Brigitte Vugs Executive functions in children with SLI: a dynamic perspective



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Executive functions in children with SLI: a dynamic perspective

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Chapter 1

General Introduction



In this thesis we focused on the role of executive functions (EFs) in children with specific language impairment (SLI). EFs are interesting to consider in children with SLI because of the complex interplay between this neurocognitive function and language development. It is well established that both EFs and language significantly change as children mature and a reciprocal relationship is often presumed. The cumulating evidence of EF deficits in children with SLI over the past years, increased the interest in the role of EFs in this group of children. This thesis not only addressed possible EF impairments in children with SLI, but also focused on developmental aspects of the associations between EFs and language as well as the possibility to improve EFs by training.

Development of executive functions

EFs are cognitive processes responsible for purposeful, goal directed behaviour. EFs are implicated in not only cognitive processes but also emotional responses and behavioural actions (Barkley, 2012; Gioia et al., 2001; Miyake & Shah, 1999). Conceptualizations of EFs vary from a unitary construct (Brydges et al., 2012) to a set of independent components (Garavan, 2002). Most recent models consider EFs a multifaceted concept with distinct subfunctions that are inter-related and function together as an integrated, supervisory control system (Miyake & Shah, 1999; Miyake et al., 2000; Stuss & Alexander, 2000). Although some uncertainties remain about the exact components of EFs, the three most frequently postulated components are working memory (WM), inhibition and cognitive flexibility (Huizinga et al., 2006; Miyake et al., 2000).

WM refers to the structures and processes used to temporarily store and manipulate information. The most frequently adopted theoretical framework in experimental studies on children with SLI is the multicomponent WM model of Baddeley (Baddeley & Hitch, 1974; Baddeley, 2003). In this model, a central executive (CE) system is assumed to be linked to three subsystems: the phonological loop, the visuospatial sketchpad, and the episodic buffer. The CE is responsible for the coordination and control of activities in WM. This system has limited attentional capacity and thus requires attentional control. The phonological loop and visuospatial sketchpad are so-called "slave" systems and responsible for the temporary storage of verbal and visuospatial information, respectively. The episodic buffer is a relatively recent addition to the model and is assumed a multidimensional representational system that binds information from different sources together to form chunks of information for further processing (Baddeley, 2003). The storage components of WM can be evaluated by simple storage tasks that require the serial recall of information. Verbal storage tasks for instance require the retention of words, digits, or letters, whereas visuospatial versions involve visual patterns or figures. The CE components of WM are typically assessed using complex memory span tasks that require significant processing activity in addition to storage. Verbal CE tasks combine the storage of verbal information with simultaneous processing of information, while

in visuospatial CE tasks processing activity is combined with the storage of visuospatial information.

Inhibition refers to the processes related to the control of attention and the ability to stop ongoing responses (Miyake et al., 2000). A distinction is often made between response inhibition and interference control (Friedman & Miyake, 2004). Response inhibition is the suppression of ongoing dominant or automatic responses and is often measured in stop-signal or go/no-go tasks requiring to stop or withhold a response (Berlin & Bohlin, 2002; Logan, 1994). Interference control refers to the resistance of distracting stimuli, which can be measured in tasks requiring children to respond to a target stimulus while ignoring distracting information as in the Flanker test (Eriksen & Eriksen, 1994). Cognitive flexibility, often also described as shifting, has been conceptualized as the ability to switch the focus of attention between activities or problem-solving strategies (Miyake et al., 2000). It is often measured in tasks that require children to shift between one or more different rules, such as the Trailmaking Test and the Wisconsin Card Sorting Test (Heaton et al., 1993).

A general issue in the field of EF research is the question of the ecological validity of the EF tasks that are used. Standardized cognitive measures of EFs have been criticized as not being sufficiently sensitive to the multidimensional nature of EFs in daily life (Chaytor et al., 2006; Anderson et al., 2002). Based on this, it is thus suggested that information should be collected in different contexts and from different sources using behavioural ratings of EFs (Gioia et al., 2001). For this purpose, Gioia and colleagues (2000) developed the Behaviour Rating Inventory of Executive Function (BRIEF). Research using the BRIEF has shown that it is a valid and reliable measure of everyday EFs (Mahone et al., 2002).

The development of EFs is a protracted process which already starts during the first years of life and extends into early adulthood. Different components of EFs show different developmental trajectories related to the neurophysiological developments of the growing brain. The ability to keep simple information in mind (i.e., WM) is already present around the age of 6 months and the development of WM undergoes enormous neurodevelopmental changes between 3 and 6 years of age (Courage & Cowan, 2009; Garon, 2008; Luciana & Nelson, 1998). Alloway and colleagues (2006) have shown a three factor model with independent verbal and visuospatial factors but a single, domain general, WM factor to provide the best account of WM in TD children between the ages of 4 and 11 years. All of the components of this model correspond to the components of the model of WM advanced by Baddeley & Hitch (1974). The components are assumed to be in place by the age of 4, and the model has been found to be quite stable up until the age of 11 years. With regard to inhibition, children generally show rapid early improvement in preschool years. There is a spurt in performance on inhibition tasks between 3 and 5 years of age, followed by more modest, linear improvements through adolescence (Best & Miller, 2010; De Luca & Leventer, 2008). The development of shifting also starts during the preschool period with children being able to shift between simple task sets. The ability to handle unexpected shifts between increasingly complex task sets develops later, with a development through adolescence (Best & Miller, 2010). During adolescence the different EF brain systems become better integrated and at age 20 to 29, EF skills are at their peak (De Luca & Leventer, 2008).

Executive functions in children with SLI

The acquisition of language is a major milestone in the development of children. While the language of the majority of children develops more or less automatically, there are also children who show marked problems and delays. When children encounter problems that can be characterized as a failure to make normal progress in language acquisition without further evidence of underlying intellectual, frank neurological, social, or emotional impairments, a diagnosis of SLI is usually made (Bishop, 2002, 2006). The prevalence of SLI is 3-6% in school-aged children (Hulme & Snowling, 2009). Children with SLI form a heterogeneous group with different profiles of language deficits. SLI can affect various linguistic domains (i.e., phonological, morphological, lexical and grammatical domains) and the language profile often changes with age and development (Bishop, 2006; Leonard, 1998). It is a persistent disorder that affects language abilities in childhood and adolescence, or even into adulthood (Brizzolara et al., 2011; McKinley & Larson, 1989). Children with SLI are also at risk for less successful academic outcomes as well as behavioural, emotional, and social difficulties (Conti-Ramsden et al., 2009; St Clair et al., 2011).

Different theories and hypotheses have been proposed to explain the underlying causes of SLI. Evidence exists for multiple determinants based on genetic and environmental factors (Bishop, 2003). Over the past years, the role of non-linguistic factors in SLI has been of increasing interest. Evidence that children with SLI have impairments in nonlinguistic factors that are not restricted to language, resulted in domain general accounts of the disorder. One factor that has been often implicated in this light is EFs. Several studies provided evidence of EF deficits in children with SLI (Archibald & Gathercole, 2006b; Im-Bolter et al., 2006; Lum et al., 2011; Marton et al., 2007). Furthermore, findings from neuroimaging studies in children with SLI showed anomalies in frontal and cingulate brain areas normally related to EFs (Dibbets et al., 2006; Jernigan et al., 1991). Recent neurobiological models of the architecture of language processing in general support the assumption that EFs are involved in linguistic processes. In the Memory-Unification-Control (MUC) model, for instance, it is assumed that language is subserved by dynamic networks of brain regions, including regions in the dorsolateral prefrontal cortex and anterior cingulate cortex which are responsible for attentional or executive control (Hagoort, 2016). These general control networks are supposed to be linked to brain regions of the core components of the language network in the temporal and frontal cortex.

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Not all components of EFs have been equally extensively studied in children with SLI and in some cases results are still somewhat contradictory. Strong links have especially been found between WM limitations and SLI (Archibald & Gathercole, 2006a; Bishop, 2006; Montgomery et al., 2010). A widely accepted account of the deficits associated with SLI, for example, is the phonological storage deficit hypothesis and the underlying assumption that a specific deficit in the temporary storage of novel phonological information underlies SLI (Archibald & Gathercole, 2007; Baddeley, 2003; Bishop, 1996; Coady & Evans, 2008; Gathercole & Baddeley, 1990). Significant group differences have been reported between children with SLI versus TD children on tasks of non-word repetition, recall of words, and recall of digits (Archibald and Gathercole, 2006b; Gray, 2003, 2006; Conti-Ramsden, 2003). In addition to these constraints on verbal storage, substantial deficits have been reported for verbal CE. Children with SLI are even more severely and consistently impaired on verbal complex memory tasks than on straightforward verbal storage tasks (Archibald & Gathercole, 2006a, 2006c; Ellis Weismer et al., 1999; Marton & Schwartz, 2003). Visuospatial WM has been less extensively investigated with somewhat contradictory results. There is as yet no consensus regarding the role of visuospatial WM in the speech and language of children with SLI. Based on studies showing children with SLI and their TD peers to perform similarly on visuospatial storage and CE tasks, several authors assume that the WM deficits of children with SLI are limited to the verbal domain (Alloway & Archibald, 2008; Archibald & Gathercole, 2006a, 2006b; Baird et al., 2009; Lum et al., 2011; Riccio et al., 2007; Williams et al., 2000). In contrast, the results of other studies have yielded evidence suggesting that the WM deficits of children with SLI may extend to the visuospatial domain (Akshoomoff et al., 2006; Bavin et al., 2005; Henry et al., 2011; Hick et al., 2005; Hoffman & Gillam, 2004; Karasinski & Ellis Weismer, 2011; Marton & Schwartz, 2003; Menezes et al., 2007; Nickisch & von Kries, 2009).

With regard to inhibition, significant group differences have been reported for children with SLI versus TD children on several tasks of response inhibition, such as go/no-go and stop-signal tasks (Bishop & Norbury, 2005; Dodwell & Bavin, 2008; Marton et al., 2007; Marton et al., 2012; Spaulding, 2010). Interference control has only been examined in a limited number of studies, showing weaker resistance of distractors in children with SLI compared to their TD peers (Marton et al., 2012; Spaulding, 2010). No group differences were found between children with SLI and their TD peers on tasks of cognitive flexibility, involving set-shifting tasks and the Trailmaking Test (Dibbets et al., 2006; Henry et al., 2012; Im-Bolter et al., 2006). However, when the cognitive flexibility tasks involved more complex stimuli such as in the Wisconsin Card Sorting Task, findings varied (Henry et al., 2012; Marton et al, 2008).

To date, research with behavioural ratings of EFs in children with SLI has been limited. Hughes and colleagues (2009) compared the parental and self-ratings of EFs for adolescents with SLI versus TD adolescents, and found more negative ratings of EFs for the SLI group with half of the parents rating their child's EF abilities in the clinically impaired range. More recently, Wittke and colleagues (2013) studied executive functioning of preschool children with SLI. The results showed that the EFs of children with SLI, aged 3 to 5 years, were rated significantly worse than those of their TD peers by both parents and teachers.

Although recent research comparing EFs between children with and without SLI showed EF deficits in children with SLI, much is still unknown about the exact associations between EFs and language in these children. Language is a complex system with different levels of processing and it is well known that various of these language abilities can be affected in children with SLI. Research in this area is still scarce. However, some studies focused on the possible associations between one of the EFs, namely WM, and language abilities in children with SLI. The verbal storage component of WM has often been proposed to be linked to word learning or vocabulary acquisition (Montgomery et al., 2010). Baddeley and Gathercole (1989) were the first to demonstrate a strong association between the functioning of the phonological loop and the acquisition of new words in TD children between 4 and 5 years. In this longitudinal study, verbal storage at the age of 4 years was significantly linked with vocabulary knowledge one year later. Since then, other studies have documented a similar association for children with SLI (Gathercole & Baddeley, 1990; Horohov & Oetting, 2004; Montgomery, 2002). It is assumed that the storage of phonological information in WM and word learning are especially linked in the early stages of vocabulary acquisition (Archibald, 2016).

Children with SLI also tend to have problems in the understanding and production of complex syntactic sentences (van der Lely, 1996; Fortunato-Travares et al., 2015). Accumulating evidence indicates that impairments in the verbal CE component of WM may account for these deficits in sentence processing (Archibald 2016; Montgomery & Evans, 2009; Noonan et al., 2014). In one study of sentence comprehension, for example, performance on a verbal CE task correlated significantly with the comprehension of complex sentences in school-aged children with SLI (Montgomery & Evans, 2009). Fortunato-Travares and colleagues (2015) investigated the association between WM and sentence comprehension through direct manipulation of WM demands, showing an effect of WM on the syntactic assignment of predicates and reflexives in sentence comprehension in children with SLI. Recently, some authors suggested that the role of verbal CE in sentence processing is influenced by the task requirements. Noonan, Redmond, and Archibald (2014) investigated the interrelations between WM deficits — measured using both verbal and visuospatial complex memory span tasks — and judgments of grammaticality. In this study, children with only language impairments and thus no WM deficits, children with deficits in both domains, and TD children completed a task in which grammatical markers occurred at different places in the sentence. Children with only language impairments performed significantly worse regardless of the location of

the marker, while children with deficits in both WM and language were only impaired for sentences with late grammatical errors, which are supposed to impose a greater WM load. Frizelle and Fletcher (2015) found that the ability to process complex sentences involving greater syntactic development was related to the verbal CE component of WM in children with SLI but not TD children. Based on these findings, it is suggested that verbal WM skills are closely linked to sentence processing when language demands are high, which is often the case for children with SLI (Archibald, 2016).

Executive function training in children with SLI

There has been much interest in recent research concerning the possibility to improve EFs by cognitive training. The underlying assumption for such interventions is that the maturation and/or efficiency of the neural circuitries underlying the trained EFs can be improved by intensive practice and training. Several novel, computer-based training programs have demonstrated promise in children and adolescents. To date, most studies particularly focused on the training of WM. Significant improvement on at least one trained WM task has been reported in several studies and meta-analytic reviews (Holmes et al., 2010; Gray et al., 2012; Green et al., 2012; Melby-Lervag & Hulme, 2012; Rapport et al., 2013; Shipstead et al., 2012). Based on this, it is generally accepted that WM training leads to positive effects on tasks closely related to the trained tasks, so called near-transfer effects. Furthermore, in a recent meta-analytic review it was found that the training effects on visuospatial WM tasks are maintained at follow-up, on average 5 months after the training (Melby-Lervag & Hulme, 2012). However, some controversy exists about the generalizability or far-transfer of the training effects on functions not closely related to trained tasks, such as other neurocognitive functions, behaviour and academic performance (Melby-Lervag & Hulme, 2012; Shipstead et al., 2012). Based on results of reviews documenting limited or negligible far-transfer effects, increasing concerns are expressed about the generalization of the trained task effects in WM training (Chacko et al., 2013; Melby-Lervag & Hulme, 2012; Rapport et al., 2013; Redick et al., 2015; Shipstead et al., 2012). In contrast, a recent systematic review and meta-analysis showed persistent training effects for inattention in daily life for children with ADHD (Spencer & Klingberg, 2015). Fewer studies have examined the trainability of inhibition and cognitive flexibility. A study of cognitive training of inhibition in TD children showed significant improvement on most of the trained tasks, but no generalization of the effect to tasks measuring WM or attention (Thorell et al., 2009). A study on the trainability of cognitive flexibility in children, adolescents and adults, showed significant improvement on cognitive flexibility tasks, and also on other EFs, including WM and interference control. White and Shah (2006) reported that adults with ADHD showed significantly improvement on both trained and non-trained tasks after task-switching training.

EF training in children with SLI has so far only received scarce attention. Wener and Archibald (2011) examined in a small scale study the effects of a treatment targeting verbal and visuospatial strategies (including a verbal CE task) in children with SLI, children with WM impairments and children with comorbid language and WM impairments. Five of the seven children with language impairments (either with or without WM impairments) showed improvement on a grammatical task after treatment and at 4-months follow-up. More recently, Holmes and colleagues (2015) investigated whether WM training could be effective in enhancing WM in a group of 12 children with low language abilities, aged 8-11 years, and 15 matched TD children. Both groups showed significant post-training gains on visuospatial storage. Further exploratory analyses revealed some predictive links between pre-training scores and training outcomes. First, visuospatial WM improved to the greatest extend following training for children with higher verbal abilities. Furthermore, children with the lowest verbal IQs at baseline made the greatest gains in verbal storage after training.

Aim and outline of the thesis

The aim of this thesis was to examine the role of EFs in children with SLI. The presented studies were conducted at Royal Dutch Kentalis, a national organization in the Netherlands specialized for persons who are deaf, hard-of-hearing or deafblind, and who have SLI. Royal Dutch Kentalis offers day-care and residential care (assessment, therapy, adapted living and work) and education to children, adolescents and young adults. Participants were recruited from special language units for preschool children with SLI, speech and language centers for school-aged children with severe language problems, and special education schools. For all these children the diagnosis of SLI was based on extensive clinical and psychometric assessment by speech and language pathologists.

This thesis not simply investigates impairments in EFs in children with SLI, but also takes into account more dynamic aspects of the relationship between EFs and SLI, like development and trainability. The thesis starts with a meta-analysis and two descriptive studies directed at impairments in EFs in children with SLI. The studies particularly focus on some topics that are underexposed in previous research, namely visuospatial WM, EF behaviours and EFs in young children with SLI. With regard to visuospatial WM, the results of former studies are highly contradictory and there is as yet no consensus on the role of visuospatial WM in children with SLI. EF behaviours were examined, because studies using rating scales in children with SLI have so far been very limited and it has been advocated to gain information on the impact of EFs in daily life besides information from cognitive tasks. EFs in preschool children with SLI also received scarce attention. As early childhood is an important period for both the development of EFs and various linguistic abilities, especially this period might be promising to examine the role of EFs in children with SLI. The fourth study not only compares performances between children with and

without SLI, but specifically addresses the associations between the components of WM and the linguistic abilities of children with SLI. Further, the development of WM in relation to language was taken into account in a longitudinal study. Previous studies mainly were cross-sectional and did not take into account developmental aspects. Longitudinal research is however needed to provide more information about the complex interplay between WM and language in children with SLI. The two final studies focus on the trainability of EFs. Given the evidence that EFs are in some way involved in SLI, it obviously will be of interest to find out whether it is possible to improve EFs in children with SLI and whether this has a positive effect on their language abilities. The research questions addressed in this thesis were as follows:

- 1. Do children with SLI show impairments in EFs?
- 2. How do the different components of WM relate to the language abilities of children with SLI?
- 3. Do impairments in EFs and its associations to language abilities change during development in children with SLI?
- 4. Is it possible to improve EFs by intensive training and does this have a positive effect on language abilities in children with SLI?

Chapter 2 focuses on the role of visuospatial WM in children with SLI. In this chapter the results are presented of meta-analyses and moderator analyses examining the magnitude of the deficits in visuospatial storage and CE, and their relation to the inclusion criteria used for SLI and the age of the children.

Chapter 3 describes a study investigating the behavioural ratings of EFs in a sample of 237 children with SLI aged 5- to 12-years. Age and sex differences were examined and behavioural ratings of EFs were related to performance on EFs tasks.

In **chapter 4**, the results are presented of a study comparing the performances of young children with SLI to that of TD children aged 4- to 5-years on measures of WM and behavioural ratings of EFs. Correlations between performances on the different components of WM and behavioural ratings of EFs were examined.

Chapter 5 describes the results of a study examining the interactions between WM and language abilities of children with SLI aged 4- to 5-years. We first examined the underlying structure of WM in children with SLI and compared it to the underlying structure of WM in TD children. Second, we examined how the different components of WM relate to the language abilities of young children with SLI.

In **chapter 6**, the results are presented of a longitudinal study investigating the developmental course of WM in children with SLI between the age of 4- to 5-years and 7- to 8-years. Further is was explored to what extent language at age 7- to 8-years can be predicted by measures of language and/or WM at age 4- to 5-years. **Chapter 7** describes the results of a pilot study on the effects of a computer-based EF training including training tasks of visuospatial WM, inhibition and cognitive flexibility (Braingame Brian) in children with SLI. Training effects were examined on tasks of the three trained EFs, tasks of other neurocognitive functions and behavioural ratings.

Chapter 8 reports a randomized controlled study on the effectiveness of the EF training Braingame Brian in children with SLI. The performances of children in the EF training group were compared to the performances of a Wait-list group. It was examined whether the training program resulted in significant near-transfer effects on tasks of the trained EFs (visuospatial WM, inhibition and verbal WM) and far-transfer effects on other untrained EFs, attention, and ratings of EF behaviours in children with SLI. Further, training effects on receptive and expressive language abilities were examined.

Chapter 9 provides a summary of the study results and an overview of the main conclusions of this thesis. Implications for clinical practice and future research are discussed.

Chapter 2

Visuospatial working memory in SLI: A meta-analysis

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ABSTRACT

We conducted a meta-analysis of the data from studies comparing visuospatial working memory (WM) in children with specific language impairment (SLI) and typically developing (TD) children. The effect sizes of 21 studies (including 32 visuospatial storage tasks and 9 visuospatial central executive (CE) tasks) were identified via computerized database searches and the reference sections of the identified studies. Meta-analyses and moderator analyses were conducted to examine the magnitude of the differences in visuospatial storage and CE, and their relation to the inclusion criteria used for SLI and the age of the children. The results showed significant effect sizes for visuospatial storage (d = 0.49) and visuospatial CE (d = 0.63), indicating deficits in both components of visuospatial WM in children with SLI. The moderator analyses showed that greater impairment in visuospatial storage was associated with more pervasive language impairment, whereas age was not significant associated with visuospatial WM. The finding of deficits in visuospatial WM suggests domain-general impairments in children with SLI. It raises questions about the language-specificity of a diagnosis of SLI. Careful attention should thus be paid to both verbal and visuospatial WM in clinical practice, and especially in those children with pervasive language impairments.

INTRODUCTION

There is growing evidence that non-linguistic factors may contribute to the problems associated with specific language impairment (SLI) and that the impairment may therefore not be exclusively linguistic (Bishop, 2006). One factor that has been implicated is working memory (WM) (Archibald & Gathercole, 2006a; Bishop, 2006; Montgomery et al., 2010). Many studies have focused on the verbal domain of children's WM. Results regarding the visuospatial domain are ambiguous at best (Alloway & Archibald, 2008; Archibald & Gathercole, 2006a; Montgomery et al., 2010). However, if children with SLI also exhibit deficits in visuospatial WM, this would implicate more general limitations, thus challenging the specificity of SLI. In the present study, we therefore conducted a meta-analysis of the results of studies that have specifically compared visuospatial WM of children with SLI and their typically developing (TD) peers.

SLI and working memory

While the language of the majority of children develops more or less automatically, there are also children who show marked problems and delays. When the problems of the children can be characterized as a selective failure to make normal language acquisition progress without further evidence of underlying intellectual, neurological, social, or emotional impairments, then a diagnosis of specific language impairment (SLI) is usually made (Bishop, 2002, 2006). This impairment affects different aspects of the children's language including phonological, morphological, lexical and grammatical aspects. In many children, moreover, the linguistic profile can change over time (i.e., with age and development); changes can then be seen to occur both within and across the different aspects of language (Bishop, 2006; Leonard, 1998).

WM refers to the structures and processes used to temporarily store and manipulate information. WM can be conceptualized somewhat differently (Courage & Cowan, 2009; Engle et al., 1999; Miyake & Shah, 1999), but the most frequently adopted conceptualization for research on the WM of children with SLI to date has been the multicomponent model of Baddeley (Baddeley & Hitch, 1974; Baddeley, 2003; 2012). In this model, a central executive (CE) system is assumed to be linked to three subsystems: a phonological loop, a visuospatial sketchpad and an episodic buffer. The phonological loop and visuospatial sketchpad are so-called "slave" systems and responsible for the temporary storage of verbal and visuospatial information, respectively. The episodic buffer is a relatively recent addition to the model and assumed to entail a multidimensional representational system that binds information from different sources together to form chunks of information for further processing (Baddeley, 2003). The CE system coordinates and controls the activities of the subsystems. It has limited attentional capacity and thus requires "attentional control."

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Engle et al. (1999) have previously suggested that WM capacity is limited by the ability to control attention and that the ability to control attention might, in fact, entirely explain the individual differences observed in WM capacity. In the Embedded-Processes model of Courage and Cowan (2009), moreover, WM is assumed to reflect the activation of information from long-term memory that is in the focus of attention. Both these views are in line with the Baddeley's model (2003, 2012) in which attentional control is part of the CE system, but focus more specifically on the executive and attentional aspects of WM.

Findings from different studies show strong links between WM limitations and SLI (Archibald & Gathercole, 2006a; Bishop, 2006; Montgomery et al., 2010). Increasing evidence indicates that the WM problems exhibited by children with SLI are diverse and may therefore involve different components of the WM system (i.e., storage and CE) (Montgomery et al., 2010). The storage component of WM can be evaluated by tasks that require serial recall of information. Verbal versions involve the retention of words, digits or letters, whereas visuospatial versions involve visual patterns or figures. The CE component is generally evaluated by tasks that require significant processing activity in addition to storage, typically using complex memory span tasks. An commonly used example of a verbal complex memory span task is listening span, in which children have to make a judgment about the meaning of each of a series of sentences, and additionally have to remember the last word of each sentence in sequence.

Most studies of the problems in different components of WM exhibited by children with SLI have focused on the verbal domain. A widely accepted account of the deficits associated with SLI, for example, is the phonological storage deficit hypothesis and the underlying assumption that verbal storage limitations lead to SLI (Archibald & Gathercole, 2007; Baddeley, 2003; Bishop, 1996; Coady & Evans, 2008; Gathercole & Baddeley, 1990). Much of the relevant evidence comes from studies of nonword repetition (i.e., the repetition of unfamiliar or nonexistent words that thus require phonological processing on the part of the child). A 16q chromosomal abnormality has even been linked to such poor nonword repetition in children with SLI and led to the suggestion that this specific verbal storage limitation might be a phenotypic marker of SLI (SLI consortium, 2004). In addition to these constraints on verbal storage capacity, substantial deficits have been reported for verbal CE. Children with SLI are even more severely and consistently impaired on verbal complex memory tasks than on straightforward verbal storage tasks (Archibald & Gathercole, 2006a, 2006c; Ellis Weismer et al., 1999; Marton & Schwartz, 2003).

In contrast to the findings for deficits in the verbal domain of WM, however, the results regarding the visuospatial domain are much less consistent. There is as yet no consensus regarding the role of visuospatial WM in the speech and language of children with SLI, for example, but several authors continue to assume that the WM deficits of such chil-

dren are limited to verbal WM. This is because children with SLI and their TD peers have been found to perform similarly on visuospatial WM tasks. Age-appropriate visuospatial WM performance among children with SLI has been found, for instance, on visuospatial storage tasks involving the immediate recall of spatial position or a sequence of visual stimuli but also on visuospatial complex memory tasks (spatial span tasks) (Alloway & Archibald, 2008; Archibald & Gathercole, 2006a, 2006b; Baird et al., 2009; Lum et al., 2011; Riccio et al., 2007; Williams et al., 2000). However, in the study by Archibald and Gathercole (2006a), the scores on the visuospatial storage task for the SLI group as a whole fell within the average range but, when the scores of the individual children were examined, 50% fell outside the average range and thus showed visuospatial storage deficits. In contrast, the results of several other studies have yielded evidence suggesting that the WM deficit observed so frequently in children with SLI may extend to the visuospatial domain of the WM system. Significant group differences have been reported for children with SLI versus children with TD language on a variety of visuospatial storage tasks including memory for hierarchical forms, pattern recognition, paired associates learning, pattern recall, the recall of locations, picture recognition, localization recall and visual symbol sequencing (Akshoomoff et al., 2006; Bavin et al., 2005; Hick et al., 2005; Hoffman & Gillam, 2004; Menezes et al., 2007; Nickisch & von Kries, 2009). Longitudinal research by Hick et al. (2005), moreover, has shown the performance of children with SLI on a visuospatial storage (pattern recall) task to develop slower than that of their TD peers. Deficits have also been demonstrated on visuospatial CE tasks, including odd-one-out, spatial WM test, space visualization and position in space (Henry et al., 2011; Hoffman & Gillam, 2004; Karasinski & Ellis Weismer, 2011; Marton & Schwartz, 2003).

A meta-analysis of visuospatial WM in children with SLI

Meta-analysis is a useful tool for statistically comparing a large set of results from — often quite divergent — individual studies (Glass, 1976). The results of a meta-analysis can help integrate research findings and, via the information provided on effect sizes, indicate the magnitude of those differences that are of interest. Within the context of the present research, this is the difference between the visuospatial WM performance of children with SLI versus their TD peers.

Assuming WM to be multicomponential, we asked ourselves the following questions. Do children with SLI show deficits in any of the components of visuospatial WM (i.e., visuospatial storage or visuospatial CE component)? When visuospatial storage or CE deficits are detected, do they relate to the inclusion criteria for SLI? And do any of the differences in the visuospatial storage or CE capacities of the children with SLI relate to their age?

The conflicting results on visuospatial WM in previous studies, raise the question of whether SLI is really language specific and thus confined to the verbal domain of WM as

has been assumed for many years now. Perhaps the problems underlying SLI are actually more general and thus related to a more pervasive impairment of the children's WM capacity. If only the verbal WM of children with SLI is found to be affected, this is in keeping with domain-specific hypotheses, maintaining that a deficit in verbal storage underlies SLI (Gathercole & Baddeley, 1990). More recently Archibald and Gathercole (2009c) argued that a combination of problems in verbal storage as well as the CE component must be assumed to underlie SLI because a verbal storage deficit cannot explain the substantial deficits found in verbal complex memory tasks. If both the visuospatial WM and verbal WM are affected in children with SLI, then this is in line with domain-general hypotheses. More generally, if children with SLI exhibit deficits in both the verbal and visuospatial components of their WM, then SLI can be assumed to arise from a limitation on the general processing capacities of children — a limitation that will manifest itself on any task with a high processing load (Ellis Weismer, 1996; Fazio, 1998; Hoffman & Gillam, 2004; Marton 2003, 2008; Montgomery, 2000, 2002). In this case, doubts can be raised about the specificity of the children's deficits to *language* and their status as having a *specific* language impairment.

Regarding our second question, the inclusion criteria for SLI used across studies are, unfortunately, not uniform. While the children identified as having SLI always show a substantial language delay, a variety of other inclusion criteria are typically also used. The number of language domains that must be affected, for example, can vary from 1 to a minimum of 3. The extent to which the children's language must be affected can vary from -1 to -1.5 standard deviations from the mean on standardized language tests. This might implicate that children with somewhat different language problems are included across studies. To complicate things further, some studies have reported greater variation in the visuospatial WM scores of children with SLI compared to their TD peers. This suggests that there might be a subgroup of children with SLI who have visuospatial WM problems (Archibald & Gatherole, 2006a; Menezes et al., 2007; Hick et al., 2005a,b). Nikisch and von Kries (2009), for example, found only visuospatial storage problems to occur in children identified as having a mixed set of language problems as opposed to a single set of language problems (i.e., both receptive and expressive language problems as opposed to only expressive language problems). Our hypothesis is therefore that the visuospatial WM performance of children with SLI will relate to the inclusion criteria used for SLI in a particular study and thereby to the nature of the language impairments. We further predict that the visuospatial WM deficit will be larger in children with more pervasive language impairments (i.e., studies that included children with multiple language deficits) than children with less pervasive language impairments (i.e., studies that included children in the SLI group with at least one language domain affected).

With regard to our final question and the course of the visuospatial WM deficits of children with SLI with age, it is well known that the language profiles of children with SLI

change over time (Bishop, 2006; Leonard, 1998). It is therefore certainly possible that if children with SLI show deficits in visuospatial WM, the extent of these problems varies across the ages of these children and thus with development. Studies with TD children show the basics for all of the components of Baddeley's WM model to be in place and clearly measurable by the age of four years. Children's WM still develops after this age, and the developmental trajectories for the different components of Baddeley's model show linear increases from four to eleven years of age (Alloway et al., 2006; Luciana & Nelson, 1998). As far as we know, only one study took changes in the visuospatial WM capacities of children with SLI into account. When Hick et al. (2005) examined performance on a visuospatial storage task over time in young children with SLI (aged 3:06 - 5;0 years), the results showed slower development of visuospatial storage performance relative to TD children. This indicates a widening gap in the visuospatial WM skills of the children with SLI relative to their TD peers over time. In line with this finding, we expected within the context of the present study, deficits in visuospatial WM to be most profound in older children. This hypothesis receives further indirect support from the assumption that inefficient verbal coding of visuospatial information contributes to the problems in visuospatial WM in older children with SLI (Gillam et al., 1998). Children from the age of seven years normally use verbal coding strategies in visuospatial WM tasks (Gathercole et al., 1994). However, it is assumed that children with SLI use minimal or inefficient verbal coding strategies due to their language problems. If inefficient verbal coding contributes to the deficits in the visuospatial WM performance of children with SLI, visuospatial WM problems should stand out most among older children, as verbal coding is known not to occur in children until around the age of seven years.

METHODS

Identification of studies

In June and July of 2012, studies investigating the visuospatial WM of children with SLI and their TD peers were identified via computerized database searches of PubMed, PsychINFO, and Web of Science. As already noted, SLI is not defined uniformly across studies. In addition, measures of visuospatial WM are not always clearly labeled as such. We therefore employed broad search terms in our initial search: *specific language impairment* and *visuospatial working memory, specific language impairment* and *working memory, language impairment* and *visuospatial working memory, language impairment* and *memory*. We further searched for papers that were judged to be relevant from the reference lists for the articles identified in our database search.

To avoid publication bias and language bias the literature search targeted published articles as well as unpublished data in the public domain in all languages. However, publication bias against nonsignificant findings is likely to be less of a problem in the study of visuospatial WM in SLI. Since several authors assume that visuospatial WM is not affected in SLI, studies in which children with SLI show performances similar to that of their TD peers are also of interest (Alloway & Archibald, 2008; Archibald & Gathercole, 2006a, 2006b; Baird et al., 2009; Williams et al., 2000).

A total of 894 papers were identified in the initial searches, of which 117 were duplicates, leaving 777 papers. The abstracts of all of the identified articles were reviewed in order to determine which of them examined the visuospatial WM of children with SLI and compared this to that of TD peers. When it was unclear that the study met this criteria, the full text for the article was reviewed. A total of 45 potentially appropriate articles were identified for further review. These articles were evaluated by the first author for inclusion in the meta-analysis. Afterwards, the total number of papers was re-evaluated by a second author, who did not take part in the initial search (JC). Agreement with the first author about whether or not a study met the inclusion criteria was 96%. Disagreements were resolved by consensus.

Inclusion criteria

For inclusion in the meta-analysis we used the following inclusion criteria:

- 1. Studies had to present original data comparing visuospatial WM of children with SLI to that of TD peers. We included studies in which children met the following criteria for SLI: impaired expressive and/or receptive language in combination with normal nonverbal intelligence. In addition to normal hearing, most studies reported no history of frank neurological impairments. In all of the studies, the children were required to show a substantial language delay, but a variety of criteria for the determination of the actual language impairment were used: clinical diagnosis by speech-language pathologist, significant discrepancy between language skills and nonverbal intelligence on standardized tests, and/or scores below age expectation on one or more standardized language tests (at least -1 sd below the mean). None of the reviewed studies included children with solely deficits in the phonological domain of language. One study compared TD children to children with a current SLI but also children with a past SLI. In this case, the results for the children with the current SLI were included in the meta-analysis (Baird et al., 2009). Studies in which the SLI group consisted of children with SLI in combination with other developmental disorders were excluded (Cohen et al., 2000; Jonsdottir et al., 2005).
- 2. At least one of the comparison groups had to be composed of typically developing children. Studies that did not have a control group were excluded (Archibald & Gathercole, 2006; van Daal, 2008) as well as studies that had a control group of children

with other developmental disorders (Alloway & Archibald, 2008; Alloway et al., 2009; Freed et al., 2012).

- 3. Each study had to include a task requiring the storage of visuospatial information or both the storage and processing of visuospatial information. The tasks had to use a span paradigm or, in other words, require the child to remember an increasing number of visuospatial stimuli — either alone or in combination with the processing of other visuospatial information; studies that did not use a span paradigm to assess the children's visuospatial abilities were excluded from our meta-analysis to facilitate comparison (Marton, 2008, 2009). In the case of visuospatial CE tasks, we only included studies in which the stimuli for storage and processing were of a visuospatial nature. Tasks requiring a combination of visuospatial stimuli with verbal stimuli were excluded (Archibald & Gathercole, 2006; Hoffman & Gillam, 2004). The exact type of visuospatial task was not further restricted; a variety of visuospatial stimuli were employed in the studies (e.g., shapes, pictures, dots, block recall, hand movements). However, we excluded some tasks, stated to be tasks of visuospatial storage, that in fact measured a different construct, like for instance visuospatial associative learning (Bavin et al., 2005). Another study was excluded because the main measure was presentation duration and not visuospatial WM (Fazio, 1998). In order to be convinced that the tasks genuinely reflected visuospatial storage, moreover, we excluded tasks that easily invited for verbal coding of the visuospatial information (for instance color identification) (Hoffman & Gillam, 2004).
- 4. Studies had to report sufficient data to calculate an effect-size for each task; that is, the number of subjects, mean scores and standard deviations or standard error.

Data extraction

A total of 21 studies from the database searches met all the inclusion criteria en were included in the meta-analyses. These 21 studies reported data of 32 separate visuo-spatial storage tasks and 9 visuospatial CE tasks. For each task reported in each study, the following information was next coded by the first author: 1) statistics regarding differences in visuospatial WM (means, standard deviations, t tests and F tests); 2) SLI diagnosis criteria; 3) number of SLI and control group participants; 4) mean age of the participants; 5) type of control group (chronologically age-matched or developmentally language-matched); 6) type of visuospatial WM task (visuospatial storage or visuospatial CE). A random sample of 5 of the studies (24%) was also coded by a second coder (JC). Coder agreement was 98 %. When disagreement occurred, consensus was achieved by re-examining the original data in the articles.

In order to have mutually exclusive (i.e., independent) samples in the meta-analysis, studies examining the same group of children were not both included in the analysis. In this case, we included data from the first study (Hick, 2005a, 2005b; Kleemans et al.,

2011, 2012). Some studies compared the children with SLI to both chronologically agematched and language-matched control children. To avoid multiple entries from the same group, only the effect sizes for the chronologically age-matched control groups were calculated; also, not all of the studies had language-matched control groups. In two studies, the SLI group was divided into subgroups and the data reported separately for these subgroups (Cowan et al., 2005; Nickisch & Von Kries, 2009). In these cases, means and standard deviations were pooled and entered for the SLI group as a whole.

Analyses

Analyses were conducted using Comprehensive Meta-Analysis version 2 (Borenstein et al., 2005). Effect sizes were calculated per task for the 21 studies included in the metaanalysis. The effect size (d) is the difference between the scores of the two groups of children divided by the pooled standard deviation for the two groups. A positive effect size indicates a higher performance of the control group on visuospatial WM. Effect sizes are considered small for d = .20, medium for d = .50 and large for d = .80 (Cohen, 1988). Given that some of the included studies had small sample sizes, which can sometimes result in extreme values, all effect sizes were multiplied by a correction factor: CF = (1 - [3 / (4 * Ncontrol + Nsli) - 9]. This correction reduced the possibility of bias from small samples by taking into account the sample size associated with each effect size (Robey & Dalebout, 1998).

For the further analysis of the data, a random-effects model (allowing for heterogeneity between studies) was used (Hunter & Schmidt, 2000). In order to answer the first research question, the weighted mean effect sizes across all included studies of visuospatial storage and visuospatial CE were calculated. For this calculation we used the inverse variance of each effect size to weight the relative contribution of each study to the overall effect size (Hedges & Pigott, 2004). To avoid multiple entries from the same group, we averaged the effect sizes obtained from each WM task in studies that used multiple tasks of visuospatial storage or CE.

Homogeneity testing was conducted for the meta-analyses of visuospatial storage and visuospatial CE to determine the extent to which there was variation in findings between studies in each component of visuospatial WM. The *I*² statistics were calculated according to Higgins et al. (2003) to describe the amount of total variation across studies due to heterogeneity. To further investigate causes of heterogeneity, moderator analyses using a mixed effects model and meta-regression techniques were performed (Hedges & Pigott, 2004; Thompson & Higgins, 2002). These analyses examined two factors that are hypothesized to affect visuospatial WM in children with SLI: inclusion criteria for SLI and age.

Publication bias was investigated using funnel plots, Egger's linear regression approach, and the trim and fill method (Duval & Tweedie, 2000; Egger et al., 1997). The

Egger's linear regression approach examines both the sample size and statistical power of each study in relation to the effect size. The trim and fill method corrects the meta-analyses by imputing the presence of missing studies to yield an unbiased pooled estimate.

RESULTS

Overall effects

The characteristics and effect sizes for the 18 studies that measured visuospatial storage are presented in Table 1. The 18 studies included 32 visuospatial storage tasks. The weighted mean effect size across the 32 effect sizes of these tasks is 0.49, with a 95% confidence interval from 0.30 to 0.68. The characteristics and effect sizes for the 7 studies that measured visuospatial CE are presented in Table 2. The 7 studies included 9 visuospatial CE tasks. The weighted mean effect size across these studies is 0.63, with a 95% confidence interval from 0.27 to 0.99. Both weighted mean effect sizes were significant, which shows the children with SLI to perform significantly below their TD peers for both the visuospatial storage (Z = 4.99, p = .000) and visuospatial CE component (Z= 3.39, p = .001).

None of the analyses that examined the presence of possible publication bias indicated publication bias for either the effect sizes of visuospatial storage or the effect sizes of visuospatial CE. Egger's linear regression for visuosatial storage: $\beta o = -1.282$, t(16) = 1.109, p = .284 and for visuospatial CE: $\beta o = -2.133$, t(5) = 1,344, p = .237. The trim and fill method detected no missing studies.

We conducted homogeneity analyses to see if the samples of effect-sizes shared a common effect size. Heterogeneity analyses revealed moderate l^2 values for the effect sizes of visuospatial storage and visuospatial CE (visuospatial storage $l^2 = 50.05$ and visuospatial CE $l^2 = 67.58$), indicating that there was substantial variability among the effect sizes within both components of visuospatial WM (Higgins et al., 2003).

Interactions

To explain the nonhomogeneity in the effect sizes found for the children's visuospatial storage, on the one hand, and their visuospatial CE, on the other hand, we examined factors that could possibly contribute to or interact with the visuospatial deficits observed in the children with SLI. To avoid a "fishing trip" or undirected search for possible correlations, we examined only two factors: the SLI inclusion criteria used in the studies and the ages of the children studied.

Table 1

Characteristics of 18 studies examining the visuospatial storage component in both children with SLI and TD peers.

Author	Tests	N SLI	N TD	Age SLI	Age TD	d
Akshoomoff et al., 2006	Hierarchical Forms Memory Task	29	26	119	115	0.774
Archibald & Gathercole, 2006b	Dot Matrix, AWMA	15	15	116	116	0.538
Archibald & Gathercole, 2006c	Visuospatial storage	14	14	122	123	0.341
Baird et al., 2009	Finger windows, WRAML	51	26	122	112	0.233
	Design memory, WRAML	51	26	122	112	0.167
Bavin et al., 2005	Pattern recognition, CANTAB	21	21	54.1	54	0.933
	Spatial recognition, CANTAB	21	21	54.1	54	0.585
	Spatial span, CANTAB	21	21	54.1	54	0.628
Briscoe & Rankin, 2007	Block recall	14	14	99.7	99.7	-0.018
Cowan et al., 2005	Corsi span	55	57	98	98	0.576
Gray, 2006	Handmovements, K-ABC	15	15	43.3	42.7	0.920
Hick et al., 2005a	Pattern recall	9	9	45	45	0.655
Hoffman & Gillam, 2004	Experimental task: No color ID	24	24	112.7	112.3	1.063
	Pointing	24	24	112.7	112.3	0.945
Hutchinson et al., 2011	Mazes memory, WMTB-C	18	24	93.2	92	0.114
	Block Recall, WMTB-C	18	24	93.2	92	0.299
Kleemans et al., 2011	Memory Span, RAKIT	61	111	73.9	72.7	1.210
Leclercq et al., 2012	Low number of dissimilar features	15	15	120.7	120.5	0.295
	Low number of similar features	15	15	120.7	120.5	1.426
	High number of dissimilar features	15	15	120.7	120.5	0.993
	High number of similar features	15	15	120.7	120.5	0.792
Lum et al., 2011	Mazes memory, WMTB-C	51	51	117.6	118.2	0.097
	Block recall, WMTB-C	51	51	117.6	118.2	0.376
Nickisch & Von Kries, 2009	Handmovements, K-ABC	42	21	108	108	0.237
	Visual Symbol Sequential Memory, ITPA-G	42	21	108	108	0.515
Petrucelli et al., 2012	Block recall, WMTB-C	24	32	63.3	63.2	0.240
	Picture Locations, CMS	24	32	63.3	63.2	0.095
Riccio et al., 2007	Dot Locations, CMS	30	30	100	100	0.095
	Picture Locations, CMS	30	30	100	100	0.090
Williams et al., 2000	Spatial recognition, CANTAB	10	10	78	78	-0.501
	Pattern recognition, CANTAB	10	10	78	78	-0.325
	Spatial span, CANTAB	10	10	78	78	-0.349

Note. N = number of included children; SLI = specific language impairment; TD = typically developing; *d* = effect size; AWMA = Automated Working Memory Assessment; WRAML = Wide Range Assessment of Memory and Learning; CANTAB = The Cambridge Neuropsychological Test Automated Battery; K-ABC = Kaufman Assessment Battery for Children; WMTB-C = Working Memory Test Battery for Children; RAKIT = Revisie Amsterdamse Kinder Intelligentie test; ITPA-G = German form of the Illinois Test of Psycholinguistic Abilities; CMS = Children's Memory Scale

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Characteristics of 7 studies examining the visuospatial CE component in both children with SLI and TD peers.

Author	Tests	N SLI	N TD	Age SLI	Age TD	d
Archibald & Gathercole, 2006b	Odd-one-out, AWMA	15	15	116	116	0.388
	Mister X, AWMA	15	15	116	116	0.685
	Spatial span, AWMA	15	15	116	116	-0.020
Archibald & Gathercole, 2006c	Visuospatial storage –visuospatial processing	14	14	122	123	-0.202
Bavin et al., 2005	Spatial working memory, CANTAB	21	21	54.1	54	0.446
Henry et al., 2011	Odd-one-out test	41	88	138.4	118	0.612
Karasinski & Weismer, 2010	Spatial working memory task	59	316	165	165	0.998
Miller & Wagstaff, 2011	Visual-spatial WM	29	20	120	123	1.530
Williams et al., 2000	Spatial working memory, CANTAB	10	10	78	78	0.289

Note. N = number of included children; SLI = specific language impairment; TD = typically developing; *d* = effect size; AWMA = Automated Working Memory Assessment; CANTAB = The Cambridge Neuropsychological Test Automated Battery

Relations to inclusion criteria for SLI

To investigate whether the variability in effect sizes related to the different inclusion criteria used for SLI, we divided the studies into two clusters: one cluster was composed of studies with the criterion of at least one affected language domain for inclusion in the SLI group; the other cluster was composed of studies with the criterion of two or more affected language domains for inclusion in the SLI group.

For the visuospatial storage component, the effect sizes for the two clusters were significant; the children in both clusters performed significantly below their TD peers (Z = 3.34, p = .001 for 1 language domain and Z = 5.24, p = .000 for 2 language domains). The cluster of studies that included children with deficits two or more domains of language yielded the largest effect size (d = 0.32 for 1 language domain and d = 0.70 for 2 language domains). The between-groups homogeneity test was also significant (Q = 5.59, p = .018), which shows the inclusion criteria for SLI to explain a significant amount of the variability in the effect sizes for visuospatial storage.

For the visuospatial CE component, the effect size was nonsignificant for the cluster of studies that included children with deficits in at least one domain of language (d =0.78, Z = 1.95, p = .052). The effect size for the cluster of studies that included children with deficits in two or more domains of language was significant (d = 0.54, Z = 2.29, p =.021). This shows the visuospatial CE of these children to be significantly below that of their TD peers. The between-groups homogeneity test was nonsignificant (Q = 0.28, p =.597). This indicates the inclusion criteria for SLI did not explain a significant amount of the variability in the effect sizes for visuospatial CE. Stated differently, the deficits in visuospatial CE are not related to the inclusion criteria for SLI.

Relations to age

We next conducted meta regression to determine if the magnitude of the effect sizes in visuospatial storage and visuospatial CE possibly declines or increases with age. The regression models for both visuospatial storage and visuospatial CE were nonsignificant ($\beta = -.003$, Z = -0.73e, p = .466 and $\beta = .005$, Z = 1.03, p = .303, respectively). These results indicate that differences in age do not produce significant differences in the effect sizes for either the visuospatial storage or visuospatial CE component.

CONCLUSIONS AND DISCUSSION

In this meta-analysis, we compared the visuospatial WM performance of children with SLI to that of TD peers. The weighted mean effect sizes for the visuospatial storage component (d = .49) and the visuospatial CE component (d = .63) were both significant. The children with SLI performed approximately one half standard deviation below their TD peers on average for both components of visuospatial WM. This finding suggests that children with SLI have not only smaller storage but also processing capacities for visuospatial information.

Although there was previously no consensus across studies on the role of visuospatial WM in children with SLI, the current findings clearly suggest that the visuospatial WM is affected in these children. This implies that the deficits in WM in children with SLI may not be not restricted to the verbal domain. However, when we compare the *magnitude* of the WM deficit in the two modalities, the deficit for visuospatial WM is not as large as the deficit for verbal WM. In a meta-analysis of non-word repetition as a measure of the verbal storage component of WM, Graf Estes et al. (2007) found the weighted mean effect size for the deficit in verbal storage to be as large as d = 1.27. As it can be assumed that children with SLI are even more severely impaired on verbal CE than on verbal storage (Archibald & Gathercole, 2006a; Ellis Weismer et al., 1999; Marton & Schwartz, 2003), this suggests that the deficit in verbal WM is two to three times larger than the deficits in visuospatial WM that we found. Fifteen of the 21 studies that we analyzed also had verbal WM tasks available for analysis. We therefore took the time to calculate the effect sizes for these verbal tasks and found a weighted mean effect size of d = 1.19 with a range of d = .57 to d = 3.14. This confirms our suspicion that the deficit in the verbal WM of children with SLI can be two to three times larger than the deficit in their visuospatial WM.

The magnitude of the effect sizes for the 21 studies (including 32 visuospatial storage tasks and 9 visuospatial CE tasks) that we analyzed varied greatly and ranged from d = -.50 (showing children with SLI to perform one-half standard deviation better than their TD peers) to d = 1.53 (showing children with SLI to perform more than one and a half

standard deviation below their TD peers). We therefore conducted moderator analyses in an attempt to explain some of this variation in the effect sizes.

The first moderator analysis addressed the question of whether the differences in the visuospatial WM for the children with SLI possibly relate to differences in the inclusion criteria used for SLI in the studies. This was found to be the case for visuospatial storage but not for visuospatial CE. The effect size for visuospatial storage was greater for studies that included children with deficits in two or more language domains (d = .70) than for studies that included children with a deficit in at least one language domain (d = .32). These results partially confirm our hypothesis that the visuospatial WM performance of children with SLI relates to the inclusion criteria used for SLI and thus the pervasiveness of language impairment. The deficit in visuospatial storage was found to be larger in children with widespread language impairment. These results are in line with a previous study showing a subgroup of children with SLI (i.e., children with more pervasive problems affecting both receptive and expressive language) to also experience visuospatial storage problems (Nikisch and Von Kries, 2009).

The results of this first moderator analysis, concerned with the relations between the inclusion criteria for SLI and the children's visuospatial WM, must be interpreted with caution. Firstly, the division of the studies in studies that included children with impairments in at least one domain of language in the SLI group versus those that included children with impairments in two or more domains of language may have allowed overlap. As not all of the studies in the first cluster included only children with an impairment in a single language domain (but, rather, at least one domain), it is possible that children with impairment in two or more language domains were inadvertently included in this first cluster. Second, the number of studies of visuospatial CE among children with SLI affected in a minimum of one domain of language was guite small. This could explain the finding of a nonsignificant effect size. Furthermore, the severity and nature (i.e., which linguistic domains are affected) of language impairments were not taken into account in this analysis although this could certainly be other important predictors of the pervasiveness of language impairment and thus relate to visuospatial WM. Finally, it cannot be completely ruled out that the included studies, besides differences in the inclusion criteria for SLI, also differed on other non-measured variables. So, the observed association between the inclusion criteria for SLI and the differences in visuospatial storage, might also reflect some other systematic influences.

The second moderator analysis addressed the question of whether age differences in the visuospatial WM capacities of children with SLI exist. We found no significant association between the age of the children with SLI and the effect sizes for either visuospatial storage or visuospatial CE. This means that age cannot explain the variation in the effect sizes across studies. This finding does not support our hypothesis that the visuospatial WM deficit in children with SLI would be larger in older children. The visuospatial WM performance of children with SLI apparently does not decline with age but, rather, remains stable. This finding is in line with other findings regarding the verbal WM of children in general and those with SLI in particular. In their meta-analysis of children's nonword repetition, Graf Estes et al. (2007) found the magnitude of the deficit in the verbal storage component of WM also remains stable across age. The present cross-sectional findings must still be interpreted with caution and further conclusions can only be drawn when we have longitudinal insight into the visuospatial WM performance of children with and without language impairments.

Taken together, the results of our meta-analysis demonstrate deficits for both visuospatial storage and visuospatial CE in children with SLI. This outcome suggests that children with SLI have a smaller capacity for both the storage and processing of visuospatial information than children without SLI. More generally, this outcomes suggests that the WM deficits of children with SLI are not restricted to the verbal domain, and that SLI may thus be associated with domain-general impairments of WM. The results of our meta-analysis also show the deficit in visuospatial WM to be stable across development. The magnitude of the deficit in visuospatial storage, in particular, correlated with the inclusion criteria for SLI: greater impairment of the children's visuospatial storage capacity was associated with more pervasive language impairment.

Our finding of apparently general WM impairments in children with SLI suggests that the language-specific nature of the diagnosis can be brought into question. "Specific language impairment" may no longer be the most appropriate term for the pattern of impairments demonstrated by children with so-called SLI. Although children with SLI show more substantial deficits on tasks requiring verbal WM than on tasks requiring visual WM (i.e., the extent of the deficit in verbal WM is two to three times larger than the extent of the deficit in visuospatial WM), their impairments are increasingly being seen to not be completely specific to language or the processing of strictly verbal information. The current results suggests that — for at least some children — the term "primary" language impairment might be more appropriate than "specific" language impairment (Edwards & Munson, 2009).

The implications are not straightforward for our finding of domain-general impairment of WM in children with SLI because different explanations are available for this outcome. One possibility is that the impairments in the visuospatial WM capacities of the children with SLI reflect limitations on their general processing capacity. Children with SLI may actually shows problems in *both* verbal and visuospatial domains when processing load is high (Ellis Weismer, 1996; Fazio, 1998; Hoffman & Gillam, 2004; Montgomery, 2000, 2002). Stated differently, children with SLI can adequately process single bits of information but encounter problems when they have to process multiple bits of information or, in other words, more complex information. Evidence for this account comes from studies showing children with SLI to indeed have problems on both verbal
and visuospatial tasks with higher processing loads (Ellis Weismer, 1996; Fazio, 1998; Hoffman & Gillam, 2004; Marton & Schwartz, 2003; Montgomery, 2000, 2002). In a recent study by Leclerq et al. (2012), children with SLI were more strongly affected by stimulus complexity defined in terms of visual similarity and the number of similar/dissimilar features in a visuospatial storage task when compared to age-matched children without SLI. The authors suggest that stimulus complexity is a critical factor of the poor visuospatial storage performances in children with SLI and may therefore explain the conflicting results of previous studies with regard to the visuospatial WM performance of children with SLI. More research is required to identify whether visuospatial WM tasks requires 'more' processing in children with SLI in some way and what the exact role of factors like stimulus complexity is.

Another possibility is that the visuospatial WM system of the children with SLI is intact but that the steering of this system by the language system is problematic due to the children's SLI. This explanation hinges on whether the performance of the children on the visuospatial WM tasks genuinely reflects their visuospatial storage and processing, as we assumed, or possibly verbal mediation of visuospatial information. Some experts have indeed hypothesized that children with SLI show inefficient verbal coding during visuospatial WM tasks (Archibald & Gathercole, 2006b; Gillam et al., 1998). Given their language problems they may use less efficient verbal strategies or rely more than other children on visual codes when actually phonological codes are preferable. Although we excluded studies that easily invite for verbal coding in this meta-analysis, the possibility of verbal coding during the performance of visuospatial WM tasks can never be completely ruled out. However, inspection of the effect sizes shows deficits in visuospatial WM in several studies of children with SLI before the age of seven (Bavin et al., 2005; Gray, 2006; Hick et al., 2005; Kleemans et al., 2011). Given that verbal coding in visuospatial tasks is known not to occur in children before the age of about seven years, this does not support the inefficient verbal encoding explanation of the children's visuospatial WM impairments (Gathercole et al., 1994).

Yet another possibility is that that the impairments in the visuospatial WM capacities of the children with SLI are the reflection of limitations in attentional control. From this perspective, attentional control is assumed to be a regulator that plays an important role in the coordination of storage and processing in WM. This view is in line with accounts of WM that highlight the notion of limited attentional resources (Courage & Cowan, 2009; Engle et al., 1999). It is known that visuospatial WM places particularly high demands on processes regulated by attentional control (Miyake et al., 2001). And, in turn, the problems encountered by children with SLI in visuospatial WM tasks may therefore stem from problems with attentional control. This possibility is supported by the finding of attentional problems in children with SLI (Dodwell & Bavin, 2008; Finneran et al., 2009; Spaulding et al., 2008; Noterdaeme et al., 2001). In addition, when Marton (2008) com-

pared children with SLI and good versus poor attentional control, the children with poor attentional control also showed greater problems on a visuospatial WM task. These findings call for additional research to explore the exact contribution of attention control to visuospatial WM in children with SLI.

The inclusion criteria used for SLI in the different studies included in our meta-analysis explained some of the variation in the effect sizes found for particularly visuospatial storage. Nevertheless, a significant amount of variation in the effect sizes remained unexplained. This indicates that there are other factors that affect the magnitude of the effect sizes found in the different studies. One additional factor that was not taken into account in the present meta-analysis is the type of control group. For inclusion in our meta-analysis, the performance of the children with SLI had to be compared to that of chronologically age-matched children without SLI. This was done because developmentally language-matched control groups were not available in all of the studies. We did not include type of control group as a factor in our moderator analyses because the inclusion of data from both age- and language-matched control groups would have violated the assumption of independent samples, which is a prerequisite for such an analysis. Four of the studies included in our meta-analysis nevertheless used both ageand language-matched control groups, which allows us to compare the effect sizes for visuospatial storage and visuospatial CE control. All of the effect sizes for the children with SLI compared to language-matched control children were negative (varying from d = -.14 to d = -2.04). This shows the children with SLI to perform better than the younger, language-matched children on visuospatial storage and visuospatial CE. In other words, children with SLI appear to be perform significantly worse than age-matched children and better than younger, language-matched children on visuospatial WM tasks. An important restriction on this conclusion is that differing linguistic skills were used across the studies to determine the language-matched control groups.

Other factors that might have affected the magnitude of the effect sizes discerned in this meta-analyses could be task characteristics, such as: type of stimuli, nature of the stimuli, duration of stimulus presentation, and task duration. Leclerq et al. (2012) have recently shown both degree of visual similarity (i.e., the overlap of visual features between two objects) and the number of features to determine visuospatial storage performance in children with SLI. Information on these variables was not included in our meta-analysis because the information was not provided in all studies and, in those studies that did provide task and stimulus information, the information was so different that systematic comparison was impossible. But in future research, this information should certainly be attended to.

In closing, the present findings obviously have some important implications for the assessment and treatment of children with SLI. For assessment in the future, visuospatial in addition to verbal WM tasks should probably be administered. The WM deficits of

children experiencing language problems may not be restricted to verbal WM, and it is obviously important to know if the problems being experienced by a child are also with visuospatial WM. For treatment, interventions should probably not focus on language alone but also on strategies for the storage and processing of both verbal and visuospatial information. It is important that WM demands be minimized during teaching and treatment in order to limit the adverse effects of the WM deficits. For the use of visual support, which is a common intervention strategy adopted for children with SLI, the current findings indicate that children with SLI might not benefit from visual support as normal children do. This means that only certain types of visual support may be suited, namely: simple visual information that does not exceed the child's WM capacity. These clinical implications may particularly be important for children with complex pervasive language impairments. As children with more widespread language impairments appear to be more resistant to interventions, and the current results show greater impairment of visuospatial WM (or at least visuospatial storage) in these children, it might be valuable additions to more traditional interventions (Boyle et al., 2010).

Finally, the present findings suggest a number of possible directions for future research. As the results of our meta-analysis show, the deficit in visuospatial storage might be larger in children with more pervasive language impairments. The associations between the impairments in visuospatial WM and different linguistic domains in children with SLI should therefore be examined in future research. Differences in the inclusion criteria for SLI reflected differences in the pervasiveness of the language impairments in the present research and were found to be associated with significant differences in the children's visuospatial WM performance. The association between receptive language problems and visuospatial WM might in particular be something to consider in future research. As the results of a previous study showed visuospatial storage to *only* be affected in children with a mixed pattern of receptive/expressive language problems, it might suggest a role of visuospatial WM in the receptive language problems of children (Nikisch & von Kries, 2009). Furthermore, the role of attentional control certainly calls for further research as such a limitation may indeed contribute to problems with visuospatial WM.

Chapter 3

Executive function behaviours in children with SLI

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ABSTRACT

Background: There is growing evidence that linguistic and non-linguistic factors may contribute to the problems associated with specific language impairment (SLI). One factor that has been implicated is executive functioning (EF). Most studies investigating EF in children with SLI use performance based tasks. Significant group differences in children with SLI are reported on the following components of EF: inhibition, working memory, planning and fluency, although not on the 'shifting' component. Correlations between performance based measurements of EF and ratings of everyday EF behaviours are often low. It is possible that standardised tests are not sufficiently sensitive to the multidimensional nature of EF. Therefore it is suggested that information on EF should be collected in different contexts and from different sources using behavioural ratings, like the Behaviour Rating Inventory of Executive Function (BRIEF) for children.

Methods and procedures: A clinical sample of 237 school aged children, aged 5–12 years, (157 boys, 80 girls) with SLI participated in this study. Behavioural and cognitive measures of EF were administered: the BRIEF-questionnaire, assessing everyday EF behaviour in a home and school setting and two EF tasks: Digit span (WISC-III-NL) and Creature Counting (TEA-Ch).

Outcomes and results: Compared to the normative sample the prevalence of EF problems in school in children with SLI is much higher than in the normal population. Teachers reported significantly more problems on almost all EF domains (i.e. Inhibition, Shifting, Emotional control, Initiate, Working memory, Plan/organise, and Monitor), except organisation of materials. Working memory and Initiate are the most impaired, since more than one third of the children had scores in the clinical range on these scales. Compared to the normative sample parents scored significantly more working memory problems. MANOVA-analyses showed developmental and gender differences on EF behaviour in school. Overall, older children had less problems in EF behaviours than younger children and boys showed more problems than girls. Like others we found low correlations between behavioural and cognitive measures (r = 0.20).

Conclusions: School aged children with SLI show substantial impairments in everyday EF behaviour in a classroom setting. Almost one third of the children scored in the clinical range on the Metacognition Index. Our findings replicate low correlations between performance based and behavioural based measures on EF. These findings indicate the importance of expanding EF assessment with behavioural measures in clinical practice for children with SLI.

INTRODUCTION

SLI and executive functions

Children with specific language impairment (SLI) encounter language problems that can be characterized as a failure to make normal progress without further evidence of underlying intellectual, neurological, social, or emotional impairment (Bishop, 2002, 2006). SLI can affect different linguistic domains including phonological, morphological, lexical and syntactical domain. The language profile of children with SLI often changes with age and these changes can occur both within and across linguistic domains (Bishop, 2006; Leonard, 1998). SLI is a persistent disorder that can affect language abilities into adolescence, or even into adulthood (Brizzolara et al., 2011; McKinley and Larson, 1989). Children with SLI are also at risk for less successful academic outcomes as well as behavioural, emotional, and social difficulties (Conti-Ramsden et al., 2009; St Clair et al., 2011).

Different theories and hypothesis are provided to explain the bases of SLI. One of these hypothesis presumes that a deficit or delay specific to language, and particularly grammar, underlies SLI (e.g. Rice and Wexler, 1996; Van der Lely, 2005). More recently however, growing evidence implicates that non-linguistic factors may contribute to the language problems associated with specific language impairment (Bishop, 2006; Montgomery et al., 2010). One factor that has been suggested is executive functioning (EF) deficits (Im-Bolter et al., 2006; Henry et al., 2011). Several studies provide evidence of domain-general executive function deficits in children with SLI (Archibald and Gathercole, 2006b; Im-Bolter et al., 2006; Lum et al., 2011; Marton et al., 2007). Findings from neuroimaging studies even suggest that children with SLI show anomalies in frontal brain areas normally related to EF, like for instance orbitofrontal, dorsolateral and medial frontal cortex (Dibbets et al., 2006; Jernigan et al., 1991).

Executive function is a broad term that comprises cognitive processes responsible for purposeful, goal directed behaviour. EF is implicated in not only cognitive processes but also emotional responses and behavioural actions (Gioia et al., 2001; Miyake and Shah, 1999). Traditionally, EF has been conceptualised as a unitary mechanism (i.e. the central executive) that does not include distinct subfunctions (Baddeley, 1986; Shallice, 1990). More recently however, EF is considered multifaceted with distinct subfunctions. These processes are inter-related and function together as an integrated, supervisory control system (Stuss and Alexander, 2000). Based on factor analysis studies, the three most frequently postulated components of EF are: inhibition, working memory and shifting (Huizinga et al., 2006; Miyake et al., 2000). Inhibition is the ability to stop prepotent or ongoing responses (Miyake et al., 2000). Working memory refers to the structures and processes used to temporarily store and manipulate information (Baddeley & Hitch, 1974; Baddeley, 2003). Shifting, sometimes also described as cognitive flexibility, has

(Miyake et al., 2000). Besides these three components, several additional components of EF have been postulated. However, some debate still remains about the exact components of EF. In this study, we used the framework of Gioia, Isquith, Guy and Kenworthy (2000). Within this model five other EF are included in addition to inhibition, working memory and shifting, namely emotional control, initiation, planning, organization and monitoring. Emotional control is conceptualised as the ability to modulate emotional responses in order to engage appropriately in social interactions. Initiation refers to the ability to initiate activities and to generate ideas, strategies and responses. Planning is typically measured using problem solving tasks and refers to children's capacity to manage task demands. Organisation is the ability to organise information. Monitoring at last, refers to the ability of a child to check his or her performance during or after a task.

The different executive functions are commonly measured using standard neuropsychological EF tasks. However, a general issue in the field of EF research is the question of ecological validity of these performance based tasks. EF tasks have been criticised since there is evidence that children with clear EF problems in their daily activities can perform well on standardised performance based tasks (Vriezen and Pigott, 2002). Additionally, several studies showed no or low correlations between performance based measures of EF and ratings of everyday EF behaviours (Anderson et al., 2002; Chaytor et al., 2006; Vriezen and Pigot, 2007). This may reflect that standardized tests are not sufficiently sensitive to the multidimensional nature of EF in daily life (Chaytor et al., 2006). Based on these findings, it is suggested that information should be collected in different contexts and from different sources using behavioural ratings of EF (Gioia et al., 2001). For this purpose Gioia and colleagues (2000) developed the Behaviour Rating Inventory of Executive Function (BRIEF). Research using the BRIEF has shown that it is a valid and reliable measure of everyday EF (Mahone et al., 2002).

With regard to the development of EF, a growing body of research indicated that it is a protracted process which extends into early adulthood. Different components of EF show different developmental trajectories related to the neurophysiological developments of the growing brain. Most research on development of EF focused on the three basic components (i.e. inhibition, working memory and shifting). With regard to inhibition, results from several studies showed that children generally show rapid early improvement in preschool years. There is a spurt in performance on inhibition tasks between 3 and 5 years of age, followed by more modest, lineair improvements through adolescence (Best and Miller, 2010; De Luca and Leventer, 2008). The ability to keep information in mind (i.e. working memory) is present around the age of 6 months (Courage and Cowan, 2009; Garon, 2008). Evidence suggests that the trajectory of working memory development is linear from 4 to 15 years of age, with the largest development between the age of 5 and 11 years (Alloway et al., 2006). The ability to shift between tasks also starts to develop during preschool with children being able to shift between simple task sets. The ability

to handle unexpected shifts between increasingly complex task sets develops later, with a development through adolescence (Best and Miller, 2010). During adolescence the different EF brain systems become better integrated and at age 20–29, EF skills are at their peak (De Luca and Leventer, 2008).

Research on age related changes in EF based on parent ratings on the BRIEF revealed a decrease in reported EF problems with increasing age. Huizinga and Smidts (2011) reported that 5–8 year old children showed significantly more behavioural EF problems on inhibition, shifting, emotional control and working memory compared to 9–11 year old children. In addition, 12–14 year olds showed more problems on inhibition and emotional control compared to 15–18 year olds.

Besides these age related changes in EF behaviours, gender differences are also reported in some studies. In normal developing children, elevated levels of behavioural EF problems assessed with the BRIEF were found in boys compared to girls aged 5 to 18 years (Gioia et al., 2000; Huizinga and Schmidt, 2011). In addition, Skogli and colleagues (2013) reported in a recent study on children with ADHD that parental rating of EF on the BRIEF could better distinguish between children with and without ADHD in boys than in girls, indicating more profound problems in EF behaviours in boys with ADHD compared to girls.

Executive functions in children with SLI

Most of the previous studies investigating EF in children with SLI used performancebased tasks. In these studies significant group differences have been reported between children with SLI and typically developing children (TD) on inhibition, working memory and planning tasks. Compared to their TD peers, children with SLI demonstrate problems with inhibition in several studies (Bishop and Norbury, 2005b; Finneran et al., 2009; Im-Bolter et al., 2006; Marton et al., 2007). However Noterdaeme and colleagues (2001) showed no difference on a go/no-go task. In addition, it is widely accepted that children with SLI show impairments in the verbal domain of working memory (Archibald and Gathercole, 2006b; 2007). Significant group differences have been reported between children with SLI versus TD children on tasks of non-word repetition, recall of words, recall of digits, and complex verbal span tasks (Archibald and Gathercole, 2006b; Gray, 2003, 2006; Conti-Ramsden, 2003). In contrast, the visuospatial domain of working memory has been less extensively investigated in children with SLI with somewhat contradictory results (Alloway and Archibald, 2008; Archibald and Gathercole, 2006a; Montgomery et al., 2010; Vugs et al., 2013). Several studies showed children with SLI to perform similarly to their TD peers on visuospatial working memory tasks (Alloway and Archibald, 2008; Archibald and Gathercole, 2006a, 2006b; Lum et al., 2011; Williams et al., 2000). The results of several other studies and a recent meta-analysis however, have yielded evidence suggesting that the working memory deficit of children with SLI may extend to the visuospatial domain (Vugs et al., 2013).

Compared to the other EF skills, planning has received considerably less attention in research on children with SLI. Problems with planning abilities in children with SLI have been reported on Towers tests and a Sorting test (Henry et al., 2011; Marton, 2008). In contrast to these findings of decreased performance on EF tasks of inhibition, working memory and planning, there is no evidence for problems in shifting in children with SLI. They perform similarly to their TD on several shifting tasks, including the Trailmaking Test and set-shifting tasks (Dibbets et al., 2006; Henry et al., 2011; Im-Bolter et al., 2006). To the best of our knowledge, performances on tasks of emotional control, initiation, organization and monitoring have not yet been examined in children with SLI.

To date, research with behavioural ratings of EF in children with SLI has been limited. In the study of Hughes and colleagues (2009) the parental and self-ratings of EF for adolescents with SLI versus TD adolescents were compared using the BRIEF. The results showed more negative ratings of EF in general for the SLI group compared to the TD group. Even half of the parents of adolescents with SLI rated their child's EF abilities in the clinically impaired range. More recently, two studies addressed the role of EF in preschool children with SLI. Wittke and colleagues (2013) studied EF of children aged 3–5 years using the BRIEF preschool version. They found that the EF of children with SLI was rated significantly worse than those of their TD peers by both parents and teachers. In the study of Vugs and colleagues (2014), behavioural ratings of EF on the BRIEF-P showed the parents of children with SLI aged 4 and 5 years to report significantly more problems relative to the parents of the TD children. These included problems with inhibition, shifting, emotional control, working memory, and planning/organisation.

Present study

Although the use of behavioural measures of EF have been widely advocated to gain more ecological valid information, studies using rating scales of EF in children with SLI has been very limited. Including behavioural ratings of children's EF could particularly be useful to gain information about the impact of EF on activities in daily life in different contexts. In addition, previous studies that did examine behavioural ratings of EF in children with SLI focused on preschool children and adolescents. Research on EF behaviours in school aged children with SLI has not yet been conducted. However, to examine the development of EF in children with SLI, it would be valuable to gain information about EF behaviours in this age group. In the present study, we therefore investigated behavioural ratings of EF in a clinical sample of children with SLI referred to our clinic (Speech and Language Centre of Royal Dutch Kentalis) aged 5–12 years. Furthermore we addressed possible developmental and gender differences. Age related and gender differences in EF behaviours have been reported in TD, but have never been examined in

children with SLI, to our knowledge. Finally we examined whether behavioural ratings of EF and performances on two EF tasks (i.e. Working Memory and Shifting) are associated in children with SLI. Our specific research questions were as follows:

- 1. Do parental and/or teachers' ratings of EF differ significantly in children with SLI versus TD peers?
- 2. Are there developmental differences in EF behaviours in children with SLI?
- 3. Are there gender differences in EF behaviours in children with SLI?
- 4. Are the behavioural ratings of EF and performances on cognitive EF tasks associated in children with SLI?

METHODS

Participants

A total of 237 children with SLI aged 5 to 12 years participated in this study: 157 boys and 80 girls. The descriptive statistics of the participants are shown in Table 1. The mean age of the children was 7 years and 7 months (SD 20 months, range 5;00 to 12;06 years). The sex ratio of our sample (with more boys included than girls), is comparable to the sex ratio in other studies on children with SLI (Robinson, 1991; Tomblin et al., 1997; Tallal et al., 1989). Most of the children went to mainstream schools (84%). 16% visited special schools for children with learning problems or schools for children with severe speech, language or hearing disorders. The proportion of parents in our sample with a bachelor or master degree was 39% compared to 28% in the Dutch working population (Statistics Netherlands, 2012). Most children were monolingual Dutch (92%) and spoke Dutch as their mother tongue at home and at school.

All children attended a special multidisciplinary treatment program for children with severe language problems. Diagnosis was based on extensive clinical and psychometric assessment by speech and language pathologists; persistent difficulties specific to language production at the phonological, word and/or sentence level were shown in all cases. Children's nonverbal IQ had to be in the normal range, and a diagnosis of hearing disorder, neurological disorder, ADHD, or autism spectrum disorder should be absent. Prior to the start of the treatment program all children received daily support for their speech or language problems for at least one year without substantial development as result of their persistent problems.

Language tasks that are used to characterise the language disorder of the included children are nonword repetition (Rispens and Baker, 2012) and naming (Renfrew, 1991; Jansonius and Borgers, 2009). Nonword repetition was used to assess the phonological working memory of the SLI group. 71% of the included children diverged from their TD peers in repeating complex nonwords of 2, 3 4 and 5 syllables and showed severe

Table 1 Descriptive statistics sample

Sample characteristics					
Number	237				
Mean age (years;months)	7;07				
Age range(years;months)	5;0 – 12;06				
Number of boys/girls	157				
NWR, mean (SD)	12.8 (6.8)				
Range NWR	0-36				
Naming, mean (SD)	31.9 (7.4)				
Range naming	12 - 47				
PPVT-III-NL, mean (SD)	95.1 (11.1)				
Range PPVT-III-NL	71 – 126				

Note. SD, standard deviation; NWR, Nonword Repetition Test (Rispens and Baker, 2012); Naming Test (Renfrew, 1991; Jansonius and Borgers, 2009); PPVT-III-NL, Peabody Picture Vocabulary Test-III-NL.

(-2SD) to moderate (-1 SD) deficits in their phonological working memory. 65% of the children showed deviant naming skills in comparison to TD children. The passive word knowledge of the SLI group, measured by the Peabody Picture Vocabulary Test (PPVT-III-NL), is age-appropriate, although the scores of the SLI group are significantly lower compared to TD children (see Table 1).

Behavioural data are collected by parental and teachers' ratings of EF (i.e. BRIEF) and cognitive measures by a short neuropsychological assessment. Data were collected from July 2009 until December 2012 on all children who visited the Speech and Language Centre of Royal Dutch Kentalis. All parents were informed about the purpose of data collection and those from who we had informed consent participated in this study. The SLI subjects were selected on age and the fact that the teacher questionnaire was returned. We were not able to collect all data in the clinical setting. Therefore the number of participants can differ.

Material

Behavioural ratings.

The BRIEF is a standardised rating scale for parents and teachers designed to measure executive function behaviours of children aged 5–18 years old (Gioia et al., 2000). We used the Dutch version, which contains of 75 items (Huizinga and Smidts, 2011). Each item pertains to specific everyday behaviour, relevant to EF. Parents and teachers were asked to indicate how often the child displayed a given behaviour in the past 6 months, based on three possible responses 'never', 'sometimes' and 'often'. The items are categorized in eight no overlapping theoretically and empirically derived clinical scales that measure different aspects of EF: Inhibit, Shifting, Emotional control, Initiate, Working

memory, Plan/organise, Organisation of materials, and Monitor. The eight clinical scales form two broader indexes: Behaviour Regulation Index (BRI) and the Metacognition Index (MI). The sum of the first three scales is referred to as the BRI; the composite score of the five remaining scales is referred to as the MI. Based on these two composite scores an overall global EF score (i.e. Global Executive Composite, GEC) is calculated. The Dutch version of the BRIEF has been normed and validated. The sample of the normative study included 847 typically developing children without a history of psychiatric disorder and/or learning disorder. The internal consistency and test-restest stability of the Dutch version are high to very high: Cronbach's a varying between.78 and.96 for the different scales, and Intraclass Coefficients between 0.73 and 0.95.

Cognitive measures

Besides the behavioural ratings of EF we also included two cognitive EF tasks for working memory and shifting. Performance of the SLI group was compared to the normative means of these tests. Working memory: Digit recall of the Dutch version of the Wechsler Intelligence Scale for Children-III: Children have to repeat a sequence of numbers forward, and in the second part backwards (Kort et al., 2002). Shifting: Creature Counting TEA-Ch: Children have to repeatedly switch between two relatively simple activities of counting upwards and counting downwards (Manly et al., 2007). They are asked to count aliens in their burrow, with occasional arrows telling them to change the direction in which they are counting. Time taken and accuracy are scored in this subtest.

RESULTS

Group comparison

The descriptive statistics for the behavioural measures of EF (BRIEF) are shown in Table 2. Performance of the SLI group was compared to the normative mean T-score of 50 in a one-sample t-test. Using the Bonferroni method, which divides the level of significance by the number of dependent variables, each t-test was tested at the 0.005 level. In addition, we calculated the percent of children with clinically impaired BRIEF scores (i.e. T-scores of 65 or higher). A percentage of 5% would be expected in a normal distribution.

For the parental rating scale, the SLI group performed significantly higher than the normative mean score of 50 on the Working memory scale: t (190) = 2.90, p = 0.004. The percentage of children with SLI with BRIEF T-scores in the clinically impaired range varied from 2 to 12%. These results show that the parents of children with SLI report significantly more problems in everyday life on the behavioural measure of working memory compared to the normative mean.

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Table 2

Descriptive statistics EF behaviours (BRIEF) for total SLI group

	Teachers (n=237)		Parents	(n=191)		
	Mean	SD	% elevated *	Mean	SD	% elevated *
BRIEF scale scores						
Inhibition	52	10	13	46	11	7
Shifting	54	11	15	46	11	7
Emotional control	52	11	15	45	11	6
Initiate	64	20	40	45	10	3
Working memory	66	18	43	52	11	12
Planning/organization	54	14	17	44	9	2
Organization of materials	50	11	10	41	11	3
Monitor	55	12	21	45	12	6
BRIEF index scores						
Behavioural regulation	53	11	12	45	12	7
Metacognition	60	16	31	45	10	3
Global executive	55	12	18	44	11	5

Note. EF, executive functioning; SD, standard deviation; * = T-score of 65 or higher (1,5 SD above the mean)

For the teachers' rating scale, the SLI group performed significantly higher than the normative mean score on all clinical scales and indexes, except the Organization of materials scale: Inhibition t (236) = 3.74, p = 0,000; Shifting t (236) = 5.91, p = 0.000; Emotional control t (236) = 3.01, p = 0.003; Initiate t (236) = 10.90, p = 0.000; Working memory t (236) = 13.52 p = 0.000; Plan/organize t (236) = 4.86, p = 0.000; Monitor t (236) = 0.16, p = 0.876; Behavioural regulation Index t (236) = 4.75, p = 0.000; Metacognition Index t (236) = 9.59, p = 0.000; Global Executive Index t (236) = 6.71, p = 0.000. The percentage of children with SLI with BRIEF T-scores in the clinically impaired range varied from 10 to 43 %. The results indicate that teachers of children with SLI report significantly more problems on almost all of the behavioural measure of EF compared to the normative mean.

Developmental related differences

Given that only teacher reports showed significant problems on a broad range of EF behaviours in children with SLI, we based the next analyses on the teachers rating scales. First, we explored whether there are age differences in EF behaviours in children with SLI, based on teacher reports.

Table 3 shows the means and standard deviations with respect to age in the SLI group. The results of the MANOVA showed that there are significant differences in means between age groups: Wilks' $\Lambda = 0.71$, F(44,851) = 1.80, p < 0.001, $\eta^2 = 0.82$. There are significant differences between the age groups on the Initiate and Working memory

	5 years (n=43)	6 years (n=55)	7 years (n=50)	8 years (n=43)	9-12 years (n=46)	Fa	Partial η²
BRIEF Scale scores		(11-3-3)	(11-3-0)	(11-13)	(
Inhibition	55 (10)	52 (9)	54 (11)	51 (9)	50 (12)	1.60	-
Shifting	54 (13)	54 (13)	56 (10)	55 (11)	54 (10)	0.30	-
Emotional control	52 (11)	53 (11)	52 (11)	52 (11)	52 (12)	0.07	-
Initiate	67 (21)	69 (21)	69 (21)	62 (20)	53 (9)	5.96 **	0.09
Working memory	68 (20)	66 (18)	73 (20)	64 (16)	56 (11)	5.89 **	0.09
Plan/organize	55 (16)	56 (14)	55 (14)	55(15)	50 (7)	1.57	-
Organization of materials	50 (11)	49 (13)	52 (12)	51 (12)	49 (8)	0.81	-
Monitor	56 (14)	55 (13)	56 (13)	54 (12)	51 (9)	1.77	-
BRIEF index scores							
Behavioural regulation	54 (12)	53 (11)	54 (11)	53 (9)	52 (13)	0.36	-
Metacognition	62 (18)	62 (17)	64 (16)	59 (16)	52 (8)	4.02 *	0.07
Global executive	57 (14)	56 (13)	57 (13)	54 (11)	52 (8)	1.53	-

Table 3 Age related Change on the BRIEF Clinical Scales and Indices(Teacher report)

Note. ^a df = (4, 232), * p < 0.05, ** p < .001

scale of the Metacognition index, with younger children showing more problems than older children.

Gender differences

The results of the MANOVA showed that there are significant differences in means of the boys and girls, with boys showing more problems in EF behaviours than girls: Wilks' $\Lambda = 0.78$, F(11,225.) = 5.67, p < 0.000, $\eta^2 = 0.22$. Table 4 shows the means and standard deviations with respect to gender. Boys scored higher on all the clinical scales of the Metacognition index, except for the Monitor scale (i.e., Initiate, Working memory, Plan/ organize, and Organization of materials).

Relations between behavioural ratings and performance based EF tasks

To explore the relations between behavioural ratings of EF and the performances on EF tasks for children with SLI, the correlations were computed between the Digit recall and Creature counting tasks and the Working memory scale, the Shifting scale and the index scores of the BRIEF (see Table 5). The working memory task Digit recall correlated significantly with the Working memory scale (r = -0.18), the Behavioral Regulation Index (r = -0.17), the Metacognition Index (r = -0.20) and the Gobal Executive Composite (r = -0.21). The shifting task Creature counting only correlated significantly with the BRIEF Shifting scale (r520.19). These results show that performance on the working memory test is associated with a broad spectrum of EF behaviours and that performance on the

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Table 4

Gender differences on the BRIEF Clinical Scales and Indices(Teachers report)

	Boys (n=157)	Girls (n=80)	Fa	Partial η²
BRIEF Scale scores				
Inhibition	53 (10)	52 (11)	0.61	-
Shifting	54 (11)	56 (12)	1.12	-
Emotional control	51 (11)	54 (13)	2.45	-
Initiate	69 (22)	55 (10)	28.14 **	0.11
Working memory	70 (19)	58 (10)	26.70 **	0.10
Plan/organize	56 (16)	50 (6)	10.08 *	0.04
Organization of materials	51 (13)	48 (6)	4.31 *	0.02
Monitor	55 (13)	53 (10)	2.20	-
BRIEF index scores				
Behavioural regulation	53 (11)	54 (12)	0.36	-
Metacognition	63 (18)	54 (9)	16.34 **	0.07
Global executive	56 (13)	53 (8)	3.04	_

Note. ^a *df* = (1,235), * *p* < 0.05, ** *p* < .001

Table 5

Correlation between cognitive and behavioural EF measures

BRIEF scores	Digit recall (n= 188)	Creature counting (n = 122)
Shifting	_	-0.19*
Working memory	-0.18*	-
Behavioural regulation	-0.17 *	-0.08
Metacognition	-0.20 **	-0.03
Global executive	-0.21 **	-0.06

Note. **p* <.05 (2-tailed); ** *p* <.01 (2 tailed)

a) WISC-Digit recall: Missing data because the task was not administered by children younger than 6 years. b) Creature Counting: Missing data because the task was not administered by children younger than 6 years. For children of 6 and 7 years administration had to be stopped often because children did not pass the practice trials.

shifting task is associated with the behavioural rating of shifting. It should however be noted that, although some significant correlations were found, all correlations should be considered low.

DISCUSSION

The purpose of this study was to examine behavioural parental and teachers' ratings of EF in a clinical sample of children with SLI aged 5–12 years. We also asked whether developmental and/or gender differences exist in the EF behaviours of children with SLI and how behavioural ratings of EF and performances on cognitive EF tasks interrelate for these children.

The results show that children with SLI have difficulties in a broad range of EF behaviours, based on teacher reports. The scores on the BRIEF of children with SLI based on teachers rating scales are significantly higher than the normative mean on the Inhibition, Shifting, Emotional control, Initiate, Working memory, Plan/organise and Monitor scales. Additionally, a greater than expected percentage of the children scored in the elevated range on all scales. 'Working memory' and 'Initiate' are the most impaired since more than one third of the children had scores in the clinical range on these scales. In contrast, parents only reported more problems on the Working memory scale. Differences between parents and teachers ratings of behaviours have been reported before. A meta-analyses on cross-informant correlations for instance, showed that the correlation between parents and teachers ratings on various measures of behaviours in children was low (Achenbach et al., 1987; Garrison and Earls, 1985). We also calculated the mean correlation between parents and teachers rating in our own sample and found a correlation of 0.37, also indicating a low correlation. It suggests that parents and teachers perceive behaviours of children differently, including EF behaviours. A possible explanation for the current finding of teachers reporting more EF problems in children with SLI than parents could be that more demands are placed on EF skills in classroom due to higher expectations on behaviour and performance.

Obviously, the high occurrence of problems to initiate goal directed behaviour in the classroom for children with SLI is a topic to discuss. While working memory problems have been reported before in previous studies, problems to initiate behaviour have not (Montgomery et al., 2010). Item analyses of the scale 'Initiate' show that it often taps getting started with a school task. The reason for high scores on this scale could be that the child has not understood the verbal instruction of the teacher. Another explanation could be that the child is not capable of initiating his goal directed behaviour, since inner speech is not adequately developed to start the required activities during school time (Winsler et al., 2003). Exploration of the ability to initiate goal directed behaviour in the classroom could be a potentially valuable direction for future research on children with SLI.

Our second research question concerned possible developmental differences in EF behaviours in children with SLI. Overall, based on teacher reports, older children showed less problems in EF behaviours than younger children. More specifically, we found sig-

nificant differences between the age groups on the Initiate and Working memory scale. These findings are in line with previous studies in typically developing children showing a decrease in EF problems when children grow older (Gioia et al., 2000; Huizinga and Smidts, 2011). One possible explanation for the current finding is that young children with SLI have a delayed development of the prefrontal lobe. With age the neural delay may decrease in these children. Future longitudinal research with brain techniques, like fMRI or ERP, should shed more light on this issue in children with SLI.

With regard to our third research question concerning possible gender differences in EF behaviours in children with SLI, we found boys to show more problems in EF functioning than girls. The results, based on teacher reports showed significant differences between boys and girls on the Initiate, Working memory, Plan/organise, and Organization of materials scales. Girls showing less EF problems compared to boys have also been a common finding in previous studies in typically developing children (Gioia et al., 2000; Huizinga and Smidts, 2011).

The final research question was whether behavioural ratings of EF and performances on cognitive EF tasks are associated in children with SLI. The results showed that performance on the working memory test correlated significantly with the behavioural rating of working memory, and performance on the shifting task correlated significantly with the behavioural rating of shifting. Besides its intercorrelation with the BRIEF Working memory scale, the working memory task Digit recall correlated also significantly with the Behavioral Regulation, the Metacognition Index and the Gobal Executive Composite. Overall, these results indicate that teachers behaviour ratings of EF of children with SLI are to some extent associated with their performance on EF tasks. Or stated more specifically, performance on the working memory task is associated with a broad range of EF behaviours and performance on the shifting task is associated with the behavioural rating of shifting. However, although some significant correlations were found, all correlations should be considered low, which is a common finding in previous research. Limited correlations between the BRIEF and cognitive measures of EF, have been reported in several studies for both TD children and other clinical groups (Anderson et al., 2010; Mahone et al., 2007; Vriezen and Pigott, 2002). This might indicate that, like in other populations, performance of children with SLI on standardised EF tests is at best weakly associated with their EF behaviours in daily life.

It is important to notice that this study had a number of weaknesses. One possible limitation concerns the number of missing values in the EF tasks data. Although the total sample of this study was large enough to draw some conclusions, data of performance on the EF tasks were not included for all children, especially for the younger age groups. This is thus a potentially valuable direction for future research. Additionally, only two performance-based tasks of EF were included (i.e., working memory and shifting). For future research, it could be valuable to add standardized tasks of other EF, like inhibition and planning. Another possible limitation on the present study is that we did not include different linguistic measures. Inclusion of measures of more linguistic domains could provide information on the associations between language and different subfunctions of EF.

In closing, the present findings may have some implications for clinical practice and future research. Based on the findings of impairments in a broad range of EF behaviours in classroom, it might be a valuable addition to include measures of EF in the assessment of children with SLI. Specifically the inclusion of rating scales of EF during daily life could be important, to assure ecological validity and complement information gleaned from cognitive measures. Regarding the treatment of children with SLI, another issue concerns whether it could be sensible to include interventions that focus on EF. Interventions and strategies aimed at reducing working memory demands, improving planning and reducing the effects of inhibition problems might be helpful. It also could be a potentially valuable direction for future research to examine whether treatment of both linguistic and EF impairments of children with SLI results in better outcomes than more traditional interventions. In general future research is needed to shed more light on the complex connections between language and executive processes in children with SLI.

Chapter 4

Working memory and executive function behaviours in young children with SLI

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ABSTRACT

The present study compared the performances of young children with specific language impairment (SLI) to that of typically developing (TD) children on cognitive measures of working memory (WM) and behavioral ratings of executive functions (EF). The Automated Working Memory Assessment was administered to 58 children with SLI and 58 TD children aged 4 and 5 years. Additionally, parents completed the Behavior Rating Inventory of Executive Function-Preschool Version. The results showed the SLI group to perform significantly worse than the TD group on both cognitive and behavioral measures of WM. The deficits in WM performance were not restricted to the verbal domain, but also affected visuospatial WM. The deficits in EF behaviors included problems with inhibition, shifting, emotional control, and planning/organization. The patterns of associations between WM performance and EF behaviors differed for the SLI versus TD groups. WM performance significantly discriminated between young children with SLI and TD, with 89% of the children classified correctly. The data indicate domain general impairments in WM and problems in EF behaviors in young children with SLI. Attention should thus be paid to WM — both verbal and visuospatial — and EF in clinical practice. Implications for assessment and remediation were discussed.

INTRODUCTION

There is growing evidence that besides linguistic factors, non-linguistic factors may contribute to the problems associated with specific language impairment (SLI) (Bishop, 2006; Montgomery et al., 2010). One factor that has been implicated is working memory (WM) (Archibald & Gathercole, 2006a; Lum et al., 2011; Montgomery, 2010). More recently, limitations on other executive functions (EF) have also been shown in children with SLI (Im-Bolter et al., 2006; Henry et al., 2011). Evaluation of WM and EF may thus contribute to assessment of children and early identification of SLI (Conti-Ramsden & Durkin, 2012; Petrucelli et al., 2012). Early identification of SLI and determination of the child's strengths and weaknesses can then facilitate intervention. However, most previous studies focused on the role of WM and EF in school-aged children with SLI, and research in preschool children is still very limited. In the present study, we therefore addressed the role of WM and EF in young children with SLI. We examined whether the performances on the different components of WM and behavioral ratings of EF differed significantly for young children with SLI versus TD peers.

SLI and working memory

The acquisition of language is a major milestone in children's development. While the development of most children's language unfolds automatically, other children show marked problems or delays. A diagnosis of specific language impairment (SLI) is made when language problems are encountered and can be characterized as a failure to make normal progress without further evidence of underlying intellectual, neurological, social, or emotional impairment (Bishop, 2002, 2006). SLI can affect different linguistic domains including phonological, morphological, lexical and grammatical domains. The language profile of children with SLI often changes with age and development; changes can occur both within and across linguistic domains (Bishop, 2006; Leonard, 1998). SLI is a persistent disorder that affects language abilities in childhood and adolescence, or even into adulthood (Brizzolara et al., 2011; McKinley & Larson, 1989). Children with SLI are also at risk for less successful academic outcomes as well as behavioral, emotional, and social difficulties (Conti-Ramsden et al., 2009; St Clair et al., 2011).

WM refers to the structures and processes used to temporarily store and manipulate information. The conceptualization that has been mostly used in research on children with SLI is the multicomponent WM model of Baddeley (Baddeley & Hitch, 1974; Baddeley, 2003). In this model, a central executive (CE) system is assumed to be linked to three subsystems: the phonological loop, the visuospatial sketchpad, and the episodic buffer. The CE is responsible for the coordination and control of activities in WM. This system has limited attentional capacity and thus requires attentional control. The phonological loop and visuospatial sketchpad are so-called "slave" systems and responsible

for the temporary storage of verbal and visuospatial information, respectively. The episodic buffer is a relatively recent addition to the model and assumed to involve the binding of information from multiple sources together into chunks (Baddeley, 2003). Other conceptualizations of WM concentrate more specifically on the executive and attentional aspects. For instance, Engle et al. (1999) have suggested that WM capacity is limited by the ability to control attention and that this ability might, in fact, entirely explain the individual differences observed in WM. In the Embedded-Processes model of Courage and Cowan (2009), WM is assumed to reflect the activation of information that is in the focus of attention from long-term memory.

Strong links have been found between WM limitations and SLI (Archibald & Gathercole, 2006a; Bishop, 2006; Montgomery et al., 2010). The evidence nevertheless suggests that the WM problems exhibited by children with SLI are diverse and may involve different components of the WM system (Montgomery et al., 2010). The functioning of the storage components can be assessed using tasks that require the serial recall of information. Verbal versions require the retention of words, digits, or letters; the visuospatial versions require the retention of visual patterns or figures. The functioning of the CE component of the WM system can be assessed using tasks that require significant processing activity in addition to storage (i.e., complex memory span tasks). In one common complex listening span task, for example, the child must make a judgment about the meaning of each sentence in a series of sentences but also remember the last word of each sentence in the order of the sentences presented.

A widely accepted account of the deficits associated with SLI is the so-called phonological storage deficit hypothesis (Archibald & Gathercole, 2007; Baddeley, 2003; Bishop, 1996; Coady & Evans, 2008; Gathercole & Baddeley, 1990) and the underlying assumption that a specific deficit in the temporary storage of novel phonological information underlies SLI. In young children with SLI, deficits in verbal storage are widely reported in studies of nonword repetition (i.e., the repetition of unfamiliar or nonexistent words that thus require phonological processing on the part of the respondent) and digit recall (Gray, 2003, 2006; Conti-Ramsden, 2003; Horohov & Oetting, 2004). Between 3 and 6 years of age, children with SLI perform significantly worse than age-matched peers on both such tasks. Nonword repetition performance is even hypothesized to be a reliable marker of SLI in young children. It differentiates between children with and without SLI from the age of 2;06 (years;months) with good results in terms of sensitivity, specificity, and overall accuracy (Chiat & Roy, 2007; Gray, 2003, 2006).

In addition to these constraints on verbal storage, substantial deficits have been reported for verbal CE. Children with SLI consistently show relatively more impairments on verbal complex memory span tasks that combine verbal storage with either verbal or visuospatial processing than on straightforward verbal storage tasks (Archibald & Gathercole, 2006b; Briscoe & Rankin, 2007). It is suggested that deficits in verbal storage

age, twinned with general processing limitations, underlie the SLI impairments on verbal complex memory span tasks (Archibald & Gathercole, 2006b). However, some controversy exists about the nature of the processing limitations of children with SLI. Some authors assume that these limitations are caused by slower processing, the so-called generalized slowing hypothesis (Kail, 1994). This hypothesis is supported by several studies showing children with SLI to have slower reaction times both in verbal and visuospatial tasks (Miller et al., 2001; Schul et al., 2004; Tallal & Piercy, 1973). Other findings indicate however that children with SLI especially struggle under conditions of high processing loads, indicating reduced processing capacity (Ellis Weismer, 1996; Fazio, 1998; Hoffman & Gillam, 2004; Montgomery, 2002).

In contrast to the verbal domain, the visuospatial domain of WM has been less extensively investigated in children with SLI and the results are ambiguous at best (Alloway & Archibald, 2008; Archibald & Gathercole, 2006a; Montgomery et al., 2010). Despite this ambiguity and the lack of consensus regarding the role of visuospatial WM in the speech and language of children with SLI, several authors continue to assume that the WM deficits are limited to the verbal domain. This is because children with SLI and their TD peers have been found to perform similarly on visuospatial storage and CE tasks (Alloway & Archibald, 2008; Archibald & Gathercole, 2006a, 2006b; Baird et al., 2009; Lum et al., 2011; Riccio et al., 2007; Williams et al., 2000). In contrast, the results of several other studies and a recent meta-analysis have yielded evidence suggesting that the WM deficit of children with SLI may extend to the visuospatial domain (Vugs et al., 2013). In young children with SLI, significant group differences have been reported for children with SLI versus TD children on a variety of visuospatial storage tasks, including pattern recognition memory, paired associates learning, pattern recall, picture recognition, and localization recall but not for spatial recognition (Bavin et al., 2005; Hick et al., 2005; Menezes et al., 2007; Nickisch & Von Kries, 2009). Longitudinal research by Hick and colleagues (2005) has further shown the performance of children with SLI (aged 3;03 to 4;05 years) on a pattern-recall task to develop slower than the performance of TD peers. Research on visuospatial CE has shown young children with SLI to perform significantly worse than TD peers on several tasks, including a spatial span task, space visualization task, and position-in-space task but not a spatial WM search task (Bavin et al., 2005; Marton, 2008).

If children with SLI also exhibit deficits in the visuospatial domain of WM, this suggests that their impairments are not restricted to language or verbal information. It would implicate more general limitations, thus challenging the specificity of SLI. Based on a twin study, Bishop (1994) also questioned the specificity of SLI as they found that SLI is not genetically distinct from less specific disorders where language impairments occur in the context of non-verbal limitations. A further domain-general account of SLI is provided by Ullman and Pierpont (2005), who propose that SLI is characterized by

abnormal development of brain structures that constitute the procedural memory brain system (procedural deficit hypothesis). This memory system serves both linguistic and non-linguistic functions, but is particularly important in the acquisition of grammar.

Finally, the episodic buffer or third subsystem assumed to compose the Baddeley's multicomponent WM model has been examined using sentence repetition tasks. Sentence repetition requires the integration of phonological information with semantic and syntactic information. Poor performance for children with SLI (compared to TD peers) on this task has been reported in several studies (Petrucelli et al., 2012; Redmond et al., 2011; Archibald & Joanisse, 2009). However, research on the episodic buffer of young children with SLI is limited.

Petrucelli and colleagues (2012) were among the first to examine the WM of young children with SLI in a multimodal context and thus using measures of the different components of WM. When they compared the performance of 5-year-old children with SLI on measures of the phonological loop, visuospatial sketchpad, central executive, and episodic buffer to that of TD children and late talkers, the children with SLI showed significantly poorer performance for the phonological loop and episodic buffer but not for the other components of WM.

SLI and executive functions

Executive function is a multidimensional construct that subsumes the processes responsible for purposeful, goal-directed behavior. EF is implicated in not only cognitive processes but also emotional responses and behavioral actions (Gioia et al., 2001; Miyake & Shah, 1999). Although some uncertainties remain about the *exact* components of EF, frequently postulated components are: inhibition, shifting, planning, fluency, and WM (Huizinga et al., 2006; Miyake et al., 2000). These inter-related processes function together to provide an integrated, supervisory control system (Stuss & Alexander, 2000).

Besides limitations in WM, significant group differences have been reported between children with SLI versus TD children on tasks of the following components of EF: inhibition, planning, and fluency. Limitations in EF shown by the children with SLI were not confined to verbal EF tasks, but also occurred for some nonverbal EF tasks. However, not all components of EF have been equally extensively studied and in some cases results are still somewhat contradictory. *Inhibition* refers to the ability to stop ongoing responses. Compared to their TD peers, children with SLI demonstrate reduced inhibition of prepotent responses in several studies (Bishop & Norbury, 2005b; Finneran et al., 2009; Im-Bolter et al., 2006; Marton et al., 2007). However, limitations in inhibition have not been confirmed by all studies investigating children with SLI to perform comparable to their TD peers on a go – no go task. A possible explanation for this finding might be that the inhibition task used in this study required inhibition of less dominant or automatic

responses than the tasks used in other studies. *Planning* is the ability to plan and organize activities and is typically measured using problem-solving tasks. Difficulties with planning have been found in children with SLI on Tower tests and a Sorting test (Henry et al., 2011; Marton, 2008). *Fluency* refers to the ability to generate new responses. In a recent study, children with SLI obtained significantly lower scores on both verbal and non-verbal fluency tests compared to their TD peers (Henry et al., 2011). Deficits in nonverbal fluency have not been consequently found in all studies, however. For instance, the study of Bishop and Norbury (2005a) showed the performance of children with SLI not to differ from that of TD children on two tasks of non-verbal fluency.

In contrast to the reported group differences on tasks of inhibition, planning and fluency, no group differences have been found on tasks of shifting. *Shifting* is the ability to switch the focus of attention between different activities. Children with SLI and their TD peers have been found to perform similarly on several shifting tasks, including the Trailmaking test and set-shifting tasks (Dibbets et al., 2006; Henry et al., 2011; Im-Bolter et al., 2006).

In addition to cognitive tasks, behavioral rating scales are often used to investigate EF in daily life (Anderson et al., 2002; Gioia et al., 2001). Hughes and colleagues (2009) compared the parental and self-ratings of EF for adolescents with SLI versus TD adolescents, and found more negative ratings of EF in general for the SLI group compared to the TD group with half of the parents of adolescents with SLI rating their child's EF abilities in the clinically impaired range. More recently, Wittke and colleagues (2013) studied executive functioning of preschool children with SLI. The results showed that the EF of children with SLI, aged 3 to 5 years, were rated significantly worse than those of their TD peers by both parents and teachers.

Assessment of WM and EF in young children

Although some studies investigated the role of the storage components of WM in young children with SLI, research on the other components of WM and EF in this age group is still very scarce. In general, the exploration of EF in young children has been minimal for long time. One reason for the limited number of studies is, that until recently, little was known about the development of EF in preschool children. However, in recent years it has been shown that the prefrontal cortex — the region of the brain that plays an important role in EF — undergoes enormous neurodevelopmental changes between the age of 3 and 6 years (Garon, et al., 2008; Luciana & Nelson, 1998). Different components of EF show different developmental trajectories. The basic components of basic EF (i.e., inhibition and WM) emerge during the first years of life. The ability to keep simple information in mind (i.e., WM) is present before the age of 6 months, for example (Courage & Cowan, 2009; Garon, 2008). The underlying structure of Baddeley's multicomponents WM model is in place by about the age of 4 years with related but separable components (Alloway

et al., 2006). Between 3 and 5 years of age, spurts in children's inhibition and WM have been observed and other components of EF such as planning and shifting can be seen to start developing at this time (Best et al., 2009; De Luca et al., 2008; Diamond, 1990; Epsy, 2004).

The majority of tasks used to study EF have been designed for use with adults and have thus not been suited for use with young children who may encounter problems with the instructions to start with. More recently, developmentally-sensitive tasks have been created and research on the WM and EF of young children has thus increased (Alloway & Gathercole, 2007; Alloway et al., 2006; Diamond, 1990). The verbal storage component of WM can now be reliably measured by the age of 2 years, for example, using nonword repetition tasks (Chiat & Roy, 2007, 2008). Alloway and Gathercole (2007) recently developed the Automated Working Memory Assessment (AWMA) to assess the different components of WM from the age of 4 years.

More general in the field of EF research is the question of the ecological validity of the EF tasks that are used. Standardized cognitive measures of EF have been widely criticized as not being sufficiently sensitive to the multidimensional nature of EF in daily life (Chavtor et al., 2006; Anderson et al., 2002). This is obviously an issue for the assessment of young children who are known to behave differently in unfamiliar contexts. That is, obtaining representative behaviors in clinical or research settings can be a major problem. To gain ecologically valid information on children's EF, it is thus suggested that information should be collected in different contexts and from different sources — including caregiver behavioral ratings of EF (Gioia et al., 2001). In a recent review of the assessment of language development in preschool children, Conti-Ramsden and Dunkin (2012) indeed advocate adoption of a multi-method, multi-informant approach. They also assert that information from caregivers may provide a more accurate assessment of young children's language problems. For the assessment of EF behavior in young children, Gioia and colleagues (2003) developed the Behavior Rating Inventory of Executive Function - Preschool Version (BRIEF-P). This guestionnaire was one of the first to provide developmentally appropriate methods to assess the multidimensional nature of the EF construct in young children. Research has shown it to be a reliable and valid measure of everyday EF (Mahone & Hoffman, 2007).

Present study

Given that research on the EF of young children with SLI is limited and that only a few studies have compared WM performance for young children with SLI versus TD children, the role of WM and EF in young children with SLI is not clear. In the present study, we therefore compared the WM performances and behavioral ratings of EF of young children with SLI to that of TD children. More specifically, we administered a battery of WM tests to assess the different components of the WM system according to Baddeley's

WM model. Additionally, we collected parental ratings of behaviors requiring a range of executive functions. We did this with children with SLI and TD children in the age range of 4- to-5 years. Our specific research questions were as follows.

- 1) Do WM performance and/or behavioral ratings of EF differ significantly for young children with SLI versus TD peers?
- 2) Do the performances on the different components of WM and behavioral ratings of EF correlate significantly for children with SLI and/or TD children?
- 3) Does WM performance discriminate between children with SLI and TD children?

METHODS

Participants

A total of 116 children aged 4- to 5-years participated in this study: 58 children with SLI (42 boys and 16 girls) and 58 age-matched TD peers (32 boys and 26 girls). The mean age of the children with SLI was 4;09 (SD = 7.41 months, range 4;0 to 5;11). The mean age of the TD peers was 4;11 (SD = 6.78 months, range 4;01 to 5;11). All of the children had average intelligence (85 or more on a nonverbal intelligence test, SON-R $2\frac{1}{2}$ -7) and were native speakers of Dutch (Tellegen & Laros, 1998). Any children with a diagnosed hearing impairment, neurological disorder, ADD/ADHD, or autism spectrum disorder were excluded.

The children in the SLI group were recruited from special language units (n = 52) or from special education schools (n = 6) in the Netherlands. All of them were receiving daily support for their speech or language problems. Diagnosis was based on extensive clinical and psychometric assessment by speech and language pathologists; persistent difficulties specific to language were shown in all cases. For most of the children, recent results for measures of language and nonverbal intelligence were available via their personal files. These results were included in the current study only when they were no more than six months old. Otherwise, assessment was repeated. Participants were included in our study when they performed 1.25 SDs or more below the mean on at least two language measures, following Tomblin et al. (1996). The language measures included the Peabody Picture Vocabulary Test-III-NL (Dunn & Dunn, 1997; Schlichting, 2005), the Reynell Developmental Language Scales (Reynell & Gruber, 1990; Eldik van et al., 2004), and tests of word and sentence development from the Schlichting Test for Language Production (Schlichting et al., 2003). The Dutch versions of these tests have all been normed. The SLI group means for expressive language were about 1.5 SDs below the standardized mean; 76% of the children with SLI performed 1.25 SDs below the mean on one of the expressive language measures; 62% performed 1.25 SDs below the mean on one of the receptive language measures; and 45% scored more than 1.25 SDs below the mean on three or more language measures.

The children in the control group were recruited from three middle-class schools in the Netherlands. The language measures examined for the control group were the *Peabody Picture Vocabulary Test-III-NL* (Dunn & Dunn, 1997; Schlichting, 2005) and the *Reynell Developmental Language Scales* (Reynell & Gruber, 1990; Eldik van et al., 2004). All of the control children performed in the normal range on both of these tests.

The SLI and control groups did not differ significantly with regard to age (ANOVA F(1,114) = 3.64, p = .059), nonverbal intelligence (ANOVA F(1,114) = 3.58, p = .061), or gender (Chi-square Test $X^2(1, N = 116) = 3.73$, p = .053). The descriptive statistics for the two groups of children are presented in Table 1. One-way analyses of variance (ANOVAs) confirmed that the SLI group had significantly lower scores on the language measures than the control group (PPVT-III-NL F(1,114) = 46.29, p < .001; Reynell F(1, 114) = 189.67, p < .001).

 Table 1

 Descriptive statistics for nonverbal intelligence and language measures

Measure	SLI (N = 58,	42 boys)	TD (N = 58, 32 boys)		
	Mean	SD	Mean	SD	
Age	57.03	7.41	59.44	6.78	
Non-verbal IQ (SON-R 21/2-7)	107.24	12.74	112.12	15.68	
PPVT-III-NL	90.86	14.86	107.05	10.37	
Reynell	84.17	12.38	112.13	12.54	
Schlichting WQ	79.14	12.59	-	-	
Schlichting ZQ	78.66	8.81	-	-	

Note. SLI = specific language impairment; TD = typically developing children; N = number of included children; SD = standard deviation; PPVT-III-NL = Peabody Picture Vocabulary Test-III-NL; WQ and ZQ Schlichting = word and sentence development Schlichting Test for Language Production

Procedure

All children were tested individually in a quiet room at their school or in a clinic. Written consent was obtained for participation in the present study from the parents of all children. Assessment took anywhere from two to four 45-minute sessions, depending on the availability of the selected language and nonverbal intelligence measures in the children's personal files. A short break was taken halfway through each session. In addition to the measures for nonverbal intelligence and language listed above, all of the children were administered the Dutch translation of the *Automated Working Memory Assessment (AWMA)* (Alloway, 2007). All of their parents completed the Dutch translation of the *Behavior Rating Inventory of Executive Function–Preschool Version (BRIEF-P)* (Gioia et al., 2003; Heijden van der et al., 2012). The AWMA was administered on a laptop. To start, the experimenter explained the task to the child. Next, practice trials were administered in which feedback was provided by the computer. If necessary, the experimenter repeated the practice items and thereby made sure that the child understood the task instructions. After the practice trials, all of the children were able to perform the trials individually without the help of the experimenter. All of the children completed the test battery, also in the order recommended. Nine of the parent questionnaires were not returned (6 for the SLI group; 3 for the control group).

Cognitive measures of WM

The AWMA (Alloway, 2007) is an automated, computerized assessment battery suitable for use with respondents who are 4 to 22 years of age. The AWMA has been validated and measures the different components of Baddeley's WM model (Gathercole & Pickering, 2000). The assessment battery includes twelve subtests which form four nonoverlapping composite scores that include three subtests of verbal storage, verbal CE, visuospatial storage, and visuospatial CE, respectively. The storage measures tap into the phonological loop or visuospatial sketchpad, depending on the nature of the information to be remembered. For the CE measures, the children must simultaneously store and process information. The processing activity is assumed to tap into the central executive component of the WM model.

Testing follows the same span procedure in all subtests. Following a practice session, a maximum of six sequences of increasing lengths are presented. The length of the sequences are increased by one after the child has correctly recalled four sequences of a particular length with a maximum of seven items for the CE tasks and nine items for the storage tasks. Testing is stopped when three sequences of a particular length are not recalled correctly. The children respond by pointing to the answer of their choice on the screen or by saying it aloud. The experimenter then imports their choice into the computer program.

Verbal storage

In the Digit recall task, the child must recall a sequence of digits in the right order. The digits can range from one to nine and are spoken at a rate of one digit per second. The sequences are randomly generated and no digits are repeated.

In the Word recall task, the child must recall a sequence of words in the right order. The words are monosyllabic, spoken at a rate of one syllable per second, and no words are repeated. When a substitution reflects the child's habitual articulation pattern for a phoneme, credit is given for the substitution and the recall of the item judged to be correct. In the Nonword recall task, the child must recall a sequence of nonwords in the right order. These nonwords are composed of the same phonemes as the words from the Word recall task. The nonwords are monosyllabic, spoken at a rate of one syllable per second, and no nonwords are repeated. As in the Word recall test, when a substitution reflects the child's habitual articulation pattern for a phoneme, credit is given for the substitution.

Verbal CE

In the Listening span task, the child is presented short sentences. The child must then judge whether the content of the sentence is correct (by saying "true" or "false") and remember the last word of the sentence. The number of sentences increases in length and the child must then recall the last words of the sentences in the correct serial order. The sentences have a simple subject-verb-object order and contain early developing vocabulary.

In the Counting recall task, the child first views red dots and blue triangles arranged in a box on the screen. The child is instructed to count the red dots, say the number aloud, and remember the total number of dots. After trials requiring the child to count the number of red dots, they must recall the number of red dots in the correct serial order.

The Backward digit recall task is the same as the Digit recall task except that the child must now recall the sequence of digits in the reverse order.

Visuospatial storage

In the Dot matrix task, a sequence of red dots is presented on a 4 x 5 grid. All of the dots appear in the grid for 2 seconds. The dots then disappear and the child must point to the position of each dot in the same serial order as presented.

In the Mazes memory task, a maze with a path drawn through it is presented to the child for 3 seconds. The same maze is then presented to the child without the path and the child must then draw the path of the line on the computer screen. Maze complexity is increased with the addition of more walls to the maze.

In the Block recall task, the child is presented a board with 9 randomly located cubes. A series of the tubes is then pointed to with an arrow. Thereafter, the child must point to the cubes in the same order.

Visuospatial CE

In the Odd-one-out task, a horizontal row of 3 boxes with a complex shape in each of them is shown to the child. The child must point to the shape that does not resemble the others. After trials in which the child identifies the odd shape, three blank boxes appear. The child is asked to point to the position of the boxes that contained the odd shapes in the correct serial order.

In the Mr. X task, the child is presented two Mr. X figures. The one on the left is wearing a yellow hat; the one on the right a blue hat. The figures are otherwise the same. Each Mr. X also has a ball in his hand, and the child must judge whether both figures have the ball in the same hand or not. In addition, the child must remember the position of the ball held by the figure with the blue hat (i.e., the figure on the right); the ball rotates to six possible positions in a circle. After trials in which the child must judge whether the ball is in the same hand or not, the Mr. X figures disappear and a circle of six dots appears. This circle reflects the possible positions of the ball. The child is asked to point to the position of the dots in the same as presented for Mr. X

In the Spatial span task, two identical shapes are presented to the child with a red dot above the right shape. The child must judge whether the two shapes are in normal or mirror image and to remember the location of the dot. The position of the dot rotates to one of three positions of a triangle. After trials requiring the child to judge the similarity of the shapes, they disappear and a triangle of three dots reflecting the possible positions of the previous dots appears. The child must point to the positions of the previous dots in the right order.

Behavioral measures of EF

The BRIEF-P is a standardized rating scale for parents and teachers designed to measure executive function behaviors of children aged 2 to 5 years old (Gioia et al., 2003, Heijden van der et al., 2012). The scale contains 63 items within five nonoverlapping theoretically and empirically derived clinical scales that measure different aspects of executive functioning: inhibition, shifting, emotional control, working memory, and planning/organization. The five clinical scales form three broader indexes of inhibitory self-control, flexibility, and emergent metacognition. An overall global EF score (i.e., global executive composite) is also calculated.

RESULTS

Group comparisons

The descriptive statistics for the cognitive measures of WM (AWMA) and behavioral measures of EF (BRIEF-P) are shown in Table 2. Performance of the SLI and TD groups were compared in multivariate analyses of variance (MANOVAs) and follow-up analyses of variance (ANOVAs). In addition, effect sizes were computed. The effect-size (*d*) is the difference between the mean of the control group and the SLI group divided by the pooled sample standard deviation. Effect sizes are considered small for *d* = .20, medium for *d* = .50, and large for *d* = .80 (Cohen, 1988).

8 Chapter 4

Table 2

Descriptive statistics for cognitive WM performance (AWMA) and EF behaviors (BRIEF-P)

	SLI		TD			
AWMA	М	SD	м	SD	F	d
Verbal storage: composite score	85.33	13.03	113.59	10.80	161.70	2.38
Digit recall	74.45	15.02	97.03	9.83	91.78	1.79
Word recall	92.81	11.31	115.76	9.04	145.58	2.26
Nonword recall	98.24	12.92	118.62	12.90	72.29	1.59
Verbal CE: composite score	89.59	9.38	113.31	12.99	127.13	2.11
Listening recall	98.09	11.32	122.68	10.96	141.33	2.23
Counting recall	87.63	11.25	109.69	15.40	77.51	1.65
Backward digit recall	89.88	7.86	101.21	13.91	29.16	1.01
Visuospatial storage: composite score	89.38	12.03	109.81	13.35	74.97	1.62
Dot Matrix	89.79	13.68	109.07	14.65	53.64	1.37
Mazes memory	93.85	11.05	110.09	11.79	58.61	1.43
Block recall	91.24	11.67	104.89	14.29	31.77	1.06
Visuospatial CE: composite score	95.74	13.84	115.14	13.45	58.57	1.43
Odd-One-Out	96.07	16.01	113.69	20.16	27.17	0.98
Mister X	94.29	13.09	110.59	18.26	30.51	1.03
Spatial recall	99.22	12.76	114.10	16.51	29.48	1.02
BRIEF-P	М	SD	м	SD	F	d
Global executive composite	59.92	14.52	48.48	10.47	22.44	0.92
Inhibition	58.89	12.69	49.91	12.04	14.38	0,73
Shifting	54.81	11.49	46.41	6.31	22.70	0.92
Emotional control	54.83	15.65	47.80	11.26	27.02	0.52
Working memory	63.94	14.54	50.84	11.69	27.02	1.01
Planning/organization	55.23	13.72	46.54	10.49	13.90	0.72

Note. SLI = specific language impairment; TD = typically developing children; M = mean; SD = standard deviation; F = ANOVA statistics; d = effect size; AWMA = Automated Working Memory Assessment; BRIEF-P = Behavior Rating Inventory of Executive Function–Preschool Version.

The SLI group performed significantly worse than the TD group on the cognitive measures of WM. We first conducted MANOVA investigating group differences on the four composite scores (i.e., verbal storage, verbal CE, visuospatial storage, and visuospatial CE). It showed a significant overall group effect: Wilks' $\Lambda = .37$, F(1,114) = 48.16, p .001, $\eta^2 = .63$. Follow-up ANOVAs were next conducted. Using the Bonferroni method, which divides the level of significance by the number of dependent variables, each ANOVA was tested at the .013 level. The outcomes for all four of the univariate comparisons were significant: verbal storage F(1,114) = 161.70, p < .001; verbal CE F(1,114) = 127.13, p < .001; visuospatial storage F(1,114) = 74.97, p < .001; visuospatial CE F(1,114) = 58.57, p < .001. The average effect size for the composite scores was d = 1.89. The largest composite effect size was found for verbal storage (d = 2.38).

Secondly, MANOVA investigating the group differences on the individual subtest scores also revealed a significant overall group effect: Wilks' $\Lambda = .31$, F(1,114) = 19.51, p < .001, $\eta^2 = .70$. All of the follow-up univariate ANOVAs (at .004 level) were significant: digit recall F(1,114) = 91.78, p < .001; word recall F(1,114) = 145.58, p < .001; nonword recall F(1,114) = 72.29, p < .001; listening recall F(1,114) = 141.33, p < .001; counting recall F(1,114) = 77.51, p < .001; backward digit recall F(1,114) = 29.16, p < .001; dot matrix F(1,114) = 53.64, p < .001; mazes memory F(1,114) = 58.61, p < .001; block recall F(1,114) = 31.77, p < .001; odd-one-out F(1,114) = 27.17, p < .001; mister X F(1,114) = 30.51, p < .001; and spatial recall F(1,114) = 29.48, p < .001. The average effect size for the differences between the performance of children with SLI and TD children on the individual subtests was d = 1.45. The largest effect size was observed for word recall (d = 2.26).

To control that intelligence, gender and age were not mediating performance on the cognitive measures of WM, multivariate analyses of covariance (MANCOVAs) and follow-up analyses of covariance (ANCOVAs) were next conducted for both the composite and individual subtest scores; nonverbal intelligence (IQ SON-R), gender and age were entered as covariates. Both the overall group effect on the composite scores (Wilks' Λ = .39, *F*(1,114) = 43.17, *p* < .001, η^2 = .62) and the univariate group effects for each of the composite scores at the .013 level (verbal storage *F*(1,114) = 137.72, *p* < .001; verbal CE *F*(1,114) = 112.73, *p* < .001; visuospatial storage *F*(1,114) = 60.78, *p* < .001; visuospatial CE *F*(1,114) = 46.65, *p* < .001) remained significant.

For the individual subtests, the overall group effect remained significant in the MAN-COVA (Wilks' $\Lambda = .33$, F(1,114) = 17.55, p < .001, $\eta^2 = .68$). And once again, all of the univariate ANCOVAs also showed significant group effects: digit recall F(1,114) = 75.73, p < .001; word recall F(1,114) = 120.54, p < .001; nonword recall F(1,114) = 65.23, p < .001; listening recall F(1,114) = 116.01, p < .001; counting recall F(1,114) = 72.00, p < .001; backward digit recall F(1,114) = 18.73, p < .001; dot matrix F(1,114) = 39.26, p < .001; mazes memory F(1,114) = 48.99, p < .001; block recall F(1,114) = 23.36, p < .001; odd-one-out F(1,114) = 18.98, p < .001; mister X F(1,114) = 24.93, p < .001; and spatial recall F(1,114) = 20.70, p < .001. The results indicate that even when intelligence, gender and age were taken into account, the SLI group performed significantly worse than the TD group on all the components of WM.

For the BRIEF-P behavioral measure of EF, an ANOVA investigating the group differences on the overall global EF score (i.e., global executive composite) revealed a significantly higher global EF score for the SLI group than for the control group: F(1,107) = 22.44, p = .002. This indicates more problems in EF behaviors in the SLI group compared to the TD group.

Table 3

MANOVAs investigating the group differences on the clinical scales of inhibition, shifting, emotional control, working memory, and planning/organization revealed a significant overall group effect: Wilks' $\Lambda = .86$, F(1,107) = 3.42, p = .007, $\eta^2 = .140$. Follow-up univariate ANOVAs showed significant group differences for all of the five clinical scales at a .01 level: inhibition F(1,107) = 14.38, p < .001; shifting F(1,107) = 22.70, p < .001; emotional control F(1,107) = 27.02 p < .001; working memory F(1,107) = 27.02, p < .001; planning/organization F(1,107) = 13.90, p < .001). The average effect size for the differences among the clinical scales was d = 0.78. The largest effect size was found for the clinical scale of working memory (d = 1.01). These results show the parents of children with SLI to report significantly more problems on all of the behavioral measure of EF compared to the parents of TD children, with medium to large effect sizes.

Relations between WM performance and behavioral ratings of EF

To explore the relations between the performances on the different components of WM and behavioral measures of EF for the SLI and TD groups of children, the correlations were computed between the WM composite scores from the AWMA, the overall global EF score from the BRIEF-P, and the clinical scales from the BRIEF-P (see Table 3). The correlations for the SLI group are displayed first and those for the TD group second.

For the SLI group, low correlations were generally found between the different components of WM and behavioral ratings of EF (r = -.004 to r = -.284). Only the correlation between the AWMA composite score of verbal storage and the BRIEF-P clinical scale of shifting was found to be significant.

For the TD group, the correlations varied between r = .004 and r = -.364. The AWMA composite score of verbal CE significantly correlated with the BRIEF-P overall global EF score (r = -.341) and the clinical scales of inhibition (r = -.348), working memory (r = -.364), and planning/organization (r = -.312). The highest correlation was between the verbal CE composite score and the clinical scale of working memory. The AWMA composite score of visuospatial CE significantly correlated with the BRIEF-P overall global

contraction between cognitive with performance (Awwink) and Er benaviors (blief 1)							
	Verbal storage	Verbal CE	Visuospatial	Visuospatial CE			
			storage				
Inhibition	.110 /066	.027 /348**	026 /244	006 /303*			
Shifting	284* /107	010 /192	004 /101	110 / .004			
Emotional control	021 /090	040 /092	021 /116	.060 /117			
Working memory	229 /192	134 /364**	270 /251	101 /348**			
Planning/organization	246 /150	044 /312*	217 /122	184 /338*			
Global executive composite	166 /144	059 /341*	151 /246	071 /339*			

Correlation between cognitive WM performance (AWMA) and EF behaviors (BRIEF-P)

Note. Correlations for SLI group displayed first; correlations for TD group displayed second. *p < .01
EF score (r = -.339) and the clinical scales of inhibition (r = -.303), working memory (r = -.348), and planning/organization (r = -.338). The highest correlation was between the visuospatial CE composite score and the clinical scale of working memory.

Identifying SLI

Table 4

Given that the measures of WM performance produced significant group differences with large effect sizes, we next explored if these measures could discriminate between young children with SLI and TD children. For this purpose, a discriminant analysis was conducted to determine whether performance on the composite scores from the AWMA (i.e., verbal storage, verbal CE, visuospatial storage, and visuospatial CE) could predict group membership. We also conducted a discriminant analysis to determine whether performance on the two language measures included in this study (i.e., PPVT-III-NL and Reynell) predicted group membership, to facilitate comparison. The results are presented in Table 4.

The first discriminant analysis explored the use of the four AWMA composite scores (entered together) as a classification function. The overall Wilks's lambda was significant, $\Lambda = .37$, X^2 (4, N = 116) = 112.71, p < .001. This shows the predictors to differentiate between the SLI and TD group. Based on this function, 90% of the children in our sample were correctly classified as SLI and 88% correctly classified as TD. Using the leave-one-out method (cross-validation) to assess how well this classification procedure would predict in a new sample, 88% of the children were next correctly classified as SLI and 86% correctly classified as TD. The sensitivity of this function is 88%, and the specificity is 90%. The positive likelihood ratio is 8.4, indicating that children with SLI are over 8 times more likely to have greater problems with WM performance than their TD peers.

The second discriminant analyses explored the use of the two language measures as classification functions. The Wilks's lambdas for both language measures were significant: PPVT-III-NL, $\Lambda = .71$, X^2 (1, N = 116) = 33.68, p < .001; Reynell $\Lambda = .38$, X^2 (1, N = 116) = 111.20, p < .001. These results show both of the language measures to successfully differentiate between the two groups of children. Based on the PPVT-III-NL, 67% of the

Classification of	Children as	SLI or TD in	Discriminant	Function	Analvsis
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Variable entered	Correctly classified SLI	Correctly classified TD	Sensitivity	Specificity	LR+	
Composite scores AWMA	52 (89.7%)	51 (87.9%)	88.1%	89.5%	8.4	
PPVT-III-R	39 (67.2%)	49 (84.5%)	81,2%	72,1%	2.9	
Reynell	51 (87.9%)	52 (89.7%)	89.5%	88.1%	7.5	

Note. LR+ = positive likelihood ratio; PPVT-III-R = Peabody Picture Vocabulary Test-III-NL

children in our sample were correctly classified as SLI and 85% correctly classified as TD. Based on the Reynell, these percentages were 88% and 90%, respectively. For the PPVT-III-NL, the sensitivity is 81%; the specificity is 72%; and the positive likelihood ratio is 2.9. For the Reynell, the sensitivity is 90%; the specificity is 88%; and the positive likelihood ratio is 7.5. The percentage of children correctly classified by the composite scores from the AWMA is comparable to the percentage of children correctly classified by the language measures.

In Table 5, we present the within-groups correlations between the predictors and the discriminant function of the AWMA composite scores as well as the standardized weights for this function. Verbal storage shows the strongest association with the discriminant function, followed by verbal CE, visuospatial storage, and visuospatial CE.

Predictors	Correlation coefficients with discriminant functions	Standardized coefficients for discriminant functions		
Verbal storage	0.90	0.65		
Verbal CE	0.80	0.39		
Visuospatial storage	0.62	0.14		
Visuospatial CE	0.54	0.02		

Table 5

Correlations and standardized coefficients of composite scores AWMA with discriminant functions

DISCUSSION

The purpose of this study was to determine if the performances of young children with SLI differ from that of TD children in terms of WM performance and EF behaviors. We also asked how the performances on the different components of WM and behavioral ratings of EF interrelate for children with SLI versus TD children and whether assessment that includes measures of WM performance discriminates between children with SLI and TD children?

With regard to our first question, namely whether WM performance and/or ratings of EF differ significantly for young children with SLI versus TD peers, we found children with SLI to perform significantly below their TD peers on all components of WM, including verbal storage, verbal CE, visuospatial storage, and visuospatial CE. The effect sizes for the different components all were large (varying from d = 1.43 to d = 2.38). We also calculated the effect sizes for the language measures included in this study, the PPVT-III-NL and Reynell, for comparison and found the effect sizes to be comparable to those for the measures of WM (d = 1.27 and d = 2.26, respectively). Taken together, these findings replicate previous findings showing clear impairments in verbal storage and verbal CE in children with SLI (Archibald & Gathercole, 2006b, 2007; Coady & Evans, 2008;

Gray, 2003; Montgomery et al., 2010). Reduced performance on visuospatial storage and visuospatial CE tasks has also been reported in most previous studies examining these in *young* children with SLI (Bavin et al., 2005; Hick et al., 2005; Menezes et al., 2007; Marton 2008). However, age-appropriate performance on visuospatial storage and visuospatial CE tasks has been shown in older children with SLI (Alloway & Archibald, 2008; Archibald & Gathercole, 2006a, 2006b; Baird et al., 2009; Lum et al., 2011; Riccio et al., 2007; Williams et al., 2000). Additionally, a recent study showed this to also be the case for young children with SLI (Petrucelli et al., 2012). The data on impairments in the visuospatial domain of WM in children with SLI are still not clear, thus.

Behavioral ratings of EF showed the parents of the young children with SLI to report significantly more problems relative to the parents of the TD children in our study. These included problems with inhibition, shifting, emotional control, WM, and planning/ organization. The effect sizes for the differences between the children with SLI and the TD children were medium to large on average (range of d = .44 to d = 1.01). The largest effect size for ratings of EF were found for WM. These results are in line with the results of Hughes and colleagues (2009) who found adolescents with SLI and their parents to report impaired EF behaviors during daily life.

Our second research question concerned the intercorrelations between performances on the different components of WM and behavioral ratings of EF for children with SLI versus TD children. The intercorrelations differed for the two groups of children. In the TD group, both verbal CE and visuospatial CE performance significantly correlated with behavioral ratings of EF in daily life. More specifically, both verbal CE and visuospatial CE performance correlated with the behavioral ratings of inhibition, WM and planning/ organization in the TD group. In contrast, in the SLI group, consistently low correlations were found for all of the components of WM with the ratings of EF in daily life; only the correlation between verbal storage performance and the behavioral rating of shifting proved significant. This pattern of findings suggests that the associations between WM performance and EF behaviors are less consistent and non-specific in young children with SLI compared to their TD peers. However, in general, limited correlations between the BRIEF and cognitive measures of EF, including WM, have also been reported in previous studies for both TD children and other clinical groups (Anderson et al., 2010; Mahone et al., 2007; Vriezen & Pigott, 2002). It is suggested that this findings are due to the lack of ecological validity of standardized cognitive measures of WM and EF (Chaytor et al., 2006; Anderson et al., 2002).

Our final research question was whether WM performance could adequately discriminate between young children with SLI versus TD peers. The composite scores from the AWMA, which measured the different components of WM, differentiated between the SLI and TD groups with 90% of the children in our sample correctly classified as SLI and 88% correctly classified as TD. The percentage of children classified correctly by the AWMA composite scores was comparable to the percentage classified correctly by the two language measures in the present study. Sensitivity and specificity were both high (i.e., 88% and 90%). The verbal storage component of WM demonstrated the strongest relationship with the discriminant function, followed by verbal CE, visuospatial storage, and then visuospatial CE. These results suggest that assessment of the cognitive measures of WM can help identify young children with SLI but that it is not sufficient on its own for accurate classification.

Taken together, the results of our study show that young children with SLI perform significantly below their TD peers on both cognitive and behavioral measures of WM. In addition to constraints on WM, the deficits in EF behaviors include problems with inhibition, shifting, emotional control, and planning/organization. The observed deficits in WM performance involved all the components of WM and were not restricted to the verbal domain; the visuospatial domain of WM was affected as well. Our results also show the patterns of associations between WM performance and EF behaviors to differ for children with SLI versus those with typical language development. Furthermore, WM performance and particularly verbal storage can adequately discriminate between young children with SLI versus typical language development.

Although consensus on the involvement of the visuospatial WM in SLI has not been found across studies to date, the current findings clearly suggest that *both* the verbal and visuospatial domains of WM are affected in young children with SLI. Stated more generally, this outcome suggests that SLI in young children may be associated with domain general impairments of WM. The impairments seem not to be completely specific to language or the processing of strictly verbal information.

Alternative explanations for this outcome are nevertheless available. One frequently offered explanation is that the visuospatial WM system may be intact but that the control of this system by the language system is problematic. This explanation hinges on whether the performance of the children on visuospatial WM tasks possibly reflects verbal mediation of visuospatial information, or genuinely reflects their visuospatial storage and processing, as we have assumed. Some experts have hypothesized that children with SLI indeed show *inefficient* verbal coding during visuospatial WM tasks (Ar-chibald & Gathercole, 2006b; Gillam et al., 1998). Due to their language problems, they may rely more on visual encoding when actually phonological codes are preferable or use less efficient verbal strategies. But such an explanation in terms of inefficient verbal coding is not likely to hold for the young children in our study because it is known that verbal coding does not emerge until around the age of seven (Gathercole et al., 1994). An alternative explanation of the domain general impairments of WM in young children with SLI must thus be sought.

Another possibility is that the visuospatial WM impairments of young children with SLI are a reflection of more general limitations on executive and attentional control. This

view is in line with accounts of WM that highlight the notion of limited executive and attentional resources (Courage & Cowan, 2009; Engle et al., 1999). Such a limitation can be expected to manifest itself on any task with a high processing load. Stated differently, young children with SLI can be expected to adequately process single bits of information but encounter problems when more complex information must be processed (Ellis Weismer, 1996; Fazio, 1998; Hoffman & Gillam, 2004; Marton & Schwartz, 2003; Montgomery, 2000, 2002). It is known that executive and attentional control greatly influence children's WM performance, and there is evidence for a stronger association between executive and attention processes and visuospatial WM than between these processes and verbal WM (Busch et al., 2005; Marton, 2008; Miyake et al., 2001).

The current finding of problems with EF together with previous documentation of attentional impairments in young children with SLI support the explanation of domain general WM impairments in terms of limitations on executive and attentional control (Finneran et al., 2009; Spaulding et al., 2008). This explanation nevertheless calls for further documentation of the exact roles of EF and attentional control in the WM performance — both verbal and visuospatial. The development of EF and attentional control over time should be documented, for example. These factors were, after all, still developing in the children included in the present study. The present findings cannot rule out that it only concerned delays in the development of these capacities. If factors like EF and attentional control are implicated, it might be specific to young children with SLI and thus not hold for older children.

Understandably, there has not been much research conducted on WM and EF of young children with SLI to date. The present study is one of the first to clearly document the WM performances and EF behaviors of young children with SLI. We used a validated and standardized test to assess the different components of WM. This multimodal approach permitted a more reliable assessment of each component than reliance on any single measure (Archibald & Gathercole, 2006a). Our study is also unique in the inclusion of behavioral ratings of EF. One possible limitation on the present study is that we did not include cognitive measures of EF. This is thus a potentially valuable direction for future research on young children with SLI. Several studies in older children with SLI have indeed revealed impairments on cognitive measures of EF, including inhibition, planning, updating, and fluency (Henry et al., 2011; Im-Bolter et al., 2006: Marton, 2008). Another possible limitation is that measures of the functioning of the episodic buffer component of WM were not included in the present study. The inclusion of such information might nevertheless be of value as impairments in this component of WM in young children with SLI have recently been reported (Petrucelli et al., 2012). Continued research on the cognitive and behavioral aspects of WM and EF of young children with SLI will provide greater insight into the relationships between linguistic and cognitive factors in language impairment.

In closing, the present findings have some potential implications for the assessment and treatment of young children with language problems. Although SLI can be reliably identified in preschool children, its diagnosis in clinical practice is sometimes difficult due to substantial variation in the range of normal language development (Conti-Ramsden & Durkin, 2012; Ellis Weismer & Evans, 2002). The present results suggest that WM measures, and particularly verbal WM measures, could be a valuable addition for the identification of young children with SLI. Furthermore, evaluation of WM and EF in young children with SLI can create more detailed profiles of the strengths and weaknesses of these children. Given the present finding of limitations on different components of WM, including the verbal and visuospatial domain, examination of WM within a multimodal approach is recommended. The WM deficits of young children experiencing language problems may not be restricted to verbal WM, and it is obviously important to know if the problems being experienced by the child are also visuospatial. In order to assure ecological validity and complement information gleaned from cognitive measures, the addition of parental ratings of the child's EF during daily life is recommended. More generally, the present findings indicate that the AWMA and BRIEF-P are efficient measures for detecting WM and EF limitations in young children.

For remediation, it is recommended that interventions should not focus on language alone but also address strategies used by the child to store and process both verbal and visuospatial information. It is also recommended that the adverse effects of impaired WM and EF be minimized during teaching and remediation by taking task demands (i.e., task complexity, amount of material, and possible distractors) into account. While the use of visual support is already a common support strategy for intervention with children with SLI, the current findings suggest that young children with SLI might not benefit as much from visual support as typically developing children do. This means that only certain types of visual support may be suited for use with young children with SLI, namely: simple visual information that does not exceed the child's WM capacity. In language impaired children with clear impairment of WM and/or EF, WM or EF training may be relevant (Klinberg et al., 2002; Prins et al., 2010). Finally, these clinical implications may be particularly important for those children showing limited response to traditional language intervention. Taking into account WM and EF in young children with SLI can create more detailed profiles of the strengths and weaknesses of these children and thus determine suitable interventions.

Chapter 5

Interactions between working memory and language in young children with SLI

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ABSTRACT

The underlying structure of working memory (WM) in young children with and without SLI was examined. The associations between the components of WM and the language abilities of young children with SLI were then analyzed. The Automated Working Memory Assessment and four linguistic tasks were administered to 58 children with SLI and 58 children without SLI aged 4 to 5 years. The WM of the children was best represented by a model with four separate but interacting components of verbal storage, visuospatial storage, verbal central executive (CE), and visuospatial CE. The associations between the four components of WM did not differ significantly for the two groups of children. However, the individual components of WM showed varying associations with the language abilities of the children with SLI. The verbal CE component of WM was moderately to strongly associated with all the language abilities in children with SLI: receptive vocabulary, expressive vocabulary, verbal comprehension, and syntactic development. These results show verbal CE to be involved in a wide range of linguistic skills; the limited ability of young children with SLI to simultaneously store and process verbal information may constrain their acquisition of linguistic skills. Attention should thus be paid to the language problems of children with SLI, but also to the WM impairments that can contribute to their language problems.

INTRODUCTION

There is growing evidence that non-linguistic factors contribute to the problems associated with what is known as specific language impairment (SLI) (Bishop, 2006; Montgomery et al., 2010). One factor that is frequently implicated is working memory (WM) (Archibald & Gathercole, 2006a; Lum et al., 2011; Montgomery et al., 2010). Several studies have indeed documented WM impairments in children with SLI, but there are still many questions to be answered about the exact role of WM in the limited language abilities of these children. The purpose of the present study was therefore to specifically investigate the interactions between WM and the language development of young children with SLI. To do this, we first examined the underlying structure of WM in young children with SLI and compared to this the underlying structure of WM in typically developing (TD) children. We then examined just how the different components of WM relate to the language abilities of young children with SLI. As early childhood is an important period for both the development of WM and various linguistic abilities, we took this period to be promising for investigating the interrelationships between WM and language abilities.

Working memory impairments in children with SLI

The acquisition of language is considered one of the milestones in the development of children. While the language of the majority of children develops automatically, there are also children who show marked problems or delays. When children encounter language problems that can be characterized as a failure to make normal progress without further evidence of underlying intellectual, neurological, social, or emotional impairment, a diagnosis of specific language impairment (SLI) is usually made (Bishop, 2002, 2006). SLI can affect different linguistic domains (i.e., phonological, morphological, lexical and grammatical domains) and the language profile often changes with age (Bishop, 2006; Leonard, 1998).

WM refers to the structures and processes used to temporarily store information, on the one hand, and manipulate it, on the other hand. Previous research has established that WM is a memory system composed of separate but interacting components. However, there is ongoing debate about the structure of WM in developing children, with a range of theoretical accounts available (Courage & Cowan, 2009; Engle, Tuholski, Laughin, & Conway, 1999; Miyake & Shah, 1999).

The theoretical account most frequently called upon in research on children with SLI is the multicomponent WM model of Baddeley (Baddeley & Hitch, 1974; Baddeley, 2003). In this model, a central executive (CE) system is assumed to link three subsystems: the phonological loop, the visuospatial sketchpad, and the episodic buffer. The CE system coordinates and controls activities in WM. It has a limited attentional capacity and requires attentional control. The phonological loop and visuospatial sketchpad are

so-called "slave" systems and responsible for the temporary storage of verbal and visuospatial information, respectively. The episodic buffer is a relatively recent addition to the model and responsible for the binding of information from multiple sources together into chunks (Baddeley, 2003).

Other theoretical accounts focus more specifically on the executive and attentional aspects of WM. For instance, Engle et al. (1999) have suggested that WM capacity is limited by the ability of people to control attention and that this ability might, in fact, entirely explain the individual differences observed in WM capacity. In the Embedded-Processes model of Courage and Cowan (2009), WM is assumed to reflect the activation of the information from long-term memory that is currently in the focus of attention. Another account has been provided by Shah and Miyake (1996), who suggest that WM resources are divided into separate pools for verbal and visuospatial information — pools that are independently capable of coordinating and keeping information active. According to this account, performance on WM tasks is largely explained by the efficiency of either verbal or visuospatial abilities.

The storage components of WM are usually assessed using tasks that require the serial recall of information. Verbal storage tasks require the retention of words, digits, or letters while visuospatial storage tasks require the retention of other patterns or figures. The CE components of WM are typically assessed using tasks that require significant processing activity *in addition to* storage (i.e., complex memory span tasks). Verbal CE tasks combine the storage of verbal information with simultaneous processing of information, while in visuospatial CE tasks processing activity is combined with the storage of visuospatial information. One example of such a verbal CE task is the so-called listening span task, in which the child must judge the meaning of each sentence in a series of sentences but *also* has to remember the first word of each sentence in the order of the sentences presented.

Up until recently, relatively little was known about the development of WM in young children. Recent research, however, has shown the development of WM to have already started during the first years of life and to undergo enormous neurodevelopmental changes between 3 and 6 years of age (Garon, et al., 2008; Luciana & Nelson, 1998). The ability to keep simple information in mind is already present before the age of 6 months (Courage & Cowan, 2009; Garon, 2008). Alloway and colleagues (2006) have shown a three factor model with independent verbal and visuospatial factors but a single, domain-general, WM factor to provide the best account of WM in TD children between the ages of 4 and 11 years. All of the components of this model correspond to the components of the model of WM advanced by Baddeley & Hitch (1974). The components are assumed to be in place by the age of 4, and the model has been found to be quite stable up until the age of 11 years. In children aged 4 to 6 years, however, the link between these in older children. The authors see this developmental dif-

ference as an indication that young children call upon executive resources (or, in other words, attentional control) for the performance of visuospatial storage tasks more than older children.

Research suggests that children with SLI can show impairments for the different components of WM (Montgomery et al., 2010). A widely accepted account of such deficits is the so-called phonological storage deficit hypothesis (Archibald & Gathercole, 2007; Baddeley, 2003; Bishop, 1996; Coady & Evans, 2008; Gathercole & Baddeley, 1990). As stated, deficits in the temporary storage of novel phonological information are assumed to underlie SLI. Much of the evidence for such phonological storage deficits comes from studies of nonword repetition (i.e., repetition of unfamiliar or nonexistent words that thus require phonological processing on the part of the respondent) and digit recall among young children with SLI (Gray, 2003, 2006; Conti-Ramsden, 2003; Horohov & Oetting, 2004). Between 3 and 6 years of age, that is, poorer performance of children with SLI relative to age-matched peers has been widely reported on both types of tasks. Performance on tasks requiring the repetition of nonwords has even been hypothesized to be a reliable marker of SLI in young children.

In addition to these constraints on verbal storage, substantial deficits have also been reported for verbal CE. Impairments on verbal complex memory span tasks, like for instance listening span tasks, have been reported in several studies (Archibald & Gathercole, 2006b; Briscoe & Rankin, 2007; Henry et al., 2012). Children with SLI consistently show more problems on tasks requiring a combination of verbal storage with the simultaneous processing of information than on straightforward verbal storage tasks. Based on these findings, it has been claimed that deficits in verbal storage together with general processing limitations underlie the SLI impairments observed for verbal complex memory span tasks (Archibald & Gathercole, 2006b). Research on performance on tasks of verbal CE in young children with SLI is understandably still scarce, however.

The visuospatial domain of WM has been less investigated in children with SLI and then with only ambiguous results at best (Alloway & Archibald, 2008; Archibald & Gathercole, 2006a; Montgomery et al., 2010). Several authors have asserted that the WM deficits of children with SLI are limited to the verbal domain. This is presumed because children with SLI have been found to perform comparable to their TD peers on visuospatial storage tasks and visuospatial CE tasks (Alloway & Archibald, 2008; Archibald & Gathercole, 2006a, 2006b; Baird et al., 2009; Lum et al., 2011; Riccio et al., 2007; Williams et al., 2000). In contrast, the results of several other studies and a recent meta-analysis have yielded evidence suggesting that the WM deficits of children with SLI may extend to the visuospatial domain, at least for some children (Henry et al., 2012; Vugs et al., 2013). Some authors hypothesized that children with SLI show problems in visuospatial WM tasks because of inefficient verbal encoding during these tasks (Archibald & Gathercole, 2006b; Botting et al., 2013; Gillam, 1998). In a recent study on the effect of verbal and

nonverbal task content in simple memory span tasks (i.e., storage tasks), Botting and colleagues (2013) found that children with SLI showed disadvantage on any task with "verbalizable" elements, suggesting that some (visuospatial) storage tasks have hidden verbal elements that children with SLI may find more difficult compared to TD peers.

In young children, significant group differences have been reported for children with SLI versus TD children on a variety of visuospatial storage and complex memory span tasks, including pattern recognition memory, paired associates learning, pattern recall, picture recognition, localization recall, spatial span, space visualization, and position-in-space but *not* for spatial recognition and a spatial WM search task (Bavin et al., 2005; Hick et al., 2005; Marton, 2008; Menezes et al., 2007; Nickisch & von Kries, 2009). Longitudinal research by Hick and colleagues (2005), moreover, has shown the performance of children with SLI (aged 3;03 to 4;05 years) on a pattern-recall task to develop slower than the performance of TD peers.

Recently, Petrucelli and colleagues (2012) examined the WM of young children with SLI in a multimodal context and drew upon the multicomponent WM model of Baddeley (2003) in doing this. They compared the performance of 5-year-old children with SLI to that of TD children and resolved late talkers on measures of the phonological loop, visuospatial sketchpad, central executive, and episodic buffer. The children with SLI showed significantly poorer performance for the phonological loop and episodic buffer but not for the other components of WM.

Associations of working memory with language abilities in SLI

The evidence for impairments of the different components of WM raises questions about the *exact* associations between these components and the language impairments of children diagnosed with SLI. Although research in this area is still scarce, some studies have specifically addressed the possible associations.

To start with, performance on verbal storage tasks has been proposed to be linked to word learning or vocabulary acquisition (Montgomery et al., 2010). Baddeley and Gathercole (1989) were the first to demonstrate a strong association between the functioning of the phonological loop and the acquisition of new words in TD children between 4 and 5 years. Since then, other studies have documented a similar association for children with SLI (Gathercole & Baddeley, 1990; Horohov & Oetting, 2004; Montgomery, 2002). In later research, however, Gathercole (2006) suggested that verbal storage deficits alone may not be sufficient to account for the diversity problems in language acquisition that are a hallmark of SLI.

In addition to problems with vocabulary development, children with SLI also tend to exhibit poor sentence comprehension (van der Lely, 1996). Some authors suggest that impairments in verbal CE may account for these language comprehension problems. In one study of offline language comprehension, for example, children with SLI performed

poorer on a verbal complex memory task compared to their TD age matched peers and comprehended fewer long sentences (Montgomery, 2000). Based on this finding, it was assumed that verbal CE deficits of children with SLI hinder language comprehension, because the additional information processing demands inherent in such comprehension paradigms, exceed their WM abilities. In another study comparing both the verbal and visuospatial WM performance of groups of children with expressive SLI, receptive-expressive SLI, and TD children, significant correlations were found between performance on a visuospatial storage task (i.e., recall of sequences of symbols) and measures of receptive language (Nickisch & von Kries, 2009). In addition, only the children with receptive-expressive SLI performed significantly poorer than the TD children on the test of visuospatial storage, which led the authors to underscore the importance of visuospatial storage capacity for language comprehension and for children with expressive-receptive SLI in particular.

Another linguistic ability presumed to be associated with limitations in WM, is that of linguistic awareness (i.e., judgments of grammaticality). Grammaticality judgment always imposes a WM demand in addition to evaluating linguistic competence. In a study of TD children, a significant linear relationship was found between WM and accuracy of grammaticality judgments for word order, present progressive tense, regular past tense, and third person agreement (McDonald, 2008). These findings were taken to suggest that the integration of verbal morphology with other information in a sentence places high demands on WM. Quite recently, Noonan, Redmond, and Archibald (2014) investigated the interrelations between WM deficits — measured using both verbal and visuospatial complex memory span tasks — and judgments of grammaticality by children with SLI. When they compared the grammaticality judgments of children with only language impairments and thus no WM deficits, children with deficits in both domains, and TD children, independent contributions of WM and language competence were found for performance on the grammaticality judgment task. The authors thus concluded that both WM and language competencies can influence language processing.

The present study

A previous study based on the current sample showed, similar to other recent studies, significant group differences between children with SLI and their TD peers on different components of WM (Montgomery et al., 2010; Vugs et al., 2014). In the current study, the associations between components of WM and the specific language abilities of young children with SLI are further examined. The following research questions were addressed.

- 1) What is the underlying structure of WM in young children with and without SLI?
- 2) How do the different components of WM relate to the language abilities of young children with SLI?

First, the underlying structure of WM in young children with SLI compared to TD peers is investigated. Multiple tasks measuring the different components of WM were called upon. Four models of WM were then tested in confirmatory factor analyses. Model 1 is a domain-specific model with two factors for all verbal tasks and all visuospatial tasks respectively. Model 2 involves four domain-specific factors: verbal storage, visuospatial storage, verbal CE, and visuospatial CE. Model 3 is a three-factor model that distinguishes two domain-specific factors (i.e., verbal storage and visuospatial storage) and a domain general CE factor incorporating both the verbal and visuospatial storage-plus-processing tasks. Model 4 resembles model 3 except that the variance on the verbal and visuospatial storage-plus-processing is not only represented by a common CE factor, but also by the verbal and visuospatial storage components. This model is based on previous research on the underlying structure of WM in TD children (Alloway et al., 2006). Second, the associations between the components of WM and language abilities of young children with SLI are investigated. Correlational analyses and regression analyses were conducted to identify significant interrelations.

METHODS

Participants

The sample included 116 children aged 4 to 5 years: 58 children with SLI (42 boys and 16 girls) and 58 age-matched peers showing typical language development (32 boys and 26 girls). The mean age of the children with SLI was 4;09 (SD = 7.41 months, range 4;0 to 5;11). The mean age of the TD children was 4;11 (SD = 6.78 months, range 4;01 to 5;11). All of the children had average intelligence as indicated by a score of 85 or more on a nonverbal intelligence test (SON-R $2\frac{1}{2}$ -7) (Tellegen & Laros, 1998). Any children diagnosed with a hearing impairment, neurological disorder, ADD/ADHD, or a autism spectrum disorder were excluded from the study.

The children in the SLI group were recruited from special language units of a Speech and Language Centre (n = 52) or from special education schools (n = 6) in the Netherlands. All of them were receiving daily support for speech or language problems. The diagnosis of SLI was based on extensive clinical and psychometric assessment by speech and language pathologists; persistent difficulties specific to language were shown in all cases. The children in the control group were recruited from three regular education schools in the Netherlands.

The SLI and control groups did not differ significantly with regard to age (ANOVA F(1,114) = 3.64, p = .059), nonverbal intelligence (ANOVA F(1,114) = 3.58, p = .061), or gender (Chi-square Test X² (1, N = 116) = 3.73, p = .053). The descriptive statistics for the two groups of children are presented in Table 1.

Measure	SLI (<i>n</i> = 58, 42 boys) Mean (SD)	TD (<i>n</i> = 58, 32 boys Mean (SD)
Age	57.03 (7.41)	59.44 (6.78)
Non-verbal IQ (SON-R 2½-7)	107.24 (12.74)	112.12 (15.68)

Table 1. Descriptive Statistics for Age and Nonverbal Intelligence

Procedure

All 116 children were administered the following tests: *SON-R 2½-7* non-verbal intelligence test (Tellegen & Laros, 1998); Dutch version of *Automated Working Memory Assessment (AWMA)* test (Alloway, 2007); Dutch version of the *Peabody Picture Vocabulary Test-III-NL* (Dunn & Dunn, 1997; Schlichting, 2005); and Dutch versions of the *Reynell Developmental Language Scales* (Reynell & Gruber, 1990; van Eldik et al., 2004). For most of the SLI group, the results of recent administrations of the aforementioned nonverbal intelligence and language tests were available in their personal files. In addition, most of the children in the SLI group had recently been administered two other language measures, namely the tests of word and sentence development from the *Schlichting Test for Language Production* (Schlichting et al., 2003). These results were included in the current study only when they were no more than three months old. Otherwise, assessment was repeated.

All of the children were tested individually in a quiet room at their school or in the clinic. Written consent was obtained for participation in the present study from the parents of the children. Assessment required anywhere from two to four 45-minute sessions, depending on the availability of the intelligence and language measures in the children's files. A short break was taken halfway through each 45-minute session.

Measures of working memory

The AWMA (Alloway, 2007) is an automated, computerized assessment battery suitable for use with respondents 4 to 22 years of age. The AWMA has been validated and measures the different components of Baddeley's model of WM (Gathercole & Pickering, 2000). The assessment battery includes twelve subtests that form four nonoverlapping composite scores with thus three subtests for each of the following: verbal storage, verbal CE, visuospatial storage, and visuospatial CE. The storage measures tap into the phonological loop or visuospatial sketchpad, depending on the nature of the information to be remembered. The CE measures require respondents to simultaneously store and process information; this processing activity is thus assumed to tap into the CE component of the WM model.

The same procedure was followed for all subtests. After an initial practice session, a maximum of six sequences of increasing length are presented. The lengths of the se-

quences are increased by one after the respondent has correctly recalled four sequences of a particular length with a maximum of seven items for the CE tasks and nine items for the storage tasks. Testing is stopped when three sequences of a particular length are not recalled correctly. Responding is done by pointing to the answer of choice on the computer screen or by stating the answer out loud. In the latter case, the experimenter registers the choice in the computer.

Verbal storage

In the Digit Recall task, the respondent must recall a sequence of digits in the right order. The sequence of digits can range from one to nine and is spoken at a rate of one digit per second. The sequences are randomly generated and no digits are repeated in a sequence.

In the Word Recall task, the respondent must recall a sequence of words in the right order. The words are monosyllabic and spoken at a rate of one syllable per second with no words within the same sequence. When a substitution reflects the respondent's habitual articulation pattern for a phoneme, credit is given for the substitution and recall of the item judged to be correct.

In the Nonword Recall task, the child must recall a sequence of nonwords in the right order. The nonwords are composed of the same phonemes as the words in the Word Recall task. The nonwords are monosyllabic and spoken at a rate of one syllable per second with no repetition of the same nonwords. As in the Word Recall test, when a substitution reflects the respondent's habitual articulation pattern for a phoneme, credit is given for the substitution and recall of the item judged to be correct.

Verbal CE

In the Listening Span task, the respondent is presented short sentences. The respondent must then judge whether the content of the sentence is correct (by saying "true" or "false") and also remember the first word of the sentence. The number of sentences increases and the respondent must recall the first word of each sentence in the order of sentence presentation as part of the task. The sentences follow a simple subject-verbobject word order and contain early developed vocabulary.

In the Counting Recall task, the respondent first views red dots and blue triangles arranged in a box on the screen. The respondent is instructed to count the red dots, say the number aloud, and remember the total number of dots on each trial. Following completion of the trials requiring the respondent to count the number of red dots, they are asked to recall the numbers of red dots in the same order as the trials were presented.

The Backward Digit Recall task is the same as the Digit Recall task except that the respondent must now recall the sequence of digits in the reverse order of trial presentation.

Visuospatial storage

In the Dot Matrix task, a sequence of red dots is presented in a 4 x 5 grid. All of the dots appear in the grid for 2 seconds. The dots then disappear and the respondent must point to the position of each dot in the same order as presented.

In the Mazes Memory task, a maze with a path drawn through it is presented to the respondent for 3 seconds. The same maze is then presented to the respondent without the drawn path, which the respondent must now draw on the computer screen. Maze complexity increases with the addition of more walls to the maze.

In the Block Recall task, the respondent is presented a board on the computer with 9 randomly located cubes. A series of cubes is then pointed to with an arrow. The respondent is asked to point to the cubes in the same order as they were presented.

Visuospatial CE

In the Odd-One-Out task, a horizontal row of 3 boxes with a complex shape in each box is shown to the respondent. The respondent must point to that shape which does not resemble the others. After an increasing number of trials in which the respondent identifies the odd shape, three blank boxes appear. The respondent is asked to point out the position of those boxes that contained the odd shapes in the order that they were previously presented.

In the Mr. X task, the respondent is presented two Mr. X figures. The one on the left is wearing a yellow hat; the one on the right a blue hat. The figures are otherwise the same. Each of the figures also has a ball in their hand. The respondent must judge whether both figures have the ball in the same hand or not. In addition, the respondent must remember the position of the ball held by the figure with the blue hat (i.e., the figure on the right); the ball rotates to six possible positions in a circle. After the trials in which the respondent must judge whether the balls are in the same hand or not, the Mr. X figures disappear and a circle of six dots appears. The respondent is then asked to point to the position of the dots — reflecting the possible positions of the balls being held — in the same order as presented for Mr. X on the right.

In the Spatial Span task, two identical shapes are presented with a red dot above the right shape. It is a complex span task, in which the respondent must judge whether the two shapes are in normal or mirror image of each other and also remember the location of the dot. The position of the dot rotates to one of three positions of a triangle. After completion of the trials requiring the respondent to judge the similarity of the shapes, the shapes disappear and a triangle composed of three dots reflecting the possible positions of the previous dots appears. The respondent must point to the positions of the previous dots in the same order that they were presented.

Measures of language abilities

Receptive vocabulary

Receptive vocabulary was assessed using the Dutch version of the *Peabody Picture Vocabulary Test-Third edition* (PPVT-III-NL) (Dunn & Dunn, 1997; Schlichting, 2005). The PPVT is a widely used, norm-referenced test of word comprehension for ages 2;06 to 90 years. The test follows a multiple choice format in which a single word is presented orally. The child must then correctly identify which of four pictures represents the word by pointing or verbalizing the number of the picture. The PPVT has been validated for the Dutch population.

Expressive vocabulary

Expressive vocabulary was assessed using the Dutch version of the Word Development Test of the *Schlichting Test for Language Production* (Schlichting et al., 2003), which has been standardized for children between 1;09 and 6;03. Expressive vocabulary skills are measured by asking the child to name objects or pictures. The Schlichting Test for Language Production has been normed and validated for the Dutch population.

Verbal comprehension

The Reynell Developmental Language Scales (RDLS) constitute a verbal comprehension test designed for use with children 1;02 to 6;03 (Reynell & Gruber, 1990; Eldik van et al., 2004). The standardized Dutch version of the test, which has been normed and validated for the Dutch population, was used. The test evaluates language comprehension abilities with increasing levels of difficulty. In the first three sections, the child has to comprehend simple instructions that require them to identify objects or pictures (e.g., Where is the ball?). In the following seven sections, the items involve brief instructions for the child to comprehend and carry out (e.g., Put the spoon in the cup).

Syntactic development

To assess sentence development, we used the Dutch version of the Sentence Development Test from the *Schlichting Test for Language Production* (Schlichting et al., 2003). This test can be used with children 1;09 to 6;03 to assess their syntactic development. Knowledge of syntactic structures is determined by having the child repeat sentences of increasing difficulty.

Statistical analyses

The intercorrelations between the WM tasks were first calculated for the total sample, the SLI group, and the TD group. We then conducted confirmatory factor analysis (CFA) using the Lisrel software (Jöreskog & Sörborn, 1996) to examine the structural organization of WM. CFA is a statistical method used to test the goodness of fit of a theoretical

model to the empirical data (Hair et al., 1998). Several fit indices were used to test the overall fit of the proposed model. Chi Squared (X^2) is a goodness-of-fit measure that compares the covariance of a proposed model to the observed covariance. For a good fit according to a chi-square analysis, the outcome value should be small and nonsignificant — showing a match between the theoretical and empirical models (Ullman, 2001). However, for larger samples, it is difficult to obtain a nonsignificant chi square value due to the sensitivity of this statistic to sample size (Kline, 1998). The Comparative Fit Index (CFI) and Normed Fit Index (NFI) were therefore also used to compare the hypothesized model to a baseline model in which the latent variables of the model are assumed to be uncorrelated and covariances are fixed to 0. To indicate a good fit, these indices should be equal to or higher than .95 (Hu & Bentler, 1999). The Root Mean Square Error of Approximation (RMSEA) is an index that attempts to remove the effects of the degrees of freedom and sample size. For this statistic, a value of .08 or lower is considered acceptable, with a value lower than .05 indicating a good fit (McDonald & Ho, 2002). In order to compare the different theoretical models, Chi Square difference tests were conducted (McDonald & Ho, 2002). To whether the factor loadings in the measurement model were invariant across the SLI and TD groups, we also used Chi Square difference tests.

To examine the interrelations between the measures of WM and the children's language abilities, Pearson correlations were calculated and regression analyses were conducted. For the regression analyses, a stepwise method was adopted, which minimized the number of variables included and controlled for collinearity: in each step, only those variables that accounted for the largest percentage of variance (i.e., significant variables) were included in the regression model. To control for the possible effects of nonverbal intelligence and age, these variables were entered in Step 1 of the regression analyses.

RESULTS

Underlying working memory structure

As can be seen from Table 2, all of the intercorrelations between the measures of WM were positive and significant *for the total sample* with values ranging from .393 to .815. The correlations for the SLI and TD groups are displayed, separately, in Table 3. *For the SLI group*, the following was found. Within the verbal storage tasks (i.e., Digit Recall, Word Recall, and Nonword Recall), significant correlations manifested themselves between Digit Recall and Word Recall (r = .673, p < .01) and between Word Recall and Nonword Recall, and Backward Digit Recall), only the correlation between Counting Recall and Backward Digit Recall), only the correlation between Counting Recall and Backward Digit Recall (r = .471, p < .01). Within the visuospatial storage tasks (i.e., Dot Matrix, Mazes Memory, and Block Recall), all three intercorrelations were significant:

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Table 2	
Correlations between Working Memor	y Tasks Scores for Total Group

	1	2	3	4	5	6	7	8	9	10	11	12
1. Digit Recall	-											
2. Word Recall	.815	-										
3. Nonword Recall	.576	.709	-									
4. Listening Recall	.633	.703	.647	-								
5. Counting Recall	.685	.625	.460	.672	-							
6. Backward Digit Recall	.598	.511	.501	.623	.647	-						
7. Dot Matrix	.571	.570	.491	.674	.618	.548	-					
8. Mazes Memory	.474	.525	.495	.645	.541	.499	.604	-				
9. Block Recall	.526	.509	.400	.583	.529	.504	.675	.514	-			
10. Odd-One-Out	.441	.560	.433	.569	.543	.509	.655	.583	.404	-		
11. Mister X	.464	.488	.450	.511	.465	.505	.566	.622	.452	.599	-	
12. Spatial Span	.483	.453	.393	.596	.589	.531	.604	.560	.451	.626	.568	-

Note. All correlation are significant at p < 0.01

Table 3

Correlations between Working Memory Task Scores for SLI Children and TD Children

	1	2	3	4	5	6	7	8	9	10	11	12
1. Digit Recall	-	.602**	.318*	.464**	.437**	.592**	.346**	.106	.408**	.292*	.176	.308*
2. Word Recall	.673**	-	.416**	.398**	.233	.398**	.127	.171	.173	.233	.184	.071
3. Nonword Recall	.249	.518**	-	.329*	.163	.469**	.256	.302*	.152	.071	.192	.014
4. Listening Recall	.174	.320*	.399**	-	.505**	.661**	.578**	.528**	.432**	.519**	.397**	.551**
5. Counting Recall	.518**	.585**	.001	.200	-	.554**	.543**	.323*	.362**	.553**	.331*	.539**
6. Backward Digit Recall	.415**	.189	.055	.114	.471**	-	.535**	.419**	.456**	.566**	.429**	.450**
7. Dot Matrix	.314*	.419**	.176	.340**	.230	.202	-	.496**	.687**	.461**	.483**	.556**
8. Mazes Memory	.181	.182	.098	.229	.199	.179	.323*	-	.297*	.481**	.580**	.518**
9. Block Recall	.292*	.401**	.166	.357**	.299*	.200	.414**	.401**	-	.330*	.271*	.474**
10. Odd-One- Out	.068	.392**	.277*	.096	.002	.010	.579**	.328.*	.086	-	.520**	.579**
11. Mister X	.355**	.347**	.328*	.123	.104	.253	.355**	.373**	.376**	.435**	-	.522**
12. Spatial Span	.304*	.414**	.416**	.228	.223	.313*	.378**	.270*	.031	.439**	.309*	-

Note. SLI group correlations in lower triangle; TD group correlations in upper triangle. *p <.05, ** p < .01

Dot Matrix with Mazes Memory (r = .323, p < .05), Dot Matrix with Block Recall (r = .414, p < .01), and Mazes Memory with Block Recall (r = .401, p < .01). Within the visuospatial CE tasks (i.e., Odd-One Out, Mister X, and Spatial Recall), all of the intercorrelations were again significant: Odd-One-Out with Mister X (r = .435, p < .01), Odd-One-Out with Spatial Recall (r = .309, p < .05). For the *TD group*, all of the intercorrelations within the verbal storage tasks, within the visuospatial Storage tasks, and within the visuospatial CE tasks were significant. The values ranged from .318 to .602 for verbal storage, .505 to .661 for verbal CE, .279 to .687 for visuospatial storage, and .520 to .579 for visuospatial CE.

When we calculated the goodness-of-fit indices for the four models of WM tested in our study for the total group of children (see Table 4), the results showed models 1 and 3 to not provide a good fit for the data. For model 1, the chi-square index was highly significant; the NFI was less than .95; and the RMSEA exceeded .08. Although the fit indices for model 3 were all above .95, the chi-square value was highly significant, suggesting a poor fit, and the RMSEA also exceeded .08. Both models 2 and 4 provided an adequate fit for the data in our study (see Figure 1). Although the chi-square values were significant, the values of the CFI and NFI were all above .95 and the RMSEA did not exceed .08 for the two models, which shows them both to provide an adequate fit. However, the goodness of fit indices for model 4 were not as good as those for model 2. A chi-square difference test of the two models then showed the fit of model 2 to be significantly better than the fit of model 4 ($\Delta X^2 = 11.57$, df = 3, p = .009). A four factor model of WM thus provides the best account of our data. Significant and positive correlations were found between all of the factors in model 2, moreover: Verbal storage – Verbal CE r = .87, p < .0001, Verbal storage –Visuospatial storage r = .75, p < .0001, Verbal storage – Visuospatial CE r = .70, p < .0001, Visuospatial storage – Visuosptial CE r = .92, p < .0001, Visuospatial storage – Verbal CE r = .92, p < .0001, and Verbal CE – Visuospatial CE r = .86, p < .0001.

Next, we calculated the goodness-of-fit indices for the four models of WM for the SLI and TD groups separately (see Table 5). For the SLI group, none of the models showed an good fit for the data; for all models the chi-square index was highly significant; the CFI and NFI were less than .95; and the RMSEA exceeded .08. For the TD group, the fit indices for model 2 were both above .95, but the chi-square value was highly significant

 Table 4

 Goodness-of-Fit Statistics for Four Models of Working Memory (Total Sample)

coouness of the statistics for four models of working memory (fotal sumple)								
Model	X²	df	Р	CFI	NFI	RMSEA		
1	130.61	53	.0000	.967	.945	.112		
2	73.27	48	.0108	.989	.971	.069		
3	109.69	51	.0000	.975	.954	.099		
4	84.84	45	.0003	.984	.966	.080		





Figure 1. Path models based on models 2 and 4 for total sample of children.

and the RMSEA exceeded .08, suggesting a poor fit. The other models all showed highly significant chi-square values; CFI and NFI less than .95; and RMSEA above .08, indicating an inadequate fit.

GOC	Goodness-of-Fit statistics for Four models of wim for SEI Children and TD Children									
Мо	del	X²	df	Р	CFI	NFI	RMSEA			
1	SLI	106.23	53	.0000	.784	.680	.147			
	TD	100.41	53	.0000	.922	.851	.124			
2	SLI	106.94	48	.0000	.809	.714	.146			
	TD	68.40	48	.0038	.971	.953	.092			
3	SLI	118.95	51	.0000	.779	682	.152			
	TD	89.70	51	.0007	.936	.867	.114			
4	SLI	107.66	45	.0000	.796	.712	.155			
	TD	67.44	45	.0168	.963	.900	.093			

Table 5 Goodness-of-Fit Statistics for Four Models of WM for SLI Children and TD Children

Given that model 2 provided the best fit to the data in the total group of children, we furthermore tested the factor loadings of this model for invariance across the SLI and TD groups. They were found to not differ significantly ($\Delta X^2 = 12.76$, df = 8, p = .124). The path coefficients between each of the factors in the model (i.e., model 2) are displayed separately for the SLI and TD groups in Figure 2. The correlations between all of the factors for the SLI group were significant and positive with exception of the correlation between Verbal CE and Visuospatial CE: Verbal storage – Verbal CE r = .54, p < .0001, Verbal storage – Visuospatial storage r = .57, p < .0001, Verbal storage – Visuospatial CE r = .60, p < .0001, Visuospatial storage – Visuospatial CE r = .85, p < .0001, Visuospatial storage – Visuospatial CE r = .23, p < .2247. For the TD group, the correlations between all of the factors were both significant and positive: Verbal storage – Verbal CE r = .77, p < .0001, Verbal storage – Visuospatial St





Figure 2. Separate path models based on Model 4 for young SLI children and TD children.

Interactions between working memory and language abilities

To explore the interrelations between the components of WM and the language abilities of the children with SLI, we again calculated a number of correlations (see Table 6). Based on the findings just summarized for the present sample — findings showing the WM of the children in our study to be best represented by a four factor model, we decided to correlate the four composite memory scores from the AMWA for verbal storage, verbal CE, visuospatial storage, and visuospatial CE with the four domains of language ability assessed as well (i.e., receptive vocabulary, expressive vocabulary, verbal comprehension, syntactic development). The results showed the intercorrelations to range from .024 to .533. The verbal storage component of WM significantly correlated with both receptive vocabulary (r = .306) and syntactic development (r = .375). The verbal CE component of WM significantly correlated with all four language abilities: receptive vocabulary (r =.533), expressive vocabulary (r = .459), verbal comprehension (r = .375), and syntactic development (r = .447). All of these correlations were moderate to strong, with the highest correlation occurring for the verbal CE component with the children's receptive vocabulary skill. The visuospatial storage component of WM significantly correlated with expressive vocabulary although this correlation was small (r = .282). The visuospatial CE component did not significantly correlate with any of the children's language abilities.

conclutions between comp	onents of white and Et	inguage nonnes	ioi sei cimarcii	
	Verbal storage	Verbal CE	Visuospatial storage	Visuospatial CE
Receptive vocabulary	.306**	.533**	.057	.131
Expressive vocabulary	.082	.459**	.282*	.115
Verbal Comprehension	.024	.375**	.192	.113
Syntactic development	.375**	.447**	.183	.264

Correlations between Com	ponents of WM and Langu	age Abilities for SLI Childrer

Note. *p <.05, ** p < .01

Table 6

To further explore the interrelations between the different components of WM and the children's language abilities, we next determined whether performance on the different components of WM could predict the children's language abilities. To control for the possible influence of nonverbal intelligence and age, we also took into account these variables. In four stepwise regression analyses with performance on one of the four language tasks as the criterion variable in each of the analyses, nonverbal intelligence and age were entered in Step 1, and the four composite scores from the AWMA as WM predictor scores in Step 2. Table 7 summarizes information for Step 2 of each regression. We first found the verbal CE component of WM and nonverbal intelligence to be significant predictors of *receptive vocabulary*: $\Delta R^2 = .147$, F(1,55) = 13.360, p = .001. The other components of WM (i.e., verbal storage, visuospatial storage and visuospatial CE)

	Total R ²	ΔR^2	β
Receptive vocabulary			
	.395	.147**	
Constant			-11.750
Nonverbal IQ			.356**
Verbal CE			.409**
Expressive vocabulary			
	.239	.138**	
Constant			12.391
Verbal CE			.396**
Verbal comprehension			
	.470	.064*	
Constant			20.748
Nonverbal IQ			.302*
Verbal CE			.270*
Syntactic development			
	.200	.200**	
Constant			40.999**
Verbal CE			.447**

 Table 7

 Results of Stepwise Regression Analyses for SLI Children

Note. For each regression nonverbal intelligence and age were entered in Step 1 (note: Step 1 of each model is not shown), and the four composite scores of the AWMA in Step 2. The information provided about Step 2 of each model involves the total variance accounted for (total R^2), change in R^2 , and the standardized beta values for each predictor.

 $^{*}\beta$ <.05, $^{**}\beta$ < .01

and age were not included in this regression model as these variables did not explain a significant percentage of the observed variance in receptive vocabulary. In addition, the verbal CE component and nonverbal intelligence were found to be significant predictors in the regression analyses of *verbal comprehension*: $\Delta R^2 = .064$, F(1,55) = 4.520, p = .038. For both the children's exepressive vocabulary and syntactic development only the verbal CE component was found to be a significant predictor in the regression analyses: *expressive vocabulary* $\Delta R^2 = .138$, F(1,55) = 9.963, p = .003; *syntactic development* $\Delta R^2 = .200$, F(1,56) = 14.010, p < .001. The verbal storage, visuospatial storage, and visuospatial CE components of WM as well as nonverbal intelligence and age were not included in the regression models for the children's expressive vocabulary or syntactic development because these components did not explain a significant percentage of the observed variance in these particular language abilities.

Taken together, these results indicate that the verbal CE component of WM was a significant predictor of expressive vocabulary and syntactic development, even when

controlling for the effects of nonverbal intelligence and age. The verbal CE component was also a significant predictor of receptive vocabulary and verbal comprehension, but for these language abilities nonverbal intelligence was an additional significant predictor.

DISCUSSION

On the basis of our analysis of the underlying structure of WM in addition to the interactions between WM and language in young children and particularly children with SLI, we can now answer our two research questions. The first concerned the similarities and differences in the underlying structure of WM in young children with and without SLI. In order to answer this question, four theoretical models of WM were tested using CFA. The first model was a two factor model that distinguished between verbal and visuospatial memory. The second model consisted of four domain-specific factors: verbal storage, visuospatial storage, verbal CE, and visuospatial CE. The third model was a three-factor model that consisted of two domain-specific components of WM — verbal storage and visuospatial storage — and a domain general CE component that incorporates both verbal and visuospatial storage-plus-processing tasks. The fourth model was comparable to the third model except that the variance in the verbal and visuospatial storage-plusprocessing tasks was not only represented by a common CE factor, but also by the verbal and visuospatial storage components.

The CFA showed both models 2 and 4 to provide an adequate fit for the data from the total group of children. Comparison of the two models subsequently showed the four-factor model (model 2) to provide the best account of the interrelationships between the components of WM. According to this model, WM is composed of four separable components: a factor for verbal storage; a factor for visuospatial storage; a verbal high executive demand processing factor (verbal CE); and a visuospatial high executive demand processing factor (visuospatial CE). Both the storage and CE tasks show domain-specificity with performance on all the tasks appearing to be controlled by the efficiency in either the children's verbal or visuospatial abilities.

In previous studies of TD children using the same battery of tests for WM, Injoque-Ricle and colleagues (2012) similarly found the four-factor model to provide the best fit for the underlying structure of WM in children 11 years of age. However, for children 6 years of age, the authors found none of the WM models to have good fit indices. And for children 8 years of age, several models provided a good fit. These findings suggest that the underlying structure of WM is undefined and has yet to be fixed in children up until the age of around 11 years. Preschool children aged 4 and 5 years were not included in the study of Injoque-Ricle et al., but when Alloway and colleagues (2006) studied children with typical language development from the age of 4 to 11 years, a three-factor model — as represented by model 4 in the present study — was found to provide the best fit. They conclude that the interrelationships between the components of WM represented in the three-factor model are thus stable across the developmental period studied. Closer examination of the data of Alloway et al., however, shows not a three-factor model but instead a four-factor model — as in our present study — to provide the best fit for the data from the 4 to 6 year olds ($\Delta X^2 = 23.35$, df = 3, p < .001). For the 7-8 year olds in addition to the 9-11 year olds, the three-factor model indeed provided the best fit, but not for the younger children. These results thus support the results of the present study and indicate that the underlying structure of WM in preschool children aged 4-5 years is best captured by a four-factor model that has domain specificity for both the storage tasks and CE tasks.

Getting to the similarities and differences in the underlying structure of WM for young children with and without SLI, we found the factor loadings for the components of the four-factor model of WM to *not* differ significantly for the SLI group compared to the TD group. This shows the interrelationships between the components of WM to be comparable for the young children in the two groups despite their different language abilities. However, the correlations between the components of WM were not identical for the two groups. In the TD group, the correlations between all of the underlying components of WM were significant and positive; in the SLI group, the correlation between the verbal CE component and visuospatial CE component was *not* significant. For the TD group, moreover, the correlation between the verbal and visuospatial CE components was high with these components sharing 72% of variance, while this was only 5% for the SLI group. These findings suggest that verbal and visuospatial CE tasks tap into more shared underlying cognitive skills in young children with TD language when compared to young children with SLI.

To answer our second research question, which addressed how the components of WM relate to the language abilities of specifically young children with SLI, correlations were first calculated. The results showed moderate to strong correlations of the verbal CE component of WM with all four language abilities (i.e., the children's receptive vo-cabulary, expressive vocabulary, verbal comprehension, and syntactic development). The verbal storage component of WM also showed significant associations with the receptive vocabulary and syntactic development of the children, but these associations were not as strong as those with the verbal CE component of WM. Finally, the visuospatial storage component of WM significantly correlated with the expressive vocabulary of the children in the SLI group, but this correlation was small.

The regression outcomes next showed the verbal CE component of WM to be a significant predictor of all four language abilities in the group of children with SLI: their receptive vocabulary, expressive vocabulary, verbal comprehension, and syntactic development. For expressive vocabulary and syntactic development, the verbal CE component was even a significant predictor when the effects of nonverbal intelligence and age were taken into account. However, nonverbal intelligence was identified as an additional significant predictor of the children's receptive vocabulary and verbal comprehension. The verbal storage and visuospatial components of WM did not explain a significant amount of variance.

The finding that the verbal CE component of WM is associated with development of a variety of language abilities, including some more complex abilities, is in keeping with a more general set of findings showing clear associations between performance on verbal CE tasks and the development of such language abilities as sentence comprehension and judgments of grammaticality (Montgomery, 2000; Noonan, et al., 2014; van der Lely, 1996). In this connection, Gathercole (2006) already has suggested that deficits in verbal storage *alone* may not account for the diversity of linguistic problems found to characterize children with SLI. It appears especially the verbal CE component of WM is involved in the acquisition of a broad range of linguistic skills. Learning a language, involving the processes of learning various linguistic skills, appears to tax constantly the ability of children to simultaneously store and process verbal information. Situations in which the child *only* has to remember verbal information for immediate recall are very rare in everyday life learning, while situations that require the maintenance of verbal information in memory while engaging in other types of information processing are much more common.

The association between the verbal storage component of WM and the receptive vocabularies of the children with SLI in our study is in line with previous findings showing a clear link between verbal storage and receptive vocabulary (or word learning). And such findings are generally taken as evidence for the assumption that the primary role of the verbal storage component of WM is to support the learning of the phonological structure of language (Gathercole & Baddeley, 1989, 1990; Horohov & Oetting, 2004; Montgomery, 2002). The present findings further suggest that the visuospatial components of WM may be involved in the language abilities of young children with SLI in some way, but a clear and meaningful association was not found. The only significant correlation in the present study was between the visuospatial storage component of WM and expressive vocabulary, but it was quite small. To our knowledge, associations between visuospatial storage and expressive vocabulary have not been reported before. The verbal components of WM are obviously more strongly related to the development of the language abilities of young children with SLI than the visuospatial components, which is in line with other findings showing the magnitude of the deficits in the visuospatial domain of WM in children with SLI to not be as large as the deficits in the verbal domain of WM (Vugs et al., 2013). Finally, it might not be surprisingly that nonverbal intelligence was associated with some of the language abilities of the SLI children in this study. Previous research has shown a strong connection between the CE components of WM and (fluid) intelligence (Engle et al., 1999). It is likely that the nonverbal intelligence test used in the current study also relied on CE skills to some extent.

To our knowledge, the present study is one of the first to identify the underlying structure of WM in children with SLI. As only preschool children were included, however, no conclusions can be drawn with regard to the stability of this structure during children's development or possible changes in the interrelationships between the components of WM with development. Given that research among children with typical language development between the ages of 6 and 15 years has shown a three-factor structure to characterize the cognitive processes underlying WM, whether or not this is also the case for children with SLI during later stages of development should be determined (Alloway et al., 2006; Gathercole et al., 2004).

The present study was also unique in the adoption of a multimodal approach of WM to examine the associations between WM and language in children with SLI. Different components of WM were examined in conjunction with each other, but also then in conjunction with the developing language abilities of the children. The individual components of WM were shown to play differing roles in the support of the language acquisition of young children with SLI (age 4-5 years). Just how the associations between the components of WM and language abilities of the children develop — and possibly shift — as the children grow older is obviously something to be determined in future research.

More specifically, in future research, a multimodal approach should be adopted to explore if and how the structure of WM changes during the development of children with SLI but also to provide more information on the complex interplay between WM and language. To do the latter, we might examine SLI groups that systematically vary on the limitations experienced with regard to the different components of WM. One possible limitation on the present study in this light is that no measures of the functioning of the so-called episodic buffer (i.e., an additional component of WM) were included in the study. As deficits in this component of WM have been demonstrated in a recent study of young children with SLI (Petrucelli et al., 2012), information on this component of WM should probably be included in future research as well.

To conclude, the present findings have some valuable implications for clinical practice. First, the significant interactions between the WM and language abilities of young children with SLI suggest that interventions focusing on both linguistic *and* WM problems might result in more optimal results than those using traditional interventions with attention to only linguistic problems. For those children showing limited response to the more common language interventions, in particular, the addition of interventions aimed at the strengthening of WM may be valuable. It can also be concluded that effort should be clearly made to minimize the demands on WM when teaching and training

children with SLI in order to reduce the adverse effects of any WM impairments that may be present and maximize attention to the alleviation of the linguistic impairments confronting these children. When demands are too high and thus exceed the limited WM capacity of young children and particularly children with SLI, WM may unnecessarily restrict the acquisition and development of new language skills.

Chapter 6

Developmental associations between working memory and language in children with SLI: a longitudinal study

Based on:

Vugs, B., Hendriks, M., Cuperus, J., Knoors, H., & Verhoeven, L. (in press). Developmental associations between working memory and language in children with specific language impairment (SLI): a longitudinal study. *Journal of Speech, Language and Hearing Research*.



ABSTRACT

Purpose: This longitudinal study examined differences in the development of WM between children with specific language impairment (SLI) and typically developing (TD) children. Further it explored to what extent language at age 7- to 8-years could be predicted by measures of language and/or WM at age 4- to 5-years.

Method: Thirty children with SLI and 33 TD children that were previously examined on measures of WM and language at age 4- to 5-years (T1) were re-examined at age 7- to 8-years (T2).

Results: The developmental course of WM was mostly similar for the two groups; only the development of the verbal storage component differed. At T1 children with SLI performed significantly below their TD peers on all components of WM (verbal storage, verbal central executive (CE), visuospatial storage and visuospatial CE), while at T2 the differences for the visuospatial components were no longer significant when age and intelligence were taken into account. Hierarchical regression showed language and verbal CE at T1 to be significant predictors of language at T2, with no differences in the developmental associations between language and WM for the two groups.

Conclusions: The results of this study suggest that particularly verbal CE is of importance for the acquisition of linguistic skills.

INTRODUCTION

Over the past years, an increasing number of studies showed that children diagnosed with specific language impairment (SLI) not only demonstrate impairments in language, but also in other cognitive domains. One factor that is often implicated is working memory (WM) (Montgomery, Magimairy, & Finney, 2010). Children with SLI at different ages demonstrate deficits in WM when compared to typically developing (TD) peers (Archibald, 2016; Henry & Botting, 2016; Montgomery et al., 2010). However, there are still many questions about the developmental associations between WM and language. To date, research on this topic is very limited. In the present study, we therefore addressed the developmental course of WM and its relation to language in children with SLI. Children with SLI and their TD peers, ages 4- to 5-years, that were examined on several measures of WM and language in a previous study, were re-examined in this follow-up study at age 7- to 8-years (Vugs, Knoors, Cuperus, Hendriks, & Verhoeven, 2014).

Working memory in children with SLI

Although the language of the majority of children develops apparently automatically, there are also children who show marked problems or delays. When these children encounter language problems that cannot be explained by other underlying impairments, such as hearing loss, intellectual problems or marked neurological problems, a diagnosis of specific language impairment (SLI) is usually made (Bishop, 2002, 2006). Children with SLI form a heterogeneous group with different profiles of language deficits. SLI can affect various linguistic domains (i.e., phonological, morphological, lexical and grammatical domains) and the language profile often changes with age and development (Bishop, 2006; Leonard, 1998).

WM refers to the structures and processes used to temporarily store and manipulate information. Although WM can be conceptualized somewhat differently (Courage & Cowan, 2009; Engle, Tuholski, Laughin, & Conway, 1999), the most frequently adopted account in research on children with SLI is the multicomponent WM model of Baddeley (Baddeley & Hitch, 1974; Baddeley, 2003). In this model, a central executive (CE) system is assumed to link three subsystems: the phonological loop, the visuospatial sketchpad, and the episodic buffer. The CE system coordinates and controls activities in WM. It has a limited attentional capacity and requires attentional control. The phonological loop and visuospatial sketchpad are so-called "slave" systems and responsible for the temporary storage of verbal and visuospatial information, respectively. The episodic buffer is a relatively recent addition to the model and responsible for the binding of information from multiple sources together into chunks (Baddeley, 2003).

The development of WM already starts during the first years of life and eventually peaks in young adulthood. Research in TD children has shown that the ability to keep

simple information in mind is already present before the age of 6 months (Courage & Cowan, 2009; Garon, Bryson, & Smith, 2008). The development of WM is often associated with structural changes in the prefrontal cortex, which undergoes enormous neuro-developmental changes between 3 and 6 years of age (Garon, et al., 2008; Luciana & Nelson, 1998). A previous study on the underlying structure of WM in young children has shown a four factor model with separate but interacting components of verbal storage, verbal CE, visuospatial storage and visuospatial CE to provide the best account of WM in children with and without SLI aged 4- to 5-years (Vugs et al., 2015).

In children with SLI, significant group differences have been reported compared to TD children on different components of WM. It is widely accepted that SLI is associated with problems in the verbal storage component of WM (i.e., the phonological loop). The socalled phonological storage deficit hypothesis assumes that a specific deficit in the temporary storage of novel phonological information underlies SLI (Archibald & Gathercole, 2007; Baddeley, 2003; Bishop, 1996; Coady & Evans, 2008; Gathercole & Baddeley, 1990). Impairments in verbal storage have been reported in children with SLI at different stages of development, varying from preschool until adolescence (Montgomery et al., 2010; Vugs, Hendriks, Cuperus, & Verhoeven, 2014). In addition to these deficits in verbal storage, substantial impairments have been reported for the verbal CE component of WM. Children with SLI are even more severely and consistently impaired on verbal complex memory span tasks that combine verbal storage with processing of information, than on straightforward verbal storage tasks (Archibald & Gathercole, 2006a; Briscoe & Rankin, 2007). Impairments in the verbal CE component of WM have been found in children with SLI at different ages (Archibald & Gathercole, 2006b; Briscoe & Rankin, 2007; Ellis Weismer, Evans, & Hesketh, 1999; Henry, Messer, & Nash, 2011; Marton & Schwartz, 2003; Vugs et al., 2014).

Regarding the role of visuospatial WM in children with SLI results are somewhat contradictory. Based on studies that showed children with SLI to perform similarly to their TD peers on visuospatial storage and CE tasks, several authors assume that the WM deficits of children with SLI are limited to verbal WM (Alloway & Archibald, 2008; Archibald & Gathercole, 2006a, 2006b). In contrast, the results of several other studies and a recent meta-analysis have yielded evidence suggesting that the WM deficit of children with SLI may extend to the visuospatial domain (Marton, 2008; Vugs, Cuperus, Hendriks, & Verhoeven, 2013). Although the results of the meta-analysis showed some impairments in visuospatial storage and visuospatial CE in children with SLI, it also revealed that the deficit for visuospatial WM is not as large as the deficit for verbal WM. The extent of the deficit in verbal WM was found to be two the three times larger than the extent of the deficit in visuospatial WM.

Associations between working memory and language

Although many studies compared WM between children with SLI and TD children, much is still unknown about the exact associations between language and WM. Language is a complex system with different levels of processing and it is well known that various of these language abilities can be affected in children with SLI. Recently, some studies more specifically addressed the possible associations between WM and language abilities in children with SLI.

The verbal storage component of WM has often been proposed to be linked to word learning or vocabulary acquisition (Montgomery et al., 2010). Baddeley and Gathercole (1989) were the first to demonstrate a strong association between the functioning of the phonological loop and the acquisition of new words in TD children between 4 and 5 years. In this longitudinal study, verbal storage at the age of 4 years was significantly linked with vocabulary knowledge one year later. Since then, other studies have documented a similar association for children with SLI (Gathercole & Baddeley, 1990; Horohov & Oetting, 2004; Montgomery, 2002). It is assumed that the storage of phonological information in WM and word learning are especially linked in the early stages of vocabulary acquisition (Archibald, 2016).

Children with SLI also tend to have problems in the understanding and production of complex syntactic sentences (van der Lely, 1996; Fortunato-Travares et al., 2015). Accumulating evidence indicates that impairments in the verbal CE component of WM may account for these deficits in sentence processing (Archibald 2016; Montgomery & Evans, 2009; Noonan, Redmond, & Archibald, 2014). In one study of sentence comprehension, for example, performance on a verbal CE task correlated significantly with the comprehension of complex sentences in school-aged children with SLI (Montgomery & Evans, 2009). Fortunato-Travares et al. (2015) investigated the association between WM and sentence comprehension through direct manipulation of WM demands, showing an effect of WM on the syntactic assignment of predicates and reflexives in sentence comprehension in children with SLI. Recently, some authors suggested that the role of verbal CE in sentence processing is influenced by the task requirements. Noonan et al. (2014) investigated the interrelations between WM deficits — measured using both verbal and visuospatial complex memory span tasks — and judgments of grammaticality. In this study, children with only language impairments and thus no WM deficits, children with deficits in both domains, and TD children completed a task in which grammatical markers occurred at different places in the sentence. Children with only language impairments performed significantly worse regardless of the location of the marker, while children with deficits in both WM and language were only impaired for sentences with late grammatical errors, which are supposed to impose a greater WM load. Frizelle and Fletcher (2015) found that the ability to process complex sentences involving greater syntactic development was related to the verbal CE component of WM in children with

SLI but not TD children. Based on these findings, it is suggested that verbal WM skills are closely linked to sentence processing when language demands are high, which is often the case for children with SLI (Archibald, 2016).

Recently, we examined how the different components of WM were related to linguistic skills in children with SLI, aged 4- to 5-years (Vugs et al., 2015). We found the verbal CE component to be moderately to strongly associated with several language abilities, including receptive vocabulary, expressive vocabulary, verbal comprehension and syntactic development. In addition, the verbal storage component significantly correlated with receptive vocabulary and syntactic development. A clear and meaningful association between the visuospatial components of WM and any of the language abilities was not found.

Present study

From the research conducted so far, there is clear evidence that WM and language are associated in children with SLI. However, most previous studies were cross-sectional and did not take into account developmental aspects. As it is well established that both WM and language significantly change cross development, longitudinal research is needed to provide more information about the complex interplay between WM and language in children with SLI. Longitudinal studies taking into account language abilities and WM will shed some light on how these skills develop and how the relationship between them may change. Do children with SLI for instance just show a continuing delay or do the developmental trajectories differ for children with SLI compared to TD children? Furthermore, longitudinal research can be useful in addressing the issue of the directional relationship between WM and language (Kapa & Plante, 2015).

In the present longitudinal study, we investigated the developmental associations between WM and language in children with SLI. Since both WM and language abilities undergo significant changes in early childhood, we specifically focused on this stage of development. Children with SLI and TD children that were examined on several measures of WM and language at age 4- to 5-years (T1) were re-examined at age 7- to 8-years (T2). The specific questions we addressed were as follows:

- 1) Does the development of the different components of WM from T1 to T2 differ for children with SLI versus TD children?
- 2) To what extent can language at T2 be predicted from the language and/or WM measures at T1 in children with SLI and TD children?

In order to answer the first research question we compared the performances of the children with SLI on the different components of WM to that of the TD children at T1 and T2, and examined differences in the development on the WM measures between the two groups. We expected the children with SLI to show a developmentally consistent pattern of WM impairments in this age range (Henry & Botting, 2016). With regard to
the second research question we expected the verbal components of WM and language abilities at T1 to predict language abilities of the SLI and TD group at T2 (Archibald, 2016; Vugs et al., 2015).

METHODS

Participants

Children were recruited from a previous study population of children with SLI and agematched TD peers aged 4- to 5-years. In this study, all of the children had average intelligence (85 or more on the Snijder-Oomen nonverbal intelligence test, SON-R 21/2-7) and were native speakers of Dutch (Tellegen & Laros, 1998). Any children with a diagnosed hearing impairment, neurological disorder, ADD/ADHD, or autism spectrum disorder were excluded. The children in the SLI group were recruited from special language units or from special education schools in the Netherlands. All of them were receiving daily support for their speech or language problems. Diagnosis was based on extensive clinical and psychometric assessment by speech and language pathologists; persistent difficulties specific to language were shown in all cases. For most of the children, recent results for measures of language and nonverbal intelligence were available via their personal files. These results were included in the study only when they were no more than six months old. Otherwise, assessment was repeated. Participants were included in the study when they performed 1.25 SDs or more below the mean on at least two language measures, following Tomblin (1996). The language measures included the Peabody Picture Vocabulary Test-III-NL (Dunn & Dunn, 1997; Schlichting, 2005), the Reynell Developmental Language Scales (Reynell & Gruber, 1990; Eldik van, Schlichting, Lutje-Spelberg, Meulen van der, & Meulen van der, 2004), and tests of word and sentence development from the Schlichting Test for Language Production (Schlichting, Eldik van, Lutje-Spelberg, Meulen van der, & Meulen van der, 2003). The Dutch versions of these tests have all been normed. The children in the control group were recruited from three middle-class schools in the Netherlands. The language measures examined for the control group were the Peabody Picture Vocabulary Test-III-NL (Dunn & Dunn, 1997; Schlichting, 2005) and the Reynell Developmental Language Scales (Reynell & Gruber, 1990; Eldik van et al., 2004). All of the control children performed in the normal range on both of these tests. For a detailed description of the study population see Vugs et al. (2014).

Three years after the initial study, all parents were contacted again and invited to participate in this follow-up study. A total of 67 children were available for follow-up; 33 children with SLI and 34 TD peers. First, we analyzed the profiles of language abilities and nonverbal intelligence of both groups at T2. At this time-point, nonverbal intelligence was measured by the subtests Categories and Patterns of the SON 6-40 (Tellegen

Table 1

& Laros, 2011). The scores on these two subtests were combined to form an estimated nonverbal IQ. For the SLI group, the language measures included tests of receptive vocabulary (Peabody Picture Vocabulary Test-III-NL; Dunn & Dunn, 1997; Schlichting, 2005), expressive vocabulary (Expressive Vocabulary subtest CELF-4), verbal comprehension (Concepts and Following Directions subtest CELF-4) and syntactic development (Formulated Sentences subtest CELF-4) (Semel et al., 2003). The TD group was again examined with tests of receptive vocabulary and verbal comprehension. Three children in the SLI group and one child in the control group did no longer meet the mentioned inclusion and exclusion criteria. Data are reported for the remaining 63 children (30 children with SLI and 33 TD peers). The descriptive statistics for the SLI and TD group at both time-points are presented in Table 1.

7- to 8- years (12).				
		T1		T2
Measure	SLI (N = 30)	TD (N = 33)	SLI (N = 30)	TD (N = 33)
Age	57.07 (7.19)	59.06 (7.00)	95.90 (5.96)	100.24 (7.29)
Gender	M: 21, F: 9	M: 20, F: 13	M: 21, F: 9	M: 20, F: 13
Non-verbal IQ				
SON-R 21/2-7	107.50 (13.14)	116.61 (14.56)	-	-
SON-R 6-40	-	-	92.80 (15.57)	103.18 (14.35)
Receptive vocabulary				
PPVT-III-NL	94.43 (14.81)	107.24 (9.73)	90.23 (13.17)	102.61 (10.15)
Verbal comprehension				
Reynell	85.83 (14.36)	117.73 (10.87)	-	-
CELF-4, Concepts and Following Directions	-	-	6.73 (2.29)	9.61 (3.08)
Expressive vocabulary				
Schlichting WQ	81.83 (12.81)	-	-	-
CELF-4, Expressive	-	-	7.10 (2.58)	-
Vocabulary				
Syntactic development				
Schlichting ZQ	78.60 (9.48)	-	-	-
CELF-4, Formulated	-	-	6.40 (2.53)	-
Sentences				

Descriptive statistics for age, gender, nonverbal intelligence and language measures at 4- to 5- years (T1) and 7- to 8- years (T2).

Note. T1 = 4- to 5-years; T2 = 7- to 8-years; SLI = specific language impairment; TD = typically developing; SON-R 2¹/₂-7 = Snijders-Oomen nonverbal intelligence test SON-R 2¹/₂-7; SON-R 6-40 = Snijders-Oomen Nietverbale Intelligentietest SON-R 6-40; PPVT-III-NL = Peabody Picture Vocabulary Test, Third Edition; Reynell = Reynell Developmental Language Scales; CELF-4 = Clinical evaluation of language Fundamentals, Fourth Edition; Schlichting WQ = Schlichting Test for Language Production, Word development; Schlichting ZQ = Schlichting Test for Language Production, Sentence development At T2, the mean age of the children SLI was 8;00 (SD = 5.96 months, range 7;02 to 8;09). The mean age of the TD peers was 8;04 (SD = 7.29 months, range 7;03 to 8;11). Oneway analyses of variance (ANOVAs) confirmed that at this time-point the SLI group had significantly lower scores on the language measures than the control group (PPVT-III-NL F(1,61) = 17.62, p < .001; CELF-4 Concepts and Following Directions F(1, 61) = 17.36, p < .001). Additionally, the SLI and control groups significantly differed with regard to age (ANOVA F(1,61) = 6.61, p = .013) and nonverbal intelligence (ANOVA F(1,61) = 7.58, p = .008). The two groups did not differ significantly with regard to gender (Chi-square Test X^2 (1, N = 63) = .610, p = .597).

Procedure

Almost all children were tested individually in a quiet room at their school. Only three children were tested at home. Written consent was obtained for participation in the present follow-up study from the parents of the children. Assessment took about two hours, with a short break halfway through. In addition to the measures for nonverbal intelligence and language listed above, all of the children were administered four subtests of the Dutch translation of the Automated Working Memory Assessment (AWMA) (Alloway, 2007). All children completed the test battery.

Measures of Working Memory

The AWMA is an automated, computerized assessment battery suitable for use with respondents who are 4 to 22 years of age (Alloway, 2007). The AWMA has been validated and measures the different components of Baddeley's WM model, including verbal storage, verbal central executive (CE), visuospatial storage, and visuospatial CE (Gathercole & Pickering, 2000). The storage measures tap into the phonological loop or visuospatial sketchpad, depending on the nature of the information to be remembered. For the CE measures, the children must simultaneously store and process information. The process-ing activity is assumed to tap into the central executive component of the WM model. In this study we included four subtests; one for each component of WM.

Testing follows the same span procedure in all subtests. Following a practice session, a maximum of six sequences of increasing lengths are presented. The length of the sequences is increased by one after the child has correctly recalled four sequences of a particular length with a maximum of seven items for the CE tasks and nine items for the storage tasks. Testing is stopped when three sequences of a particular length are not recalled correctly. The children respond by pointing to the answer of their choice on the screen or by saying it aloud. The experimenter then imports their choice into the computer program.

Verbal storage

In the Digit recall task, the child must recall a sequence of digits in the right order. The digits can range from one to nine and are spoken at a rate of one digit per second. The sequences are randomly generated and no digits are repeated.

Verbal CE

In the Listening recall task, the child is presented short sentences. The child must then judge whether the content of the sentence is correct (by saying "true" or "false") and remember the last word of the sentence. The number of sentences increases in length and the child must then recall the last words of the sentences in the correct serial order. The sentences have a simple subject-verb-object order and contain early developing vocabulary.

Visuospatial storage

In the Dot matrix task, a sequence of red dots is presented on a 4 x 5 grid. All of the dots appear in the grid for 2 seconds. The dots then disappear and the child must point to the position of each dot in the same serial order as presented.

Visuospatial CE

In the Spatial span task, two identical shapes are presented to the child with a red dot above the right shape. The child must judge whether the two shapes are in normal or mirror image and to remember the location of the dot. The position of the dot rotates to one of three positions of a triangle. After trials requiring the child to judge the similarity of the shapes, they disappear and a triangle of three dots reflecting the possible positions of the previous dots appears. The child must point to the positions of the previous dots in the right order.

Statistical analyses

First, differences between the SLI and TD group at T1 and T2 were tested using multivariate analyses of variance (MANOVAs) and follow-up analyses of variance (ANOVAs). Using the Bonferroni method, which divides the level of significance by the number of dependent variables, each ANOVA was tested at the .013 level. In addition, effect sizes were computed. The effect-size (*d*) is the difference between the mean of the control group and the SLI group divided by the pooled sample standard deviation. Effect sizes are considered small for d = .20, medium for d = .50, and large for d = .80 (Cohen, 1988). To control that age and intelligence were not mediating performance on the WM measures, multivariate analyses of covariance (MANCOVAs) and follow-up analyses of covariance (ANCOVAs) were next conducted for the four WM measures; nonverbal intelligence (IQ SON) and age were entered as covariates. Then, 2 x 2 repeated measures ANOVAs were conducted with time of assessment as within factor (T1, T2) and group as between factor (SLI, TD). Effect sizes (Cohen's η^2) were reported for all analyses. Following the criteria of Cohen (1988) effect sizes were considered small for $\eta^2 = .01$, medium for $\eta^2 = .06$, and large for $\eta^2 = .14$. To control that age and intelligence were not mediating possible time or interaction effects, repeated measures analyses of covariance (ANCOVAs) were next conducted for the four WM measures; nonverbal intelligence (IQ SON) and age were entered as covariates.

To examine the interrelations between the measures of WM and language at T1 and language at T2, first principal component analyses with varimax rotation was conducted on the language tasks of the total group of children. Further, Pearson correlations were calculated and a hierarchical regression analysis was conducted with language at T2 as the criterion variable. The predictor variables and a Group variable (SLI = 1, TD = 0) were included in Step 1 and in Step 2 all corresponding interaction terms were included. To control for multicollinearity, we centered all predictor variables before conducting the regression analysis (Jewel, 2003).

RESULTS

Development of Working Memory

The descriptive statistics for the WM measures (AWMA) are shown in Table 2. First, performances of the SLI and TD group were compared. MANOVA investigating group differences on the four WM measures at T1 (i.e., verbal storage, verbal CE, visuospatial storage, and visuospatial CE) showed a significant overall group effect: Wilks' $\Lambda = .33$, F(4,58) = 29.93, p < .001, $\eta^2 = .67$. Follow-up ANOVAs showed significant group effects for all four WM measures at a .013 level. The effect sizes ranged from 1.03 to 2.30. On all WM measures the SLI group scored at a significantly lower level than the TD group.

At T2 there also was a significant overall group effect on the WM measures, Wilks' Λ = .56, *F*(4,58) = 11.60, *p* < .001, η^2 = .44. Follow-up ANOVAs again revealed significant group differences for each of the individual WM measures, with effect sizes ranging from .67 to 1.63.

To control that age and intelligence were not mediating performance on the WM measures, MANCOVAs and follow-up ANCOVAs were conducted. At T1, the overall group effects on the WM measures remained significant: Wilks' $\Lambda = .58$, F(4,56) = 10.41, p < .001, $\eta^2 = .42$). And once again, the univariate ANCOVAs showed significant group effects for the four WM measures: Verbal storage F(1,59) = 60.92, p < .001; Verbal CE F(1,59) = 54.95, p < .001; Visuospatial storage F(1,59) = 10.87, p = .002, Visuospatial CE F(1,59) = 7.59, p = .008).

			SLI					Ð				Time			ime X G	roup
Variable	1	T2	t	d	þ	1	T2	t	р	q	_	d	η²	Ŀ	d	ŋ²
Verbal storage	71.40 (12.99)	81.50 (11.88)	3.045	.005	2.30	98.39 (10.86)	96.18 (13.56)	-1.367	.181	1.17	4.84	.032	.073	11.78	.001	.162
Verbal CE	99.60 (11.04)	98.43 (12.34)	678	.678	2.14	123.36 (11.51)	116.61 (10.42)	-2.489	.018	1.63	4.15	.046	.064	2.066	.156	.033
Visuospatial Storage	91.93 (11.97)	100.73 (11.19)	3.017	.005	1.15	106.42 (13.52)	109.42 (14.67)	1.086	.286	.68	8.63	.005	.124	2.085	.154	.033
Visuospatial CE	98.53 (12.30)	98.37 (9.70)	063	.950	1.03	113.03 (16.73)	107.15 (15.92)	-2.056	.048	.67	2.38	.128	.038	2.129	.150	.034

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At T2 the overall group effect again was significant for the WM measures (Wilks' Λ = .58, F(4,56) = 10.41, p < .001, $n^2 = .42$). Follow-up ANCOVAs showed significant group effects for verbal storage and verbal CE, but not for visuospatial storage and visuospatial CE: Verbal storage F(1,59) = 12.99, p = .001; Verbal CE F(1,59) = 36.99, p < .001; Visuospatial storage F(1,59) = 3.26, p = .076, Visuospatial CE F(1,59) = 1.71, p = .196. Nonverbal intelligence was significantly related to visuospatial CE, F(1,59) = 15.09, p < .001.

To investigate differences between the SLI and TD group on the WM measures over time, repeated measures ANOVAs were conducted (see Table 2). The results showed a significant effect of time for visuospatial storage, with a medium effect size. A significant Time x Group interaction effect was found for verbal storage, with a large effect size. Children in the SLI group showed an improvement in verbal storage over time, while the performances of the TD group remained stable. The other WM measures did not show significant effects.

When we controlled for possible mediating effects of age and nonverbal intelligence, repeated measures ANCOVAs showed similar results as described above (data available from first author).

Interactions between working memory and language abilities

To start, principal component analyses with varimax rotation on the language tasks revealed one language factor at T1 as well as T2. At T1, this factor explained 84.08 percent of the variance in language tasks and at T2 82.99 percent.

Pearson correlation coefficients were computed to explore how WM and language abilities are related over time for the total group of children. We correlated the predictor measures (i.e., verbal storage, verbal CE, visuospatial storage, visuospatial CE and language at T1) and the criterion measure (i.e., language at T2). The resulting correlation matrix is shown in Table 3. Nearly all predictor measures at T1 were significantly

Table 3.

Correlations between components of working memory and language at 4- to 5- years (T1) and language at 7- to
8- years (T2) (n = 63).

Variable	1	2	3	4	5	6	
1. Language T1	-						
2. Verbal storage T1	.601*	-					
3. Verbal CE T1	.669*	.640*	-				
4. Visuospatial storage T1	.542*	.541*	.590*	-			
5. Visuospatial CE T1	.386*	.448*	.506*	.552*	-		
6. Language T2	.671*	.501*	.622*	.445*	.211	-	

Note. T1 = 4- to 5-years; CE = central executive; T2 = 7- to 8-years * *p* < .001

correlated to language at T2. Only the visuospatial CE component of WM at T1 did not significantly correlate with language at T2.

To further explore the developmental associations between WM and language, we next conducted a hierarchical regression analysis with language at T2 as the criterion variable. The predictor variables and a Group variable were included in Step 1. In Step 2, all corresponding interaction terms were included to explore differences between the SLI and TD group. Table 4 summarizes the information of the regression analysis.

The results showed language and the verbal CE component of WM at T1 to be significant predictors of language at age T2: $\Delta R^2 = .536$, F(6,56) = 10.773, p < .001. Step 2 of the regression analysis did not show significant interactions: $\Delta R^2 = .067$, F(5,51) = 1.731, p = .144. These results indicate that the developmental associations between language and WM at T1 and language at T2 do not differ for the SLI versus TD children.

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Results of Hierarchical Regression Analysis on language at age 7- to 8-years.

Variable	Total R ²	ΔR^2	β
Step 1			
	.536***	.536***	
Language T1			.459**
Verbal storage T1			.112
Verbal CE T1			.372*
Visuospatial storage T1			.078
Visuospatial CE T1			203
Group			.100
Step 2			
	.603	.067	
Language T1			.481**
Verbal storage T1			.158
Verbal CE T1			.362*
Visuospatial storage T1			.092
Visuospatial CE T1			228
Group			.119
Group x Language T1			038
Group x Verbal storage T1			203
Group x Verbal CE T1			026
Group x Visuospatial storage T1			.251
Group v Visuospatial CE T1			094

Note. T1 = 4- to 5-years; CE = central executive

* *p* <.05, ** *p* < .01, *** *p* < .01

DISCUSSION

The aim of this longitudinal study was to examine the developmental associations between WM and language from the age of 4- and 5-years to 7- and 8-years in children with SLI and TD children. We first evaluated differences in the developmental course of WM between children with SLI and their TD peers. Furthermore, we examined whether language at T2 could be predicted from the language and/or WM measures at T1.

With regard to our first research question, namely whether the development of the different components of WM from T1 to T2 differed for children with SLI versus TD children, we found the developmental course of WM to be mostly similar. No significant differences were found between the two groups with regard to the development of the verbal CE, visuospatial storage and visuospatial CE components of WM. Only the development of the verbal storage component significantly differed for the children with SLI versus TD children. The children with SLI showed an improvement in verbal storage, while performances of the TD children remained stable. Group comparison, however, revealed that the children with SLI still performed significantly worse on verbal storage compared to their TD peers at T2.

The children with SLI were found to perform significantly below their TD peers on all components of WM (i.e., verbal storage, verbal CE, visuospatial storage and visuospatial CE) at both T1 and T2. At T1, the effect sizes for the different components all were large (range of d = 1.03 to d = 2.30). At T2, effect sizes were large for the verbal components of WM (d = 1.17 and d = 1.63) and medium for the visuospatial components (d = .68 and d = .67). These results are in line with previous findings showing clear impairments in verbal storage and verbal CE in children with SLI at different ages (Archibald & Gathercole, 2006b, 2007; Coady & Evans, 2008; Gray, 2003; Montgomery et al., 2010). Reduced performance on visuospatial storage and visuospatial CE tasks in young children with SLI has also been reported in most previous studies (Bavin, Wilson, Maruff, & Sleeman, 2005; Hick, Botting, & Conti-Ramsden, 2005; Menezes, Takiuchi, & Befi-Lopes, 2007; Marton 2008). At T2 we also found the children with SLI to perform below their TD peers on the visuospatial components of WM, but the magnitude of these deficits is not as large as the deficits in the verbal components of WM. This replicates findings of a recent meta-analysis of visuospatial WM, showing that the deficit in verbal WM of children with SLI is two to three times larger than the deficit in visuospatial WM (Vugs et al., 2013). Furthermore, the differences between the children with SLI and TD children in visuospatial storage and visuospstial CE at T2 were no longer significant when we took into account the mediating effects of age and nonverbal intelligence. Especially nonverbal intelligence was significantly related to visuospatial CE. These results suggest that in children with SLI in the age of 7- to 8-years reduced performance on visuospatial storage and visuospatial CE might not be a specific problem in visuospatial WM, but rather an indirect effect of nonverbal intelligence. The relationships between WM, language and intelligence in children with SLI is something that needs further investigation. Research in TD children has shown that executive functions, including WM, are correlated with crystalized and fluid intelligence (Arffa, 2007; Engle, 1999). A recent meta-analysis on non-verbal intelligence furthermore showed children with SLI to perform on average 0.69 standard deviations lower than their TD peers (Gallinat & Spaulding, 2014). It has to be determined in future research whether intelligence influences the associations between WM and language abilities in children with SLI.

Our second research question addressed to what extent language at T2 could be predicted from the language and/or WM measures at T1 in children with SLI and TD children. To answer this question correlations were first calculated. Language, verbal storage, verbal CE and visuospatial storage at T1 all were found to be significantly correlated to language at T2. Only the visuospatial CE component of WM did not show a significant correlation with language at T2. Hierarchical regression further showed language and the verbal CE component of WM at T1 to be significant predictors of language at T2. The verbal storage and visuospatial components of WM did not explain a significant amount of variance for language at T2. Moreover, it was found that these developmental associations between language and WM were similar for the children with SLI and TD children.

This study was one of the first to take into account the developmental associations between WM and language abilities in children with SLI in a longitudinal study. Taken together, the current results indicate a developmentally consistent pattern of WM impairment in children with SLI in early childhood. However, the verbal components of WM at age 4- to 5-years are more strongly related to language abilities three years later than the visuospatial components in both children with SLI and TD children. Particularly, the verbal CE component of WM was found to be a significant predictor of language. These findings are in line with previous research, showing clear links between verbal CE and various language abilities in children with SLI (Archibald 2016; Montgomery & Evans, 2009; Noonan et al., 2014; Vugs et al., 2015). The current results suggest that verbal CE is important for the acquisition of linguistic skills. It seems plausible that the ability to simultaneously store and process verbal information (i.e., verbal CE) is involved in almost all everyday situations of learning new language abilities and that problems in verbal CE affect the processes of learning various linguistic skills. However, the present findings do not rule out that language abilities also affect WM. It is likely that WM and language develop in reciprocal interaction with changing effects on each other over time. More systematic research is needed to disentangle this complex interplay between WM and language in children with SLI. To gain more information about the directionality of the relationship between WM and language, it could be of interest to investigate whether linguistic training affects WM performance. Or, the other way around, whether WM training influences language abilities. Likewise, studies that identify groups of children whose WM and language skills dissociate may prove useful to draw more firm conclusions about the directional relationship between WM and language in children with SLI (Noonan et al., 2014).

Of course several limitations apply to the present study. One possible limitation for instance is that no measures of other executive functions (EFs) like inhibition and cognitive flexibility were included. Most recent models typically considered EFs a multifaceted concept with distinct subfunctions, and WM is one of the most frequently postulated components of EFs (Huizinga et al., 2006; Miyake et al., 2000). Recent studies showed impairments in other EFs like for instance inhibition in children with SLI (Pauls & Archibald, 2016; Henry et al., 2011). How WM is related to other EFs in children with SLI is something to be determined in future research. Further, no measures of the functioning of the episodic buffer component of WM were included. The inclusion of such information might be of value for future research as impairments in this component of WM in children with SLI have recently been reported (Petrucelli, Bavin, & Bretherton, 2012). Another concern is the relative small sample size of the study, especially in relation to the hierarchical regression analyses that was conducted. Given the number of predictor variables and interaction terms, it is possible that some of the interactions would have reached statistical significance in a larger sample. A last limitation is that the limited age range of the children included in this study. Based on this, no conclusions can be drawn with regard to the stability of the associations between WM and language abilities for children older than 8 years of age. Just how the associations between the components of WM and language abilities of the children develop — and possibly shift — as the children grow older is obviously something that has to be examined in future research. Continued research will provide greater insight in the role of WM in the language acquisition of children with SLL

In closing, the present findings have some valuable implications for clinical practice. First of all, it seems important to include WM tasks in the assessment of children with SLI. Attention should not only be paid to the language problems of these children, but also to possible WM impairments that can contribute to their language problems. It is specifically recommended to adopt a multimodal approach of WM given the current findings of impairments in both verbal and visuospatial WM. Although children with SLI show more substantial impairments in verbal WM, it is obviously important to know if the WM problems being experienced by a child are also visuospatial. For instance for the use of visual support, which is a common intervention strategy adopted for children with SLI. Likewise, it seems sensible to pay attention to WM in treatment. Given the developmentally consistent WM impairments of children with SLI, it is important that WM demands be minimized during teaching and treatment in order to limit the adverse effects of the WM deficits. Further, interventions directed at improving WM and teaching children effective strategies to cope with their WM limitations (e.g., rehearsal, grouping,

visualization) may be valuable additions to more traditional linguistic interventions. Interventions focusing on both language and WM problems might result in more optimal results than those with attention to only linguistic abilities.

Chapter 7

Executive Function Training in Children with SLI: A Pilot Study

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ABSTRACT

The aim of this study was to evaluate the effectiveness of a computer-based executive function (EF) training in children with SLI. A total number of 10 children with SLI, ages 8 to 12 years, completed a 25-session training of visuospatial working memory, inhibition and cognitive flexibility over a 6-week period. Treatment outcome was examined directly after training and at 6 months follow-up by tasks of the three trained EF, tasks of other neurocognitive functions (attention, planning and fluency), and ratings of EF and behavioral problems by parents and teachers. Directly after training, results showed significant improvement on cognitive flexibility and a positive trend for visuospatial storage and inhibition. At 6 months follow-up, children with SLI performed significant improvement on sustained EF. Furthermore, the results showed significant improvement on sustained attention, attention control, parent- and teacher-rated attention behavior and parent-rated EF and externalizing behavior. Effect sizes were medium for all these outcome measures. The result of this pilot study highlight the importance of a large-scale, randomized controlled trial examining the possible effects of EF training in children with SLI.

INTRODUCTION

Growing evidence implicates that executive functions (EFs) are in some way involved in the problems associated with specific language impairment (SLI). Several studies reported limitations in EFs in children with SLI (Im-Bolter et al., 2006; Henry et al., 2011). This raises the question whether training of EF might be a meaningful intervention for these children. Especially since EF training has proven to be a promising intervention in other groups of children characterized by impairments in EF (Klingberg et al., 2002; 2005, Melby-Lervag & Hulme, 2012). To date, however, research describing interventions directed at improving EF in children with SLI is very limited. In the present study, we therefore explored the possible effect of EF training in children with SLI.

Executive functions in children with SLI

Executive function (EF) is a broad term that comprises cognitive processes responsible for purposeful, goal directed behavior and is typically considered a multifaceted concept with distinct subfunctions (Miyake & Shah, 1999; Miyake et al., 2000). These processes are inter-related and function together as an integrated, supervisory control system (Stuss & Alexander, 2000). The three most frequently postulated components of EF are: working memory, inhibition and cognitive flexibility (Huizinga et al., 2006; Miyake et al., 2000). Working memory (WM) refers to the structures and processes used to temporarily store and manipulate information (Baddeley & Hitch, 1974; Baddeley, 2003). Inhibition is the ability to stop prepotent or ongoing responses (Miyake et al., 2000). Cognitive flexibility, often also described as shifting, has been conceptualized as the ability to switch the focus of attention between activities (Miyake et al., 2000). However, still some debate remains about the exact components of EF. Various additional components of EF have been postulated, for instance planning, fluency, emotional control, initiation and monitoring (Gioia et al., 2000; Piatt et al., 1999). Another issue in the field of EF research is the question of ecological validity of the performance-based tasks used to measure EF. Some evidence exists that these standardized tests may not be sufficiently sensitive to the multidimensional nature of EF in daily life (Chaytor et al., 2006). Based on these findings, it is suggested that behavioral ratings of EF should be used to collect information in different contexts and from different sources (Gioia et al., 2001).

Children with specific language impairment (SLI) show a selective failure to make normal progress in language acquisition without further evidence of underlying intellectual, neurological, social, or emotional impairments (Bishop, 2002, 2006). They form a heterogeneous group with different profiles of language deficits; the impairment can affect different linguistic domains including phonological, morphological, lexical and grammatical domains. Moreover, the linguistic profile changes with age and development in many children (Bishop, 2006; Leonard, 1998). To explain the underlying causes of SLI, different theories and hypotheses exist. Some of these hypotheses presume that SLI originates from a deficit or delay specific to language, and particularly grammar (Rice and Wexler, 1996; Van der Lely, 2005). More recently, however, there is growing evidence that also non-linguistic factors are involved in SLI (Bishop, 2006; Montgomery et al., 2010). One factor that has been often implicated is EF (Im-Bolter et al., 2006; Henry et al., 2011). Several behavioral studies provided evidence of EF deficits in children with SLI (Archibald & Gathercole, 2006b; Im-Bolter et al., 2006; Lum et al., 2011; Marton et al., 2007). In similar vein, findings from neuroimaging studies concluded that children with SLI show anomalies in frontal brain areas normally related to EF (Dibbets et al., 2006; Jernigan et al., 1991).

In children with SLI, significant group differences have been reported compared to typically developing (TD) children on several components of EF, including WM, inhibition, cognitive flexibility, planning and fluency. These impairments were not confined to verbal EF tasks, but occurred for some nonverbal EF tasks as well. However, not all components of EF have been equally extensively studied and in some cases results are still somewhat contradictory. Strong links have especially been found between working memory limitations and SLI (Archibald & Gathercole, 2006a; Bishop, 2006; Montgomery et al., 2010). It is widely accepted that children with SLI show impairments in verbal WM. Significant group differences have been reported between children with SLI versus TD children on tasks of non-word repetition, recall of words or digits, and complex verbal span tasks (Archibald and Gathercole, 2006b; Gray, 2003, 2006; Conti-Ramsden, 2003). In contrast, visuospatial WM has been less extensively investigated with somewhat contradictory results. Several studies showed children with SLI to perform similarly to their TD peers on visuospatial WM tasks (Alloway and Archibald, 2008; Archibald and Gathercole, 2006a, 2006b; Lum et al., 2011; Williams et al., 2000). The results of some other studies and a recent meta-analysis however, have yielded evidence suggesting that the WM deficits of children with SLI may extend to the visuospatial domain (Vugs et al., 2013). In addition, significant group differences have been reported between children with SLI versus TD children on several tasks of inhibition, like go/no-go tasks and tasks requiring resistance of distractors (Bishop & Norbury, 2005; Finneran et al., 2009; Im-Bolter et al., 2006; Marton et al., 2007; Spaulding 2010). With regard to cognitive flexibility, studies in preschool children with SLI (age 4 and 5 years) showed poorer performance of the SLI group in comparison with their TD peers (Farrant et al., 2012; Coello et al., 2015). In contrast, group differences between children with SLI and their TD peers were not found in school-aged children on several shifting tasks, including the Trailmaking test and setshifting tasks (Dibbets et al., 2006; Henry et al., 2011; Im-Bolter et al., 2006). Planning has received considerably less attention in research on children with SLI. Problems with planning abilities in children with SLI have been reported on Towers tests and a Sorting test (Henry et al., 2011; Marton, 2008). With regard to *fluency*, children with SLI obtained significantly lower scores on both verbal and non-verbal fluency tests compared to their TD peers in a recent study (Henry et al., 2011). Deficits in non-verbal fluency have not been consequently found in all studies, however. For instance, the study of Bishop and Norbury (2005) showed the performance of children with SLI not to differ from that of TD children on two tasks of non-verbal fluency.

Regarding behavioral ratings of EF in daily life, Hughes and colleagues (2009) compared the parental and self-ratings of EF for adolescents with SLI versus TD adolescents. They found more negative ratings of EF behaviors for the SLI group compared to the TD group, with half of the parents of adolescents with SLI rating their child's EF abilities in the clinically impaired range. In recent studies, also the parents and teachers of preschool and school-aged children with SLI reported significantly more problems relative to TD children on everyday EF behaviors (Cuperus et al., 2014; Vugs et al., 2013; Wittke et al., 2013). These included problems with inhibition, WM, shifting, emotional control, initiation, and planning. In school-aged children with SLI, most problems have been reported on behavioral ratings of WM and initiation in classroom settings, with one third of the children scoring in the clinically impaired range (Cuperus et al., 2014).

Training of Executive functions

Increasing evidence suggests that it is possible to improve EF by cognitive training. The underlying assumption for these kind of interventions is that the maturation and/ or efficiency of the neural circuitries underlying the trained EFs can be improved by intensive practice and training. Several novel, computer-based training programs have demonstrated promise in children and adolescents. To date, most studies particularly focused on the training of WM. Convincing evidence has been found for the trainability of WM in children with ADHD (Beck et al., 2010; Green et al., 2012; Holmes et al., 2010; Klingberg et al., 2002, 2005). Klingberg and colleagues (2002, 2005) were among the first to show in a randomized controlled study in children with ADHD that a computer-based WM training improved the trained visuospatial WM of the children, Later on, significant improvement on at least one trained WM task has also been reported in several other studies and meta-analytic reviews (Holmes et al., 2010; Gray et al., 2012; Green et al., 2012; Melby-Lervag & Hulme, 2012; Rapport et al., 2013; Shipstead et al., 2012). Furthermore, some studies showed that in children with ADHD these positive effects tend to be stable after training (Holmes et al., 2010; Klingberg et al., 2005). In a recent meta-analytic review it was found that the training effects on visuospatial WM tasks are maintained at follow-up, which was on average of 5 months after the training (Melby-Lervag & Hulme, 2012). Based on this, it is generally accepted that WM training leads to positive effects on tasks closely related to the trained tasks, so called near-transfer effects.

However, quite some controversy exists about the generalizability or far-transfer of the training effects on functions not closely related to the trained tasks, like for instance other

neurocognitive functions, ADHD behavior and academic performance (Melby-Lervag & Hulme, 2012; Shipstead et al., 2012). Based on results of several reviews documenting limited or negligible far-transfer effects, increasing concerns are expressed about the generalization of the trained task effects in WM training (Chacko et al., 2013; Melby-Lervag & Hulme, 2012; Rapport et al., 2013; Redick et al., 2015; Shipstead et al., 2012). In contrast, a recent systematic review and meta-analysis showed persistent training effects for inattention in daily life for children with ADHD (Spencer & Klingberg, 2015).

Fewer studies examined the trainability of inhibition and cognitive flexibility. One study on the effect of cognitive training of inhibition in TD children, showed significant improvement on most of the trained tasks, but no generalization of the effect to tasks measuring WM or attention (Thorell et al., 2009). A study on the trainability of cognitive flexibility in a sample with children, adolescents and adults, showed significant improvement on cognitive flexibility tasks, but also on other EFs, including WM and interference control. In another study, adults with ADHD showed significant improvement on both trained and non-trained tasks after task-switching training (White & Shah, 2006).

Recently, van Oord et al. (2012) examined the effect of a computerized EF training in which three EFs were trained (i.e., WM, inhibition and cognitive flexibility) in children with ADHD. In this training, game elements were added in order to increase children's motivation and potentially optimize their cognitive performance during training. Add-ing game elements has proven to enhance the cognitive performance of children on EF tasks (Dovis et al., 2012; Prins et al., 2011). The results of this study showed significant improvement on parent-rated EF and ADHD behavior. Effects were maintained at 9 week follow-up. A positive effect of this EF training has also been found in a study in obese children, showing significant improvement on a WM task and childcare workers reports of WM and metacognition. In addition, children were more capable of maintaining their weight at 8 weeks after the training (Verbeken et al., 2013).

Cognitive training in children with SLI

To date, studies examining the possible effects of EF training in children with SLI are scarce. Whether cognitive nonlinguistic training in general could have a positive effect on language abilities in children with primary (or specific) language impairment was investigated in two exploratory single-subject design studies. Ebert and Kohnert (2009) trained processing speed and auditory memory in two school-aged children with SLI. The results indicated that the participants made gains in processing speed and some language abilities, including sentence formulation and grammatical morpheme production. These results were replicated and extended to bilingual children with SLI using a training of processing speed and sustained attention (Ebert, Rentmeester-Disher, & Kohnert, 2012). Both children made gains in nonlinguistic skills, as well as in Spanish and English. More recently, Ebert and colleagues (2014) compared the effects of the three

different treatment programs in fifty-nine bilingual school-aged children with SLI. The participants received either nonlinguistic cognitive, English, bilingual (Spanish-English) or deferred treatment. All three active treatment groups outperformed the deferred treatment control group and showed gains in the skills directly trained in the treatment. Children in the nonlinguistic cognitive treatment group significantly improved on processing speed and improvements on sustained attention approached statistical significance. Furthermore, cross-domain effect occurred: children in the nonlinguistic cognitive treatment in the nonlinguistic cognitive treatment some of the language tasks from pre-test to post-test.

Holmes and colleagues (2015) investigated whether WM training could be effective in enhancing verbal WM in children with low language abilities. A group of 12 children with low language abilities aged 8-11 years and 15 matched TD children completed Cogmed WM training (Cogmed, 2005). Both groups showed significant post-training gains on visuospatial storage (or visuospatial STM). Further exploratory analyses revealed some predictive links between pre-training scores and training outcomes. First, visuospatial WM improved to the greatest extend following training for children with higher verbal abilities. Furthermore, children with the lowest verbal IQs at baseline made the greatest gains in verbal STM after training.

Taken together, these results indicate that cognitive training could have positive effects on both nonlinguistic and language outcomes in children with SLI. Although cumulating evidence shows limitations in EF in children with SLI, the possible effect of cognitive training directed at improving EF has not yet been examined in these children.

Present study

The aim of the present pilot study was to explore the possible effects of EF training in children with SLI. We evaluated the effectiveness of a 6-weeks computerized EF training program suitable for children aged 8-12 years, in which visuospatial WM, inhibition and cognitive flexibility are trained. The performances of the children with SLI before and after training were compared on tasks measuring the three EFs trained in the training program, tasks of other neurocognitive functions and ratings of EF and behavioral problems by parents and teachers. Since it was the first study of the possible effects of EF training in children with SLI, the research questions were mainly exploratory. Our specific research questions were as follows.

- 1) Does EF training produce significant improvement on tasks of the trained EFs (visuospatial WM, inhibition, cognitive flexibility) in children with SLI?
- 2) Does EF training produce significant improvement on other neurocognitive functions in children with SLI?
- 3) Does EF training produce significant improvement in parents and/or teachers' ratings of EF and behavioral problems in children with SLI?

METHODS

Participants

A total number of 10 children with SLI aged 8- to 12-years (8 boys/2 girls) participated in this pilot study. The mean age of the children was 9;05 (SD = 15.61 months, range 8;05 to 12;05). All children were recruited from a Speech and Language Centre for children with severe language problems in the Netherlands. Diagnosis was based on extensive clinical and psychometric assessment by speech and language pathologists; all participants scored 1.25 SD or more below the mean on at least two language measures, following Tomblin (1996). Any children with a diagnosis of hearing disorder, frank neurological disorders, or autism spectrum disorder were excluded. All children had average nonverbal intelligence (mean = 101.78, SD = 12.96) and were native speakers of Dutch. Prior to attending the treatment center the children received daily support for their speech or language problems for at least one year without substantial development as result of their persistent problems. Children who showed clear impairments in EF based on their personal files or clinical evaluation (e.g., -1 SD on at least one task of WM, inhibition or cognitive flexibility) were sent information about the research project and were asked to participate. Those from whom we received informed consent participated in this pilot study. During the treatment period children did not receive any other treatment directed at improving EF or other neurocognitive functions.

Procedure

The study design included a pretest before the start of the training, a post-test at the conclusion on the training period and a follow-up.

One week before the beginning of the training the pre-test was conducted. The children were assessed in the Speech and Language Centre with tasks of the three trained EF (visuospatial WM, inhibition, cognitive flexibility) and tasks of other neurocognitive functions. Additionally, parents and teachers completed questionnaires about the behavior of the child. The tests and questionnaires used in the pre-test are described below.

The treatment program started with an introduction session with the child and his or her parents. They received practical information about the training and it was checked whether the training program worked well on the child's computer. Furthermore, it was ensured that the computer was placed at a location with limited distraction and that no contact was possible with the internet or other software. Session 1, 2, 10 and 20 were led by a research assistant in the Speech and Language Centre. The other sessions were done at home. All children kept a diary of their experiences with the game, in which time spent on the training, the number of sessions played in a week, and difficulties or problems were reported for each session. Time spent on the training sessions and frequency of the sessions were evaluated in the meetings with the research assistant. After completion of the training children did no longer have access to the program.

One week after treatment, the post-test was conducted, in which the children completed the tasks of the trained EFs. Follow-up was conducted 6 months after treatment and included all tasks and questionnaires of the pre-test session. Tasks were administered by neuropsychologists with expertise in assessment of children with SLI. All participants completed the post-test. At the follow-up session, one of the participants did not complete three of the included tasks. In addition, all questionnaires were returned at post-test. For the follow-up, the teacher questionnaires of one participant were missing.

Treatment

The intervention is the EF training "Braingame Brian." In this training three EFs (visuospatial WM, inhibition and cognitive flexibility) are trained, embedded in a game like environment (Prins et al., 2013; Ten Brink et al., 2013). In this game world the main character is Brian. He is a creative boy that likes to invent things. The training consists of 25 sessions of about 45 minutes, played by the child four times a week over a period of 6 weeks. The child does not play more than one session of 45 minutes each day of training. All sessions contain two blocks (of about 15 minutes) of the three training tasks in a fixed order; first the WM training task, second the inhibition training task and third the cognitive flexibility training task. After each block of training tasks, the difficulty level of the training tasks is automatically adjusted to the child's level of performance. Before, after and between training tasks, the child can walk around in the game world. To enhance motivation, every completed block of training tasks results in an elaboration of the game world and extra powers for Brian. With these extra powers, he can create interventions to help people in his village, resulting in happier village people (the more Brian helps them, the more they smile).

The WM training

The WM training, embedded in the game world, combines different types of WM training (Dovis et al., 2008a). It consists of five levels: (a) training of short-term memory, (b) training of short-term memory, updating and keeping information online, (c) training of short-term memory and manipulation/updating, (d) training of short-term memory and keeping information online during a delay, and (e) training of short-term memory, keeping information online and manipulation of information/updating. Each level is trained for 5 of the 25 sessions. At each level, the training consists of a 4×4 grid of equally sized rectangles. The rectangles light up in a random sequence. The first rectangle light up for 900 ms, and after 500 ms, the next one lights up. After each sequence, the child has to reproduce the sequence by clicking the rectangles in the right order with the computer mouse. The first level of the training requires the child to reproduce the sequences of rectangles that lightened up. In the other levels, tasks are more complex. The child for instance has to hold information online about the position of side bars that lightened up before the rectangles appeared or to remember the position of rectangles in different colors (i.e., repeat first orange and then purple rectangles). The child finishes a session if he or she has reproduced the required amount of sequences. During the training, the sequence length is adapted to the child's individual level of performance.

The inhibition training

This task was designed to train prepotent response inhibition (Dovis et al., 2008b). The task is visually designed as a factory, in which the child has to respond as quickly and accurately as possible to an arrow on a machine. In the first block of trials (practice block), a stimulus lights up on the left or right side of the machine. These are the "go trials." If the stimulus lights up on the left, the child has to press the left button (Q key), and if the stimulus lights up on the right, the child has to press the right button (P key). It is not a matter of responding as quickly as possible, but to respond within a certain range; a stimulus at the top of the screen shows the range within which the child has to respond (a bar colored green between 700 and 1,000 ms and red before 700 ms and after 1,000 ms). In the next block the "stop trials" are introduced. In these stop-trials, after presentation of the stimulus, a stop-signal is given (a tone, and the stimulus on the machine turns red). Then the child has to inhibit his or her ongoing response. The time a child needs to stop his or her response is the Stop Signal Reaction Time. This reaction time is progressively shortened; the presentation of the stop-signal is automatically adjusted to the level of the child's performance. In total, 25% of the trials are stop trials and 75% are go trials. A block has to be replayed if the child has more than 20% errors on the go trials and 30% errors on the stop trials.

The cognitive flexibility training

This task, based on the training described by Karbach and Kray (2009), was designed to train cognitive flexibility (Dovis et al., 2008b). The child has to sort various objects according to the instruction given, in a task which is also visually designed as a factory. The first two blocks are practice blocks. In the first block, the child is instructed to sort objects according to color, and in the second block according to shape (non-switch trials). In the subsequent blocks switch trials are introduced, in which the child has to switch the rule of sorting the parts from color to shape, or from shape to color; 25% of the time are switch trials and 75% of the time are non-switch trials. The interval to respond is progressively shortened based on the child's level of performance on the task. The switch cost is the time needed for switch trials subtracted from the time needed for non-switch trials, and the training is intended to reduce the switch cost. If the child has more than 30% errors on switch or non-switch trials, the test block has to be replayed.

Regarding training compliance, most participants (n = 9) completed all 25 sessions of the training; one participant completed 24 sessions. Because a minimum of 20 training sessions was required to complete the training program, all children were considered to have finished the EF training. The duration of the training period varied from 39 to 50 days. On average participants spend 46.45 minutes on one training session (SD = 9.12).

Outcome measures

Trained executive functions

<u>Visuospatial working memory</u>: The Automated Working Memory Assessment (AWMA) is an automated, computerized assessment battery suitable for use with respondents who are 4 to 22 years of age (Alloway, 2007). The AWMA has been validated and measures the different components of Baddeley's WM model, including verbal storage, verbal central executive (CE), visuospatial storage, and visuospatial CE (Gathercole & Pickering, 2000). The storage measures tap into the phonological loop or visuospatial sketchpad, depending on the nature of the information to be remembered. These tasks appeal to the serial recall of information. For the CE measures, the children must simultaneously store and process information. The processing activity is assumed to tap into the CE component of the WM model. In this study we included one subtest of each component of WM. In all subtest the same span procedure is followed. The length of the sequences increases by one after the child has correctly recalled four sequences of a particular length. Testing is stopped when three sequences of a particular length are not recalled correctly. For each of the subtests standard scores were calculated with a normative mean of 100 (SD = 15).

The subtest *Dot Matrix* is a task of visuospatial storage. In this task a sequence of red dots is presented on a 4 x 5 grid. All of the dots appear in the grid for 2 seconds. The dots then disappear and the child must point to the position of each dot in the same serial order as presented.

The subtest *Spatial Span* is a task of visuospatial CE. In this subtest two identical shapes are presented to the child with a red dot above the right shape. The child must judge whether the two shapes are in normal or mirror image and to remember the location of the dot. The position of the dot rotates to one of three positions of a triangle. After trials requiring the child to judge the similarity of the shapes, they disappear and a triangle of three dots reflecting the possible positions of the previous dots appears. The child must point to the positions of the previous dots in the right order.

Inhibition: The subtest *Walk Don't Walk* of the Test of Everyday Attention for Children (TEA-Ch) requires the periodic and unpredictable withholding of a routine response (Manly et al., 2007). Children are given a sheet showing "paths", each made up of 14 squares. They are asked to listen to a tape that will play one sound (go tone) if the move to the next square should be made and another (no-go tone) if not. The moves are made

by "dotting" each square with a marker pen. The go and no-go tones are identical for the first ms, requiring the child to listen to the entire sound before making their response. The scoring of this tasks is based on the number of trials in which the child marked the correct number of moves. For all tasks included in test protocol except the AWMA, standard scores were calculated with a normative mean of 10 (SD = 3).

<u>Cognitive flexibility</u>: The *Trail Making Test* (TMT) is a test of task switching (Delis et al., 2001). It consists of five parts in which the child is instructed to connect dots with numbers or letters in the right order as fast as possible while still maintaining accuracy. It also includes a number-letter switching condition, in which the child has to switch between numbers and letters. For this study we used this switching condition.

Other neurocognitive functions

<u>Verbal working memory</u>: The subtest *Digit Recall* of the AWMA is a task of verbal storage. In this task the child must recall a sequence of digits in the right order. The digits can range from one to nine and are spoken at a rate of one digit per second. The sequences are randomly generated and no digits are repeated.

The subtest *Listening Span* of the AWMA is a task of verbal CE. In this subtest the child is presented short sentences. The child must then judge whether the content of the sentence is correct (by saying "true" or "false") and remember the first word of the sentence. The number of sentences increases in length and the child must then recall the first words of the sentences in the correct serial order. The sentences have a simple subject-verb-object order and contain early developing vocabulary.

<u>Attention</u>: The subtest Sky Search of the TEA-Ch is a task of selective attention. In this task children are given a laminated A3 sheet depicting rows of paired spacecraft. Four distinctive types of crafts were presented, with most pairs being of mixed type. The children are instructed to find all of the target items, defined by a pair of identical craft, as quickly as possible. Twenty targets are distributed among 108 distractors. Both accuracy and time taken to complete the task are recorded.

The subtest Score! from the TEA-Ch is a task of sustained attention. In this subtest the child has to count identical tones which are separated by silent interstimulus intervals of variable duration. Children are asked to silently count the tones (without assistance from fingers) and to give the total at the end.

The subtest Creature Counting of the TEA-Ch is a measure of attention control. Children have to repeatedly switch between two relatively simple activities of counting upwards and counting downwards. They are asked to count aliens in their burrow, with occasional arrows telling them to change the direction in which they are counting. Time taken and accuracy are scored in this subtest. <u>Planning</u>: The subtest *Six Elements* of the Behavioural Assessment of the Dysexecutive Syndrome for Children (BADS-C) is a test of planning, task scheduling and performance monitoring (Emslie et al., 2003). In this task the children are instructed to do three tasks each of which is divided into two parts. They have to schedule their time on these six subtasks over a five minute period. They are not allowed to do two parts of the same task consecutively. Scoring is based on the number of tasks attempted, and score penalties are given for rule infractions or not spending an equal amount of time on each task.

<u>Fluency</u>: The subtest *Verbal Fluency* of the Developmental Neuropsychological Assessment Battery (NEPSY) is designed to assess verbal productivity through the ability to generate words within specific semantic and initial letter categories. The child is given a semantic or initial letter category and asked to produce as many words as possible in 60 seconds (Korkman et al., 1998).

Behavioral ratings

<u>EF behaviors</u>: The *Behavior Rating Inventory of Executive Function* (BRIEF) is a standardized rating scale for parents and teachers designed to measure EF behaviors of children aged 5–18 years old (Gioia et al., 2000). We used the Dutch version, which contains 75 items (Huizinga & Smidts, 2011). Each item pertains to specific everyday behavior, relevant to EF. The items are categorized in eight no overlapping theoretically and empirically derived clinical scales that measure different aspects of EF: Inhibit, Shifting, Emotional control, Initiate, Working memory, Plan/organise, Organisation of materials, and Monitor. The eight clinical scales form two broader indexes: Behaviour Regulation Index (BRI) and the Metacognition Index (MI). Based on these two composite scores an overall global EF score (i.e. Global Executive Composite, GEC) is calculated. For this study we used the subscales Inhibit, Shift, Working memory, the Metacognition index, and the Total scale as dependent variables. For all the included rating scales the normative mean is 50 (SD = 10), with higher scores indicating more problems.

Behavioral problems: The Child Behavior Checklist (CBCL) is a standardized rating scale for parents to detect emotional and behavioral problems in children and adolescents aged 6 to 18 years (Achenbach & Rescorla, 2001). The questionnaire is made up of eight syndrome scales, including Anxious/depressed, Depressed, Somatic complaints, Thought problems, Attention problems, Rule-breaking behavior and Aggressive behavior. These group into three higher order factors: Internalizing, Externalizing and Total Problems. For this study we used the scales Thought problems, Attention problems. Besides the version for parents, we also included the version for teachers (Teacher Report Form, TRF).

Statistical analyses

Data were analyzed using SPSS Version 21.0. All data were complete for 80% of the children at pre-test, post-test and follow-up. Missing data were imputed using multiple imputation (Rubin, 1987). For all parameters, mean (M) and standard deviation (SD) were calculated. Because of the small number of participants in this pilot study the data cannot be assumed to be normally distributed. For this reason we used the nonparametric Wilcoxon signed-rank test to examine changes in each study period. Scores at the posttest and follow-up were compared to the pre-test scores. Additionally, effect-sizes were calculated using the following equation: $r = z / \sqrt{N}$, in which N is the total number of observations on which z is based. The effect size is a measure of the magnitude of the mean difference. Effect sizes are considered small for r = .20, medium for r = .50 and large for r = .80 (Cohen, 1988).

RESULTS

Pre-Test Results

The descriptive statistics for the pre-test are shown in Table 1. Performances of the participants were compared to the normative mean scores of the different tests in one-sample t-tests. In addition, the number of children scoring –1 standard deviation (SD) or more below the normative mean was calculated.

For the EFs trained in the training program, 2 participants scored –1 SD or more below the normative mean for Dot Matrix, 3 for Walk Don't Walk and 1 for TMT Number-letter switching. However, the average pre-test scores of the participants did not significantly differ from the normative mean scores for Dot Matrix, Walk Don't Walk, and TMT Numberletter switching. None of the participants scored –1 SD below the normative mean for Spatial Span and the average pre-test score for this task was even significantly higher than the normative mean.

Regarding the performances of the participants on the other neurocognitive functions at pre-test, the average pre-test scores of Digit Recall and Score! were significantly below the normative mean, with 7 of the participants scoring –1 SD or below on both tasks. The other pre-test scores did not significantly differ from the normative mean scores.

For the parental rating scale of EF behaviors (BRIEF), only the average pre-test score of Working Memory was significantly higher than the normative mean, with 5 of the participants scoring +1 SD or more. The scores on the other scales and indexes did not significantly differ from the normative mean. For the teacher's rating scale of EF behaviors, the average pre-test scores for GEC, MI, and Working Memory were significantly higher than the normative mean. A total number of 5 participants scores +1 SD or more

Table '	Ta	b	le	1
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Descriptive statistics of the pre-test scores

Measure	 Task	Mean (SD)	т	p	> -1 sd
Trained executive functi	ons				
Visuospatial storage	Dot matrix	97.40 (15.70)	492	.631	2
Visuospatial CE	Spatial recall	108.94 (10.71)	2.506	.034	0
Inhibition	Walk Don't Walk	8.89 (3.29)	-1.011	.338	3
Cognitive flexibility	TMT Number-letter switching	9.63 (3.02)	351	.735	1
Other neurocognitive fu	nctions				
Verbal storage	Digit recall	80.55 (7.42)	-7.864	.000	7
Verbal CE	Listening span	103.29 (17.67)	558	.591	2
Selective attention	Sky Search, correct	9.80 (2.74)	231	.823	2
	Sky Search, time	9.50 (2.22)	711	.495	2
Sustained attention	Count!	6.40 (2.84)	-4.014	.003	7
Attentional control	Creature Counting, correct	8.33 (4.12)	-1.213	.256	4
	Creature Counting, time	7.44 (4.36)	-1.758	.113	5
Planning	Six elements	10.83 (3.49)	.585	.575	1
Fluency	Verbal fluency	9.56 (3.57)	373	.718	3
Behavioral ratings					
EF behaviors	BRIEF parents GEC	54.50 (9.22)	1.544	.157	1
	BRIEF parents MI	55.50 (7.29)	2.232	.053	2
	BRIEF parents inhibition	56.70 (11.66)	1.817	.103	3
	BRIEF parents cogn. flexibility	48.00 (11.81)	535	.605	2
	BRIEF parents working memory	63.10 (10.08)	4.109	.003	5
	BRIEF teacher GEC	62.33 (13.09)	2.827	.020	5
	BRIEF teacher MI	70.78 (24.29)	2.566	.030	6
	BRIEF teacher inhibition	55.44 (7.27)	2.248	.051	2
	BRIEF teacher cogn. flexibility	55.67 (8.70)	1.953	.083	2
	BRIEF teacher working memory	76.44 (23.39)	3.391	.008	6
Behavioral problems	CBCL Total	60.90 (10.21)	3.375	.008	5
	CBCL Internalizing	55.20 (10.92)	1.506	.166	3
	CBCL Externalizing	59.40 (10.63)	2.797	.021	6
	CBCL Thought problems	62.50 (13.08)	3.021	.014	5
	CBCL Attention problems	66.20 (8.44)	6.067	.000	7
	TRF Total	55.40 (8.78)	1.944	.084	2
	TRF Internalizing	52.80 (8.56)	1.034	.328	2
	TRF Externalizing	51.80 (5.87)	.970	.357	0
	TRF Thought problems	55.40 (6.31)	2.706	.024	3
	TRF Attention problems	59.20 (4.24)	6.866	.000	4

Table 2

on GEC, 6 on MI and 6 on Working Memory. The scores for Inhibition and Cognitive Flexibility did not significantly differ from the normative mean.

Regarding the rating scale for behavioral problems, significantly more problems were reported on the CBCL compared to the normative mean for the Total Score, Externalizing, Thought Problems, and Attention Problems. A total number of 5 participants scored +1 SD or more on the Total Score, 6 on Externalizing, 5 on Thought Problems and 7 on Attention Problems. For the TRF, the average pre-test scores differed from the normative mean for Thought Problems, and Attention Problems, with 3 of the participants scoring +1 SD or more on Thought Problems and 4 on Attention Problems. The average pre-test scores on the other scales did not significantly differ from the normative mean.

Trained Executive Functions outcomes

The descriptive statistics for the trained EFs at pre-test, post-test and follow-up are displayed in Table 2. At the post-test, the sample showed significant improvement on the TMT Number-letter Switching, with a medium effect size. The other EF measures did not show significant improvement, although a trend was found for Dot Matrix and Walk Don't Walk, with small to medium effect sizes.

From the pre-test to the follow-up, there was a significant increase in the scores of Dot Matrix, Walk Don't Walk, and TMT Number-letter Switching. The effect sizes all were medium. The other measures did not show significant improvement.

Measure	Pre-test	Post-test	Follow-up	Post-te	est	Follow	-up	
	Mean (SD)	Mean (SD)	Mean (SD)	р	r	р	r	
Dot matrix	97.40 (15.70)	118.55 (14.34)	113.95 (11.16)	.069	.429	.025	.528	
Spatial span	108.94 (10.71)	112.86 (14.84)	114.563 (7.88)	.204	.299	.161	.330	
Walk Don't Walk	8.89 (3.29)	11.50 (3.21)	12.25 (3.33)	.078	.415	.034	.499	
TMT Number-letter switching	9.63 (3.02)	11.38 (1.92)	11.75 (2.92)	.047	.467	.017	.563	

Scores on the trained executive functions at pre-test, post-test and follow-up.

Other neurocognitive functions outcomes

The descriptive statistics for the other neurocognitive functions and behavioral ratings at pre-test and follow-up are shown in Table 3. Regarding the measures of other neuro-cognitive functions, the sample showed significant improvement at the follow-up on Score! and Creature Counting correct, with medium effect sizes. The other neurocognitive functions measures did not show significant improvement.

Tab		2
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Scores on the neurocognitive functions and behavioral ratings at pre-test and follow-up.

Measure	Pre-test	Follow-up	р	r
Digit recall	80.55 (7.42)	77.50 (6.61)	.107	.380
Listening span	103.29 (17.67)	106.06 (13.47)	.866	.040
Sky Search, correct	9.80 (2.74)	9.50 (2.12)	.833	.047
Sky Search, time	9.50 (2.22)	9.80 (2.57)	.675	.094
Count!	6.40 (2.84)	8.20 (3.12)	.040	.460
Creature Counting, correct	8.33 (4.12)	11.30 (1.95)	.018	.545
Creature Counting, time	7.44 (4.36)	9.40 (4,65)	.234	.273
Six elements	10.83 (3.49)	9.33 (2.50)	.414	.192
Verbal fluency	9.56 (3.57)	10,14 (3.13)	.854	.043
BRIEF parents GEC	54.50 (9.22)	51.20 (7.07)	.066	.411
BRIEF parents MI	55.50 (7.29)	51.20 (6.05)	.018	.528
BRIEF parents inhibition	56.70 (11.66)	53.10 (7.91)	.102	.365
BRIEF parents cogn. flexibility	48.00 (11.81)	47.50 (8.18)	.721	.080
BRIEF parents working memory	63.10 (10.08)	54.40 (5.87)	.005	.628
BRIEF teacher GEC	62.33 (13.09)	51.20 (9.78)	.091	.388
BRIEF teacher MI	70.78 (24.29)	54.30 (11.34)	.263	.257
BRIEF teacher inhibition	55.44 (7.27)	52.10 (9.15)	.202	.293
BRIEF teacher cogn. flexibility	55.67 (8.70)	47.10 (7.11)	.119	.358
BRIEF teacher working memory	76.44 (23.39)	56.60 (11.61)	.093	.385
CBCL Total	60.90 (10.21)	55.78 (10.10)	,038	.476
CBCL Internalizing	55.20 (10.92)	52.56 (9.75)	.352	.214
CBCL Externalizing	59.40 (10.63)	55.67 (10.25)	.028	.505
CBCL Thought problems	62.50 (13.08)	57.33 (7.00)	.027	.506
CBCL Attention problems	66.20 (8.44)	60.22 (5.83)	.034	.486
TRF Total	55.40 (8.78)	54.50 (8.21)	.546	.137
TRF Internalizing	52.80 (8.56)	52.10 (12.28)	.721	.080
TRF Externalizing	51.80 (5.87)	51.20 (8.89)	.885	.038
TRF Thought problems	55.40 (6.31)	54.80 (5.41)	.733	.076
TRF Attention problems	59.20 (4.24)	55.30 (4.76)	.021	.518

Behavioral ratings outcomes

For the behavioral rating of EF, parents reported at the follow-up significantly less problems on the Metacognition Index and Working Memory scale of the BRIEF (see Table 3). The effect sizes both were medium. The other scales and indexes of the parents version of the BRIEF did not show significant improvement. In addition, none of the scales or indexes of the teacher version of the BRIEF did show significant improvement. With the exception of the Internalizing scale, parents reported less behavioral problems on all the scales of the CBCL The effect sizes all were medium. On the TRF, teachers reported less Attention Problems, with a medium effect size. The other scales or indexes did not show significant improvement.

DISCUSSION

The purpose of this pilot study was to explore the possible effects of a computer-based EF training (Braingame Brian) in children with SLI. We examined whether EF training produced significant improvement on tasks of the trained EFs (visuospatial WM, inhibition, cognitive flexibility), other neurocognitive functions (verbal WM, attention, planning and fluency) and/or parents and teachers' ratings of EF and behavioral problems in a sample of 10 children with SLI.

To start, we examined the performances of the included children at pre-test. The initial performances of the children with SLI on measures of the verbal storage component of WM and sustained attention were significantly below average compared to peers of the same age. These results are in line with results from former research showing clear impairments in verbal storage in children with SLI (Archibald and Gathercole, 2006b; Conti-Ramsden, 2003; Gray, 2006; Montgomery et al., 2010). Deficits in attention in children with SLI have also been reported in some previous studies (Finneran et al. 2009; Spaulding et al., 2008). Additionally, both parents and teachers reported at the pre-test more problems in WM, thought problems and attention problems on behavioral rating scales compared to the normative mean. Parents also reported significantly more overall and externalizing behavior problems, while teachers reported more overall problems in EF. Problems on everyday EF behaviors in children with SLI have been reported before in several studies (Cuperus et al., 2014; Hughes et al., 2009; Vugs et al., 2013).

With regard to our first research question, namely whether EF training produced significant improvement on tasks of the trained EFs in children with SLI, results showed significant improvement on a task of cognitive flexibility directly after training and a positive trend for the visuospatial storage component of WM and inhibition. At 6 months follow-up children with SLI performed significantly better on the visuospatial storage component of WM, inhibition and cognitive flexibility. The magnitude of improvement was moderate for all the outcome measures showing a significant training effect: effect sizes all were medium, varying from r = .467 to r = .563. These findings replicate the results of former studies showing near-transfer effects (Holmes et al., 2010; Gray et al., 2012; Klingberg et al., 2002, 2005). Meta-analyses on the effectiveness of WM training in children with ADHD also reported medium effect sizes for visuospatial WM directly

after training and at follow-up (Melby-Lervåg & Hulme, 2012; Rapport et al., 2013). Based on previous research is generally accepted that computerized training leads to positive effects on tasks closely related to the trained tasks.

Our second and third research questions concerned possible far-transfer effects, namely whether EF training produced significant improvement on tasks of other neuro-cognitive functions and/or parents and teachers' ratings of EF and behavioral problems in children with SLI. At 6 months follow-up, we found significant improvement on two tasks of neurocognitive functions that were not trained in the program: sustained attention and attention control. Regarding the behavioral ratings, both parents and teachers reported significantly fewer attention problems. Moreover, parents reported significantly less problems in WM and metacognition, thought problems, externalizing behavioral problems, and overall behavioral problems. The magnitude of improvement was moderate for all these outcome measures: effect sizes were medium, varying from r = .460 to r = .628.

So, besides the near-transfer effects, some generalization of the training effects to other neurocognitive functions and behavior occurred in children with SLI in the current study. Although far-transfer effect were also found in some previous studies (Egeland et al., 2013; Klingberg et al., 2005), results concerning the generalization of the trained task effects are certainly not uniform. The generalization of effects on functions not closely related to the trained tasks is still a point of discussion. In contrast to the current findings, a recent meta-analysis showed negligible far-transfer effects of WM training (Melby-Lervag & Hulme, 2012). This difference might be explained by the nature of the interventions. Whereas the interventions examined in the meta-analysis only included WM training, the training in the current study was a broad EF training for WM, inhibition and cognitive flexibility. In a recent study using the same EF training, Van der Oord and colleagues (2012) also found medium to large training effects for behavioral ratings of EF and attention problems in children with ADHD.

Closer inspection of the WM tasks in the current study showed only a significant training effect on the visuospatial storage task, which is closely related to the visuospatial WM task in the training. Tasks measuring the verbal storage, verbal CE and visuospatial CE components of WM did not show significant improvement after training. This suggests that the training effect on the visuospatial storage component of WM does not generalize to other non-trained components of WM. Although previous research showed some limitations in visuospatial WM in children with SLI, deficits are definitely most profound in verbal WM in these children. In the current study, the largest impairment was also found on a verbal WM task: the participants scored significantly below the normative mean on Digit recall before and after the training. Therefore, the inclusion of a verbal WM training task in the training program should be considered for children with SLI. Such a domain-specific WM training might be more effective, but further research will be necessary to examine this (Kroesbergen et al., 2012).

There are some important methodological limitations of this pilot study that should be considered in interpreting the results. First, the sample size was small, which makes the conclusions sensitive to random effects. It also is possible that some of the nonsignificant results (especially those showing a trend) would have reached statistical significance in a larger sample. Furthermore, as an initial pilot examining EF training in children with SLI, no control group was included. For this reason, test-retest effects and other experimental confounds like maturation effects cannot be ruled out (Shadish et al., 2002). Another concern is the use of unblinded raters. Parents and teachers all were aware the children received the EF training, which may have led to biased reports. It cannot be ruled out that improvement on behavioral ratings also reflected expectations of outcome rather than changes solely produced by the training. On the other hand however, it is often argued that parents are more biased in their ratings than teachers, because they are involved in supporting and motivating their child during the training while teachers are not. Previous studies on WM training in children with ADHD often have not found significant effects on the behavioral ratings of teachers (Beck et al., 2010; Klingberg et al., 2005). Although the current results also showed most improvement in the behavioral ratings by parents, it is promising that we did find a significant effect on the teachers rating of attention problems.

Although the current pilot study must be considered exploratory due to the mentioned methodological limitations, the results support the importance of a large-scale, randomized controlled trial investigating the possible effects of the current training program in children with SLI. Further research with a robust study design should reveal whether EF training is really an appropriate and effective intervention for children with SLI. In the first place the current training effects have to be replicated, but future studies should also disentangle these effects in order to explore the specific contributions of the different trained EFs and to examine which children with SLI respond best to this intervention.

The substantial limitations in EF in children with SLI reported in previous studies, support the premise that EF training might be effective in this population. It seems plausible that good EF are an important condition for the processes of learning various linguistic skills and that it is effective to improve these conditions. The ability to hold information in mind (working memory), to tune out irrelevant information (inhibition), and to switch the focus of attention between activities (cognitive flexibility) are involved in almost all everyday situations of learning new language abilities. If it is proven possible to improve EF in children with SLI by computerized training, this might also have an (indirect) effect on the linguistic skills of these children. Near-transfer is namely the mechanism through wich far-transfer to other functions is hypothesized to occur (Shipstead et al., 2012). In the current pilot study however, no linguistic tasks were included. The inclusion of these tasks to examine whether the training effects generalize to nontrained language outcomes could be a valuable addition for future research.

Chapter 8

Executive Function Training Effects in Children with SLI

Based on:

Vugs, B., Cuperus, J., Hendriks, M., Knoor, H., & Verhoeven, L. Executive Function Training Effects in Children with SLI. *Submitted*



ABSTRACT

Background: Growing evidence suggests that executive functions (EFs) are in some way involved in the problems associated with specific language impairment (SLI). This raises the question whether EF training might be a meaningful intervention for these children. The present study examined the effectiveness of a computerized EF training in children with SLI, ages 8 to 12 years.

Methods: Forty-four children with SLI were randomized to either an EF training group or a Wait-list group. The EF-training consisted of 25 sessions over a period of 6 weeks and included training tasks of visuospatial working memory (WM), inhibition and verbal WM. Performances of the EF training group were compared to the Wait-list group on the three trained EFs (near-transfer) and tasks of cognitive flexibility, attention and behavioral ratings of EFs (far-transfer) directly after training. Further, training effects on receptive and expressive language abilities were examined. The EF training group also completed a follow-up 3 months after training.

Results: Clear near-transfer effects were found for the three trained EFs directly after training. The effect-sizes of these training effects all were large and effects appeared long-lasting for visuospatial WM and one of the verbal WM tasks. In addition, positive trends with medium effect-sizes occurred for both the receptive and expressive language tasks directly after treatment, but these effects were no longer significant when corrected for multiple testing. At 3-months follow-up, the EF training group showed significant improvement on a grammar task. The result did not show significant far-transfer effects for cognitive flexibility, attention or the behavioral ratings of EFs.

Conclusions: The current results of near-transfer effects on the trained EFs and generalization of the training effects on grammatical abilities, support the premise that EF training could be a promising intervention in children with SLI.
INTRODUCTION

Growing evidence suggests that executive functions (EFs) are in some way involved in the problems associated with specific language impairment (SLI). Several studies reported deficits in EFs in children with SLI (Pauls & Archibald, 2016; Henry et al., 2011; Im-Bolter et al., 2006). This raises the question whether EF training might be a meaningful intervention for these children: Is it possible to improve EFs in children with SLI by extensive training? And if so, does it have a positive effect on their language abilities? To date, research examining interventions directed at improving EFs in children with SLI is very limited. The present study therefore evaluates the effects of EF training in children with SLI.

Executive functions in children with SLI

Children with SLI fail to make normal progress in language development despite the absence of underlying intellectual, frank neurological, social, or emotional impairments (Bishop, 2006). They form a heterogeneous group with individual children showing different profiles of language deficits; the impairment can affect various linguistic domains including phonological, morphological, lexical and grammatical domains. Moreover, the linguistic profile often changes with age and development (Bishop, 2006; Leonard, 1998). Different theories and hypotheses have been proposed to explain the underlying causes of SLI. Some hypotheses presume that SLI originates from a deficit or delay specific to language, and particularly grammar (Rice and Wexler, 1996; Van der Lely, 2005). More recently, however, growing evidence indicates that non-linguistic factors are also involved in SLI (Bishop, 2006; Montgomery et al., 2010). Evidence that children with SLI have impairments in non-linguistic factors that are not restricted to language, resulted in domain-general accounts of the disorder. One factor that has been often implicated in this light is EFs. Several studies provided evidence of EF deficits in children with SLI (Archibald & Gathercole, 2006); Henry et al., 2011; Im-Bolter et al., 2006; Lum et al., 2011)

EFs are cognitive processes responsible for purposeful, goal directed behavior. Most recent models considered it a multifaceted concept with distinct subfunctions that are inter-related and function together as an integrated, supervisory control system (Miyake & Shah, 1999; Miyake et al., 2000; Stuss &Alexander, 2000). Although some uncertainties remain about the exact components of EFs, the three most frequently postulated components are working memory (WM), inhibition and cognitive flexibility (Huizinga et al., 2006; Miyake et al., 2000). WM refers to the structures and processes used to temporarily store and manipulate information (Baddeley & Hitch, 1974; Baddeley, 2003). Inhibition refers to the processes related to the control of attention and the ability to stop ongoing responses (Miyake et al., 2000). Cognitive flexibility, often also described as shifting, has

been conceptualized as the ability to switch the focus of attention between activities or problem-solving strategies (Miyake et al., 2000).

Strong links have especially been found between WM limitations and SLI (Archibald & Gathercole, 2006a; Bishop, 2006; Montgomery et al., 2010). A widely accepted account of the deficits associated with SLI, for example, is the phonological storage deficit hypothesis and the underlying assumption that a specific deficit in the temporary storage of novel phonological information underlies SLI (Archibald & Gathercole, 2007; Baddeley, 2003; Bishop, 1996; Coady & Evans, 2008; Gathercole & Baddeley, 1990). Significant group differences have been reported between children with SLI versus TD children on tasks of non-word repetition, recall of words, and recall of digits, (Archibald and Gathercole, 2006b; Gray, 2003, 2006; Conti-Ramsden, 2003). In addition to these constraints on verbal storage, substantial deficits have been reported for verbal CE. Children with SLI are even more severely and consistently impaired on verbal complex memory tasks than on straightforward verbal storage tasks (Archibald & Gathercole, 2006a, 2006c; Ellis Weismer et al., 1999; Marton & Schwartz, 2003). Visuospatial WM has been less extensively investigated with somewhat contradictory results. There is as yet no consensus regarding the role of visuospatial WM in the speech and language of children with SLI. Based on studies showing children with SLI and their TD peers to perform similarly on visuospatial WM tasks, several authors assume that the WM deficits of children with SLI are limited to the verbal domain (Alloway & Archibald, 2008; Archibald & Gathercole, 2006a, 2006b; Baird et al., 2009; Lum et al., 2011; Riccio et al., 2007; Williams et al., 2000). In contrast, the results of other studies and a recent meta-analyses have yielded evidence suggesting that the WM deficit of children with SLI may extend to the visuospatial domain (Vugs et al., 2013). Although medium effect sizes for both visuospatial storage and visuospatial CE tasks were found in the meta-analyses, results also showed that the deficit for visuospatial WM is not as large as the deficit for verbal WM in children with SLI. The deficit in the verbal WM of children with SLI is two to three times larger than the deficit in their visuospatial WM.

Significant group differences have also been reported between children with SLI versus TD children on several tasks of inhibition, such as go/no-go tasks and tasks requiring resistance of distractors (Bishop & Norbury, 2005; Dodwell & Bavin, 2008; Marton et al., 2007; Spaulding, 2010). Recently, Pauls & Archibald (2016) conducted a meta-analysis of EFs in children with SLI, including inhibition. The results showed children with SLI to perform significantly below their TD peers on inhibitory control tasks (g = -.56). In most studies, no group differences were found between children with SLI and their TD peers on tasks of cognitive flexibility, involving set-shifting tasks and the Trailmaking Test (Dibbets et al., 2006; Henry et al., 2012; Im-Bolter et al., 2006). However, when the cognitive flexibility tasks involved more complex stimuli such as in the Wisconsin Card Sorting Task, findings varied (Henry et al., 2012; Marton et al, 2008). Results of a recent meta-analysis of cognitive flexibility in children with SLI showed a small but reliable effect-size (g = -.27) (Pauls & Archibald, 2016). However, the authors considered the difference in cognitive flexibility between children with SLI and their TD peers to be not clinically significant.

Executive function training

In recent research, there has been growing interest in the possibility to improve EFs by cognitive training. The underlying assumption for such interventions is that the maturation and/or efficiency of the neural circuitries underlying the trained EFs can be improved by intensive practice and training. Several novel, computer-based training programs have demonstrated promise in children and adolescents. To date, most studies particularly focused on the training of WM. Klingberg and colleagues (2002, 2005) were among the first to show, in a randomized controlled study in children with ADHD, that a computer-based WM training improved the trained visuospatial WM of the children. Significant improvement on at least one trained WM task has also been reported in several studies and meta-analytic reviews (Holmes et al., 2010; Gray et al., 2012; Green et al., 2012; Melby-Lervag & Hulme, 2012; Rapport et al., 2013; Shipstead et al., 2012). Based on this, it is generally accepted that WM training leads to positive effects on tasks closely related to the trained tasks, so called near-transfer effects. Furthermore, in a recent meta-analytic review it was found that the training effects on visuospatial WM tasks are maintained at follow-up, on average 5 months after the training (Melby-Lervag & Hulme, 2012).

However, quite some controversy exists about the generalizability or far-transfer of the training effects on functions not closely related to trained tasks, such as other neurocognitive functions, behavior and academic performance (Melby-Lervag & Hulme, 2012; Shipstead et al., 2012). Based on results of reviews documenting limited or negligible far-transfer effects, increasing concerns are expressed about the generalization of the trained task effects in WM training (Chacko et al., 2013; Melby-Lervag & Hulme, 2012; Rapport et al., 2013; Redick et al., 2015; Shipstead et al., 2012). In contrast, a recent systematic review and meta-analysis showed persistent training effects for inattention in daily life for children with ADHD (Spencer & Klingberg, 2015).

Fewer studies have examined the trainability of inhibition and cognitive flexibility. A study on the effectiveness of an inhibition training in TD children showed significant improvement on most of the trained tasks, but no generalization to tasks measuring WM or attention (Thorell et al., 2009). A study on the trainability of cognitive flexibility in children, adolescents and adults, showed significant improvement on cognitive flexibility tasks, and also on other EFs, including WM and interference control. White and Shah (2006) reported significant improvement on both trained and non-trained tasks in adults with ADHD after task-switching training.

Van Oord and colleagues (2012) examined in children with ADHD the effectiveness of a computer-based EF training that combined three EF training tasks of WM, inhibition and cognitive flexibility. In this training program, game elements were added in order to increase children's motivation and potentially optimize their cognitive performance during training. Adding game elements has proven to enhance the cognitive performance of children on EF tasks (Dovis et al., 2012; Prins et al., 2011). The results showed significant improvement on parent-rated EFs and ADHD behavior, with effects maintained at 9 week follow-up. A positive effect of this EF training has also been found in obese children, showing significant improvement on a WM task and childcare workers' reports of WM and metacognition (Verbeken et al., 2013).

Executive function training in children with SLI

EF training in children with SLI has so far only received scarce attention. Whether cognitive nonlinguistic training could have a positive effect on language abilities in children with SLI was investigated in an exploratory single-subject design study by Ebert and Kohnert (2009). They reported significant gains in expressive language abilities after a 5-week training of auditory memory and processing speed. Wener and Archibald (2011) examined in a small scale study the effects of a treatment targeting verbal and visuospatial strategies (including a verbal CE task) in children with SLI, children with WM impairments and children with comorbid language and WM impairments. Five of the seven children with language impairments (either with or without WM impairments) showed improvement on a grammatical task after treatment and at 4-months followup. More recently, Holmes and colleagues (2015) investigated whether WM training could be effective in enhancing verbal WM in a group of 12 children with low language abilities, aged 8-11 years, and 15 matched TD children. Both groups showed significant near-transfer effects on visuospatial storage after treatment. Further exploratory analyses revealed some predictive links between pre-training scores and training outcomes. Children with the lowest verbal IQs at baseline made the greatest gains in verbal STM after training, while visuospatial WM improved to the greatest extend for children with higher verbal abilities.

In a previous study, we evaluated the effectiveness of the EF training "Braingame Brian" in children with SLI. Ten children with SLI, ages 8 to 12 years, completed a 25-session training of visuospatial WM, inhibition and cognitive flexibility over a 6-week period (Vugs et al., 2016). The results showed significant improvement on cognitive flexibility and a positive trend for visuospatial WM and inhibition directly after training. At 6 months follow-up, the children performed significantly better on tasks of all three trained EFs. Furthermore, significant improvement was found on sustained attention, attention control, parent- and teacher-rated attention behavior and parent-rated EFs and externalizing behavior with medium effect sizes. Although the results of this pilot

study had to be considered exploratory due to several methodological limitations, it highlighted the importance of a large-scale, randomized controlled trial investigating the possible effects of EF training in children with SLI.

Present study

The aim of the present study was to examine the effects of the EF training "Braingame Brian" in children with SLI. In its original form, this training program included three EF training tasks of visuospatial WM, inhibition and cognitive flexibility. However, for this study a verbal WM training tasks was included, which replaced the cognitive flexibility training task. As outlined earlier, EF problems are most profound in verbal WM in children with SLI, while evidence for deficits in cognitive flexibility are less compelling (Archibald & Gathercole, 2006a, 2006c; Pauls & Archibald, 2016). The clear links between verbal WM and SLI suggest that especially verbal WM may be an effective area to target in EF training in children with SLI. Furthermore, Wener & Archibald (2011) reported domain- and treatment specific effects of WM training in children with SLI.

The performances of an EF training group were compared to the performances of a Wait-list group on tasks measuring the three trained EFs (near-transfer) and tasks of cognitive flexibility, attention and behavioral ratings of EFs (far-transfer). Given the central role of language abilities in children with SLI, we further examined whether intensive EF training can lead to positive gains in language abilities in children with SLI. The specific research questions were as follows:

- 1) Does EF training result in significant near-transfer effects on the trained EFs and/or far-transfer effects on untrained EFs, attention and parents and/or teachers' ratings of EFs in children with SLI?
- 2) Does EF training result in significant training effects on receptive and/or expressive language in children with SLI?

METHODS

Participants

Children were recruited from two special education school for children with speech and language problems in the Netherlands. Inclusion criteria for participation in the study were: a diagnosis of SLI, age between 8 and 12 years and impairments in WM or inhibition. The diagnosis of SLI was based on extensive clinical and psychometric assessment by speech and language therapists; all participants scored 1.25 SD or more below the mean on at least two language measures (following Tomblin, 1996). Any children with a diagnosis of hearing disorder, frank neurological disorders, or autism spectrum disorder were excluded and all children had average nonverbal intelligence. Seventy-one chil-

dren and their parents received information about the research project and were invited to participate.

Written consent was obtained for 50 children. These children were randomly assigned to either an active EF training condition or a control condition (using random number generators by a person blind to the study). Two children were excluded after the pre-test because they did not show impairments in WM or inhibition. One child in the training group withdrew after the first training session, and one child in the control group moved schools after the pre-test. Data are reported for the remaining 44 children.

Children in the training group completed the EF training Braingame Brian. During the training period children did not receive any other treatment directed at improving EFs. The control group was a Wait-list group with the possibility to start the EF training after completion of the study. The training group included 10 boys and 12 girls with a mean age of 10;02 years (SD 14.63 months) and the control group included 8 boys and 14 girls with a mean age of 9;08 years (SD 10.38). Descriptive statistics for both groups are shown in Table 1. The training group and control group did not differ significantly on age, gender, non-verbal IQ, PPVT, CELF Formulated Sentences or CELF Expressive Vocabulary (Dunn & Dunn, 1997; Kort et al., 2008; Schlichting, 2005).

Measure	EF training (N=22)	Wait-list (N = 22)	Group co	omparison
	Mean (SD)	Mean (SD)	F/ X ²	р
Age	122.23 (14.63)	116.09 (10.38)	2.575	.116
Gender	M: 10, F: 12	M: 8, F: 14	1.467	.116
Non-verbal IQ	98.57 (12.49)	95.05 (9.86)	.843	.365
PPVT	80.50 (11.22)	82.92 (11.34)	.330	.570
CELF, Formulated Sentences	4.07 (1.42)	4.12 (1.90)	.006	.940
CELF, Active Vocabulary	4.82 (1.62)	4.96 (2.60)	.024	.877

Table 1. Demographic characteristics

Note. SD = standard deviation.

Procedure

The study design included a pretest one week before the start of the training and a post-test at the conclusion of the training period for both the training group and control group. Additionally, the training group completed a follow-up 3 months after completion of the training. The researchers conducting the pre-training, post-training and follow-up were blind to group membership.

At the start of the training, the child and his/her parents were given practical information about the training at an introduction session at their home. It was ensured that the program worked on the child's computer and that it was placed at a location with limited distraction. Sessions 1 was led by a research assistant. After the first session there was weekly contact between the research assistant and the parents. All children kept a diary of their experiences with the game, time spent on the training, the number of sessions played in a week, and difficulties or problems were reported. Time spent on training and frequency of the sessions were evaluated in the contacts with the research assistant. After completion of the training participants no longer had access to the program.

One week after treatment, the post-test was conducted, which included all tasks and questionnaires of the pre-test session. The follow-up of the training group 3 months after the treatment included the same tasks and questionnaires. Tasks were administered by psychologists with expertise in assessment of children with SLI. All participants completed the post-test and all children in the training group completed the follow-up. At the post-test, the teacher questionnaires of one participant in the training group and two children in the control group were missing. At the follow-up, only nine parents and eight teachers returned the questionnaires. Due to the large number of missing data, results of the EF questionnaires were not included in analyses of the follow-up.

Treatment

The intervention is the EF training "Braingame Brian." In this training three EFs (visuospatial WM, inhibition and verbal WM) are trained, embedded in a game like environment (Prins et al., 2013; Ten Brink et al., 2013). In this game world the main character is Brian. He is a creative boy that likes to invent things. The training consists of 25 sessions of about 40 minutes, played by the child four times a week over a period of 6 weeks. The child does not play more than one session of 45 minutes each day of training. All sessions contain two blocks (of about 15 minutes) of the three training tasks in a fixed order; first the visuospatial WM training task, second the inhibition training task and third the verbal WM training task. After each block of training tasks, the difficulty level of the training tasks is automatically adjusted to the child s level of performance. Before, after and between training tasks, the child can walk around in the game world. To enhance motivation, every completed block of training tasks results in an elaboration of the game world and extra powers for Brian. With these extra powers, he can create interventions to help people in his village, resulting in happier village people (the more Brian helps them, the more they smile).

In its original form, the EF training 'Braingame Brian" included three EF training tasks of visuospatial WM, inhibition and cognitive flexibility. For this research project a verbal WM training tasks was designed, which replaced the cognitive flexibility training task.

The visuospatial WM training

The visuospatial WM training, embedded in the game world, combines different types of WM training (Dovis et al., 2008a). It consists of five levels: (a) training of short-term

memory, (b) training of short-term memory, updating and keeping information online, (c) training of short-term memory and manipulation/updating, (d) training of short-term memory and keeping information online during a delay, and (e) training of short-term memory, keeping information online and manipulation of information/updating. Each level is trained for 5 of the 25 sessions. At each level, the training consists of a 4×4 grid of equally sized rectangles. The rectangles light up in a random sequence. The first rectangle light up for 900 ms, and after 500 ms, the next one lights up. After each sequence, the child has to reproduce the sequence by clicking the rectangles in the right order with the computer mouse. The first level of the training requires the child to reproduce the sequences of rectangles that lightened up. In the other levels, tasks are more complex. The child for instance has to hold information online about the position of side bars that lightened up before the rectangles appeared or to remember the position of rectangles in different colors (i.e., repeat first orange and then purple rectangles). The child finishes a session if he or she has reproduced the required amount of sequences. During the training, the sequence length is adapted to the child's individual level of performance.

The inhibition training

This task was designed to train prepotent response inhibition (Dovis et al., 2008b). The task is visually designed as a factory, in which the child has to respond as quickly and accurately as possible to an arrow on a machine. In the first block of trials (practice block), a stimulus lights up on the left or right side of the machine. These are the "go trials." If the stimulus lights up on the left, the child has to press the left button (Q key), and if the stimulus lights up on the right, the child has to press the right button (P key). It is not a matter of responding as quickly as possible, but to respond within a certain range; a stimulus at the top of the screen shows the range within which the child has to respond (a bar colored green between 700 and 1,000 ms and red before 700 ms and after 1,000 ms). In the next block the "stop trials" are introduced. In these stop-trials, after presentation of the stimulus, a stop-signal is given (a tone, and the stimulus on the machine turns red). Then the child has to inhibit his or her ongoing response. The time a child needs to stop his or her response is the Stop Signal Reaction Time. This reaction time is progressively shortened; the presentation of the stop-signal is automatically adjusted to the level of the child's performance. In total, 25% of the trials are stop trials and 75% are go trials. A block has to be replayed if the child has more than 20% errors on the go trials and 30% errors on the stop trials.

The verbal working memory training

The verbal WM training resembles the structure of the visuospatial WM training, combining different types of WM training in five levels. The first level is a training of short-term memory and requires the child to reproduce a sequence of auditory presented words. The words are spoken at a rate of one digit per second. After each sequence, the child has to reproduce the sequence by clicking the pictures of the words in a 3 x 3 grid in the right order with the computer mouse. In the second level, the child has to reproduce a sequence of auditory presented numbers in the reverse order by clicking the numbers in a 3 x 3 grid. The digits range from one to nine and are spoken at a rate of one digit per second. The third level contains sequences of words and numbers. The child must remember the numbers first and afterwards the words in the right order. In the fourth level, a keyword has to be hold online while a sequence of words is presented. The child has to reproduce the sequence numbers first and afterwards the keyword. In the last level, the child judges whether the content of short sentences is correct (true or false) and must remember the first word of the sentences. After a series of sentences have been presented the child recalls the first words of the sentences in the correct serial order. The number of sentences in each set increases. Each level is trained for 5 of the 25 sessions. The child finishes a session if he or she has reproduced the required amount of sequences. During the training, the sequence length is adapted to the child's individual level of performance.

Outcome measures

Trained executive functions

<u>Visuospatial working memory</u>: The subtest Spatial Span of the Wechsler Nonverbal Scale of Ability (WNV) requires the child to point to blocks in the same serial order as presented. In the second part of the test the child has to point to the blocks in the reverse order (Wechsler & Naglieri, 2006).

Inhibition: The subtest Walk Don't Walk of the Test of Everyday Attention for Children (TEA-Ch) requires the periodic and unpredictable withholding of a routine response (Manly et al., 2007). Children are given a sheet showing "paths", each made up of 14 squares. They are asked to listen to a tape that will play one sound (go tone) if the move to the next square should be made and another (no-go tone) if not. The moves are made by "dotting" each square with a marker pen. The go and no-go tones are identical for the first ms, requiring the child to listen to the entire sound before making their response.

<u>Verbal working memory</u>: The 15-Words Test requires the child to remember as many words as possible from a list of 15 words. After the child responded the words are read again with a total of 5 trials. Twenty minutes after the last trial the child once more has to remember the words (delayed recall).

In the subtest Digit recall of the Dutch version of the Wechsler Intelligence Scale for Children-III children have to repeat sequences of numbers forwards, and in the second part backwards (Kort et al., 2002).

Untrained executive functions and attention

<u>Cognitive flexibility</u>: The subtest Creature Counting of the TEA-Ch is a measure of attention control (Manly et al., 2007). Children have to repeatedly switch between two relatively simple activities of counting upwards and counting downwards. They are asked to count aliens in their burrow, with occasional arrows telling them to change the direction in which they are counting. Time taken and accuracy are scored in this subtest.

<u>Attention</u>: The subtest Score! from the TEA-Ch is a task of sustained attention (Manly et al., 2007). In this subtest the child has to count identical tones which are separated by silent interstimulus intervals of variable duration. Children are asked to silently count the tones (without assistance from fingers) and to give the total at the end.

Language

<u>Receptive</u>: The subtest Comprehension of Instruction of the NEPSY-II is designed to process instructions of increasing syntactic complexity (Korkman et al., 2007). In this subtest children have to point to appropriate stimuli in response to oral instructions.

<u>Expressive</u>: In the subtest Recalling sentences of the CELF-4 the child has to repeat sentences of increasing length and complexity (Kort et al., 2008). The task calls on the child's grammatical skills as it requires knowledge of the rules of grammar and sentence structure.

Behavioral ratings

<u>EF behaviors</u>: The Behavior Rating Inventory of Executive Function (BRIEF) is a standardized rating scale for parents and teachers designed to measure EF behaviors of children aged 5–18 years old (Gioia et al., 2000). We used the Dutch version, which contains of 75 items (Huizinga & Smidts, 2011). Each item pertains to specific everyday behavior, relevant to EFs. The items are categorized in eight no overlapping theoretically and empirically derived clinical scales that measure different aspects of EFs: Inhibit, Shifting, Emotional control, Initiate, Working memory, Plan/organise, Organisation of materials, and Monitor. The eight clinical scales form two broader indexes: Behaviour Regulation Index (BRI) and the Metacognition Index (MI). Based on these two composite scores an overall global EF score (i.e. Global Executive Composite, GEC) is calculated. For this study we used the subscales Inhibit, Working memory, the Metacognition index, and the Total scale as dependent variables.

Statistical analyses

Baseline differences between the EF training condition and Wait-list condition were tested using ANOVAs and chi-square tests. Then, differences between the EF training

group and control group were tested with ANOVAs for repeated measures analyses with time of assessment as within factor (prestest, posttest) and group as between factor (EF training or Wait-list), To assess long-term effects for the children randomized to the EF training condition a within group ANOVA for repeated measures analyses was conducted with time as within factor (pretest, posttest, follow-up). The Bonferroni method was used to correct for multiple testing. Thresholds for statistical significance were *p* < .01 for the trained EFs, *p* < .025 for the untrained EFs and attention, *p* < .006 for the behavioral ratings, and *p* < .025 for the language measures. Effect-sizes were calculated for all analyses. Effect-sizes are considered small for η^2 < .06, medium for η^2 between .06 and .14 and large for η^2 > .14 (Cohen, 1988).

RESULTS

Pre-training

First, ANOVAs were conducted to examine baseline differences between the EF training group and Wait-list group. Descriptive statistics for the pretest are shown in Table 2. No significant differences were found between the two treatment conditions on any of the baseline variables.

Training effects

Regarding the trained EFs, the repeated measures analyses showed significant interaction effects for Spatial Span, Walk Don't Walk, 15WT total and Digit Recall, with large effect-sizes. In all cases children in the EF training group improved more after training than children in the Wait-list group (see Table 3).

No significant interaction effects occurred for cognitive flexibility, attention or the behavioral ratings of EFs. With regard to the behavioral ratings, the results showed both parents and teachers of children in the EF-training group to report less behavioral problems after treatment on the WM scale of the BRIEF compared to children in the Wait-list group. Although the effect-sizes were medium in magnitude, gains on the parents and teacher WM scale of the BRIEF were no longer significant when corrections were made for multiple comparisons: BRIEF parent WM p = .043 and BRIEF teacher WM p = .040.

Two trends emerged on the receptive and expressive language tasks with medium effect-sizes, but interaction effects were no longer significant at the Bonferroni threshold: Comprehension of Instructions p = .041 and Recalling Sentences p = .041. Children in the EF training group improved more on both language tasks after treatment than children in the Wait-list group.

Table 2	
Baseline comparison between EF training group and Wait-list group	

Measure	Task	EF Training (N=22)	Wait-list (N=22)	Group co	omparison
		Mean (SD)	Mean (SD)	F	р
Trained executive	functions				
Visuospatial WM	Spatial Span (WNV)	11.68 (2.88)	11.68 (3.01)	.000	1.000
Inhibition	Walk Don't Walk (TEA-Ch)	11.00 (4.11)	10.36 (3.20)	.329,	.569
Verbal WM	15WT total	41.91 (10.86)	37.41 (5.94)	2.906	.096
	15WT recall	9.27 (2.79)	8.50 (2.39)	.972	.330
	Digit recall (WISC III)	8.68 (1.86)	8.56 (1.97)	.099	.755
Untrained execution	ive functions and attention	I			
Cognitive flexibility	Creature Counting (TEA-Ch)	5.57 (2.18)	5.14 (1.85)	.471	.496
Attention	Score! (TEA-Ch)	6.91 (2.72)	6.14 (2.77)	.872	.356
Language					
Receptive	Comprehension of Instructions (NEPSY-II)	23.00 (3.11)	22.81 (3.58)	.031	.860
Expressive	Recalling Sentences (CELF-4)	31.24 (9.96)	29.09 (12.71)	.378	.542
Behavioral rating	S				
EF behaviors	BRIEF parents GEC	47.68 (8.79)	50.68 (8.67)	1.300	.261
	BRIEF parents MCI	48.59 (9.41)	49.31 (7.27)	.082	.776
	BRIEF parents inhibition	56.32 (9.57)	56.86 (8.14)	.041	.840
	BRIEF parents WM	46.91 (8.82)	51.23 (9.29)	2.497	.122
	BRIEF teacher GEC	53.39 (12.61)	51.43 (7.68)	.455	.504
	BRIEF teacher MCI	54.05 (12.34)	51.33 (8.59)	.693	.410
	BRIEF teacher inhibition	59.18 (13.08)	56.57 (9.14)	.570	.455
	BRIEF teacher WM	50.91 (10.73)	51.57 (9.01)	.048	.828

scores at pretest and	t positiest for the EF training gr	oup ana wait-i	upup.											
Measure	Task	Pre-	test	Post	:-test	-	ime		שֿ	dno.		Time	x Grou	đ
		EF-Training Mean (SD)	Wait-list Mean (SD)	EF-Training Mean (SD)	Wait-list Mean (SD)	LL.	٩	² ר	ш	٩	² ר	ш	٩	² ل
Trained executive f	unctions													
Visuospatial WM	Spatial Span (WNV)	11.68 (2.88)	11.68 (3.01)	14.68 (2.83)	11.45 (3.29)	17.951	000	299	3.630	064 .(080	24.319	000	367
Inhibition	Walk Don't Walk (TEA-Ch)	11.47 (3.53)	10.36 (3.20)	13.14 (3.61)	9.43 (4.11)	.631	.432 .0	015	5.739	021	123	8.012	.007	163
Verbal WM	15WT total	41.91 (10.86)	37.41 (5.94)	45.45 (9.44)	34.96 (8.79)	.246	.622	306	9.288	004	181	7.422	600.	151
	15WT recall	9.27 (2.79)	8.50 (2.39)	9.95 (3.18)	7.95 (2.94)	.028	. 869	100	3.398	072 .(075	2.243	.142	.051
	Digit recall (WISC III)	8.68 (1.86)	8.50 (1.97)	10.50 (1.89)	8.63 (2.28)	10.151	.003	195	3.836	057 .(084	7.516	600.	.152
Untrained executiv	e functions and attention													
Cognitive flexibility	Creature Counting (TEA-Ch)	5.57 (2.18)	5.14 (1.85)	5.48 (1.66)	5.24 (1.89)	000	1.00 .0	000	406	528 .(010	.130	.721	003
Attention	Score! (TEA-Ch)	6.91 (2.72)	6.14 (2.77)	7.77 (1.60)	6.00 (2.29)	1.349	.252 .(331	3.841	057 .(084	2.550	.118	.057
Language														
Receptive	Comprehension of Instructions (NEPSY-II)	23.05 (3.27)	22.81 (3.58)	24.68 (3.04)	22.64 (3.66)	2.845	.100	968	1.358	251 .(034	4.452	.041	102
Expressive	Recalling Sentences (CELF-4)	31.24 (9.96)	29.09 (12.71)	36.14 (9.14)	29.50 (11.49)	6.241	.017	132	1.929	172 .(045	4.467	.041	860
Behavioral ratings														
EF behaviors	BRIEF parents GEC	47.68 (8.79)	50.68 (8.67)	46.82 (7.49)	50.09 (8.42)	.514	.477 .(012	1.849	181 .(042	.018	.894	000
	BRIEF parents MCI	48.59 (9.41)	49.32 (7.22)	46.95 (7.63)	49.91 (6.77)	.323	.573 .(308	.716	402 .(017	1.465	.233	.034
	BRIEF parents Inhibition	46.91 (8.82)	51.23 (9.29)	45.56 (6.41)	51.27 (9.25)	.726	. 399	217	4.191	047 .(091	.829	.368	.019
	BRIEF parents WM	56.32 (9.57)	56.86 (8.14)	52.14 (6.69)	57.63 (7.85)	2.056	.159 .(347	1.989	166 .(045	4.343	.043	.094
	BRIEF teacher GEC	51.90 (10.07)	50.95 (7.83)	49.95 (9.69)	53.42 (13.25)	.020	. 889	100	.216	645 .(006	1.923	.240	.036
	BRIEF teacher MCI	52.38 (9.79)	48.43 (7.39)	50.74 (8.81)	51.32 (8.51)	1.827	.185 .(046	.065	801 .(002	3.296	.077	080
	BRIEF teacher Inhibition	50.76 (10.98)	48.61 (8.22)	51.68 (9.06)	51.84 (8.71)	2.750	.105 .(267	514	478 .(013	3.695	.062	089
	BRIEF teacher WM	57.33 (10.04)	50.90 (7.72)	56.37 (9.59)	56.00 (8.84)	5.661	.022	130	.684	413 .(018	4.500	.040	.106

Scores at pretest and posttest for the EF training group and Wait-list group.

Table 3

Table 4

Long-term effects

Repeated measures ANOVAs were conducted with Time (pre-test, post-test, follow-up) as within-group factor for the children in the EF-training group. The results of these analyses are displayed in Table 4. For the trained EFs, there were significant effects of time for Spatial Span (WNV) and Digit Recall (WISC-III) with large effect sizes. Post hoc tests revealed that performance on the Spatial Span task significantly improved from pre-test to post-test (p < .001) and from pre-test to follow-up (p < .001). Performance on the Digit Recall task also improved significantly from pre-test to post-test (p = .001) and from pre-test to follow-up (p = .001) and from pre-test to follow-up (p = .029).

With regard to cognitive flexibility and attention, no significant effects of Time occurred for Creature Counting (TEA-Ch) and Score! (TEA-Ch).

For the language measures, significant effects of Time were found for Comprehension of Instructions (NEPSY-II) and Recalling Sentences (CELF-4) with large effect-sizes. Post hoc tests revealed that performance on the Comprehension of Instructions task improved significant from pre-test to post-test (p = .034), but not from rom pre-test to follow-up (p = .162). Performance on the Recalling Sentences task improved significant from pre-test (p = .014) and from pre-test to follow-up (p = .002).

Measure Task Pre-test Post-test Follow-up Time Mean (SD) Mean (SD) Mean (SD) F η² р **Trained executive functions** Visuospatial WM Spatial Span (WNV) 11.68 (2.88) 14.68 (2.83) 15.69 (2.51) 28.70 .000 .577 Inhibition Walk Don't Walk 11.47 (3.53) 13.14 (3.61) 12.68 (4.32) 3.070 .057 .133 (TEA-Ch) Verbal WM 15WT total 41.91 (10.86) 45.45 (9.44) 45.09 (9.89) 2.928 .065 .122 15WT recall 9.27 (2.79) 9.95 (3.18) 10.64 (2.32) 3.049 .058 .127 Digit recall (WISC III) 8.68 (1.86) 10.09 (2.51) 10.50 (1.89) 8.883 .001 .297 Untrained executive functions and attention Cognitive Creature Counting 5.57 (2.18) 5.48 (1.66) 5.59 (1.76) .057 .935 .003 flexibility (TEA-Ch) Attention Score! (TEA-Ch) 6.91 (2.72) 7.77 (1.60) 7.23 (1.98) 2,789 .073 .117 Language Receptive Comprehension of 23.05 (3.27) 24.68 (3.04) 24.64 (2.97) 4.324 .021 .194 Instructions (NEPSY-II) Expressive **Recalling Sentences** 31.24 (9.96) 36.14 (9.14) 38.23 (10.21) 8.745 .001 .304 (CELF-4)

Scores at pre-test, post-test and follow-up for the EF-training group.

DISCUSSION

This study evaluated the effectiveness of a computerized EF training in children with SLI. The performances of 22 children with SLI, ages 8 to 12 years, who followed the training program were compared to the performances of 22 children in a Wait-list group. It was examined whether EF training resulted in both near-transfer effects on the three trained EFs and far-transfer effects on tasks of cognitive flexibility and attention, and behavioral ratings of EFs. Further, it was examined whether the EF training resulted in positive gains in language abilities in children with SLI.

Near-transfer effects were found on tasks of visuospatial WM, inhibition and verbal WM directly after training. The effect-sizes of these training effects all were large. The training effects also appeared long-lasting for visuospatial WM and one of the verbal WM tasks: the significant improvements of the EF training group on Spatial Span and Digit Recall were maintained at 3-months follow-up. It should be considered that the gains made in the trained EFs are unlikely to be due to a practice effect, because the features of the stimuli in the tasks used as outcome measures were different from that in the training tasks. The current findings of near-transfer effects replicate previous studies showing positive effects on tasks closely related to the trained tasks (Holmes et al., 2010; Gray et al., 2012; Green et al., 2012; Melby-Lervag & Hulme, 2012; Rapport et al., 2013; Shipstead et al., 2012). It indicates that it is possible to improve EFs in children with SLI through intensive training.

Positive training effects were also found on the language tasks. Directly after treatment, positive trends with medium effect-sizes occurred for the EF training group compared to the Wait-list group on both the receptive and expressive language tasks. However, these effects were no longer significant at the Bonferroni threshold. At 3-months follow-up, the EF training group showed significant improvement on the Recalling Sentences task. These results implicate that the improvements in the trained EFs also have a positive effect on the linguistic skills of the children with SLI. It seems plausible that remediation of impaired EFs leads to gains in processes supported by these EFs such as language learning: the ability to hold information in mind (WM) and to tune out irrelevant information (inhibition) are involved in almost all everyday situations of learning new language abilities. Recent neurobiological models of the architecture of language processing support the assumption that EFs are involved in linguistic processes. In the Memory-Unification-Control (MUC) model, for instance, it is assumed that language is subserved by dynamic networks of brain regions, including regions in the dorsolateral prefrontal cortex and anterior cingulate cortex which are responsible for attentional or executive control (Hagoort, 2016). These general control networks are supposed to be linked to brain regions of the core components of the language network in the temporal and frontal cortex.

More specifically, the present results show a positive training effect on the grammatical abilities of children with SLI. After all, sentence recall requires knowledge of the rules of grammar and sentence structure. These findings are in line with the results of a previous small scale study showing children with SLI to improve on a grammatical test after a verbal memory intervention (Wener & Archibald, 2011). A positive effect on grammatical abilities might not be surprisingly given the accumulating evidence that WM skills are related to sentence processing in children with SLI (Archibald 2016; Fortunato-Travares et al., 2015; Frizelle and Fletcher, 2015; Montgomery & Evans, 2009; Noonan et al., 2014). Further support for an association between WM and grammar comes from the Procedural Deficit Hypothesis (Ullman & Pierpont, 2005). According to this theory, the grammatical problems of children with SLI can be explained by abnormalities in brain structures underlying procedural long term memory (i.e., frontal/basal ganglia circuits and the cerebellum). It is further assumed that children with SLI have not only procedural memory deficits, but also WM deficits as both functions rely at least partly on the same affected frontal/basal ganglia circuits (Lum et al., 2011).

With regard to far-transfer effects on cognitive flexibility, attention and/or behavioral ratings of EFs, no significant effects were found. Although positive trends with medium effect-sizes emerged for the parents and teachers' ratings of WM directly after training, gains were no longer significant when corrections were made for multiple testing. The generalizability of training effects on functions not closely related to trained tasks is a general concern in previous research on WM training (Chacko et al., 2013; Melby-Lervag & Hulme, 2012; Rapport et al., 2013; Redick et al., 2015; Shipstead et al., 2012). Due the relatively low power of the current study it could however be premature to draw firm conclusions about far-transfer effects in children with SLI. It should further be noted that most previous studies did not specifically examine training effects in groups of children with low scores on WM. It is likely that improving skills from the impaired range to the average range results in more positive learning advantages than moving skills from the average range to somewhat higher (Archibald, 2016). Recent studies demonstrated that WM training in children with low WM did result in far-transfer effects in math and English (Holmes et al., 2009: Holmes and Gathercole, 2014). Given the reported impairments in WM in children with SLI, it could be hypothesized that training of WM may also result in some far-transfer effects in these children.

Taken together, the results of this study indicate that it is possible to improve EFs in children with SLI by extensive training and that this also has a positive effect on their grammatical abilities. It supports the premise that EF training could be a promising intervention in children with SLI. Further research in this area is however certainly needed. First of all, the current training effects have to be replicated in studies that control for possible non-specific training effects and expectancy effects. Further, future studies should explore the specific contributions of the different trained EFs. Although not explicitely investigated in this study, particularly verbal WM should probably be considered an important component of EF training. Previous research after all showed that verbal

WM was clearly related to the linguistic skills of children with SLI (Archibal 2016; Vugs.et al., 2015). Future research is also needed to reveal which children with SLI respond best to this type of intervention. Children with SLI can show different profiles of strengths and weaknesses in EFs, which could be of importance when considering the effectiveness of EF training. Finally, it could be a potentially valuable direction for future studies to investigate the exact effects of EF training on different linguistic skills in children with SLI. It is recommended to include not only grammatical tasks, but tasks of various linguistic skills. Given the present effects on the effortful sentence recall task, positive effects might possibly also be expected on other, less effortful tasks. It could particularly be of interest to examine the generalization of training effects on vocabulary. Based on the phonological storage deficit hypothesis it is assumed that that limitations in verbal WM contribute to the vocabulary learning difficulties of children with SLI (Gathercole & Baddeley, 1990; Archibald & Gathercole, 2007). In line with this, interventions that target verbal WM may be expected to ameliorate the encoding deficits of children with SLI and to have potential for gains in vocabulary learning.

Some important methodological limitations should be considered in interpreting the present results. First, the study design included a wait-list group and not an active control group (e.g., a training with non-adaptive computer tasks). Therefore, non-specific treatment effects (like for instance attention of the parents) were not controlled for. Moreover, it cannot be ruled out that the training effects found in the present study are due to an 'expectancy' effect. Because children in the EF training group were aware that they were investing in a training program, they may have exerted extra effort during the assessments at post-test and follow-up, as a result of the investment and expectancy during the training (Morrison & Chein, 2011). Additionally, parents and teachers were also aware the children received the EF training, which may have led to biased reports. It is possible that improvement on behavioral ratings reflected expectations of outcome rather than changes solely produced by the training.

In closing, the present findings obviously have some important implications for clinical practice. Interventions directed at improving EFs might be a valuable addition to more traditional interventions that focus solely on linguistic problems. Based on the current results, computerized EF training should be considered a promising intervention to support EFs in children with SLI, particularly for children with impairments in WM or inhibition and grammatical abilities. An alternative approach to support EF difficulties is to train children in the use of effective strategies to cope with their limitations in EFs (e.g., rehearsal, grouping, visualization). Furthermore, it could be effective to minimize the adverse effects of impaired EFs by taking task demands (i.e., task complexity, amount of material, and possible distractors) into account. Perhaps the next step in supporting EFs in children with SLI is to offer an integral treatment combining computerized EF training, strategy training and adjustment of the environment.

Chapter 9

General discussion



The acquisition of language is a complex task placing demands on various cognitive processes, each of which could potentially constrain language learning. The objective of this thesis was to study the role of EFs in children with SLI, taking into account dynamic aspects of the relationship between EFs and SLI like development and trainability. Four main topics were addressed in six clinical studies and a meta-analysis: impairments in EFs in children with SLI, the associations between WM and language abilities, the development of EFs and its relation to language, and the trainability of EFs in children with SLI. In this concluding chapter, an overview of the main results and conclusions will be presented. Furthermore, the strengths and limitations of this thesis will be discussed. Finally, directions for future research and clinical implications are provided.

Impairments in executive functions in children with SLI

Chapter 2 focused on the role of visuospatial WM in children with SLI. A meta-analysis comparing the visuospatial WM performance of children with SLI to that of TD peers, showed significant effect sizes for visuospatial storage (d = 0.49) and visuospatial CE (d = 0.63), indicating deficits in both components of visuospatial WM in children with SLI. This implies that the deficits in WM in children with SLI may not be not restricted to the verbal domain. However, when we compared the magnitude of the WM deficits in the two modalities, the deficit for visuospatial WM is not as large as the deficit for verbal WM: the deficit in the verbal WM of children with SLI is two to three times larger than the deficit in their visuospatial WM. The moderator analyses showed that greater impairment in visuospatial storage was associated with more pervasive language impairment, whereas age was not significant associated with visuospatial WM.

In **chapter 3** behavioural parental and teachers' ratings of EFs on the BRIEF were examined in children with SLI aged 5 to 12 years. The results showed that the prevalence of EF problems in classroom settings in children with SLI was much higher than in the normal population. Compared to the normative mean, teachers reported significantly more problems on almost all EF domains (i.e., Inhibition, Shifting, Emotional control, Initiate, Working memory, Plan/organize, and Monitor). WM and Initiation of behaviour were the most impaired, since more than one third of the children had scores in the clinical range on these scales. Parents reported significantly more WM problems. Furthermore, developmental and gender differences on EF behaviours were found. Overall, older children had less problems in EF behaviours (Initiation of behaviour and WM) than younger children and boys showed more problems than girls. Performance on a WM task was associated with a broad range of EF behaviours and performance on a shifting task was specifically associated with the behavioural rating of shifting. However, all correlations should be considered low.

Chapter 4 focused on young children with SLI. The performances of children with SLI aged 4- and 5-years were compared to that of TD children on measures of WM and

behavioural ratings of EFs. The results showed children with SLI to perform significantly below their TD peers on all components of WM, including verbal storage, verbal CE, visuospatial storage, and visuospatial CE. Parents of the young children with SLI reported significantly more problems in EFs, including problems with inhibition, shifting, emotional control, WM, and planning/organization with most problems reported for WM. Performance on the WM task significantly discriminated between young children with SLI and TD, with 89% of the children classified correctly. The patterns of associations between WM performance and EF behaviours differed for the SLI versus TD groups, showing less consistent and non-specific associations in young children with SLI compared to their TD peers.

Taken together, the children with SLI showed impairments on several EFs at different ages. These impairments did not only reveal at a cognitive level, but also affected EF behaviours in daily situations. Of particular interest are the EF deficits found in preschool children with SLI. To date, the role of EFs in young children with SLI received only scarce attention. However, as early childhood is an essential period for the acquisition of language, it seems especially important to address limitations in EFs in this developmental phase. The current results indicate that in young children with SLI careful attention should be paid to EFs in assessment and treatment.

Specifically focusing on WM, deficits were found in both the verbal and visuospatial domain. So, the impairments of children with SLI seem not to be completely specific to language or the processing of strictly verbal information. These results are in line with other recent studies showing impairments in a range of verbal and nonverbal EFs, and support domain general accounts of SLI (Henry et al., 2015; Kapa & Plante, 2015). It might even bring into question whether "*specific* language impairment" is the most appropriate term for the pattern of impairments demonstrated by children with so-called SLI. However, we also found the verbal WM deficit in children with SLI to be two to three times larger than the deficit in visuospatial WM, indicating that problems are most profound in the verbal domain.

A possible explanation for these findings is that not all children with SLI show deficits in visuospatial WM. In the meta-analysis, the deficit in visuospatial WM was after all found to be larger in children with more widespread language impairment. These results are in line with a previous study showing only a subgroup of children with SLI (i.e., children with more pervasive problems affecting both receptive and expressive language) to experience visuospatial WM problems (Nikisch and Von Kries, 2009) Another possibility is that the impairments in the visuospatial WM capacities of children with SLI mainly reflect domain general processing limitations on executive and attentional control. It suggests that children with SLI especially encounter problems in visuospatial WM tasks when processing load is high, and that the specific ability to process simple visuospatial information is relatively intact. Such an account is in accordance with the assumption that WM performance of children with SLI is constrained by domain general processing deficits together with domain specific verbal deficits, and thus not domain specific visuospatial deficits (Archibald & Gathercole, 2006c; Im-Bolter et al., 2006).

Associations between working memory and language abilities

Chapter 5 examined the underlying structure of WM in young children with and without SLI and analyzed the associations between the components of WM and the language abilities of children with SLI aged 4 and 5 years. The results demonstrated that WM was best represented by a model with four separate but interacting components of verbal storage, visuospatial storage, verbal CE, and visuospatial CE. The associations between the four components of WM did not differ significantly for the SLI children versus TD children. However, the individual components of WM showed varying associations with the language abilities of the children with SLI. The verbal CE component of WM was moderately to strongly associated with all the language abilities, including receptive vocabulary, expressive vocabulary, verbal comprehension, and syntactic development. In addition, a moderate association was found between the verbal storage component of WM and any of the language abilities were not found.

So, the verbal components of WM were obviously more strongly related to the language abilities of young children with SLI than the visuospatial components. The findings of an association between the verbal storage component of WM and the receptive vocabularies of the children with SLI are in line with previous studies showing a clear link between verbal storage and receptive vocabulary (or word learning). Such findings are generally taken as evidence for the assumption that the primary role of the verbal storage component of WM is to support the learning of the phonological structure of language (Gathercole & Baddeley, 1989, 1990; Horohov & Oetting, 2004; Montgomery, 2002). Further, particularly the verbal CE component was found to be significantly related to a wide range of linguistic skills, including some more complex abilities. This is in keeping with a more general set of findings showing clear associations between performance on verbal CE tasks and the development of various language abilities such as sentence comprehension and judgments of grammaticality (Archibald, 2016; Montgomery, 2000; Noonan, et al., 2014; van der Lely, 1996). In this connection, Gathercole (2006) already has suggested that deficits in verbal storage alone may not account for the diversity of linguistic problems found to characterize children with SLI. It seems plausible that the ability to simultaneously store and process verbal information (i.e., verbal CE) is involved in the processes of learning various linguistic skills. Situations that require the maintenance of verbal information in memory while engaging in other types of information processing are very common in everyday life learning.

Developmental perspective

The results of a longitudinal study in which children that were previously examined on several measures of WM and language at age 4- to 5-years (T1), were re-examined at age 7- to 8-years (T2) were described in **chapter 6**. Differences in the development of WM between children with SLI and TD children were examined, and it was explored to what extent language atT2 could be predicted by measures of language and/or WM at T1. The result showed the developmental course of WM to be mostly similar for the children with SLI and their TD peers. There were no differences in the development of the verbal CE, visuospatial storage and visuospatial CE components of WM between the two groups. Only the development of the verbal storage component differed significantly: the children with SLI showed an improvement in verbal storage, while performances of the TD children remained stable. Group comparison, however, revealed that the children with SLI still performed significantly worse on verbal storage compared to their TD peers at T2. Hierarchical regression showed language and verbal CE at T1 to be significant predictors of language at T2, with no differences in the developmental associations between language and WM for the two groups.

Of particular interest are the findings that the verbal CE component of WM at age 4- to 5-year was a significant predictor of language three years later. This suggests a causal relation between verbal WM and language during childhood in children with SLI. Limitations in verbal WM may constrain language development in these children. As mentioned before, it seems plausible that verbal WM is involved in almost all everyday situations of learning new language abilities. However, the present findings do not rule out that language abilities also affect WM. Children for instance use language (internal speech and verbal strategies) to mediate WM performance. It is likely that WM and language develop in reciprocal interaction with changing effects on each other over time. The directionality of the relationship between WM and language abilities is defintely something that needs further investigation. More systematic research is needed to disentangle this complex interplay in children with SLI.

Further, developmental differences were found on behavioural ratings of EFs in children with SLI aged 5 to 12 years, with older children showing less problems in EF behaviours of initiation and WM than younger children (see **chapter 3**). It is known from research in TD children that there is more variability in EFs in early childhood when these skills emerge, whereas skills become more stable when children grow older (Huizinga et al., 2006). However, a significant effect of age was not found in the meta-analysis of visuospatial WM (see **chapter 2**). In addition, other recent meta-analyses comparing the performance of children with SLI to that of TD children on several EFs also did not show significant age effects: impairments on tasks of the verbal storage component of WM (Graf Estes et al., 2007), the CE components of WM (Henry & Botting, 2016), and inhibition (Pauls & Archibald, 2016) in children with SLI were found to be invariant with

age. These results suggest that children with SLI consistently lag behind their TD peers in the performance on several EFs throughout development. A possible explanation for the current age effect on behavioural EF ratings could simply be that decifis in EFs are less expressed in daily behaviours when children with SLI grow older. Moreover, it is a common finding in research on EFs that correlations between performance-based measures of EFs and ratings of EF behaviours are low (Anderson et al., 2002; Chaytor et al., 2006; Vriezen & Pigot, 2007).

Trainability of executive functions

Chapter 7 presented the results of a pilot study on the effects of a computer-based EF training including training tasks of visuospatial WM, inhibition and cognitive flexibility in ten children with SLI ages 8 to 12 years. Treatment outcome was examined directly after training and at 6 months follow-up by tasks of the three trained EFs, tasks of untrained neurocognitive functions (attention, planning and fluency), and ratings of EFs and behavioural problems by parents and teachers. Directly after training, results showed significant improvement on cognitive flexibility and a positive trend for visuospatial storage and inhibition. At 6 months follow-up, the children performed significant improvement on tasks of all three trained EFs. Furthermore, the results showed significant improvement on sustained attention, attention control, parent- and teacher-rated attention behavior, and parent-rated EFs and externalizing behaviour with medium effect sizes.

Chapter 8 focused on the effectiveness of a computer-based EF training including training tasks of visuospatial WM, inhibition and verbal WM. In a randomized controlled study the performances of children with SLI who followed the EF training were compared to the performances of children in a Wait-list group on the three trained EFs (near-transfer) and tasks of cognitive flexibility, attention and behavioural ratings of EFs (far-transfer). Further, training effects on receptive and expressive language abilities were examined. Clear near- transfer effects were found on tasks of all three trained EFs directly after training. Effect-sizes of these effects were large and training effects appeared long-lasting for visuospatial WM and one of the verbal WM tasks. In addition, positive trends with medium effect-sizes occurred for both the receptive and expressive language tasks directly after treatment, but these effects were no longer significant when corrected for multiple testing. At 3-months follow-up, the EF training group showed significant improvement on a grammar task. The results did not show significant far-transfer effects for cognitive flexibility, attention or the behavioural ratings of EFs.

The results of these studies support the premise that EF training could be a promising intervention for children with SLI. The findings of near-transfer effects replicate previous research showing positive effects on tasks closely related to the trained tasks (Holmes et al., 2010; Gray et al., 2012; Green et al., 2012; Melby-Lervag & Hulme, 2012; Rapport et al., 2013; Shipstead et al., 2012). It indicates that it is possible to improve EFs in children

with SLI through intensive training. The results further show that the improvements in the trained EFs also have a positive effect on the linguistic skills of the children with SLI, suggesting that remediation of impaired EFs may lead to gains in processes supported by these EFs such as language learning. More specifically, the results show a positive training effect on the grammatical abilities of children with SLI, which is in line with the results of a previous small scale study showing children with SLI to improve on a grammatical test after a verbal memory intervention (Wener & Archibald, 2011). A positive effect on grammatical abilities might not be surprisingly given the accumulating evidence that verbal WM skills are related to sentence processing in children with SLI (Archibald 2016; Fortunato-Travares et al., 2015; Frizelle and Fletcher, 2015; Montgomery & Evans, 2009; Noonan et al., 2014). Further support for an association between WM and grammar comes from the Procedural Deficit Hypothesis (Ullman & Pierpont, 2005). According to this theory, the grammatical problems of children with SLI can be explained by abnormalities in brain structures underlying procedural long term memory (i.e., frontal/basal ganglia circuits and the cerebellum). It is further assumed that children with SLI have not only procedural memory deficits, but also WM deficits as both functions rely at least partly on the same affected frontal/basal ganglia circuits (Lum et al., 2011).

Strengths and limitations

A major strength of this thesis is that not only studies were included comparing EFs between children with SLI and TD children, but that also the developmental associations between EFs, in particular WM, and language abilities in children with SLI were taken into account in a longitudinal study. This study was one of the first to offer some information on how WM develops in children with SLI across time and how the complex relationship between WM and language abilities may change. Moreover, intervention studies examining the trainability of EFs in children with SLI were included. Although previous research already showed substantial limitations in EFs in children with SLI, studies investigating the effects of EF training were to date very scarce. The results of the current studies therefore lead directly to recommendations for the treatment of children with SLI in clinical practice. Another strength is that a multimodal approach of WM was adopted to examine the role of WM and its relation to language in children with SLI. Different components of WM were examined in conjunction with each other, but also then in conjunction with the developing language abilities of the children. This multimodal approach permitted a more reliable assessment of each WM component than reliance on any single measure (Archibald & Gathercole, 2006a).

Some limitations to the studies presented in this thesis also need to be considered. First of all, only WM tasks were included in most studies and no tasks of other EFs like inhibition and cognitive flexibility. As EFs is typically considered a multifaceted concept with distinct subfunctions, it is important to examine how the different EFs are related to one another in children with SLI. It may particularly be valuable to examine this within a theoretical framework taking into account the relationships between the various EFs during the development of children, like for instance the Developmental Integrative Framework Model proposed by Garon and colleagues (2008). This model posits a hierarchical relationship between the different EFs based on the order in which they emerge. Furthermore, in none of the studies measures of the functioning of the episodic buffer component of WM were included. The inclusion of such information might nevertheless be of value as impairments in this component of WM have recently been reported in children with SLI (Petrucelli et al., 2012). Another concern is the limited age range of the children included in the longitudinal study. Based on this, no conclusions can be drawn with regard to the stability of the associations between WM and language abilities for children older than 8 years of age. Finally, some important limitations of the intervention study have to be considered. One concern is that the study design included a wait-list group and not an active control group (e.g., a training with non-adaptive computer tasks). Therefore, non-specific treatment effects (like for instance attention of the parents) and possible 'expectancy' effects were not controlled for. Another concern is the use of unblinded raters. Parents and teachers were aware the children received the EF training, which may have led to biased reports.

Directions for future research

Like already mentioned, future research is needed to further disentangle the complex interplay between EFs and language in children with SLI. To gain more information on the directionality of the relationship between EFs and language, it may be valuable to examine not only whether EF training influences language abilities, but also whether training of linguistic skills affects EFs. Further, it will be of interest to explore children's use of language during EF tasks. In addition, future research is required on the underlying structure of WM and the developmental associations between WM and language in older children with SLI than the children inculded in the present studies. Changes in the degree and nature of linguistic impairments when children with SLI grow older could in turn change the underlying structure of WM and its associations with language. Just how the associations between the components of WM and language abilities of the children develop — and possibly shift — as the children grow older is obviously something to be determined in future research. Continued research will provide greater insight in the role of WM in the language acquisition of children with SLI.

In one of the studies, a mediating effect of non-verbal intelligence was found. The relationships between EFs, language and intelligence in children with SLI is another topic that needs further investigation. Research in TD children has shown that EFs are correlated with crystalized and fluid intelligence (Arffa et al., 2007; Engle, 1999). A recent meta-analysis on non-verbal intelligence furthermore showed children with SLI

to perform on average 0.69 standard deviations lower than their TD peers (Gallinat & Spaulding, 2014). It has to be determined in future research whether intelligence influences the associations between EFs and language abilities in children with SLI.

A last topic that certainly calls for further research in children with SLI is the effectiveness of EF training. To start, the current training effects have to be replicated in studies that control for possible non-specific training effects and expectancy effects. Further, future studies should explore the specific contributions of the different trained EFs. Although not explicitely investigated in the intervention studies, particularly verbal WM should probably be considered an important component of EF training. The results of this thesis after all showed that especially verbal WM was clearly related to the linguistic skills of children with SLI. Future research is also needed to reveal which children with SLI respond best to this type of intervention. Children with SLI can show different profiles of strengths and weaknesses in EFs, which could be of importance when considering the effectiveness of EF training. Finally, it could be a potentially valuable direction for future studies to investigate the exact effects of EF training on the different linguistic skills of children with SLI. It is recommended to include not only grammatical tasks, but tasks of various linguistic skills. Given the present effects on the effortful sentence recall task, positive effects may possibly also be expected on other, less effortful linguistic tasks. It may particularly be of interest to examine the generalization of training effects on vocabulary. Based on the phonological storage deficit hypothesis it is assumed that limitations in verbal WM contribute to the vocabulary learning difficulties of children with SLI (Gathercole & Baddeley, 1990; Archibald & Gathercole, 2007). In line with this, interventions that target verbal WM may be expected to ameliorate the encoding deficits of children with SLI and to have potential for gains in vocabulary learning.

Clinical implications

In closing, the present findings obviously have some important implications for clinical practice. First of all, it seems important to include EF tasks in the assessment of children with SLI. Attention should not only be paid to the language problems of these children, but also to possible EF impairments that can contribute to their language problems. Evaluation of EFs in children with SLI creates more detailed profiles of their strengths and weaknesses, which in turn can guide appropriate interventions. As the present results with regard to WM showed that the deficits of children with SLI were not restricted to the verbal domain, it is recommended to administer verbal as well as nonverbal tasks. It is obviously important to know if the WM problems being experienced by a child are also visuospatial. For instance for the use of visual support, which is a common intervention strategy adopted for children with SLI. Furthermore, it might be a valuable addition to include behavioural measures of EFs. The inclusion of rating scales of EFs during daily life could be important to assure ecological validity and complement information

gleaned from cognitive measures. The current results also showed that WM performance significantly discriminated between young children with SLI and TD. Although SLI can be reliably identified in preschool children, its diagnosis in clinical practice is sometimes difficult. Possibly, verbal WM measures can contribute to the identification of young children with SLI.

It further seems sensible to pay attention to EFs in the treatment of children with SLI. Interventions focusing on both language and EF problems might result in more optimal results than those using traditional interventions with attention to only linguistic abilities. Although the studies in this thesis only focused on school-aged children with SLI, interventions directed at supporting EFs may also be valuable for younger children. After all, clear impairments in EFs were found in preschool children with SLI and especially this developmental phase is critical for the acquisition of various linguistic skills. Based on the current results, computerized EF training should be considered a promising intervention to support EFs in children with SLI, particularly for children with impairments in WM or inhibition and grammatical abilities. It is further recommended that the adverse effects of impaired EFs be minimized during teaching and remediation by taking task demands (i.e., task complexity, amount of material, and possible distractors) into account. It also could be valuable to train children in the use of effective strategies to cope with their limitations in EFs (e.g., rehearsal, grouping, visualization). Perhaps the way forward to improve interventions for children with SLI is to offer a combination of computerized EF training, strategy training and adjustment of the environment.

Appendices

References

Nederlandse samenvatting

Dankwoord

Publications

Curriculum vitae



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NEDERLANDSE SAMENVATTING

Hoewel de taalontwikkeling van de meeste kinderen ogenschijnlijk als vanzelf verloopt, zijn er ook kinderen met specifieke problemen op dit gebied. Wanneer er sprake is van een achterstand in de taalontwikkeling, die niet kan worden toegeschreven aan een verstandelijke beperking, gehoorproblemen, aantoonbaar neurologisch letsel of ernstige emotionele en psychische problemen wordt gesproken van een taalontwikkelingsstoornis (TOS). Problemen in de taalontwikkeling kunnen zich op verschillende gebieden manifesteren en naarmate kinderen ouder worden, kunnen de aard en de ernst van de taalproblemen veranderen. De prevalentie van TOS wordt geschat op 3-6% bij kinderen in de basisschoolleeftijd.

De laatste jaren wordt steeds meer onderzoek gedaan naar de rol van niet-linguïstische factoren bij kinderen met TOS. Van een aantal neurocognitieve functies is bekend dat ze van invloed zijn op het leren van taal. Diverse studies laten zien dat kinderen met TOS problemen hebben met executieve functies (EF). EF zijn cognitieve processen die verantwoordelijk zijn voor doelgericht en efficiënt gedrag. Ze worden vaak beschouwd als 'het regelcentrum van ons brein'. Er zijn verschillende EF te onderscheiden die onderling gerelateerd zijn en samen fungeren als een geïntegreerd controlesysteem. Als belangrijkste EF worden doorgaans inhibitie, werkgeheugen en cognitieve flexibiliteit genoemd. Inhibitie is het vermogen om niet direct te reageren op een impuls en gedrag te remmen wanneer dat nodig is. Werkgeheugen maakt het mogelijk om informatie gelijktijdig te onthouden en te bewerken. Cognitieve flexibiliteit (ook wel shifting genoemd) is het snel en flexibel kunnen aanpassen van gedrag aan een veranderende situatie.

In dit proefschrift is de rol van EF bij kinderen met TOS nader onderzocht, waarbij ook is gekeken naar meer dynamische aspecten van de relatie tussen EF en TOS, zoals ontwikkeling en trainbaarheid. Om te beginnen zijn EF problemen bij kinderen met TOS nader in kaart gebracht (**hoofdstuk 2 – 4**). In **hoofdstuk 5** is vervolgens gekeken naar relaties tussen werkgeheugen en specifieke taalvaardigheden. **Hoofdstuk 6** beschrijft een longitudinale studie gericht op de ontwikkeling van werkgeheugen in relatie tot taal. Tenslotte komt de mogelijkheid om EF te verbeteren door middel van training aan de orde in **hoofdstuk 7 en 8.** De verschillende studies zijn uitgevoerd binnen Koninklijke Kentalis. Kentalis biedt in Nederland diagnostiek, zorg en onderwijs voor mensen met een TOS en voor mensen die slechthorend, doof of doof-blind zijn.

In **hoofdstuk 1**, de algemene inleiding, worden recente wetenschappelijke bevindingen omtrent EF bij kinderen met TOS besproken. Ook worden de onderzoeksvragen en opzet van dit proefschrift beschreven.

Problemen met executieve functies bij kinderen met TOS

Hoofdstuk 2 richt zich op de rol van visueel-ruimtelijk werkgeheugen bij kinderen met TOS. Hoewel eerder onderzoek duidelijke problemen laat zien op het gebied van het verbaal werkgeheugen, bestaat in de literatuur geen consensus over de rol van het visueel-ruimtelijk werkgeheugen. Een meta-analyse waarin de prestaties van kinderen met TOS en normaal ontwikkelende kinderen op het gebied van visueel-ruimtelijk werkgeheugen werden vergeleken laat zien dat kinderen met TOS vaker dan hun leeftijdsgenoten een probleem hebben met visueel-ruimtelijk werkgeheugen. Deze bevindingen impliceren dat kinderen met TOS niet uitsluitend problemen hebben in het verbale domein. Het bleek echter ook dat de visueel-ruimtelijke werkgeheugenproblemen niet zo ernstig zijn als de problemen op het gebied van het verbaal werkgeheugen: de problemen met verbaal werkgeheugen van kinderen met TOS zijn 2 tot 3 keer groter dan de problemen met visueel-ruimtelijk werkgeheugen. Op basis van deze bevindingen kan worden geconcludeerd dat het van belang is werkgeheugen in kaart te brengen bij kinderen met TOS, waarbij ook aandacht is voor visueel-ruimtelijk werkgeheugen. Indien er visueel-ruimtelijke werkgeheugenproblemen zijn is het relevant dit te weten, onder andere voor de inzet van visuele ondersteuning.

Vervolgens is in **hoofdstuk 3** gekeken naar gedragsvragenlijsten voor EF bij 237 kinderen met TOS in de leeftijd van 5 tot 12 jaar. EF kunnen niet alleen in kaart worden gebracht door middel van cognitieve testen, maar ook door vragenlijsten die kijken naar EF gedrag in dagelijkse situaties. Onderzoek met EF vragenlijsten bij kinderen met TOS is echter nog erg schaars. De resultaten van huidig onderzoek laten zien dat leerkrachten bij kinderen met TOS meer problemen rapporteren ten aanzien van vrijwel alle EF (inhibitie, cognitieve flexibiliteit, emotieregulatie, initiatief nemen, werkgeheugen, plannen en ordelijkheid), waarbij de meeste problemen worden gemeld op het gebied van werkgeheugen en initiatief nemen. Ouders rapporteren meer problemen met werkgeheugen. Conclusie op basis van dit hoofdstuk is dat de EF problemen van kinderen met TOS niet uitsluitend cognitief van aard zijn, maar ook EF gedrag in dagelijkse situaties beïnvloeden. EF gedragsvragenlijsten kunnen dan ook worden gezien als een zinvolle aanvulling in de diagnostiek van kinderen met TOS.

Hoofdstuk 4 beschrijft een studie waarbij de prestaties van 4- en 5-jarige kinderen met TOS op testen voor werkgeheugen en EF gedragsvragenlijsten zijn vergeleken met normaal ontwikkelende kinderen. EF zijn nog nauwelijks onderzocht bij kinderen in de voorschoolse leeftijd, terwijl juist deze periode belangrijk is voor de ontwikkeling van zowel taal als EF. Uit het onderzoek komt naar voren dat jonge kinderen met TOS slechter presteren op zowel verbaal als visueel-ruimtelijk werkgeheugen. Daarnaast rapporteren ouders meer problemen met inhibitie, cognitieve flexibiliteit, emotieregulatie, werkgeheugen en plannen in dagelijkse situaties. Deze resultaten ondersteunen dat ook bij jonge kinderen met TOS aandacht besteed dient te worden aan EF in diagnostiek en behandeling.

Relaties tussen werkgeheugen en taalvaardigheden

In **hoofdstuk 5** zijn de relaties tussen werkgeheugen en de verschillende taalvaardigheden (passieve woordenschat, actieve woordenschat, taalbegrip en zinsontwikkeling) onderzocht bij 4- en 5-jarige kinderen met TOS. Er worden hierbij geen duidelijke relaties gevonden tussen het visueel-ruimtelijk werkgeheugen en specifieke taalvaardigheden. Het verbaal werkgeheugen is daarentegen matig tot sterk geassocieerd met alle onderzochte taalvaardigheden, terwijl het verbaal korte termijngeheugen een samenhang laat zien met passieve woordenschat. Samengevat is het verbaal werkgeheugen dus sterker gerelateerd aan de taalvaardigheden van kinderen met TOS dan het visueel-ruimtelijk werkgeheugen. Het is ook aannemelijk dat het vermogen om verbale informatie gelijktijdig op te slaan en te bewerken (verbaal werkgeheugen) een rol speelt bij het leren van taal in dagelijkse situaties.

Ontwikkeling

Hoofdstuk 6 richt zich op de ontwikkeling van werkgeheugen bij kinderen met TOS. De kinderen die werden onderzocht op de leeftijd van 4- en 5-jaar (hoofdstuk 4) zijn opnieuw onderzocht op de leeftijd van 7- en 8-jaar. De resultaten van deze longitudinale studie laten zien dat de ontwikkeling van werkgeheugen tussen 4 en 8 jaar nagenoeg vergelijkbaar is voor kinderen met TOS en normaal ontwikkelende kinderen. Verder blijkt verbaal werkgeheugen op de leeftijd van 4- en 5-jaar een significante voorspeller voor taalvaardigheden 3 jaar later. Op basis van deze bevindingen wordt geconcludeerd dat problemen in verbaal werkgeheugen van invloed zijn op de ontwikkeling van taalvaardigheden bij kinderen met TOS. Het sluit echter niet uit dat taalvaardigheden ook de ontwikkeling van werkgeheugen beïnvloeden. Het is aannemelijk dat werkgeheugen en taal zich in wederzijdse interactie met elkaar ontwikkelen met wisselende effecten op elkaar naarmate kinderen ouder worden Aanbeveling voor toekomstig onderzoek is dan ook de directionaliteit van de relatie tussen werkgeheugen en taal verder in kaart te brengen.

Executieve functies training

Hoofdstuk 7 beschrijft de resultaten van een pilotstudie naar het effect van een computertraining voor EF bij kinderen met TOS in de leeftijd van 8 tot 12 jaar. Gezien het feit dat EF betrokken zijn bij TOS, is het zinvol om te weten of het mogelijk is EF te verbeteren bij deze kinderen en of dit mogelijk ook een positief effect heeft op hun taalvaardigheden. In deze studie is gebruik gemaakt van de EF training 'Braingame Brian', waarin trainingstaken voor visueel-ruimtelijk werkgeheugen, inhibitie en cognitieve flexibiliteit zijn opgenomen. Direct na de training bleken de kinderen beter te presteren op het gebied van cognitieve flexibiliteit. Bij de follow-up na 6 maanden presteerden ze beter op taken voor alle drie de getrainde EF, taken voor aandacht en vragenlijsten voor EF ingevuld door ouders en leerkrachten. De resultaten van deze pilotstudie ondersteunen het belang van een grootschaligere, gecontroleerde studie naar het effect van EF training bij kinderen met TOS.

In **hoofdstuk 8** is het effect van de EF training 'Braingame Brian' nader onderzocht in een gerandomiseerd gecontroleerd onderzoek. Voor deze studie is de cognitieve flexibiliteit taak uit de originele Braingame Brian training vervangen door een nieuw ontwikkelde verbaal werkgeheugen trainingstaak. Aangezien de EF problemen van kinderen met TOS het meest prominent zijn op het gebied van het verbaal werkgeheugen, is het aannemelijk dat met name training van het verbaal werkgeheugen effect laat zien. Uit het onderzoek komt naar voren dat kinderen met TOS die de training hebben doorlopen beter presteren in vergelijking met de wachtlijstgroep op alle drie de getrainde EF (visueel-ruimtelijk werkgeheugen, inhibitie en verbaal werkgeheugen). De trainingseffecten voor visueel-ruimtelijk en verbaal werkgeheugen werden hierbij ook nog gezien bij follow-up, 3 maanden na de training. Daarnaast laat de trainingsgroep bij de follow-up verbetering zien op een taak voor grammatica. Er werden geen trainingseffecten gevonden voor andere niet-getrainde EF en aandacht. Positieve effecten op de getrainde functies, zogenaamde near-transfer effecten, werden ook veelvuldig gevonden in studies naar EF training bij andere doelgroepen. Over de generalisatie van de trainingseffecten naar andere, niet-getrainde functies (far-transfer) bestaat momenteel echter veel discussie. Bij kinderen met TOS laten de huidige resultaten wel enig far-transfer effect zien op het gebied van taalvaardigheden. Het suggereert dat training van EF mogelijk een positief effect heeft op de processen die ondersteund worden door deze EF, zoals het leren van taal. Op basis van huidige bevindingen kan EF training beschouwd worden als een veelbelovende interventie voor kinderen met TOS en het pleit dan ook voor verder onderzoek op dit gebied.

In **hoofdstuk 9**, de algemene discussie, worden de conclusies op basis van de verschillende onderzoeksbevindingen besproken. Bovendien worden er aanbevelingen gegeven voor de klinische praktijk en vervolgonderzoek.

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CURRICULUM VITAE

Brigitte Vugs was born on December 22, 1977 in Haaren, The Netherlands. She completed her pre-university education at Maurick College, Vught (1996) and obtained a Master in Mental Health at Maastricht University (cum laud, 2000). During this period, she conducted a combined clinical and research internship at Franciscusoord, Adelante Rehabilitation for children and adolescents. Her master thesis examined the neuropsychological profile of boys with Duchenne muscular dystrophy. Afterwards she worked successively as a psychologist at Franciscusoord and HSK, an organization for psychological employee care. In 2002 and 2003, she obtained her post-Master degree as health care psychologist at the GGzE, child and adolescents psychiatry in Eindhoven, and Royal Dutch Kentalis (fomer Sint Marie) in Eindhoven, a national organization specialized in diagnostic, care and educational services to people who are deaf, hard of hearing or deafblind, as well as to people with severe speech/language impairments or autistic spectrum disorders accompanied by severe speech and language difficulties. After this study, she continued working at Royal Dutch Kentalis. In 2008 she enrolled in the specialized clinical neuropsychology postgraduate residency. Part of this education program was to set up a scientific study, which formed the basis of the present thesis. In 2013, she was registered as a certified clinical neuropsychologist. In March 2013, Brigitte started her PhD research at Royal Dutch Kentalis in collaboration with the Radboud University. Since March 2016, she is working at Máxima Medical Centre in Veldhoven.

