eggers et al., 2010; Ntourou et al., 2018) but is in agreement with the reported finding of an association between increased IC and decreased overt stuttering symptoms by Harrewijn et al. (2017). Since there are no other studies that have investigated any possible associations between IC performance and the different types of speech disfluencies in CWS using experimental paradigms, the finding of Study III is also compared to those studies on AWS and non-stuttering populations. This finding is in accordance with that of Bakhtiar and Eggers (2023) who reported a correlation between IC weaknesses (in speed) and stuttering frequency in AWS. In addition, it aligns with findings of two neurocognitive studies in AWS. The first one suggests a link between the neural pathways associated with IC and stuttering severity (Neef et al., 2018), while the second one suggests that AWS have difficulties in inhibiting a planned speech response (Ning et al., 2017). When compared with studies in non-stuttering populations, Engelhardt et al. (2010) also concluded that the speech production system relies on IC and Lee et al. (2017) reported lower IC to be related to a higher incidence of SLDs.

In conclusion, findings from Studies I and III on IC illustrate that, as a group, 6- to 9-year-old CWS exhibit reduced IC compared to age- and gender-matched controls. This weakness is found to be associated with increased SLD (but not OD) production.

6.2 Cognitive flexibility in children who stutter

CF was investigated in all three studies. In Studies I and II, it was elicited by comparing the speed and accuracy results between (a) the compatible block and the compatible trials of the mixed block, and (b) the incompatible block and the incompatible trials of the mixed block (with and without set-shifting in both comparisons). In other words, two different measures of CF were used. Study I investigated CF across two classification groups (CWS and CWNS), while Study II investigated CF across two classification groups and two age groups (younger and older). For Study III, where we wanted to examine possible associations between CF and disfluency production (to that previously and similarly described for IC), we used the CF measure of Study I that yielded the highest performance costs in speed and accuracy, namely the difference between the compatible block and the compatible trials of the mixed block with set-shifting.

Studies I and II focused on comparing the performance of CWS and CWNS under CF task-conditions. Similar inclusion criteria and behavioural tasks were used, facilitating comparisons between the two studies. Study II had a larger number of participants from a wider age range. Having two age groups within each classification group allowed, to a certain extent, an investigation of CF development. In Study I, it was hypothesised that CWS compared to CWNS would be slower and less accurate under CF task-conditions. In Study II, it was hypothesised that CWS, in both younger and older groups, would be slower compared to the corresponding CWNS age group and differences would be more evident in the comparisons of the incompatible trials. The hypotheses of the two studies were partially supported and the findings were, to a certain extent, similar. Overall, as expected, and in line with the ANT literature, all participant groups were slower in the mixed block because additional time is required for task-set reconfiguration (Rogers & Monsell, 1995; Sohn & Carlson, 2000) and also due to greater uncertainty usually experienced in mixed blocks (Los, 1996).

Regarding group comparisons, as hypothesised, in both studies the CWS presented with reduced CF. In Study I, CWS compared to CWNS, slowed down more when moving from the no set-shifting to the set-shifting trials. This was also evident in Study II, but only for the older CWS and not for the younger CWS, since the performance costs of the two younger subgroups (CWS-Y and CWNS-Y) were comparable. In addition, and contrary to the result hypothesised, in Study II, CWS-O compared to the CWNS-O, were also found to be less accurate.

The finding of a longer processing time for the CWS observed in Study I and in Study II (for the CWS-O subgroup in the incompatible trial comparisons) is in agreement with the findings of Eichorn and Pirutinsky (2021) who reported longer response times under set-shifting conditions in 8- to 11-year-old CWS when compared to their age and gender-matched controls. It is also partially in agreement with the findings of Rocha et al. (2019) who found 7- to 9-year-old CWS to be slower compared to the CWNS under CF task-conditions. However, the comparable speed/accuracy results found in Study II between the younger subgroups (CWS-Y and CWNS-Y) do not align with the findings of Anderson et al. (2020) and Eichorn et al. (2018), who reported slower performance in 3- to 6-year-old CWS. It is possible though that this discrepancy occurred due to the different age groups between studies and the different CF aspects investigated. For example, in Study II, 4- to 6-year-old rather than 3- to 6-year-old children were included. Moreover, the studies by Anderson et al. (2020) and Eichorn et al. (2018) focused on investigating switching (sorting things based on different dimensions) and not set-shifting (adjusting the motoric response based on changing rules). Given that the response times measured in EF tasks are considered good indicators of processing speed (Frischkorn et al., 2019), Study I and II provide evidence that CWS, as a group, compared to CWNS, require additional processing time during set-shifting.

The additional findings of Study II, revealed that CWS-O, compared to CWNS-O, made more errors (compatible trial comparisons) when moving from no set-shifting to set-shifting trials. These findings are consistent with the findings of Eggers and Jansson-Verkasalo (2017) who reported more errors in 6- to 9-year-old CWS and also partially agree with the findings of Rocha et al. (2019) who reported 7- to 9-year-old CWS being both slower and less accurate compared to their controls. The finding of more errors in Study II suggests that older CWS, who are less likely to spontaneously recover from stuttering, may be unable to adjust their response style, i.e., to slow down to avoid errors, as previously reported for 4- to 10-year-old CWS by Eggers et al. (2013). Even though this claim is speculative, it receives support from two studies that investigated CF in CWS and CWNS. In the first one (Eichorn et al., 2018), 3- to 7-year-old CWS traded speed over accuracy, while in the latter, (Eichorn & Pirutinsky, 2021) in which 8- to 12-year-old CWS were included, no speed-accuracy trade-off was observed. Adjusting the response style to shift from speed to accuracy, or vice versus, is a strategy of pure control (Gopher et al., 2000) and was not as clearly observed in the older CWS subgroup, something required in demanding communicative situations. Another possible explanation lies in the slight differences between the tasks employed in the two studies. In Study I, the stimulus was static, whereas in Study II, the stimulus was dynamic, and each block had more trials, making the task more challenging and possibly more sensitive in revealing the differences among the different subgroups.

Study III investigated the relationship between CF task performance and the different types of produced disfluencies (SLDs and ODs) and is the first study to examine this association with both types of disfluencies in CWS. The finding of Study III showed that there was a higher effect of the performance costs in accuracy on SLD production for the CWS group, compared to the CWNS group. In other words, for the CWS group, the increase in errors during set-shifting was associated with the increase in SLD (but not OD) production. As with IC, only two studies have explored the potential association between reduced CF and overt stuttering symptoms (Eggers et al., 2010; Ntourou et al., 2018), reporting no link between the two. No other studies with preschool/school-age stuttering children or adults have investigated the relationship between CF and disfluencies. The inconsistency in findings may be attributed to the different age groups, the use of parental questionnaires versus experimental paradigms and the different paradigm types used (computerised versus noncomputerised). The findings of Study III may also be compared to the few studies conducted in non-stuttering populations. For example, in an unpublished study, Hart et al. (2021) investigated CF in 3- to 6-year-old children and reported a correlation between CF (teacher-questionnaire) results and revisions. Furthermore, Dayalu et al. (2022) reported an inverse relationship between

CF task results and disfluencies/articulation errors in adults diagnosed with Parkinson's disease.

In conclusion, the findings from Studies I, II, and III on CF, illustrate that 4- to 6-year-old CWS exhibit comparable CF with their age-matched controls, while 6- to 9- and 7- to 9-year-old CWS present reduced CF, suggesting that CF weaknesses may play a role in stuttering appearance and/or persistence. Lastly, the identified association between reduced CF and increased SLD production for the 6- to 9-year-old CWS group illustrates that reduced CF contributes to SLD but not OD production.

6.3 Theoretical and clinical implications

In this thesis, we observed reduced IC and CF in 6- to 9- and 7- to 9-year-old CWS when compared to their age- and gender-matched CWNS. Further investigation revealed that the reduced IC/CF in the 6- to 9-year-old CWS was associated with SLD (but not OD) production. Moreover, there were no differences in CF between 4- to 6-year-old CWS and CWNS. This finding illustrates that reduced CF is present in older CWS, who are less likely to undergo spontaneous recovery.

Both IC and CF play crucial roles in modulating and regulating cognitive and behavioural responses efficiently. The findings of this thesis may be linked to the covert repair hypothesis (Kolk, 1991; Postma & Kolk, 1993; Postma et al., 1990) and the vicious circle hypothesis (Vasić & Wijnen, 2005), both aetiological models of stuttering, specifically highlighting monitoring issues in PWS. In the covert repair hypothesis, it is proposed that PWS demonstrate slower phonological processing systems (Anderson et al., 2022), leading to vulnerabilities in their phonetic plans. This vulnerability creates opportunities for covert self-repairs, subsequently hindering fluent speech production (i.e., SLD and/or OD production). Deficiencies in IC may lead to less robust phonological representations of words in the mental lexicon (Anderson & Wagovich, 2017; Ofoe et al., 2018). The identified association between performance costs (speed) and SLD production in the CWS group may be attributed to the increased slowing down, preventing the language production system from having sufficient time to withhold the detected error before articulation. Moreover, deficiencies in CF may impede the capacity to adapt to new speech plans, as observed in instances where CWS demonstrated a slowing down when confronted with such challenges (Anderson et al., 2020), a pattern that also emerged in this thesis. In the vicious circle hypothesis, it is speculated that the internal monitoring systems of PWS are hypervigilant, resulting in unnecessary repairs during the articulatory phase. Deficiencies in IC may impede the ability to halt these unwarranted repairs, thereby exacerbating disfluencies (Vasić & Wijnen, 2005). Similarly, deficiencies in CF could hinder the adjustment of the articulatory plan prior to the production phase, again leading to disfluencies. One of the predictions of the vicious circle hypothesis is that disfluencies are related with the increased focus and awareness to speech production (Vasić & Wijnen, 2005). A key finding of this thesis is that older CWS (7- to 9-year-olds), who are less likely to spontaneously recover from developmental stuttering, exhibit reduced CF compared to their younger controls (4- to 6-year-olds). It is possible that the reduced CF interferes with the older CWS subgroup's ability to shift their focus away from a subcomponent of speech production, thus impacting their ability to produce fluent speech.

The findings of this thesis may also be linked to the executive function model (Anderson & Ofoe, 2019), since an association was found between reduced IC/CF and SLD production. This model posits a bidirectional relationship between the domain-general processes of IC and CF and domain-specific processes (i.e., emotion, sensory, motor, and language). Deficits in IC/CF lead to deficits in domain-specific processes, which, in turn, contribute to deficits in IC/CF. Lastly, the findings may also be linked to a more recent theoretical account proposed by Usler (2022). He proposed that cognitive processes, such as IC and CF, may play a role in the onset and persistence of developmental stuttering and that SLDs could result from heightened cognitive conflict and control during speech.

IC is recognised as a fundamental aspect of self-regulation (Gärtner et al., 2018), with a substantial overlap between IC and self-regulation (Diamond, 2013). Additionally, CF plays a crucial role in managing negative thoughts and disengaging from emotional aspects within a given situation (Gabrys et al., 2018). Both IC and CF are vital for developing and maintaining strategies to cope with maladaptive behaviour, including inner speech, which facilitates self-regulation (Miyake et al., 2004). This is particularly important because maladaptive thought processes can lead to negative biases and contribute to social difficulties (Burton et al., 2022).

Formulating possible clinical suggestions might seem premature, given that CWS experiencing weaknesses in IC and CF may struggle with managing emotional arousal and negative thoughts, particularly in demanding communicative situations; however, stuttering interventions could include parental counselling that aims to improve their children's self-regulation and building a supportive communicative environment, as predictable environments can promote the development of self-regulation (Carlson, 2003). There are some existing programmes for preschoolers counselling parents on this topic (de Sonneville-Koedoot et al., 2015; Kelman & Nicholas, 2020). These programmes advise parents to incorporate such behaviours that aid the improvement of their child's self-regulation and the reduction of environmental barriers to effective communication. For example, they recommend enhancing turn-taking behaviours by allowing ample time for the child to talk, providing advance notice of upcoming changes, and implementing structures in conflict situations.

For older (school-aged) children, several authors have described therapeutic techniques, in addition to parental counselling, aiming at managing cognitive, affective and behavioural challenges, associated with developmental stuttering (e.g., Boyle, 2011; Harley, 2018; Kelman et al., 2022). For instance, the proposed training of CWS involves increasing awareness of bodily sensations, altering their relationship with thoughts, and acquiring the ability to pause and be mindful instead of reacting impulsively during moments of stuttering. These techniques not only enhance IC, CF skills, and self-regulation (Moore & Malinowski, 2009; Oberle et al., 2012), but also contribute to regulating ruminating thinking and addressing social anxiety (Goldin et al., 2009; Ramel et al., 2004), which is something that PWS may exhibit higher levels of, compared to the general population (Craig et al., 2003; Craig & Tran, 2014).

6.4 Limitations and future research

The potential limitations of this thesis include a male-to-female ratio (18, 1) in Studies I and III, which was higher than typically reported for the investigated age group (Yairi & Ambrose, 2013), thus, potentially limiting the generalisability of these findings to girls who stutter. While the absence of standardised language testing for Greek-speaking children may initially seem like a limitation in Studies I and III, the children's language abilities were assessed using micro- and macro-analyses of speech samples produced during the Bus Story Test (Renfrew, 2010). Despite being unstandardised in Greek, this test is widely used in studies with Greek-speaking school-aged children (e.g., Kambanaros & Grohmann, 2013; Theodorou et al., 2016). Additionally, the use of slightly different inclusion criteria between Studies I and II could be seen as a limitation. However, when children who scored very mildly on the SSI-3 were excluded from the CWS group in Study II, the results remained consistent. Importantly, across all studies, only children for whom there was parental concern about stuttering and had been diagnosed with stuttering by a speech-language pathologist were included. Lastly, no formal assessment of ADHD was conducted. However, there were no reported concerns from parents, teachers, or speech-language pathologists regarding ADHD for any of the participants in the three studies.

To establish the generalisability of the results that suggest a link between reduced IC/CF and increased SLD production, future research may consider studies which meet the typically reported male-to-female ratio. Furthermore, it would be beneficial to conduct cross-sectional (or longitudinal studies), similar to Study II, to facilitate comparisons between different age and classification groups.

7 Conclusions

This thesis examined IC in 6- to 9-year-old and CF in two distinct datasets encompassing both 6- to 9-year-old and 4- to 9- year-old CWS and CWNS. In addition, speech samples, collected exclusively from the 6- to 9-year-olds, were analysed to explore potential associations between IC/CF performance costs and disfluency production. The results revealed lower task performance (higher performance costs) for the CWS, suggesting that, as a group, they tend to exhibit lower IC/CF proficiency compared to CWNS. In a more in-depth examination of CF in two age groups (4- to 6-year-olds and 7- to 9-year-olds), older CWS were both slower and made more errors compared to their controls. This allows for the speculation that CF weaknesses contribute to the development and/or persistence of developmental stuttering. Furthermore, both IC and CF performance costs were associated with SLDs (but not ODs) in the 6- to 9-year-old CWS group, indicating a potential contributing role of IC/CF weaknesses in SLD production. In summary, the findings of this thesis suggest that CWS exhibit weaknesses in IC and CF, highlighting the importance of considering these factors as potential contributors in the development and/or persistence of developmental stuttering.

Abbreviations

ANCOVA	analyses of covariance
ANT	Amsterdam neuropsychological tasks
AWS	adults who stutter
CF	cognitive flexibility
CRI	complex response inhibition
CWS	children who stutter
CWNS	children who do not stutter
EFs	executive functions
IC	inhibitory control
ODs	other disfluencies
PWS	people who stutter
ROO	response organization objects
SLDs	stuttering-like disfluencies
SSI-3	stuttering severity instrument 3rd edition
SSI-4	stuttering severity instrument 4th edition
SSV	shifting attentional set-visual

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