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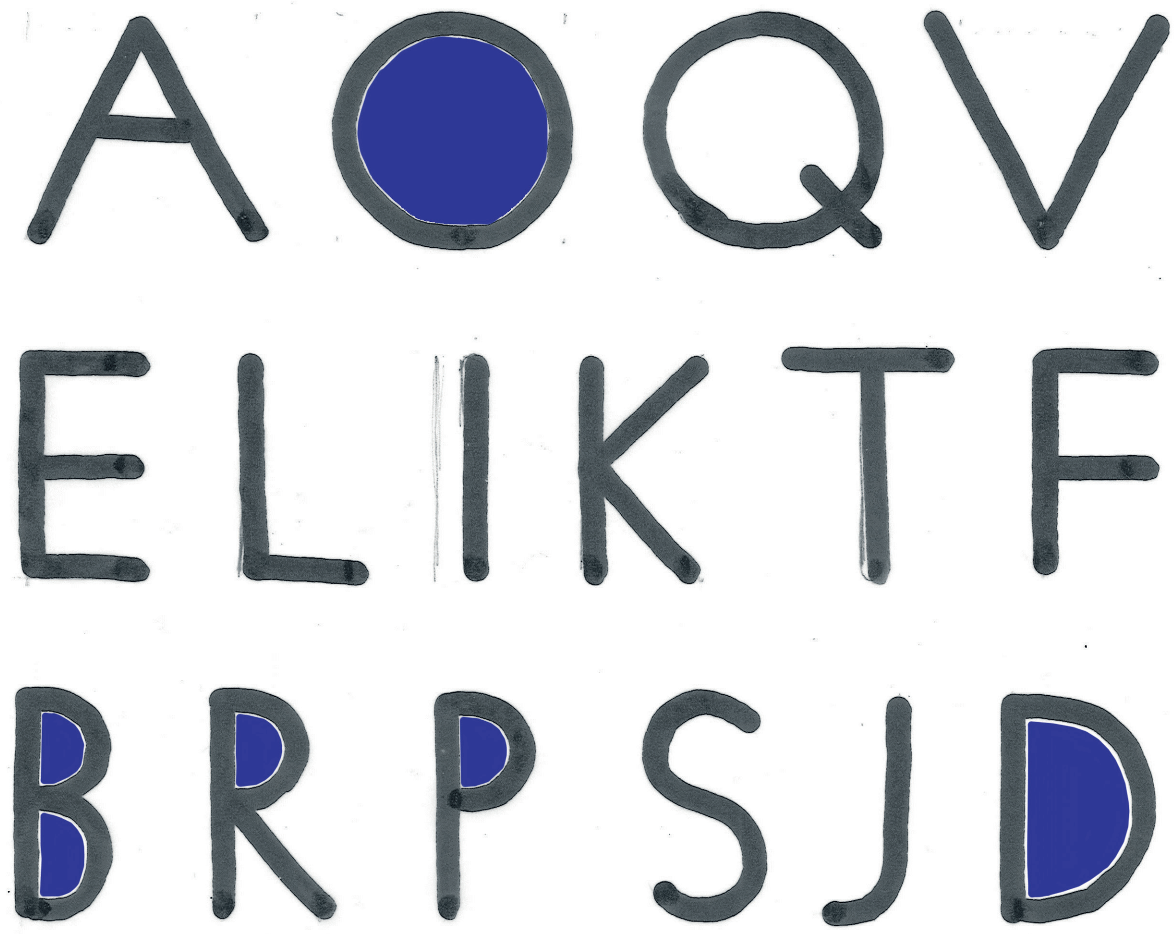

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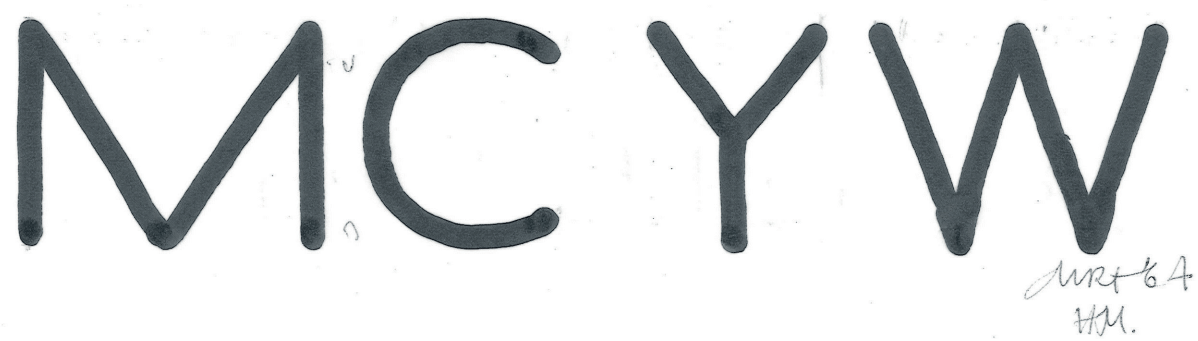
Femke Scheltinga



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Reading problems and dyslexia:
Identification, intervention and treatment within a RTI framework

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Faculteit	Faculteit der Maatschappij en Gedragswetenschappen	

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CHAPTER 1

GENERAL INTRODUCTION

General introduction

Reading development and reading difficulties have drawn interest of many researchers. Not surprisingly, since reading skill is considered one of the most important basic skills that children have to master from the start of formal education. Proficient reading skills support learning in other academic domains and relate to later academic skills (Guthrie, VanMeter, McMann, & Wigfield, 1996). Although the ultimate goal of reading education is comprehension of what has been read, the focus of a large body of research has been on the development of decoding skills and related difficulties. Accurate and fast word decoding is important for reading fluency and this in turn contributes to reading comprehension (Lai, Benjamin, Schwanenfugel, & Kuhn, 2014). Therefore, decoding skills can be considered the foundation for reading development. In the case of reading difficulties, reading comprehension is also likely to be affected. Consequently, various studies have looked at the ethology, course of development and remediation of reading difficulties. This thesis contributes to this field of research.

Recognition of reading problems and dyslexia: from education to diagnosis

Dyslexia is a learning disorder prevalent in 4% in the population (Blomert, 2006). It is a specific learning disorder, limited to reading and/or spelling difficulties. In the Netherlands, two definitions are used. The Stichting Dyslexie Nederland (Dutch Dyslexia Foundation, www.stichtingdyslexienederland.nl) provides a definition that describes dyslexia at the behavioural level. Dyslexia is defined as a disorder characterized by a persistent problem to acquire accurate and/or fluent decoding and/or spelling at the word level. A more extensive definition is given by Blomert (2006), used within the clinical setting. It says that dyslexia refers to a specific reading and spelling disorder with a neurobiological basis, caused by cognitive deficits in phonological and orthographic processing. These result in severe difficulties with reading and spelling in spite of regular education. The latter definition incorporates factors at the neurobiological, cognitive and behavioural level as explanation for the reading problems (see Hulme & Snowling, 2010).

At the behavioural level, the most striking sign of reading disorders is the effortful reading process. The reading problem can be best characterized by poor reading fluency, in particular for reading words (Torgesen, 2005). This is in particular the case for relatively transparent orthographies. In a transparent orthography, graphemes are generally associated with one sound and therefore letter-by-letter decoding enables accurate reading. In relatively transparent orthographies, poor readers often obtain high levels of reading accuracy after only a few months of reading instruction, but their reading rate remains slow (e.g. de Jong & van der Leij, 2003; Holopainen, Ahonen, & Lyttinen, 2001; Landerl & Wimmer, 2008).

Within the Dutch educational setting, reading problems are first identified on the basis of behavioural data. In recent years, a data-driven approach has been promoted as means to monitor and register learning progress. Data that are usually collected within the school encompass test scores for both word reading fluency and text reading fluency. Schools are encouraged to collect data on reading development and to use this information to adjust the instruction to the educational needs of individual children. If children do not benefit from the classroom teaching methods, the instruction and support should be intensified. This in line with another important aspect of the definition of dyslexia, that is *resistance* to regular teaching methods. When a pupil stays behind in reading skill and does not catch up with extra otherwise effective teaching methods, one can question whether a diagnosis of dyslexia is in order.

To diagnose dyslexia, the diagnostician collects additional information. Cognitive deficits underlying dyslexia are tested such as phonological processing. Difficulties in phonological processing are generally acknowledged as the core deficit of dyslexia (e.g. Vellutino et al., 2004). Phonological processing involves the use of speech sounds (Duff & Clarke, 2011) and difficulties are manifested with different skills. Phonological skills that have often been associated with reading ability in the literature on reading problems are phonological awareness, phonological memory and rapid naming. Besides phonological processing, orthographic processing has also been put forward as essential factor for the development of reading ability. Evidently, one needs to understand how sounds map systematically onto letters to enable accurate decoding (Ehri, 2005).

One objective to collect information at different levels is to exclude other explanations for the reading difficulties. Diagnostics serve to confirm the presence of markers specific to dyslexia. On the other hand, the possibility of comorbidity is examined. Dyslexia co-occurs with several other learning disorders. In particular, the overlap between dyslexia and specific language impairment (SLI) has been topic of several studies. Both disorders have been associated with phonological processing deficits, but the reading profiles differ (e.g. McArthur, Hogben, Edwards, Heath, & Mengler, 2000). Therefore, it has been suggested that the phonological deficits might not be identical in severity or exact nature (Joanisse, 2004).

Response to intervention

The response to intervention (RTI) approach has been proposed as model to prevent, help and identify children with reading disabilities. It describes levels or tiers of instruction, support and intervention. Tier 1 refers to high quality teaching within the classroom setting. For those children falling behind despite high-quality instruction, extra intervention is provided. Tier 2 intervention is defined as additional interventions that are limited in time and delivered one-to-one or in small groups. Children who show no or poor progress are identified for additional tier 3 defined as additional, highly personalised intervention, delivered on an one basis and providing ongoing support for learning. So within a RTI approach, responsiveness determines the intervention intensity that is needed. The model takes into account the degree to which a pupil at risk for reading difficulties responds to intervention and reduces the reading delay, ensuring persistence of the reading difficulties and resistance to instruction and intervention (e.g. Linan-Thompson, Vaughn, Prater, & Cirino, 2006). A similar model has also been adopted in the Netherlands ('*Onderwijscontinuüm*', Struiksma & Rurup, 2008). It is an organizational model that describes how to fulfil the educational needs of children with reading difficulties.

Table 1. Overview Dutch RTI framework '*Onderwijscontinuüm*'

Level 1	High-quality classroom teaching
Level 2	Extended instruction, within the classroom (lowest 25%)
Level 3	Intensive, individual instruction (lowest 10%)
Level 4	Specialized treatment in the clinical setting (4%)

The RTI framework is mainly proposed as means to identify the hard-to-remediate or non-responsive children and to reduce the number of children that develop severe reading difficulties (e.g. Burns, Appleton, & Stehouwer, 2005; Vellutino, Scanlon, Zhang, & Schatschneider, 2008). It also has been questioned whether response to intervention can be predicted at pretest. Only a few studies have explored the skills that can predict responsiveness to reading intervention (e.g. Al Otaiba & Fuchs, 2002; Nelson, Benner, & Gonzalez, 2003). Individual differences in rapid naming skill, phonological awareness, alphabetic knowledge and memory have been shown to be partly related to intervention outcome. Contrary, Zijlstra (2015) found that responders and non-responders were comparable on cognitive skills. If responsiveness is not or only limited predicted by initial reading and reading related skills, the use of RTI as means to identify children with severe reading difficulties is further supported.

Effective components of reading remediation

Within the RTI framework, progress is monitored according to different tiers of intervention. An important assumption is that the intervention was validated and of high-quality. Reviews and meta-analyses have reported on the evidence-based features of reading interventions (e.g. Elbaum, Tejero Hughes and Watson Moody, 2000; Reynolds, Whedall & Madelaine, 2011).

Overall, it has been argued that interventions need to be focused, explicit, systematic and intensive (Shaywitz, Morris, & Shaywitz, 2008). The intervention should focus on strengthening the connections between orthography and phonology during reading practice (Bus & IJzendoorn, 1999). Explicit instruction in grapheme-phoneme correspondences (i.e. structured, explicit phonics instruction), combined with segmentation skills, enables children to grasp the alphabetic principal (e.g. Lovett, Barron & Benson, 2003). Reading of new words is supported by drawing attention to the orthographic patterns within words (Conrad & Levy, 2011). This is most effective when it is embedded within broader reading context (Swanson, 1999), with word reading and text reading exercises so children can practice their knowledge and strategies with new materials (Wanzek & Vaughn, 2008).

Moreover, intervention should provide children with many repeated opportunities to practice reading fluency and comprehension (Lovett et al., 1994). To improve word and text reading fluency, repeated reading has been shown to be effective (Kuhn & Stahl, 2003; Therrien, 2004). Repeated reading is often practiced by reading one text being read multiple times, but repeated reading of different texts with similar reading level appears to be evenly effective (Kuhn, et al, 2003; O'Connor, White, & Swanson, 2007). To speed up word recognition, also other techniques have been used. Words and letter patterns are presented with limited exposure time, i.e. flashed presentation, to speed up processing (Bar-

Kochva & Hasselhorn, 2015; van den Bosch, Bon & Schreuder, 1995; Das-Smaal, Klapwijk & van der Leij, 1996).

Although interventions with these components resulted in gains for the majority of children, gains in word reading fluency appear more difficult to obtain. Very poor readers can be effectively taught decoding skills (Lyon & Moats, 1997), but reading fluency remains poor (Torgesen, 2005). Remediation of the word reading fluency difficulties is challenging and was one of the objectives of this thesis.

Outline thesis

The main purpose of the present study was to explore the remediation of dyslexia within a RTI framework. The remediation focused on the improvement of word reading fluency since the reading difficulties of children with severe dyslexia can be best characterized by poor reading fluency (e.g. Fletcher, 2009). In addition, we examined reading related cognitive skills that are typical for dyslexia and response to intervention.

In *chapter 2*, we first explore the specificity of reading skills and underlying reading related skills in dyslexia. In general, phonological processing difficulties are considered causally related to the decoding difficulties in dyslexia (Vellutino, Fletcher, Snowling, & Scanlon, 2004). This has led to the universal phonological core deficit theory of dyslexia. Phonological deficits have also been suggested as underlying deficit of the language difficulties in specific language impairment (SLI). However, children with SLI do not always develop decoding difficulties. This raises questions about the commonalities and differences in reading skills and phonological processing in children with dyslexia and SLI.

The objective of the study in *chapter 3* was to investigate possible predictors of responsiveness to a tier 2 school-based reading intervention. Intervention at school aims to prevent severe reading difficulties. Intensive reading intervention can help children to improve their reading skills (Torgesen, 2000), but not all children respond adequately to interventions (e.g. Struiksma, van der Leij, & Stoel, 2009). These children are referred to as so called non-responders (McMaster, Fuchs, Fuchs, & Compton, 2005). The response to intervention approach helps to identify these hard-to-remediate children. The aim of this study was to explore whether and to what extent reading related subskills explain differences in responsiveness to intervention.

In *chapter 4*, we present the outcomes for an outpatient treatment in the clinical setting. Children who showed no or poor response to an intensive intervention at school (tier 2) were referred to a clinic for reading disorders. Children received an experimental or control treatment. The experimental treatment was designed to improve the word reading fluency of children with severe dyslexia by focusing on the mapping between phonology and orthography of sublexical features of words. A computerized word training program was. implemented as part of 50 weekly treatment sessions.

Chapter 5 presents a more detailed investigation of the effect of word training program that was used in the experimental treatment condition. The word training program consisted of practice in reading words with orthographic sublexical patterns. We examined the progress in word reading rate and accuracy scores across and within sessions. The aim was to determine whether the training resulted in better reading for trained words and transfer to untrained words.

The *final chapter* provides a summary of the findings from the preceding chapters. Furthermore, findings are discussed in the light of practical implications and suggestions for future research.

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CHAPTER 2

COMMONALITIES AND DIFFERENCES IN PHONOLOGICAL PROCESSING IN CLINICAL GROUPS OF CHILDREN WITH DYSLEXIA AND SLI

Abstract

In this study, Dutch children with dyslexia (DYS), children with specific language impairment (SLI) and children with typical reading and language development (CONTR) were compared on several measures of phonological processing and memory. The central question was whether children with dyslexia and SLI differ on the phonological core characteristics. In addition, we explored the association of reading skills and language impairment to phonological processing tasks. Children with DYS and SLI showed poor phonological awareness and rapid naming skills. Although level of performance was most strongly related to reading level in both groups, children with SLI showed (mild) deficits on phoneme awareness and serial rapid naming that could not be traced to their reading level. They were more severely hampered on tasks with a phonological short-term memory component that seemed largely independent of reading achievement. Gradient differences in performance on phonological decoding were best explained by reading level. We conclude that the core deficit of dyslexia is best characterized by tasks that involve phoneme awareness and serial rapid naming, whereas SLI is associated with phonological short-term memory impairments. Moreover, part of the phonological abilities appear independent of reading level in SLI.

Background

A great deal of research places a strong emphasis on the association between phonological processing and reading. Phonological processing refers to a wide range of linguistic operations that make use of information about the speech sound (i.e. phonological) structure of the language (Catts, 1989), including phonological awareness, verbal short-term memory and lexical retrieval of phonological information (Wagner & Torgesen, 1987). The strong relationship between different phonological processing skills and reading has been found across orthographies, although the exact manifestation and effect depend on the orthographic characteristics of the language (e.g. Caravolas, Volín, & Hulme, 2005; Furness & Samuelsson, 2010; Landerl, Wimmer, & Frith, 2000). Separate phonological processing skills contribute to individual differences in specific reading skills (Boets, De Smedt, Cleuren, Vandewalle, Wouters, & Ghesquière, 2010). More in particular, deficits are considered causally related to dyslexia, known as a specific reading disorder in word reading (Vellutino et al., 2004). However, phonological processing deficits have also been mentioned as an underlying deficit of specific language impairment (SLI) (Chiat, 2001; Bortolini & Leonard, 2000; Orsolini, Sechi, Maronato, Bonvino & Corcelli, 2001), which is defined as a developmental language disorder that manifests itself on a broad range of language skills. A number of studies have reported similar reading problems in both developmental disorders as a consequence of phonological processing deficits. However, children with SLI do not always develop word-reading problems (McArthur, Hogben, Edwards, Heath & Mengler, 2000). Therefore, it has been suggested that phonological deficits possibly occur in both dyslexia *and* SLI, but these deficits might not be identical in severity or exact nature (Joanisse, 2004), resulting in different reading profiles. In this study, the occurrence of phonological processing deficits and word reading difficulties in children with dyslexia and SLI in learning to read Dutch is further explored. The Dutch case is interesting since it could give more insight into the importance of phonological processing for reading a relatively transparent orthography.

Phonological processing deficits as a cause of reading problems

The understanding that spoken words consist of individual speech sounds (phonemes), i.e. phoneme awareness appears an important prerequisite for reading development, in particular reading accuracy, enabling mapping sounds to letters. Its predictive influence seems limited to the first years of literacy acquisition and less important in transparent orthographies (Furness & Samuelsson, 2010; de Jong & van der Leij, 1999, 2003; Landerl & Wimmer, 2000). It has been argued that phoneme awareness improves by learning to read as a consequence of the regularities between letter-sound correspondences. However, tasks which require phoneme awareness are difficult for children with dyslexia (Vellutino et al., 2004) and also at a later age children with dyslexia remain performing poorly on phoneme awareness tasks as task complexity is increased (Boets, et al., 2010; de Jong & van der Leij, 2003; Caravolas, Volín & Hulme, 2005).

Limitations in verbal short-term memory or storage of phonological information are also often mentioned as a correlate of dyslexia (for an overview see Hulme & Roodenrys, 1995; Montgomery, 2002). It has been suggested that phonological short-term memory puts constraints on the number of sounds that can be kept in memory during the process of analyzing and blending. Difficulties are manifested by poor performance on tasks that tap memory span for words, digits (Avons & Hanna, 1995; de Jong, 1998; Elbro, 1996; Kramer, Knee, & Delis, 2000) and nonwords of varying length (Roodenrys & Stokes, 2001; Snowling, 2000; Wimmer, Mayringer, Landerl, 1998). Deficits in phonological short-term memory can be regarded as a correlate of dyslexia (de Jong, 2006), although its contribution has been found to be small (Boets et al., 2010) and findings of poor performance on memory tasks are not consistently found across studies (van der Sluis, van der Leij, and de Jong, 2005; Vaessen et al., 2010). Moreover, its independent, causal role has been questioned (de Jong & van der Leij, 1999), as it relates strongly to measures of phonological awareness.

Another skill that has been associated with reading ability is rapid serial naming (RAN). There has been some debate whether RAN can be considered a component of phonological processing. It has been questioned whether the task measures the ability to retrieve phonological information from long term memory (Vellutino, et al., 2004) or the ability to integrate visual and phonological information rapidly (Wolf & Bowers, 1999) which is assumed to be important in orthographic learning. Nonetheless, there is abundant evidence showing that rapid naming is a strong predictor of reading and reading difficulties across orthographies. Some studies have suggested that RAN is the most or even only skill that is important (Wimmer, 2001). Others have found that also other phonological processing skills contribute (Boets, et al., 2010). Rapid naming has a single and independent contribution besides phonological awareness and phonological memory (Boets et al., 2010; de Jong & van der Leij, 2003). Its contribution is particularly strong to reading fluency (Boets, et al., 2010; Furness & Samuelsson, 2010; Wimmer, Mayringer & Landerl, 2000; de Jong & van der Leij, 2003).

Phonological processing and SLI

Several studies have reported that many if not all children with SLI show deficits on tasks involving phonological processing similar to deficits found in dyslexia. Children with SLI perform poorly on measures of phoneme awareness (Bishop & Adams, 1990; Briscoe, Bishop & Norbury, 2001; Leitão, Hogben & Fletcher, 1997; Nithart, et al., 2009; Rispens, 2004; Snowling, Bishop & Stothard, 2000; Vanderwalle, Boets, Ghesquière & Zink, 2010). They are slow at naming pictures of common objects (Lahey & Edwards, 1999; Leitõa, Hogben, Fletcher, 1997; McGregor, Newman, Reilly & Capone, 2002). Poor performance is also reported on tasks of verbal short-term memory, tapped by span tasks and nonword repetition (Baddeley, Gathercole & Papagno, 1998; Botting & Conti-Ramsden, 2001; de Bree, Wilsenach & Gerrits, 2004; Weismer, Tomblin, Zhang, Buckwalter, Chynoweth & Jones, 2000; Gathercole, 2006; Gathercole & Baddeley, 1990; Rispens, 2004). Deficits in phonological short-term memory appear most severe in SLI (Nithart et al., 2009), in particular when measured by nonword repetition tasks (Rispens, 2004; de Bree et al., 2004; Bishop, North & Donlan, 1996).

Considering the high incidence of phonological deficits in SLI and the strong relation between measures of phonological processing and reading development, it may be expected children with SLI experience reading problems similar to children with dyslexia. Indeed, several authors have found that children with SLI are at risk for literacy problems (e.g. Bird et al. 1995, Snowling, 2005; van Weerdenburg, 2005). On the other hand, not all children with language impairment develop reading problems (Carroll & Meijers, 2010; Simkin & Conti-Ramsden, 2006). McArthur et al. (2000) reported that only about half of the children with SLI develop reading problems. Catts et al. (2005), using less stringent selection criteria, found an even lower incidence (17%). These findings have led to different possible explanations in which SLI and dyslexia are considered distinct or identical disorders stemming from one or several deficits (e.g. Catts, 1993; Bishop & Snowling, 2004; Pennington & Bishop, 2008).

Overlap in deficits between dyslexia and SLI

Catts, Adlof, Hogan and Ellis Weismer (2005) suggested that reading problems in both dyslexia and SLI stem from the phonological processing deficits. They compared children with dyslexia, SLI and combined SLI *and* dyslexia on performance at tasks of phoneme awareness and nonword repetition. Both groups with reading problems performed worse on these tasks in comparison to the SLI group without reading problems. The pure SLI group exhibited only mild phonological processing deficits. Catts et al. concluded that phonological deficits do not necessarily occur in all children with SLI, only in case of co-occurrence with dyslexia.

In contrast, Vandewalle et al. (2010) showed that all children with SLI performed poorly on measures of phonological awareness and verbal short-term memory in kindergarten. However, these measures did not predict their reading and spelling development one year later. Children who developed reading problems in the first year of literacy education could not be differentiated from children without reading problems on tasks of phonological processing measures in kindergarten. Vandewalle et al. (2010) suggest that the lack of an association between phonological processing skills – i.e. phoneme awareness and verbal short-term memory – might be the result of the relatively transparent orthography of Dutch. Only RAN predicted reading level in both the SLI group and in the control group. Children with SLI *and* reading problems showed poor performance on RAN in comparison to the control group.

A similar result was also reported by Bishop, McDonald, Bird and Hayiou-Thomas (2009). At the age of 9 years, RAN differentiated between groups of SLI with or without dyslexia. The SLI group without dyslexia performed similarly to the control group. Although all children with SLI performed poorly on measures of phonological processing at the age of 4, the children with SLI but no reading problems showed only mild phonological processing problems in later grades (see also Catts et al., 2005). Bishop et al. argue that although dyslexia and SLI show overlap in some deficits as phonological processing, a reading problem might only occur when there is a combination of deficits in phonological processing and rapid serial naming skill. Moreover, early deficits on phonological tasks of phoneme awareness or nonword repetition do not necessarily cause reading problems. The initial phonological processing deficits in some children with SLI will diminish as their phonological skills improve by learning to read because of the reciprocal relationship between reading and phonological skills.

On the other hand, Nithart et al. (2009) suggest that the phonological problems in SLI and dyslexia are of a different nature. They compared a group with SLI and reading problems with a group with reading problems but no history of language difficulties on a wide range of phonological measures. Although both groups were comparable on the reading measures, they showed different profiles of phonological deficits. The children with SLI showed problems on tasks of phonological discrimination and phonological short-term memory that were not found in dyslexia. Both groups showed problems on measures of phoneme awareness, that were more pronounced in SLI, which led Nithart et al. to conclude that problems in phoneme awareness give rise to reading problems and that SLI may be regarded as a disorder of phonological short-term memory as measured by nonword repetition (see also Scuccimarra et al., 2008).

These results show that phonological difficulties do not always result in reading problems and phonological problems of varying severity can be associated with reading problems of the same degree. Moreover, if children are not selected on the basis of reading problems, it has been shown that phonological problems can exist without causing reading problems (Højen-Tengesdal & Tønnesen, 2010). In contrast, Zourou, Ecalle, Magnan and Sanchez (2010), who conducted a study in French, showed that children with SLI improved their phoneme awareness skills to a normal level, although word decoding skills remained poor. Children with SLI appeared to be able to acquire phoneme awareness over time, but they were not able to use these skills in tasks as reading and spelling.

In this study, we further explored the commonalities and differences in reading skills and phonological processing in Dutch children with SLI and dyslexia. In comparison to the Vandewalle et al study, the children in our study were older and already had received some years of literacy education. It may be assumed that literacy problems become more pronounced in later years when task demands increase. However, the influence of phonological processing on reading skills might be limited because of the relatively transparent orthography of Dutch. Two questions were investigated. Do children with dyslexia or SLI exhibit comparable deficits in phonological processing? If so, are phonological processing deficits associated with reading problems in both disorders to the same extent?

We expected that dyslexic and SLI groups would perform poorly on phonological tasks in comparison to normal controls. Differences between the dyslexic and SLI groups might arise as a consequence of varying degrees of reading problems. The severity of the phonological deficit would be commensurate with the degree of reading deficit in both SLI and dyslexia. In particular, children with relatively better reading skills would perform better on skills of phoneme awareness, and rapid serial naming. However, we expected it also to be possible that the SLI group would show a more pronounced deficit on nonword repetition, in particular with increasing word length (see for review Gathercole, 2006). In addition, as it has been suggested that some SLI children do not develop dyslexia, we expected the SLI group to be more heterogeneous with regard to the reading ability.

Method

Participants

Two clinical groups participated in this study, one with the diagnosis of dyslexia and one with the diagnosis of specific language impairment. Children with dyslexia were selected to have a lag in reading accuracy of at least two years measured by a task of word reading fluency indicating poor reading level that is comparable to beginning readers (TMRT, see below for a description). They had no reported history of difficulties in non-phonological language skills (in particular receptive and/or productive semantics, syntax and morphology). Their nonverbal and verbal intelligence had to lie within the normal range, i.e. not more than 1 SD below the norm. The dyslexic subgroup (DYS) consisted of fourteen children from which seven children were recruited at regular primary schools and seven children were recruited at schools children with special educational needs.

SLI was defined as a severe language impairment that was not the direct result of global intellectual, sensory, motor, emotional or physical impairments. All children with SLI attended special schools for language-impaired children. In the Dutch educational system, children are admitted to these schools only in case of normal or low-average nonverbal intelligence, weak language scores on criterion-referenced tests (2 SD below the mean), no sensori-motor deficits and no psychiatric disorder. A psychologist or a speech-language pathologist refers the children to such services after extensive clinical and psychometric examination. Every two years, it is evaluated whether the criteria for the diagnosis of SLI are still met. At our request, specialists at two schools selected children on the basis of recent diagnostic data. Fifteen children were selected.

Table 1. Characteristics of subjects

		CONTR	DYS	SLI	ANOVA	
					F	η_p^2
	N	24	14	15		
	Gender	14b	9b	11b		
		10g	5g	4g		
CA	M	10;6	10;7	10;7	.14	.01
	SD	8.0	11.4	10.3		
NVIQ	M	16.29	14.21	13.07	3.69_b	.13
	SD	4.50	2.36	2.74		
VIQ	M	17.67	14.21	9.87	14.00_{bc}	.36
	SD	3.57	3.20	2.64		
TMRT	M	87.43	42.55	65.75	59.94_{abc}	.70
	SD	9.94	15.91	12.15		
	Range	69-105.7	17.3-64	47.7-81		

Note: Number of participants per group; gender (b = boys, g = girls); chronological age (CA) in years with standard deviations in months; mean standard scores (M) and standard deviations (SD) for Nonverbal IQ (NVIQ) with a standard mean of 15; standard scores for Verbal IQ (VIQ) with a standard mean of 15; Three Minute Reading test is a composite mean score of speeded word reading accuracy on three lists of different word complexity; including Standard Deviations (SD). ANOVA *F*- with degrees of freedom; significant post hoc (Tukey-Kramer) between-group differences are indicated by subscripts: $_a$ CONTR-DYS; $_b$ CONTR-SLI; $_c$ DYS-SLI.

The groups were compared to a control group. All groups were matched for age. We selected subjects to perform within the normal range on a test of nonverbal intelligence. We excluded children with problems other than reading or language problems. The control group consisted of twenty-four children. They were selected at regular primary schools. Their reading ability, nonverbal and verbal intelligence were within the normal range.

Specifications of the participants are summarized in table 1. Univariate analyses of variance (ANOVA) were used to examine group means. Groups did not differ in age ($F(2,50) = .14$, *n.s.*). There was a significant group difference in scores on the task of word reading fluency (TMRT) ($F(2,50) = 59.3$, $p < 0.05$, $\eta_p^2 = .70$). Tukey-Kramer post hoc test was used to assess which groups differed significantly from each other. The SLI group performed significantly better than the DYS group ($p<.001$), but less well than the control group ($p<.001$). Groups performed significantly different on the task of fluent pseudoword reading ($F(2,50) = 74.26$, $p < 0.001$, $\eta_p^2=.75$). Both DYS and SLI obtained significant lower scores in comparison to the CONTR ($p<.001$), but the SLI group outperformed the DYS group ($p<.001$). Groups performed significantly different on the nonverbal intelligence task ($F(2,50) = 4.02$, $p < 0.05$, $\eta_p^2=.14$). Although the scores of the SLI group were not more than 1 *SD* below the norm, the SLI group obtained significantly lower scores than the control group ($p<0.05$). Other group comparisons indicated no significant differences in nonverbal intelligence. We found a significant group effect on the verbal intelligence test ($F(2,50) = 30.2$, $p<0.001$, $\eta_p^2=.55$). The SLI subgroup performed significantly poor in comparison to the DYS subgroup ($p<0.001$) and the control subgroup ($p<0.001$).

Procedure

The first author administered the tests to all children individually. Each child was tested in four separate sessions of half an hour within a period of about one and a half month. Tests were presented in the same order to each child.

Assessment

Verbal and nonverbal intelligence measures. Figural exclusion test was used to measure nonverbal intelligence. This test is part of the Revised Amsterdam Child Intelligence Test (Bleichrodt, Drenth, Zaal & Resing, 1987). This task requires determining within 30 seconds which abstract picture does not fit to 3 other similar pictures. The score is the total number of correct answers. We converted the raw score to a standard.

A verbal analogies test was used to indicate verbal intelligence. This test is also part of the Revised Amsterdam Child Intelligence Test (Bleichrodt et al., 1987). We expected that the SLI group would perform less well since language abilities play a significant role in solving verbal analogies accurately (Masterson, Evans & Aloia, 1993). The test requires inductive reasoning in which discovery of semantic relations is involved. The task involves presentation of pictured objects expressing the relation "A is to B as C is to D" where the D item has to be chosen from four alternatives. The test consists of 30 items but administration stops after 4 successive mistakes. The score is the total number of correct answers.

Reading measures. The Three Minutes Reading Test was used to measure fluency of word reading (TMRT; Verhoeven, 1995). The TMRT is a standardized test, which consists of three lists of words of increasing difficulty. The test requires children to read the words as accurate and fast as possible. Per list, the score is the number of accurately read words after one minute. We computed the average score for the three lists per child. In addition, we converted the score into age related norm scores to determine the delay in reading.

Fluency of pseudoword reading was measured by a standardized test (Klepel; van den Bos, Lutje-Spelberg, Scheepstra en de Vries, 1994) to indicate fluency in reading of pseudowords. The test requires to read the words correctly and as quickly as possible. The test consists of 116 pseudowords of increasing difficulty in both length and complexity. The score was the number of words that were correctly read in two minutes.

Reading related measures: dependent variables. Phoneme Awareness was measured by a phoneme deletion test (de Jong & van der Leij, 2003). The test consists of three parts of nine pseudowords each. At the first and second part the child has to delete one consonant from one-syllable words (part I: /skoom/ without /s/) and two-syllable words (part II: /memslos/ without /s/). The third part requires leaving out twice one consonant from two-syllable pseudowords (/f/ from /fiembamf/). Pseudowords are presented twice on an audiotape. Two items for practise precedes the first and third part of the test. The final score is the number of correct responses.

The Rapid Serial Naming Task was used to measure retrieval speed of symbol information. The child has to name as rapidly as possible a visual array of stimuli presented on a chart. Four different charts with stimuli of a given category randomly repeated were used: numbers (1, 3, 5, 6, 8), letter names (A, D, O, P, S), objects with phonologically dissimilar names (/mes/, /oog/, /boek/, /deur/, /jas/ i.e. knife, eye, book, door and jacket) and objects with phonologically confusable names (/broer/, /broek/, /bloed/, /bloem/ i.e. brother, trousers, blood, flower) (Messbauer & de Jong, 2001). Confusability of items puts extra load on phonological processing. Sample trials ensure that the individual stimuli in each category are known. The child has to name the stimuli from left to right starting at the top of the chart as fast as possible, but without making any mistakes and without skipping any. A stopwatch is used to record the time taken for each chart. We computed the mean number of seconds per picture per chart. The correlations between tasks were moderate to high ranging from .61 to .82 and therefore we decided to compute a composite score by averaging across naming tasks.

A Nonword Repetition test (de Jong & van der Leij, 1999; see also Gathercole & Baddeley 1990) was administered to test phonological short-term memory. The test requires the repetition of pseudowords, varying from one to four syllables. Each word is presented once on audiotape after a short beep. The final score is the number correctly repeated words with a maximum score of 48.

A Word Span task was designed to measure verbal memory span (e.g. de Jong, 1998). Children listen to a sequence of high frequency CVC adjectives which they repeat in the same order of presentation. The number of words per sequence increased from three to eight words. A sequence of equal length was presented twice with a total of 12 items. The words were presented on audiotape with a 1 second interval between words. Two items for practise preceded the test items. The administration stopped when a child fails on two lists of equal length. The final score was the number correctly repeated sequences.

Results

Differences between children with dyslexia or SLI and normal controls

Means and standard deviations for all variables are displayed in table 2. We conducted multivariate analysis of variance with group (DYS, SLI and CONTR) as the between subjects factor. Bartlett's test of sphericity justified multivariate analysis of variance. Assumptions of equal covariances and equal variances across groups were met. Multivariate analysis revealed that groups differed overall on the set of seven variables (Wilks's λ =.11, $F(10,92)=19.17$, $p<.001$, $\eta_p^2=.68$). We examined the univariate F-values to determine on which variables groups performed significantly different. To control for inflated Type I error because of multiple testing, we applied a Bonferroni-Holm step down procedure to adjust alpha levels while maintaining maximal power for each comparison (Holm, 1979; Aickin & Gensler, 1996). The univariate F-values and effect sizes are also presented in table 2. The results of the univariate analyses revealed significant difference between-group differences on all measures. We pursued to discover which group means differed by employing Tukey-Kramer *post hoc* pair wise comparisons. Significant differences are indicated by subscripts in table 2 and described below.

Post hoc analysis on phoneme awareness revealed a significant difference between CONTR and DYS ($p<.001$) and between CONTR and SLI ($p<.001$). There was no significant difference between the DYS and the SLI group. The DYS and SLI group needed significantly more time on the rapid serial naming task in comparison to the CONTR (p<.01 for both comparisons), but there was no difference between SLI and DYS. On nonword repetition, the DYS and SLI subgroup differed significantly from the CONTR group (resp. p<.01 and p<.001), but the DYS subgroup obtained higher scores than the SLI subgroup (p<.001). We found the same results on word span. The DYS group differed from the CONTR (p<.01) and outperformed the SLI group (p<.01). Evidently the difference between the SLI and CONTR subgroup was also significant (p<.001).

Table 2. Overview of the performance of three subgroups

	CONTR		DYS		SLI		ANOVA	
N	24		14		15			
	M	SD	M	SD	M	SD	$F_{(2,50)}$	η_p^2
Tasks								
phonological awareness	18.33	3.50	9.21	4.12	9.00	3.68	40.45_{ab}	.62
serial rapid naming	.66	.10	.82	.14	.81	.14	10.83_{ab}	.30
nonword repetition	41.96	3.59	37.43	3.25	30.33	5.42	36.88_{abc}	.60
word span	10.33	1.97	7.64	2.10	4.93	1.87	34.92_{abc}	.58
articulation rate	5.90	.49	5.10	.46	4.75	.50	28.99_{ab}	.54

Note. Mean overall scores on each task (M) and standard deviation (SD; mean number of pictures per second on the serial rapid naming task, mean number of syllables per second on the articulation task. ANOVA F- with degrees of freedom; significant post hoc (Tukey-Kramer) between-group differences are indicated by subscripts: $_a$ CONTR-DYS; $_b$ CONTR-SLI; $_c$ DYS-SLI. Partial eta's are reported in the last column as measure of effect size.

Association between phonological processing skills and fluent word reading

Next, we considered how performance on the various aspects of phonological processing relates to reading achievement, in particular to what extent phonological deficits co-occur with similar levels of reading achievement in both disorders. We conducted hierarchical regression analyses to determine the unique variance contributed to phonological processing tasks by fluency of word reading and the presence of specific language impairment. Presence of SLI was entered as a dummy variable (1 = SLI, 0 = no SLI). Raw scores were converted to standardized scores. Data were screened on outliers. Outliers on the dependents were defined as cases with a standardized residual greater than 3.3. Mahalanobis

Distance was used to determine outliers on the set of predictors. Furthermore, we examined the influence of data points in affecting the regression equation. A Cook's distance >1 was considered large. No cases were deemed to be (influential) outliers.

Pearson correlations among predictors and between predictors and the dependent variables are reported in table 3. Eta reflects the association between the nominal dummy variable and other variables. The interpretation of the dummy variable 'SLI vs. no SLI' is relative to the DYS group and the CONTR group. Contrary to Pearson's r, eta has no direction. The dummy shows an association with nonverbal intelligence which is not surprising considering the reported lower group mean. The predictors fluency of word reading and the dummy 'SLI vs. no SLI ' were not correlated (η =.11, p = .45). The dummy was associated with all dependent variables except with fluency of pseudoword reading. Fluency of word reading was significantly correlated with all tasks tapping phonological processing. The correlations were moderate varying from .43 to .67. The negative correlation with rapid serial naming indicates that children who obtain high reading scores name pictures more rapidly.

Table 3. Correlations between predictors and dependent variables

Variable	1	2	3	4[a]	5	6	7	8
1 Age (months)	—							
2 Nonverbal IQ	-.09	—						
3 Fluent Word Reading	.12	.23	—					
4 SLI vs. No SLI	.05	.30*	.11	—				
5 Phon. Awareness	.14	.36**	.67**	.46**	—			
6 Serial Rapid Naming	-.30*	-.21	-.62**	.28*	-.60**	—		
7 Nonword Repetition	.11	.39**	.43**	.72**	.45**	.56**	—	
8 Word Span	.17	.51**	.44**	.67**	.46**	.59**	.76**	—
9 Pseudoword reading	.04	.27*	.91**	.16	.68**	-.63**	.46*	.46*

Results on the hierarchical regression analyses are reported in table 4. After controlling for age and nonverbal intelligence in the first step, we entered fluency of word reading and the dummy variable. In separate analyses, the predictors were subsequently entered at the second and third step to determine the relative contribution of both factors. If the dummy variable appeared to contribute unique variance in the equation after controlling for reading performance, this would indicate that children with SLI performed poorly on the variable irrespective of their word reading skills. On the other hand, entering the dummy variable at the first step, we could evaluate whether reading scores still explained variance after the difference between children with and without SLI had been partialled out. The dummy 'SLI vs. no SLI' accounted for unique variance of most variables after partialling out the contribution of fluency of word reading (model 1). Children with SLI irrespective of their word reading skills performed poorly on tasks tapping phonological processing. In addition, fluency of word reading accounted for a significant amount of variation in all criterion variables over and above the diagnosis of SLI (model 2). The degree of phonological difficulties was related to reading level across the range of phonological skills.

Table 4. Results hierarchical regression analysis on standardised scores

Model		Phonological recoding	Phoneme Awareness	Serial Rapid Naming	Nonword Repetition	Wordspan	Articulation Speed
Variables		ΔR^2					
1	age in months PIQ (figural exclusion)	.074	.158*	.147*	.173**	.306***	.131*
2	Reading score	.765***	.345***	.356***	.112**	.092**	.250***
3	SLI vs. no SLI	.002	.123***	.046*	.381***	.286***	.219***
2	SLI vs. no SLI	.007	.142**	.058*	.398***	.300***	.239***
3	Reading score	.760***	.327***	.345***	.094**	.078**	.229***
	N	53	53	53	53	53	53

Notes: **p*<.05, ***p*<.01, ****p*<.001

Fluency of word reading and diagnosis of SLI did not account for equal amounts of variation in each variable. In comparison to the dummy variable, word reading explained most of the variance in phoneme awareness (32.7% vs. 12.3%) and rapid serial naming (34.5% vs. 4.6%). These findings support our expectations that phonological awareness and rapid serial naming are the strongest indicators of the phonological core deficit of dyslexia. In comparison to word reading, the dummy variable explained most of the variance in nonword repetition (38,1% vs. 9,4%) and word span (28,6% vs. 7,8%). Although poor performance was partly explained by reading level, poor performance was more strongly associated with SLI. The SLI subgroup exhibited more severe memory deficits irrespective of reading level.

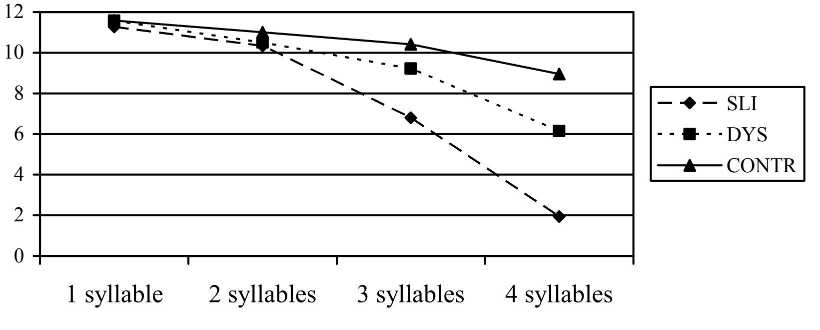
Differences within the SLI group related to fluent word reading skill

The performance on the phonological processing tasks of the SLI group was further examined taking into account the heterogeneity with regard to fluency of word reading. A cluster analysis was run to classify the children into groups of normal and disabled readers according to reading score. The reading scores of eight children from the SLI group resembled the (lower) reading scores from the control group (SLI-NR). The reading scores of the other seven children of the SLI group were comparable to the scores from the DYS group (SLI-RD). Differences between means on phonological tasks between groups were further investigated using two-tailed t-tests for independent samples. The difference between the SLI subgroups on the selection variable fluent word reading was confirmed (t=6.938, df=13, p<0.001, d=1.91). SLI-NR outperformed SLI-RD on fluency of pseudoword reading (t=3.065, df 13, p<0.01, d=1.58). The two subgroups, however, did not differ on any of the other measures. Our expectation that the subgroup with only SLI might outperform the subgroup with SLI and dyslexia (SLI-RD) on the variables that belong to the core deficit of dyslexia was only supported by a difference phonological decoding, but not in phoneme awareness and rapid serial naming.

Additional considerations; memory processes in SLI

MANOVA and hierarchical regression results revealed more severe deficits based on mean scores in the SLI group on tasks in which (phonological) memory processes were involved. If limited memory processes determined difficulties we would expect to find increasing difficulties with an increase of word length instead of poor performance irrespective of word length. Figure 1 shows a relative large decrease in performance for the SLI subgroup on words consisting of 3 and 4 syllables. This effect was tested by a MANOVA for repeated measures with Group as between subjects factor and Length (1, 2, 3 and 4 syllables) as within subject factor. The interaction effect between group and Length was significant ($F(6,150) = 19.80$, $p<0.001$). We specified contrasts, which showed that groups only differed on repeating pseudowords of 3 or 4 syllables. The SLI group performed worse than DYS and CONTR on repeating nonwords with three syllables ($p<0.01$) and four syllables ($p<0.001$). DYS only performed worse than CONTR on repeating pseudowords with four syllables ($p<0.05$).

Figure 1. Mean Performance on the Nonword Repetition Test per syllable length/group



Discussion

Overall, the results indicate that the group with dyslexia and the SLI group performed poorer than the typical group on all variables tapping phonological processing. Most notably, the atypical groups exhibited deficits in phoneme awareness and serial rapid naming to the same extent. However, the SLI group performed worse on tasks in which phonological short-term memory was involved, i.e. on tasks of nonword repetition and wordspan. The group with dyslexia displayed a more severe deficit in fluency of pseudoword reading (phonological decoding) than the SLI group which is in agreement with the different average scores on the fluency of word reading task used for selection.

Hierarchical regression analysis showed that the gradient differences in performance on phonological awareness and serial rapid naming were most strongly associated with differences in reading skills, which confirms the strong associations between these phonological abilities and reading achievement reported by many others. However, poor performance of the SLI group was not fully accounted for by reading level. For rapid naming, only a small portion was not explained by reading in the group with SLI (4.6%). As we found that the rapid naming deficit is mainly associated with reading level in SLI, our results confirm that the rapid naming deficit in SLI is similar to that found in dyslexia. Lahey and Edwards (1999) found that children with receptive language deficits made more semantic-associated errors in a rapid naming task and they interpreted this finding as evidence of poorly differentiated and organized semantic-lexical representations. Although in this study the role of semantics was minimized by task presentation (the items were presented and practised before testing), poor semantic-lexical representations might be an explanation of the difficulties in SLI irrespective of reading level.

In phoneme awareness, reading achievement and the SLI factor explained 32.7% and 12.3% of unique variance, respectively. Reading shares the strongest association with phoneme awareness, but all children in the group with SLI performed poorly after controlling for reading achievement. The poor and better readers within the SLI group showed no difference in performance. The finding of poor phoneme awareness independent of reading level replicates the findings of others. Nauclér and Magnusson (2000) reported that a group with language impairment managed to learn to decode words at the same level as their normal controls, despite their poorly developed phonological awareness at pre-school age and the first years of education. It is clear from our findings that the group with SLI performed significantly worse than the group with dyslexia and the control group on tasks in which verbal short-term memory and phonological memory was involved, i.e. on tasks of Nonword Repetition and Wordspan. Reading level accounted for 9.4% and 7.8% variance in performance on nonword repetition and wordspan, whereas the SLI factor accounted for 38.1% and 28.6%, respectively. These results support findings of other studies. Children with SLI perform poorly on memory tasks involving word and nonword repetition (Marton & Schwartz, 2003; Rispens & Parigger, 2010). Consistent finding of a severe nonword repetition deficit in SLI has led to the suggestion of its use as behavioural marker of SLI (Conti-Ramsden & Hesketh, 2003). The deficit becomes strikingly clear with increasing word length (Gathercole & Baddeley, 1990; Archibald & Gathercole, 2006; Marton & Schwartz, 2003). Same results were obtained in this study. Performance of the SLI group decreased relatively more with increasing word length in nonword repetition, indicating a limitation in phonological storage capacity (Gathercole, 2006).

Although the group with dyslexia outperformed the group with SLI, they also were poor at verbal memory tasks, word span and nonword repetition in comparison to the control group. Low performance on simple span tasks and nonword repetition reflect that children with dyslexia suffer from limitations in phonological storage capacity although these differences seem less severe in comparison to SLI. Other studies also report differences between the disorders with respect to severity in phonological storage capacity. Rispens (2004) found that children with dyslexia performed poorly on nonword repetition, but in comparison to children with SLI, the deficit appeared less severe. Goulandris, Snowling and Walker (2000) report a similar result. The present study supports the view that nonword repetition shares a stronger association with language than with reading ability because children with SLI are more affected than children with dyslexia (Gathercole, 2006). The same pattern was found for verbal memory span. Dyslexia was associated with poor word span, but children with dyslexia outperformed children with SLI. Poor memory span has been reported by some, contrary to others. Rispens (2004) found that the group with dyslexia did not perform poorly on a digit span task in comparison to a control group, whereas the group with SLI did. Van der Sluis, de Jong and van der Leij (2005) also did not find problems on a simple digit span task for a group with dyslexia. One explanation for these contradicting results might be that the digit and word span task differ in amount of support from long-term memory. Word-specific characteristics as item familiarity, frequency and imageability influence performance on span tasks. Span for digits, comprising a set of very familiar and highly discriminable short words, can be normal in spite of large difficulties with the recall of lists of slightly less familiar words (Hulme & Roodenrys, 1995).

Although reading achievement appears most strongly associated with phonological core deficits, phoneme awareness and serial rapid naming, the children with SLI showed (mild) problems on these phonological processing skills irrespective of word reading level. On measures of verbal short-term memory and phonological memory, children with SLI show severe problems, which are barely accounted for by reading level. Our results seem at odd with results reported by

Catts et al. (2005) who found that a well-reading SLI group performed better than poor-reading SLI group on tasks of phoneme awareness and nonword repetition. The subgroup of well-reading SLI displayed, however, mild phonological processing difficulties. They suggest that the underlying deficits of dyslexia and SLI are likely to involve continuously distributed abilities and the co-morbidity of the disorders should spread its effects to the borderline of each disorder. Our results also suggest that performance on phoneme awareness is most strongly associated with reading achievement in both disorders. However, the same reasoning does not apply to our findings on nonword repetition, which was not as strongly associated with reading as with SLI. It might have been that the sample in our study differs from the sample by Catts et al. (2005) that was selected at preschool. Some children showing language difficulties at preschool age grow over their problems. The group with persistent language problems might reflect

It appears possible that children with the same cognitive impairments present with different profiles of reading. It may be assumed that the developmental outcome depends on the other cognitive resources available to the child. Deficits in phonological decoding, or in the underlying ablity to map graphemes to phonemes at a sublexical level, obviously are complementary to deficits in phoneme awareness and rapid serial naming in explaining dyslexia. It may be argued that deficits in phonological decoding are a distinctive condition for developmental dyslexia. because the children with SLI but without dyslexia of the present study outperformed the group with SLI and dyslexia in phonological decoding, but not in phoneme awareness and rapid serial naming (Table 2). Difficulties with nonword reading have been reported by other researchers (see for a meta-analysis Hermann, Matyas, & Pratt, 2006). The connection between the development of phonological decoding and of orthographic skills (the 'self-teaching mechanism'; Share 1995) may play a role. As a piece of indirect evidence, poor phonological awareness seems to have less impact on reading skills in a child with high orthographic skills than in one with poor orthographic skills. Sparks (2001) reports cases of hyperlexia who exhibit poor phonological processing skills, but who appear to compensate these difficulties by exceptional orthographic skills. The developmental relation between phoneme awareness, phonological decoding, orthographic processing, and reading disabilities deserves further study.

The present study has some limitations. The sample size of the group of children with SLI was small because we were not able to select more within a reasonable amount of time and distances. Our wish to exclude cases with obvious co-morbidity with other disorders than dyslexia (e.g. autism; ADHD; mental retardation) added to the problem. We are, however, quite convinced that the sample may be called representative for relatively 'pure' SLI with or without dyslexia. The group of children with dyslexia was easier to find because of a higher prevalence. Nevertheless, one should be cautious not to over-interpret the findings of the present study. Next, although tasks were included with appropriate psychometric validity to be used with children at this age (for example, the phoneme deletion test was certainly not too easy), it would have been better to include some kind of orthographic processing task to explore the possible relation with differences in word reading within the SLI group. Furthermore, because the different processes and skills do not act on their own but rather interact in development (Bishop & Snowling, 2004), variables that bootstrap reading skills in SLI in face of poor phonological skills should longitudinally be studied.

In sum, this study reveals that both dyslexia and SLI share deficits in phonological processing. Deficits in phonological awareness, rapid naming and phonological decoding are most strongly related to reading level in both disorders and can therefore be considered best candidates for the phonological core. Deficits in phoneme awareness and rapid naming, however, cannot fully be accounted for by reading level in SLI. Children with SLI show additional (mild) problems on both tasks irrespective of reading level. Furthermore, our study supports findings of other studies that SLI is characterized by severe verbal short-term memory capacity limitations that seem for most part independent of reading achievement.

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CHAPTER 3

PREDICTORS OF RESPONSE TO INTERVENTION OF WORD READING FLUENCY IN DUTCH[1]

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Abstract

The objective of this study was to investigate the contribution of rapid digit naming, phonological memory, letter sound naming and orthographic knowledge to the prediction of responsiveness to a school-based, individual intervention of word reading fluency problems of 122 Dutch second and third graders whose reading scores were below the 10^{th} percentile in comparison with the normative group. Degree of responsiveness was determined by comparison of a pre- and posttest measure of word reading fluency with a 6 month interval. At posttest, 38% of the children had improved their reading scores above the 10^{th} percentile. Maintenance scores revealed no significant growth on average, confirming that word reading fluency skills of poor readers are hard to remediate. Except rapid digit naming, none of the measures predicted responsiveness after controlling for the autoregressive effect of initial performance on fluency of word reading. A large part of the variance remained unexplained, supporting the advantage of a response to intervention approach above traditional psychometric testing to identify severe reading disabilities.

Background

Intensive reading intervention can support children with reading problems to improve their rapid, context-free word reading skills (Torgesen, 2000). However, children receiving special instruction show large differences in responsiveness to evidence-based reading interventions (Denton, Fletcher, Anthony & Francis, 2006; O'Shaugnessy & Swanson, 2000; Torgesen et al., 1999; Torgesen, Alexander, Wagner, Rashotte, Voeller & Conway, 2001; Vellutino, Scanlon & Tanzman, 1998; Vellutino, Scanlon, & Lyon, 2000). The preventive model of response to instruction (RTI) takes into account the degree to which a student at risk for reading difficulties benefits from a validated intervention and reduces his/her reading delay and risk status in comparison with the normative group (e.g. Linan-Thompson, Vaughn, Prater & Cirino, 2006). Generally, a RTI approach involves close monitoring of student progress, implementation of group or individual interventions and referral to special educational services only if a child does not respond to intervention (Fuchs, Mock, Morgan & Young, 2003; Fuchs & Fuchs, 2006). A response to intervention approach is considered as a method to prevent, help and identify children with dyslexia or severe reading disabilities (Burns, Appleton & Stehouwer, 2005; Fletcher, Coulter, Reschly & Vaughn, 2004; Fuchs, Compton, Fuchs, Bryant & Davis, 2007; Vaughn & Fuchs, 2003; Vaughn, Linan-Thompson & Hickman, 2003; Vellutino et al., 2000). Instead of a 'wait-to-fail' procedure, children receive intervention if they are at risk of reading difficulties. Those children who make minimal progress or do not meet benchmarks after intervention – so called nonresponders (McMaster, Fuchs, Fuchs, & Compton, 2005) or hard-to-remediate (Torgesen, 2000) – are identified as most likely learning disabled or reading disabled and may need more intensive, long-term intervention.

In contrast to methods that focus on individual skills at one time point to identify children with learning disabilities, the RTI approach takes progress in learning in response to adapted instruction into consideration (Case, Speece and Molloy, 2003), reducing the role of environmental aspects. This suits an important aspect of current definitions of dyslexia, as for example expressed by the British Dyslexia Association (BDA; www.bdadyslexia.org.uk) and the Stichting Dyslexie Nederland [Dutch Dyslexia Foundation] (SDN; www.stichtingdyslexienederland.nl), which have incorporated the concepts of both *specificity* and *resistance* to conventional teaching methods. Besides testing to specify the cognitive deficits underlying the dyslexia such as difficulties in phonological and orthographic processing, poor response to evidence-based intervention indicating resistance is a necessary criterion for diagnosis of dyslexia.

The major objective of this study was to investigate the contribution of reading related subskills to the prediction of responsiveness to a school-based, individual intervention of word reading fluency problems of Dutch second and third graders. For the current study, we adapted an intervention from a prior study (Struiksma, van der Leij, & Stoel, 2009) for large-scaled implementation at schools delivered by remedial teachers. Struiksma et al. investigated the intervention for effectiveness, but the study did not investigate the relation between response to intervention and learner characteristics at pretest that have been associated with reading achievement. We first examined the degree of variability in response to intervention. The second aim was to assess whether and to what extent reading related subskills contribute to differential responsiveness to the intervention. The answers to these questions can help to improve the early identification of children who are likely to develop reading disabilities and give insights into mechanisms that underlie the reading disabilities of the severely hampered group.

Response to Intervention and learner characteristics

Two recent reviews summarize learner characteristics that are associated with responsiveness (Al Otaiba & Fuchs, 2002; Nelson, Benner & Gonzalez, 2003). Based on 23 studies, Al Otaiba and Fuchs (2002) report seven factors that showed a relation with response to intervention: phonological awareness, verbal or phonological memory, rapid naming, intelligence, attention, orthographic skills and demographics. Including seven additional studies, a meta-analysis by Nelson et al. (2003) determined the magnitude and relative contribution of these learner characteristics to RTI. With the exception of demographics, the findings of Nelson et al. (2003) were consistent with the learner characteristics identified by Al Otaiba and Fuchs (2002).

In this study, we focused on reading related subskills, in particular phonological and orthographic processing skills. General cognitive factors such as attention and intelligence were not taken into account. Although attention contributed to RTI in some studies (e.g. Dally, 2006), its role is limited in computer-based interventions with short training sessions and immediate feedback from performance (Clarfield & Stoner, 2005; Hintikka, Aro & Lyytinen, 2005). Hintikka et al. (2005) found that children with poor attention showed larger benefits from computer-based intervention in comparison to regular teacher-directed instruction. Intelligence was found to influence RTI in some studies (Berninger, Abbott, Zook, Ogier, Lemos-Britton & Brooksher, 1999) but its role in fluency of word reading has been disputed (Hatcher & Hulme, 1999; Stanovich & Siegel, 1994). Ricketts, Nation and Bishop (2007) showed in a group of 9 year olds that vocabulary knowledge accounted for unique variance in reading comprehension and exception word reading but not in text reading accuracy, regular word reading and nonword reading. It should be noted that in the language under study – Dutch, with a relatively transparent orthography (Seymour et al., 2003) – exception words are practically non-existing. The influence of (verbal) intelligence and vocabulary appears to be limited to reading comprehension (Hatcher & Hulme, 1999; Ricketts, Nation and Bishop, 2007), which is not within the scope of this study.

The critical role of phonological processing skills in reading development is generally acknowledged (e.g, Snowling, 2000). Deficiencies in three kinds of phonological processing abilities are most frequently reported in the literature on reading problems: 1) phonological awareness, 2) phonological memory, and 3) rapid naming. These abilities have all been found to contribute to the responsiveness to instruction. Unlike phonological awareness, however, phonological memory and rapid naming were included in a relatively small number of studies (Al Otaiba et al., 2002).

Phonological memory is suggested to be a relatively pure measure of phonological processing, in particular if measured by nonword repetition (Bishop & Snowling, 2004). Nonword repetition tasks require the temporal storage of phonological sequences without explicit knowledge of the phonemes. Since nonwords are used, support from long-term memory is thought to be limited. Moreover, the influence of reading achievement on nonword repetition tests is thought to be less strong than it is on phonological awareness. Especially in transparent orthographies, phonological awareness has a strong reciprocal relation with reading achievement but its predictive role is limited to the first grades of reading education (de Jong & van der Leij, 1999; 2002; Wimmer, 1996; Wimmer, Mayringer & Landerl, 2000), unless the task of phonological awareness is sufficiently difficult (Patel, Snowling & de Jong, 2004; de Jong & van der Leij, 2003). This can be explained since in a transparent orthography, the letter-sound correspondence is relatively predictable and apparent. Most sounds are represented by one letter or grapheme and the other way around. As children learn to read, their knowledge of letters within words increases and this helps children to increase awareness of the separate sounds within words.

The involvement of phonological memory in reading achievement has several explanations. Phonological memory plays a role in the acquisition of word- and sound-specific knowledge; i.e., it is involved in learning to associate letters with their sounds and names and in the construction of novel well-specified and detailed phonological representations (de Jong & Olson, 2004; Mauer & Kamhi, 1996). In addition, phonological memory is involved in phonological recoding of words, by holding the letter sounds in memory before the sounds can be blended into words. According to the self-teaching hypothesis of Share (1995), phonological recoding is crucial for the acquisition of orthographic knowledge. Therefore, phonological memory might be involved in learning to read through the process of phonological recoding. Measures of phonological memory, among which nonword repetition tests, have been found to contribute to the prediction of RTI (Berninger et al., 1999; Uhry & Sheperd, 1997; Vellutino, et al., 1996; 2000). However, this finding was not always replicated (Hatcher & Hulme, 1999; O'Shaugnessy & Swanson, 2000).

As a subskill of reading, serial rapid naming appears to be relatively independent from phonological awareness and phonological memory (de Jong and van der Leij, 1999) and a strong predictor of reading difficulties (de Jong & van der Leij, 2003). Serial rapid naming tasks assess the ability to name series of common items as rapidly as possible. An explanation for the predictive role of serial naming speed on fluency of word reading measures is that naming a limited set of symbols, letters or digits rapidly reflects the ability to retrieve the letter sounds or whole word sounds while reading (Wolf and Bowers, 1999; van den Bos, Zijlstra, & lutje Spelberg, 2002). Furthermore, rapid naming skill is suggested to affect the ability to construct orthographic representations of letter names, sublexical letter units and words (Bowers & Wolf, 1993; Bowers, Golden, Kennedy & Young, 1994; de Jong & Vrielink, 2004). Bowey (2005) suggests that rapid naming skills can be best interpreted to reflect the degree of over-learning of letter and/or digit names and the efficiency of phonological processing. Although it was only included in a limited number of studies (Al Otaiba & Fuchs, 2002), rapid naming was identified as a stronger predictor of responsiveness than phonological awareness or phonological memory in the meta-analysis conducted by Nelson et al. (2003). Studies demonstrated that children with poor versus good response to intervention could be differentiated by rapid naming skills prior to intervention (e.g. Berninger et al., 1999; Vaughn, Linan-Thomson & Hickman, 2003).

Besides phonological processing skills, the understanding of how sounds map systematically onto letters is essential for the acquisition of accurate word decoding skills (Ehri, 2005). Letter knowledge involves both knowledge of phonemes and the recognition of the visual features of letters. Knowledge of letter names and sounds at a preliterate stage appear both powerful predictors of reading acquisition in different languages (van der Leij & de Jong, 1999; 2002; McBride-Chang, 1999; Näslund & Schneider, 1996). In comparison to letter name knowledge, letter sound knowledge becomes important at a later stage (McBride-Chang, 1999; Foulin, 2005) and takes longer to reach optimal performance (Ritchey & Speece, 2006). In particular, the *fluency* of naming letter sounds, appears associated with reading skills (Badian, 1998; Ritchey and Speece, 2004, 2006). Ritchey and Speece (2006) argue that fluent recognition of letter-sound relationships may serve as a mechanism to support phonological recoding, blending and accurate word identification, which is prerequisite for self teaching resulting in building knowledge of orthographic representations (Share, 1995). Thus, knowledge of letters or graphemes (including digraphs) involves orthographic knowledge at the sub-lexical level which is necessary to acquire orthographic knowledge at the lexical level.

Sensitivity to regularities of letter sequences might be helpful for children to learn to read fluently. Instead of relying on a grapheme-by-grapheme recoding, recognition of recurring sublexical letter patterns speeds up the word recognition process. Orthographic knowledge at the lexical level enables reading by sight, i.e., the recognition of words without decoding them. Already after a short period of reading education, children become gradually sensitive to regularities in letter patterns (Cassar & Treiman, 1997). This knowledge of orthographic representations has been found to be related to reading speed independent of phonological skills (Cunningham, Perry & Stanovich, 2001; Hagiliassis, Pratt, & Johnston., 2006; Olson, Forsberg & Wise, 1994), already during early reading development (Booth, Perfetti & MacWhinney, 1999).

Al Otaiba and Fuchs (2002) reported in their review that a limited number of studies included measures of orthographic skills. Orthographic skills encompassed both letter knowledge and knowledge of spelling patterns, i.e., orthographic knowledge. Only 3 out of 7 studies found evidence for a predictive role of orthographic skills. Whereas Vellutino et al. (2000) found that phonological awareness, rapid naming and phonological memory reliably differentiated between the most poorly and most readily remediated poor readers, measures of orthographic skills did not. In contrast, Berninger et al. (1999) found that differences in orthographic skills differentiated children with good and poor intervention outcome. Also Stage et al. (2003) using a composite of letter retrieval and orthographic knowledge concluded that orthographic skills predicted growth in word reading skill after intervention

Present Study

The majority of studies that were aimed at the identification of learner characteristics related to degree of responsiveness have been conducted in the English language domain, which has an opaque orthography. Studies focused on responsiveness to intervention in a language with more transparent orthographies are scarce. As an exception, the effect of RTI in Spanish was studied by Linan-Thompson and colleagues (e.g., Linan-Thompson, Cirino, & Vaughn, 2007; Vaughn, Mathes, Linan-Thompson, & Francis, 2005), and the earlier Dutch study of Struiksma et al. (in press) focused on learning mechanisms during intervention, but both studies did not include pretest learner characteristics to predict responsiveness. Instead, the present study investigates the predictive value of reading subskills in Dutch. In the present study, we aimed for examining learning characteristics of *second and third graders* that could predict responsiveness defined as *fluency of word reading.* The choice for grade level was made because after first grade, norm scores of reading become more stable (Spira, Bracken & Fischel, 2005) and children in need of reading intervention can be more readily identified. The focus was on word reading fluency since it has been suggested that poor reading fluency, in particular with context-free words, characterizes best the reading difficulties of children with severe dyslexia (Lovett et al., 1990; Torgesen, 2000), which also applies to transparent orthographies with mostly unambiguous relationships between graphemes and phonemes. Poor readers obtain high levels of accuracy after only a few months of education, but remain reading in slow rate (Dutch: Yap & van der Leij, 1993; Italian: Zoccolotti, De Luca, Di Pace, Judica, Orlandi & Spinelli, 1999; Finnish: Holopainen, Ahonen, Lyttinen, 2001; German: Landerl, Wimmer, & Frith, 1997; Landerl & Wimmer, 2008). Moreover, fluent word recognition skill is an important correlate of text reading fluency (Torgesen, Rashotte & Alexander, 2001). Problems with fluent word recognition likely affect fluency in text reading and reading comprehension (e.g. Perfetti, 1992).

To predict responsiveness to intervention we included measures of phonological skills, phonological memory and serial rapid naming. In addition a measure of letter sound knowledge was included, since letter sound knowledge has been found to be a strong predictor of word reading development. Furthermore, as a measure of orthographic knowledge, an orthographic choice task was administered. In a study with English speaking children in grade 3 to 5, Hagiliassis et al. (2006) showed that orthographic choice tasks tap skills that are independent from phonological skills. In a factor analyses, orthographic choice tasks loaded highly on an orthographic processing factor without cross-loads on the phonological factor. They argued that orthographic choice tasks measure skills that are independent from phonological skills because the recognition of the correct spelling cannot be made on the basis of a sound-based strategy when homophones are used. We examined the contribution of these reading-related skills to the degree of responsiveness in hierarchical regression analyses after controlling for the autoregressive effects of reading at the earlier point in time as recommended by Bowey (2005). When reading effects at pretest are ignored, the contribution of other variables that correlate with reading at pretest may be overestimated.

Research design

We first explored the proportion of second and third graders showing good versus poor response to an intensive, well-controlled, research-based intervention of 20 weeks in a pretest-, posttest design and whether reading status remained stable at retest 5 months after intervention was finished. Next, we questioned whether the degree of RTI could be predicted by pretest measures of rapid digit naming, phonological memory, letter sound naming and orthographic knowledge. Results will be reported in separate sections.

Method

Subjects

The subjects came from ten regular elementary schools. Criteria for school selection included the use of good instructional reading methods (see Blok, Otter, Overmaat, de Glopper & Hoeksma, 2003) and the use of a system for monitoring reading development. Moreover, distribution of reading scores from the school population did not deviate from the normative distribution, i.e., there was a maximum of 10 percent of children with reading difficulties. Three successive years, teachers at school nominated children for intervention. Teachers based their nomination on scores of a word reading fluency test. The successive years will be referred to as separate cohorts 1, 2 and 3. Children were only selected if they obtained a score on word reading fluency at or below the 10th percentile based on norm scores of grade 2 or grade 3. This benchmark was chosen because of its practical value in the Dutch educational system. At the majority of schools in the Netherlands, gains in reading achievement are monitored using standardized tests at fixed moments from the start of grade 1 when formal reading instruction starts. At each time point, teachers and special educators evaluate the learning progress in comparison to the norm. The benchmark of 10^{th} percentile is nation-wide used to indicate whether children need additional and more intensive instruction outside the classroom. For children in the beginning of grade 2 with an educational age of 1.2 years, this benchmark means that they do not read more than 15 words correct per minute of a word list with one-syllabic words with consonant clusters, in contrast to an average performance of between 37 and 40 words per minute (see below for description of tests). For children in grade 3 with an educational age of 2.2 years, this means that they do not read more than 36 words per minute on the same word list in contrast to an average performance of 64 of 65 words per minute.

In this study, all selected children were enrolled in an intensive school-based intervention. The intervention was studied for effectiveness in a prior study (Struiksma, Scheltinga & van Efferen, 2006; Struiksma et al, in press) and will be further described below. The number of children selected per year was 35, 42, and 45 for cohort 1, 2 and 3, respectively, resulting in a total of 122 children (64 boys and 58 girls). The mean age at pretest was 95.20 (sd. 7.82). 113 children were in grade 2 during intervention; 90 of them had received reading instruction for 1.2 school years; 23 had duplicated grade 1 and had received reading instruction of 2.2 school years; 9 children were in grade 3 and had received reading instruction of 2.2 school years. An overview of the subject characteristics at start of intervention is reported in table 1. The three cohorts did not differ on fluency of word reading scores at the entry of intervention ($F(2,119) = 2.13$, n.s., $\eta_p^2=.04$), neither on chronological age ($F(2,119)=.46$, n.s., $\eta_p^2=.01$).

Table 1. Overview Study; Descriptive Statistics and Distribution of Children Across Cohorts

Cohort	N	Sexe	Age	Word Reading Fluency
I	35	21 b	94.31	12.00
		14 g	(5.96)	(5.87)
II	42	18 b	96.02	13.24
		24 g	(7.60)	(7.62)
III	45	25 b	95.13	15.09
		20 g	(9.25)	(6.40)

Note. Age in months; b for boys, g for girls; fluency of word reading based on part 2 of the DMT at pretest, score is mean number of correctly read words in one minute; Standard deviations between brackets.

Table 2 shows which additional tests were administered per cohort to predict RTI. Scoring data on some tests were missing for a few children because of illness or time limitations. Exact numbers of means and standard deviations per test are reported in the results section.

Table 2. Overview Study; Administration of Task Across Cohorts

Cohort	N	Tasks Administered
I & III	80	Nonword Repetition
		Rapid Digit Naming
II & III	87	Orthographic Choice Task
III	45	Letter Sound Naming

Intervention Based on prior study

The intervention was based on prior research (Struiksma, 2003; Struiksma, van der Leij, & Stoel, 2009) that comprised experiments of context-free word reading fluency training in first and second grade. In sum, the study of Struiksma et al. revealed that reading accuracy was already high at start of the intervention (80%) and fluency improved under favourable conditions of training closed sets of orthographically similar words. However, individual learning curves showed that the differences in fluency gains between children increased over sessions. The response to intervention was related to the acquisition and generalization of knowledge at the sublexical level. Although Dutch has a relatively transparent orthography with regular letter-sound correspondences, it also includes complexities as consonant clusters (Seymour et al., 2003). The poor responders were less able to generalize the trained complex sublexical units ('st' in 'step') to fluently reading of new words with the same units ('stop').

In the present study the intervention of Struiksma et al. was adjusted for large scale implementation. The intervention comprised individual sessions of half an hour, four times per week over a period of 20 weeks. The intervention combined word and text fluency practice as recommended by Lovett et al. (1994). Each session, the child trained word reading fluency during 15 to 20 minutes with a computerized program. Each session of the computer training started with an initial of reading 12 one-syllabic words. The remedial teacher pressed a key to indicate whether the word was read correctly. A voice key was used to register response latencies. Next, the child trained reading the words correctly and as quickly as possible in 6 to 12 trails. Each trial the same 12 words were presented with limited exposure time but in random order. After the child had named the word, the computer generated the spoken form of the word after 750 msec. and the word was again displayed on the screen. The child also received feedback of performance per trial that was indicated by coloring bars at the bottom of the screen. After a child had read at least 10 out of 12 words correctly for 6 trials, the program proceeded with the concluding test that was similar to the initial test. In accordance to performance, difficulty level increased over sessions by decreasing exposure time and increasing word complexity. Word complexity was increased by including words with consonant clusters (for example 'stap' (step) and by decreasing orthographic overlaps between the presented words ('*sta*p', '*sta*m' (trunk) and '*sta*l' (stable) versus 'stap' and 'stok' (stick) and 'slim' (smart)).

The remaining time of a session was spent on reading connected text. The child chose a book of interest at its instructional level which is the level just beyond mastery indicated by a 'sufficient' score on the text reading test. The remedial teacher offered guidance using the method Pause Prompt Praise which is developed according to the behavioral approach (McNaughton et al., 1987; and see Struiksma, 2001 for a Dutch adaptation). While reading, the therapist uses praise as reinforcement for correct reading of a sentence or for self-corrections by the child; the remedial teacher pauses to allow children to self-correct, encouraging independent reading behavior; and the therapist gives prompts, for example meaning-oriented, to assist the child to self-correct. Moreover, to improve fluency, repeated readings of text passages were practiced both timed and untimed.

Training of remedial teachers and treatment fidelity

Remedial teachers at school delivered the interventions. The remedial teachers were trained in a course that comprised 8 meetings that took place in the period September until end of May. At these meetings a range of relevant topics were dealt with: theories of reading, reading development and reading difficulties, screening and testing for reading disabilities and basic behavioral principles. Also experiences and difficulties that were run into were discussed. Remedial teachers were observed at least once at school during a session to ensure that the intervention was implemented consistently. Observations focused on amount of time that was spent effectively on reading ('time on task'), use of the Pause Prompt Praise approach, instruction and scoring. Observations were directly discussed afterwards. Directions for improvement concerned most often the application of Pause Prompt Praise approach. However, in general the intervention was implemented correctly which may be attributed to the highly structured design by use of computer and behavioral instructions.

Procedure

The reading intervention lasted a total of 40 hours provided over 20 weeks during one school year, starting in October. Level of fluency of word reading was determined at the beginning and end of intervention. These measurements at pre- and posttest were taken by remedial teachers at school. Measurements took place in October for pretest, in April for posttest. After participation in intervention, children from cohort 1 and 2 were followed in subsequent years until the end of a three year period of study. The first measurement after posttest took place about 5 months later in October. This measurement was considered for maintenance of reading level.

The measures of rapid digit naming, phonological memory, letter sound naming and orthographic knowledge were administered to different cohorts (see table 2), as part of smaller sub-studies. Tests of phonological processing, nonword repetition and rapid digit naming, were administered to two cohorts; the first and third year of the intervention study. An orthographic knowledge task was administered to two cohorts of the intervention study; the second and third year of the intervention study. A test of letter sound naming fluency was added to the last cohort, the third year, of the intervention study.

Remedial teachers administered the rapid naming tasks together with the reading measures in October. Remedial teachers in the second year administered the orthographic knowledge task in the period from November to December. Administration of the tasks in the first and third year was completed by the first author and took place in the period from November to December. In the third year, the first author was assisted by a test assistant. In the third year, the nonword repetition test, the orthographic knowledge task and letter sound fluency were administered all together in one session of about 40 minutes. Testing took place at school. All testing took place individually in a quiet room. It was ensured that children felt comfortable during testing. It was made clear that they could not do it 'wrong' and the purpose of testing was explained.

Measures for screening and evaluation

Word reading fluency. This was measured at pre- and posttest by presenting a word list of the Drie-Minuten-Test (DMT) [Three-Minute-Test] (Verhoeven, 1995). The DMT is a standardised Dutch test which comprises three cards of word lists of increasing difficulty. For the current study one card was used containing a word list of one-syllabic words with consonant clusters (CCVC, CVCC and CCVCC), which were not identical, but similar in word structure as the trained words during intervention. The reading card is made up of five columns with 30 words in each column. Children are instructed to read the words as accurate and as fast as possible within one minute. The score is the number of accurately words accurately read in one minute. The manual reports the reliability for the card 1 and card 2 varies from .90 and .94 for grade 2 and grade 3. This is based on normative testing with 3431 Dutch children (Moelands, Kamphuis & Verhoeven, 2004).

Predictor variables.

Rapid Naming. The *rapid naming task* (Struiksma, van der Leij & Vieijra, 2004) required children to name as quickly as possible digits on a card. The numbers 1 until 9 were each presented 4 times in random order in 3 columns. The total time taken was recorded in seconds and the number of items named correctly was also recorded. The total number of items named correctly was divided by the total time taken to name the 36 items. The score used for analysis represented the number of items named correctly per second.

Nonword Repetition. A Dutch version of a *nonword repetition test* (Scheltinga, 2003) was developed (see Gathercole, Willis, Baddeley & Emslie, 1994). The test required the repetition of pseudowords, varying in word length from 2 to 5 syllables. Twelve words of each length were presented, resulting in a total of 48 words. Neither nonwords, nor constituent syllables corresponded to existing words. Low-frequent CVC syllables were selected of which multi-syllabic words were constructed. All vowels were lax vowels. Within a nonword, a vowel occurred only once. Word complexity and phonological similarity within each length category were controlled for. The before last syllable of all words is pronounced with stress. Stress of the 2-syllabic words is on the first syllable. A professional speech therapist pronounced all nonwords in consistent rate. All nonwords were recorded by using the computer program CE2000 (Syntrillium Software Corporation). Two test items preceded the test. Each nonword was preceded by a short beep of 800 Hz of 200 msec. The child heard the word through a headphone and had to repeat the word directly by microphone. The answers were recorded in CE2000 to enable detailed analysis of the number of correctly repeated syllables. Due to a programming error, the answers on two 5-syllabic words were missing for a number of children (n=32) resulting in a total of 46 responses. Therefore, the total score was based on 46 responses for all children. Reliability was Cronbachs Alpha .88 based on the group sample of the present study.

Orthographic Knowledge. An *orthographic knowledge* task was developed and administered which required choosing the correct written word from a pair of words with the same pronunciations (homophones). The test was computerized. The child was presented with a spoken form of a word, directly followed by two written alternatives on the screen. The child was required to choose the correct written word form as quickly as possible. The cursor was fixed on the middle of the screen until appearance of the written words. The child had to click on the correct word form. The test consisted of three parts of which the first two parts were considered for practice. In the first part, the child heard a word, which was shown on the screen left or right from the cursor. The child had to click on the word as quickly as possible. This part was used to determine the mean reaction time needed to move the cursor to the word. Reaction time in subsequent parts was corrected for the mean reaction time needed to click on a word. In the second part, the child was presented with the spoken word, after which two written alternatives were presented on the left and right of the cursor. One word was presented in the correct form. The incorrect word form sounded differently after correct phonological decoding. In the third part, two orthographically different written alternatives, which sounded identical after correct decoding, were presented. This part consisted of 60 test items. The score was the number of correct responses on the third part. Reliability was Cronbach's Alpha .82 based on the group sample of the present study. Mean reaction time was also registered. Response times were corrected for the average moving speed of the cursor determined by the first part of the test.

Letter and grapheme knowledge. In addition, a *letter sound naming* task was developed to measure fluency of grapheme-phoneme knowledge. During six trials, the child was required to pronounce the sound of the grapheme as fast as possible. The first 5 trials were considered as practice, the sixth trial was considered as test trial. Each trial, 28 graphemes were consecutively presented in the centre of the screen. During the practice trials the computer generated the correct pronunciation of the grapheme 250 msec. after the child had given a response. Feedback on accuracy was given on the screen. The test administrator pressed a key to record accuracy. A voice key registered naming times. The set of 28 graphemes comprised 17 graphemes for vowels, both single graphemes (5) and digraphs (12), and 11 graphemes for consonants. Because of unreliable registration by voice key, the fricatives /s/ and /z/ and /f/ and voiceless plosives /p/ and /t/ were excluded from presentation. For analyses mean response time at the sixth trial was used. Reliability was Cronbach's Alpha .82. Accuracy scores approached ceiling (94% of the graphemes were named correctly).

Results

Response to Intervention

To give an indication of the variation in response to intervention, responsiveness was defined according to progress in word reading fluency on the word list with CCVC and CVCC words (card 2 of DMT). Reading scores that were still at or below the lowest 10^{th} percentile after intervention were classified as poor response. To obtain a reading score above the 10^{th} percentile, a child had to make gains equal to more than 3 months of education which means that a child should read at least 12 words more compared to pretest score. This criterion was based on norm scores which indicate what reading score is expected after the number of months of received education. The norm scores are computed from the raw scores (i.e., words read correctly per minute) as provided in the manual of the standardized reading test.

Gains ranged from no improvement to an increase of 47 words on the word reading fluency score. From the 122 children enrolled in intervention, 46 children

(37.7%) obtained reading scores above the 10^{th} percentile. 79 Children (62.3%) still performed below or at the 10^{th} percentile after intervention (37 boys, 39 girls). It should be noted that classification of good versus poor responders is dependent on the adopted criteria (Mcmaster, Fuchs, Fuchs & Compton, 2005). The benchmark of the 10^{th} percentile however was chosen because of its practical relevance in the Dutch educational system. Descriptive statistics for the groups classified by using the cut-off score at the 10^{th} percentile boundary are presented in table 3. Means and standard deviations on the fluency of word reading task at pre- and posttest are given. In addition, growth conveyed as number of months of improvement is given. The progress made by the good responders was on average 23.30 words per minute (standard deviation of 7.22) versus 8.91 words per minute (standard deviation of 4.98) in the group of poor responders which equals average growth of 6.26 months and 2.54 months respectively. The good and poor responders did not differ in chronological age (t=.83, df=120, n.s.) or educational age (t=1.3, df=120, n.s.). The dependent measure of word reading fluency was analyzed via a 2 (group: poor vs. good responders) X 2 (time: pretest, posttest) multivariate analysis of variance (MANOVA) with repeated measures on the second factor. The MANOVA determines whether a significant difference exists between the two groups when compared on the reading measures at pre- and post-testing simultaneously. There was a main effect of group ($F(1,120)=55.41$, $p<.05$, $\eta_p^2=.32$) and a main effect of time ($F(1,120)=849.52$, $p<.001$, $\eta_p^2=0.88$). The group by time interaction reached significance indicating that the good responders made more progress than the poor responders ($F(1,120)=169.69$, $p<0.001$, $\eta_p^2=0.59$). In absolute terms the increase in words per minute was larger in the good responders than in the poor responders. On average the good responders obtained higher reading scores at pretest. This difference however was not significant ($F(1,120)=3.87$, $p=.052$, $\eta_p^2= .031$). The good and poor responders differed significantly at posttest ($F(1,120)=118.47$, $p<.05$, $\eta_p^2= .50$) which is of course a expected result since we nominated children as good or poor responder on the base of their posttest scores.

Table 3. Descriptive Statistics and Mean Performance Scores on Reading for Good versus Poor Responders

				Word Reading Fluency			
Group	N	Sex	Age	Pretest	Posttest	Growth	Maintenance
Good	46	27 b	95.96	15.11	38.41	6.26	36.14[a]
responders		19 g	(7.56)	(7.05)	(9.88)	(2.10)	(13.76)
Poor	76	37 b	94.75	12.63	21.54	2.54	23.14[b]
responders		39 g	(7.59)	(6.55)	(7.19)	(1.57)	(9.35)
Total group	122	64 b	95.20	13.57	27.90	3.94	26.90[c]
		58 g	(7.82)	(6.82)	(11.65)	(2.54)	(12.13)

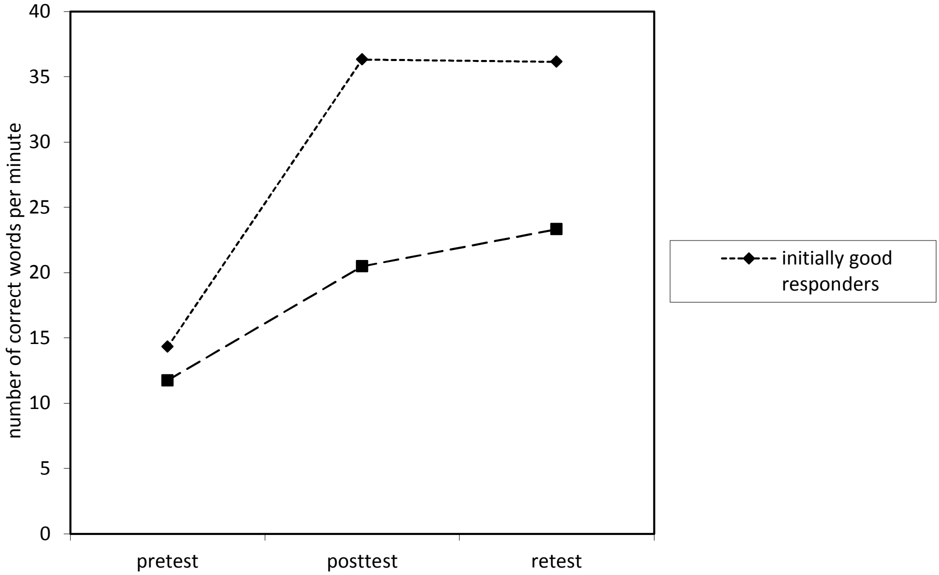
Note. Age in months; word reading fluency is based on chart 2 of the DMT, score is mean number of correctly read words in one minute; growth, increase in words per minute equal to the number of months on average needed to make the progress; standard deviations between brackets.
[a] N=22, [b] N=57, [c] N=79.

Since the good responders were already better readers at pretest, the interaction effect might merely reflect a proportional increase in words per minute. In other words, it might be that the increase in words per minute is of similar growth percentage in both groups. To check whether the significant interaction reflects a proportional effect, the scores in the various conditions are subjected to a logarithmic transformation (Levine, 1993; Martens & de Jong, 2008) and a MANOVA is performed on the transformed scores. The significant interaction effect remained ($F(1,120)=30.52$, $p<0.001$, $\eta_p^2=0.20$) and it is therefore safe to conclude that the two groups make different progress that is not caused by a proportional effect.

From 67 out of 77 children in cohort 1 and 2, scores at maintenance testing were available 5 months after posttest. Since the study stopped after three school years, maintenance data from cohort 3 were not available. Means and standard deviations are also reported in table 3. The difference between posttest and maintenance testing was considered for the groups of good and poor responders as defined at posttest. The dependent measure of word reading fluency was analyzed in a 2 (group: poor vs. good responders as defined at posttest) X 2 (time: posttest, retest) multivariate analysis of variance (MANOVA) with repeated measures on the second factor. There was a main effect of group ($F(1,65)=56.46$, $p<.001$, $\eta_p^2=.47$), no main effect of time ($F(1,65)=1.65$, n.s, $\eta_p^2=0.04$) and no group by time interaction ($F(1,65)=.56$, n.s., $\eta_p^2=0.01$). On average, there was no significant progress between post- and maintenance testing, but the difference between good and poor responders remained stable.

Additionally, children were classified as poor vs. good readers based on their reading scores in comparison to the normative group at maintenance testing. Children with reading scores below the 10th percentile were classified as 'poor'. At the moment of retesting 1 of the poor responders (n=45) had improved fluency in word reading and could be classified as good responder. From the 22 readers that were classified as good responders directly after intervention, 14 did no longer meet that criterion at retest; they did not improve their reading scores at a similar rate as during intervention. At maintenance testing, only 9 classified as good responders according to the criterion, indicating both the severity of the reading problems and the need for continued individual intervention.

Figure 1. Responders versus non-responders



Note. N=79, response of poor versus good responders whose word reading fluency scores were available at pre-, post- and maintenance testing

Prediction of response to intervention.

In hierarchical regression analyses with reading scores at posttest as dependent variable controlling for age and reading scores at pre-test, the predictor variable was entered in the last step in the regression analysis (Cohen & Cohen, 2003). Data were screened on outliers (Stevens, 1996). Outliers on the dependents were defined as cases with a standardized residual greater than 3.3. Mahalanobis Distance was used to determine outliers on the set of predictors. Furthermore, we examined the influence of data points in affecting the regression equation. A Cook's distance >1 was considered large.

Measures of phonological processing as predictor of RTI

The phonological processing tasks were administered to the first and third year groups of the intervention study (see table 2). Due to illness (n=3) or poor sound recording of the answers (n=2), the data of 5 children were not available for analyses. In addition, one case was identified as an outlier on the rapid naming task. A total of 74 children were included in the analyses of phonological processing tasks. There was no ceiling effect on the nonword repetition test. The descriptive statistics are presented in table 4.

Table 4. Descriptive Statistics and Intercorrelations Between Age, Word Reading Fluency and Phonological Skills

Variable	Age	Word Reading Fluency (WRF)		Rapid Digit Naming	Nonword Repetition
		Pretest	Posttest		
Age	—	.48**	.32**	.41**	.14
WRF Pretest		—	.52*	.55**	.34*
WRF Posttest			—	.55**	.31*
Rapid Digit Naming				—	.21
Nonword Repetition					—
M	94.85	13.77	27.09	1.32	102.05
SD	6.13	6.13	10.32	.33	18.58

Note. N = 74; Fluency of word reading based on chart 2 of the DMT, number of correct words per minute; Rapid Digit Naming, number of digits per second; Nonword Repetition, number of correct syllables.
* $p< 0.05$, ** $p<0.001$

Table 4 shows also the correlations between the predictors with dependent and control variables. The scores on the nonword repetition test correlated significantly but not very highly with reading at pretest (.34) and reading at posttest (.31). Rapid naming task scores and reading were moderately correlated (-.55 and -.55 at pretest and posttest, respectively). The negative sign indicates that better reading scores are associated with faster naming. The rapid naming task and nonword repetition test were not significantly correlated, which is in agreement with findings of others (de Jong and van der Leij, 2002) that the rapid naming task and nonword repetition test are independent measures of phonological processing.

The rapid naming task and nonword repetition test were separately entered into a regression model after controlling for age (in months) and reading score at pretest. The results are reported in table 5. Both age and reading at pretest explained unique variance in reading scores at posttest (ΔR^2=.10 and ΔR^2=.17, respectively). Children who are better readers at pretest are better readers at posttest. Rapid naming (ΔR^2=.096) but not nonword repetition (ΔR^2=.022) explained additional variance in reading at posttest. However, it does not imply that phonological processing measured by nonword repetition does not contribute to reading. Instead, reading at pretest mediated the contribution of nonword repetition on reading at posttest. Nonword repetition explained variance in reading at pretest (ΔR^2=.087, p<.05) and reading at pretest explained reading at posttest after controlling for nonword repetition scores (ΔR^2=.197, p<.001). The test statistic Sobel was computed to examine the significance of mediating effect of reading at pretest on the relationship between nonword repetition scores and reading at posttest. The test statistic for the Sobel test was 13.66, p<.05 indicating that the relationship between nonword repetition scores and reading at posttest was significantly mediated by reading at pretest. After controlling for the mediating effect of reading at pretest, the correlation between reading at posttest and nonword repetition performance disappeared, indicating full mediation.

Table 5. Summary of Hierarchical Regression Analysis

	Variable	Posttest Word reading fluency			
	Order	ΔR^2	ΔF	Beta	t
		Model 1			
1	Age	.101	8.06*	.02	.16
2	Pretest Word Reading Fluency	.170	16.59**	.30	2.49*
3	Rapid Digit Naming	.096	10.59*	.38	3.26*
		Model 2			
1	Age	.101	8.09*	.10	.86
2	Pretest Word reading fluency	.170	16.59**	.42	3.48*
3	Nonword Repetition	.022	2.22	.16	1.49

Note. N= 74; Rapid Digit Naming, number of digits per second; Nonword Repetition, number of correct syllables per word. Beta refers to the beta in the final model.
* $p< 0.05$, ** $p<0.001$

Measures of Letter Sound Fluency and Orthographic Knowledge as Predictors.

The orthographic knowledge task was administered to the second and third cohort whereas the letter sound fluency task was only administered to the third cohort (see table 2). The results are presented in subsequent order. The orthographic knowledge task was administered to 80 children from the second and third cohorts. Results were missing for 7 children due to unknown reasons. The means and standard deviations on accuracy scores and response times are presented in table 6.

Table 6. Descriptive Statistics and Intercorrelations

Variable	Age	Word Reading Fluency (WRF)		Orthographic Choice (OC)	
		Pretest	Posttest	Reaction Time	Accuracy
Age	—	.50**	.48*	-.22	.46**
WRF Pretest		—	.66**	-.33*	.60**
WRF Posttest			—	-.24*	.46*
OC Reaction Time				—	-.22
OC Accuracy					—
M	94.96	13.85	28.68	2.21	38.25
SD	8.19	6.76	11.50	1.21	8.18

Note. N = 80; Word Reading Fluency based on chart 2 of the DMT, number of correct words per minute; Reaction time, mean reaction time per item in seconds; Accuracy, number of correct choices with a maximum score of 60. * $p< 0.05$; ** $p<0.001$.

The analyses of the response times on the orthographic knowledge task were based on response times for valid and correct responses only. In addition for each child, a mean response time and a standard deviation were computed. Response times that deviated from the child's mean more than 3 standard deviations were considered outliers and excluded from the analyses. Response times of incorrect responses on the orthographic knowledge tasks amounted to a percentage of 37.28% of the responses. Taking into account the outliers on response times, the excluded data on reaction times totaled 38.17 % of the data. There was no ceiling effect on accuracy scores. The highest number of correct answers was 55 out of 60.

The correlations between reading measures and orthographic measures are presented in table 6. The accuracy scores on the orthographic knowledge task were correlated with fluency of word reading at pretest and post-test (r=0.60 and r=0.46, respectively). The response times also correlated to fluency of word reading scores at pretest and posttest (r= -.33 and r= -.34). The negative correlation between the response times on the orthographic knowledge task and reading indicates that a slow response time on the orthographic knowledge task correlated with a low number of words read per minute.

All 80 cases were included in the hierarchical regression analysis (see table 7). After controlling for age and reading at pretest, neither accuracy nor response time on the orthographic knowledge task explained additional variance. Although there was no significant effect of orthographic knowledge on reading at post-test, the influence of orthographic knowledge on later reading was likely to be mediated by earlier reading skills. To test for mediation effect, the test statistic Sobel was computed to examine the mediating effect of reading at pretest on the relationship between accuracy scores and response times on the orthographic knowledge task and reading at posttest. For the accuracy scores, the test statistic Sobel was .51, $p<.001$ which means that the relationship between accuracy scores and reading at posttest was significantly mediated by reading at pretest. For the response times, the test statistic for the Sobel test was -.002, $p<.05$, indicating that reading at pretest mediated the relation between response times and reading at posttest. After controlling for the mediating effect of reading at pretest, the correlation between reading at posttest and accuracy scores and response times of the orthographic knowledge test disappeared, indicating full mediation.

Table 7. Summary of Hierarchical Regression Analysis

	Variable	Posttest Word reading fluency			
	Order	ΔR^2	ΔF	Beta	t
		Model 1			
1	Age	.225	23.66**	.19	1.95
2	Pretest Word Reading Fluency	.236	29.78**	.56	5.52**
3	Orthographic Choice, RT	.000	.04	-.02	-.21
		Model 2			
1	Age	.225	23.66**	.18	1.81
2	Pretest Word reading fluency	.236	29.78**	.53	4.81**
3	Orthographic Choice, Acc	.002	1.20	.06	.52

Note. N= 80, Word Reading Fluency is based on chart 2 of the DMT; RT, Reaction Times; Acc, Accuracy. Beta refers to the beta in the final model.
* $p< 0.05$, ** $p<0.001$

The letter sound naming task was administered to 45 children. Data on both accuracy and response times were collected. For analyses, only the response times on correct and valid responses were used. Response times on incorrect responses were not included in the analyses (5.36%). In addition, response times were considered to be invalid if the response time was below 500 msec. This criterion was determined by visual inspection of the scatter plot of response times. Furthermore, for each child, a mean response time and a standard deviation were computed. Response times that deviated from the child's mean more than 3 standard deviations were considered outliers and excluded from analyses. In all, the incorrect responses and invalid response times amounted to a percentage of 11.41% of the total data. Descriptive statistics are reported in table 8.

Table 8. Descriptive Statistics and Intercorrelations

Variable	Age	Word Reading Fluency (WRF)		Letter Sound Fluency (LSF)	
		Pretest	Posttest	Reaction Time	Accuracy
Age		.55**	.48*	-.25	.15
WRF Pretest		—	.58**	-.36*	.22
WRF Posttest			—	-.22	.05
LSF RT				—	-.01
LSF Accuracy					—
M	95.13	15.09	29.49	.88	.94
SD	9.25	6.40	10.55	.14	.05

Note. N=45; Word Reading Fluency based on chart 2 of the DMT, number of correct words per minute; RT=Reaction time in seconds per item; Accuracy, percentages correctly named letter sounds.
* $p< 0.05$, ** $p<0.001$.

The correlations between reading measures and letter sound fluency task are presented in table 8. The accuracy scores were not related to word reading scores at pretest and posttest (r=0.22 and r=0.05, respectively), but this might be due to ceiling effect as 94% of the letters were pronounced correctly. The response times correlated significantlybut not very highly with word reading scores at pretest (r= -.36) but not with reading scores at posttest (r= -.22). Because of absence of correlations with reading scores at posttest, hierarchical regression analyses with the letter sound naming task as predictor were not performed.

Discussion

Response to Intervention.

The present study supports findings that response to intervention as indicated by fluency of word reading varies largely among poor readers in the lowest range of reading performance. After an intervention of 20 weeks, the range in individual growth of word reading fluency score varied from no gain at all to an increase of 47 words per minute. The latter score corresponds to a progress normally expected after 15 months according to norm scores. To illustrate variation in response to intervention, responsiveness was defined using a cut-off score on fluency of word reading based on norm scores directly after intervention. Improvement of reading skills above the 10^{th} percentile was considered as good response which was obtained by 46 out of 122 children. The reading scores of the other 76 children fell still in the lowest decile. At retest after five months, good and poor responders showed on average no significant improvement in reading scores. It is important to note that the retest took place about one month after a 6-week summer vacation during which children received no formal reading instruction. Although struggling readers are in need of more practice (Torgesen et al., 2001), they are less willing to read voluntarily. Nevertheless, at the individual level, some children continued to improve their reading skills, although most of them at a lower rate than during the intervention. As a consequence, some children did no longer meet the criterion for good responder at retest; 14 out of 22 children who were initially classified as good responders at posttest did not keep up reading growth with the normative group. In contrast, 1 child showed the opposite pattern with poor reading score directly after intervention followed by an increase afterwards. Children who changed from responsiveness status could not be distinguished from the children who remained stable.

The results support earlier findings that word reading fluency skills of poor readers are hard to remediate (e.g. Torgesen, 2000). Whereas the intensive one-to-one intervention resulted in a significant progress in word reading fluency skills for a substantial number of children, an even larger proportion did not profit, at least not in the sense that their word reading performance was improved up to a higher norm-based level. This latter result may not be surprising considering the stringent selection criterion of reading scores at or below the 10^{th} percentile. The results at maintenance testing suggest that many children, even children who showed initially good response to intervention, may remain in need of continued intervention and might not respond to mainstream or classroom instruction.

Overall, however, the results on the effectiveness of the intervention on context-free word reading fluency should be considered with caution. It can not be ascertained how much gain is due to natural growth, unmeasured factors or intervention since a control group was not included (Yoder & Compton, 2004). On the other hand, it should be noted that the group of children under study had obtained no or poor reading achievement after at least one year of formal reading instruction during which reading process was monitored following similar testing procedures as in the present study. Considerable gains and change in rate of learning would not likely have occurred without intensive intervention. Therefore, it can be argued that the progress made can be largely ascribed to the intervention. Future studies to the effectiveness of similar interventions could be improved by including control groups receiving no intervention or standard practice condition. The reliability of the growth parameter might also improve by including more frequent assessments of reading measures before, during and after intervention.

Prediction of response to intervention.

The autoregressive effect of word reading fluency at pretest explained most of the variance in intervention outcome indicated by word reading fluency at posttest, replicating findings by other researchers (Lovett et al., 1990; O'Shaughnessy & Swanson, 2000). Rapid digit naming contributed unique variance (10%), after the variance explained by age and word reading fluency at pretest (27%) was partialled out. Letter sound fluency was correlated to word reading fluency at pretest, but not at posttest. Nonword repetition and orthographic knowledge were correlated to word reading fluency at pre- and posttest, but both skills did not add unique variance to the prediction of responsiveness. The effect on word reading fluency at posttest was mediated by reading at pretest.

The finding that serial rapid naming of digits predicted variance in growth of word reading fluency after intervention, corresponds with the finding of Nelson et al. (2003) who identified rapid naming as the learner characteristic most strongly related to RTI in their meta-analysis. Moreover, it supports the suggestion by de Jong and van der Leij (2003) that differences in rapid naming influence the extent to which first graders benefit from instruction or become poor readers. This relationship may be assumed to be particularly strong in case of responsiveness defined according to fluency of word reading (Torgesen et al., 2001), presumably because the ability to name a limited set of symbols, letters or digits rapidly reflects the ability to retrieve the letter sounds or whole word sounds while reading

(Wolf and Bowers, 1999; van den Bos, Zijlstra, & Lutje Spelberg, 2002). As is suggested by Pennington (2006), serial rapid naming relates to precise timing mechanisms that characterize the intermodal processing component necessary for achieving automaticity in more advanced stages of reading development.In the acquisition of word reading fluency in transparent orthographies rapid naming is the most consistent correlate in comparison with other phonological skills (Holopainen, Ahonen, & Lyytinen, 2001; de Jong & van der Leij, 1999; Wimmer, 1993; Wimmer, Mayringer & Landerl, 1998). The finding of the strong contribution of rapid naming skills to reading extends to opaque orthographies as well (Bowers, 1995, Mc-Bride-Chang & Manis, 1996, Savage & Fredrickson, 2005). With regard to RTI, Wise et al. (2000) found that rapid naming skill predicted gains in rate of word reading after intervention. They suggested that rapid naming skills affect the ultimate reading speed. Since naming speed is not easily improved through training (de Jong & Oude Vrielink, 2004), rapid naming skills at start of intervention may give an indication of the ultimate ability to improve the rapid retrieval of letter sounds and whole word sounds during reading (e.g. Torgesen et al., 2001).

Fluency in naming letter sounds correlated less strongly to reading skill at pretest than rapid digit naming, which was not a consequence of smaller group sample. In addition, letter sound fluency did not significantly correlate with reading at posttest. These results might be surprising, since in other studies the relation between reading and rapid naming has been found to be stronger in case of letter naming than of digit naming (de Jong & Oude Vrielink, 2004; Wile & Borowsky, 2004). In comparison with rapid naming tasks, it should be noted that our letter sound fluency task differs in task design. Instead of serial presentation of a limited set of letters in a sequence, 28 graphemes, including consonants, vowels and digraphs were presented on distinct trials in random order. Contrary to the usual rapid naming tasks, our letter sound fluency task did not merely reflect the speed of retrieval of over-learned symbols, but also knowledge of a variety of grapheme-sound relationships. Moreover, the relation between naming of items and reading has been shown to be stronger when the naming task requires continuous naming of items presented in a sequence (Bowers, 1995; Kail, Hall & Caskey, 1999). As in serial rapid naming, reading involves rapid sequential processing of individual symbols. Furthermore, the importance of letter sound naming to reading appears dependent on the reading task demand. Wile and Borowsky (2004) showed that letter sound naming is uniquely related to pseudohomophone and nonword naming latencies, whereas rapid naming of digits and letters was related to exception word naming. Finally, the contribution of letter sound fluency might be stronger at an earlier stage of reading development. Studies using tasks of letter knowledge and letter sound fluency to explain variance in RTI (Ritchey & Speece, 2006; Stage et al., 2003) focused on children in an initial stage of reading acquisition, from kindergarten to first grade. At pretest, letter sound fluency and word reading fluency were related.

Considering phonological memory, its contribution to RTI might have been stronger when *accuracy* and not *fluency* in word reading was taken as outcome measure. Phonological memory appears to be specifically involved in accurate reading of pseudowords in which elaborate phonological recoding is required (Gathercole and Baddeley, 1993). Indeed, Torgesen et al. (1999) found that phonological memory skills explained growth on the unspeeded reading measure of word attack, but not word identification. Hatcher and Hulme (1999) also did not find a predictive role of phonological memory in RTI indexed by growth in word identification. The focus of the present study, however, was on fluency, since in relatively transparent orthographies; in particular differences in reading rate and not in accuracy determine the differences between good and poor readers (Wimmer, 1996). Additionally, the contribution of phonological memory has been found stronger at an initial stage of reading development (de Jong and van der Leij, 1999). In a study of RTI in at risk kindergarten children, Schneider et al. (1999) found that phonological memory was the best predictor of growth through training. However, when reading achievement proceeds, the strength of the predictive role diminishes since phonological memory starts to develop in interaction with reading skills and the relation between reading and phonological memory becomes stable (de Jong & van der Leij, 1999). Accordingly, after the autoregressive effect of reading is taken into account, no variance is left for phonological memory to explain.

Similarly, orthographic knowledge defined as knowledge of spelling patterns develops in interaction with word reading skills. Caravolas, Hulme and Snowling (2001) showed that reading practice and experience are important determinants of conventional spelling skills. It can be argued that orthographic knowledge develops as a product of reading practice. Accordingly, after taking reading skills into account, orthographic knowledge does not add unique variance to the prediction of responsiveness. The accuracy and speed of recognition of correct spellings was related to word reading fluency. It may be argued that the extent of ability to learn orthographic patterns through reading practice will put constraints on the ability to improve word reading fluency.

Overall, contrary to other studies we did not find much evidence for predictive contribution by reading related subskills. In addition to initial fluency of word reading only rapid naming accounted for a small amount of the variance in responsiveness. Possibly, the use of single tests to measure an underlying construct with relatively small participant-to-test ratios is not very powerful. It may also be argued that the use of a factor comprising different measures of a single construct may have been a better predictor, more sensitive for individual differences, while less sensitive to measurement error (Bowey, 2005). Moreover, more than one skill is likely to predict the response to intervention (Stage et al., 2006). On the other hand, including more measures of one or more underlying constructs may not have resulted in a larger amount of explained variance. An intervention study in German – similar to Dutch in orthographic complexity – reported that a range of predictive skills only accounted for small amounts of individual differences in kindergarten (e.g. Schneider, Ennemoser, Roth & Kuspert, 1999).

In addition, it is important to note that for the present study children were selected within a quite restricted range in comparison with other studies. Only children with fluency of word reading scores at or below the 10th percentile at pretest were included. Other studies report selection criteria of below the 15th percentile (Vellutino et al., 1996) or below the 20th percentile (Berninger, 1999). Taking a larger range of reading skills at the start of intervention will result in larger variances in scores on the predictor variables, increasing the possibility of finding higher correlations and differences between poor and better readers. Furthermore, studies differed in selection criteria related to the moment of identification. Studies that were conducted in kindergarten used phonological predictors (e.g. Torgesen et al., 1999), whereas studies that included children in later grades used actual reading scores as criterion (e.g. Vellutino et al., 1996, 2006). This latter criterion is likely more reliable in the identification of at-risk children than phonological processing skills or early reading skills. At a younger age, reading skills are instable (Spira et al., 2005) and reading disabilities may not be reliably identified. Consequently, selection at a preliterate or early reading stage may include false positives. Obviously, those children who are incorrectly identified as reading disabled at the start of intervention will show better reading outcomes in combination with better reading related skills.

The present study underscores that controlled intensive intervention can generate considerable, though individually variable improvement in word reading fluency within a group of very poor readers. Pretest reading and reading related measures merely accounted for a modest part of the variance at posttest. This finding gives strong support to the response to intervention approach as skill-specific means to identify severe reading disabilities, i.e., dyslexia, meanwhile ruling out inadequate instruction as a cause and helping the milder cases. The intervention included an investment per student of 40 hours of one-to-one practice and instruction, provided by an expert tutor following an evidence-based instruction method. Costly as it may be, identification by monitoring response to intervention appears a better approach than reliance on measurement of reading related subskills. Although phonological and orthographic subskills are universally recognized as important correlates of reading acquisition and differences in reading development, these subskills are not likely to be reliable in predicting at the individual level which struggling readers in the lowest range will develop severe reading problems under specialized instructional conditions.

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CHAPTER 4

IMPROVING READING FLUENCY IN CASES WITH SEVERE DYSLEXIA

Abstract

The aim of this study was to improve the word reading fluency of Dutch children with severe dyslexia in a clinical setting. Word reading fluency deficits are most characteristic for children with dyslexia learning to read in a relatively transparent orthography. However, this deficit has appeared hard to remediate. Studies report mixed results on effectiveness of treatments which might be partly due to different selection criteria to qualify children for treatment. In this study, the response-to-intervention (RTI) approach was used to select poor readers who showed no or limited progress after lengthy, intensive school-based intervention. An experimental treatment method focusing attention on the mapping between phonology to orthography of sublexical features of words was compared with a neuropsychological treatment method that is often applied in the Dutch clinical setting. Results show that continued outpatient treatment leads to further, though small, improvement of both word and text reading fluency. No differences were found between the two treatment conditions. Follow-up testing showed that the majority of children sustained the reading level that was obtained directly after treatment.

Background

Insight in effective ways to remediate reading problems is increasing (Alexander & Slinger-Constant, 2004; Torgesen, 2005). Different types of intervention have successfully reduced the reading problems of a substantial proportion of poor readers (e.g. Duff, & Clarke, 2011; Lovett, Steinbach, & Frijters, 2000; Regtvoort et al., 2013; Scanlon et al., 2004; Torgesen et al., 2001; Torgesen, 2005). Moreover, improvement of reading through intervention has been shown to be related to changes in brain activity (Simos, et al., 2007). Amongst the instructional principles that have been put forward as effective components of intervention (see for reviews Duff & Clarke, 2011; Swanson et al., 1999), are phonological training, decoding practice and reading strategy instruction. Generally, interventions need to be focused, explicit, systematic and intensive (Shaywitz, Morris, & Shaywitz, 2008). In addition, intervention should provide children many repeated opportunities to practice reading fluency and comprehension (Foorman & Torgesen, 2001; Lovet et al., 1994). However, it is well established that not all children respond adequately to interventions that are generally effective for the majority of the children (e.g., McMaster, Fuchs, Fuchs, & Compton, 2002; Struiksma, Stoel, & van der Leij, 2009; Torgesen, 2000; Torgesen et al., 2001; Vellutino et al., 1996). This proportion of 'nonresponders' is estimated at 2 to 6% of the children (Torgesen, 2000). Knowledge about how to remediate effectively the severe reading problems of these children remains limited (Torgesen et al., 2001). In the present study, we aimed to examine treatment response of very poor Dutch readers who demonstrate minimal response to previous intervention.

The reading difficulties of children with severe dyslexia can be best characterized by poor reading fluency, in particular with context-free words (Fletcher, 2009; Lovett, Benson, & Olds, 1990; Torgesen, 2000). Findings suggest that very poor readers can be effectively taught decoding skills (Lyon & Moats, 1997), but their reading rate remains slow (e.g. Torgesen, 2005). Comparable findings have been reported for studies conducted in English as in other languages with more transparent orthographies. A relatively transparent orthography with many regular correspondences between letters and sounds allows even poor readers to obtain high levels of accuracy after only a few months of reading instruction. However, their reading rate remains slow and does not improve easily (Dutch: Yap & van der Leij, 1993; Italian: Zoccolotti, De Luca, Di Pace, Judica, Orlandi & Spinelli, 1999; Finnish: Holopainen, Ahonen, Lyttinen, 2001; German:

Landerl, Wimmer, & Frith, 1997; Landerl & Wimmer, 2008). Intervention studies also show that reading fluency is hard to improve (e.g. McMaster et al., 2005; Torgesen et al., 2001; Vadasy, Sanders & Tudor, 2007; Vaugh et al., 2009). A Dutch intervention study that was directed at training word reading fluency by an intensive, school-based, one-to-one intervention (Struiksma, van der Leij & Stoel, 2009) reported that about one third of the children made virtually no progress. Although accuracy scores were high, fluent recognition of single words remained poor. Since fluency of word recognition will also affect text reading fluency and ultimately reading comprehension (e.g. LaBerge & Samuels, 1974; Perfetti, 1992; Schwanenflugel et al., 2006), it appears evident to focus on remediation of word reading fluency problems of children with severe reading problems.

Response to intervention

It has recently been advocated to take into account responsiveness to intervention to identify children with severe reading problems. A response to intervention (RTI) approach is considered as a method to control for poor instruction at school by monitoring reading progress of poor readers in interaction with remedial instructional conditions in order to differentiate between children who profit from such activities and children with persistent dyslexia (Burns, Appleton & Stehouwer, 2005; Fletcher, Coulter, Reschly & Vaughn, 2004; Fuchs, Compton, Fuchs, Bryant & Davis, 2008; Vaughn & Fuchs, 2003; Vaughn, Linan-Thompson & Hickman, 2003; Vellutino, Scanlon, & Lyon, 2000). Careful monitoring of the responsiveness to intervention limits the role of poor teaching or lack of experience as cause of reading difficulties (Case, Speece, & Molloy, 2003). According to the definition of dyslexia by the International Dyslexia Association (2002), the severe reading disability should not be caused by lack of provision of effective classroom instruction (e.g. International Dyslexia Association, 2002). In the DSM-5, the learning disorder is additionally characterized by '*symptoms that have persisted for at least 6 months, despite interventions that target those difficulties'* in DSM-5 (2013, p. 66), Within the RTI approach, instruction, guidance and practice are adapted to the needs of the children. Ideally, RTI includes consecutive layers or tiers of increasingly intensive intervention. In most RTI models, three tiers are differentiated. After more general high-quality class-level instruction (tier 1), children with poor progress qualify for supplemental intervention that is more intensive, provided in small-groups or one-to-one at school (tier 2). Children who fail to acquire word reading skills despite participating in intervention programs at the tier 2 level – so called nonresponders (McMaster et al., 2005), treatment resisters or hard-to-remediate children (Torgesen, 2000) – are identified as most likely dyslexic readers who are in need for more intensive, individual, long-term intervention (Fuchs, Mock, Morgan & Young, 2003) at tier 3. As the focus of this study is on children with severe reading problems, the remediation of their reading difficulties is studied within a RTI framework.

A number of studies have examined the effects of supplemental intervention for non-responders who were selected after a period of tier 2 intervention (Berninger et al., 2002; O'Connor, Hartry & Fulmer, 2005; O'Connor, Hartry, Fulmer, & Bell, 2005; McMaster, Fuchs, Fuchs & Compton, 2005; Vadasy, Sanders, & Jenkins, 2002; Vadasy, Sanders & Abbott, 2008; Vellutino et al., 1996; 2002; Wanzek & Vaughn, 2008). Wanzek and Vaughn (2008) found that fluency continued to be poor for first graders who received continued daily small group intervention that was directed at teaching phonics and word recognition as well as fluency of word and text reading. O'Connor et al. (2005) followed a group of at risk kindergarten children during three tiers of school-based intervention until grade 3. After tier 3 intervention, 8% of the initial sample still failed the mastery criteria and qualified for even more specialized treatment. Overall, these studies suggest that it is difficult to successfully remediate the reading problems of all children who showed poor response to prior tier 2 intervention. Nevertheless, although not all tier 2 nonresponders may profit from tier 3 treatment, the majority of participants showed significant – albeit in some cases rather small – progress.

To our knowledge, there are no treatment studies (tier 3) conducted within the RTI framework in the Netherlands. However, two Dutch specialized clinical treatment methods have been well-described and evaluated. One is based on a psycholinguistic theory, and the other on a neuropsychological theory about the origins of subtypes of dyslexia.

The treatment method based on neuropsychological theory has been executed for more than fifteen years in several outpatient clinics, including the institute of the present study. The method has also gained international support outside The Netherlands (e.g. review Bakker, 2006). The treatment combines various reading activities based on the theory of the balance model which supposes that reading problems originate from an over-reliance on the right or left hemisphere (Bakker, 1990). One of the main activities is to read words presented on a computer screen with limited exposure time in the left visual half-field or right visual half-field to stimulate activity of the contra-lateral hemisphere.

The treatment method has been evaluated by Kappers (1997) and van Daal and Reitsma (1999). The study of Kappers (1997) showed that after weekly treatment for at least half a year overall reading fluency increased more during treatment than before treatment was given. However, there was large variability in response to treatment. The degree and rate of progress was related to the reading level at entrance and the rate of progress prior to treatment. After treatment, the rate of gain 'leveled off', but performance was maintained on the posttest level which was still quite low. Comparable to the findings of O'Connor et al. (2005), about 12% did not make any progress. Van Daal and Reitsma (1999) replicated the study but also differentiated between 'pure' dyslexics and children who combined reading disability with cognitive deficits or psychiatric symptoms. They concluded that both groups benefited from the treatment, but the children with pure dyslexia profited most. Neither group could catch up the reading deficit. Individual differences in treatment outcome were related to the absolute level of word reading and age at intake.

In addition, experimental studies were conducted to investigate the assumptions underlying the neuropsychological treatment method. Results suggest that progress in fluency is made regardless of locus of visual presentation (Berends & Reitsma, 2006; Dryer, Beale, & Lambert, 1999; Lorusso, Facoetti, & Molteni, 2004). Instead, effective components of the treatment may have been the reading of words with limited exposure duration (van den Bosch, van Bon & Schreuder, 1995; Das-Smaal, Klapwijk & van der Leij, 1996; Tressoldi, Vio, & Iozzino, 2007; Wentink, van Bon & Schreuder, 1997), or the repeated reading of words (Berends & Reitsma, 2005; Judica, De Luca, Spinelli, & Zoccolotti, 2002; Thaler, Ebner, Wimmer & Landerl, 2004).

In recent years, the psycholinguistic approach has received much support. The treatment method aims to remediate both reading and spelling difficulties by teaching step-by-step systematic knowledge of all linguistic elements and rules needed to transform a spoken word into a correct orthographic word form (e.g. Tijms, Hoeks, Paulussen-Hoogeboom, & Smolenaars, 2003; Tijms, 2007; Tijms, 2011). Studies of Tijms et al. (2003) and Tijms & Hoeks (2005) showed that the effects of treatment were most pronounced for reading accuracy in comparison with fluency. During the first six months of treatment most gains were on reading accuracy and spelling, however, reading rate appeared to improve during the second half of the treatment (Tijms, 2007). In a more recent study, Tijms (2011) found that treatment resulted in greater gains for the treatment group in comparison with a waiting list control group on measures of reading and spelling skills. Reading accuracy and spelling skills reached the mean normative level, indicating age-adequate skills. Although word and text reading fluency improved, it stayed behind corresponding with the lower bound of the normal range.

Although the Dutch treatments reported by Kappers (1997), van Daal and Reitsma (1999) and Tijms and his colleagues certainly qualify as specialized tier 3 interventions that differ from tier 2 interventions, the treatment methods were not evaluated within a RTI framework. That is, none of the studies took responsiveness to prior intervention into account. As a result, little is known about the way the reading problems have developed in the period before the children were referred to the clinical setting. It may be assumed that there were differences in referral procedures because the studies vary considerably in characteristics of the participants. In terms of norm scores, the samples of Kappers (1997) and van Daal and Reitsma (1999) contained children with more severe reading disabilities in comparison to Tijms and colleagues (on average in the 3^{rd} versus $10\text{-}12^{th}$ percentile respectively on a fluency of word reading tasks; see also van der Leij, 2006). Kappers reports that children were only accepted for treatment if they had received some kind of remedial help or special education earlier. In the first studies by Tijms and colleagues, children were referred to the institute by teachers, parents or psychological of educational services but there is no history reported. However, Tijms (2011) selected participants based on criteria for severe dyslexia in the Dutch health care system (Blomert, 2006). One important criteria is that special remedial teaching was already provided at school before treatment. Although this criterion limits the possible factor of poor education, poor education could have contributed to the poor reading abilities of the children in other studies. Moreover, the criterion does not ensure that children received remediation of equal quality. Moreover, for both treatment evaluations, progress was only evaluated in comparison to criteria derived from normative samples without the use of a control group design with randomised assignment to treatment conditions. As a consequence, no strong conclusions about treatment specific effects can be made. A comparison between effects of treatment methods across studies is complicated since it has been suggested that mixed results on effectiveness of treatments are related to differences in selection criteria (Lyon & Moats, 1997). Moreover, there may have been bias when a clinical treatment study is studied by researchers attached to the clinic. The present study was designed to overcome some of these limitations.

Present study

Considering the limited gains in word reading fluency reported by Kappers (1997), van Daal & Reitsma (1999) and Tijms et al. (2003), it is challenging to investigate whether there may be more effective ways to improve fluency in Dutch dyslexic children. As it has been argued that fluency skill develops as children start to recognize larger orthographic patterns within words (Share, 1995), a focus on the mapping of orthographic units to phonological units seems to be a promising method to enhance fluency. Recognition of orthographic patterns larger than the single grapheme is considered important for the development of fluent word and text reading. Instead of processing graphemes separately, recognizing graphemes as chunks is more efficient resulting in more fluent word recognition. Therefore, stimulation of fluency should involve training that emphasizes the recognition of orthographic patterns at a sublexical level. Indeed, treatment that involves training at both the word level and sublexical level has been found to be more effective than training at the word level alone (Berninger et al., 2000; Levy, Bourassa, & Horn, 1999; McCandliss, Beck, Sandak & Perfetti, 2003; Wolf & Bowers, 1999). Training with a focus on small units within words in contrast to whole words results in larger gains and transfer effects (Levy, et al., 1999). Although some studies have shown that training of recognition of orthographic patterns without explicit attention to phonology can be effective to enable children to draw analogies between words (Berends & Reitsma, 2007, Irausquin et al., 2007), training in which the connection between orthography and phonology is practiced explicitly is likely to be more effective (Bus & IJzendoorn, 1999; Hatcher, Hulme & Ellis, 1994; Marinus & de Jong, 2008; McCandliss, et al., 2003) in particular in reading unknown words (Harm, McCandliss & Seidenberg, 2003). In addition to the mapping of orthographic and phonological patterns at the sublexical level, treatment should include speeding up the process. Severe dyslexics experience difficulties in reading when they are forced to identify words, in particular pseudowords, under time pressure (Yap & van der Leij, 1993). It has been argued that such processing can be speeded up by presenting sublexical units with limited exposure duration (van den Bosch, et al., 1995; Das-Smaal, et al., 1996).

In sum, the present study was designed as an experimental study of the effect of a special tier 3 treatment on young Dutch children with severe and 'pure' dyslexia. First, to control for poor instruction at school in the selection of very poor readers, in the present study children who did not respond very well to a tier 2 intervention of individual remedial teaching at school reported by Struiksma, van der Leij and Stoel (2009), were selected to investigate the effects of a special treatment method in a reading clinic outside the school. As a result, only very poor readers with a well-documented history of intervention resistance qualified to join the study. Moreover, influence of educational deprivation was controlled for. Second, because we wanted to test our hypothesis that the experimental treatment would be more effective than other special treatments, it was decided to compare two treatment conditions. Participants were randomly assigned to the experimental treatment, or to the neuropsychological treatment described in the former section. The neuropsychological treatment was chosen because it has been successfully applied to children with severe dyslexia (see Bakker, 2006). Third, the experimental treatment focused primarily on fluency of sublexical processing and phonological recoding in order to stimulate fluency of word reading, independent of frequency or familiarity.

Method

Participants

During three consecutive years, 46 children were referred for treatment at the outpatient clinic for reading disorders. All participants were identified as non-responders to former intensive school-based intervention if their word reading fluency scores were still at or below the 10^{th} percentile based on norm scores, corresponding to a delay in reading of at leat 1 year (see also Scheltinga, van der Leij, & Struiksma, 2010). From the 46 referred children, the parents from 2 children did not agree on the conditions for participation (see below), and 2 children dropped out before finishing one or two full periods of treatment. Data of these children were not included in the present study resulting in a final sample of 42 children. All children were diagnosed as dyslexic at the Child Psychiatric Institute of Rotterdam by administration of a large battery of diagnostic tests including measures of intelligence and reading skills. Children were matched on outcomes of the reading and intelligence tasks. Children were matched to be comparable on reading skills and intelligence. Next, matched children were randomly assigned to one of two treatment conditions. The test results serve as participant characteristics at pretest (see table 1). Multivariate analyses of variance (MANOVA) were used to examine group means on the selection variables. Groups did not differ in age ($F(1,40) = .09$, *n.s.,* $\eta_p^2=.002$), word reading fluency ($F(1,40) = .23$, *n.s.,* $\eta_p^2=.006$), text reading fluency ($F(1,40) = .07$, *n.s.,* $\eta_p^2=.002$), Full Scale IQ ($F(1,40) = 1.94$, *n.s.,* $\eta_p^2=.046$), Verbal IQ ($F(1,40) = .32$, *n.s.,* $\eta_p^2=.008$) and spelling ($F(1,40)=.64$, *n.s.,* $\eta_p^2=.016$).

Table 1. Participant characteristics at intake

	Treatment condition					
Variable	SWRF (N=20)			HSS (N=22)		
	N	M	SD	N	M	SD
Gender ratio (boys: girls)	20	13 : 7		22	10 : 12	
Genetically at risk (yes: no)	20	12 : 8		22	16 : 6	
Age (in months)	20	106.6	(7.9)	22	105.8	(9.1)
Educational age (in months)	20	26.0	(6.3)	22	24.8	(5.6)
Full Scale IQ	20	93.4	(12.5)	22	98.4	(10.6)
Verbal IQ	20	94.1	(14.9)	22	96.2	(9.8)
Word reading fluency	20	27.7	(9.1)	22	26.4	(8.1)
Text reading	20	2.3	(1.2)	22	2.18	(1.7)
Spelling	20	40.8	(15.9)	22	37.3	(12.7)
Word Span	17	13.0	(2.7)	21	12.4	(2.8)
Digit Span	16	9.9	(.9)	20	9.8	(1.9)
Phoneme deletion	13	11.2	(2.7)	16	10.9	(2.0)
Phonological Synthesis	15	12.7	(2.3)	16	11.5	(2.9)

In addition, measures of memory and phonological awareness were administered to the majority of children. These data are provided to give a more comprehensive description of the group sample. A measure of word span from the Kaufmann Assessment Battery for Children was administered to 38 children. The digit span task from the same battery was administered to 36 children. Treatment groups did not differ on word span ($F(1,36)=.40$, *n.s.*, $\eta_p^2=.011$) or digit span ($F(1,34)=.07$, *n.s.*, $\eta_p^2=.002$). Measures of phonological awareness comprised tests of the FIK-2 testing battery (Aukes & Eg, 1993). One subtest involved phoneme deletion (n=29) and the other test involved phonological synthesis (n=31). Both subtests had a maximum score of 16. Treatment groups did not differ on phoneme deletion (F(1,27)=.11, *n.s.*, η_p^2=.004) and phonological synthesis (F(1,29)=.63, *n.s.*, η_p^2=.021).

From the record, information provided by parents on a written questionnaire was also collected. These data are used to describe the groups of participants in more detail. Parents were asked for the highest level of taken education as indication of SES. High education was indicated by university or college education, secondary vocational education was classified as average, primary school and low vocational training was classified as low. Education taken by the fathers ranged from low (n=18), average (n=16) to high (n=5) and was in three cases education not reported, education taken by the mothers ranged from low (n=16), average (n=23) to high (n=3). Moreover, parents reported whether dyslexia run in the family. The majority, i.e. 28 out of 42 parents, reported that other cases of dyslexia in the family were known.

Procedure

Children were selected after they had received an intensive school-based intervention delivered by remedial teachers. The remedial teachers were trained in a course that comprised 8 meetings that took place in the period September until end of May. In addition, remedial teachers were at least once observed at school during a session to ensure that the intervention was implemented consistently and to control for treatment fidelity (Troja, 1999). Based on outcome measures, teachers nominated poor responding children for referral to the clinical setting for treatment.

Preceding enrollment for treatment, a multidisciplinary team composed of a psychologist, psychiatrist and special educators at the Child Psychiatric Institute of Rotterdam assured the presence of dyslexia by administration of a large battery of diagnostic tests including measures of intelligence and reading skills (table 1). Diagnostic testing took place according to the standard procedure applied by the Child Psychiatric Institute. The measures at intake were included as learner characteristics to describe the different reader profiles. When dyslexia was diagnostically established, the children were subscribed for treatment in the clinic for reading disorders at the Centre for Child Studies (Child Psychiatric Institute Rotterdam). On average the time span between diagnostic testing and start of treatment lasted 12.24 (5.43) weeks, with a minimum of 2 and maximum of 28 weeks.

Following similar reasoning as Torgesen et al. (2001), we applied a two-group design. It was considered unethical to keep children identified with severe reading disorders from intensive reading instruction as would have been the case with a no-treatment control group. An alternative treatment or an a-specific placebo-treatment to control for effects of attention was also considered unethical, since it would have been at the expense of time focused on their primary reading difficulties. To enable examination of treatment effects, pre-treatment assessments served as baseline condition to compare with outcomes of treatment. Based on the available data from diagnostic testing, children were matched on reading level, educational age, spelling, IQ and mother tongue. Next, they were randomly assigned to one of two treatment groups. One of these groups received the neuropsychological method with Hemisphere Specific Stimulation (HSS), and the other group received a newly developed program called Sublexical Word Reading Fluency (SWRF). The HSS method served as a control condition to examine the additional effects of SWRF. Treatment conditions were comparable in time spent per session, guidance and instruction by professionals. Moreover, comparable sets of words were used for practice. Both treatment conditions focused on training word reading fluency without explicit instruction in spelling. Each treatment session took place weekly during 45 minutes. The methods are described in more detail below.

Professional therapists, i.e. psychologists, remedial educationalists or speech therapists carried out the treatments. Each child had its own therapist. The children were referred to other therapists only occasionally due to holidays, illness or changing time schedules. The therapists worked according to a protocol. The protocol was slightly changed once during the first year of study; the rate of meanwhile testing was increased from after every 10 to after every 9 treatment sessions. At testing sessions, progress in reading skills was evaluated through administration of standardized measures of text reading and word reading fluency. The standardized measures consisted of different versions that were used to avoid effects of repeated testing. The measures are described in detail below.

Since children all received the same intervention at school in the previous school year, the effects of past instruction were largely controlled. To minimize individual effects following from different concurrent interventions being used at school, children were not involved in specialized instruction at school during the treatment period. They took part in curriculum based reading administration. Clinicians kept the school informed on progress made in clinical settings by sending reports of evaluation each period. In addition, parents were involved in the treatment process. Parent support was provided through several consultations during which different topics were discussed.

Compliance, attrition and treatment duration

Before start of treatment, parents were pointed out that continuity of treatment was an important condition. Therefore, sessions should be skipped as few as possible. During summer holidays only, it was permitted to skip three sessions in a row. In addition, parents agreed on their child's participation during the complete period of 50 sessions. Occasionally, a session was skipped because of illness or other set backs. In some cases treatment was terminated earlier, because the requirement of continuity was not met by parents. In table 2 an overview of treatment compliance, the duration of a period in weeks and the number of treatment sessions per period is given. On average, treatment took place during 66.1 weeks (s.d. 15.09).

Table 2. Treatment compliance, duration and intensity

	Treatment condition									
	SWRF					HSS				
Period	N	Weeks		Intensity		N	Weeks		Intensity	
1	20	11.4	(1.4)	8.3	(1.3)	22	12.4	(2.1)	9.1	(.9)
2	20	13.9	(4.9)	8.5	(1.0)	22	12.5	(2.0)	9.1	(.7)
3	20	14.1	(4.3)	9.0	(1.3)	19	13.5	(2.2)	8.9	(.4)
4	17	12.8	(2.8)	8.8	(.8)	17	12.4	(2.3)	9.0	(.0)
5	15	12.7	(1.9)	8.5	(.7)	17	12.3	(3.4)	9.5	(2.8)
6	12	11.9	(1.2)	8.0	(1.7)	15	12.1	(2.6)	8.6	(.9)

Note. Weeks reflects the number of weeks between two evaluation time points; intensity reflects the number of treatment sessions per period.

Description of intervention and treatment

The school-based intervention served as criterion to select non-responders. Next, the children were assigned to one of the treatment methods which were based on different exercises in word reading fluency. In both treatment conditions, about 30 minutes per session were spent on exercises at the word reading level. The intervention and treatment methods combined word and text fluency practice as recommended by Lovett et al. (1994). In all conditions the same method for oral text reading practice was used. Details are described below.

School-based intervention. The intervention was based on prior research (Struiksma, 2003). The intervention comprised individual sessions of half an hour 4 times per week over a period of 20 weeks. Fluency was trained with a computerized program. Each session the child practiced in reading a set of 12 one-syllabic words accurately and as quickly as possible during 15 to 20 minutes. Words were repeatedly presented with limited exposure time. In accordance to performance, difficulty level increased over sessions by decreasing exposure time and increasing word complexity. Word complexity was increased by including words with consonant clusters and by decreasing orthographic overlaps between the presented words. The remaining time of a session was spent on reading connected text (see for a more detailed description Struiksma, van der Leij, & Stoel, 2009).

Hemisphere specific stimulation (HSS). This neuropsychological method (e.g. Kappers, 1997) has been widely used in the Dutch clinical setting and it was the standard treatment method in the outpatients' clinic for reading disorder at the Centre for Educational Services in Rotterdam where the study took place. This method served as a control condition to study the effects of a newly developed method described below. This clinical treatment uses integrated treatment methods of neuro-psychological and cognitive psychological origin (Kappers, 1997) and is based on the balance model (Bakker, 1990). According to this model, initial reading is predominantly mediated by the right hemisphere and gradually the left hemisphere becomes more involved. It is hypothesized that reading problems result from over-reliance on one of the cerebral hemispheres which is indicated by reading strategy. Over-reliance on the right hemisphere results in slow and effortful reading strategy, whereas over-reliance on the left hemisphere results in hasty and guessing reading strategy. The main principle is that the treatment combines various activities that aim to stimulate the left or right hemisphere. In each session, 20 minutes are spent on reading words presented with limited exposure duration on the computer screen using the computer program Hemstim (Moerland & Bakker, 1993). Dependent of the reading strategy of the child, words are presented in the right or left visual field. In addition, tactile exercises are included. Words are composed of plastic letters and fastened to the grooves of a board out of sight. Words have to be read aloud. The remaining time was spent on oral text reading as described below in more detail.

Sublexical Word Reading Fluency training (SWRF). The activities that formed the basis of the computerized experimental treatment were aimed at strengthening connections between orthography and phonology at the sublexical and lexical levels, which has been proven to be effective (Berninger et al., 2000; Bus & IJzendoorn, 1999; Harm, McCandliss & Seidenberg, 2003; McCandliss, Beck & Perfetti, 2003). Each computer session consisted of several exercises with a focus on the sublexical structure of words. A small set of 8 keywords was used each session. Each session was started with a pretest of reading the 8 keywords, 8 pseudowords that had overlap with the keywords and 8 pseudowords with no overlap. The keywords were first trained in six trials to ensure that the words were recognized by sight. Next, exercises at the sublexical level were based on these words. First, attention was focused on the position of letters within words by word building exercises (e.g. McCandliss et al., 2003). The child was required to replace one letter in the word by another that was provided on the screen by moving the letter with the mouse. From the 8 keywords an additional 16 words were constructed. The total of 24 minimally distinct words was used in the following exercises. Next, a sublexical feature was auditorily primed before visual presentation (Savage & Stuart, 1998). And last, the sublexical feature was presented within the word with limited exposure time to stimulate rapid recognition (e.g. Das-Smaal et al., 1996; van den Bosch et al., 1995). Each session was finished by reading the words from pretest, followed by training pseudowords with overlap in six trials. Exercises were presented in fixed order. The remaining time was spent on guided oral text reading.

Text reading fluency. In both treatment conditions, each session of about 15 minutes were spent on oral text reading practice. The child chose a book of interest at its instructional level which is the level just beyond mastery indicated by a 'sufficient' score on the text reading test. The therapist offered guidance using the method Pause Prompt Praise which is developed according to the behavioural approach (McNaughton, Glynn & Robinson, 1987; and see Struiksma, 2001 for a Dutch adaptation). While reading, the therapist uses praise as reinforcement for correct reading of a sentence or for self-corrections by the child; the therapist pauses to allow children to self-correct, encouraging independent reading behaviour; and the therapist gives prompts, code-oriented or meaning-oriented, to assist the child to self-correct and to focus attention on correct decoding. Moreover, to improve fluency, repeated readings of text passages were practiced both timed and untimed. If timed, the time needed was recorded and plotted in a graph to motivate the child by making progress visual.

Progress Measures

Word reading fluency. This was measured by presenting a word list of the Drie-Minuten-Test (DMT) [Three-Minute-Test] (Verhoeven, 1995). The DMT is a standardized Dutch test which comprises three cards of word lists of increasing difficulty. The first word list comprises five columns of 30 one-syllabic words of the type CV (koe), VC (uil) and CVC (pen). The second word list comprises five columns of 30 one-syllabic words of the type CCVC (spin), CVCC (bank), CCVCC (krant), CCCVC and CVCCC(C) (herfst). The third word list comprises four columns of 30 words with 2, 3 or 4 syllables. Children are instructed to read the words as accurate and as fast as possible within one minute. If the child hesitates more than 5 seconds, the teacher is allowed to read the word aloud stimulating the child to proceed reading. The score is the number of accurately words accurately read in one minute. The teacher uses a digital timer to record time. In addition, the proportion of accurately read words is computed. The manual reports the reliability for the separate word lists varying from .86 and .96 for different grades (Moelands, Kamphuis & Verhoeven, 2003).

Text Reading Fluency. This test (AVI; Visser et al., 1998). consists of nine cards, each with a written story (connected text). With each card, the text increases in difficulty in terms of word type and sentence length. Norms per card indicate a maximum number of mistakes and a maximum reading time, which determine the score 'good', 'sufficient' or 'insufficient'. The 'good' score implies that the child has mastered the level of difficulty. The 'sufficient' score implies that the child does not fully master the level of difficulty yet and needs more instruction, while the 'insufficient', score implies that the text is yet too difficult to read. Testing proceeds to a higher card number as long as the child reaches a 'sufficient' score and stops after a child reaches an 'insufficient' score on a card. The highest card number on which a 'good' score was reached is considered the text reading level. To enable comparison of gains in word reading fluency with text reading fluency, we transformed the scores in grade equivalents based on published norms for the DMT and AVI.

Data analysis

Data on the progress in fluency of reading words and texts were collected using standardized tests at the start, during and at the end of treatment for both treatment conditions. For analyses 5 or 6 evaluation measurements were included. The number of measurements depended on whether testing took place after 10 or 9 weeks (see procedure). Moreover, data on prior reading skill before treatment were available for analyses. Follow-up measurement took place on average a half-year after treatment had stopped. This resulted in a total number of 8 to 9 measurements per child that were included in data analyses. Separate multilevel analyses were run on sets of data of word and text reading measures, including all measurement time points to estimate overall growth.

Multilevel models are suitable for analyzing repeated measures or longitudinal data at fixed or varying occasions (Snijders & Bosker, 1999). Multilevel analysis has several advantages above multivariate analyses with repeated measures. First, multilevel modeling has a lower risk of capitalization on chance (type I errors) (Quené & van den Bergh, 2004). Second, multilevel techniques can handle missing data. Third, multilevel techniques can deal with measurements at varying time points. Fourth, regression is fitted with information from the individuals score and from all the members in the group. Finally, this approach enables to estimate individual growth curves in comparison to growth pattern of the entire group. The software MlWin for multilevel analysis was used (Rashbash et al., 2000). In the present study, a two-level hierarchical nesting can be defined with time-varying measurement occasions nested within subjects. In the next section, we elaborate on the models in detail.

Results

Progress on word reading fluency measures

First we calculated the proportion of words read accurately on the three DMT word reading fluency cards at the start of treatment. The number of incorrectly read words was subtracted from the total number of words read. At start of treatment the number of words read correctly was 91%, 87% and 77% for card 1, 2 and 3 respectively which confirms that high levels of accuracy are obtained in a transparent orthography even by poor readers (Landerl & Wimmer, 2008). In subsequent analyses, the number of words read correctly per minute was used as measure of word reading fluency for analyses. Since the correlation between scores on the three separate cards was high (correlation coefficients between cards varied from .77 to .81 at intake), we computed the mean number of words to obtain a single score for word reading fluency. In 86 out of 420 measurements data on card 3 were missing. These scores were estimated by a missing value analysis using a maximum likelihood estimation procedure. A summary of descriptive data on a selection of measurement points per treatment condition is given in table 3: at pretest of school intervention, at start of treatment and at 5th evaluation point. For data analyses 5 or 6 evaluation points were included.

Table 3. Progress on reading measures

		Treatment condition					
		SWRF			HSS		
Period	Variable	N	M	SD	N	M	SD
Pre-Intervention	DMT	20	14.59	6.87	22	14.31	7.50
	Text Fluency	11	.82	.75	14	.71	1.07
Start-Treatment	DMT	20	38.22	12.43	22	39.26	10.82
	Text Fluency	20	3.75	1.62	22	3.59	1.65
Period 5	DMT	15	51.18	16.03	17	48.31	11.77
	Text Fluency	15	6.07	2.19	17	5.35	1.41

Note. Word reading fluency DMT (Drie-Minuten-Test) mean score based on three separate word lists; Text Fluency, mastery level

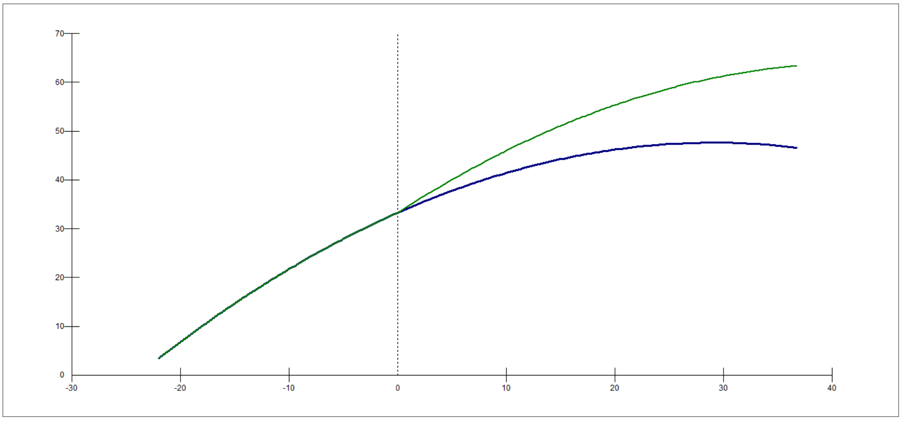
Next, multilevel analyses were run. Table 4 presents the results for the significant parameters in subsequent steps. The significance of each models' fit is determined by the difference in deviance between two models which has a chi-squared distribution with the difference in number of parameters as degrees of freedom (Hox, 2002). The first step concerned the estimation of a two-level intercept-only model, with an intercept and no explanatory variables. The intercept 38.07 in this two-level null-model reflects the overall mean of the word reading fluency score across the measurement occasions of all children. Next, the intercept and slope were estimated by entering time as predictor to the model to determine overall growth for the word reading outcome. The number of weeks between measurement occasions were counted and then divided by 4. Each point at the time scale reflects approximately one month. The time scale was centered at the start of treatment, i.e. the first measurement at intake for treatment. Models were tested with linear and curvilinear growth. In comparison with the null-model, inclusion of the linear and curvilinear parameters results in a model with better fit of the data (χ^2=668.60, df=2, p<.001). The intercept (34.36) now reflects the overall mean test score at start of treatment across all children. The fixed slope estimates a significant linear progress of 1.19 which indicates the progress in words read per minute per 4-week period. However, the progress levels off as indicated by the significant negative curvilinear variable. By adding the time variables to the fixed part of the model, the between occasions variance becomes 35.39 and at the student level, the variance is 97.28. This model is used as comparison for next models.

4. Summary of the model estimates for the two-level analyses of the word reading fluency scores.

	Model							
ter	0		1		2		3	
pt	38.065	(1.44)	34.360	(1.565)	34.268	(1.540)	33.270	(1.585)
near			1.185	(.031)	1.208	(.067)	.983	(.101)
urvilinear			-.010	(.002)	-.008	(.002)	-.017	(.004)
ent							.456	(.154)
n								
2								
t	64.832	(18.912)	97.280	(22.021)	97.040	(21.747)	98.126	(21.973)
					.168	(.041)	.170	(.041)
vilinear					.0001	(.000)	.0001	(.000)
pt*time					2.731	(.790)	2.787	(.799)
pt*timeCurvilinear					-.069	(.025)	-.074	(.026)
neCurvilinear					-.003	(.001)	-.003	(.001)
	212.103	(15.448)	35.391	(2.578)	15.478	(1.269)	15.24	(1.24)
ce (IGLS)	3492.17		2823.57		2627.26		2618.72	
			668.60		196.31		8.54	
			2		5		1	
			<.001		<.001		<.01	
nce model			0		1		2	

Vord reading fluency, mean number of words read correctly per minute; Time, 4-week periods; Treatment, 4-week periods from start
ent added as incremental factor.

Figure 1. Model estimation for the two-level analyses of the word reading fluency scores



Note. Horizontal axis reflects 4-week periods, centred around start of treatment.

After defining the growth in reading fluency over time, a number of explanatory variables were added to the fixed part of the model, in separate steps. To explore whether treatment effects were different for the two conditions, a dummy indicating treatment condition was entered. This variable did not reach significance and was not included in the model. Next, number of treatment sessions and IQ were added in subsequent steps. Both main effects as well as the interactions were included but they were not significant.

Text reading fluency

A similar procedure was followed for the text reading measure. Table 5 presents the results for the significant parameters in subsequent steps. The score on the text reading measure reflects a mastery level which ranges from 0 to 9. Measures were missing in 9.3% of the cases. However, multilevel analyses can handle missing data as noted earlier. The first step of analyses was construction of the intercept-only model (model 0). The average reading level of all children was 4.04.

Next, the time variable was entered as a linear trend. With inclusion of the time variable the model improved significantly (χ^2=532.36, df=1, p<.001). The intercept (3.15) reflects the estimated average reading level at start treatment. By adding the time variable to the fixed part of the model, the between occasions variance reduced with 80%. The curvilinear trend was also included in the model.

Growth leveled slightly off. To take into account that growth varies across children the time variable was allowed to vary randomly. The time variable varied across children. The curvilinear trend did not converge as a random factor. The covariance between time and intercept was significant (M=.023, *sd*=.007) indicating that a higher intercept was related to larger growth. Variance around the intercept and at the occasion level decreased (21% and 12% respectively). Overall, the model with the time variable was a significant improvement in comparison with the null-model (χ^2=30.86, df=2, p<.001).

This model (2) served as benchmark for the next model (3) in which treatment was included as an incremental factor. The effect of treatment was significant. Growth in reading level increased with .166 (*sd*=.031) each four-week period in addition to baseline growth of .083 (*sd*=.013). The model improved significantly (χ^2=27.16, df=1, p<.001). The treatment variable was not significant as a random factor.

Next, a number of explanatory variables were added to the fixed part of the model, in separate steps. Explanatory variables that were included comprised treatment condition, treatment intensity, and IQ. Treatment intensity did not converge. Moreover, not one of the factors was significant as fixed or random variable.

5. Summary of the model estimates for the two-level analyses of the text reading fluency scores

	Model							
ter	0		1		2		3	
pt	4.037	(.188)	3.150	(.222)	3.140	(.182)	2.776	(.193)
near			.169	(.006)	.169	(.007)	.083	(.017)
urvilinear			-.001	(.000)	-.001	(.000)	-.004	(.001)
ent							.166	(.031)
m								
2								
pt	1.048	(.325)	1.595	(.368)	1.264	(.299)	1.252	(.294)
					.001	(.000)	.001	(.000)
pt*time					.023	(.007)	.024	(.007)
1								
	3.867	(.297)	.792	(.061)	.695	(.057)	.648	(.053)
nce *(IGLS)*	1648.24		1115.88		1085.02		1057.86	
			532.36		30.86		27.16	
			1		2		1	
			<.001		<.001		<.001	
nce Model			0	1	1		2	

ext reading fluency, mastery level; Time, 4-week periods; Treatment, 4-week periods from start of treatment added as incremental fa

Word reading versus text reading fluency

Growth in word reading fluency was compared to growth in text reading fluency during treatment. Multivariate multilevel analyses were run on measurements of word and text reading from start until end of treatment. To enable comparison between the two different measures of reading, the raw scores were converted into norm scores. Norm scores reflect the educational age in months at which the reading score is obtained on average. At the lowest level, dummy variables were defined reflecting word reading level and text reading level. In the model the intercepts of both dummy variables and the slopes of the reading variables over time were added. The intercepts for word reading and text reading were 11.32 (.33) and 11.09 (.58) respectively, reflecting the average reading level in months at the start of treatment. The variable time x 'dummy word reading' reflected a progress of .21 (.03) during treatment. For text reading the progress was .54 (.06). The difference between the slope variables was significant (χ^2=26.40, df=1, *p*<.001). Text reading increased more than word reading. There was no significant curvilinear trend.

Gains in comparison with the norm group

We classified children as good versus poor responders based on scores at the final evaluation moment of clinical treatment. For each treatment condition, we determined the number of children, who performed still within the lowest 10th percentile according to norm scores, indicating poor response. For the HSS condition, the word reading fluency scores of 18 out of 22 children (82%) still fell within the lowest 10th percentile. For the SWRF condition, 13 out of 20 children (65%) were classified within the lowest 10th percentile. There was no difference between treatment conditions (χ^2=1.53, df=1, *n.s.*). For text reading fluency, 6 (27.3%) and 8 (40%) children in the HSS and SWRF condition respectively obtained reading scores within the lowest 10th percentile. The number of children in the lowest 10th percentile did not differ per treatment condition (χ^2=.76, df=1, *n.s.*).

At follow-up, we determined whether children who obtained reading scores above the 10th percentile directly after treatment were able to keep up their reading level. Retest scores were available on 36 cases, 21 and 15 in the conditions HSS and SWRF respectively. Follow-up testing took place after an average of 36.4 weeks (s.d.10.3). For the HSS condition, the word reading fluency scores of 16 out of 21 children (76%) fell within the lowest 10th percentile. At follow-up testing, one child moved from above the 10th percentile to below that criterion, but the word reading fluency scores of two children further improved above the criterion. For the SWRF condition, 10 out of 15 children (67%) were classified within the lowest 10th percentile at follow-up. One child was not able to sustain reading level above the 10th percentile. There was no difference between treatment conditions (χ^2=1.45, df=1, *n.s.*). For text reading fluency, 15 (71%) and 8 (53%) children in the HSS and SWRF condition respectively obtained reading scores within the lowest 10th percentile. The number of children in the lowest 10th percentile did not differ per treatment condition (χ^2=.61, df=1, *n.s.*).

Discussion

The present study examined progress on measures of reading fluency in children with persistent dyslexia within a multi-tiered RTI approach. We investigated whether a lengthy, intensive treatment focusing on sublexical orthographic structure of words and phonological recoding by mapping phonology to orthography would add to progress after no or poor response to school-based tier 2 intervention. The effects of this experimental method were examined in comparison with a traditionally applied method.

Overall, results indicate that continued specialized treatment in a clinical setting contributed to overall growth on measures of word reading fluency and text reading fluency. Larger effects were expected for the training that focused on the mapping of orthographic units to phonological units since the recognition of orthographic units larger than the single grapheme is considered important for the development of fluent word and text reading. For the present study, we argued that the explicit focus on orthographic units linked to phonology might result in larger gains and transfer effects (e.g. Levy, et al., 1999; Bus & Ijzendoorn, 1999). Contrary to our expectations, the sublexical treatment condition did not explain additional variance in gains on word reading fluency; both treatment conditions resulted in similar gains.

Accordingly, Wanzek and Vaughn (2008) suggested that there is no evidence to prefer one method over the other as long as methods are focused, explicit, systematic and intensive, although not all instructional components are equally effective (Swanson & Hoskyn, 1998). The present study design did not allow exploring the effectiveness of separate components of both treatments. However, both treatment conditions in the present study included components that likely contributed to treatment effect, such as repeated reading of words, use of computer, providing feedback, intensive and lengthy one-to-one tutoring and controlling task difficulty in text reading practice (e.g. Shaywitz, Morris, & Shaywitz, 2008). Moreover, assisted-reading with feedback, setting realistic targets for each treatment period and providing rewards in both conditions were likely effective components resulting in gains in reading.

In addition, our results showed that children made relatively more progress on standardized measures of text reading fluency than word reading fluency. In contrast, Torgesen et al. (2001) found that children made more gains in word reading efficiency than in text reading fluency after intervention. These contrasting findings could be related to differences in task requirements. In the study by Torgesen et al. children were required to read *and* comprehend the story during testing. This might have slowed down the reading speed since attention was explicitly paid to comprehension of the text. The standardized measure used in our study required the accurate and fluent reading of a passage without explicit instruction for comprehension. However, language skills including comprehension may have contributed to the difference between word and text reading in our study. Possible explanations are postulated by interactive theories of reading processes (e.g. Stanovich, 1984). It has been suggested that individuals who experience reading difficulties use an interactive reading process that is more affected by context (Corkett & Parrila, 2008). By using contextual information, dyslexic children may compensate for their difficulties with word recognition (Nation & Snowling, 1998). Accordingly, Alexander and Slinger-Constant (2004) argue that children with strong cognitive and semantic abilities can offset the severity of decoding deficit in contextualized text reading. They may benefit from context when decoding unknown words and they may exhibit greater strength in comprehension than would be expected based on their word reading skills. These benefits possibly occur only after a certain level of text reading fluency is obtained (Burns et al, 2011). Our results showed that growth in text reading fluency was related to the initial text reading level. Although speculative, children in this study who were better at reading texts fluently at the start could possibly make more use of compensating language skills that helped them to improve their text reading fluency.

Although children showed overall gains that resulted in more age-appropriate reading level for some children, progress was limited, in particular for word reading fluency. One possible explanation could be that the standardized reading tasks used to measure progress were not sensitive enough to detect specific learning effects since these tasks can be considered ‘far-transfer’ measures. That is, the words of the standardized tasks showed overlap with the words with regard to trained sublexical orthographic patterns, but the words were not identical to the trained words and contained additional sublexical complexities. Besides the difference between trained word material and the tested word material, the task demands were different. During training the words were presented in isolation on the computer screen. Progress was assessed by presenting words in lists. Martin-Chang and Levy (2006) showed that transfer after training is dependent on the congruency between processes employed during training and testing which is referred to as transfer appropriate processing.

On the other hand, the finding that only a small number of children improved to a more age-appropriate level converges with the results reported by others indicating that in particular fluency is hard to improve in children who responded poorly to prior interventions (e.g. O’Connor et al., 2002; Vaughn & Wanzek, 2008; Torgesen et al. 2001). Kappers (1997) studied the treatment method that was identical to the control condition in our study. Kappers reported that word reading fluency progressed on average 2.4 words in a period of 8 weeks. On text reading, children made a steady growth of 0.5 levels per assessment with more progress on text reading than on word reading. Progress decreased over time, with -0.036 and -0.019 for word and text reading respectively. In our study, there was a steady growth of 1.44 words on word reading and of 0.25 levels on text reading per 4 week period during treatment, with a curvilinear trend of -0.017 for word reading. It should be noted that in our study growth was estimated over a longer time period with the intercept set at the initial time point whereas Kappers estimated growth during treatment with time centered around the mean number of assessments. Although the differences in analyses estimating the curvilinear growth complicate a direct comparison between studies, the results are similar, showing limited gains. In contrast to the Kappers’ study, children in this study were selected after no or poor response to an intensive school-based intervention, ensuring the severity and persistency of the reading problems. Children were only selected if their reading scores fell within the lowest decile according to norm scores. These children were very poor readers in comparison to age peers who had followed the same though less intensive reading education. Taking into account that stringent criteria were used, it might not be surprising that treatment gains were not sufficient for catching up with reading levels of age peers.

Considering the stringent criteria for participation and the severity of de reading difficulties, one might argue that treatment should have been more intensive to be more effective. In the present study, children attended the reading clinic once per week for 45 minutes. Harn, Linan-Thompson and Roberts (2008) showed that increasing remediation time per session (30 vs 60 minutes) had a significant impact on reading outcomes. Although treatment on a more frequent basis seems preferable, this was difficult to realize within the current research setting since children had to attend the clinic. Since children with reading problems are not likely enjoying reading, they tend to get less reading practice. Torgesen et al. (2001) argued that children with reading fluency problems need more reading practice to close the gap in reading acquisition. One way to increase reading practice would have been to provide children with homework. Tressoldi (2007) combined treatment with homework assignments. In the study by Tijms (2011), participants had to practice at home three times a week for 15 minutes. Royer and Walles (2008) reported encouraging results on fluency training after a period in which children practiced at home daily. Although in the present study children were encouraged to read at home as well, they were not provided with homework.

It has also been suggested that adjustments of intervention content in response to children' needs might be more influential than intensifying intervention (Scarborough, 2005). Accordingly, Chard et al. (2003) concluded that (text) fluency appears to develop more quickly if the difficulty level of text is adjusted as children progress. In the present study, the children practiced reading text according to their nearby (or instructional) reading level. However, most time was spent on practice of word reading fluency. Although the content of word reading practices corresponded to the level of school-based intervention on which children experienced sustained problems, all children practiced with the same materials at the same pace in both conditions as a consequence of the quasi-experimental design of the study. If treatment materials would be adjusted to the individual needs through a process of dynamic assessment during practice, gains might have been larger. Our results showed that initial reading level was related to growth during intervention and treatment. Therefore, some of the children would have benefited from more intensive guidance, i.e. in duration, homework, et cetera.

Limitations of the study

Some limitations of the study should be mentioned. Since it was considered unethical to keep children identified with severe reading problems from treatment, a no-treatment control group was not included. Therefore, the effectiveness of treatment versus no treatment could not be determined. However, studies have shown that the gap between good and poor readers is likely to become larger (Torgesen, 2000) unless intervention has been provided. Therefore, it is likely that the reading delay of the children under study would have increased when no treatment had taken place. Another comment can be made on the sample size over all treatment sessions. Although parents and children were encouraged to continue to participate the full period of 50 sessions, not all children completed the 50 treatment sessions and some resigned after insufficient periods of treatment. There were various reasons for earlier termination. Most commonly, treatment was terminated if children had obtained a mastery level on text reading that was considered sufficient to cope with extended instruction within the class room setting. In a few cases, treatment was terminated because parents were not able to take care of continuity in attending the clinic. A last critical comment could be made on the evidence of treatment fidelity which was not quantified. Professional therapists delivered the treatment following a protocol. Moreover, by using a computer program, treatment delivery was largely standardized. However, no observations or monitoring were carried out.

Summary and implications

In spite of the limitations, this study adds to a growing number of studies conducted within the RTI approach which has not been studied in the Netherlands before. Longitudinal quasi-experimental training studies including an experimental and control condition are relatively rare. Studies often include only one treatment condition, using a baseline or standard scores to determine the effects. In addition, effects are often measured over a short period of time including a pre- and post-test. Longitudinal studies require both effort and time from researchers and participants. It appears difficult to prevent preliminary dropout of participants, to collect data on exact same time points. By using techniques like multilevel analyses, it was possible to follow children over different periods of time with varying measurement occasions. The present study shows that supplemental tier 3 remediation of severe reading problems results in small improvements of both word and text reading fluency. It should be noted that children improved there text reading score from approximately level 3 to level 6 during training. These levels indicate increasing complexity and correspond to norm levels. In general, level 6 is considered a functional reading level. That is, children are able to read written information in daily use. It is a basic level that enables children to read average texts independently, which can be considered an important outcome. Furthermore, the finding of only small improvements may have been partly the result of stringent selection criteria, taking response to tier 2 intervention into account. Participants had severe and persistent reading difficulties. Since tier 3 remediation in the clinical setting is time-consuming and costly, it is important to consider carefully which children are in need of this professional help. The RTI framework seems an effective approach to select those children who are in need of specialized remediation. Additionally it is important to note that continued support and guidance will still be necessary to prevent further increase of reading delay in comparison to age peers. Support should also be directed towards compensating and coping strategies that help children to deal with their reading difficulties in their further academic achievement.

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CHAPTER 5

FOCUS ON SUBLEXICAL FEATURES TO IMPROVE WORD READING FLUENCY IN SEVERE DYSLEXIA: TRAINING, TRANSFER AND GENERALIZATION

Abstract

The objective of this study was to examine the effect of a word fluency training in detail. The word fluency training focused on practice of orthographic patterns, i.e. consonant clusters and open versus closed syllables. It was argued that knowledge of orthographic patterns would support the recognition of words and new (pseudo)words. Children practiced their word reading skills with a computer program that consisted of various exercises. All children were selected to be very poor readers. They had shown no or limited progress after lengthy, intensive school-based intervention and were referred to the clinical setting for treatment. The word training in the present study was part of n experimental treatment condition. The aim of a detailed, in-depth study was to examine the learning and transfer effects during word fluency training. The training lasted 50 sessions. Results show that continued training results in further improvement of word reading accuracy and fluency, but this was in particular the case for trained words. There was transfer to new words, but only limited.

Background

Failure to acquire rapid context-free word recognition skills is one of the most reliable indices of severe dyslexia across orthographies (Lovett, et al., 1990; Torgesen, 2000). Poor reading fluency forms the most salient and persistent deficit in children with dyslexia learning to read a transparent orthography whereas high levels of reading accuracy can be obtained (e.g. de Jong & van der Leij, 2003; Landerl & Wimmer, 2008; Zoccolotti et al., 1999). This is the case with Dutch which has relatively many consistent grapheme-phoneme correspondences. Apart from some specific difficulties of Dutch orthography, children with dyslexia learn to read relatively accurate but with a slow reading rate. This slow rate in word reading is assumed to hinder fluent reading of text and ultimately reading comprehension (e.g. Lai, Benjamin, Schwanenfugel & Kuhn, 2014). Ample studies were aimed at improving the reading skills of children with dyslexia. The majority of studies have focused on the improvement of accuracy in decoding skills, but focus has shifted to the improvement of reading fluency as well. Studies have reported moderate effects of fluency training (see for reviews Chard, Vaughn & Tyler, 2003; Kuhn & Stahl, 2003; Therrien, 2004). Gains in reading rate contrary to reading accuracy appear more difficult to establish through intervention (Torgesen, et al., 2001), in particular with hard-to-remediate or severe cases of dyslexia. Effortful word recognition processes put constraints on the attention resources that are required for other aspects of reading comprehension. Consequently, the deficit in word reading fluency likely affects the academic success during educational age and later on.

In order to become a fluent reader, the recognition of words should be fast and effortless. Reading fluency has a multidimensional nature with various contributing subskills (Katzir et al., 2006). Both lexical and sublexical processes are involved in reading fluency (Kame'enui & Simmons, 2001). It has been suggested that reading fluency needs automated processing at the level of phonology, orthography and semantics (Perfetti, 1992). In normal development, children already make use of both sublexical and lexical representations from the early start of reading (Burani et al., 2002). As children learn to read, they learn to map the phonological representations of words to the written orthographic patterns. Cuetos et al. (2009) showed that children make use of orthographic units larger than single graphemes as indicated by more accurate and rapid reading of pseudowords if these are composed of familiar groups of letters. Through reading, children build orthographic representations. As more words are known, word representations with similar spellings and pronunciation are clustered in the lexicon, which enables the use of regularities in orthographic patterns, facilitating automatic word recognition and nonword reading (Harm, McCandliss & Seidenberg, 2003). The development of orthographic knowledge is a factor related to reading rate (Cunningham, Perry & Stanovich, 2001; Hagiliassis, Pratt, & Johnston, 2006; Olson, Forsberg & Wise, 1994). Recognition of orthographic patterns larger than the single phoneme enables efficient and fluent word recognition (Bowers, Sunseth, Golden, 1999; Cuetos & Suarez-Coalla, 2009; Rey, 1998; Ziegler & Goswami, 2003). Skilled fluent readers make use of small and large orthographic units in parallel (Peereman, Brand & Rey, 2006; Ziegler & Perry, 1998).

Orthographic depth influences the process of decoding (Seymour et al., 2003; Ziegler & Goswami, 2003). In a relatively transparent orthography as Dutch, learning to read starts with mapping sounds to letters, i.e. alphabetic decoding which is an effective reading strategy for accurate word recognition. Although, it is possible to recognize words by decoding single letters, readers in transparent orthographies also seem to benefit from using sublexical units larger than letters (Burani, Marcolini & Stella, 2002; Cuetos et al., 2009; Pagliuca & Monaghan, 2010). De Jong (2006) suggested that skilled readers in a transparent language also read by using orthographic units larger than a single grapheme to enable more rapid word processing. This might be especially true for certain linguistic complexities. Also in transparent orthographies, the linguistic structure has an impact on the reading process of children learning to read (Duncun, Colé, Seymour & Magnan, 2006; Søvik, Samuelsteun & Svarva, 1996). For example, developing young readers are able to read monosyllabic words adequately, but they are less efficient at decoding monosyllabic words with consonant clusters and polysyllabic words (Verhoeven & van Leeuwe, 2009). It has been suggested that word recognition becomes more efficient with the recognition of letter patterns such as consonant clusters and syllables (Verhoeven & Perfetti, 2003).

In contrast to normal readers, it has been suggested that children with dyslexia make use of non-componential, holistic representations (Harm, McCandliss & Seidenberg, 2003) which hamper fast and effortless access to word representations (Perfetti, 1992). For transparent orthographies, it has been suggested that children with dyslexia remain depending on decoding as a dominant strategy whereas normal readers make use of both sublexical and lexical strategies (Davies, Rodriguez-Ferreiro, Suárez & Cuetos, 2012). This appears from word length effects that are more prominent in dyslexia than in normal reading (Van der Leij & van Daal, 1999; Zoccolotti, et al., 2005; Martens & De Jong, 2008). Moreover, efficiently processing of letter sequences as a larger orthographic unit appears troublesome for children with dyslexia (Di Filippo, De Luca, Judica, Spinelli & Zoccolotti, 2006; Seymour et al., 2003; Struiksma, 2003; Ziegler & Goswami, 2003). Struiksma (2003; Struiksma, van der Leij & Stoel, 2009) showed that children with dyslexia experience continuing difficulties with reading words with consonant clusters (see also: Jimenez et al., 2007; Magnan & Biancheri, 2001; Thaler et al., 2004; 2009). In addition, polysyllabic words with contextual dependent vowel sounds appear difficult (Steenbeek, et al., 2013; Treiman, et al., 2006) because contextual information is needed to decode the vowel accurately. Multisyllabic words are usually not read by sight but require sublexical processing (Spinelli et al., 2005). In spite of these findings, also dyslexics seem able to make use of orthographic representations (Barca, Burani, Di Filippo & Zoccolotti, 2006). In the present study, we aimed to examine whether the word reading fluency of very poor readers could be improved by training, focusing on (sub)lexical word characteristics or orthographic patterns within words.

Several methods have been used to improve fluent word recognition through training. As repeated reading has been shown to be effective in improving reading fluency (Kuhn & Stahl, 2003; Therrien, 2004), this method has also been adopted to improve single word reading. When a child reads words repeatedly, mapping phonology to orthography, representations are formed in the orthographic lexicon (e.g. Landi, Perfetti, Bolger, Dunlap & Foorman, 2006; Perfetti, 1992; Share, 1995). Likewise, representations of re-occurring letter combinations are formed. Training with repeated reading of words has been shown to be effective for dyslexics, but it might lead to word specific learning (Berends & Reitsma, 2006). In contrast to training in whole word reading, transfer might be obtained after training with explicit focus on orthographic patterns (Benson, Lovett & Kroeber, 1997). Levy, Bourassa and Horn (1999) concluded that poor readers showed weakest generalization following training with whole words in comparison with segmental word training. Teaching children the mapping between phonology and orthography at the sublexical level could enable them to transfer this knowledge to unknown words or pseudowords (Harm et al., 2003). Training can focus on different sizes of orthographic units. The units used might determine the effectiveness of training. Jiménez et al. (2007) studied the effectiveness of training different spelling-to-sound units (phonemes, syllables, onset-rime and whole words) in relatively transparent orthography of Spanish. Training of both phonemes and syllables resulted in improved word and pseudoword reading.

Studies have explored the effectiveness of training with focus on orthographic characteristics. These studies differ in duration, the type of orthographic units trained and type of training and exercises given. McCandliss, Beck, Sandak & Perfetti (2003) examined an intervention called Word Building. The intervention aimed drawing attention to the letter-sound correspondences and the position of graphemes within words. Exercises consisted of making words by changing single graphemes, requiring more fully engagement in the reading process. The experimental group obtained larger gains in comparison with the control group.

Conrad and Levy (2011) also suggested that drawing attention to the orthographic commonalities within words supports the reading acquisition of new words. Moreover, they argued that practice with words sharing orthographic patterns may result in orthographic awareness, making children notice orthographic consistencies. In their training study, words were presented together in blocks with shared orthographic patterns. Two- and three letter patterns at the beginning, middle and end of words were practiced with repeated reading. The training resulted in gains for reading the trained words, but generalization to new words with trained patterns was limited. This is consistent with the findings by others (Berends & Reitsma, 2006; Thaler, Ebner, Wimmer, & Landerl, 2004). Thaler et al. (2004) trained onset consonant clusters within a limited set of words by repeated reading. After a great amount of repetitions, the reading speed of the trained words improved but it was not yet age adequate. Moreover, generalization to untrained words with similar onset consonant clusters was only small.

It has been suggested that children with reading difficulties may need more explicit instruction (Shaywitz et al., 2008), probably also with respect to instruction in the recognition of orthographic patterns within words (Conrad & Levy, 2011). Benson et al. (1997) concluded that explicit instruction on orthographic patterns in comparison with reading words resulted in larger transfer effects to new words with trained patterns. Accordingly, Berends and Reitsma (2007) found transfer effects to new words after training words with focusing on orthography. Their training focused on drawing analogies between words with shared orthographic patterns of consonant clusters. After training focused on orthographic characteristics of the words resulted in generalization to untrained words with shared orthographic patterns.

Hintikka et al. (2008) assessed the effects of training German sublexical patterns (onset consonant clusters as *schl)* in a computer game compared to general book reading. The children that trained with the sublexical patterns improved their reading of these patterns more than the comparison group. However, there were no large generalization effects to words including the trained sublexical units. Marinus, de Jong and van der Leij (2012) also evaluated repeated reading with focus on four consonant clusters. Training of consonant clusters resulted in an improvement of consonant cluster naming speed. However, explicit training of high frequent consonant clusters did not result in substantive gains in word reading speed after a training. They suggest that a more promising orthographic training unit might be the syllable or morpheme.

Indeed, several studies have found promising results for training syllables (Tressoldi, Vio, Iozzino, 2007; Wentink, van Bon & Schreuder, 1997; Ecalle, Kleinsz & Magnan, 2013; Huemer, Aro, Landerl & Lyytinen, 2010; Heikkilä, Aro, Närhi, Westerholm & Ahonen, 2013). For example, Ecalle, Kleinsz and Magnan (2013) explored the benefits of a grapho-syllabic training with a grapho-phonemic computerized training in French. Poor readers practiced speeded recognition of syllables at different word positions. Grapho-syllabic training contributed more to word reading after training than the grapho-phonemic training. They showed positive effects on silent word recognition, word reading aloud and reading comprehension after a training focused on syllables. Heikkilä et al (2013) also concluded that poor readers showed gains in reading speed after syllable training. However, gains were again specific for trained syllables, contrary to findings by Huemer et al. (2010) who found a transfer effect to pseudowords.

In general, it appears challenging to obtain transfer to untrained materials. Previous studies were sometimes training studies of relatively short duration with limited sets of words and/or orthographic patterns. Berends and Reitsma (2007) suggested that training effects might be enlarged when training takes place with groups of similar words instead of single words. Instead of focusing on one size of orthographic units, it might be important to practice at different levels of orthographic presentations, i.e. at letter, multiletter patterns, syllables and words to increase orthographic awareness (Conrad & Levy, 2011). In addition, training should provide sufficient repetition and practice Reitsma, 1997).

Present study

The present study was an in-depth study taking a closer look at the effect of an experimental computerized training program. The training was part of a longitudinal remediation study (see Chapter 4). We aimed to examine the progress in reading rate and accuracy scores during training sessions. The training focused on strengthening the connections between orthography and phonology at the sublexical and lexical levels as shown to be effective (Bus & IJzendoorn, 1999; Castles, Coltheart, Wilson, Valpied, & Wedgewood, 2009; Ecalle et al., 2013; Harm, McCandliss & Seidenberg, 2003; McCandliss, Beck & Perfetti, 2003). Exercises at the sublexical level were based on core words that were trained first. As these words could be recognized by sight, it was suggested that it would support the recognition of new words with overlap at the sublexical level (Struiksma, 2003). To obtain transfer to new and, ultimo, unrelated words, instruction was not limited to whole words, but also directed attention to segmentation skills (Benson et al., 1997; Lovett, Barron & Benson, 2003), focusing on orthographic patterns. Both monosyllabic words with consonant clusters as polysyllabic words were included.

The study distinguished from other studies as it was long in duration. A greater amount of repetition may promote the retention and generalization of the training effects (Lemoine et al., 1993) especially in the case of dyslexia (Thaler et al., 2004). Moreover, trained material contained large amounts of variants of two kinds of orthographic complexities and sublexical units were trained in several word contexts. The study was quasi-experimental; it took place in the clinical setting as a field trial to investigate its contribution for the practical implementation.

Method

Participants

All 21 participants were identified as non-responders to a former intensive school-based intervention. The reading difficulties were considered persistent if their word reading fluency scores were still at or below the 10^{th} percentile based on norm scores after intensive intervention. They were referred for experimental treatment at the outpatient clinic for reading disorders. All children were diagnosed as dyslexic at the Child Psychiatric Institute of Rotterdam. School-based intervention took place in grade 2, treatment started in grade 3. Mean age was 8 years and 9 months (s.d. 7.5 months). On average 42.43 sessions (10.44) were completed.

Procedure

The experimental study was part of a larger intervention study that investigated the effects of clinical treatment within a RTI framework (see for description Scheltinga, Struiksma & van der Leij, 2010). Children were referred for treatment within the clinical setting after they had shown poor response to an intensive school-based intervention. Teachers nominated the children for referral to the clinical setting for treatment. Preceding enrollment for treatment, a multidisciplinary team composed of a psychologist, psychiatrist and special educators at the Child Psychiatric Institute of Rotterdam assured the presence of dyslexia by administration of a large battery of diagnostic tests. When dyslexia was diagnostically established, the children were subscribed for treatment in the clinic for reading disorders at the Centre of Child Studies (Pedologisch Institute Rotterdam). Children were randomly assigned to one of two treatment methods of which one, the newly developed program Sublexical Word Reading Fluency was focus of the present study. The treatment method is described in more detail below.

Professional therapists, i.e., psychologists, remedial educationalists or speech therapists carried out the treatment. Each child trained with one own therapist during the full period. Therapists only changed occasionally due to holidays, illness of changing schedules. The first author instructed the therapists on using the program and written instructions were available. The activities within each treatment session were conducted in vast order. See for detailed description below.

Compliance, attrition and treatment duration

Before start of treatment, parents were pointed out that continuity of treatment was an important condition for inclusion. It was emphasized that sessions should be skipped as few as possible. During summer holidays only, it was permitted to skip three sessions in a row. In addition, parents agreed on their child's participation during the complete period of 50 sessions. Occasionally, a session was skipped because of illness or other setbacks. In some cases, treatment was terminated earlier, because the requirement of continuity could not be observed.

Description of the program, word material and treatment sessions

For training during treatment, a computer program was developed. The computer program included 50 sessions. Each week, practise took place with one session. Children practised words with consonant clusters during the first 20 session, which were ordered randomly. Next, 30 sessions were used to practise open and closed syllables. Each session comprised several computer-based exercises, which were presented in vast order.

Word material. For each session 8 keywords for training, 8 transfer words and 8 generalization words were chosen. Exercises were based on the keywords. For the first twenty sessions, fifty-three keywords were chosen to represent Dutch consonant clusters. Twenty-five words contained a consonant cluster at the beginning of the word; twenty-eight words contained a consonant cluster at the end of a word. Each keyword was used 3 to 4 times over sessions. The same keyword was never used two sessions in a row.

For the last 30 sessions, keywords were chosen to present open vs. closed syllables. The difficulty of open vs. closed syllables in Dutch is determined by a change in the pronunciation of the vowel, which is presented by the same grapheme. The vowel is tense when it is followed by a single consonant; the vowel is lax when it is followed by double consonants. For example, the word *bomen* is pronounced with a tense vowel, while the word *bommen* is pronounced with a lax vowel, i.e., /bɔ:mən/ versus /bɔmən/ respectively. Words were chosen with letters presenting different vowels in the context of closed vs. open syllables. For the sessions 21 to 30, 8 words were selected to make 4 minimal pairs of open and closed syllables. Pairs like bomen-bommen were selected. In each session two different vowels were used to construct 2 pairs of 2 minimal pairs. In the sessions 31-50, vowels were presented in different consonantal contexts. Again, pairs of -CCVC and -CVC were held constant but the onset varied (ex. ballen - delen and maten - letten). In addition, a list of transfer words, corresponding in trained sublexical structure (i.e., identical consonant clusters of syllable structure) was composed. These words were pseudowords. To evaluate generalization, each session words with untrained sublexical units were automatically selected. See table 1 for example lists of used keywords and transfer words.

Table 1. Example of session material

Key words	Transfer words	Generalization words
Session 1-20		
Dorp	Karp	Kals
Worm	Garm	Guts
Zalm	Zilm	Dint
Berg	Norg	Nust
Park	Durk	Jaks
Pink	Vank	Kisp
Melk	Rolk	Meft
Session 21-30		
Dekken	Jekkel	Heffer
Deken	Jekel	Hefer
Wetten	Geter	Rebel
Weten	Getter	Rebel
Zonnen	Monner	Hubber
Zonen	Moner	Hober
Koppen	Joppel	Dosser
Kopen	Jopel	Dosel
Session 31-50		
Remmen	Demmel	Dezzel
Bomen	Gomer	Tozel
Repen	Feper	Jeser
Doppen	Foppel	Wossel
Wedden	Gedder	Beppel
Daden	Padel	Table
Delen	Zeler	Rezel
Ballen	Haller	Jazzel

Exercises per training session.

Training keywords. Next the pupil had to read all keywords repeatedly in six trials for training of rapid recognition. Each word was presented once per trial. Presentation of words was similar to the pre-test. The pupil was instructed to read the words as fast and accurate as possible. The tutor scored correct and incorrect on the keyboard. After the pupil had named the word, the written form was generated together with the spoken form. In the left bottom of the screen a column per trial was filled with yellow blocks for each word read correctly so the pupil could see how many words were read correctly.

Word building. The next exercise was aimed at drawing attention to sub-lexical units within words (according to McCandliss et al., 2003). The pupil had to make minimal changes in words by moving letters on the screen. Starting point was a keyword from which one letter had to be changed by a new letter that was provided on the screen. The letter that had to be changed within the word turned grey and the pupil had to drag the new letter into the word. The pupil read the word and the tutor scored correct or incorrect. From 4 keywords 8 new words, most real words and some pseudowords, were formed. After building 12 words, the words were presented in a list and the child had to read the list as quickly and accurately as possible. The same exercise of building and reading was done with the remaining 4 key words. Both *keywords* and newly made words from the word building exercise, resulting in 24 words, were used for subsequent exercises.

Priming task. The representation of the sub-lexical structures was aimed to stabilize by providing both auditory and orthographic information in a priming task (Savage, et al, 1998). The pupil heard the pronunciation of the sublexical pattern followed by presentation of the written word. For practise with consonant clusters, the pronunciation of the cluster was provided. For practise with open and closed syllables, the pronunciation of the tense or lax vowel preceded the written presentation. The pupil named the word. The tutor scored correct or incorrect. Feedback was provided on the screen. In the left bottom of the screen one column is filled with yellow blocks for each word read correctly with a maximum of 24. In addition, spoken feedback was generated on the computer. The spoken form of the sub-lexical unit and the whole word were generated together with the written word form 750 ms after the pupil named the word.

Speeded reading. Reading with limited exposure time was trained to speed up word recognition (Bar-Kochva & Hasselhorn, 2015; van den Bosch, Bon & Schreuder, 1995; Das-Smaal, Klapwijk & van der Leij, 1996). Again the focus was on parts of the word. For words with consonant clusters the cluster was flashed on the screen during 200, 500 or 800 ms.. For words with open vs. closed syllables, the single or double consonant following the vowel was flashed. The fixed part of the word was presented on the screen during one second. After the flashed presentation, the complete word disappeared from the screen. Presentation duration of the flashed part was lengthened or shortened according to performance. Duration increased when more than 3 out of last 5 words were read incorrectly.

Dependent measures

Training, transfer and generalization. Each session, progress in word recognition was measured on reading keywords, transfer words and generalization words at pre- and post-test. Each session started with a pre-test during which the pupil had to read all 8 keywords, transfer words and generalization words. After practice the pupil read the same words again in the post-test. The post-test was identical to the pre-test. Words appeared in random order in the middle of the screen during 2500 ms.. The pupil was instructed to read the word before it disappeared from the screen. The pupil had to read the word aloud. Reaction time was recorded by voice key. The pupil was instructed to speak loudly and clearly. When the voice key detected sound, a picture of an ear appeared in the right down corner of the screen. The distance between the pupil and microphone was adjustable in case of false or no registration. The tutor scored correct or incorrect by pressing a key on the keyboard. The computer generated the correctly spoken word as feedback. The spoken word was generated 750 ms after the pupil named the word or after the word disappeared from the screen. Both accuracy and latency scores were registered per word.

Data analysis

For analyses, we used multilevel models to enable analyses of repeated measures at fixed or varying occasions (Snijders & Bosker, 1999). In comparison to multivariate analyses with repeated measures, multilevel modeling has a lower risk of capitalization on chance (Quené & van den Bergh, 2007). Moreover, multilevel techniques can deal with missing data and measurements at varying time points. The multilevel approach also enables to estimate individual growth curves in comparison to growth pattern of the entire group. The software MlWin for Multilevel analysis, version 2.31, was used (Rashbash, et al., 1999).

In the present study, a two-level hierarchical nesting is defined with sessions nested within subjects. Separate multilevel analyses were separately run on the data from the first 20 and remaining 30 sessions. For each pretest and post-test per session, we calculated the mean response time in milliseconds for correctly read keywords, pseudowords with shared and orthographic pattern and pseudowords with non-shared orthographic pattern. The dependent measures were the average response time per word type at pretest and posttest and the sum of accuracy scores. The multilevel model was scaled in such a way that the intercept reflects the response time at the first session of each training phase. An overview of parameters is given in table 2.

Table 2. Overview on coding explanatory parameters

Parameter	Explanation
Session	Scaled at first of session for each training phase
SessionCurvilinear	Quadratic function of the session variable
Within-session	Pre-test (coded 0) versus posttest (coded 1)
Orthographic pattern	Consonant cluster: session 1-20; syllables: session 21-50
Consonant cluster	Cluster at beginning of word (coded 0); Cluster at ending (coded 1)
Syllable	Closed (coded 0) versus open (coded 1)
Pseudowords	Keywords function as reference category (coded zero)
TrainedPs	Dummy variable: Pseudowords with shared orthographic pattern (coded 1)
UntrainedPs	Dummy variable: Pseudowords without shared orthographic pattern (coded 1)

Multilevel analyses were run by including the parameters step-by-step to the fixed part of the intercept-only model. At the first step, a time parameter (session), linear and curvilinear, was added to the fixed model. The time parameter was also added to the random part of the model to allow variance across children. At the next steps, explanatory parameters were added to the fixed part of the model. To examine learning effect within sessions, a dummy variable was constructed reflecting pretest (coded zero) versus posttest (coded 1). The influence of orthographic pattern was explored by adding a dummy variable for consonant cluster (session 1-20) and syllable (session 21-50) to the model. At pretest and posttest, pseudowords were used to control for transfer to new words. We defined two dummy variables that were entered as a set of variables with keywords defined as reference category. The dummy trainedPs refers to the pseudowords with the trained orthographic pattern. In other words, these pseudowords show overlap (cluster or syllable) with the trained keywords. The dummy untrainedPs refers to the pseudowords that have no overlap with the trained keywords in that specific session. In addition, interaction terms were constructed to explore transfer across and within sessions for the word categories. For example, a positive interaction between trainedPs and within-session would indicate that knowledge from trained keywords is used in reading new words.

At each step with the inclusion of new parameters, we determined if the model fit improved. The significance of each models' fit was determined by the difference in deviance between two models which has a chi-squared distribution with the difference in number of parameters as degrees of freedom (Hox, 2002). In addition, a parameter was only maintained in the model if it was significant.

Results

The results are presented in two sections. In the first section the results on the first 20 training sessions, the gains in reading words with consonant clusters are reported. In the second section, the results on the last 30 sessions, gains in reading words with open vs. closed syllables, are reported. The main sections are further divided in subsections to report on results on accuracy and latency scores for trained keywords, transfer words and generalization words.

Progress on reading words with consonant clusters

Response times. The analyses on the response times were based on correct and valid answers. Response times were considered invalid when the response time was below 500 msec. or 2500 msec., indicating that the voice key had been triggered by another sound or was not set off properly. On the first 20 sessions 23% of the answers were invalid at pretest and posttest.

Multilevel analyses were run. The final model is given in table 3. The intercept reflects the average response time at the first session (M=1337.236). The parameter session was included as predictor to the model estimating the decrease in response times over the first 20 sessions. Including session only to the fixed part resulted in a significant mean decrease of -3.561 (sd=.0957). Session was also included as a curvilinear parameter but it reached no significance and was not retained in the model. To take into account that the response times over sessions can vary across children, we allowed the session parameter to vary randomly across children resulting in a full multivariate model with regard to repeated measures (Snijders & Bosker, 1999). The fixed parameter was no longer significant, but the variance was significant and the model improved. A number of dummy coded explanatory variables were included in the model. The parameter pretest-posttest indicates an average decrease in response time (-58.297) within a session which indicates a short-term learning effect. Words with a consonant cluster at the end of the word were read faster than words with a cluster at the beginning (-44.144). Overall, pseudowords were read with slower rate. This is indicated by the parameters untrainedPs (240.401) en trainedPs (227.976). To check for transfer and generalization effects within and across sessions, we added interaction terms. The interactions between word category and session were not significant. With keywords as reference category, both categories of pseudowords showed poor progress within sessions from pretest to posttest. Interactions between within-session and word categories were significant. The significant decrease in average reaction time from pretest to posttest is mainly explained by an effect for trained real words.

Accuracy scores. The sum of correctly read words per word category at pretest and posttest was taken as dependent measure. Multilevel analyses were run by including the parameters step-by-step to the model. We followed the same procedure as described above. Table 3 presents the results for the parameters included in the final model.

The intercept indicates that the average accuracy scores were already high at the start of the training. On average 6.889 words were read correctly. The parameter session estimates the increase in accuracy scores over the first 20 sessions. There was a small but significant main effect of session. Accuracy scores improved with .029 (sd=.012) each session. The session parameter was allowed to vary randomly across children. The significant covariance between intercept and session indicates that the children with high accuracy scores at the beginning made less progress. This is likely due to a ceiling effect. Session was also included as a curvilinear parameter but it reached no significance and was not retained in the model. The parameter pretest-posttest was significant indicating a learning effect. In addition, more errors were made at reading words with a cluster at the end (-.106). Overall, more errors were made at reading pseudowords in comparison to real words, reflected by the significant negative parameters for trainedPs and untrainedPs. However, across sessions, children improved their pseudoword reading indicated by the significant interactions between session and word categories trainedPs and untrainedPs. This effect was small. On average the accuracy score improved at posttest after practice (sd=.378). The learning effect within session was mostly accounted for by reading key words, indicated by the negative interactions between within-session and word categories. However, the interaction between within-session and trainedPs was not significant.

Table 3. Results on session 1-20

	Response times		Accuracy scores	
Parameter				
Fixed				
Intercept	1337.236	(49.929)	6.889	(.250)
Session	-4.235	(2.351)	.029	(.012)
Session Curvilinear				
Within-session	-58.297	(15.889)	.378	(.086)
Cluster	-44.144	(9.657)	-.106	(.052)
TrainedPs	227.976	(15.986)	-1.298	(.131)
UntrainedPs	240.401	(16.150)	-1.427	(.134)
Session*trainedPS			.022	(.011)
Session*untrainedPs			.023	(.011)
Within-session*trainedPs	48.974	(22.552)	-.210	(.121)
Within-session*untrainedPs	53.201	(22.788)	-.466	(.122)
Random				
Level 2				
$\sigma^2_{intercept}$	47863.906	(15275.682)	1.111	(.357)
$\sigma^2_{session}$	101.276	(35.647)	.002	(.001)
$\sigma^2_{intercept*session}$			-.028	(.013)
Level 1				
σ^2_{e}	52119.930	(1510.834)	1.531	(.0

Progress on reading multisyllabic words

Response times. The analyses on the response times were based on correct and valid answers. Response times were considered invalid when the response time was below 500 msec. or 2500 msec., indicating that the voice key had been triggered by another sound or was not set off properly. On the last 30 sessions 17.8% of the answers were invalid at pretest and posttest.

We made a distinction between three word categories that were further divided according to orthographic pattern. Each session consisted of eight keywords were real words with open and closed syllables, eight pseudowords that showed overlap with the keywords and eight keywords that showed less overlap. The average response times were calculated for each category and within each category for open versus closed syllables. The average response time per word type at pretest and posttest was taken as dependent measure. The multilevel model was scaled in such a way that the intercept reflects the response time at the first session of this part of the training. The first session was coded zero. Multilevel analyses were run following the same procedure as described above. The parameters were added to the model step-by-step. Table 4 presents the results for the parameters included in the final model.

Table 4. Results on session 21-50

	Response times		Accuracy scores	
Parameter				
Fixed				
Intercept	1635.086	(36.144)	2.975	(.147)
Session	-9.034	(1.967)	.044	(.007)
Session Curvilinear			.001	(.000)
Within-session	-45.322	(14.363)	.034	(.041)
Syllable			-.107	(.024)
TrainedPs	65.057	(22.294)	-.415	(.041)
UntrainedPs	30.282	(22.906)	-.388	(.042)
Session*trainedPS	1.828	(1.256)		
Session*untrainedPs	3.159	(1.281)		
Within-session*trainedPs	47.815	(20.979)	-.138	(.058)
Within-session*untrainedPs	60.157	(21.397)	-.188	(.060)
Random				
Level 2				
$\sigma^2_{intercept}$	21404.807	(7212.703)	.394	(.128)
$\sigma^2_{session}$	52.272	(20.093)	.0004	(.0001)
$\sigma^2_{intercept*session}$				
Level 1				
σ^2_{e}	91272.273	(1873.172)	.786	(.0

The intercept reflects the average response time at the first session (1601.466). Including session to the fixed part resulted in a significant decrease of reaction time over session. On average, reaction time decreased with -9.034 (sd=1.967) per session. Session was also included as a curvilinear parameter but it reached no significance and was not retained in the model. To take into account that the response times over sessions can vary across children, we allowed the session parameter to vary randomly across children. The covariance with the intercept was not significant. There was no effect for syllable (open versus closed). Overall, pseudowords were read with slower rate, indicated by the parameters trainedPs and untrainedPs. Across sessions, the reading of pseudowords did not improve in comparison with the real keywords. Within sessions, there was an overall decrease of average reaction times (-45.221). The reading rate improved within sessions. This effect was accounted for by the trained real keywords.

Accuracy scores. The sum of correctly read words for three categories, divided into open and closed words, at pretest and posttest was taken as dependent measure. The maximum sum score was 4. Table 4 presents the results for the parameters included in the final model.

The intercept indicates that the average accuracy scores were already high at the start. On average 2.975 (sd=1.147) were read correctly per word category. There was a small but significant main effect of session. Accuracy scores improved with .0443 (sd=.007) each session. The curvilinear parameter session was also significant. The session parameter was allowed to vary randomly across children. There was a significant variance, but the covariance with intercept was deleted from the model. Overall, more errors were made at reading words with an open syllable (-.107). This was also the case with reading pseudowords as indicated by the parameters trainedPs and untrainedPs (-.42 and -.39, respectively). There was no significant interaction between session and word categories. The parameter within session was included to check for a learning effect. With the inclusion of the interactions between within-session and word categories, the main effect was no longer significant, but the parameters were retained in the model since it improved overall. Within session, the accuracy scores did not improve but showed a decline.

Discussion

The present study was an in-depth study to examine the specific effects of a word reading fluency training. The training was part of a longitudinal remediation study. The training focused on repeated reading of words focusing on orthographic patterns. Words with consonant clusters and two-syllable words were included. We investigated the overall effects on training measures of word reading reaction times and accuracy scores.

Overall, reading rates increased over sessions. Per session the progress appears small (-4.24 for clusters and -9.034 for multisyllabic words). However for reading multisyllabic words, the estimated progress in reading rate is 270 msec. over 30 sessions. The reaction time at the start decreases with 16%. The improvement in reading words for consonant clusters was smaller. Reaction times were already lower at the start, so there might have been on average less room for improvement. The improvement in reading rate was similar for reading words and pseudowords with consonant clusters over sessions. In general, pseudowords were read with lower reading rate and this did not improve relatively more than reading real words over sessions. Pronounced difficulties with reading pseudowords in children with dyslexia have been reported by other (e.g. van Daal, & van der Leij, 1999; Hermann, Matyas, & Pratt, 2006; Verhoeven & Keuning, 2014). Teaching children the mapping between phonology and orthography at the sublexical level did not enable them to read pseudowords as quickly as real words.

For accuracy outcomes, it must be noted that words were already read relatively accurately. This was the case for both training phases. This is in line with studies that have shown that even children with dyslexia can obtain reasonable levels of accurate reading (e.g. de Jong & van der Leij, 2003; Landerl & Wimmer, 2008; Zoccolotti et al., 1999). The accuracy scores improved across sessions for cluster words and two-syllabic words. Small progress was made on reading cluster words, with poor initial readers making larger gains. In general, pseudowords were read more poorly but accuracy scores improved slightly more than for real words. This was the case for pseudowords with as well trained as untrained consonant clusters.

It should be noted that during the first 20 sessions all Dutch consonant clusters were trained. Those pseudowords with untrained consonant clusters had a consonant cluster that was not trained within the particular session, but it was trained later on. Therefore, progress indicates that training helped children to improve decoding skills at reading unknown words. Over sessions, this could also be the result of repeated practice. Reitsma (1997) showed that the number of repetitions of orthographically similar words is related to the number of new words that are read correctly.

Interestingly, words with consonant cluster at the beginning of a word were read more accurately, but slower. Difficulties with final consonant clusters have been reported by others, in particular for spelling (e.g. Van Bon & Uit de Haag, 1997; Treiman, Zukowski, Richmond-Welty, 1995). In our data, reaction times reflect the start of reading aloud. A consonant cluster at the beginning of a word slows down the start of reading, but this seems to support accurate decoding. Children started reading the words with consonant clusters faster, but made more errors. Davies et al. (2012) argued that a word's pronunciation might not be fully prepared at the response onset. They showed that children with dyslexia take longer to prepare and read aloud words accurately as indicated by measures of reaction time and reaction duration. In our study, children had to read words as quickly and adequately as possible at pretest and posttest. This might have hindered them at decoding reading words with final consonant clusters.On the other hand, although speculative, it seems to indicate that they did not make use of orthographic patterns but were reading letter-by-letter.

Since transfer effects across sessions were difficult to determine as all orthographic patterns were practiced during the period of treatment, learning or transfer effects were examined by analyzing progress within sessions. Within sessions, there was progress in reading rate and accuracy scores for cluster words. The effect was largest for real words and only small for pseudowords. Accuracy at posttest improved relatively more for pseudowords with trained clusters (.168) in comparison with untrained clusters. This indicated a small transfer effect from trained words to similar untrained pseudowords within a session. For reading two-syllabic words, we only found an effect for reaction times but this effect was specific for trained real keywords.

Overall, the training resulted in gains for both word reading rate and word reading accuracy, but transfer as measured within sessions was limited. This was especially the case for two-syllable words. Also other training studies reported limited generalization to new words (e.g. Thaler et al. 2004). However, several studies reported promising results for syllable training (e.g. Ecalle, et al, 2013; Heikkillä et al., 2013; Hintikka et al., 2008; Huemer et al, 2010; Tressoldi et al., 2007; Wentink et al., 1997), although the size of training effect varied. One explanation for our findings could be that the chosen type of two-syllable words (open vs. closed syllables) is less suitable as orthographic training unit in Dutch. The syllabic words that were trained in the present study can be considered opaque instead of transparent. In order to read words of the type CVCVC with a single vowel in the first syllable (e.g. /o/ in /bomen/), one must be aware of the orthographic rule which determines that the vowel has a long (or tensed) pronunciation within this context. The training studies in for example Finnish, French, German and Italian used transparent syllables that could be used as an unit for rapid recognition. In those studies, the trained syllables could be considered as 'fixed' orthographic patterns.

One study in Dutch by Wentink et al. (1997) reported an increase in the speed of reading words after flash card practice with pseudowords of different length. The authors argue that children acquired syllable-bound processes after practice as shown by transfer to standardized measures of word reading. Taking a closer look at the used word material, it shows that only consonant clusters and words with a fully transparent pronunciation were chosen for practice. With respect to the words that were trained in the present study, explicit instruction in orthographic rules (Tijms, 2011) might have been more effective. In future studies, the selection of trained units should be considered more carefully, taking into account the linguistic structure of the language which has an impact on the reading process of children learning to read (Duncan, Colé, Seymour & Magnan, 2006; Søvik, Samuelsteun & Svarva, 1996).

Although the word structure CVCVC might be less suited as a sublexical unit for training of syllable recognition, children increased their reading speed of trained real words across and within sessions. This might be an effect of repeated exposure and practice. This is in line with the finding by Steenbeek-Planting et al. (2013). They found improved reading accuracy and speed after repeated reading of bisyllabic words, including those with context-dependent vowel pronunciation. Moreover, they argued that feedback about failures might help in particular poorest readers to improve their reading scores. In our study, the training program generated feedback with repeated reading practice.

Moreover, similarly to the study by Steenbeek-Planting et al., words were trained in mixed sets. By contrasting two-syllable words with CVCCVC and CVCVC words, we expected to draw attention to the orthographic patterns within words. The different exercises were aimed at increasing the awareness or sensitivity for mappings between orthography and phonology. However, our data do not demonstrate that these exercises enabled children to grasp the orthographic consistency (or rule) since the accuracy scores only improved for trained keywords. Conrad and Levy (2011) argued that practice with words sharing orthographic patterns may result in orthographic awareness, making children notice orthographic consistencies. This might be more efficient to teach children to draw analogies.

The finding of a small transfer effect of trained cluster words should be interpreted with caution. From our data, it is not possible to conclude that children used the consonant cluster as an orthographic unit for rapid word recognition. Marinus et al. (2012) demonstrated that after training with a small set of frequent consonant clusters, fast recognition of isolated consonant clusters improved but it did not result in substantive gains in word reading. Moreover, training of consonant clusters did not yield different results than letter training. Our study was longer of duration and was not limited to a set of four consonant clusters. Nevertheless, it could be the case that children simply became more efficient at letter decoding.

An overall limitation of the present study was that we did not include a control group without or with alternative practice. Comparisons are therefore only possible across and within sessions. The progress across sessions merely reflects reading improvement during the training period. Since we included all possible letter combinations, drawing conclusions about transfer effects across sessions is limited. Related to this issue, it should be emphasized that this study is different from other training studies because it was part of a clinical treatment study. The training replaced the standard or regular treatment that functioned as the control condition (see chapter 4). Consequently, this has put some practical and ethical constraints on the experimental set-up for the training.

The main overall purpose of the training under study was to investigate the remediation of reading fluency in a clinical group of children. In this chapter, we described an in-depth study supplemental to the previous study (chapter 4) to examine the learning effect of word reading fluency training. We hypothesized that training with a focus on orthographic pattern would help children to improve their word reading skills. It should be kept in mind that participants had severe and persistent reading difficulties. All children had already received an intensive school-based intervention. Although gains and transfer were small, it appeared possible to increase word reading speed by training.

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CHAPTER 6

EPILOGUE

Epilogue

The main focus of the four studies in this thesis was on the remediation of word reading fluency deficits in dyslexia. In addition, we examined whether reading related cognitive skills are typical for dyslexia (chapter 2) and predictive of response to intervention (chapter 3). In separate studies we reported the results of a school-based intervention (chapter 3) and of outpatient treatment in a clinical setting (chapter 4 and 5). The intervention and treatment were part of a response to intervention framework, respectively at tier 2 and tier 3. After one year of formal education, poor readers enrolled an intensive school-based intervention in grade 2. The intervention focused on improving word reading fluency using a computer program that systematically offered word reading practice. Moreover, the children practiced their text reading fluency through guided text book reading.

The school-based intervention was effective for some but not all children. The so-called non-responders were referred to the clinical setting for intensive, specialized treatment (tier 3). Overall, degree of responsiveness to intervention and treatment varied substantially among children. This variation was only partially related to initial reading level and underlying reading related skills as rapid naming. Children who received treatment after school-based intervention continued to improve their reading skills, with relatively better gains for text reading than for word reading. In general, gains seemed to be limited. Considering that only severe cases of dyslexia were selected for treatment, the finding that they benefited from treatment is rather positive. In this final chapter we will discuss some additional issues that were only briefly mentioned in the previous chapters.

Gains but no full remedy

From the studies described in chapter 3 and 4, we concluded that poor readers can benefit from school-based intervention and outpatient treatment. Those who did not or did only partially profit from intervention made gains later on following continued specialized treatment. The reading scores were compared with norm scores to consider the extent of normalization. Regarding norm scores, the reading scores of some children did not longer fell within the lowest 10th percentile. This was particularly true for text reading scores. That is, growth was most clearly shown in text reading rate. Although a control group was lacking, it is likely that reading delay in comparison to age-related norm groups would have been increased without continued treatment (e.g. Tijms, 2011). Notably, on average children improved their text reading skills to a functional reading level.

This level is minimally needed to access printed and written information for daily use within relevant contexts. It is a basic reading level that enables children to read average texts independently. It has been suggested that a certain level of text reading fluency is necessary to benefit from further practice (Burns et al., 2011). The obtained level might enable children to improve their reading skills further within the educational setting with sufficient practice and support.

In both studies, we examined the progress made after intervention and treatment. Retention scores were collected in both studies. Five months after the school-based intervention was terminated, most children had retained their reading level, with a few exceptions making additional improvements. Similar findings were reported at follow-up, 36 weeks, after treatment. Most children were able to sustain their reading skills at the norm level, albeit additional growth did not occur. In our studies we did not check what type of guidance was offered after intervention or treatment, but it has been suggested that support and guidance is often absent in the higher grades of elementary schools (Snow & Moje, 2010). This is somewhat surprising because reading skills are still developing in later grades (Verhoeven & van Leeuwe, 2009) and practice and support should also be provided to older pupils. Of course, this is particularly the case for children with reading difficulties since they need more repetition and practice. Within the Dutch RTI model described by Struiksma and Rurup (2008), the importance of ongoing support for children during and after treatment is described. Tier 3, corresponding to level four in the Dutch model (specialized treatment in the clinical setting; see introduction), should include collaboration between the clinical institute and the school sharing information about progress and coaching of children with severe dyslexia. With regard to our findings, it can also be concluded that ongoing support and guidance is needed to maintain the reading level. Since reading difficulties are not likely to be fully resolved by treatment, it is important to support children with dyslexia to further develop their reading skills afterwards at school. In general, the practitioner from the clinical institute should provide advice to teachers at school. Knowledge about how to support and stimulate the reading development must be carried over. After treatment, children with dyslexia should be provided with sufficient opportunities for practice with feedback. Moreover, as children grow older they should learn how to use compensatory strategies to overcome their decoding deficit (e.g. Nation & Snowling, 1998).

To maintain and to improve reading skills, self-initiated reading or print exposure is important (Chard, Vaughn, & Tyler, 2002; Guthrie, Schafer, & Huang, 2001). After a certain level of text reading fluency is obtained (Burns et al, 2011), children may compensate for their difficulties with word recognition by using contextual information (Nation & Snowling, 1998). This helps children to make further gains similar to self-teaching processes (Share, 1995). Sufficient practice with familiar texts will improve reading fluency and increase reading vocabulary (Torgesen et al., 2001). Indeed, leisure time reading has been shown to be related to further improvement of reading skills. However, children with reading difficulties are less likely to read on their own (Mol & Bus, 2011). Poor readers with a long history of difficulties and negative experiences are more likely develop an aversion and negative emotions towards reading. They may perceive reading as a threatening activity (Thielen, Mol, Sikkema-de Jong & Bus, 2015). Therefore, tasks to stimulate reading motivation with appropriate support are important, also in later grades (Guthrie, et al. 2006; Snow & Moje, 2010). Several instructional practices have been suggested to increase motivation for reading (e.g. Guthrie, Wigfield, Metsala & Cox, 1999; Thielen, et al., 2015). These practices include 1) affording own choices for reading, 2) adjustment to own topic interests, 3) extrinsic rewards and praise, 4) cooperative learning, and 5) teacher involvement, i.e. care for progress and support. Moreover, the use of e-books and other technologies can support poor readers during reading. Several text-to-speech software packages are available (Callebaut, 2004). The use of this software can be helpful for children with severe dyslexia to overcome severe decoding deficits during reading comprehension activities. Meanwhile, they get feedback about the correspondences between written and spoken language which in turn supports the further development of reading skills.

In addition, development of adaptive coping strategies help them to be successful later in life despite their learning disabilities (Navalany, Carawan, & Renwick, 2010). It is important that children learn to take control of their situation. Firth, Frydenburg, Steeg and Bond (2013) examined the effect of a coping strategies program *Success and Dyslexia*. The program concentrates on assertion skills to fulfill needs, ask for support if needed and on active strategies and problem solving by working directly on the problem rather than worrying about or ignoring of them.

In the methods for intervention (chapter 3) and treatment (chapter 4), principles of applied behavior analyses were incorporated. The aim was changing (reading) behavior through behavior modification techniques. For example, the remedial teacher at school learned about recognizing behavior to avoid reading and stimulating on task behavior. In addition, guidance was offered using the adjusted version of the method Pause, Prompt and Praise (e.g. Struiksma, 2001) to reinforce correct reading, stimulate self-correction and encourage independent reading behavior. More general coping strategies were not explicitly part of intervention or treatment, although the professional practitioners paid attention to for example inter-personal relationships, autonomy and problem solving. We suggest that attention for coping both during as well as after treatment could be an important addition to current educational practices.

Individual or small-group intervention within the school setting

In chapter 3 we found that responsiveness was only partially predicted by initial word reading level and rapid naming skill. Based on these findings, we suggested that response to intervention is supported as method to identify children with severe reading difficulties. Moreover, RTI can be used to rule out inadequate instruction as a cause of reading difficulties. Thereby it responds to an important aspect of the definition of dyslexia that the reading disability should not be caused by a lack of effective classroom instruction. The reading disability should be persistent despite specific intervention. Another important aim of RTI is the prevention of reading difficulties. Provision of early intervention prevents that at risk children fall farther behind their peers.

It is important to note that the school-based intervention described in chapter 3 was very intensive. It took place four times a week for 30 minutes during 20 weeks in grade 2. The intervention was implemented by a remedial teacher who received ongoing training during the intervention period. The intervention required an investment of 40 hours for individual training with an expert tutor. In recent years, schools were confronted with lower budgets and downsizing. As a consequence, schools are not always able to employ specialized personnel, such as remedial teachers. In the Dutch educational RTI framework (see general introduction), schools are required to offer intensive intervention. High-quality, intensive intervention should precede the identification of severe reading difficulties for referral to diagnostic practice. Schools often mention that is difficult to organize one-to-one tutoring on an almost daily basis. They prefer interventions that do no not require individual teaching (Begeny & Silber, 2006).

It has been argued that small-group interventions (usually ranging from two to four children) can be equally effective (Alexander & Slinger-Constant, 2004; Begeny, Krouse, Ross, & Mitchell, 2009; Elbaum, Vaughn, Hughes, & Moody, 2000; Helf, Cooke, & Flowers, 2009). Grouping practices with more proficient readers guiding less able readers has also been shown to be effective to build fluency (Chard et al., 2002). A combination of both group-based fluency training and individual instruction has also been suggested (Hatcher, Hulme, et al., 2006). Moreover, teachers but also highly trained volunteers can effectively implement interventions (e.g. Blok, Oostdam & Boendemaker, 2012; Elbaum et al., 2000; Zijlstra, 2015). Training assistants and/or the use of scripted intervention procedures may solve some of the bottlenecks in organizing intensive interventions for children with reading disabilities within the school setting. These possibilities merit further research. The effectiveness of interventions that are more time-efficient without loss of quality should be subject of future research.

Finally, we would like to stress that prevention of reading problems is likely more cost-effective than 'wait-to-fail'. It is well known that without intervention the reading delay of poor readers increases in comparison with age peers (e.g. Tijms, 2011; Stanovich, 1986). In older struggling readers, it may become more challenging to repair the large reading delay. Moreover, children often build negative experiences with reading if they don't receive support. This can be prevented by early intervention. In our studies, children enrolled school-based intervention in second grade. Recent studies (Regtvoort et al, 2013; Zijlstra, 2015) suggest that intervention starting earlier (late kindergarten or beginning of grade 1) may prevent the development of reading difficulties in a substantial group of at risk children. The intervention should be of long duration, continuing in second grade. The possibilities of time-efficient interventions starting at young age and prolonging during subsequent grades should be explored within the educational system.

One size fits all? Taking into account individual differences

Intervention (chapter 3) as well as treatment (chapter 4 and 5) used standardized, replicable procedures. The main focus of the intervention and the treatment was on improvement of word reading fluency. All children practiced with the same word material at similar pace. During intervention, the number of training trials within a session was adjusted to the needs of each individual child. This means that the training was ended once the child had read eight out of ten words correctly and within the maximum response time on three consecutive trials. However, all children progressed through the lessons at the same pace. This means that flexibility in instruction was limited. On the other hand, children practiced text reading fluency according to their nearby reading level. This is in line with the suggestion that text reading fluency develops more quickly if the difficulty is adjusted to children's developmental level (Chard et al., 2002).

Our findings demonstrated variation in gains made by children. We suggested that gains might have been larger if the content would have been adjusted to the individual instructional needs. This has been referred to as the zone of proximal development to maximize learning (Vygotsky, 1978). It reflects the level which is not mastered yet, but can be achieved with sufficient instruction and practice. It can be said that during intervention and treatment, children practiced their text reading fluency within this zone of proximal development. After each treatment period, text reading proficiency was assessed. If a child had improved reading fluency, he was allowed to choose a new reading book at a higher level. However, growth during word training was not used to adjust the program to an advanced level.

Future research could examine the effects of integrating RTI approaches with dynamic assessment (DA) procedures (e.g. Gustafson, Svensson, & Fälth, 2014). DA is supposed to assess the individual learning potential through intervention, whereas within RTI assessment is used to determine the most suitable intervention. Both approaches use intervention and assessments to adjust to the educational needs of children. Lidz and Peña (2009) therefore suggest that RTI and DA could be used complementary. Within tier 2 and tier 3, DA could be used to make decisions about more individualized intervention trajectories. Monitoring progress during intervention and treatment might inform about mastery levels, weaknesses, strengths and possible compensatory routes (Navarro & Mora, 2011). In our study, it would have been interesting to use the sublexical word fluency training for dynamic assessment. In chapter 5, data from the sublexical word fluency training were examined to explore learning effects. Learning growth during training could have directed the content and the pace of training using a dynamic algorithm. DA could help to design interventions that allow to progress from basic reading skills to more advanced levels at a child's own pace.

Learner characteristics

In chapter 2 and 3, we examined underlying skills in relation to reading ability in dyslexia and specific language impairment and as predictor for responsiveness to intervention. In both studies, rapid naming skill showed the strongest relationship. Phonological memory was not related to responsiveness.

Similarly, Tijms (2011) reported small to moderate associations between treatment responsiveness, rapid naming skill but also phonological memory. As variance in responsiveness is only partially explained by underlying cognitive skills, we argued that a RTI approach is recommended.

Nevertheless, the question remains how the relation between underlying skills and reading may affect remediation outcomes. We underscore the suggestion by Tijms, that future research disentangling the exact relation between RAN and responsiveness would be interesting to explore ways to increase intervention effectiveness. We do not argue for training rapid naming skills, because this has shown not to be effective (de Jong & Oude Vrielink, 2004). Explicit training of underlying skills does not affect reading (van der Leij, 2013), nor does improved reading result in progress on underlying reading related skills (Zijlstra, 2015). However, focus on shared aspects between RAN and reading, such as efficient processing letter-speech sound integration processing might improve responsiveness (Tijms, 2011).

Additionally, other learner characteristics might be good candidates for prediction. We only included reading related underlying skills to predict responsiveness. Nelson, Benner, & Gonzalez (2003) found that also problem behavior influenced the magnitude of responsiveness. Attention and motivation are no specific correlates of reading disabilities, but they may be associated or even be a consequence of the experienced reading difficulties. These aspects might be altered using an appropriate remedial approach to increase responsiveness to intervention. Behavioral aspects may be taken into account when assigning children to intervention conditions. For example, Vaughn, Linan-Thompson and Hickman (2003) reported that lengthy intervention sessions sometimes elicit challenging problem behavior indicating that for some children adjustments should be made.

Concluding remarks: longitudinal studies within the educational setting

Longitudinal quasi-experimental remediation studies as described in this thesis are relatively rare. Effects of remediation are most often measured over a short period of time including pretests and posttests without maintenance scores. The training studies are often targeted to explore specific issues and therefore limited in content and duration. Furthermore, training is often delivered by researchers or trained research assistants. Our study distinguished itself with respect to those aspects. The school-based intervention was delivered by trained teachers. The fidelity was controlled for by observations, feedback and ongoing training during intervention periods. The treatment was delivered by professional practitioners within the clinical setting. Procedures and progress of individual children were reviewed at regular meetings. Although longitudinal studies are costly, we suggest that this type of practice-based studies is valuable because it gives insight into the implementation of remedial practices within real-life settings. Moreover, intervention at tier 2 ensures that only very poor readers with a well-documented history of intervention resistance are included for costly treatment at tier 3, controlling for the influence of educational deprivation. Ideally, instruction at tier 1 should also be included within the research design because the quality of classroom instruction is also of importance. Additionally, the support and guidance that is provided at school during and after treatment should be taken into account when evaluating reading skills at follow-up. From our studies, it appeared difficult to prevent preliminary dropout of participants. This might not be surprising, because longitudinal studies require both effort and time from researchers and participants. By using techniques like multilevel analyses, it was possible to follow children over different periods of time with varying measurement occasions and handling missing data. Future studies should also consider appropriate analyzing techniques to explore different learner and environmental characteristics within an extended RTI framework.

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SUMMARY

This thesis focuses on the characteristics of reading problems and dyslexia and its treatment. Reading development and reading difficulties have drawn interest of many researchers. Accurate and fast word decoding is important for reading fluency and this in turn contributes to reading comprehension. Difficulties with accurate and effortless decoding affect the reading process and ultimately school success. Consequently, various studies have looked at the ethology, course of development and remediation of reading difficulties or dyslexia. This thesis contributes to this field of research.

In the general introduction (**chapter 1**) the topics of the thesis are described. Dyslexia and the case of comorbidity are introduced. Response to Intervention is proposed as an approach to support the identification of children with dyslexia. Children that show no or poor progress after intervention are in need of more intensive treatment. Some components of intervention and treatment have already been suggested to be effective.

In the first study (**chapter 2**), the central question was whether children with dyslexia and SLI differ on the phonological core characteristics. Children with dyslexia, children with specific language impairment (SLI) and children with typical reading and language development (control group) were compared on several measures of phonological processing and memory. In addition, we explored the association of reading skills and language impairment to phonological processing tasks. Children with dyslexia and SLI showed poor phonological awareness and rapid naming skills. Although the level of performance was most strongly related to reading level in both groups, children with SLI showed (mild) deficits on phoneme awareness and serial rapid naming that could not be traced to their reading level. They were more severely hampered on tasks with a phonological short-term memory component that seemed largely independent of reading achievement. Gradient differences in performance on phonological decoding were best explained by reading level. We concluded that the core deficit of dyslexia is best characterized by tasks that involve phoneme awareness and serial rapid naming, whereas SLI is associated with phonological short-term memory impairments. Moreover, part of the phonological abilities appear independent of reading level in SLI.

The objective of the second study (**chapter 3**) was to investigate the contribution of rapid digit naming, phonological memory, letter sound naming and

orthographic knowledge to the prediction of responsiveness to a school-based, individual intervention of word reading fluency problems of 122 Dutch second and third graders whose reading scores were below the 10^{th} percentile in comparison with the normative group. Degree of responsiveness was determined by comparison of a pre- and posttest measure of word reading fluency with a 6 month interval. At posttest, 38% of the children had improved their reading scores above the 10^{th} percentile. Maintenance scores revealed no significant growth on average, confirming that word reading fluency skills of poor readers are hard to remediate. Except rapid digit naming, none of the measures predicted responsiveness after controlling for the autoregressive effect of initial performance on fluency of word reading. A large part of the variance remained unexplained, supporting the advantage of a response to intervention approach above traditional psychometric testing to identify severe reading disabilities.

In the third study (**chapter 4)** we aimed to improve the word reading fluency of Dutch children with severe dyslexia in a clinical setting. Studies report mixed results on effectiveness of clinical treatments focusing on word reading fluency which might be partly due to different selection criteria to qualify children for treatment. In this study, the response-to-intervention (RTI) approach was used to select poor readers who showed no or limited progress after lengthy, intensive school-based intervention. An experimental treatment method focusing attention on the mapping between phonology to orthography of sublexical features of words was compared with a neuropsychological treatment method that is often applied in the Dutch clinical setting. Results showed that continued outpatient treatment leads to further, though small, improvement of both word and text reading fluency. No differences were found between the two treatment conditions. Follow-up testing showed that the majority of children sustained the reading level that was obtained directly after treatment.

The objective of the fourth study (**chapter 5**) was to examine the effect of a word fluency training in more detail. The word fluency training focused on practice of orthographic patterns, i.e. consonant clusters and open versus closed syllables. It was argued that knowledge of orthographic patterns would support the recognition of words and new (pseudo)words. The aim of a detailed, in-depth study was to examine the learning and transfer effects during word fluency training. Results showed that continued training results in further improvement of word reading accuracy and fluency, but this was in particular the case for trained words. There was transfer to new words, but only limited.

In the epilogue (**chapter 6**), some additional issues that were only briefly mentioned in the previous chapters are discussed. The intervention and treatment under study resulted in gains but no full remedy. Direct effects and transfer effects were limited. Intensive continued guidance after treatment, self-initiated reading and text exposure and adaptive coping strategies might be important ingredients for improving the reading skills after treatment ends. In order to make continued guidance possible, small-group interventions could also be evenly effective and could overcome the limited time that is available for teachers. Within this study, intervention and treatment were standardized. That is, children practiced in the same pace. Adjustments to the individual learning needs of the pupils could result in more positive effects. Dynamic assessment procedures could help to design interventions that take into account the individual differences. In accordance, learning characteristics other than examined in chapter 3 could be good candidates to design individualized interventions.

SAMENVATTING

De studies uit dit proefschrift richten zich de kenmerken en de behandeling van leesproblemen en dyslexie. Leesontwikkeling en leesproblemen hebben de aandacht van veel onderzoekers gekregen. Goede en vlotte woordherkenning is belangrijk voor leesvloeiendheid en dit draagt bij aan leesbegrip. Moeilijkheden met goed en vlot decoderen beïnvloeden het leesproces en uiteindelijk het schoolsucces. Om die reden hebben verschillende studies zich gericht op de oorzaken van leesproblemen, het ontstaan en het verloop ervan en de remediëring. Dit proefschrift draagt bij aan dit onderzoeksterrein.

In de algemene introductie (**hoofdstuk 1**) worden de verschillende onderwerpen uit het proefschrift kort beschreven. Er wordt kort ingegaan op wat dyslexie is en de comorbiditeit met andere taalstoornissen. De benadering *Response to Intervention* is in eerder onderzoek voorgesteld om kinderen met dyslexie te identificeren. Kinderen die niet of weinig profiteren van interventie komen in aanmerking voor meer intensieve behandeling. Voor zowel interventie als behandeling van leesproblemen en dyslexie zijn al effectieve componenten aangedragen.

In de eerste studie (**hoofdstuk 2**) luidde de centrale vraag of kinderen met dyslexie en taalontwikkelingsstoornissen (TOS) verschillen in fonologische kenmerken. Kinderen met dyslexie, kinderen met TOS en kinderen met een normale taal- en leesontwikkeling zijn met elkaar vergeleken op verschillende maten van fonologische verwerking en geheugen. We zijn nagegaan wat de relatie van leesvaardigheid en taalstoornis met fonologische verwerkingsvaardigheden is. Kinderen met dyslexie en TOS lieten een zwak foneembewustzijn en trage benoemsnelheid zien. Hoewel in beide groepen het niveau op de taken het sterkst samenhing met de leesvaardigheid, lieten kinderen met TOS (milde) tekorten op zowel foneembewustzijn en benoemsnelheid zien die los stonden van leesvaardigheid. Ze waren bovendien zwakker op taken die een beroep deden op het fonologisch geheugen en dit bleek voor een groot deel onafhankelijk van leesvaardigheid. Graduele verschillen in scores op een taak van fonologisch decoderen (lezen van onzinwoorden) werden het best verklaard door leesniveau. We concludeerden dat het kernprobleem van dyslexie het best gekenmerkt wordt door taken als foneembewustzijn en benoemsnelheid, terwijl TOS gerelateerd is aan problemen met fonologisch geheugen. Ook bleek dat fonologische vaardigheden bij kinderen met TOS deels onafhankelijk zijn van hun leesniveau.

Het doel van de tweede studie (**hoofdstuk 3**) was om te onderzoeken in hoeverre benoemsnelheid, fonologisch geheugen, benoemen van letterklanken en orthografische kennis de respons op een schoolinterventie voorspellen. Het ging om een interventie die 122 leerlingen met een zwakke leesvaardigheid (E-score) uit groep 4 en 5 individueel kregen aangeboden. De mate van responsiviteit op de interventie werd bepaald door de vlotte woordherkenning voor en na 6 maanden interventie te vergelijken. Op de nameting had 38% van de leerlingen hun leesvaardigheid verbeterd: zij scoorden niet meer op E-niveau maar daarboven. In de periode na de interventie lieten de leerlingen over het algemeen geen significante groei meer zien, wat bevestigt dat de vlotte woordherkenning moeilijk te remediëren is. Behalve benoemsnelheid voorspelde geen van de taken de mate van responsiviteit. Een groot deel van de variatie tussen leerlingen werd niet verklaard. We suggereren dat dit de inzet van een *respons to intervention* benadering ondersteunt in vergelijking met een meer traditionele benadering van testen.

In de derde studie (**hoofdstuk 4**) beoogden we de vlotte woordherkenning van leerlingen met ernstige dyslexie in een klinische setting te verbeteren. Eerdere studies laten wisselende resultaten van effectiviteit zien als het gaat om het verbeteren van de woordherkenning in een behandeling. Dit zou deels kunnen komen door de verschillende selectiecriteria die in de studies gebruikt zijn. In deze studie selecteerden we leerlingen die geen of weinig vooruitgang lieten zien na een lange en intensieve interventie op school. Een experimentele behandelmethode werd vergeleken met de standaard behandelmethode in de leeskliniek. De experimentele methode richtte de aandacht op de koppeling tussen fonologie en orthografie op sublexicaal niveau. Uit de studie bleek dat de behandeling leidde tot verdere verbetering (alhoewel beperkt) van de leesvaardigheid op woord en tekstniveau. Er werden geen verschillen gevonden tussen de twee behandelmethoden. Leerlingen werden een half jaar tot een jaar na afronding van de behandeling nog eens getoetst. De meerderheid van de leerlingen had het leesniveau vastgehouden dat ze direct na behandeling behaalden.

Het uitgangspunt voor de vierde studie (**hoofdstuk 5**) was een meer gedetailleerde analyse te geven van de experimentele behandelmethode. Deze behandelmethode richtte zich op het oefenen van orthografische patronen binnen woorden, zoals medeklinkerclusters en open versus gesloten lettergrepen. We beargumenteerden dat kennis van deze orthografische patronen de woordherkenning van woorden en nieuwe (pseudo)woorden zou ondersteunen.

Het doel van deze studie was om na te gaan wat de leer- en transfereffecten tijdens de behandeling waren. De resultaten lieten zien dat oefening leidde tot verdere verbetering van goede en vlotte woordherkenning, maar dit geldt vooral voor de woorden die de leerlingen in de training geoefend hadden. Er was slechts beperkte transfer naar nieuwe woorden met getrainde orthografische patronen.

In de epiloog (**hoofdstuk 6**) worden een aantal aanvullende punten, die slechts kort in de voorgaande hoofdstukken werden aangestipt, besproken. De interventie en behandeling die werden onderzocht resulteerden in vooruitgang in leesvaardigheid, maar het leidde niet tot volledige remediëring. Directe effecten en transfereffecten waren beperkt. Voortzetting van intensieve begeleiding na de behandeling, veel en vaak lezen en aangepaste coping strategieën zijn mogelijk nodig om de leesvaardigheid verder te verbeteren nadat de behandeling is afgerond. Om deze begeleiding mogelijk te maken zou dit in kleine groepen kunnen plaatsvinden om zo tegemoet te komen aan de onderwijspraktijk waarin tijd kostbaar is. Ook zou de interventie en behandeling misschien meer op maat geboden moeten worden. In de hier besproken studies waren de interventie en behandeling gestandaardiseerd. Aanpassingen aan de individuele leerbehoeften van de leerlingen zou mogelijk meer effectief zijn. Dynamische toetsprocedures zouden kunnen bijdragen aan de opzet van dergelijke interventies. Leerlingkenmerken zouden hierbij kunnen worden meegenomen.

DANKWOORD

Het schrijven van het dankwoord is een van het meest dankbare deel van al het werk dat er aan vooraf ging, mits de maat daarvoor het aantal lezers is. Bij het ontvangen van een proefschrift bladert iedereen immers direct naar die laatste pagina's. Het is het meest gelezen hoofdstuk, vermoed ik. Mijn dankwoord valt tegen voor hen die op een klassiek dankwoord hoopten, met namen en persoonlijke noten. Zo lang als mijn weg naar de uiteindelijke promotie was, zo beknopt zal ik het dankwoord houden. Als een traject zo'n ruime tijd beslaat, neemt het aantal mensen aan wie je dank betuigen kunt en wilt toe. En de herinneringen en anekdotes zijn ontelbaar. Ik zou er een boek met de omvang van een proefschrift aan kunnen wijden. Ik kies voor een samenvatting. Dankwoordlezer, herken jezelf!

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Mijn promotor en copromotor zijn nog altijd actief op het gebied van alles dat met dyslexie te maken heeft, al zou dat inmiddels niet meer hoeven. Ik doe nu met mijn promotie alweer een beroep op tijd die zij ook in opera- of concertzalen zouden kunnen doorbrengen. Ik zou het in hun plaats wel weten!

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De strikte deadline heeft me uiteindelijk het meest geholpen, zoals mij dat vaker helpt. Ik had het geluk dat het schrikkeljaar mij een dag extra bood. Bovendien stonden Aryan, Chris en leescommissie paraat om het op korte termijn te beoordelen.

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