The only way is up

Risk factors, protective factors, and compensation in dyslexia

van Viersen, S.

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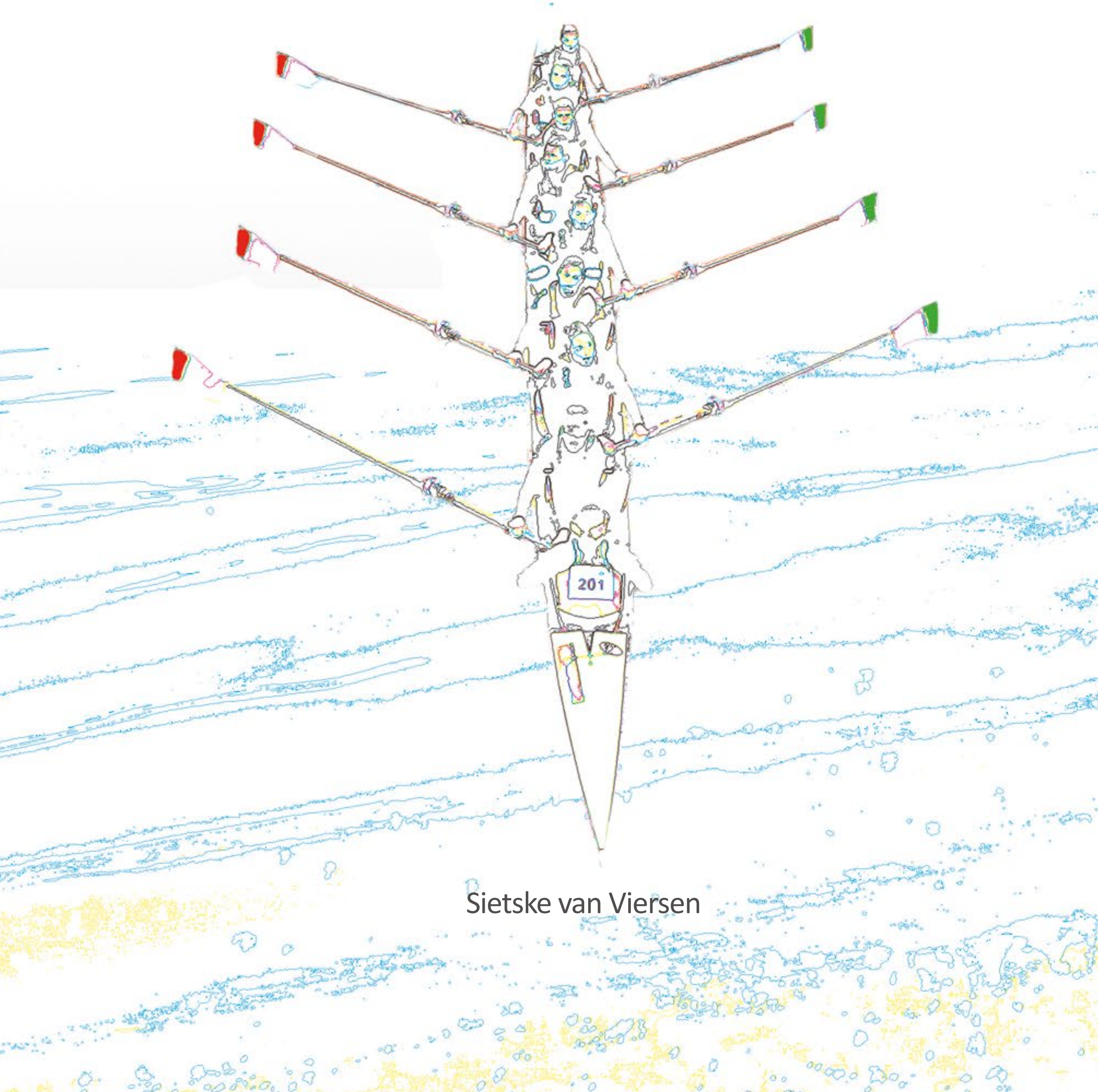
RISK FACTORS, PROTECTIVE FACTORS, AND COMPENSATION IN DYSLEXIA



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CONTENTS

CHAPTER 1

GENERAL INTRODUCTION

This introduction starts with two short case descriptions. The first case description has been derived from multiple similar cases and pays special attention to risk factors for the development of dyslexia. The second case description is based on a single, very typical case of dyslexia in a gifted child and illustrates how dyslexia may develop when both risk and protective factors are present. Both cases provide a first glimpse at the main issues that will be addressed in this dissertation. This is what life has been like for child X with dyslexia:

> *Before X went to school, her development progressed well. Her vocabulary might have been a bit behind, but it was not considered a problem. Learning to name colors was consistently difficult. In kindergarten, learning songs and rhymes was not a particular strength. In Grade 1, learning became much more complicated: learning the letter sounds was difficult and when X started learning to read, she was slow in reading words and made many errors. Throughout the early grades, poor accuracy turned into persistent slowness in reading and basic interventions in and outside the classroom did not seem to be effective. Reading exercises at school made her cringe and X went to school with dread, as she was slow on all tasks that demanded reading. As her father was not really a reader either, X's parents started to become quite worried about the situation. Still, it took some time before both teacher and school were convinced to hand documentation over for clinical assessment. Finally, at the end of Grade 3, there was an official diagnosis of dyslexia! The reading difficulties still provide a daily struggle for X, as reading remains problematic, burdensome, and a main source of frustration. However, at least she now gets specialized help to reduce the reading difficulties and cope with the difficulties that are caused by the reading disabilities, both at an educational and personal level. This allows X to focus on the (near) future again.*

This is what life has been like for Y, a gifted child with dyslexia:

> *From birth onward, Y's curiosity, ability to learn, and cleverness stood out. Already alert from the cradle, Y was quick to walk and talk, always inquisitive, and asking questions. Y couldn't wait for his first day of kindergarten; meeting new people, learning new things. Kindergarten was a good first step, but his creativity was not always valued; learning rhymes based on content was apparently not what Y was supposed to do. Once in school, Y's eagerness and enthusiasm peaked and faded with the years. His lagging reading and (most notably) spelling skills make him doubt himself. They may be noticed more than his precocious abilities, of which he is actually quite proud. As a result, Y is not admitted to a gifted program, thereby taking away the opportunity to feed his knowledge and build his talents. Boredom kicks in due to a lack of challenges and underachievement is lurking due to increasingly severe reading difficulties, for which he struggles to get sufficient support. Referral for diagnostic assessment has been put off by Y's teacher for quite some time now, as she has seen children doing worse than Y, and help from a professional seems far away. Combined, this poses a real threat to Y's happiness, both inside and outside school, and future school career.*

Both cases are similar but at the same time vastly different. Whereas the first case highlights (early) risk factors that are characteristic for the development toward dyslexia, the second case illustrates the interaction between both risk factors related to dyslexia and protective factors associated with giftedness. As a result, despite having the same learning disability, both children differ in their developmental trajectories, the difficulties that they experience (both educationally and personally), and their prospects for the future.

Recent developments in education have led to some major improvements in how schools (are supposed to) deal with diversity in the classroom, make room for tailor-made interventions, and provide opportunities for gifted curricula (e.g., van Bijsterveldt-Vliegenthart, 2012). Moreover, knowledge about identification, diagnosis, and intervention of dyslexia is becoming more advanced, and awareness of giftedness and gifted education is also growing. However, the case descriptions above illustrate where issues still exist. These include the role of family risk in relation to dyslexia, *early* identification of risk factors (specifically oral language), presence of protective factors, their interaction with risk factors and possible compensation, and consequences for diagnostic assessment. These issues are the focus of this dissertation.

DEFINITION OF DYSLEXIA

Most researchers agree that dyslexia concerns a severe and persistent difficulty with acquiring word-level reading skills (e.g., Snowling, 2000). However, this seemingly simple definition of dyslexia has been hotly debated (e.g., Stanovich, 1991). For decades, aptitude-achievement discrepancy definitions were widely accepted. Aptitude was generally conceived as intelligence. These definitions were based on the assumption that the reading disabilities of children with high and children with low aptitude are qualitatively different. Yet, falsification of this hypothesis (Stanovich, 1996; Stanovich & Siegel, 1994) has slowly moved the field toward low achievement definitions and more recently the response-to-intervention approach toward dyslexia (Fuchs, Fuchs, & Compton, 2004, 2012). Currently, it is still debated whether and/or what literacy skills should be part of the definition of dyslexia besides word-level reading (i.e., spelling). In addition, definitions may show variation in the extent to which they state exclusion criteria or specify underlying causes of the literacy difficulties. Despite this variation in definitions and criteria, in The Netherlands prevalence estimates of dyslexia are thought to range between 4-7% in primary education (Blomert, 2006; Kleijnen et al., 2008) and 12% in secondary education (Sontag & Donker, 2012).

ETIOLOGY OF DYSLEXIA

According to the multiple deficit model (MDM; Pennington, 2006) the etiology of developmental disorders is probabilistic and multifactorial in nature. In this model, no single risk factor is sufficient or necessary to develop a disorder. Instead, developmental disorders are assumed to arise as the result of the interaction between multiple environmental and genetic risk and protective factors. Risk and protective factors alter the development of neural systems and thereby affect the cognitive functions that underlie behavior. Because a disorder results from multiple cognitive deficits, which can differ in severity and interact with each other as well as with protective factors, there are an infinite number of pathways toward developing a disorder.

A consequence of the probabilistic and multifactorial nature of developmental disorders is that the liability to develop a developmental disorder is continuous. Risk factors increase the liability to a disorder whereas protective factors decrease its liability. The continuous nature of the liability fits with the behavioral manifestation of many developmental disorders, which are continuous as well. As a result, many disorders can be considered as dimensional rather than as a distinct category. Therefore, disorders such as dyslexia are often defined as the lower end of a (normal) distribution. The cut-offs between normal and abnormal behavioral manifestations are relatively arbitrary.

Over the years, the MDM has replaced models that assume that one unique factor determines dyslexia. In contrast to these single factor models, the MDM can account for dyslexia in ways that single factor models cannot. Research has long focused on finding one single factor that might cause dyslexia and explain all its associated behavioral symptoms. Phonological processing had been proposed to be this factor for dyslexia (Lundberg & Høien, 1989; Snowling, 1998; Wagner, 1986; see Stanovich, 1988, for an alternative suggestion). However, in the meantime, multiple studies have shown that a phonological deficit is neither necessary nor sufficient for causing dyslexia; not all individuals with a phonological deficit have dyslexia (Birch & Chase, 2004; Snowling, 2008) and not all individuals with dyslexia have a phonological deficit (Heim et al., 2008; Pennington et al., 2012; Valdois et al., 2011). Moreover, studies have convincingly shown that reading is dependent on multiple factors (e.g., McGrath et al., 2011; Peterson et al., 2016), that dyslexia often arises as the result of multiple risk factors (e.g., Aro et al., 2009; van Bergen, van der Leij, & de Jong, 2014; Nag & Snowling, 2011), and that the experienced problems may increase in severity as risk factors accumulate (e.g., Nash, Hulme, Gooch, & Snowling, 2013; Snowling, 2008; Wolf & Bowers, 1999).

Moreover, the MDM can better account for comorbidity between disorders than single factor models. This is important because comorbidity rates are substantial for children with dyslexia. More specifically, 25 to 40% of children with dyslexia also have attention-deficit hyperactivity disorder (ADHD; Willcutt & Pennington, 2000; see also Gooch, Snowling, & Hulme, 2011; McGrath et al., 2011), about 50% also have a specific language impairment (SLI; McArthur, Hogben, Edwards, Heath, & Mengler, 2000; see also Ramus, Marshall, Rosen, & van der Lely, 2013; Rispens & Parigger, 2010), up to 60% have comorbid dyscalculia (Landerl & Moll, 2010; see also Moll, Göbel, & Snowling, 2015; Slot, van Viersen, de Bree, & Kroesbergen, 2016; van der Sluis, van der Leij, & de Jong, 2005), and around 60% also have Developmental Coordination Disorder (DCD; Kaplan, Wilson, Dewey, & Crawford, 1998; see also Chaix et al., 2007; Lingam et al., 2010). In single factor models, each disability is caused by its own unique underlying deficit. As such, in the case of comorbidity between disorders, prevalence is assumed to be even lower than the prevalence rates of the separate disorders. When prevalence rates are high, comorbidity would have to be explained by a separate underlying risk factor, unrelated to those that are specific to the individual disabilities. There is no research to support this, however. Within the MDM, these high comorbidity rates align with the assumed multifactorial nature of disorders. For example, the high comorbidity between dyslexia and SLI is thought to arise from a shared deficit in phonology (Catts, Adlof, & Hogan, 2005). Comorbidity is thus thought to result from the overlap in underlying risk factors between disorders.

This dissertation will focus on cognitive risk and protective factors associated with dyslexia. The main risk and protective factors that are included in this dissertation are further explained below.

RISK FACTORS

The cognitive factors involved in literacy acquisition develop from infancy all the way through early adolescence. In most cases, literacy development progresses normally. In some cases, however, literacy development may be severely hampered. Here, several key cognitive skills that are deemed important for literacy acquisition and may be impaired in children with dyslexia are discussed.

LANGUAGE SKILLS

Oral language skills are often regarded as the foundation of literacy development (e.g., Storch & Whitehurst, 2002). Vocabulary is one of the first language skills to develop (Scarborough, 1990; Torppa et al., 2010), and expanding vocabulary size is assumed to be a prerequisite for the further development of higher order language skills, such as syntactic skills and morphological awareness (see Rispens, 2004, for an overview). These language skills are also believed to affect the development of word reading ability through their effect on preliteracy skills, such as phonological awareness, verbal short-term memory, and grapheme-phoneme knowledge (Bates & Goodman, 1997; Lee, 2011; Metsala & Walley, 1998; Nation & Snowling, 1998; Share, 1995). Vocabulary has been assumed to have a more direct effect on the development of reading comprehension than on word decoding, as the knowledge of words has been found to be crucial for the understanding of texts (Lee, 2011; Muter, Hulme, Snowling, & Stevenson, 2004; Oakhill & Cain, 2012; Ricketts, Nation, & Bishop, 2007).

Several studies have shown that children who later go on to become dyslexic may show early deficits in their vocabulary and/or syntactic development (Scarborough, 1990; Torppa et al., 2010). For example, Scarborough (1990) found impairments in sentence length, syntactic complexity, and pronunciation accuracy of spoken language in two-and-a-half-year-old children who later became dyslexic. In addition, these children started to show receptive vocabulary deficits from the age of three years. Torppa et al. (2010) found an expressive language deficit in dyslexic children at the age of 1.5 years, expressive vocabulary and sentence length deficits at 2 years, and sentence length and receptive language impairments at 2.5 years. For both studies, the presence of deficits was not stable over time. Moreover, deficits may have partly resolved around preschool age (see also Rescorla, 2011). Thus, despite the common belief that early language skills are important for learning to read, clear empirical evidence for its status as a risk factor for dyslexia is lacking.

PRELITERACY SKILLS

Research has shown that across languages of varying orthographic complexity, there are three independent preliteracy skills that predict variation in literacy development; phonological awareness (PA), rapid automatized naming (RAN), and letter knowledge (Caravolas et al., 2012). PA can be defined as the ability to detect and manipulate speech sounds in spoken words, whereas RAN pertains to the access to and quick retrieval of phonological representations from long-term memory (Boets et al., 2010; de Jong & van der Leij, 2003; Snowling, 2001; Wagner & Torgesen, 1987). Letter knowledge simply refers to the knowledge of letter-sound correspondences necessary for building alphabetic skills (Byrne, 1998; Caravolas et al., 2012). Verbal short-term memory (VSTM), i.e., the temporary storage of phonological representations, has also been suggested as an important predictor (e.g., Snowling, 2000; Wagner & Torgesen, 1987). However, several studies have shown that it is a weak and inconsistent predictor (Vaessen, Bertrand, Denes, & Blomert, 2010; Ziegler et al., 2010) and that it is not a unique predictor of later reading and spelling skill across languages (e.g., Caravolas et al., 2012; de Jong & van der Leij, 1999).

Impaired preliteracy skills have been found to be considerable risk factors for the development of dyslexia. Children who go on to become dyslexic often show low performance on PA tasks, slow RAN, limited VSTM, and have considerable difficulty with learning the letters of the alphabet (Snowling, 2000). However, the hallmark deficits of children with dyslexia may differ between languages of different orthographic depth. In very opaque languages, such as English, children with dyslexia often show severe underlying deficits in PA (and VSTM), resulting in poor word reading accuracy (Seymour, Aro, & Erskine, 2003). In more transparent languages, such as Dutch, underlying deficits of children with dyslexia mostly pertain to RAN, often resulting in poor word reading fluency (Landerl & Wimmer, 2000). PA deficits are not visible in simple tasks, but remain present during development when more difficult or speeded tasks are used (de Jong & van der Leij, 2003; Patel, Snowling, & de Jong, 2004). The differences between languages in the main underlying deficits of children with dyslexia, and RAN and PA in particular, can have consequences for the connection between early language and preliteracy skills. This connection could, for example, be stronger in English than in Dutch due to the more dominant position of the PA-deficit in these children with dyslexia.

FAMILY RISK

A specific risk factor that may precede all risk factors above in the development of dyslexia is family risk. Dyslexia is a highly heritable disorder (e.g., Byrne et al., 2009). Children are generally considered to have an increased genetic risk, or a so called family risk (FR), when they have a first degree relative (usually a parent) with a history of severe literacy difficulties. Both genetic and environmental risk factors are transferred from parents to their children through genetic and cultural transmission. These risk factors negatively impact neural systems and underlying cognitive abilities associated with literacy development (van Bergen, van der Leij et al., 2014). As a result, FR children have an increased liability to develop dyslexia; the chance to develop dyslexia is 3 to 4 times higher for FR children than for children without an FR (van Bergen et al., 2011; Elbro, Borstrøm, & Petersen, 1998; Pennington & Lefly, 2001; Snowling, Gallagher, & Frith, 2003; see Snowling & Melby-Lervåg, 2016, for an overview).

For children at FR of dyslexia, a further distinction can be made between FR children who go on to become dyslexic (FR-dyslexic) and children who do not succumb to severe and persistent literacy problems (FR-nondyslexic). However, as liability for dyslexia is continuous, based on the MDM (Pennington, 2006), mild deficits in literacy and underlying cognitive skills are to be expected in FR-nondyslexic children as well. This translates into a stepwise pattern of performance (i.e., FR-dyslexic < FR-nondyslexic < typically developing [TD] control) on literacy and literacy-related cognitive factors. There is an increasing body of research supporting these expectations for both FR groups (see Snowling & Melby-Lervåg, 2016, for an overview). Because genes that are related to FR often affect general processes (e.g., neuronal migration; Carrion-Castillo, Franke, & Fisher, 2013), an FR can cause an impairment in a factor that is unrelated to reading. Consequently, impairment in children with dyslexia might be due to their FR and not be related to their reading status. The distinction between FR-dyslexic and FR-nondyslexic children can help to pinpoint what factors are related to later reading outcomes and what factors are merely associated with FR (e.g., Hakvoort, van der Leij, van Setten, Maurits, Maassen, & van Zuijen, 2016; Moll, Loff, & Snowling, 2013).

Previous research has shown that the presence of a stepwise pattern of performance may vary between literacy-related factors, including the previously mentioned (early) language and preliteracy skills. FR-nondyslexic children have, for example, been found to show mild deficits in measures of PA (van Bergen, de Jong, Maassen, & van der Leij, 2014; Moll et al., 2013; Pennington & Lefly, 2001). However, results for letter knowledge and RAN are less unanimous and may partly depend on the specific point in development (i.e., preliterate or not; see Snowling & Melby-Lervåg, 2016). Some studies have indicated mild impairments in RAN (Pennington & Lefly, 2001) and letter knowledge (Snowling et al., 2003), whereas other studies indicated that RAN performance (van Bergen, de Jong, Maassen, & van der Leij, 2014; Moll et al., 2013; Snowling et al., 2003) and letter knowledge (van Bergen, de Jong, Maassen, & van der Leij, 2014; Pennington & Lefly, 2001) of FR-nondyslexic children is comparable to that of TD children. Concerning early language skills, studies consistently showed that FR-nondyslexic children generally have no impairments in early vocabulary and syntactic skills (Scarborough, 1990; Snowling et al., 2003; Torppa et al., 2010). Yet, findings for verbal abilities in a broader sense are more mixed. Some studies find mild impairments in FR-nondyslexic children (e.g., van Bergen, de Jong, Maassen, Krikhaar, Plakas, & van der Leij, 2014) whereas others do not (e.g., Moll et al., 2013).

Several issues warrant further attention. So far, little is known about the role of early oral language skills in the development of dyslexia and how having an FR may influence this. The first issue concerns early oral language deficits during infancy. Previous FR studies focusing on early deficits in vocabulary and syntactic skills always considered vocabulary development at individual points in time, thereby often switching to a different instrument per developmental phase (e.g., Scarborough, 1990). This resulted in inconsistency in findings between occasions during early development. Torppa et al. (2010) used the same instrument at three consecutive occasions during infancy, but findings for (expressive) vocabulary deficits remained inconsistent over time. Therefore, an important next step is to focus on investigating growth trajectories, using the same instrument at consecutive time points, to make clearer statements about early vocabulary development and possible differences between FR-dyslexic and FR-nondyslexic children. Mapping growth could also reveal more about the severity and persistence of early language deficits and their significance as a risk factor for dyslexia.

The second issue concerns how early oral language proficiency subsequently influences trajectories in literacy development. Only a few FR studies have mapped the developmental trajectories toward literacy, and particularly toward reading comprehension, the ultimate outcome of literacy development, while including early oral language skills (e.g., Duff, Reen, Plunkett, & Nation, 2015; Hulme, Nash, Gooch, Lervåg, & Snowling, 2015; Torppa et al., 2010). However, these studies focused on one particular part of development, covering a limited age range or only one possible pathway, or included a limited set of mediating variables. Consequently, it has remained unclear *if* there is an effect of early language on reading comprehension, and if so, *how* this effect is mediated by other literacy-related skills. In addition, as the inclusion of FR in these studies varied considerably, it is unknown at *which* points during development and *how* the relation between early language and reading comprehension may be influenced by FR, and whether this relation is also mediated by other literacy-related skills. A large longitudinal FR study, including both preliteracy and word decoding skills as well as early and later language skills, is necessary to gain more insight into the development of literacy and dyslexia.

PROTECTIVE FACTORS

Besides specific risk factors, children with dyslexia may also possess protective factors. As with risk factors, these may lie within the environment (e.g., Kiuru et al., 2013), or concern underlying cognitive factors. Here, the focus is specifically on *cognitive* protective factors, in terms of highly developed skills, that are relevant for literacy development and exist in presence of cognitive risk factors for dyslexia.

GIFTEDNESS

A proposed but very much underresearched context for potential protective factors is giftedness. Here, giftedness is defined as 'academic giftedness' or high intelligence, which is generally equivalent to having an IQ score above 130 (two standard deviations above average; e.g., Winner, 1997). Of course, giftedness entails much more than just high IQ. For example, creativity and motivation are also considered important characteristics of children displaying extraordinary talents (e.g., Mönks & Katzko, 2005; Renzulli, 2002). However, as both creativity and motivation are still hard to measure in a standardized way (Lovett & Lewandowski, 2006), and the link with basic academic skills such as learning to read is not very obvious, high intelligence is considered the most relevant part for the topic addressed in this dissertation.

TWICE-EXCEPTIONALITY FRAMEWORK

According to the twice-exceptionality framework, high intelligence has an influence on the manifestation of learning disabilities. Twice-exceptionality is defined as the concomitant occurrence of high intelligence and a learning disability within an individual (Brody & Mills, 1997; McCoach, Kehle, Bray, & Siegle, 2001). Prevalence rates for twice-exceptionality have been found very hard to obtain due to serious identification problems arising, for example, from the lack of consistency in definitions of both giftedness and learning disabilities (Nielsen, 2002). A rough yet conservative estimate, mostly based on discrepancy accounts of twice-exceptionality (e.g., McCoach et al., 2001), is that about 1-5% of children with a learning disability are twice-exceptional (McCoach, Kehle, Bray, & Siegle, 2004; Nielsen, 2002; Silverman, 1989).

The main idea of the twice-exceptionality framework is that the cognitive profile of twice-exceptional children consists of both strengths related to their high intelligence and weaknesses associated with their learning disability (Brody & Mills, 1997; see Foley Nicpon, Allmon, Sieck, & Stinson, 2011, for an overview). The assumption is that these cognitive strengths, enabling compensation, influence performance at the behavioral level and thereby alter the expression of the learning disability (Assouline, Foley Nicpon, & Whiteman, 2010; Brody & Mills, 1997). However, empirical data supporting this assumption is very limited, mostly as the result of problems with (early) identification of twice-exceptional children (this issue is also addressed below).

COMPENSATION

Atypical expression of learning disabilities in twice-exceptional children is thought to result from compensation. Within the twice-exceptionality framework, the term compensation is derived from the assumed cognitive profile in twice-exceptional children, consisting of giftedness-related strengths and disability-related weaknesses (Foley Nicpon et al., 2011). In the specific context of dyslexia, the term compensation is easily used as an explanation when children show higher than expected performance, but it is difficult to pinpoint what compensation means. There are several possible definitions of compensation. For example, Snowling et al. (2003) state that compensation occurs when a cognitive strength or protective factor moderates the effect of a cognitive risk factor on literacy development. Following this definition, a protective factor can have a straight impact on the risk factor itself or on the relation between the risk factor and the literacy skill at hand. This can be considered as 'direct compensation'. A different way of thinking about compensation is through compensatory *mechanisms* (e.g., Reis, McGuire, & Neu, 2000). In this case, a protective factor provokes or stimulates the emergence of a compensatory mechanism during development that helps to circumvent the underlying deficit or subdue its negative effect on literacy development. This could be called 'indirect compensation', as it influences the impact of a risk factor by altering reading processes. However, both definitions of compensation are speculative and not based on empirical data; to the best of our knowledge there is neither hard evidence showing the existence of compensation nor proof of how it may (or may not) work.

Findings from brain studies have suggested the existence of (indirect) compensation. Individuals with dyslexia who had partly overcome their literacy difficulties at a later age (compensated dyslexics) showed increased activation in additional (but unassociated) brain regions during phonological processing. This overactivation would indicate the use of additional skills to process phonology. These regions were not activated in TD controls and individuals who still had persistent problems (uncompensated dyslexics; see Ozernov-Palchik, Yu, Wang, & Gaab, 2016, for an overview). In addition, Gilger, Talavage, and Olulade (2013) found that in nonverbally gifted/dyslexic students the pattern of functional activation in both hemispheres during reading-related tasks (e.g., rhyming) as well as during math-related tasks (e.g., spatial processing) resembled that of nongifted/dyslexic students. Moreover, compared to gifted readers without reading disabilities, the gifted/dyslexic students showed underactivation in *right* hemisphere brain regions during the same tasks. Gifted/dyslexic students were thus similar to nongifted/dyslexic students in terms of patterns of brain activation associated with dyslexia, but different from gifted students without reading disabilities in terms of patterns of brain activation associated with giftedness. These findings were explained in terms of loss of gifted potential in gifted/dyslexic students as a result of compensation for linguistic difficulties that are part of their dyslexia.

Previous research on compensation at the cognitive and behavioral level is limited to the identification of high performance on cognitive factors relevant for literacy development in presence of cognitive deficits associated with dyslexia. Possible factors involved in compensation include language skills (i.e., grammar and vocabulary; Nation & Snowling, 1998; Snowling, 2008; Snowling et al., 2003), orthographic knowledge (van der Leij & van Daal, 1999; Stanovich & Siegel, 1994) and orthographic learning (Wang, Castles, Nickels, & Nation, 2011), and/or visual memory and perceptual speed (Snowling, 2001; but see Winner et al., 2001).

MASKING

As stated above, compensation at the cognitive level may lead to a certain degree of masking at the behavioral level (i.e., in terms of affected performance characteristics). Masking may take several forms, affecting a child's ability and/or disability (see Brody & Mills, 1997, for an overview). First, masking may occur when a child's reading disability hampers overall academic achievement, resulting in not being recognized as gifted (Silverman, 2003). Second, giftedness-related strengths may influence achievement in such a way that the literacy level is higher than expected based on the presence of an underlying deficit, resulting in a missed diagnosis of dyslexia (Lovett & Lewandowski, 2006). Finally, masking may occur both ways when giftedness-related strengths enable compensation to a point that a learning disability is not recognized and yet the disability restricts achievement to an extent that giftedness is not acknowledged either (Hernández Finch, Speirs Neumeister, Burney, & Cook, 2014).

All three types of masking seriously complicate (early) identification of twice-exceptional children. Therefore, several studies stress the importance of comprehensive assessment, i.e., gaining diagnostic information from multiple sources, to 'unmask' twice-exceptionality (e.g., Assouline et al., 2010; Hernández Finch et al., 2014; Lovett & Lewandowski, 2006). However, the issue that remains is that empirical studies, especially concerning specific learning disabilities such as dyslexia, are lacking (Foley Nicpon et al., 2011).

The main assumptions of the twice-exceptionality framework are at odds with the work of Stanovich and colleagues. For decades, having a high IQ has been discarded as a relevant factor in the development of dyslexia. The argument behind this was that the core phonological deficits associated with dyslexia were found to be unrelated to intelligence (Stanovich & Siegel, 1994). Put differently, children of various intelligence levels are assumed not to differ in the cognitive processes that underlie their reading disability (Stanovich, 1996). Yet, as a cognitive skill is further away from the 'phonological core' and moves more toward general elements influenced by intelligence, this may lead to some variation in the cognitive profiles of children with dyslexia, but not in those skills associated with the condition, dyslexia, itself (Stanovich & Siegel, 1994). Consequently, intelligence has been thought to be irrelevant in relation to dyslexia. However, the role of cognitive strengths, in particular those associated with high intelligence, and their impact on reading has never been examined in gifted children. Gifted children with literacy problems often fail to reach the threshold for dyslexia based on low achievement criteria and/or fail to show underlying deficits associated with dyslexia. As a result, they miss out on either a diagnosis or reimbursed treatment. It is relevant to know whether missed diagnoses in these children are justified or could be due to compensation. In addition, as stated above, it has remained unclear what the exact behavioral and cognitive characteristics of these children are, and how these issues relate to practical and theoretical knowledge about dyslexia.

This dissertation will readdress the question whether there is any role for IQ in literacy development. Although Stanovich has shown that the lower end of the intelligence spectrum does not matter in relation to dyslexia, it is unknown whether this is also the case for the higher end of the distribution. Most importantly, as main assumptions coming from the field of twice-exceptionality and leading views from the field of dyslexia are largely contradictive, empirical data will be collected to properly evaluate both theories. Questions surrounding the combination between giftedness and dyslexia, in terms of behavioral and cognitive characteristics and associated strengths and weaknesses, will be addressed. Related, the existence of compensation will be tested and possible mechanisms explored. Consequences for diagnostic assessment of dyslexia in different subgroups of gifted children and the role of protective factors will be considered.

CURRENT STUDIES

The studies presented in this dissertation are based on data from two separate research projects; the Dutch Dyslexia Program and the Giftedness & Dyslexia Study. Both projects will be briefly introduced below. Next, an overview of the content of each chapter is presented.

DUTCH DYSLEXIA PROGRAM

The Dutch Dyslexia Program (DDP) is a longitudinal research project that started in 1999 and focuses on literacy development in children with and without an FR of dyslexia (van der Leij, van Bergen, van Zuijen, de Jong, Maurits, & Maassen, 2013). The DDP project is set up around three major universities in The Netherlands; the University of Amsterdam, the University of Groningen, and Radboud University Nijmegen. The three components of the project, a prospective longitudinal study, intervention, and genetic studies, make it multidisciplinary and combine research in the fields of genetics, neurophysiology, neurology, education, psychology, and linguistics. Children with and without an FR of dyslexia were followed from the age of two months until (now) age 16 years. Couples who were expecting a baby and had a family history of literacy difficulties were recruited to participate in the study. Children were considered to be at FR of dyslexia when they had one parent and one close relative with dyslexia. The dyslexia status of the children themselves was determined at several occasions as the project continued. In this dissertation, Chapter 2 and 3 are based on the DDP data, which were collected when children were between 17 months and 12 years old.

GIFTEDNESS & DYSLEXIA STUDY

The Giftedness & Dyslexia Study (HBDYS) is a cross-sectional study focusing on dyslexia in gifted children. It was set up in 2012 at Utrecht University and continued in collaboration with the University of Amsterdam in 2014. The study is divided into two part projects; one covering primary school children who are learning to read (ages 7 to 10, Grades 2 to 4), and the other involving students in the early years of secondary education (ages 11 to 14, Grades 7 to 9) starting to learn foreign languages (English, French, and German). The part project in secondary education is longitudinal and follows a subsample of students for two consecutive years. The main aim of the HBDYS study was to map the behavioral and cognitive characteristics of multiple subgroups of highly intelligent children with various degrees of literacy difficulties during two crucial school phases. We thereby hoped to gain more insight into risk factors associated with dyslexia, protective factors associated with high intelligence but relevant for literacy development, and possible effects of compensation and masking of literacy difficulties in this special population of children. Chapters 4, 5, and 6 are each based on data from the HBDYS study.

OUTLINE

The studies reported in Chapter 2 and 3 concern two FR studies that focus on early oral language skills as a risk factor for dyslexia.

CHAPTER 2 focuses specifically on very early vocabulary development. Parent reports were used to assess receptive and expressive vocabulary. Growth in receptive and expressive vocabulary development was modeled between the ages of 17 and 35 months. The aim of this study was to determine whether early vocabulary as a risk factor is related to FR status and/or associated with later reading ability. In addition, we wanted to know whether early vocabulary growth was different in children with an FR who later became dyslexic, compared to children with an FR and no dyslexia and/or TD children without an FR, and if so, whether differences point toward delay or deviance of growth.

CHAPTER 3 investigates the influence of early oral language on pathways toward reading comprehension in children with and without an FR of dyslexia. The study covers multiple phases of literacy development between the ages of 4 and 12 years. Mediating variables include both preliteracy skills and word decoding ability as well as later language abilities. Importantly, FR is included as a separate predictor to assess whether the effect of FR on reading comprehension is fully mediated by preliteracy skills or whether FR also has an independent effect via language abilities.

In Chapter 4, 5, and 6 we focus on the combination between giftedness and dyslexia. The studies reported here provide more insight into profiles of dyslexia-related risk factors and giftedness-related protective factors of gifted children with dyslexia. Reference groups include averagely intelligent children with dyslexia, TD children, and gifted children without literacy difficulties. Possibilities for compensation are also addressed. While Chapter 4 and 5 focus on children in primary education, Chapter 6 focuses on students in the first grades of secondary education.

CHAPTER 4 contains the first study to report empirical findings on the behavioral and cognitive characteristics of gifted children with dyslexia learning to read in the early to middle grades of primary education. Findings are supposed to stimulate a new outlook on current theories on dyslexia as well as practice in diagnostic assessment and intervention of dyslexia.

CHAPTER 5 compares underlying cognitive deficits associated with dyslexia in gifted and averagely intelligent children with dyslexia. In addition, differences in cognitive profiles between gifted children with dyslexia and gifted children with relative reading difficulties (borderline-dyslexic children) are investigated in terms of depth and breadth of deficits. A first attempt is made to specifically test hypotheses of (direct) compensation and to present a guideline regarding which gifted children are dyslexic and which are not.

CHAPTER 6 examines the literacy profiles of gifted students with dyslexia in their native language and three foreign languages. This study provides more insight in the degree to which dyslexia in the native language might influence foreign language learning in different groups of children. Furthermore, the set-up of this study offers the opportunity to obtain empirical evidence for the existence of compensation in foreign language learning.

CHAPTER 7 provides a synthesis of the research that is presented. The findings are discussed in light of current views and progression in the field as well as the aims of this dissertation. Theoretical and practical implications are presented along with directions for future research. Finally, recommendations for diagnostic practice and interventions are provided.

CHAPTER 2

DELAYED EARLY VOCABULARY DEVELOPMENT IN CHILDREN AT FAMILY RISK OF DYSLEXIA

This study aimed to gain more insight in the relation between vocabulary and reading acquisition by examining early growth trajectories in vocabulary of children at family risk (FR) of dyslexia longitudinally. The sample included 212 children with and without an FR from the Dutch Dyslexia Program. Parents reported on their children's receptive and expressive vocabulary size at ages 17-23-29 and 35 months using the Dutch *MacArthur Communicative Development Inventories* (CDI). Dyslexia status at the end of Grade 2 (8 years) rendered three groups; FR-dyslexic (n = 51), FR-nondyslexic (n = 92), and typically developing (TD) control children (n = 69). Repeated measures analyses showed that FR-dyslexic children had lower receptive vocabulary scores from 23 months onward and lower expressive scores from 17 months onward than FR-nondyslexic children. Latent growth curve modeling showed lower initial growth rates in FR-dyslexic children, followed by partial recovery, indicating a delayed increase in receptive and expressive vocabulary. FR-nondyslexic and TD children did not differ. Early deficits in receptive and expressive vocabulary are associated with later reading. Early vocabulary growth of FR-dyslexic children is characterized by a delay, but not deviance of growth. Vocabulary can be considered an additional but small risk factor for dyslexia.

Keywords: Vocabulary, receptive/expressive, family risk, dyslexia, early language, CDI.

van Viersen, S., de Bree, E. H., Verdam, M., Krikhaar, E., Maassen, B., van der Leij, A., & de Jong, P. F. (in press). Delayed early vocabulary development in children at family risk of dyslexia. *Journal of Speech, Language, and Hearing Research*.

INTRODUCTION

Vocabulary is a fundamental part of the development of (early) language skills. It is proposed to be a prerequisite for the emergence of higher order language skills such as grammar and morphological awareness (e.g., Bates & Goodman, 1997; Torppa, Lyytinen, Erskine, Eklund, & Lyytinen, 2010). Vocabulary is also believed to influence word reading acquisition through its effects on literacy-related abilities such as phonological awareness, phonological memory, and the acquisition of letter knowledge (e.g., Bates & Goodman, 1997; Lee, 2011; Metsala, 1999; Metsala & Walley, 1998; Nation & Snowling, 2004; Share, 1995; Torppa et al., 2010). In the current study, we focused on the role of early vocabulary development in later word reading ability; particularly, we examined differences in vocabulary development between children at family risk of dyslexia who did or did not go on to become dyslexic.

Dyslexia is a developmental learning disability characterized by severe and persistent reading and/or spelling difficulties at word level (e.g., Vellutino, Fletcher, Snowling, & Scanlon, 2004). Dyslexia has been found to be highly heritable and to run in families (van Bergen, van der Leij, & de Jong, 2014; Snowling & Melby-Lervåg, 2016). Deficits in phonological processing are generally assumed to lie at the core of dyslexia (de Jong & van der Leij, 2003; Vellutino et al., 2004). Especially impairments in phonological awareness, i.e., detection and manipulation of speech sounds (Snowling, 2001), are thought to hamper reading acquisition. Although it is still debated what underlies this phonological deficit, several theories link early vocabulary growth to (impairments in) the development of phonological skills and later literacy difficulties.

At least two prominent theories predict a relation between early vocabulary development and later word reading ability. The lexical restructuring theory (LRT; e.g., Metsala & Walley, 1998) proposes that vocabulary growth is instrumental in the development of phonological awareness through phonological representations. As new words are added to the mental lexicon of a child and neighborhood density increases, segmental restructuring of lexical items occurs, resulting in more detailed phonological representations (Metsala, 1999). These fine-grained phonological representations facilitate a child's discrimination between words with a similar sound and store these in a more discernable form in long-term memory (Metsala, 1999). According to the LRT, the phonological problems associated with dyslexia would result from impaired or delayed growth in early vocabulary. The segmentation theory (e.g., Boada & Pennington, 2006) proposes auditory perception/speech processing as the foundation of later phonological skills and word reading ability, instead of vocabulary growth. As phonological representations become more detailed through a process of increasing segmentation of speech sounds, phonological awareness emerges, also enabling vocabulary growth. In line with this theory, delays or deviations in speech sound segmentation lie at the basis of the (linguistic) problems generally associated with the development of dyslexia. The segmentation problems negatively affect the quality of the phonological representations, leading to significant differences in phonological awareness and vocabulary growth (Boada & Pennington, 2006).

Although both theories are similar in many respects and become increasingly intertwined during development due to the reciprocal relation between vocabulary and phonological skills, there are some important nuances. First, they ascribe a different role/position to vocabulary growth in the development of phonological awareness and later reading. Secondly, they propose a different implied causality in the relationship between vocabulary growth and emerging phonological awareness. LRT explicitly proposes that more detailed/segmented phonological representations are constructed because of vocabulary growth. In contrast, segmentation theory suggests that vocabulary growth may be more dependent on the quality of the phonological representations as based on speech segmentation skills. Furthermore, LRT suggests that vocabulary growth has a strong effect on the emergence of phonological awareness during development, whereas segmentation theory interprets vocabulary growth and the emergence of phonological awareness *both* to be outcomes associated with speech sound segmentation. Consequently, both theories have their own implications for expected differences in early vocabulary development and its association with later reading outcome. As lexical restructuring is assumed to take place as vocabulary size increases, which may extend across childhood and vary in intensity between developmental phases (Metsala, Stavrinos, & Walley, 2009; Rispens & Baker, 2012), differences may only emerge or become more pronounced as development progresses. On the other hand, segmentation theory assumes impairments to be present from the onset of vocabulary development onward. Here, we investigated possible differences in vocabulary development in the context of a family risk (FR) study.

FR studies, especially those that are longitudinal and start at an early age, are well suited to test and further specify risk factors of dyslexia. Children who are at FR of dyslexia have a first degree relative (parent or sibling) with severe and persistent literacy problems. FR children are expected to have both a higher genetic and environmental liability to develop dyslexia than children without an FR (van Bergen, de Jong, Maassen, & van der Leij, 2014; Pennington, 2006). FR studies provide more insight in precursors of literacy difficulties and thereby enable early identification and intervention. Research has shown that children at FR of dyslexia may vary from typically developing (TD) children in reading-related cognitive skills, such as language development, already from a very young age (Snowling & Melby-Lervåg, 2016). Within the FR group a further distinction can be made between FR children who do (FR-dyslexic) and do not (FR-nondyslexic) succumb to severe literacy problems. For example, previous research has shown that FR-nondyslexic children may show mild impairments in literacy and underlying skills, such as phonological awareness (e.g., van Bergen, de Jong, Maassen, & van der Leij, 2014), which is illustrated by a stepwise pattern of performance (i.e., FR-dyslexic < FR-nondyslexic < TD control; e.g., Moll, Loff, & Snowling, 2013). As such, this distinction between both FR groups provides the opportunity to determine to what extent the relation between a cognitive skill and word reading ability is associated with FR status and/or with later literacy outcome (hereafter dyslexia status).

FR studies focusing on (the predictive value of) general language development in the preschool years (i.e., 4 to 7 years; e.g., Carroll, Mundy, & Cunningham, 2014; Snowling, Gallagher, & Frith, 2003) have shown that poor early language skills are associated with later literacy impairments in FR children. Several FR studies have focused on vocabulary development in an even younger age group (i.e., range 14 - 31 months) using *MacArthur Communicative Development Inventories* (CDI) parent reports, which were also used in the current study (i.e., Aro, Eklund, Nurmi, & Poikkeus, 2012; de Bree, Zamuner, & Wijnen, 2014; Kerkhoff, de Bree, de Klerk, & Wijnen, 2013; von Koss Torkildsen, Syversen, Simonsen, Moen, & Lindgren, 2007; Koster et al., 2005; Lyytinen et al. 2004). Although all studies show that TD children had higher vocabulary scores than FR children, only one study found significant differences for receptive vocabulary (i.e., at 18 months; Kerkhoff et al., 2013) and one for expressive vocabulary (i.e., at 17 months; Koster et al., 2005). The nonsignificant results are possibly due to the relatively small sample sizes and associated lack of power in most of these studies. Moreover, the FR groups in these studies may show large differences in the percentages of children that later become dyslexic. If early vocabulary deficits are only found in FR-dyslexic children, these children may form the minority of the FR group, which affects the found effect sizes (see Snowling & Melby- Lervåg, 2016, for an overview of FR studies and effect sizes). Thus, the role of early vocabulary in the etiology of dyslexia has remained equivocal based on FR-only studies.

So far, only a few studies have focused on early vocabulary while discerning children with an FR who do and do not go on to become dyslexic. Scarborough (1990) found that impairments in both receptive and expressive vocabulary in FR-dyslexic children emerge at 42 months. Torppa et al. (2010) showed that differences were already attested at a younger age: vocabulary production differentiated FR-dyslexic children from FR-nondyslexic and control children from the age of 2 years onward. Using a continuous approach to reading, Duff, Reen, Plunkett, and Nation (2015) found that the relation between vocabulary (at ages 16 to 24 months) and word reading ability (at ages 4 to 9 years) remained significant after controlling for FR status. This finding illustrates the importance of taking both children's FR status and dyslexia status into account when investigating the role of early risk factors.

Overall, the results of these studies suggest that vocabulary is mainly related to later reading outcome, as early deficits were only found in FR children who became dyslexic. However, some issues remain unresolved. The ages at which deficits in the FR-dyslexic group could be identified varied across the studies mentioned above. In addition, it is unclear whether the deficits that were found were persistent or might have been resolved at later ages closer to school entry. Moreover, although these studies are longitudinal, they did not focus on vocabulary *development*, which was often restrained due to the use of different instruments between time points. To gain more insight in vocabulary development, we examined growth trajectories in groups of FR children with and without dyslexia and a group of typical readers without an FR.

Studies on TD children suggest that vocabulary development is not (entirely) linear during infancy and toddlerhood (i.e., 0 to 4 years; e.g., Fenson et al., 1994; Hamilton, Plunkett, & Schafer, 2000; Kauschke & Hofmeister, 2002). For example, a sudden and rapid increase in vocabulary size during the second year of life (Bates & Goodman, 1997), the vocabulary spurt, has been regularly observed. The exact pattern of growth and acceleration is still debated (e.g., see Goldfield & Reznick, 1990; Mervis & Bertrand, 1995) and might depend on whether receptive or expressive vocabulary is involved and the time span that is covered. Fenson et al. (1994), using parent reports, have demonstrated that receptive vocabulary shows a linear increase in American children between the ages of 8 and 28 months, whereas expressive vocabulary shows a more exponential increase toward the end of the second year. These results have been replicated by Hamilton et al. (2000) in British children between the ages of 12 and 25 months. A German study by Kauschke and Hofmeister (2002), using spontaneous speech at 13, 15, 21, and 36 months showed that the exponential increase in expressive vocabulary toward the end of the second year is followed by a further expansion, but with a decreased growth rate in the third year. Based on these studies, a vocabulary spurt and subsequent deceleration can be expected for expressive vocabulary, whereas receptive vocabulary seems to grow more gradually. However, information about atypically developing populations is scarce.

In sum, we examined differences in early vocabulary development among FR-dyslexic children, FR-nondyslexic children, and TD children without an FR. Growth of early receptive and expressive vocabulary was measured longitudinally on four occasions between the ages of 17 and 35 months using CDI (Fenson et al., 1994) parent reports. Both expressive and receptive vocabulary were taken into account to get a better overview of the extent/depth of possible impairments and increase possibilities for early identification and intervention. The first aim of the study was to determine whether receptive and expressive vocabulary are associated with children's FR status and/or dyslexia status (risk versus reading). The second aim was to examine whether and/or how the growth rates of FR-dyslexic children might differ from FR-nondyslexic children and/or TD control children (delay versus deviance of growth).

Although LRT and segmentation theory were not specifically framed for the context of FR studies, expectations for both groups of FR children can be deduced from these language models. For FR-dyslexic children, impairments in vocabulary development would be expected based on both LRT and segmentation theory. However, LRT would predict more severe impairments than segmentation theory: the consequence of poor lexical restructuring is poor phoneme awareness, which is a core deficit associated with dyslexia. Furthermore, for FR-nondyslexic children, mild impairments would be anticipated according to LRT. Previous studies reported stepwise patterns (i.e., FR-dyslexic < FR-nondyslexic < TD control) for phonological awareness and later reading (e.g., van Bergen, de Jong, Maassen, & van der Leij, 2014; Moll et al., 2013). This pattern would also be expected to surface for the assumed catalyst of phoneme awareness, early vocabulary. This stepwise pattern might be present from the outset or emerge during development (e.g., Metsala & Walley, 1998). However, as speech sound segmentation is thought to be impaired only in children who later become dyslexic (e.g., Boada & Pennington, 2006), no (mild) impairments would be expected for FR-nondyslexic children throughout (early) development.

METHODS

PARTICIPANTS

The total sample consisted of 212 children from the Dutch Dyslexia Program (DDP). The DDP is a longitudinal study of children with and without an FR of dyslexia (see van der Leij et al., 2013, for a detailed description). Parents with and without a history of dyslexia were recruited when they were expecting a baby and the children first came to the lab when they were 2 months old. FR was defined as having at least one parent and one close relative (i.e., first degree) with dyslexia. A detailed account of FR assessment in this data set is provided by van Bergen, de Jong, Plakas, Maassen, and van der Leij (2012). Dyslexia status was determined based on low achievement criteria at the end of Grade 2 (8 years). In line with Dutch dyslexia protocols (e.g., Kleijnen et al., 2008), children had to show word reading fluency scores that belonged to the lowest 10% of the population (based on individual assessment and with IQ ≥ 70 at time of testing). Application of these criteria resulted in three groups; 1) FR-dyslexic children (FRD; n = 51, 56.9% boys), 2) FR-nondyslexic children (FRND; n = 92, 58.7% boys), and 3) TD control children (TDC; n = 69, 60.9% boys). Four children were categorized as having dyslexia but had no FR and were omitted from the study, as this group was too small to include separately in the analyses. All parents signed written informed consent and the project was approved by the ethical committee.

The background characteristics of the groups in the sample are displayed in Table 1. The word reading performance at the end of Grade 2 indicates a stepwise pattern of performance, with FR-nondyslexic children scoring in between FR-dyslexic and TD control children. The vocabulary measures clearly indicate that the smaller vocabulary sizes in FR-dyslexic children than in both nondyslexic groups were present both at the preschool age (i.e., before literacy acquisition) and after several years of literacy instruction (i.e., end of Grade 3). Since both FR groups showed about equal levels of parental education (see Table 1), which was used as a proxy for socio-economic status (SES) and significantly lower than in the TD group, it can be ruled out that possible differences in early vocabulary size between both FR groups were caused by differences in SES.

Note that this study was conducted on largely the same sample as described in van Bergen et al. (2012), van Bergen, de Jong, Maassen, Krikhaar, Plakas, and van der Leij (2014), and van Bergen, de Jong, Maassen, and van der Leij (2014). Small differences in the total number of participants per study and the number of participants per group are the result of occasion used for the dyslexia diagnosis (i.e., Grade 2 or Grade 3) and the available data per child for the relevant variables included in a particular paper. The data described in the current study has not been reported on in the above mentioned studies within the DDP.

TABLE 1

Background Characteristics of the Three Groups in the Sample

	FRD		FRND		TDC	
Variable	*M*	*SD*	*M*	*SD*	*M*	*SD*
Word reading						
end Grade 2	34.57_a	13.11	69.98_b	16.13	75.18_c	14.73
Vocabulary						
53 months (VIQ)	102.55_a	11.55	109.06_b	10.39	111.91_b	9.10
end Grade 3	10.72_a	2.57	12.03_b	2.44	12.32_b	2.53
Parental education						
Mother	2.65_a	1.15	2.95_a	1.17	3.68_b	0.75
Father	2.47_a	1.20	2.58_a	1.20	3.57_b	0.96
Average	2.26_a	0.75	2.58_a	1.04	3.62_b	0.72

Note. Means in the same row that do not share subscripts differ at $p < .05$. FRD = family risk dyslexia; FRND = family risk no dyslexia; TDC = typically developing control; VIQ = verbal IQ.

INSTRUMENTS

MAIN VOCABULARY MEASURE. The Dutch version (Zink & Lejaegere, 2002) of the CDI (Fenson et al., 1994) was used to assess vocabulary size at 17, 23, 29, and 35 months. The N-CDI Words and Sentences (normed for ages 16 to 30 months) consists of 22 semantic categories covering 702 lexical items. The N-CDI was developed in Flanders (Belgium) and adjusted slightly for the DDP study to ensure that Flemish-specific words were not used (see Koster et al., 2005, for specifics). Parents reported on their children's receptive and expressive vocabulary within a two-week window of the targeted ages. The receptive score consisted of all words their child understood. The expressive score represented all words their child both understood and said. Internal consistency of the receptive and expressive scales is .99 and .98, respectively (Zink & Lejaegere, 2002). The correlations between the receptive and expressive vocabulary scores at 35 months and the verbal IQ score at age 4.5 years (as reported on in van Bergen, de Jong, Maassen, Krikhaar et al., 2014) are .52 and .53 ($p < .001$), which we take to mean that the validity of the N-CDI is sufficient.

BACKGROUND MEASURES. Verbal intelligence at the age of 53 months was estimated using the language comprehension subtest of the *Reynell* (van Eldik, Schlichting, lutje Spelberg, van der Meulen, & van der Meulen, 2001) and the expressive vocabulary, expressive syntax, and verbal short-term memory subtests of the *Schlichting* (Schlichting, van Eldik, lutje Spelberg, van der

Meulen, & van der Meulen, 2003). All four subtests had a discontinuation rule. Raw scores were the number of correct items per subtest, for which norm-based standard scores were derived from the manual. The standardized scores of the four subtests were averaged to obtain a verbal IQ score. A more elaborate description of the subtests and the resulting verbal intelligence score is provided by van Bergen, de Jong, Maassen, Krikhaar et al. (2014). Cronbach's α for the subtests is .90, .90, .86, and .79, respectively.

Vocabulary knowledge/verbal reasoning ability at the end of Grade 3 (age 9) was assessed using the Dutch version (Kort et al., 2005) of the vocabulary subtest of the *Wechsler Intelligence Scale for Children – Third Edition* (WISC-III; Wechsler, 1991). Children were asked to describe the meaning of a given word. The subtest contains 35 items that increase in difficulty and level of abstraction. Correct answers were awarded with 2 points, incomplete answers with 1 point. Raw scores are transformed into age-referenced Wechsler standard scores (M = 10, SD = 3). Reliability of the subtest for this age group is .80.

To determine the dyslexia status of the children at the end of Grade 2, word reading fluency was measured using the Dutch timed *Drie-minuten-toets* (DMT; Verhoeven, 1995). The task comprised a list of 150 monosyllabic words, with all words containing at least one consonant cluster. Children had one minute to read aloud as many words as possible. The raw score was the number of correctly read words within the time limit, which was transformed into a norm-based standard score for classification purposes. Reliability of the task for this age is .96.

ANALYSES

First, repeated measures analyses with planned contrasts (α = .05) were conducted on the N-CDI parent report measures to investigate group differences in receptive and expressive vocabulary between 17 and 35 months. For each vocabulary type, group membership was the between-subjects factor and time point (1 to 4) the within-subject factor. Two follow-up contrasts were specified, comparing 1) FR-dyslexic children to both nondyslexic groups (i.e., FRD vs. FRND and TDC), and 2) both nondyslexic groups (i.e., FRND vs. TDC). Cohen's d was used as a measure of effect size (i.e., .2 = small, .5 = medium, .8 = large; Cohen, 1992).

Next, separate latent growth curve models were fitted to describe receptive and expressive vocabulary development across the four time points using the R package lavaan (R Core Team, 2015) for structural equation modeling (SEM). The baseline model contained the four time points as indicator variables (i.e., N-CDI measures at 17, 23, 29, and 35 months), an initial status factor (i.e., intercept) and a linear change factor (i.e., linear slope). The intercept represents the initial vocabulary size of the children at the start of the study, with equal factor loadings for all four indicators (i.e., fixed at 1). The linear slope reflects children's average linear growth in vocabulary between time points throughout the study, with factor loadings indicating equal intervals between time points (i.e., 0, 1, 2, 3). If the fit of the baseline model was considered insufficient, factors representing nonlinear growth (e.g., quadratic term) could be added to the model to better approximate the average growth curve of the children in the study. When model fit was considered acceptable, we regressed the latent growth curve factors on the (dummy coded) independent variables FR status (FR vs. no FR), dyslexia status (dyslexia vs. no dyslexia), and gender (boy vs. girl). Thereby we could determine the differences between the three groups (i.e., FRD, FRND, and TDC) in their vocabulary development. To evaluate exact and approximate model fit, we used the chi-square value (χ^2) with associated *p*-value, root mean square error of approximation (RMSEA) including *p*close, comparative fit index (CFI), and standardized root mean square residual (SRMR; Kline, 2011; Little, 2013). For good model fit, chi-square should have a non-significant *p*-value (i.e., > .05), RMSEA should be < .05 (< .08 is acceptable), with *p*close > .05, CFI being > .95 (> .90 is acceptable), and SRMR being < .05 (< .08 is acceptable; Kline, 2011; Little, 2013).

RESULTS

DATA SCREENING

Outlier analysis showed no significant univariate or multivariate outliers. Each occasion contained missing receptive and expressive vocabulary scores (T1: 33 children, range across groups 8.7-21.6%; T2: 24 children, 8.7-13.7%; T3: 15 children, 5.8-8.7%; T4: 20 children, 5.4-14.5%). For the repeated measures analyses, raw scores were used and missing data were imputed using expectation maximization. Means and standard deviations were similar for the original and imputed data. The imputed data were also transformed into gender-based norm-referenced percentile scores, which were available for the first three time points (norms are available up to 30 months). For the SEM analyses, missing data were dealt with using full information maximum likelihood estimation. Raw scores were transformed into sample-based z-scores over all four time points combined to facilitate the estimation of the growth models.

MULTIVARIATE ANALYSES

Table 2 displays the receptive and expressive vocabulary scores of the three groups at the four time points. The group contrasts are reported for the main and interaction effects. For receptive vocabulary, the FRD group had marginally lower vocabulary scores than both nondyslexic groups over all time points taken together, Wilks' $\lambda = 0.96$, $F(4, 206) = 1.98$, $p = .098$, as well as a lower increase in vocabulary size over time, Wilks' $\lambda = 0.97$, $F(3, 207) = 2.50$, $p = .06$. The FRND and TDC groups did not differ on either of the effects. For the individual time points, both nondyslexic groups outperformed the FRD group in receptive vocabulary size at 23 ($p = .02$), 29 ($p = .02$), and 35 months ($p = .04$), but not at the youngest age of 17 months ($p = .35$). The effect sizes are all considered small. There were no significant differences between the FRND and the TDC groups at any time point (see also Figure 1).

For expressive vocabulary, the FRD group had significantly lower vocabulary scores than both nondyslexic groups for all time points overall, Wilks' $\lambda = 0.94$, $F(4, 206) = 3.04$, $p = .02$, as well as a lower increase in vocabulary size over time, Wilks' $\lambda = 0.94$, $F(3, 207) = 4.00$, $p = .008$. The FRND and TDC groups did not differ significantly on either of the effects. Contrasts at the individual time points confirmed that the FRD group had significantly smaller expressive vocabulary size than both nondyslexic groups at all four occasions ($p < .05$). Effect sizes were small to medium. There were no significant differences between the FRND and TD control group (see also Figure 1).

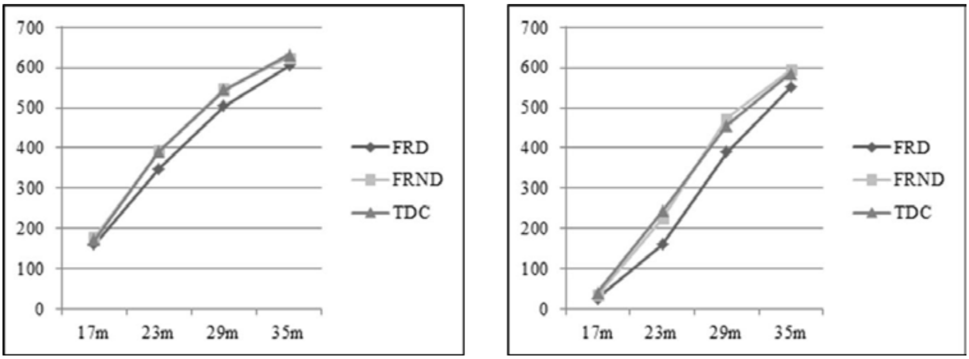

Figure 1. Mean development of receptive (left) and expressive (right) vocabulary size in the three groups across four time points. *Note*. See also Table 2. FRD = family risk dyslexia; FRND = family risk no dyslexia; TDC = typically developing control.

The small mean group differences in vocabulary size were partly supported by the mean percentages of children with mild (i.e., ≤ 25th percentile) and severe (i.e., ≤ 10th percentile) vocabulary impairments (Table 2). For receptive vocabulary, the FRD group contained significantly more children with mild vocabulary impairments than both nondyslexic groups ($p = .03$), but not more children with severe impairments. For expressive vocabulary, differences between the FRD group and both nondyslexic groups were marginally significant for mild vocabulary impairments (p = .06), and not significant for the percentage of severe impairments.

LATENT GROWTH CURVE MODELING

Table 3 provides the correlations among receptive and expressive vocabulary at the four time points, FR status, dyslexia status, and gender. The correlations of receptive and expressive vocabulary across consecutive occasions were strong, indicating substantial stability of individual differences in vocabulary development. Except for receptive vocabulary at 17 months, dyslexia status correlated significantly with receptive and expressive vocabulary. FR status was associated with expressive vocabulary at the first two time points only and also related to dyslexia status. Gender was significantly related to expressive vocabulary at the last three time points.

TABLE 2

Means, Standard Deviations, and Group Comparisons of the Receptive and Expressive Vocabulary Scores and Percentiles

	FRD (n = 51)		FRND (n = 92)		TDC (n = 69)		FRD vs. FRND	FRD vs. TDC	FRND vs. TDC
Variable	*M*	*SD*	*M*	*SD*	*M*	*SD*	Cohen's *d*		
Receptive vocabulary									
17 months[$]	161.39_a	89.65	178.49_a	95.99	172.07_a	89.54	-0.18	-0.12	0.07
23 months	348.40_a	133.90	393.80_b	116.53	391.77_b	113.02	-0.36	-0.35	0.02
29 months	504.59_a	114.11	548.18_b	105.88	543.32_b	96.70	-0.40	-0.37	0.05
35 months	605.29_a	78.95	625.08_b	69.43	631.19_b	60.46	-0.27	-0.37	-0.09
Percentiles									
% ≤ 25 percentile[#]	41.8_a		25.0_b		25.6_b				
% ≤ 10 percentile[#]	17.0_a		9.8_a		8.2_a				
Expressive vocabulary									
17 months[$]	25.74_a	22.78	34.93_b	37.36	41.37_b	33.19	-0.30	-0.55	-0.18
23 months	160.33_a	124.75	225.52_b	145.52	243.48_b	143.46	-0.48	-0.62	-0.12
29 months	389.87_a	155.44	473.37_b	134.32	455.61_b	141.77	-0.28	-0.44	0.13
35 months	550.82_a	110.37	594.36_b	85.66	585.49_b	102.17	-0.44	-0.33	0.09
Percentiles									
% ≤ 25 percentile[#]	39.2_a		26.1_a		24.1_a				
% ≤ 10 percentile[#]	18.9_a		11.9_a		10.1_a				

Note. Means in the same row that do not share subscripts differ at $p < .05$. FRD = family risk dyslexia; FRND = family risk no dyslexia; TDC = typically developing control.
[$] Data of this time point are partly described in Koster et al. (2005). [#] Average percentage over the first three time points.

TABLE 3

Pearson's Correlations between Receptive and Expressive Vocabulary Measures and Independent Variables per Time Point

Variables	1.	2.	3.	4.	5.	6.	7.	8.	9.	10.	11.
1. Receptive 17m	1.00										
2. Receptive 23m	.79**	1.00									
3. Receptive 29m	.58**	.80**	1.00								
4. Receptive 35m	.46**	.68**	.84**	1.00							
5. Expressive 17m	.52**	.54**	.40**	.32**	1.00						
6. Expressive 23m	.56**	.74**	.61**	.51**	.76**	1.00					
7. Expressive 29m	.53**	.72**	.85**	.71**	.54**	.75**	1.00				
8. Expressive 35m	.38**	.61**	.77**	.84**	.34**	.56**	.85**	1.00			
9. Family risk status	.00	-.06	-.05	-.09	-.14*	-.14*	-.04	-.03	1.00		
10. Dyslexia status	-.07	-.16*	-.17*	-.14*	-.15*	-.22**	-.22**	-.17*	.39**	1.00	
11. Gender	.08	.12	.11	.14*	.08	.15*	.17*	.16*	.03	.02	1.00

Note. Correlations are based on EM imputed data. For family risk status, 0 = no family risk, 1 = family risk; for dyslexia status, 0 = no dyslexia, 1 = dyslexia; for gender, 0 = boy, 1 = girl. m = months.
$^*p < .05$. $^{**}p < .01$.

RECEPTIVE VOCABULARY

The baseline model for receptive vocabulary could not be fitted due to a negative variance (i.e., Heywood case) of the T4 indicator. Therefore, the residual variance of T4 was constrained to be larger than zero. The fit of this baseline model was poor, $\chi^2(6) = 321.49$, $p < .001$, RMSEA = .50, *p*close = .00, CFI = .39, SRMR = .26. Based on the expected developmental trajectory and the shape of the growth curves displayed in Figure 1, a quadratic term was added to the model (factor loadings fixed at 0, 1, 4, and 9), indicating nonlinear growth. This model could not be fitted due to a negative variance of the T1 indicator. After restricting this variance to be larger than zero, the model fitted the data significantly better than the model without the quadratic growth parameter, $\Delta\chi^2(3) = 312.76$, $p < .001$, and demonstrated the following fit, $\chi^2(3) = 8.73$, $p = .03$, RMSEA = .095, *p*close = .12, CFI = .99, SRMR = .03. Although the model fit is not optimal given the significant chi-square value and the RMSEA exceeding .08, it was considered acceptable for the purposes of our analyses based on the good CFI and SRMR values (Kline, 2011; Little, 2013). In addition, the parameter estimates showed no indication of systematic patterns of misfit.

The variance (0.24, *SE* = .03) of the intercept was significant ($p < .001$), indicating that the children showed significant variability in receptive vocabulary size at 17 months. Also, the variances of the linear (0.18, *SE* = .03) and the quadratic slope (0.02, *SE* = .003) were significant ($p < .001$), indicating individual differences in developmental trajectories. Overall, the quadratic growth model explains about 85% of the observed total standardized variance in receptive vocabulary growth between 17 and 35 months.

For the final model, the latent growth curve factors were regressed on the independent variables FR status, dyslexia status, and gender to assess group differences in receptive vocabulary development. This final model demonstrated good model fit, $\chi^2(6) = 10.79$, $p = .10$, RMSEA = .06, *p*close = .32, CFI = .99, SRMR = .02. There was a significant effect of dyslexia status on both the linear slope and the quadratic slope, but not on the intercept. There were no significant effects of FR status and gender on receptive vocabulary development. The standardized coefficients in Figure 2 indicate that, when controlling for FR status and gender, FR-dyslexic children had lower levels of linear growth in receptive vocabulary than nondyslexic children. However, they also had higher overall quadratic growth, indicating more change in the rate of growth over time (Bollen & Curran, 2006). Overall the three independent variables explained 1.1% of the variance in the intercept, 4.1% in the linear slope, and 5.0% in the quadratic slope. A detailed overview of the parameter estimates is provided in Appendix A.

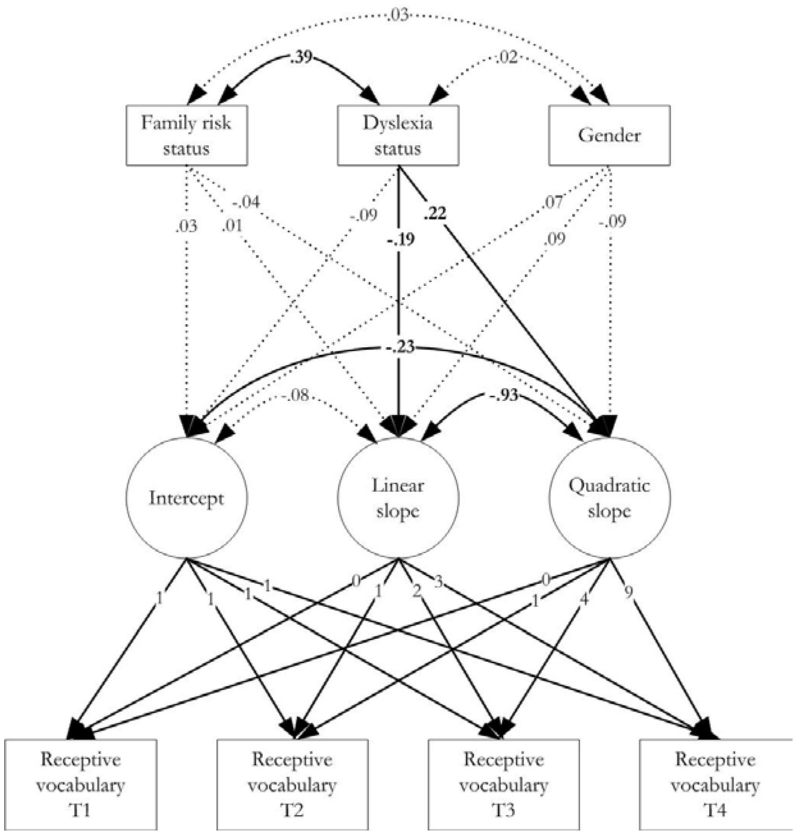

Figure 2. Latent growth curve model of receptive vocabulary with family risk status, dyslexia status, and gender as predictors. *Note.* (Cor)relations between latent factors and predictors are standardized. To aid visibility, error terms are not displayed. Solid lines and bold numbers indicate significant effects ($p < .05$). T = time point.

EXPRESSIVE VOCABULARY

The fit of baseline model for expressive vocabulary, after constraining the residual variances of T1 and T4 to be larger than zero, was poor, $\chi^2(7) = 245.33$, $p < .001$, RMSEA = .40, *p*close < .001, CFI = .50, SRMR = .27. In line with the model for receptive vocabulary, a quadratic growth parameter was added to the model (factor loadings fixed at 0, 1, 4, and 9). This model fitted the data significantly better than the model without the quadratic term, $\Delta\chi^2(4) = 170.98$, $p < .001$, but the fit was still unacceptable, $\chi^2(3) = 74.35$, $p < .001$, RMSEA = .34, *p*close < .001, CFI = .85, SRMR = .08. Given the severe nonlinearity of the mean development displayed in Figure 1, a separate logistic curve[1] (following an S-shape) was fitted to the data using the R package OpenMx (R Core Team, 2015). This logistic curve model, which was not nested under the previous model (so without the linear and quadratic slopes), fitted the data better that the previous model. Based on the CFI and SRMR values and plausible parameter estimates (Kline, 2011; Little, 2013), this model fit was considered sufficient for the final baseline model, $\chi^2(1) = 17.58$, $p < .001$, RMSEA = .28, *p*close < .001, CFI = .96, SRMR = .07.

The variance of both the intercept (0.02, *SE* = .002) and change factor (2.06, *SE* = .75) were significant ($p < .05$), indicating that children showed significant variability in expressive vocabulary size at 17 months as well as individual variability in developmental trajectories. Overall, the logistic curve model explains about 82% of the observed total standardized variance in expressive vocabulary growth between 17 and 35 months.

The latent growth curve factors were regressed on FR status, dyslexia status, and gender to assess group differences in expressive vocabulary development. The fit of this final model was considered acceptable[2], $\chi^2(4) = 20.38$, $p < .001$, RMSEA = .14, *p*close = .006, CFI = .97, SRMR = .05. The results showed a significant effect of both gender and dyslexia status on the change factor. There was no significant effect of FR status on expressive vocabulary development. The standardized coefficients in Figure 3 show that girls had higher growth rates than boys. When controlling for FR status and gender, FR-dyslexic children had lower levels of growth in expressive vocabulary than nondyslexic children. Overall the three independent variables explained 0.4% of the variance in the asymptote, 3.8% in the intercept, and 8.8% in the change factor. See Appendix B for an overview of the parameter estimates.

[1] Browne (1993) shows how a logistic curve can be used in latent growth curve modeling. The logistic latent curve model is defined using three latent factors, where the parameterization is chosen such that the first factor (alpha) represents potential performance (i.e., asymptote), the second (beta) represents initial performance (i.e., intercept), and the third (rho) represents learning speed (i.e., change factor). The factor loadings of these three factors are modelled using a target function and three specific basic functions that are given by Browne (1993, p. 179).

[2] Given that linear, quadratic, and logistic growth factors cannot be combined into one latent growth curve model, and that the mean development of the groups in Figure 1 shows that the S-curve is not as clearly defined in the TD control group as in both FR groups, optimal model fit cannot be obtained. The logistic curve model was chosen as the final model because it fitted the data significantly better than the linear + quadratic curve model, model fit was acceptable based on the CFI and SRMR, and the parameter estimates showed no indication of systematic patterns of misfit.

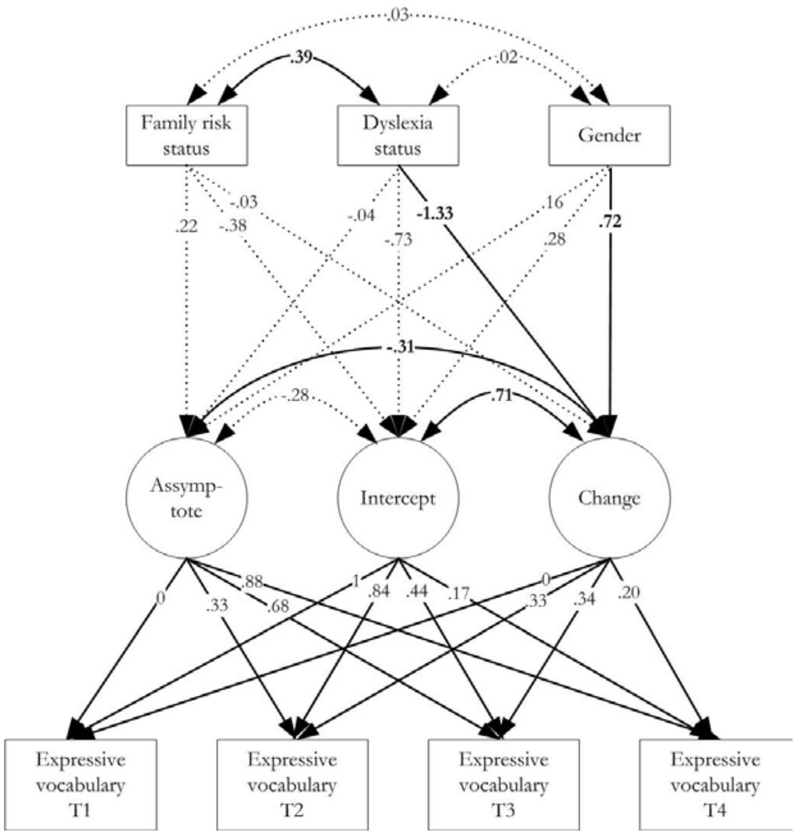

Figure 3. Logistic latent growth curve model of expressive vocabulary with family risk status, dyslexia status, and gender as predictors. *Note.* (Cor)relations between latent factors and predictors are standardized. To aid visibility, error terms are not displayed. Solid lines and bold numbers indicate significant effects ($p < .05$). T = time point.

DISCUSSION

This study examined vocabulary development among children with a family risk (FR) of dyslexia who became dyslexic (FR-dyslexic), FR children who did not become dyslexic (FR-nondyslexic), and typically developing (TD) control children. Growth of early receptive and expressive vocabulary was assessed longitudinally across four occasions between the ages of 17 and 35 months using parent reports. The first aim of the study was to determine whether receptive and expressive vocabulary were associated with FR or with dyslexia status (risk versus reading). The second aim was to examine whether and how the vocabulary growth curves of FR-dyslexic children differ from FR-nondyslexic children and/or TD control children (delay versus deviance of growth).

Consistent with the results of previous studies (Duff et al., 2015; Scarborough, 1990; Torppa et al., 2010), neither receptive nor expressive vocabulary development differed between FR-nondyslexic and TD control children. The overall vocabulary size of FR-dyslexic children was smaller than in both nondyslexic groups (with small effect sizes for receptive vocabulary and small to medium for expressive vocabulary). These results support earlier findings that infant vocabulary development is related to dyslexia status and not to FR. However, unlike previous studies (Scarborough, 1990; Torppa et al., 2010) we found lower receptive vocabulary in FR-dyslexic children already at 23 months and lower expressive vocabulary already at 17 months. Moreover, our results showed that the differences between the FR-dyslexic children and both nondyslexic groups were persistent, at least between the ages of 17 and 35 months, as lower scores were present at all subsequent time points after the first occurrence.

In addition to these mean differences, latent growth curve modeling showed differences in growth trajectories between FR-dyslexic and FR-nondyslexic/TD children. For receptive vocabulary, growth curves were nonlinear and showed a gradual increase and subsequent shallowing of the slope. This suggests that our study started somewhere halfway in the vocabulary spurt and extended past the peak in growth rate toward the end of the spurt. Based on the finding that FR-dyslexic children showed more change in the rate of growth over time, it can be concluded that receptive vocabulary development in these children was characterized by lower initial growth followed by a weaker deceleration of the growth rate (see also Figure 1). This pattern was supported by the change in effect sizes between time points, indicating an increase in differences between FR-dyslexic and nondyslexic children in receptive vocabulary size, followed by a decrease in mean differences. Thus, the expansion of receptive vocabulary during the second year of life was slower in the group of FR-dyslexic children. Their growth was protracted and the deceleration reduced toward the start of their third year. As a result, they partly resolved their lag. These findings are not in line with previous studies by Fenson et al. (1994) and Hamilton et al. (2000), who found a linear increase in early receptive vocabulary growth. However, both studies focused on a different time span (i.e., 8 to 28 months and 12 to 25 months, respectively), partly involving younger children than in our study. As a result, these studies might have captured neither the start nor the end of the vocabulary spurt, and thus missed the deceleration in vocabulary growth after the 28 month mark.

For expressive vocabulary, growth curves were also nonlinear and seemed to cover the full vocabulary spurt, with an acceleration in growth during the second year and subsequent deceleration during the third year. Expressive vocabulary development in FR-dyslexic children was characterized by a later onset of the spurt. Receptive and expressive vocabulary growth curves of FR-dyslexic children differ from those of FR-nondyslexic and TD children based on lower initial growth rates, indicating delayed vocabulary development, followed by partial recovery. This nonlinearity in early expressive vocabulary was also attested in studies by Fenson et al. (1994), Hamilton et al. (2000) and Kauschke and Hofmeister (2002). Only the latter found the full vocabulary spurt. This is probably due to the time window included (i.e., 13 to 36 months) , which is more comparable to the one covered in the current study.

The findings of the present study speak to theories linking early vocabulary growth to (impairments in) the development of phonological skills and later literacy. The smaller vocabulary size and delayed development in FR-dyslexic children was expected based on both the lexical restructuring theory (LRT) and segmentation theory. However, our findings on the severity of these deficits and findings for the FR-nondyslexic children do not seem to be in line with the direct causal relation between vocabulary growth and emerging phonological awareness proposed in the LRT (e.g., Metsala & Walley, 1998). Because FR-nondyslexic children have no vocabulary impairment, LRT predicts that these children would not have phonological awareness (PA) deficits either. Yet, multiple studies have shown a stepwise pattern for phonological awareness (i.e., FR-dyslexic < FR-nondyslexic < TD control; e.g., Moll et al., 2013), including our own longitudinal study on largely the same sample (van Bergen, de Jong, Maassen, & van der Leij, 2014). The mild phonological awareness impairments that may arise in FR-nondyslexic children thus do not seem to be caused by vocabulary only but also by other cognitive skills. This is further supported by the continual absence of mild vocabulary deficits in the FR-nondyslexic group at later ages (e.g., van Bergen, de Jong, Maassen, Krikhaar et al., 2014; see also Table 1).

Furthermore, on the basis of the LRT the vocabulary delay of the FR-dyslexic children would be anticipated to be more severe or even clinical to causally relate the development of phonological awareness to vocabulary growth. However, the majority of the children in our study did not show clinical levels of vocabulary impairment. Although lexical restructuring is an ongoing process which extends across childhood and may vary per developmental phase (Metsala et al., 2009; Rispens & Baker, 2012), it seems unlikely that small deficits in infant vocabulary increase during early childhood, explaining the larger deficits in phonological awareness; the FR-dyslexic children had partly recovered from their acquired lag toward the end of their third year. Accordingly, the relation between vocabulary and phonological awareness seems to be less straightforward than suggested in LRT.

The patterns of vocabulary development observed in the current study seem more in line with the segmentation theory (e.g., Boada & Pennington, 2006). For FR-dyslexic children, problems with speech sound segmentation hamper the formation of high-quality phonological representations, which is associated with more negative outcomes for vocabulary growth and phonological awareness. The vocabulary deficits in this group, despite their stability, are based on small effects. These results, combined with the absence of mild impairments in FR-nondyslexic children, are in line with the limited causality implied by this theory. Consequently, it could be that phonological representations only partly account for vocabulary and that other factors, such as syntactic complexity, lexical richness, and input clues to word meaning (see Hoff, 2006, for an overview), play a substantial role. The segmentation theory could also account for the finding that not all children with dyslexia have vocabulary difficulties and that vocabulary might even be a source of compensation for some children (e.g., Snowling et al., 2003; van Viersen, Kroesbergen, Slot, & de Bree, 2014).

The results of the current study also suggest reasons why early vocabulary deficits were often not found in previous FR studies. First, the current findings clearly show that delays in vocabulary development are only found in FR-dyslexic children. A comparison between only TD and FR children is less likely to yield group differences; the effect of early vocabulary is small and only found in a small part of the FR group. Secondly, previous FR studies often suffered from lack of power due to small sample sizes and probably large variety in the percentage of FR children that would later become dyslexic in the at risk group (see Aro et al., 2012 for an exception), affecting replicability of effects. Our results show that, based on a developmental approach and a larger sample than in previous studies, vocabulary is indeed a small, additional risk factor for the development of later reading impairment.

Concerning practical implications, our study suggests that early screening of vocabulary development based on parent reports may be valuable for detecting language delays, but might be of lower predictive value for the detection of (being at risk for) later word reading deficits. Yet, the co-occurrence of deficits in receptive and expressive vocabulary and/or accumulation of vocabulary deficits and other risk factors might increase the impact on later literacy development. As vocabulary is not only relevant for word reading ability, but also for reading comprehension (Lee, 2011; Muter, Hulme, Snowling, & Stevenson, 2004; Ricketts, Nation, & Bishop, 2007), early screening might be beneficial. Although the delay in vocabulary growth of FR-dyslexic children may seem to be (partly) resolved toward the end of the third year of life (see also Rescorla, 2011), deficits may increase in predictive value for literacy development around the age of school entry (e.g., Thompson et al., 2015). They may also arise again when children start learning to read, illustrating the reciprocal nature of the relation between vocabulary, phonology, and literacy at later ages (e.g., Snowling, Hulme, & Nation, 1997; see also Snowling, Duff, Nash, & Hulme, 2015).

Overall, this study has shown that vocabulary is weakly related to the etiology of dyslexia. Early deficits are associated with children's dyslexia status and not with their FR. Early vocabulary development of FR-dyslexic children is characterized by a delay compared to FR-nondyslexic and TD control children and does not seem deviant. As such, vocabulary may function as an additional risk factor for the development of later reading impairments rather than as a proximal cause of dyslexia.

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CHAPTER 3

PATHWAYS INTO LITERACY: THE EFFECTS OF EARLY ORAL LANGUAGE ABILITIES AND FAMILY RISK FOR DYSLEXIA

The present study investigated the influence of early oral language and family risk (FR) for dyslexia on the two developmental pathways toward reading comprehension, that is through word reading and through (oral) language abilities. The sample included 237 children (164 at FR) from the Dutch Dyslexia Program. Longitudinal data covered seven occasions between 4 and 12 years. The effect of early oral language ability on reading comprehension was mediated by preliteracy skills and word decoding ability for the first pathway and by later language abilities for the second pathway. FR affected literacy development through its subsequent effects on preliteracy skills, word decoding, and reading comprehension. FR did not have an effect on the development of (early) oral language abilities.

Keywords: Early language, family risk, dyslexia, literacy development, reading comprehension.

van Viersen, S., de Bree, E. H., van der Leij, A., Maassen, B., & de Jong, P. F. (submitted). Pathways into literacy: The effects of early oral language abilities and family risk for dyslexia. *Psychological Science*.

INTRODUCTION

Early oral language abilities are believed to form the foundation of two pathways into literacy development that both lead to the acquisition of reading comprehension (Oakhill & Cain, 2012; Storch & Whitehurst, 2002; see also Hoover & Gough, 1990). In the first pathway oral language abilities affect the development of preliteracy skills, such as phonological awareness (PA) and letter knowledge (Caravolas et al., 2012), which function as precursors to the acquisition of word decoding skills. Word decoding skills are in turn involved in reading comprehension. The second pathway continues to hinge on oral language abilities, such as vocabulary and grammar development, to foster linguistic comprehension, which is also essential for reading comprehension (Storch & Whitehurst, 2002). The main aim of the present study was to examine the effect of early language on developmental trajectories toward reading comprehension in children with and without a family risk (FR) of dyslexia. As children at FR of dyslexia have a higher risk of developing word decoding problems, a second aim was to assess whether the effect of FR on reading comprehension is fully mediated by preliteracy and word decoding skills or whether FR also affects the pathway that runs independently via language abilities toward the development of reading comprehension.

Multiple studies have shown that having an FR of dyslexia affects literacy development and can be considered an important risk factor for the development of literacy difficulties (see Snowling & Melby-Lervåg, 2016, for an overview). Only a few longitudinal FR studies have focused on how early language skills influence literacy development through either preliteracy skills or later language abilities, or both. In one of these studies, Duff, Reen, Plunkett, and Nation (2015) measured early vocabulary in British children during infancy at the age of 16 to 24 months and later vocabulary, PA, reading accuracy, and reading comprehension during a follow up when children were 5 to 9 years old. Their study showed that early vocabulary predicted later vocabulary and PA as well as reading accuracy and reading comprehension. These findings indicate that early language has an effect on both word decoding and reading comprehension at some point during development.

A Finnish study by Torppa, Lyytinen, Erskine, Eklund, and Lyytinen (2010) gives a more detailed account of the pathway from early language toward word reading accuracy and fluency. In this study language and preliteracy skills were assessed at several intermediate occasions between the age of 2.5 and 9 years. They found that early language skills at 2.5 years predicted language and preliteracy skills (i.e., PA, rapid automatized naming [RAN], and letter knowledge) at 3.5 years, whereas expressive language skills at 3.5 years predicted preliteracy skills at 5.5 years. Preliteracy skills at 5.5 years in turn predicted word reading ability at 9 years. This study shows that the effect of early language on word decoding skills runs via preliteracy skills and that there were no direct effects of early language on word decoding ability.

An even more extensive study was reported by Hulme, Nash, Gooch, Lervåg, and Snowling (2015), investigating both pathways toward reading comprehension in a British sample of children and tracking performance on several occasions between 3.5 and 8 years. This study focused on the influence of early speech and language factors on literacy development and included preliteracy skills and word-level literacy as mediating variables. Concerning the first path, their model showed that language skills at 3.5 years predicted preliteracy skills (i.e., PA, RAN, and grapheme-phoneme knowledge) at 4.5 years, which in turn predicted word-level literacy at 5.5 years. A significant indirect effect of language skills on word-level literacy through preliteracy skills was also found. Early language at 3.5 years and word-level literacy around 5.5 years both had an independent effect on reading comprehension. This study shows that early language also has a direct effect on reading comprehension, besides the indirect effect through preliteracy skills and word decoding ability. This suggests the existence of an independent pathway based on language abilities.

Overall, these studies show a clear role for (early) language skills as a foundation for literacy development. However, the results for the effects of having an FR of dyslexia on the two pathways into literacy are less clear. Although all studies included children at risk for dyslexia, they differed in the way the effects of FR were modeled. Torppa et al. (2010) combined the samples of children with and without an FR of dyslexia and therefore could not trace the effects of having an FR. Hulme et al. (2015) modelled literacy development longitudinally for typically developing control and FR children separately. Their findings showed that both pathways in literacy development were present, for control children as well as for children with an FR of dyslexia. Duff et al. (2015) explicitly modelled the effect of FR during different stages of literacy development by including it as a predictor in their model. They showed that FR status only had an effect on later literacy outcomes (i.e., reading accuracy and reading comprehension), but not on (early) vocabulary or PA. Thus, the results of Duff et al. (2015) suggest that the effect of FR on literacy development does not run through early language. Taking this approach to modeling the effect of FR while including more mediating occasions may reveal more about whether and/or how FR influences the development toward reading comprehension through either or both pathways.

The goal of the current study was to track developmental trajectories toward literacy in a large sample of children with and without an FR of dyslexia. We add to findings of previous studies in three ways. First, we investigated the influence of early language abilities on reading comprehension through two independent pathways, building on word decoding and language abilities, while including mediating variables at multiple time points. Related, as early language abilities are associated with general abilities, we controlled for nonverbal IQ to be able to examine whether it is specifically the oral language abilities that influence both pathways. Second, we conducted the study in Dutch. Although most previous studies only included word reading accuracy, we also assessed word reading fluency as this is deemed a better indicator of word reading ability in transparent orthographies, such as Dutch (e.g., de Jong & van der Leij, 2003). Third, we examined whether the effect of FR on reading comprehension is fully mediated by preliteracy skills and word decoding ability or whether FR also has an independent effect through language abilities.

METHOD

DESIGN

The current study is based on data from the Dutch Dyslexia Program (DDP), a prospective longitudinal study of children with and without an FR of dyslexia (see van der Leij et al., 2013, for an overview). Data were collected throughout development, from age 2 months until the end of 6^{th} grade. Data from assessments before the age of 4 years are not reported on here. In this study, data from seven assessments are reported. Here, T1 and T2 were during the first year of kindergarten (at 4 and 4.5 years), T3 at the end of the second year of kindergarten (6 years), T4 at the beginning of Grade 2 (7.5 years), T5 at the end of Grade 2 (8 years), T6 at the end of Grade 3 (9 years), and T7 in Grade 6 (12 years). Recruitment of families with and without an FR who were expecting a baby was centered around three major urban areas across The Netherlands (i.e., Amsterdam, Nijmegen, and Groningen). The sample included twice as much at risk families as control families.

PARTICIPANTS

The total sample consisted of 237 children, including 164 FR children and 73 control children. Children were considered to be at FR when they had at least one parent and one first degree relative with dyslexia. FR status was further confirmed by assessing the (non)word reading fluency and verbal reasoning skills of the parents. Van Bergen, de Jong, Plakas, Maassen, and van der Leij (2012) provide a more detailed account of FR assessment in this sample. Six control children without an FR turned out to be dyslexic and were excluded from the study. All parents signed written informed consent. Ethical approval for the study was provided by the faculty's ethical committee.

INSTRUMENTS

An overview of the measures per time point and the corresponding Grades is provided in Table 1.

IQ AT T1 AND T2. Nonverbal intelligence was assessed at T1 using six subtests of the *SON-R* test battery (Tellegen, Winkel, Wijnberg-Williams, & Laros, 1998). The subtests patterns, object assembly, and block design are all performance tasks, which measure visuospatial processing, visual-motor coordination, and persistence. The reasoning tasks, tapping fluid reasoning about concrete and abstract categories, are the picture completion, analogies, and categories subtests. Raw scores per subtest were transformed into norm-based standard scores (M = 10, SD = 3) and used for the analyses. Test-retest reliabilities of the subtests vary between .48 (picture completion) and .64 (block design and categories; Evers et al., 2009-2012).

At T2, verbal intelligence was measured using the subtest language comprehension of the *Reynell* (van Eldik, Schlichting, lutje Spelberg, van der Meulen, & van der Meulen, 2001) and the subtests expressive syntax, expressive vocabulary, and verbal short-term memory of the *Schlichting* (Schlichting, van Eldik, lutje Spelberg, van der Meulen, & van der Meulen, 2003). Norm-based standard scores (M = 100, SD = 15) per subtest were used for the analyses. A more elaborate description of the (non)verbal intelligence tests and the details of the individual subtests is provided by van Bergen, de Jong, Maassen, Krikhaar, Plakas, and van der Leij (2014). Reliabilities of the subtests were .90, .90, .86, and .79, respectively.

PRELITERACY SKILLS AT T3 AND T4. Letter knowledge at T3 was measured using a grapheme production task (Verhoeven, 1992) and a letter recognition task (van Otterloo, 2000). Children had to read a list of 34 Dutch graphemes as quickly and accurately as possible for the production task. For the recognition task, children were verbally presented with a letter sound, for which they had to choose the correct grapheme out of six alternatives. This task contained 32 items. The number of correctly produced or recognized graphemes for the separate tasks were used in the analyses.

PA at T3 was measured using a phoneme blending task (Verhoeven, 1993a) and a phoneme segmentation task (Verhoeven, 1993b). For the blending task, children had to form a word out of several consecutive sounds, e.g., /*aa*/ /*p*/ → *aap* (monkey). In the segmentation task children had to divide a given word into its separate phonemes, e.g., *uur* (hour) → /*uu*/ /*r*/. Both tasks contained 20 one-syllable items of increasing difficulty (i.e., from C-V to C-V-C-C-C items). The task was terminated when a child had no correct answers within a set of items of equal difficulty. Raw scores (i.e., number of correct items) were used in the analyses.

At T3, RAN of colors (red, green, black, blue, yellow) and objects (tree, duck, chair, scissors, bicycle; van den Bos, 2003) was used to measure naming speed. For both subtests, children had to name items as quickly and accurately as possible. Each subtest contained a list of 50 items. The time to completion was used to compute the number of named items per minute, which was the score used in the analyses. At T4, the objects subtest was replaced with the digits subtest (2, 4, 5, 8, 9) to include both an alphanumeric and non-alphanumeric subtest in the analyses. Internal consistency of the subtests varied between .87 and .93 (Evers et al., 2009-2012).

PA at T4 was measured using a phoneme deletion task (de Jong & van der Leij, 2003). Children had to delete a target phoneme from a nonword, which resulted in another nonword. The test consisted of 18 items, covering nine monosyllabic and nine disyllabic nonwords and was preceded by two practice items. Testing was terminated after six consecutive errors. Raw scores were the number of correct items.

LANGUAGE SKILLS AT T6 AND T7. At T6, vocabulary knowledge was assessed using the Dutch version (Kort et al., 2005) of the vocabulary subtest of the *Wechsler Intelligence Scale for Children – Third Edition* (WISC-III; Wechsler, 1991). The subtest contained 35 concepts, increasing in level of abstraction, for which children had to describe its meaning. Children received 2 points for correct answers and 1 point for incomplete answers. The subtest was terminated after four consecutive errors. Raw scores were transformed into age-referenced standard scores (M = 10, SD = 3), which were used for the analyses. The reliability is .80 for this age group (i.e., 9 years).

At T7, we tested vocabulary knowledge using version A of the *DiaWoord*, which is part of the online *Diataal* test battery (Hacquebord, Stellingwerf, Linthorst, & Andringa, 2005). Children had to provide the definition of 50 words that were presented in minimal context sentences. The items varied in difficulty. The outcome is a competence score, based on the sum of all correct items and their difficulty weights, that reflects the vocabulary level of the children. The reliability is .82.

WORD READING AT T4 THROUGH T7. (Non)word reading accuracy at T4 and T5 was measured using a word and a nonword task (de Jong & Wolters, 2002). Children were asked to read a list of (non)words, consisting of 40 items each. Items on both lists increase in difficulty from one to four syllables. There was no time limit; the tasks were terminated when six errors in the last eight items occurred. Number of correctly read (non)words was the score used in the analyses. At T6, only the nonword accuracy task was administered due to a lack of complexity in the word accuracy task for children at this age.

Word reading fluency at T4 and T5 was measured using two lists of the *Drie-minuten-toets* (DMT; Verhoeven, 1995). Both lists contain 150 monosyllabic words. The first list covers simple words with a CV, VC, or CVC pattern. The second list includes more difficult words with at least one consonant cluster. Raw scores were the number of correctly read words within one minute per list, which were used in the analyses.

At T6 and T7, reading fluency was measured using a word reading task (EMT; Brus & Voeten, 1999) and a nonword reading task (Klepel; van den Bos, lutje Spelberg, Scheepstra, & de Vries, 1994). Both tasks consist of a list of 116 items, increasing in difficulty from one to four syllables. Children had to quickly and accurately read as many (non)words as possible in one (words) or two (nonwords) minutes, respectively. Raw scores (i.e., number correctly read) were used in the analyses. Internal consistency of both tasks is .90 and .92, respectively (Evers et al., 2009-2012).

READING COMPREHENSION AT T7. We used *Diatekst* of the *Diataal* test battery (Hacquebord et al., 2005) to assess reading comprehension in Grade 6. Children had to read five texts, each consisting of about 200 words, and answer 10 to 13 questions per text. Questions target reading comprehension at word and sentence level (micro), combining information from sentences and passages (meso), and distilling the main message of the text (macro). The outcome is a competence score, based on a weighing of the number of correct and incorrect responses, indicating the overall comprehension level. The reliability is .69.

PROCEDURE

Children were tested individually by trained and supervised (post)graduate students. Testing took place at home or at the university in a quiet room during one-to-two-hour sessions for each occasion. Ample breaks were provided during the assessments. Tasks were administered in a fixed order.

ANALYSES

The longitudinal data that were used to model literacy development were analyzed with structural equation modeling (SEM) using the R lavaan package (R Core Team, 2016) version 3.3.0. For single-indicator factors, the error variance of the indicator was specified based on the instrument's reliability. Subsequently, the model was built up hierarchically. First, the foundation of the model was formed by the second-order factor model previously reported on by van Bergen, de Jong, Maassen, Krikhaar et al. (2014), regressing the reasoning and performance nonverbal IQ factors and verbal IQ factor on a general IQ factor. Second, the adjacent occasion was added, for which the specified direct effects were theory driven. All correlations between latent variables within an age phase were included initially and removed one by one if statistically nonsignificant. Third, if necessary, additional across-time paths were added to the model based on the modification indices before the next measurement occasion was added. Factor loadings of identical constructs with identical indicators were fixed to be equal across time points, with residual correlations between the identical indicators across time points.

RESULTS

All children in the sample had complete parent data, necessary to establish FR status, and complete data from at least one of the seven measurement occasions. Missing data due to attrition and nonresponse were dealt with using full information maximum likelihood (FIML; see Little, 2013). Data points for univariate (0.4%, as based on 3 standard deviations above or below the mean) and multivariate (0.05%) outliers were also coded as missing. Distributions for all variables were approximately normal. However, since for some variables (e.g., word reading accuracy at T5) distributions differed somewhat between the control and FR group, robust maximum likelihood (MLR) estimation was used (Little, 2013).

Group means on all IQ, cognitive, and literacy measures are provided in Table 1. Although most group comparisons on the early (non)verbal IQ measures did not yield significant results (e.g., nonverbal IQ measures), children in the control group generally had higher scores than in the FR group based on the positive effect sizes. From kindergarten on the control group clearly outperformed the FR group on all cognitive (except for vocabulary at T7) and literacy measures, as indicated by the medium to large effect sizes. Appendix C shows the correlations between the IQ, cognitive, and literacy measures in the full sample. T3 preliteracy skills correlated weakly with T1 nonverbal abilities and moderately with T2 verbal abilities. Correlations between T3 preliteracy skills and later word reading variables were moderate to strong. T7 reading comprehension correlated moderately with word reading variables, whereas correlations with vocabulary increased in strength from weak to strong over time. FR is weakly correlated with T2 verbal abilities and later vocabulary skills, and moderately to strongly with later reading skills.

A full structural regression model was fitted to the whole sample, including FR as a separate predictor. The model showed a good fit to the data, $\chi^2(557) = 997.31$, $p < .001$, root mean square error of approximation (RMSEA) = .058, 90% confidence interval [.052 - .063], *p*close = .02, comparative fit index (CFI) = .93, standardized root mean square residual (SRMR) = .10. The standardized path weights are displayed in Figure 1.

Two pathways toward reading comprehension are present in the model. The first pathway, through preliteracy skills and word decoding ability, shows high stability from the moment word decoding was assessed (at T4). The standardized regression coefficients between consecutive time points were large for word reading accuracy (T4 through T6) as well as word reading fluency (T4 through T7). The correlation between both word reading components decreased from .80 at T4 to .30 at T6. The effect of T7 word reading fluency on reading comprehension just missed significance ($p = .053$)[3]. As a second pathway we found that early language abilities have an effect on T7 reading comprehension through vocabulary skills at T6 and T7.

Both pathways are influenced by early (non)verbal abilities. At T1 and T2, the second-order IQ model, placing the nonverbal and verbal IQ factors under a general IQ factor, was optimized by a residual correlation between the object assembly and categories indicators (see also van Bergen, de Jong, Maassen, Krikhaar et al., 2014). For the first pathway, this general IQ factor predicted three preliteracy factors (i.e., letter knowledge, PA, and RAN) at T3, which functioned as mediating variables for the development of word decoding ability. Letter knowledge and PA correlated strongly with each other and weakly with RAN. Subsequently, T3 letter knowledge and PA predicted T4 word reading accuracy, whereas T3 letter knowledge and RAN predicted T4 word reading fluency. Furthermore, there was a direct effect of verbal IQ on PA at T3, indicating that verbal abilities predict additional variance in PA beside general

[3] The effect of T7 word reading fluency on T7 reading comprehension is significant ($\beta = .29$, $p = .005$) when the FR predictor is not included in the model.

abilities represented by the IQ factor. RAN and PA at T4 were predicted by their respective counterparts at T3, but had no *additional* effect on literacy development from this point onward. The negative direct effect of verbal IQ on RAN at T4 was considered a suppression effect, which is thought to result from the high stability of RAN between T3 and T4. For the second pathway, verbal abilities at T2 predicted T6 vocabulary, which in turn predicted T7 vocabulary. In other words, early language had a direct influence on later language abilities, whereas their effect on word decoding ability would run via the general IQ factor and preliteracy skills, and T3 PA in particular.

Concerning the influence of FR, we found effects on several variables within the word decoding pathway, but not on the language pathway toward reading comprehension. Children at FR of dyslexia had lower general cognitive abilities (but not verbal abilities) at T1/T2, lower preliteracy skills at T3, and showed lower PA, word reading accuracy, and word reading fluency at T4. Nonetheless, statistical testing of indirect effects of FR on reading comprehension at T7, through T3 preliteracy skills (i.e., letter knowledge as well as RAN) and word reading fluency, showed no significant results. This is reasonable based on the nonsignificant relation between word reading fluency and reading comprehension at T7. However, FR turned out to have a direct effect on reading comprehension at T7, indicating that FR children have lower reading comprehension levels by the time they are in 6th grade.

Overall, the model accounted for 78% of the variance in word reading accuracy at T6, 95% of the variance in word reading fluency at T7, and 72% of the variance in reading comprehension at T7. This indicates that we could model a large amount of the variance in literacy development between the age of 4 and 12 years.

TABLE 1

Performance of Family Risk and Control Groups on IQ, Preliteracy, and Literacy Measures

Measure (max)	FR group			Control group				
	n	*M*	*SD*	*n*	*M*	*SD*	*p*	Cohen's *d* [95% CI]
T1 (start Kindergarten year 1)								
Patterns (19)	163	10.90	3.23	73	11.66	3.43	.11	0.23 [-0.05, 0.51]
Block design (19)	163	11.69	3.06	73	12.07	3.12	.38	0.12 [-0.15, 0.40]
Object assembly (19)	162	12.43	2.63	73	12.86	2.86	.26	0.16 [-0.12, 0.44]
Picture completion (19)	161	12.56	3.06	73	12.67	3.29	.80	0.04 [-0.24, 0.31]
Analogies (19)	163	12.16	2.79	73	12.56	3.01	.32	0.14 [-0.14, 0.42]
Categories (19)	162	11.83	3.17	73	12.52	3.10	.12	0.22 [-0.06, 0.50]
T2 (halfway Kindergarten year 1)								
Language comprehension (145)	161	108.03	11.89	72	112.49	11.38	.008	0.38 [0.10, 0.66]
Expressive syntax (145)	157	108.31	13.81	71	111.75	13.04	.08	0.25 [-0.03, 0.54]
Expressive vocabulary (145)	161	106.53	15.15	72	109.17	13.68	.21	0.18 [-0.10, 0.46]
Verbal STM (145)	161	103.81	14.25	71	110.59	12.83	.001	0.49 [0.21, 0.77]
T3 (end Kindergarten year 2)								
Active letter knowledge (34)	157	13.89	8.52	69	18.58	8.48	< .001	0.55 [0.26, 0.84]
Passive letter knowledge (32)	158	17.96	7.66	69	22.54	7.41	< .001	0.60 [0.32, 0.89]
Phoneme blending (20)	155	9.79	6.39	69	14.28	5.26	< .001	0.74 [0.45, 1.03]
Phoneme segmentation (20)	155	7.47	6.32	69	12.14	6.12	< .001	0.75 [0.45, 1.04]
RAN colors	152	39.53	10.51	66	45.69	10.99	< .001	0.58 [0.28, 0.87]
RAN objects	94	37.96	9.16	48	44.39	10.21	< .001	0.68 [0.32, 1.03]
T4 (start Grade 2)								
Phoneme deletion (18)	142	9.43	4.48	64	12.73	3.75	< .001	0.77 [0.47, 1.08]
RAN colors	142	51.74	10.91	64	55.86	12.39	.02	0.36 [0.07, 0.66]
RAN digits	141	78.09	17.87	62	89.06	16.24	< .001	1.15 [0.83, 1.47]
Word accuracy (40)	143	23.61	11.74	63	34.30	6.34	< .001	1.03 [0.72, 1.34]
Nonword accuracy (40)	143	11.61	9.14	62	19.35	9.14	< .001	0.85 [0.54, 1.16]
Word reading fluency 1 (150)	145	41.60	20.03	64	60.52	19.25	< .001	0.96 [0.65, 1.26]
Word reading fluency 2 (150)	145	29.63	20.33	63	50.46	21.89	< .001	1.00 [0.69, 1.31]
T5 (end Grade 2)								
Word accuracy (40)	140	31.16	8.71	65	37.95	2.46	< .001	0.93 [0.62, 1.23]
Nonword accuracy (40)	143	15.74	10.30	66	25.03	8.94	< .001	0.94 [0.64, 1.24]
Word reading fluency 1 (150)	144	56.92	22.26	66	75.39	15.17	< .001	0.91 [0.61, 1.21]
Word reading fluency 2 (150)	143	45.21	23.51	66	68.53	18.84	< .001	1.05 [0.74, 1.36]
T6 (end Grade 3)								
Nonword accuracy (40)	138	19.42	10.07	62	29.71	8.08	< .001	1.08 [0.77, 1.40]
Word reading fluency (116)	138	43.70	16.33	63	60.94	12.46	< .001	1.13 [0.81, 1.45]
Nonword reading fluency (116)	135	28.12	12.78	62	45.06	15.11	< .001	1.25 [0.93, 1.58]
Vocabulary (19)	142	11.56	2.67	64	12.45	2.52	.02	0.34 [0.04, 0.64]
T7 (Grade 6)								
Word reading fluency (116)	91	65.71	16.21	52	78.92	12.32	< .001	0.89 [0.53, 1.24]
Nonword reading fluency (116)	91	51.24	18.37	52	69.02	14.02	< .001	1.05 [0.69, 1.41]
Vocabulary (90)	84	64.24	9.09	42	67.29	6.23	.05	0.37 [-0.00, 0.74]
Reading comprehension (90)	85	64.73	10.88	39	71.77	3.66	< .001	0.76 [0.37, 1.15]

Note. Cohen's *d* is adjusted for unequal sample size (Hedge's *g*). FR = family risk; CI = confidence interval; T = time point; STM = short-term memory; RAN = rapid automatized naming.

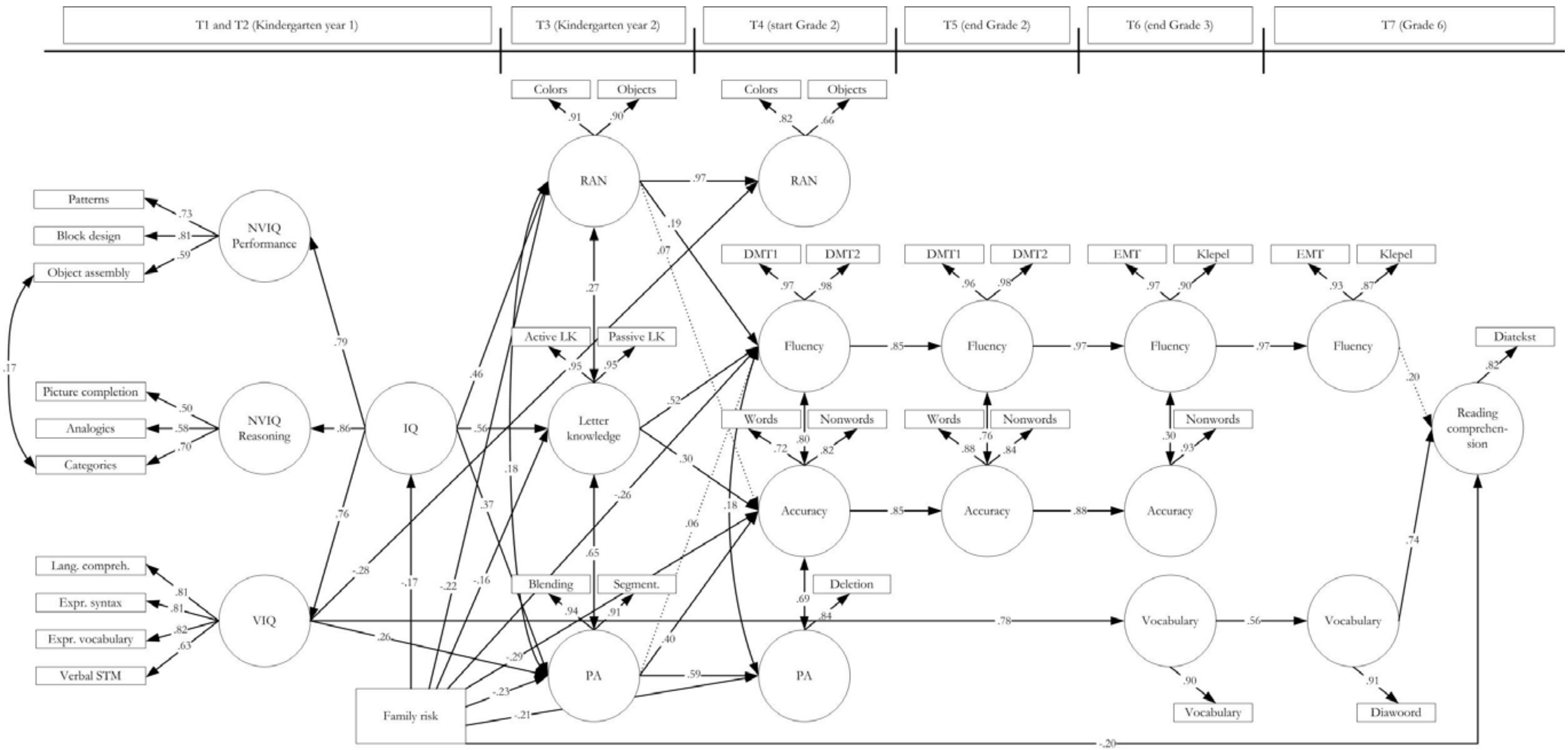

Figure 1. Structural equation model showing the longitudinal relations between T1 and T2 (non)verbal IQ measures, T3 preliteracy skills, T4 and T5 reading accuracy and fluency skills, T6 reading accuracy and fluency skills and vocabulary, and T7 reading fluency, vocabulary, and reading comprehension, including the effect of family risk status. *Note.* Rectangles indicate observed variables and ellipses represent latent variables. Values on single-headed arrows from latent to observed variables are standardized factor loadings. Values on single-headed arrows between latent variables are standardized regression coefficients. Double-headed arrows indicate correlations (standardized covariances). Solid lines indicate significant effects ($p < .05$). Error terms are not displayed to aid visibility. NVIQ = nonverbal IQ; VIQ = verbal IQ; RAN = rapid automatized naming; PA = phonological awareness.

DISCUSSION

The results of this study showed that both pathways toward reading comprehension, one through preliteracy skills and word decoding ability and the other through language abilities, build on early oral language skills. For the first pathway, early language (here verbal IQ) had a direct effect on PA. Importantly, this effect was independent of the effect of general IQ which affected all preliteracy skills (RAN, letter knowledge, and PA). The preliteracy skills subsequently predicted word decoding ability. In the second pathway, early language had a direct effect on later language abilities, which in turn predicted reading comprehension. These findings are in line with previous FR studies, showing that early oral language has an effect on word decoding as well as on reading comprehension at some point during development (Duff et al., 2015). More specifically, they confirm that the effect on word decoding is mediated by preliteracy skills (Torppa et al., 2010), and that an additional independent pathway from early language toward reading comprehension exists (Hulme et al., 2015), that may run via later language abilities.

The influence of word decoding ability on reading comprehension has been suggested to decrease over time, but not disappear altogether (Hoover & Gough, 1990). However, we did not find an effect of word reading fluency on reading comprehension. This could be due to inclusion of FR as a predictor in the model. Modeled effects are then estimated based on the variance within the FR group instead of the variance of the total sample. This leads to a restriction of range and thereby underestimation of the effect of word decoding ability on reading comprehension. Indeed, when FR was left out of the model, and all variance was taken into account, we did find an effect of word reading fluency on reading comprehension (β = .29, p = .005).

A specific feature of this study was that we modeled word reading accuracy *as well as* word reading fluency longitudinally. Both word reading abilities were predicted by different preliteracy skills. As in many studies, PA was more important for accuracy, while RAN was more important for fluency (Kirby, Desrochers, Roth, & Lai, 2008). We also found that both word reading accuracy and fluency showed high stability from the start of Grade 2 on. Interestingly, the effect of word reading accuracy on reading comprehension was fully accounted for by word reading fluency. This illustrates that fluency is more important in trajectories toward literacy in transparent orthographies, such as Dutch.

Concerning the role of FR, we found a small effect of FR on overall IQ, but not a specific effect on verbal abilities. Stronger effects of FR were found on the preliteracy skills, that is, from the onset of literacy acquisition. However, the effects of FR on preliteracy skills did not fully mediate the effect of FR on word reading accuracy and fluency. Thus after the start of literacy acquisition, additional effects of FR are found when word reading development sets off. This finding sets an important upper limit for the prediction of word decoding from preliteracy skills, as there are effects of parental reading abilities on children's word reading skills that are apparently not yet present in children's preliteracy skills (van Bergen, de Jong, Maassen, & van der Leij, 2014).

FR did not have an effect on reading comprehension through the language pathway. However, unexpectedly, we did find a direct effect of FR on reading comprehension. Accordingly, children at FR of dyslexia not only have lower word decoding levels throughout primary education but also end up with lower reading comprehension levels in Grade 6. This finding is in line with Duff et al. (2015), who found lower reading comprehension levels in FR children between the age of 5 and 9 years. A possible explanation could be that our measure, i.e., reading lists of words, does not cover all aspects of word reading fluency. For example, oral reading speed of texts, on which FR children might also show impairments, could be more important for reading comprehension. A second explanation, not necessarily incompatible with the first, could be that children who show somewhat lower word decoding ability read less and have therefore less exposure to texts (Mol & Bus, 2011), leading to lower levels of reading comprehension.

Overall, the present study clearly shows that there are two pathways toward reading comprehension. Early oral language ability is foundational for both pathways. Family risk for dyslexia did not influence the language pathway, but affected literacy development through its subsequent effects on preliteracy skills, word decoding, and reading comprehension.

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CHAPTER 4

HIGH READING SKILLS MASK DYSLEXIA IN GIFTED CHILDREN

This study investigated how gifted children with dyslexia might be able to mask severe literacy problems and the role of possible compensation. The sample consisted of 121 Dutch primary school children that were divided over four groups; averagely intelligent children with dyslexia, gifted children with dyslexia, typically developing (TD) children, and gifted children. The test battery included measures of literacy (reading/spelling) and cognitive abilities related to literacy and language (phonological awareness [PA], rapid automatized naming [RAN], verbal short-term memory [VSTM], working memory [WM], grammar, and vocabulary). It was hypothesized that gifted children with dyslexia would outperform children with dyslexia on literacy tests. Additionally, a core-deficit model including dyslexia-related weaknesses and a compensational model involving giftedness-related strengths were tested using Bayesian statistics to explain their reading/spelling performance. Gifted children with dyslexia performed on all literacy tests in between children with dyslexia and TD children. Their cognitive profile showed signs of weaknesses in PA and RAN and strengths in VSTM, WM, and language skills. Findings indicate that phonology is a risk factor for gifted children with dyslexia, but this could be moderated by other skills such as WM, grammar, and vocabulary, providing opportunities for compensation of a cognitive deficit and masking of literacy difficulties.

Keywords: giftedness, dyslexia, twice-exceptionality, literacy, language, working memory, Bayes.

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INTRODUCTION

Within the field of giftedness, there is increasing interest in understanding twice-exceptionality, the concomitant occurrence of giftedness and a learning disability (LD) within an individual (Brody & Mills, 1997). As the field of twice-exceptionality still heavily relies on anecdotal information, it is in need of empirical research to support evidence-based practices regarding the identification and education of gifted students with LD (Lovett & Lewandowski, 2006; Nielsen, 2002), and more information on individual exceptionalities is required (Foley Nicpon, Allmon, Sieck, & Stinson, 2011). The present study aims to provide empirical data on the achievement and cognitive characteristics of gifted children with dyslexia, a learning disability characterized by severe reading and/or spelling difficulties (Vellutino, Fletcher, Snowling, & Scanlon, 2004). This empirical data will increase our understanding of both giftedness and dyslexia, as well as shed more light on the possibilities to mask underachievement and to compensate a cognitive deficit. These insights may provide a new step toward better identification and service of twice-exceptional children.

Although empirical data are not available, the estimates for the prevalence of giftedness and LD range from 1% to 5% of the total population of children with learning disabilities (McCoach, Kehle, Bray, & Siegle, 2004; Nielsen, 2002; Silverman, 1989), which is comparable to the prevalence in the general population. However, these estimates are based on a wide variety of definitions of giftedness and LD as well as twice-exceptionality and can be considered rather conservative (Nielsen, 2002). Moreover, many twice-exceptional children might remain undetected as they may not positively or negatively stand out compared to the general population (Nielsen, 2002). A lack of a concrete definition of twice-exceptionality, underachievement, underestimation of intellectual abilities, and masking effects are considered the main reasons for the problems with adequate identification and early intervention of twice-exceptionality (Brody & Mills, 1997; Foley Nicpon et al., 2011; McCoach, Kehle, Bray, & Siegle, 2001).

In the present study, the definition of McCoach et al. (2001) was used for twice-exceptionality, stating that “gifted/learning disabled students are students of superior intellectual ability who exhibit a significant discrepancy in their level of performance in a particular academic area” (p. 405). In addition, giftedness was defined as ‘high intelligence’ or academic giftedness, which is typically classified with an IQ score above 130 (Winner, 1997). However, since the achievement-ability discrepancy (i.e., a significant difference between general learning ability and outcomes in a specific academic ability, such as reading) has been heavily debated (e.g., Lovett & Lewandowski, 2006), it was only used to nominate children for participation in the study, as it is the only definition that leaves room for the possibility of masking or compensation of LD in a high IQ population (see Assouline, Foley Nicpon, & Whiteman, 2010, for an elaborate discussion). Further inclusion was based on a comprehensive evaluation of a child's academic *and* cognitive strengths and weaknesses, integrating multiple sources of information, which may unveil specific underlying deficits needed for correct identification of LD, here dyslexia, in gifted children (Assouline et al., 2010; Brody & Mills, 1997; Lovett & Lewandowski, 2006; McCoach et al., 2001; Nielsen, 2002). Prior to focusing on the combination of giftedness and dyslexia, both elements will be introduced in more detail.

Dyslexia is defined as a learning disability characterized by severe reading and/or spelling difficulties at word level (Snowling, 2000). Depending on the transparency of a language's orthography, reading difficulties are either marked by poor accuracy or slowness in naming of words (Frith, Wimmer, & Landerl, 1998). Nonword reading is particularly impaired and often seen as the first indicator of broader underlying decoding problems (van den Bos & Scheepstra, 1993; Griffiths & Snowling, 2002). One of the main cognitive deficits proposed to underlie dyslexia is a phonological deficit leading to difficulties in connecting sounds to letters (Vellutino et al., 2004), but the breadth/range of phonological areas involved is still under investigation. According to Snowling (2000), phonological awareness (PA), verbal short-term memory (VSTM), and rapid automatized naming (RAN) tasks are a reflection of the phonological skills demanded for successful literacy acquisition. Although it is still a matter of debate whether RAN should be viewed as part of the phonological deficit, there is convincing evidence that RAN is related to reading skills (e.g., Vaessen, Gerretsen, & Blomert, 2009; Warmington & Hulme, 2012).

Gifted children often show specific academic and cognitive strengths that are relevant in relation to the weaknesses associated with dyslexia described above. Even though the relation between IQ and reading achievement is not perfect (Naglieri, 2001; Vellutino, Scanlon, & Lyon, 2000), research on the literacy skills of gifted children has shown that most of them learn to read earlier than their peers do and they have been reported to read at least two grade levels above their chronological grade (Kaplan, 1999). In addition, gifted children have been described as having textual information understanding capacities that are well above the level of their age-matched peers (Reis et al., 2004), as they use both their advanced vocabulary and grammar to enhance understanding and accelerate their literacy development (Hoh, 2005). Gifted children rely on metacognitive skills, such as analysis, synthesis, and evaluation while reading and they automatically integrate prior knowledge and experience into their reading, allowing an intuitive development of reading skills (Catron & Wingenbach, 1986). Furthermore, gifted children display more efficient working memory (WM) and higher speed of processing than typically developing (TD) children (Alloway & Elsworth, 2012; Conway, Cowan, Bunting, Therriault, & Minkoff, 2002; Dark & Benbow, 1991; Johnson, Im-Bolter, & Pascual-Leone, 2003). Several studies have shown that intelligence and working memory are (highly) correlated, but they also form separate cognitive skills with unique contributions to learning outcomes (Ackerman, Beier, & Boyle, 2005; Alloway & Alloway, 2010; Conway, Kane, & Engle, 2003). Working memory has been found to be the stronger predictor for academic progress in literacy (Alloway, 2009) and can therefore, besides general language abilities, be an important compensatory factor in the literacy development of gifted children with dyslexia. For gifted children, these precocious abilities allow them to make associations between words faster and more frequently and recall language facts from their memory faster, resulting in better performance on speeded tasks, such as timed word reading tests as well as RAN tasks (Dark & Benbow, 1991; Johnson et al., 2003).

It is as yet unknown how the combination of giftedness and dyslexia manifests itself within one child. However, we do know that dyslexia is a learning disability that can be found across intelligence levels, including high IQ children (Snowling & Hulme, 2012; see e.g., Hatcher & Hulme, 1999; Vellutino et al., 2004, on the role of IQ in dyslexia). Moreover, previous research has shown that children displaying positive indicators for dyslexia such as academic underachievement, slow literacy development, and poor phonological skills need not necessarily have literacy problems that are severe enough to reach the diagnostic threshold (Miles, Wheeler, & Haslum, 2003; Snowling, 2008). This identification problem specifically arises with gifted or high-functioning children, whose reading deficits are proposed to be mild or compensated (Snowling & Hulme, 2012). Research on gifted children with LD in general has shown that the academic achievement of these students might not be as low as that of other students with LD; *average* achievement is sometimes considered sufficient to suspect an underlying deficit (Assouline et al., 2010; Bireley, Languis, & Williamson, 1992; Brody & Mills, 1997). In addition, a student's giftedness and learning disabilities can both lie in related academic areas. For example, a student can show reading levels well above grade level but experience great difficulty with spelling and writing (Bireley et al., 1992; Brody & Mills, 1997).

At the cognitive level, it can be expected that both the underlying deficit(s) associated with dyslexia and the precocious abilities of gifted children that were described above, also occur in the cognitive profile of gifted children with dyslexia. Yet, it is unknown how these specific strengths and weaknesses affect each other. Empirical research on cognitive characteristics of gifted children with LD in general has indicated that they show underlying perceptual and memory deficits in visual and auditory discrimination, sequencing, decoding skills, short-term auditory memory, and spatial abilities (Waldron & Saphire, 1990, 1992). In contrast, they show more advanced verbal abilities than nonverbal abilities, high metacognitive skills, and rely more on verbal comprehension, reasoning, and abstract thinking (Assouline et al., 2010; Hannah & Shore, 2008; Waldron & Saphire, 1990). Generally, their WM and processing speed abilities are similar to those of their age mates (Assouline et al., 2010). However, these studies do not provide any information about the implications of certain patterns of cognitive strengths and weaknesses for the behavioral expression of a specific LD or possible compensation.

The present study tested the performance differences between groups to answer the question what the achievement and cognitive characteristics of gifted children with dyslexia are compared to averagely intelligent children with dyslexia, TD children, and gifted children. The assessment battery used to measure performance covered five domains, i.e., literacy, phonology, verbal and visuospatial WM, and language skills. Based on previous research, it was hypothesized that gifted children with dyslexia would show higher reading and spelling performance overall than the averagely intelligent children with dyslexia, but lower performance than TD children and considerably lower than gifted children. Additionally, a 'core-deficit' model of dyslexia and a 'compensational' model of giftedness were tested to explain group differences in reading and spelling performance. These models serve to illustrate that gifted children with dyslexia may have specific cognitive weaknesses that are related to their dyslexia, but also possess strongly developed skills that are related to their giftedness, which might form protective factors that could be involved in compensation of a cognitive deficit. Therefore, mapping the behavioral and cognitive profile of gifted children with dyslexia will shed more light on the possibility to compensate a phonological deficit and mask literacy difficulties.

METHOD

PARTICIPANTS

The sample consisted of 121 Dutch primary school children from Grades 2 to 4. Informed consent was obtained from all participants and their parents. The study consisted of four groups; (a) averagely intelligent children with dyslexia (D; n = 33), (b) gifted children with dyslexia (GD; n = 26), (c) typically developing children (TD; n = 31), and (d) gifted children (G; n = 31). Children were first nominated for the gifted and dyslexia group based on a significant discrepancy between their IQ and reading and/or spelling ability of at least two standard deviations (Snowling, 1998). Subsequently, the three inclusion criteria for *both* dyslexia groups were in line with the criteria for an official diagnosis in The Netherlands (Kleijnen et al., 2008). Children had to show (a) at most average scores on both reading and spelling (standard score ≤ 12), (b) below average scores on reading or spelling (lowest 10-15% or a standard score ≤ 6), and (c) below average performance on at least one of the three cognitive factors that have been proposed to underlie dyslexia; PA, RAN, and VSTM (standard score ≤ 7; Snowling, 2000). Of the 43 twice-exceptional children that were nominated based on a significant discrepancy, 26 children met the inclusion criteria for the GD group and 17 children turned out to be borderline-dyslexic cases (i.e., criteria a and c were met, but they failed to meet criterion b). The borderline cases were excluded from further analyses. Giftedness was defined as a high IQ score on a validated intelligence test (see Lovett & Lewandowski, 2006). The cut-off value was set at a full IQ score > 125 or a 95% reliability interval tapping at least 130 in case of a short form. Table 1 shows the division of age, intelligence, and sex in the four groups.

TABLE 1

Sample Size, Percentage of Boys, and Means and Standard Deviations for Age and IQ score per Group

	D	GD	TD	G
Variable	(n = 33)	(n = 26)	(n = 31)	(n = 31)
% Boys	48.5	65.4	29.0	41.9
Age (months)	113.85_{a} (12.39)	108.77_{ab} (8.14)	103.45_{bc} (8.92)	100.58_{c} (10.43)
IQ (total)	98.70_{a} (11.41)	132.50_{b} (8.05)	108.23_{c} (9.41)	134.10_{b} (8.95)

Note. The group means are given with the standard deviations in parentheses. Means in the same row that do not share subscripts differ at $p < .05$. D = dyslexia; GD = gifted + dyslexia; TD = typically developing; G = gifted.

Out of the 33 children in the dyslexia group, 20 had an official diagnosis of dyslexia (60.6%); for the gifted and dyslexia group this was 13 of the 26 children (50.0%). This relatively low percentage is due to the fact that Dutch children in primary education are only eligible for reimbursed treatment of dyslexia if they meet strict criteria, including poor performance on nonword reading as well as an extensive period of remedial instruction prior to a possible diagnosis (Blomert, 2006). The latter criterion is most influential on the number of diagnoses in the current study because of the young age of the children in the sample. To address this issue, the children without a diagnosis all had to be referred by their teachers based on relative academic underachievement and persistent and continuous problems with reading and/or spelling, and children should have been *at least* in the process of being tested for dyslexia.

INSTRUMENTS

INTELLIGENCE. To estimate the general cognitive abilities of the participants, a short form of the Dutch version (Kort et al., 2005) of the *Wechsler Intelligence Scale for Children – Third Edition* (WISC-III; Wechsler, 1991) was used, consisting of the verbal subtests similarities and vocabulary and the performance subtests picture completion and block design. The reliability and validity quotients are all reported to be above .83 (Kaufman, Kaufman, Balgopal, & McLean, 1996).

LITERACY. Timed word reading was measured using *Eén Minuut Test* (EMT; Brus & Voeten, 1999) and decoding speed of nonwords was measured using *Klepel* (van den Bos, lutje Spelberg, Scheepstra, & de Vries, 1994). In these tests, the child has one and two minutes, respectively, to accurately read as many (non)words as possible. Word length increases from one to four syllables. Raw scores are the number of correctly read words or nonwords, with a maximum of 116 words. Age-referenced norm scores (M = 10, SD = 3) were used for the inclusion criteria of dyslexia. Internal consistency is .90 for EMT and .92 for Klepel (Evers et al., 2009-2012).

Timed text reading was measured using *AVI* (Visser, van Laarhoven, & ter Beek, 1996). In this test, the child had to read several texts that correspond in difficulty to grade levels (i.e., mid Grade 1 to end Grade 6). Both the reading time (seconds) and the number of errors were recorded. Scores on the highest 'mastery' text were transformed into a number of words correct per minute ratio for the analyses. Reliability is evaluated as 'acceptable' (Evers et al., 2009-2012).

Spelling at word level was measured using a short form of the *PI-dictee* (Geelhoed & Reitsma, 2000). The short form contains eight blocks of seven words, with each block representing specific spelling categories (P. F. de Jong, personal communication, September 2012). The test continued until the child made six or more errors in one block. The raw score is the total number of correctly written words. After computation of the approximate total score on the full version (i.e., 9 blocks of 15 words), using the formula raw score + 7 * (15/7), age-referenced norm categories (A - E) were used for the inclusion criteria of dyslexia. Internal consistency of the full version varies between .90 and .93 (Evers et al., 2009-2012).

PHONOLOGY. PA was assessed using the *Fonemische Analyse Test* (FAT; van den Bos, lutje Spelberg, & de Groot, 2011). This is a computerized test measuring the ability to analyze and manipulate phonemes. The first subtest targets phoneme deletion (e.g., *kraal* 'bead' without /*k*/ is *raal*), and the second subtest targets phoneme transposition (e.g., transposing onset phonemes of *Kees Bos* to *Bees Kos*). Raw response time and accuracy scores were transformed into a number correct per minute ratio score for the analyses. Internal consistency of the test is .93 (Evers et al., 2009-2012).

RAN was measured using the *Continu Benoemen & Woorden Lezen* (CB&WL; van den Bos & lutje Spelberg, 2007). This test includes four subtests (colors, digits, pictures, and letters) assessing the child's naming speed. Average raw scores for the colors and pictures subtests and the digits and letters subtests were computed for the analyses, resulting in a 'non-alphanumeric' score and an 'alphanumeric' score. Internal consistency of the test varies between .79 and .87 (Evers et al., 2009-2012).

VSTM was measured by the subtest digit recall of the *Automated Working Memory Assessment* battery (AWMA; Alloway, 2007). The subtest consists of several series of digits of increasing length that were presented through the computer and recalled by the child. Raw scores were used for the analyses. Test-retest reliability is .89 (Alloway, Gathercole, Kirkwood, & Elliot, 2009).

WORKING MEMORY. WM was also measured using subtests of the AWMA (Alloway, 2007). All WM subtests were discontinued after three incorrect answers. Verbal WM was measured using backward digit recall, in which the child recalled increasing series of digits backwards. Raw scores were used in the analyses. Test-retest reliability of this subtest is .86 (Alloway et al., 2009).

Visuospatial WM was measured by two subtests. Spatial span demands the child to evaluate figures by mental rotation, classify them as 'the same' or 'opposite', and recall the place of a red dot in an empty figure. Odd-one-out requires the child to indicate in increasingly complex sequences which figure out of three is odd, and recall the odd figures in a matrix. The raw scores of the visuospatial subtests were combined into a composite score for the analyses. Test-retest reliabilities are .79 and .88, respectively (Alloway et al., 2009).

LANGUAGE. Grammar and vocabulary were measured using the *Clinical Evaluation of Language Fundamentals-4 NL* (CELF; Kort, Schittekatte, & Compaan, 2010). The child's grammar skills were assessed by the subtest formulated sentences of the language structure index, in which the child formulates a sentence about visual stimuli using a targeted word or phrase. The subtest word classes 2 of the language content index, in which the child chooses two related words and describes their relationship, measured vocabulary. Raw scores were used for the analyses. Internal consistency of the subtests is .78 and .87, respectively (Evers et al., 2009-2012).

PROCEDURE

Participants were recruited through advertisements on the websites of educational magazines and clinical institutions and through contacts with school psychologists. Trained and supervised graduate students performed the assessments using the test battery described above. All children were tested either in a clinic, at school, or in their homes within one session that lasted for two to three hours. After the assessment, the test results were summarized in a short report and evaluated by a licensed school psychologist. Any diagnostic uncertainties were resolved during joint evaluation meetings.

ANALYSES

DATA SCREENING

Missing data analyses showed several missing data points (i.e., AVI [4], digit recall [1], backward digit recall [1], visuospatial WM [1], grammar [3], and vocabulary [3]). Since the software for the analyses does not allow missing data and only 0.8% of the total number of data points in the analyses were missing (equally distributed across groups), single imputation based on the series mean was applied. The data contained no univariate or multivariate outliers. Further data screening showed no violations of assumptions for multivariate analysis of variance. Finally, since the age of the children was not equally distributed across groups (see Table 1), this variable was centered and added to the analyses as a covariate.

MAIN ANALYSES

Instead of using traditional frequentist analyses, Bayesian statistics were used to compare all four groups on literacy skills and cognitive components. Bayesian model selection offers the possibility to use prior knowledge to formulate and evaluate informative hypotheses using equality and inequality constraints between groups and compare competing hypotheses that are based on specific expectations (Klugkist, Laudy, & Hoijtink, 2005). The outcome of the analysis is a Bayes factor (BF), representing the amount of evidence in favor of one hypothesis compared to another (Kass & Raftery, 1995). The Bayesian framework is an effective alternative for standard frequentist analyses. It offers solutions to important analytical problems concerning multiple testing, such as alpha inflation and loss of power after correcting the alpha level (Klugkist, van Wesel, & Bullens, 2011). In addition, Bayesian analyses are not based on normality or asymptotic assumptions, making it a suitable approach for relatively small sample sizes (Gill, 2008). As such, Bayesian analyses were used to test informative hypotheses about the literacy skills and cognitive components and obtain parameter estimates to make further inferences. The analyses were performed using the BIEMS software package (Mulder, Hoijtink, & Klugkist, 2010; Mulder, Hoijtink, & de Leeuw, 2012; Mulder et al., 2009).

For *literacy*, we tested two competing hypotheses about the literacy skills of gifted children with dyslexia compared to averagely intelligent children with dyslexia, TD children, and gifted children. Each hypothesis was translated into a statistical hypothesis with (in)equality constrained parameters (Klugkist et al., 2005). Here, the parameters were the group means on the word reading, nonword reading, text reading, and spelling tasks. Based on the inclusion criteria for dyslexia, the first hypothesis stated that gifted children with dyslexia would score about equally low on literacy skills as averagely intelligent children with dyslexia, lower than TD children, and gifted children would outperform all groups, that is, $\mu_{D} = \mu_{GD} < \mu_{TD} < \mu_{G}$ (Model 1). The second hypothesis stated that gifted children with dyslexia would score *higher* on literacy skills than children with dyslexia, but lower than TD children, and gifted children would outperform all groups, that is, $\mu_{D} < \mu_{GD} < \mu_{TD} < \mu_{G}$ (Model 2). These informative hypotheses were compared to the alternative hypothesis, or unconstrained model, that is, μ_{D}, μ_{GD}, μ_{TD}, μ_{G} (Model 0), to protect against incorrectly choosing a wrong or poorly formulated hypothesis (van de Schoot et al., 2011).

For the underlying *cognitive components* (i.e., PA, RAN, VSTM, verbal and visuospatial WM, grammar, and vocabulary), two different sets of informative hypotheses were tested. First, we formulated two informative hypotheses about the phonology measures that would fit a core-deficit model of dyslexia, as proposed by Snowling (2000; see also Stanovich, 1988; Stanovich & Siegel, 1994). The first hypothesis stated that gifted children with dyslexia would score about equally low on all phonology measures as the averagely intelligent children with dyslexia, lower than the TD children, and gifted children would outperform all groups, that is, $\mu_{D} = \mu_{GD} < \mu_{TD} < \mu_{G}$ (Model 1). The second hypothesis stated that gifted children with dyslexia would score higher on all phonology measures than children with dyslexia, but lower than TD children, and gifted children would outperform all groups, that is, $\mu_D < \mu_{GD} < \mu_{TD} < \mu_G$ (Model 2). For RAN, the informative hypotheses had to be formulated in the opposite direction, since low scores indicated high performance on these tasks. Second, a compensational model was used to formulate informative hypotheses about the cognitive components that were expected to be giftedness-related strengths. However, since previous research on these components in twice-exceptional children in general showed highly mixed results, not all group differences could be specified in the informative hypotheses. Consequently, the first hypothesis stated solely that gifted children would outperform all groups on the WM and language measures, that is, $\mu_G > \mu_{TD,}\ \mu_{GD,}\ \mu_D$ (Model 1). The second hypothesis stated that both gifted children and gifted children with dyslexia would outperform the TD children as well as the averagely intelligent children with dyslexia on the WM and language measures, that is, $\mu_{G,}\ \mu_{GD} > \mu_{TD}, \mu_D$ (Model 2).

Generally, the first step of the analysis involves calculating the $BF_{i,u}$ for the informative hypothesis (H_i) versus the unconstrained alternative (H_u). When one of the informative hypotheses receives more support from the data than the unconstrained model ($BF_{i,u} > 1$), a second step could be to compare several models by dividing the $BF_{i,u}$ of each model by the sum of BFs of the other models of interest. Assuming prior probabilities to be equal for all models, this results in a posterior model probability (PMP), representing the relative support for a specific hypothesis within a set of hypotheses (Klugkist et al., 2011). It should be borne in mind that, in a Bayesian framework, the definition of 'probability' is defined as a degree of belief. In the case of a PMP, 'probability' relates to the probability that a hypothesis is true (Klugkist et al., 2011). In addition to the BFs and PMPs, the obtained parameter estimates were used to make more detailed inferences about the group differences.

RESULTS

LITERACY

Table 2 shows the BFs and PMPs for all three models in the analysis, presenting the results for each literacy skill separately. Recall that in this approach Model 0 was the alternative hypothesis ($\mu_D, \mu_{GD}, \mu_{TD}, \mu_G$), Model 1 stated that gifted children with dyslexia would show literacy skills comparable to averagely intelligent children with dyslexia ($\mu_D = \mu_{GD} < \mu_{TD} < \mu_G$), and Model 2 stated that gifted children with dyslexia would perform better than averagely intelligent children with dyslexia but worse than TD children ($\mu_D < \mu_{GD} < \mu_{TD} < \mu_G$). Model 2 received most support from the data for all literacy skills, on average about 23 times more than the alternative hypothesis. The probabilities that the hypothesis under Model 2 is true vary between .87 and .96. As displayed in Table 3, the posterior means of the unconstrained model indeed indicate that gifted children with dyslexia scored higher than averagely intelligent children with dyslexia on every aspect of literacy (reading and spelling), but lower than TD children, and gifted children outperformed all groups.

TABLE 2

Bayes Factors (BF) and Posterior Model Probabilities (PMP) of the Three Models for the Literacy Skills and Cognitive Components

	Model 0		Model 1		Model 2	
Skill/component	BF	PMP	BF	PMP	BF	PMP
Literacy						
Word reading	1.00	.04	0.19	.01	**23.94**	.95
Nonword reading	1.00	.04	2.50	.09	**23.15**	.87
Text reading	1.00	.04	0.05	.00	**24.19**	.96
Spelling	1.00	.04	0.27	.01	**23.87**	.95
Core-deficit model						
Phoneme deletion	1.00	.04	0.02	.00	**21.31**	.95
Phoneme transposition	1.00	.04	1.01	.04	**23.36**	.92
RAN alphanumeric[a]	1.00	.03	7.62	.25	**22.01**	.72
RAN non-alphanumeric[a]	1.00	.04	4.81	.18	**20.90**	.78
VSTM	1.00	.26	0.22	.06	**2.59**	.68
Compensational model						
Verbal WM	1.00	.11	**3.93**	.44	**4.05**	.45
Visuospatial WM	1.00	.10	3.10	.31	**5.90**	.59
Grammar	1.00	.10	3.97	.38	**5.43**	.52
Vocabulary	1.00	.11	2.57	.29	**5.26**	.60

Note. Bold face indicates BFs of models that received most support from the data. RAN = rapid automatized naming; VSTM = verbal short-term memory; WM = working memory.
[a] Hypotheses were formulated in the opposite direction for both RAN components, as higher numbers indicate worse performance for these measures.

COGNITIVE COMPONENTS

Table 2 also shows the BFs and PMPs for the core-deficit model of dyslexia and the compensational model of giftedness-related strengths, presenting the results for each cognitive component separately. Recall that in the core-deficit approach, Model 1 stated that gifted children with dyslexia would show phonology levels comparable to averagely intelligent children with dyslexia ($\mu_D = \mu_{GD} < \mu_{TD} < \mu_G$), and Model 2 stated that gifted children with dyslexia would show higher phonology levels than children with dyslexia but lower than TD children ($\mu_D < \mu_{GD} < \mu_{TD} < \mu_G$). For all phonology measures, Model 2 received most support from the data, about 22 times more than the alternative hypothesis. For the PA and RAN measures, the results are confirmed by the posterior means under the unconstrained model that are displayed in Table 3. Findings show that the gifted children with dyslexia scored higher on the phonology measures than the averagely intelligent children with dyslexia, but lower than the TD children, and the gifted children showed the highest scores (PMPs .72-.95). However, the BF and PMP of the second model are considerably lower for VSTM than for the PA and RAN measures. The posterior group means of VSTM indicate that one of the constraints in the second model was imposed incorrectly, explaining the lower BF. In contrast to PA and RAN, the gifted children with dyslexia outperformed not only the children with dyslexia, but also the TD children on the VSTM component. The gifted children outperformed all groups.

For the compensational model, recall that Model 1 stated that the gifted children would outperform all groups on WM and language skills ($\mu_G > \mu_{TD,}\ \mu_{GD,}\ \mu_D$) and that Model 2 stated that both the gifted children and the gifted children with dyslexia would outperform the TD children as well as the averagely intelligent children with dyslexia on WM and language skills ($\mu_{G,}\ \mu_{GD} > \mu_{TD}, \mu_D$). Although for some measures the differences in BFs between Model 1 and Model 2 were relatively small, Model 2 received about five times more support from the data for all components than the alternative hypothesis (Table 2; PMPs .45-.60). The posterior means in Table 3 show that gifted children with dyslexia outperformed averagely intelligent children with dyslexia as well as TD children on both WM components, although there seems to be no difference between the gifted children with dyslexia and the TD children on verbal WM. The gifted children still outperformed all other groups. Similarly, gifted children with dyslexia outperformed both children with dyslexia and TD children on the language component grammar, but the gifted children showed even higher scores. For vocabulary, however, the posterior means show that the gifted children with dyslexia not only outperformed the averagely intelligent children with dyslexia and the TD children, but also scored about equal to the gifted children.

Table 3

Posterior Means (PM) and Standard Deviations (PSD) of the Literacy Skills and Cognitive Components Adjusted for Age

	Dyslexia		Gifted + Dyslexia		Typically Developing		Gifted	
Skill/component	PM	PSD	PM	PSD	PM	PSD	PM	PSD
Literacy								
Word reading	30.14	4.11	39.57	4.53	56.59	3.95	70.55	4.17
Nonword reading	19.16	4.98	26.31	5.62	47.74	4.75	58.85	5.12
Text reading	76.48	8.93	91.80	10.07	103.03	8.74	113.28	9.23
Spelling	16.76	1.90	22.80	2.10	30.67	1.82	36.42	1.93
Core-deficit model								
Phoneme deletion	9.67	1.92	18.18	2.15	20.68	1.84	27.68	1.97
Phoneme transposition	1.21	0.31	3.15	0.34	5.03	0.30	8.54	0.31
RAN alphanumeric[a]	35.71	1.05	33.57	1.18	29.40	1.02	25.30	1.07
RAN non-alphanumeric[a]	58.57	2.74	54.60	3.03	49.07	2.65	46.28	2.81
VSTM	22.57	0.68	25.89	0.77	24.56	0.66	26.54	0.70
Compensational model								
Verbal WM	9.79	0.42	12.54	0.48	12.09	0.42	14.68	0.44
Visuospatial WM	14.08	0.52	19.86	0.57	17.50	0.51	20.67	0.53
Grammar	24.68	0.83	26.87	0.93	25.00	0.80	30.76	0.86
Vocabulary	12.56	0.92	18.20	1.04	16.41	0.88	18.73	0.94

Note. RAN = rapid automatized naming; VSTM = verbal short-term memory; WM = working memory.
[a] Higher numbers indicate worse performance on the RAN alphanumeric and non-alphanumeric cognitive components.

DISCUSSION

This study compared literacy skills and the cognitive profiles between gifted children with dyslexia, averagely intelligent children with dyslexia, typically developing (TD) children, and gifted children. The hypothesis that gifted children with dyslexia would show higher reading and spelling performance overall than the averagely intelligent children with dyslexia, but lower performance than TD children and considerably lower than gifted children, was accepted. Furthermore, the hypotheses that gifted children with dyslexia have a specific cognitive profile of dyslexia-related weaknesses (core-deficit model) and giftedness-related strengths (compensational model) that may provide possibilities for compensation of underlying deficits, were largely confirmed.

Assumptions based on anecdotal information and previous research on twice-exceptionality were confirmed; the performance of gifted children with dyslexia on reading and spelling tests was in between that of averagely intelligent children with dyslexia and TD children. Gifted children with dyslexia, classified based on the same low achievement criteria for dyslexia as their nongifted counterparts, were not found to display literacy performance as poor as averagely intelligent children with dyslexia. This illustrates the difficulty of recognizing literacy difficulties in these children in class; based on their achievement they might not appear to fulfil criteria for dyslexia at first sight, because their averagely intelligent peers with dyslexia may show even lower achievement. Consequently, they are less likely to be referred for diagnostic assessment. These results are also important for the borderline cases that were excluded from the analyses because they did not meet the diagnostic criteria for dyslexia. From the twice-exceptionality framework, it is often assumed that this group compensates most and that these children might have reached the diagnostic threshold if they had been averagely intelligent. We contend that more research into this borderline-dyslexic group is warranted. Also, it should be considered to provide these children with an *appropriate* intervention without a diagnosis of dyslexia. Although not all children in both dyslexia groups had received a diagnosis of dyslexia prior to the study, these children were all referred by their teachers because of serious concerns about their literacy development and were in process of being tested. Limiting inclusion to children with a (double) diagnosis would have made it virtually impossible to conduct this study, especially because of the aforementioned difficulty of identification of gifted children with LD and their relatively young age.

Furthermore, since nonword reading plays such an essential role in diagnosing dyslexia in The Netherlands, it is important to highlight that nonword reading performance of the gifted children with dyslexia was also better overall than of the averagely intelligent children with dyslexia. Hence, nonword reading does not sufficiently differentiate children with dyslexia from typical readers in a gifted/high IQ population. This might be due to compensation that gifted children with dyslexia possess in skills related to nonword reading; as they often possess increased inhibition (Johnson et al., 2003), for instance, they might be able to ignore the associations with real words better during nonword reading. Likewise, gifted children with dyslexia may show relatively better performance on the phonological awareness (PA) and rapid automatized naming (RAN) tasks because they perform better on speeded tasks (Catts, Gillispie, Leonard, Kail, & Miller, 2002; Johnson et al., 2003). In addition, recent research on the role of visual attention span in reading development and dyslexia might shed more light on possibilities for compensation of a (non)word reading deficit (e.g., van den Boer, de Jong, & Haentjens-van Meeteren, 2012; Valdois, Lassus-Sangosse, & Lobier, 2012). Consequently, the finding that gifted children with dyslexia generally outperform children with dyslexia on a nonword reading test implies that nonword reading *alone* is not a suitable screening method for dyslexia in gifted children.

In line with the proposed core-deficit model of dyslexia, the gifted children with dyslexia showed weaknesses in PA and RAN. However, they performed better on the PA and RAN tasks than the averagely intelligent children with dyslexia. Remarkably, the gifted children with dyslexia seemed to show a strength in VSTM rather than a deficit compared to their averagely intelligent peers with dyslexia. Concerning the compensational model of giftedness-related strengths, the gifted children with dyslexia indeed showed high performance on verbal and visuospatial WM tasks and language skills. It can be concluded that the high grammar scores and the exceptionally high scores on vocabulary indicate a major advantage compared to averagely intelligent children with dyslexia. General language skills possibly form an important area of compensation for gifted children with dyslexia. Although better language skills have been found a protective factor for all children with dyslexia (Nation & Snowling, 1998; Snowling, 2008), gifted children with dyslexia might benefit even more because they can rely on virtually excellent language skills.

To the best of our knowledge, this is the first study that has empirically examined the clinical expression and underlying cognitive profile of dyslexia in a gifted/high IQ population. It shows that masking of literacy difficulties can cause dyslexia to be hard to detect in gifted children for a protracted time, despite achievement also being considerably lower than anticipated on the basis of the intellectual capacities of the child. In addition, it shows that phonology can be considered a risk factor for the development of possible reading and/or spelling difficulties, but that its effect could be moderated by many other skills. In the case of gifted children with dyslexia, large WM capacity and excellent language skills may function as important protective factors. These findings fit the ideas of a multiple-deficit model of developmental learning disabilities (Pennington, 2006). Overall, the emergence of a specific learning disability, such as dyslexia, depends on a complex interplay of risk and protective factors, which are unique to specific populations and even differ per individual child (Pennington, 2006).

An additional novelty was the use of Bayesian statistics instead of standard frequentist statistics. Bayesian statistics allow the integration of prior knowledge in the evaluation of hypotheses and are especially suitable for studies were small sample sizes are an issue (Gill, 2008; Klugkist et al., 2005). Although the sample size in this study cannot be considered particularly small, it might cause power problems in relation to multiple testing when taking into account the amount of skills and cognitive components under investigation. Consequently, Bayesian statistics are a perfect fit to our data compared to traditional frequentist statistics, providing important solutions to multiple testing problems (Klugkist et al., 2011).

Practical implications of the study mainly involve raising awareness about the ways in which dyslexia might occur in gifted children. Even though it is premature to derive new diagnostic guidelines from these findings, it can be stated that teachers and diagnosticians should be more conscious about gifted children showing signs of underachievement, sudden deterioration in their school performance, or demotivation. Moreover, teachers always have the responsibility to take action when they notice a child is falling behind in a specific domain. However, this is often neglected when dealing with twice-exceptional children (Assouline et al., 2010). More alertness will hopefully improve the possibility of early intervention and prevent increasing severity of the impairment in a child's future school career, as well as promote quicker referral to gifted programs. The aim of early recognition by teachers or diagnosticians should not be to provide a label for the child but to identify the child's strengths and weaknesses and utilize this knowledge for the purpose of remediation and better service.

Future research should focus on cross-linguistic studies as well as longitudinal or cross-sectional studies of gifted children with dyslexia to assess development and outcomes at adolescence and adulthood. Furthermore, case series analyses could provide more insight in the etiological differences between gifted children with dyslexia at the individual level, including the borderline cases that were excluded from the analyses in the current study. Using larger sample sizes, including more children with diagnoses, and adding more/other cognitive components (e.g., executive functions or processing speed) will extend knowledge of the behavioral and cognitive risk and protective factors of gifted children with dyslexia. In combination with the replication of findings, this will hopefully result in earlier identification of gifted children with dyslexia and improve intervention and programming practices.

In summary, this study showed that gifted children with dyslexia outperform averagely intelligent children with dyslexia on literacy skills and that they have a unique cognitive profile characterized by both deficits related to their dyslexia and strengths associated with their giftedness. Findings suggest that weaknesses in phonology could be moderated by strengths in WM and general language ability. This renders reading and spelling ability levels that are not as low as in averagely intelligent children with dyslexia, which in turn, frustrates early signaling and referral. Overall, it can be stated that gifted children with dyslexia form a special group within the population of children with dyslexia as well as the population of gifted children with LD. More research into their characteristics and consequences for diagnosis and intervention is necessary to elucidate (potential) effects of masking and compensation.

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CHAPTER 5

RISK AND PROTECTIVE FACTORS IN GIFTED CHILDREN WITH DYSLEXIA

This study investigated risk and protective factors associated with dyslexia and literacy development, both at the group and individual level, to gain more insight in underlying cognitive profiles and possibilities for compensation in high-IQ children. A sample of 73 Dutch primary school children included a dyslexic group, a gifted-dyslexic group, and a borderline-dyslexic group (i.e., gifted children with relative literacy problems). Children were assessed on literacy, phonology, language, and working memory. Competing hypotheses were formulated, comparing the core-deficit view to the twice-exceptionality view on compensation with giftedness-related strengths. The results showed no indication of compensation of dyslexia-related deficits by giftedness-related strengths in gifted children with dyslexia. The higher literacy levels of borderline children compared to gifted children with dyslexia seemed the result of both fewer combinations of risk factors and less severe phonological deficits in this group. There was no evidence for compensation by specific strengths more relevant to literacy development in the borderline group. Accordingly, the findings largely supported the core-deficit view, whereas no evidence for the twice-exceptionality view was found. Besides practical implications, the findings also add to knowledge about the different manifestations of dyslexia and associated underlying cognitive factors at the higher end of the intelligence spectrum.

Keywords: dyslexia, giftedness, risk/protective factors, literacy, case series, Bayes.

van Viersen, S., de Bree, E. H., Kroesbergen, E. H., Slot, E. M., & de Jong, P. F. (2015). Risk and protective factors in gifted children with dyslexia. *Annals of Dyslexia, 65*, 178-198. doi: 10.1007/s11881-015-0106-y

INTRODUCTION

An abundant body of research focused on the underlying cognitive deficits associated with dyslexia, the severe and persistent reading and/or spelling difficulties at word level (Snowling, 2000). Research into these cognitive factors has covered various populations of children with dyslexia, including children of different intelligence levels (e.g., Elbro, 2010; Snowling, 2008; see Vellutino, Fletcher, Snowling, & Scanlon, 2004, for a review). However, high-IQ children (i.e., academic giftedness; Winner, 1997) have remained underresearched in this area of study. Results on averagely intelligent children are generalized to high-IQ populations based on the assumption that literacy and intelligence are only moderately related (e.g., Vellutino, Scanlon, & Lyon, 2000). Yet, gifted children are often believed to have a specific cognitive profile of dyslexia-related weaknesses and giftedness-related strengths, affecting the (clinical) expression of dyslexia (e.g., Brody & Mills, 1997; Foley Nicpon, Allmon, Sieck, & Stinson, 2011). The present study aims to test and extend theories on dyslexia and investigate possibilities for compensation by providing more insight in the underlying cognitive profiles of gifted children through the assessment of risk and protective factors at the group and individual level.

Current understanding of the etiology of learning disabilities such as dyslexia is best explained by the multiple deficit model (e.g., Bishop & Snowling, 2004; Pennington, 2006; Pennington et al., 2012; Ramus et al., 2003; Wolf & Bowers, 1999). According to this model, learning disabilities arise as a result of the interaction between multiple cognitive risk and protective factors that are influenced by both genes and the environment during development. The model is probabilistic in nature, with risk factors increasing the probability of the emergence of a learning disability and protective factors decreasing this probability. Considering that this model is the one of few to specifically take protective factors into account as part of the development of learning disabilities (Pennington, 2006), the multiple deficit model provides the most suitable framework for investigating the role of protective factors in the development of dyslexia in the current study.

Risk factors associated with dyslexia are generally believed to lie in phonological processing, which is proposed as a primary source of reading disabilities (Vellutino et al., 2004). The most important deficits concern phonological awareness (PA; i.e., detection and manipulation of speech sounds in spoken words), verbal short-term memory (VSTM; i.e., the temporary storage of phonological representations), and rapid automatized naming (RAN; i.e., access to and quick retrieval of orthographic and phonological representations from long-term memory;

Boets et al., 2010; de Jong & van der Leij, 2003; Snowling, 2001; Wagner & Torgesen, 1987). The dominant and commonly accepted view regarding these underlying cognitive risk factors is that they are the core deficits of dyslexia and are unaffected by intelligence (hereafter the 'core-deficit view'; Stanovich & Siegel, 1994). Children of high and low intelligence are assumed to display no marked differences in the underlying cognitive processes that lie at the basis of their reading disability (Stanovich, 1996). Nonetheless, despite its strong claims that have both theoretical and clinical consequences, the core-deficit view has never been tested in a gifted population.

A contrasting view on the role of intelligence in dyslexia can be found in the literature on twice-exceptionality (i.e., the co-occurrence of giftedness and a learning disability within an individual; Brody & Mills, 1997; see Foley Nicpon et al., 2011, for a review), which specifically addresses the role of protective factors. Protective factors are generally only mentioned in relation to 'compensation'. Compensation pertains to the moderation of the effect of a cognitive risk factor by an additional or co-occurring protective factor (e.g., Snowling et al., 2003). According to this 'twice-exceptionality view', the cognitive profile of twice-exceptional children is assumed to consist of high ability-related protective factors (providing possible resources for compensation) and disability-related risk factors (e.g., Assouline, Foley Nicpon, & Whiteman, 2010; Brody & Mills, 1997; Foley Nicpon et al., 2011). The combination of risk and protective factors may result in higher literacy levels than might be expected based on the underlying deficit, which is also referred to as 'masking' (e.g., Lovett & Lewandowski, 2006; Silverman, 2003). Accordingly, compensation is attested when, while matched on literacy performance, the cognitive profiles of gifted children with dyslexia are characterized by *more severe* deficits than averagely intelligent children with dyslexia, as otherwise there would be no room for protective factors to compensate. Protective factors proposed to be involved in compensation of phonological deficits include visual memory and perceptual speed (Snowling, 2001), orthographic knowledge (van der Leij & van Daal, 1999; Stanovich & Siegel, 1994), and/or general language skills (Nation & Snowling, 1998; Snowling, 2001, 2008; Snowling et al., 2003).

So far, only two studies have investigated risk and protective factors in gifted children with dyslexia. Berninger and Abbott (2013) found that *verbally* gifted children with dyslexia outperformed verbally average-performing children with dyslexia on reading, spelling, and language skills, but not on the cognitive deficits associated with dyslexia; phonological storage and processing, orthographic storage and processing, and RAN. A recent study by van Viersen et al. (2014) on the behavioral and cognitive characteristics of a group of overall gifted children with dyslexia, showed that this group exhibited weaknesses in PA and RAN, but not in VSTM, and strengths in verbal WM, visuospatial WM, and language skills compared to typically developing (TD) children. Furthermore, their reading and spelling performance was in between that of averagely intelligent children with dyslexia and TD children.

The findings of Berninger and Abbott (2013) indeed point toward compensation of underlying deficits by specific strengths, resulting in a certain degree of masking of literacy difficulties compared to averagely intelligent children with dyslexia. Based on their results, more severe cognitive deficits can be expected in gifted children with dyslexia when both groups are matched on literacy performance. The findings by van Viersen et al. (2014) are less easy to interpret, as the underlying deficits were reported to be less severe in gifted children with dyslexia and results were not controlled for literacy level. This is necessary for assessing real differences between cognitive profiles of both dyslexic groups and making direct claims about compensation or masking. Accordingly, more research into the cognitive profiles of gifted children with dyslexia, and specifically the combination between risk and protective factors, is needed.

An even more interesting group of gifted children to investigate in the context of risk and protective factors as well as compensation and masking concerns gifted children who read poorly compared to their IQ, but do not meet standards of low achievement needed for a diagnosis (i.e., 'borderline-dyslexic' children). These children are often not eligible for treatment. In other words, these borderline children show *relative* literacy problems, while their intelligence levels, and therefore general 'compensatory resources', are equal to gifted children that do reach the diagnostic threshold for dyslexia. Charting the cognitive risk and protective factors of both groups (and individual profiles within both groups) provides important insights in possibilities for compensation and masking, and explains their differences in literacy performance, thereby putting both the core-deficit view and twice-exceptionality view to the test.

In the present study, we aimed to test and extend theories on dyslexia and possibilities for compensation by investigating profiles of underlying cognitive risk and protective factors of two populations of gifted children, both at the group and individual level. First, the cognitive profiles of gifted children with dyslexia and averagely intelligent children with dyslexia were compared controlling for differences in literacy levels, by reanalyzing the data of two subgroups presented in van Viersen et al. (2014). Based on the core-deficit view, it was hypothesized that gifted children with dyslexia and averagely intelligent children with dyslexia would show equally large underlying deficits when controlling for literacy, with protective factors being present but independent of the risk profile. In contrast, the concomitant occurrence of larger deficits and more pronounced strengths in gifted children with dyslexia would support the twice-exceptionality view on compensation. Second, the cognitive profiles of gifted children with dyslexia and borderline-dyslexic children were compared controlling for differences in overall intelligence. Given the higher literacy level in the borderline group, it was hypothesized that this difference could be explained by either smaller deficits (core-deficit view), or the presence of *specific* protective factors more relevant for compensation, such as language skills (twice-exceptionality view), or both. An individualized approach toward both gifted groups was required for providing more information on *combinations* of deficits within children. It was hypothesized that the groups would not differ in the numbers of risk and protective factors overall. Instead, the breath/depth of underlying deficits (core-deficit view) and/or specific strengths (twice-exceptionality view) may account for differences between both groups in literacy development.

METHODS

PARTICIPANTS

A total of 73 Dutch primary school children (56.2% boys) from Grades 2 to 4 participated in the study. The children were recruited through contacts with school teachers or school psychologists, calls on the websites of educational magazines, and clinical institutions. Informed consent was obtained from all participants and their parents. The inclusion criterion for giftedness was set at a full IQ score > 125 or a 95% reliability interval tapping at least 130 in case of a short form. In addition, the criteria for dyslexia were set at 1) a significant discrepancy between IQ and reading *or* spelling performance of at least 2 SD (Snowling, 1998) and 2) below average scores on reading *or* spelling (lowest 10-15% or a standard score $\leq$ 6). The latter criterion is in line with an official diagnosis of dyslexia in The Netherlands (Kleijnen et al., 2008) and was the specific criterion that had to be fulfilled by the averagely intelligent children with dyslexia. The gifted children were divided in a group of children that met both criteria (i.e., gifted children with dyslexia; n = 26, 65.4% boys) and a borderline-dyslexic group (n = 14, 57.1% boys) that only met the first criterion. The children in the borderline group showed relative literacy problems compared to their high IQ but did not reach the cut-off for a dyslexia diagnosis.

Table 1 displays the division of age, intelligence, and sex in the groups and mean literacy levels. The groups significantly differ in age ($F[2, 70] = 9.52$, $p < .001$); the borderline group is significantly younger than both other groups ($p < .01$). As expected, the groups also differ significantly in intelligence ($F[2, 70] = 126.76$, $p < .001$). The gifted groups have about equal intelligence levels ($p = .37$), which differ significantly from the averagely intelligent dyslexic group ($p < .001$). All three groups significantly differ in word reading ($F[2, 70] = 26.83$, $p < .001$) and nonword reading levels ($F[2, 70] = 24.91$, $p < .001$), following the pattern borderline > gifted/dyslexic > dyslexic. There was a main effect of spelling level ($F[2, 70] = 10.26$, $p < .001$); the averagely intelligent children with dyslexia showed significantly lower scores than both gifted groups ($p < .01$).

TABLE 1

Sample Size and Means and Standard Deviations for Age, IQ score, and Literacy Measures per Group

	Gifted + Dyslexia (n = 26)		Borderline (n = 14)		Dyslexia (n = 33)	
Variable	*M*	*SD*	*M*	*SD*	*M*	*SD*
Age (months)	108.77	8.14	99.00	10.49	113.85	12.39
IQ (total)	132.50	8.05	135.29	4.58	98.70	11.41
Word reading[a]	5.77	2.75	9.64	1.69	4.06	2.33
Nonword reading[a]	6.27	2.20	9.14	1.61	4.64	2.00
Spelling[b]	3.12	12.08	10.56	13.54	-6.93	13.19

Note. Data of the gifted + dyslexia group and dyslexia group are also reported in van Viersen et al. (2014).
[a] Standard score ($M = 10$, $SD = 3$). [b] Instruction-residual.

INSTRUMENTS

INTELLIGENCE. A short form of the Dutch version (Kort et al., 2005) of the *Wechsler Intelligence Scale for Children – Third Edition* (WISC-III; Wechsler, 1991) was used to estimate the general cognitive abilities of the children. The short form consisted of the performance subtests picture completion and block design and the verbal subtests similarities and vocabulary. Full IQ scores ($M = 100$, $SD = 15$) were computed based on the formula provided by Kaufman, Kaufman, Balgopal, and McLean (1996) and used for inclusion. The reported reliability and validity quotients of this short form are all greater than .83 (Kaufman et al., 1996). Children were not re-assessed and recent results were used if a complete version of the WISC-III had already been administered in the past two years.

LITERACY. Timed word reading and decoding speed of nonwords were measured using *Eén Minuut Test* (EMT; Brus & Voeten, 1999) and *Klepel* (van den Bos, lutje Spelberg, Scheepstra, & de Vries, 1994), respectively. The child had one (words) and two minutes (nonwords) to accurately read as many (non)words as possible. In both tests word length increased from one to four syllables. Raw scores were the number of correctly read words or nonwords, with a maximum of 116, which were transformed into age-referenced norm scores (M = 10, SD = 3) for the inclusion. Internal consistency is .90 for EMT and .92 for Klepel (Evers et al., 2009-2012).

A short form of the *PI-dictee* (Geelhoed & Reitsma, 2000) was used to measure spelling dictation at word level. The short form contained eight sets of seven words, with each set representing specific spelling categories (P. F. de Jong, personal communication, September 2012). The test was discontinued when the child made six or more errors in one set. Raw scores are the total number of correctly written words. For the analyses, the total raw score was transformed into an instruction-residual after partialling out the number of months instruction in spelling (i.e., computed by 10 months of instruction per year of education). Internal consistency of the full version varies between .90 and .93 (Evers et al., 2009-2012).

PHONOLOGY. PA was assessed using the *Fonemische Analyse Test* (FAT; van den Bos, lutje Spelberg, & de Groot, 2011), which measured the ability to quickly analyze and manipulate phonemes in words in two subtests with 12 items each. The first subtest targets phoneme deletion (e.g., *kraal* 'bead' without /*k*/ is *raal*) and the second subtest is a spoonerism task (e.g., transposing onset phonemes of *Kees Bos* to *Bees Kos*). For both tasks, raw response time in seconds was transformed into an age-referenced norm score (M = 8-12). In addition, the number of correct answers per minute was computed based on raw speed and accuracy scores per task and used for the analyses. Internal consistency of the test is .93 (Evers et al., 2009-2012).

The task *Continu Benoemen & Woorden Lezen* (CB&WL; van den Bos & lutje Spelberg, 2007) was used to assess RAN. The test consisted of four subtests (colors, digits, pictures, and letters) measuring the child's naming speed. The child is asked to correctly name 50 items (5 types per subtest; e.g., red, yellow, blue, green, and black) as quickly as possible. Raw scores (i.e., naming time in seconds) of the subtests could be transformed into age-referenced norm scores (M = 8-12). The raw scores of the letters and digits subtests were combined into a (mean) alphanumeric RAN variable and the colors and pictures subtests were combined into a (mean) non-alphanumeric RAN variable for the analyses. Internal consistency of the test varies between .79 and .87 (Evers et al., 2009-2012).

The subtest digit recall of the *Automated Working Memory Assessment* battery (AWMA; Alloway, 2007) was used to measure VSTM, which required the child to recall several series of digits of increasing length. All subtests of this instrument were discontinued after three incorrect answers. Raw accuracy scores were used in the analysis and could also be transformed into age-referenced norm scores (*M* = 85-115). Test-retest reliability is .89 (Alloway, Gathercole, Kirkwood, & Elliot, 2009).

WORKING MEMORY. The verbal and visuospatial short-term and WM components were measured using subtests of the AWMA (Alloway, 2007). Backward digit recall, in which the child recalled increasing series of digits backwards, was used to measure verbal working memory (VWM). Visuospatial working memory (VSWM) was measured by the subtests spatial span (SS), in which the child had to evaluate figures by mental rotation, classify them as 'the same' or 'opposite', and recall the position of a red dot in an empty figure, and the subtest odd-one-out (OO), in which the child had to indicate the odd figure out of increasingly complex sequences of three figures and recall the odd figure in a matrix. Raw scores on both subtests were combined into a mean VSWM variable. Visuospatial short-term memory (VSSTM) was measured using the subtest dot matrix, in which the child had to recall the position of increasingly complex series of red dots in an empty matrix. Raw accuracy scores were used in the analyses, but could also be transformed into age-referenced norm scores (M = 85-115). Test-retest reliabilities were .86, .79, .88, and .85, respectively (Alloway et al., 2009).

LANGUAGE. The *Clinical Evaluation of Language Fundamentals-4 NL* (CELF; Kort, Schittekatte, & Compaan, 2010) was used to measure grammar and vocabulary. The subtest formulated sentences (FS) was used to assess the child's grammar skills. Here, the child had to formulate sentences about actions or situations that were expressed in pictures using a targeted word or phrase. The subtest word classes 2 (WC2), in which the child chose two related words and described their relationship, measured vocabulary. Raw accuracy scores (FS max = 40, WC2 max = 40) were used in the analyses and could also be transformed into age-referenced norm scores (*M* = 8-12). Internal consistency of the subtests is .78 and .87, respectively (Evers et al., 2009-2012).

PROCEDURE

The assessments were performed by supervised graduate students, who all received an extensive training in using the test battery described above. Children were tested either in a clinic, at school, or in their homes within one two-to-three-hour session, with plenty breaks in between tests. Duration of the assessment depended on the availability of recent and relevant test results for the instruments included in the test battery (i.e., IQ not older than 2 years, literacy not older than 6 months). The test results were summarized in a short report, evaluated by a licensed school psychologist, and reported back to the parents of the participating children. Any diagnostic uncertainties were resolved during joint evaluation meetings.

ANALYSES

GROUP ANALYSES

To test the first two hypotheses concerning the group comparisons between the gifted children with dyslexia and the averagely intelligent children with dyslexia (first hypothesis) and between the gifted children with dyslexia and the borderline-dyslexic children (second hypothesis), Bayesian statistics were used instead of traditional frequentist statistics (i.e., here the frequentist alternative would be MANCOVA). Bayesian statistics provide important benefits over traditional methods for the current study. First, in contrast to null hypothesis testing, Bayesian model selection offers the possibility to formulate and evaluate informative hypotheses based on prior knowledge by using equality and inequality constraints between groups and compare competing hypotheses that represent specific expectations or views (Klugkist, Laudy, & Hoijtink, 2005). Second, the outcome of the analyses is a Bayes factor (BF), which represents the amount of evidence in favor of one hypothesis compared to another (Kass & Raftery, 1995), instead of a traditional p-value. As such, Bayesian model selection offers solutions to analytical problems related to multiple testing, such as alpha inflation and loss of power after correcting the alpha level (Klugkist, van Wesel, & Bullens, 2011). Third, Bayesian analyses are not based on normality or asymptotic assumptions, making it a useful alternative when working with small sample sizes (Gill, 2008) or samples drawn from one side of the distribution (e.g., children with disabilities). Finally, the obtained parameter estimates (i.e., posterior means and posterior standard deviations) include the effect of the covariates and can directly be used to make inferences about groups differences. The analyses were conducted with the BIEMS software package (Mulder, Hoijtink, & Klugkist, 2010; Mulder, Hoijtink, & de Leeuw, 2012; Mulder et al., 2009).

For the analyses, each hypothesis was translated into a statistical hypothesis with (in)equality constrained parameters (Klugkist et al., 2005). In this study, the parameters were the group means (i.e., μ) on the cognitive risk and protective factors. The statistical hypotheses are displayed and further explained in Table 2. First, each of the informative hypotheses are compared to the alternative hypothesis by calculating the $BF_{i,u}$'s. A BF larger than 1 indicates support in favor of the informative hypothesis, whereas a BF smaller than 1 provides evidence in favor of the alternative hypothesis. A rule of thumb for the interpretation of the BF is provided by Kass and Raftery (1995), stating that a BF between 1 and 3 represents a small effect, a BF between 3 and 10 can be considered as substantial evidence supporting the informative hypothesis under investigation, and a BF above 10 indicates a large effect.

If one or more of the informative hypotheses received more support from the data than the alternative hypothesis, these competing hypotheses can be compared by calculating the posterior model probability (PMP) for each model (assuming prior probabilities to be equal for all models). This is done by dividing the $BF_{i,u}$ of each model by the sum of BFs of the other relevant models. The PMP represents the relative support for a specific hypothesis within a set of hypotheses (Klugkist et al., 2011). Note that in a Bayesian framework, the term 'probability' refers to a degree of belief and relates to the probability that a hypothesis is true (Klugkist et al., 2011). When the difference between the PMPs of two models is smaller than .05, the models are considered to be equally supported by the data. The parameter estimates were used to make more detailed inferences about differences between the groups. Group differences on cognitive risk and protective factors were evaluated using both multivariate and univariate testing.

To adjust for differences in literacy levels between the gifted children with dyslexia and the averagely intelligent children with dyslexia, age and a literacy composite were used as covariates. The literacy composite consisted of the average z-scores of the word reading and nonword reading norm scores and the instruction-residual of the raw spelling score. Age and IQ score were used as covariates for the comparison of the cognitive profiles between the gifted children with dyslexia and the borderline-dyslexic children.

INDIVIDUAL ANALYSES

A case series analysis was conducted on the data of the gifted children with dyslexia and the borderline children for assessing individual differences between cognitive profiles of strengths and weaknesses of these children (third hypothesis). For each cognitive risk factor, impaired performance was defined as a standard score on the relevant measures below a cut-off of -1 standard deviation relative to the norm-based mean. This criterion is also used in the official diagnosis of dyslexia in The Netherlands (Kleijnen et al., 2008) and has been found useful in previous research on subgroups of poor readers (e.g., Catts, Hogan, & Fey, 2003; Nag & Snowling, 2011; Ramus et al., 2003). A cut-off of +1 standard deviation relative to the norm-based mean was used to identify cognitive protective factors. In addition, risk and protective factors were combined into strengths and weaknesses, representing dyslexia-related and giftedness-related cognitive domains. Below average performance on either or both the deletion and spoonerism task was counted as a weakness in PA. Similarly, impaired performance on either or both alphanumeric and non-alphanumeric RAN was considered a weakness in RAN. As VSTM has been found to be a potential strength in gifted children with dyslexia (van Viersen et al., 2014), it could be mapped as either a risk or a protective factor per child. Above average performance on either or both VSTM and VWM was considered a strength in verbal WM abilities and high performance on either or both VSSTM and VSWM was counted as a strength in visual WM skills. High scores on either or both grammar and vocabulary were considered a strength in language skills.

TABLE 2

Tested Models, Statistical Hypotheses, and Translation for Bayesian Analyses

Model	Statistical hypothesis	Translation
Gifted + Dyslexia vs. Dyslexia		
Weaknesses		
Model 0	μ_D, μ_{GD}	Alternative hypothesis or unconstrained model
Model 1	$\mu_D > \mu_{GD}$	**Twice-exceptionality view:** Dyslexic children have higher scores on the cognitive risk factors than gifted/dyslexic children, indicating more severe deficits in the gifted/dyslexia group
Model 2	$\mu_D = \mu_{GD}$	**Core-deficit view:** Both groups have about equal scores on the cognitive risk factors, indicating that the groups are comparable
Strengths		
Model 0	μ_D, μ_{GD}	Alternative hypothesis or unconstrained model
Model 1	$\mu_D < \mu_{GD}$	**Twice-exceptionality view:** Dyslexic children have lower scores on the cognitive protective factors than gifted/dyslexic children, indicating more pronounced strengths in the gifted/dyslexia group
Model 2	$\mu_D = \mu_{GD}$	Both groups have about equal scores on the cognitive protective factors, indicating that the groups are comparable
Gifted + Dyslexia vs. Borderline-dyslexic		
Weaknesses		
Model 0	μ_B, μ_{GD}	Alternative hypothesis or unconstrained model
Model 1	$\mu_B > \mu_{GD}$	**Core-deficit view:** Borderline-dyslexic children have higher scores on the cognitive risk factors than the gifted/dyslexic children, indicating less severe deficits in the borderline group
Model 2	$\mu_B = \mu_{GD}$	Both groups have about equal scores on the cognitive risk factors, indicating that the groups are comparable
Strengths		
Model 0	μ_B, μ_{GD}	Alternative hypothesis or unconstrained model
Model 1	$\mu_B > \mu_{GD}$	**Twice-exceptionality view:** Borderline-dyslexic children have higher scores on the cognitive protective factors than the gifted/dyslexic children, indicating more pronounced strengths in the borderline group
Model 2	$\mu_B = \mu_{GD}$	Both groups have about equal scores on the cognitive protective factors, indicating that the groups are comparable

Note. μ represents the group mean. D = dyslexia; GD = gifted + dyslexia; B = borderline-dyslexic.

RESULTS

GROUP COMPARISONS

For the first part of the analyses, cognitive profiles of gifted children with dyslexia and averagely intelligent children with dyslexia were compared, controlling for differences in literacy level. Table 3 shows the posterior means and standard deviations of the gifted children with dyslexia and the children with dyslexia, as well as the BFs and PMPs for the three models in the analyses of the cognitive factors.

The multivariate results show that overall the alternative hypothesis received most support from the data for the *risk* factors (Model 0; PMP = .84). The univariate results show that this is the case for both PA tasks and VSTM (PMPs .98-.94). The posterior means of the unconstrained model indicate that the gifted children with dyslexia had neither equal nor lower scores on PA and VSTM compared to the children with dyslexia, but instead had higher scores. This indicates that gifted children with dyslexia may have *less* severe deficits in these areas. For RAN, Model 2 received most support from the data, about two times more than the alternative hypothesis, with the probabilities that the hypothesis under Model 2 is true varying between .56 and .53. The posterior means show that controlling for literacy level, the gifted children with dyslexia and the children with dyslexia have similar scores on RAN.

The multivariate results for the *protective* factors show that overall Model 1 received most support from the data, about 16 times more than the alternative hypothesis, indicating a large effect (PMP = .94). This is supported by univariate results, showing that indeed Model 1 received most support from the data for all protective factors (PMPs .67-.63), except for grammar where the difference between both models is too small to interpret (ΔPMP < .05). The posterior means show that on verbal WM, visuospatial STM and WM, and on vocabulary the gifted children with dyslexia obtained higher scores than the children with dyslexia, whereas on grammar both groups had comparable scores when controlled for differences in literacy levels. Overall, the results show that the gifted children with dyslexia have equal or even less severe deficits on risk factors than averagely intelligent children with dyslexia, and indeed strengths on a broad range of protective factors.

TABLE 3

Posterior Means (PM) and Standard Deviations (PSD) Adjusted for Age and Literacy Performance and Bayes Factors (BF) and Posterior Model Probabilities (PMP) of the Three Models for the Cognitive Risk and Protective Factors of the Gifted/Dyslexic and Dyslexic Children

	Dyslexia (n = 33)		Gifted + Dyslexia (n = 26)		Model 0 (μ_D, μ_{GD})		Model 1		Model 2 ($\mu_D = \mu_{GD}$)	
Skill/component	PM	PSD	PM	PSD	BF	PMP	BF	PMP	BF	PMP
Risk factors							($\mu_D > \mu_{GD}$)[a]			
Multivariate					**1.00**	.84	0.00	.00	0.19	.16
Univariate										
Phoneme deletion	11.32	1.34	17.51	1.68	**1.00**	.98	0.00	.00	0.02	.02
Phoneme transposition	1.90	0.07	3.08	0.09	**1.00**	.96	0.00	.00	0.03	.03
RAN alphanumeric	33.71	1.09	32.84	1.38	1.00	.28	0.58	.16	**2.00**	.56
RAN non-alphanumeric	55.68	3.01	53.33	3.72	1.00	.35	0.36	.12	**1.53**	.53
VSTM	22.63	0.62	25.98	0.77	**1.00**	.94	0.01	.01	0.05	.05
Protective factors							($\mu_D < \mu_{GD}$)			
Multivariate					1.00	.06	**16.1**	.94	0.03	.00
Univariate										
Verbal WM	10.40	0.47	12.76	0.58	1.00	.32	**1.98**	.63	0.17	.06
Visuospatial STM	21.34	0.61	26.10	0.77	1.00	.33	**2.00**	.67	0.00	.00
Visuospatial WM	14.93	0.64	19.99	0.81	1.00	.33	**2.00**	.67	0.00	.00
Grammar	26.32	0.83	27.50	1.04	1.00	.24	**1.62**	.39	**1.52**	.37
Vocabulary	13.50	0.67	17.70	0.84	1.00	.33	**2.00**	.66	0.01	.00

Note. μ represents the group mean. Bold face indicates BFs of models that received most support from the data. RAN = rapid automatized naming; VSTM = verbal short-term memory; WM = working memory; STM = short-term memory; D = dyslexia; GD = gifted + dyslexia.
[a] $\mu_D < \mu_{GD}$ for both RAN variables, because higher numbers indicate slower performance.

For the second part of the analyses, gifted children with dyslexia and borderline-dyslexic children were compared on their cognitive profiles, controlling for differences in overall intelligence. Table 4 displays the posterior means and standard deviations of the gifted children with dyslexia and the borderline children, as well as the BFs and PMPs for the three models in the analyses of the risk and protective factors.

For the *risk* factors, the multivariate results show that overall Model 2 received most support from the data, about three times more than the alternative hypothesis indicating a small to medium effect (PMP = .46). Yet, the univariate results show that Model 1 received most support from the data for the PA tasks (PMPs .42-.45). The posterior means indicate that the borderline children indeed have higher scores on both PA tasks than the gifted children with dyslexia, indicating less severe deficits. For RAN, Model 1 and 2 received about equal support from the data (ΔPMP < .05), although the posterior means show that the gifted children with dyslexia were somewhat slower on the tasks than the borderline children. In other words, both groups have comparable RAN levels, but the borderline group has significantly better literacy skills. This indicates that the children in the borderline group have *relatively* larger RAN deficits than the gifted children with dyslexia. For VSTM, Model 2 received most support from the data (PMP = .48), indicating that both groups obtained comparable scores.

For the *protective* factors, the multivariate results show that overall Model 2 received most support from the data, about four times more than the alternative hypothesis, indicating a medium effect (PMP = .61). The univariate results show that indeed Model 2 received most support for the visuospatial STM and language factors (PMPs .50-.39), but not for verbal WM, for which, as for the risk factors, Model 1 received most support from the data (PMP = .41). For visuospatial WM, Model 1 and 2 received about equal support from the data (ΔPMP < .05). The posterior means indicate that the gifted children with dyslexia have lower scores on verbal WM than the borderline children. On the other protective factors, both groups appear to have comparable scores, showing that borderline-dyslexic children have neither more pronounced strengths in general, nor more pronounced strengths in, for example, language skills that are considered more relevant to literacy development. Overall, the results show that borderline children have (at most) equal or less severe deficits on risk factors than gifted children with dyslexia, and a comparable and about equally strong set of protective factors, apart from verbal WM.

Table 4

Posterior Means (PM) and Standard Deviations (PSD) Adjusted for Age and IQ and Bayes Factors (BF) and Posterior Model Probabilities (PMP) of the Three Models for the Cognitive Risk and Protective Factors of the Gifted/Dyslexic and Borderline-Dyslexic Children

	Gifted + Dyslexia (n = 26)		Borderline (n = 14)		Model 0 (μ_B, μ_{GD})		Model 1 ($\mu_B > \mu_{GD}$)[a]		Model 2 ($\mu_B = \mu_{GD}$)	
Skill/component	PM	PSD	PM	PSD	BF	PMP	BF	PMP	BF	PMP
Risk factors										
Multivariate					1.00	.14	2.89	.40	**3.28**	.46
Univariate										
Phoneme deletion	17.94	2.21	20.19	3.98	1.00	.26	**1.64**	.42	1.28	.33
Phoneme transposition	3.10	0.16	3.77	0.28	1.00	.27	**1.69**	.45	1.06	.28
RAN alphanumeric	34.01	2.51	32.32	4.50	1.00	.25	**1.50**	.38	**1.48**	.37
RAN non-alphanumeric	55.98	4.62	53.36	8.12	1.00	.25	**1.55**	.39	**1.38**	.35
VSTM	25.91	0.56	25.45	0.98	1.00	.30	0.69	.21	**1.59**	.48
Protective factors										
Multivariate					1.00	.16	1.36	.22	**3.76**	.61
Univariate										
Verbal WM	12.55	0.34	13.31	0.60	1.00	.26	**1.58**	.41	1.24	.33
Visuospatial STM	25.18	0.72	25.52	1.29	1.00	.25	1.19	.30	**1.83**	.46
Visuospatial WM	19.31	0.58	20.03	1.04	1.00	.25	**1.44**	.36	**1.55**	.39
Grammar	26.56	0.92	25.76	1.66	1.00	.31	0.62	.19	**1.65**	.50
Vocabulary	18.29	0.76	18.03	1.32	1.00	.27	0.86	.24	**1.78**	.49

Note. μ represents the group mean. Bold face indicates BFs of models that received most support from the data. RAN = rapid automatized naming; VSTM = verbal short-term memory; WM = working memory; STM = short-term memory; B = borderline-dyslexic; GD = gifted + dyslexia.

[a] $\mu_B < \mu_{GD}$ for both RAN variables, because higher numbers indicate worse performance.

CASE SERIES

Finally, a case series analysis was performed on the gifted children with dyslexia (Table 5) and the borderline-dyslexic children (Table 6)[4]. Instead of showing differences in group averages on risk and protective factors (size/severity), this case series provides more information about heterogeneity within both gifted groups and individual differences in numbers of risk and protective factors and breadth/depth of strengths and weaknesses.

The case series analysis shows that both groups contain about equal percentages of children with a deficit in PA (i.e., GD = 46.2%, B = 50.0%) and the gifted/dyslexia group has a higher percentage of children with a RAN deficit than the borderline group (i.e., GD = 76.9%, B = 57.1%). In other words, despite the more severe PA deficit for the gifted/dyslexia group compared to the borderline group, the gifted/dyslexia group did not include *more* children with a deficit. Only 7.7% of the gifted/dyslexic children and 14.3% of the borderline children show a deficit in VSTM. On the other hand, none of the borderline children and only 15.3% of the gifted children with dyslexia show a strength in VSTM.

Besides individual deficits, combinations of deficits can play a role; the gifted/dyslexia group might show accumulation of risk factors for the development of severe literacy difficulties. Table 5 and Table 6 show that both groups have about the same number of weaknesses on average. Although the groups are comparable in their percentages of children with no (GD = 15.4%, B = 14.3%) or three deficits (GD = 7.7%, B = 7.1%), the borderline group contains more children with a single deficit (GD = 46.2%, B= 57.1%) and the gifted/dyslexia group more children with a double deficit (i.e., mostly a combination between PA and RAN; GD = 30.8%, B = 21.4%). This higher percentage of children with a double deficit in the gifted/dyslexia group directly translates to the higher percentage of children with a RAN deficit in this group mentioned above.

[4] Please note that, although the differences in profiles between the gifted/dyslexia group and the borderline group do not seem to be large, both gifted groups significantly differ from averagely intelligent children with dyslexia in their number of risk and protective factors as well as the breadth/depth of their weaknesses and strengths. Summary of the results for the case series analysis of the averagely intelligent children with dyslexia: PA deficit = 87.9%, RAN deficit = 84.8%, VSTM deficit = 39.3%; no deficits = 0.0%, one deficit = 18.2%, two deficits = 51.5%, tree deficits = 30.3%; Verbal WM strength = 0.0%, Visual WM strength = 27.3%, Language strength = 12.1%; no strengths = 60.6%, one strength = 39.4%, two strengths = 0.0%, three strengths = 0.0%. The complete table is displayed in Appendix D.

TABLE 5

Findings from the Case Series showing the Cognitive Risk and Protective Factors observed among Gifted/Dyslexic Children with Severe Literacy Difficulties

	Risk factors					Protective factors								
Case no.	PA deletion SS < 8	PA spoonerism SS < 8	RAN alpha-numeric SS < 8	RAN non-alphanumeric SS < 8	VSTM SS < 85 SS > 115	VWM SS > 115	VSSTM SS > 115	VSWM SS > 115	Gram-mar SS > 12	Voca-bulary SS > 12	IQ score	Literacy compo-site	Number of weak-nesses	Number of strengths
208	-		-	-	+		+	+	+		133	-0.53	2	3
6			-	-			+		+		>145	-0.41	1	2
30		-	-	-			+	+	+	+	138	-1.08	2	2
197	-		-	-	+	+	+	+			133	-0.65	2	2
22				-				+	+	+	126	-1.19	1	2
124	-	-	-	-			+				133	-0.64	2	1
29	-	-	-	-			+	+			124	-0.57	2	1
83	-		-	-	+		+				123	-1.03	2	2
93			-	-			+				124	-1.00	1	1
87					+	+	+	+	+	+	>145	0.10	0	3
49							+				130	-0.15	0	1
91	-					+	+	+		+	133	-0.21	1	3
35			-				+	+			137	0.19	1	1
67	-	-						+	+		125	-0.20	1	2
109		-	-	-			+		+		>145	-0.08	2	2
1			-	-			+	+			129	-0.56	1	1
51			-	-			+				121	-0.19	1	1
15	-	-	-				+				123	-0.94	2	1
204				-			+		+	+	139	0.80	1	2
60				-							137	0.05	1	0
53	-	-		-	-		+	+		+	145	0.19	3	2
193			-	-		+	+	+	+		142	-0.11	1	3
125	-		-		-		+	+			135	0.49	3	1
65			-				+	+			121	-0.09	1	1
13							+		+	+	124	-0.05	0	2
34									+		135	0.47	0	1
Total	10	7	16	16	2/4	4	22	14	11	7	132.5	-0.29	1.31	1.65

Note. PA = phonological awareness; RAN = rapid automatized naming; VSTM = verbal short-term memory; VWM = verbal working memory; VSSTM = visuospatial short-term memory; VSWM = visuospatial working memory; SS = standard score.

TABLE 6

Findings from the Case Series showing the Cognitive Risk and Protective Factors observed among the Borderline-Dyslexic Cases with Relative Literacy Problems

Case no.	Risk factors					Protective factors					IQ score	Literacy compo-site	Number of weak-nesses	Number of strengths
	PA deletion SS < 8	PA spoonerism SS < 8	RAN alpha-numeric SS < 8	RAN non-alphanumeric SS < 8	VSTM SS < 85 SS > 115	VWM SS > 115	VSSTM SS > 115	VSWM SS > 115	Gram-mar SS > 12	Voca-bulary SS > 12				
7			-		-	+	+	+	+	+	132	1.04	2	3
5	-	-					+	+		+	138	0.86	1	2
130		-	-	-		+					135	-0.21	2	1
131	-	-				+		+			137	0.70	1	2
69				-					+		135	0.55	1	1
14	-		-	-	-			+		+	136	-0.32	3	2
92		-									130	0.68	1	0
31			-	-			+	+		+	137	-0.17	1	2
185			-	-							130	0.25	1	0
132			-	-				+		+	131	0.37	1	2
173	-	-	-	-			+	+			138	1.28	2	1
129	-	-					+	+			141	1.15	1	1
149						+	+			+	>145	0.05	0	3
177						+	+	+			129	1.20	0	2
Total	5	6	7	7	2/0	5	7	9	2	6	135.3	.53	1.21	1.57

Note. PA = phonological awareness; RAN = rapid automatized naming; VSTM = verbal short-term memory; VWM = verbal working memory; VSSTM = visuospatial short-term memory; VSWM = visuospatial working memory; SS = standard score.

The case series analysis on the *protective* factors largely confirms the pattern that the group comparison of the gifted children with dyslexia and the borderline children showed. The gifted/dyslexia group contains a lower percentage of children with a strength in verbal WM (i.e., GD = 23.1%, B = 35.7%). Furthermore, in both groups, half of the children show a strength in language skills, including grammar, of which the group comparisons indicated that it might not be strength in gifted/dyslexic children overall. Remarkably, visual WM is a strength for many of the children in the gifted/dyslexia group, even compared to the borderline group (i.e., GD = 92.3%, B = 71.4%). These percentages show that the borderline children do not have more or more relevant protective factors that may explain their higher literacy levels, which is supported by the percentages of combinations of strengths. Both groups have about equal percentages of children with two (i.e., GD = 38.5%, B = 42.9%) or three strengths (i.e., GD = 15.4%, B = 14.3%). However, the gifted/dyslexia group contains fewer children with no strengths at all (i.e., GD = 3.8%, B = 14.3%) and more children with a single strength (i.e., GD = 42.3%, B = 28.6%). This higher percentage of children with a single strength is directly related to the higher number of children with a strength in visual WM in the gifted/dyslexia group.

DISCUSSION

In the present study, we aimed to test and extend theories on cognitive risk and protective factors in dyslexia, by investigating cognitive profiles of gifted children at the group and individual level. To gain more insight in possibilities for compensation and masking of learning disabilities, three groups were compared in the light of two competing views; the core-deficit view (e.g., Stanovich, 1996) and the twice-exceptionality view (e.g., Assouline et al., 2010; Foley Nicpon et al., 2011). First, a comparison was made between gifted children with dyslexia and averagely intelligent children with dyslexia, controlling for literacy level. Second, gifted children with dyslexia and borderline-dyslexic children (i.e., gifted children with relative literacy problems) were compared, controlling for differences in overall intelligence. Third, individual profiles within both gifted groups were investigated to provide more information about differences in overall numbers of risk and protective factors, the breadth/depth of underlying deficits, and the specificity of strengths.

The first comparison showed that gifted children with dyslexia had similar (low) scores on RAN compared to averagely intelligent children, but also seemingly less severe deficits in PA and VSTM. Evidently, gifted children with dyslexia do not have larger underlying deficits than children with dyslexia, indicating no support for the twice-exceptionality view. The similar scores on RAN in both groups provide partial support for the core-deficit view. Moreover, the gifted children with dyslexia showed high performance across a broad range of protective factors. The equal scores on grammar, which is assumed to be more strongly related to literacy than most of the other factors, suggests that the high scores on the other protective factors are relatively independent of the scores on the risk factors and the literacy performance. This is also largely in line with the core-deficit view. The finding that RAN levels of gifted children and averagely intelligent children with dyslexia were similar indicates that a RAN deficit is an important risk factor underlying the literacy deficits in both groups.

The second comparison indicates that the higher literacy levels of the borderline-dyslexic children compared to those of the gifted children with dyslexia could be explained by less severe underlying cognitive deficits of the borderline group, which is in line with the core-deficit view. Although the multivariate results indicated that the profiles of risk factors were comparable between both groups, the univariate results showed that gifted children with dyslexia on average have more severe deficits in PA and RAN. In addition, the case series analyses indicated that the phonological problems of the gifted children with dyslexia are not only on average more severe, but also more often tend to concern a combination of deficits than in the borderline children. Both groups had about equal scores on VSTM. Coupling the VSTM findings to the low percentage of children with a deficit in the case series analysis, VSTM does not seem to be a general weakness in most gifted children with literacy difficulties. This is in line with previous findings by van Viersen et al. (2014) as well as findings in the general population (Moll, Loff, & Snowling, 2013). However, the low percentage of children with above average scores on VSTM shows that it might not be a strength either and illustrates the relatively strong performance of gifted children on this task compared to averagely intelligent children with dyslexia.

The hypothesis that borderline children might have higher literacy levels because they possess specific strengths more relevant to literacy development (e.g., language skills), as suggested by the twice-exceptionality view, was not supported. In line with their equal intelligence levels, borderline children and gifted children with dyslexia largely share their profile of cognitive strengths and showed comparable percentages of children with a strength in language skills, reducing the possibility of domain-specific compensation. The borderline children only outperformed gifted children with dyslexia on verbal WM, both in group means as well as number of borderline children with a strength in this area. Verbal WM may not be a strength in gifted children with dyslexia because it is related to phonological memory (Vellutino et al., 2004), which is more impaired in gifted children with dyslexia than in the borderline children. On the other hand, for almost all gifted children with dyslexia visual WM was considered a strength. Apparently there are relatively more children in the gifted/dyslexia group than in the borderline group who have a single strength in visual WM, but broader impairments in phonology and language-related skills, which provides an indication of the breadth and specificity of the problems of these children.

Based on these findings it can be stated that, in line with the core-deficit view, literacy performance (measured by core tasks generally used in diagnosing dyslexia) and the associated underlying cognitive deficits seem to be largely independent of intelligence (Stanovich, 1996), *also* in gifted populations with literacy problems. The counterevidence, i.e., the higher scores on PA and VSTM of the gifted children with dyslexia compared to the averagely intelligent children with dyslexia, may (partly) be the result of the 'task impurity problem' (van der Sluis, de Jong, & van der Leij, 2007), which concerns compensation on task-related aspects. Skills involved in on-task compensation may either be related to literacy ability, such as a larger vocabulary leading to higher familiarity with words in the task, or unrelated to literacy ability, i.e., higher processing speed (Catts, Gillispie, Leonard, Kail, & Miller, 2002; Johnson et al., 2003). For example, both PA tasks were timed and 'phonological speed' of the children played a prominent role in the assessment of task performance. This possibility is supported by the findings of the comparison between both gifted groups. The borderline children showed less severe deficits and no specific strengths, explaining their higher literacy levels, while resources for compensation on task-related aspects were equal across both groups.

As stated earlier, most studies appear to agree that compensation pertains to the moderation of the effect of a cognitive risk factor by an additional or co-occurring protective factor, but it remains unclear how this might actually work. The current study found no evidence for compensation by IQ-related factors as proposed by the twice-exceptionality view. Accordingly, compensation does not entail a balance between risk and protective factors (at a specific point in time), resulting in remediation of the defective underlying skill and better literacy performance at the outcome level. However, other types of compensation remain relevant in relation to gifted children. For example, even though the task impurity problem involves compensation on task-specific aspects and not compensation in relation to a defective skill, it is clear that this type of compensation poses serious challenges to diagnostic practice. The higher task performance ensuing from task-related compensation frustrates finding and proving the existence of a severe underlying deficit, providing an incorrect picture of the child's skills and deficits.

Another interesting option, next to on-task compensation, is compensation by a protective factor *during* development, resulting in the emergence of compensatory mechanisms or strategies that can be used in more complex literacy tasks, such as text reading and reading comprehension (e.g., Nation & Snowling, 1998). This type of compensation implies that there may be alternative ways to cope with problems that are involved in literacy performance, resulting in better performance at the outcome level than would be predicted based on the deficient underlying skill. This type of compensation is also explicitly proposed within the twice-exceptionality framework (e.g., Reis, McGuire, & Neu, 2000). Possible factors involved in this type of compensation might be the amount of print exposure (Mol & Bus, 2011) and orthographic learning (Wang, Castles, Nickels, & Nation, 2011). For example, when gifted children with dyslexia are better able to learn orthographic knowledge (i.e., letter clusters) than averagely intelligent children with dyslexia, they can read and recognize longer words and/or words of larger complexity (Bekebrede, van der Leij, & Share, 2009). This orthographic knowledge can be used as a compensatory mechanism to *circumvent* their phonological deficit and improve their performance on the literacy task (van der Leij & van Daal, 1999). This type of compensation is more in line with literacy outcomes determined by the multiple deficit model, because it underlines the developmental nature of interactions between risk and protective factors and illustrates why protective factors should be taken into account in theories on dyslexia.

The present results suggest that borderline children do not compensate underlying deficits, masking their literacy difficulties to an extent that they unjustly miss a dyslexia diagnosis. The term 'borderline-dyslexic' is thus appropriate, as the children's literacy problems may not be severe enough to qualify for dyslexia. Yet, borderline children experience educational problems directly related to the *consequences* of the discrepancy between their high intelligence and lower literacy levels. Elbro (2010) refers to this issue by differentiating between dyslexia as a disability, i.e., poor ability, and dyslexia as a handicap, i.e., the consequences of poor ability. For these children, their handicap may predominantly lie in the discrepancy between their literacy level and the difficulty of the texts they are supposed to be able to read based on their intelligence.

The results do provide implications for diagnostic practice. Given that the relation between phonological deficits and literacy difficulties is not causal (e.g., Pennington, 2006), we contend that it is important to continue to diagnose dyslexia based on literacy performance at the behavioral level and *not* include literacy-related cognitive deficits as a requirement for a diagnosis. Mapping of risk and protective factors should solely be used for underpinning a diagnosis and/or providing clues for intervention. With respect to the borderline children, adhering to diagnosis at the behavioral level would confirm that these children are indeed not dyslexic based on their literacy levels. However, future research should establish whether these children might actually experience educational problems as the result of their relative literacy problems or 'handicap' (e.g., Assouline, Foley Nicpon, & Huber, 2006; Elbro 2010), especially since the decision as to whether a child is eligible for remediation or intervention is dependent on the diagnosis of dyslexia.

There are some limitations to this study. First, due to the proximal analysis of literacy and underlying cognitive abilities, it was not possible to *directly* test mechanisms of compensation and masking in the present study. Instead, inferences were based on patterns of literacy and cognitive skills across groups and individual children. More information is needed about the way skills included here contribute to literacy processes to make more detailed statements about compensation and masking in future studies. Furthermore, our sample size was relatively small, including a limited group of borderline children, and the effects attested were not very large. Our choice for a Bayesian approach instead of traditional statistics allowed for the comparison of two opposing views and effectively dealt with the problems associated with small groups that are generally found at the end of the normal distribution. Combined with a detailed case series analyses, it allowed us to visualize the patterns of risk and protective factors and showed a clear trend toward distinctive cognitive characteristics in both gifted groups. Nonetheless, the results should be interpreted with caution and warrant replication in larger samples and different age groups.

In summary, this study confirmed that the profiles of underlying cognitive risk factors of children with dyslexia are largely independent of intelligence, also in gifted populations. We found no clear evidence for direct compensation of dyslexia-related cognitive risk factors by giftedness-related (and literacy-related) protective factors in gifted children with dyslexia. Moreover, gifted children with dyslexia and borderline-dyslexic children can be distinguished based on their literacy levels and the severity and breadth/depth of their underlying cognitive deficits. There was no evidence for compensation through specific protective factors that would further explain the differences in literacy levels between both gifted groups. Yet, other types of compensation remain possible, illustrating the importance of further investigating the role of protective factors and extend theories as well as aid diagnostic practice.

CHAPTER 6

FOREIGN LANGUAGE READING AND SPELLING IN GIFTED STUDENTS WITH DYSLEXIA IN SECONDARY EDUCATION

A few studies suggest that gifted children with dyslexia have better literacy skills than averagely intelligent children with dyslexia. This finding aligns with the hypothesis that giftedness-related factors provide compensation for poor reading. The present study investigated whether, as in the native language (NL), the level of foreign language (FL) literacy of gifted students with dyslexia is higher than the literacy level of averagely intelligent students with dyslexia and whether this difference can be accounted for by the difference in their NL literacy level. The sample consisted of 148 Dutch native speaking secondary school students divided in four groups: dyslexia, gifted/dyslexia, typically developing (TD), and gifted. All students were assessed on word reading and orthographic knowledge in Dutch and English when they were in 7th or 8th grade. A subsample (n = 71) was (re)assessed on Dutch, English, French, and German literacy one year later. Results showed that Dutch gifted students with dyslexia have higher NL literacy levels than averagely intelligent students with dyslexia. As in the NL, a stepwise pattern of group differences was found for English word reading and spelling, i.e., dyslexia < gifted/dyslexia < TD < gifted. However, it was not found for French and German literacy performance. These results point toward compensation: the higher English literacy levels of gifted/dyslexic students compared to their averagely intelligent dyslexic peers result from mechanisms that are unique to English as an FL. Differences in results between FLs are discussed in terms of variation in orthographic transparency and exposure.

Keywords: dyslexia, giftedness, foreign language, secondary education, Bayes.

van Viersen, S., de Bree, E. H., Kalee, L., Kroesbergen, E. H., & de Jong, P. F. (2017). Foreign language reading and spelling in gifted students with dyslexia in secondary education. *Reading and Writing: An Interdisciplinary Journal*. Advance online publication. doi: 10.1007/s11145-016-9717-x

INTRODUCTION

A few studies have suggested that gifted children with dyslexia, despite having severe and persistent word level literacy difficulties, read and spell better than their averagely intelligent peers with dyslexia (Berninger & Abbott, 2013; van Viersen, Kroesbergen, Slot & de Bree, 2014). These studies have mainly concerned primary school children's literacy levels in the native language (NL). The current study focused on secondary education students' reading and spelling acquisition in a foreign language (FL). We examined whether, as in the NL, the level of FL literacy of gifted students with dyslexia is higher than the literacy level of averagely intelligent students with dyslexia. In addition, we investigated whether such a difference can be accounted for by the difference in literacy level in their NL.

There are two possible explanations for the attested higher literacy level of gifted children with dyslexia compared to averagely intelligent children with dyslexia. The first is that these children have a less severe underlying (cognitive) deficit. In other words, gifted children with dyslexia have somewhat higher literacy levels simply because they have slightly better literacy-related skills (core-deficit account; Stanovich & Siegel, 1994). Such findings have indeed been reported (van Viersen, de Bree, Kroesbergen, Slot, & de Jong, 2015). However, van Viersen et al. (2015) argue that the higher performance of gifted children with dyslexia on tasks measuring underlying deficits is not the result of better underlying skills, but is likely due to giftedness-related strengths (e.g., processing speed; Johnson, Im-Bolter, & Pascual-Leone, 2003) impacting on task-specific aspects unrelated to the targeted skill. An alternative explanation would be that gifted students with dyslexia actually have equally severe deficits but have higher literacy levels due to compensation (compensation account; e.g., Foley Nicpon, Allmon, Sieck, & Stinson, 2011). Here, compensation refers to the development of compensatory mechanisms or strategies associated with specific protective factors that are relatively unrelated to the underlying deficit of dyslexia but known to influence literacy (e.g., orthographic compensation, see below; van der Leij & van Daal, 1999). These compensatory mechanisms could be used to circumvent an underlying deficit or subdue its negative effect and thereby improve literacy performance (see also van Viersen et al., 2015). Unfortunately, both accounts are hard to separate based on descriptive data in the NL. As we will argue below, investigating both dyslexic groups on their FL skills may provide support for the compensation account and disentangle it from the core-deficit account.

A comparison between the FL literacy outcomes of gifted students with dyslexia and averagely intelligent students with dyslexia could yield similar findings as in the NL. According to the Linguistic Coding Differences (LCD) hypothesis (Sparks & Ganschow, 1991, 1993, 1995) NL skills are fundamental for the acquisition of an FL. A large body of empirical research found strong associations between NL and FL literacy performance (e.g., Morfidi, van der Leij, de Jong, Scheltinga, & Bekebrede, 2007; Sparks, Patton, Ganschow, Humbach, & Javorsky, 2006). Several studies have demonstrated that this association also holds for students with dyslexia (e.g., Crombie, 1997; Morfidi et al., 2007). For example, Crombie (1997) showed that secondary school students with dyslexia show lower performance than nondyslexic students in both NL and FL reading and writing, as well as in FL speaking and listening. Morfidi et al. (2007) also found lower FL performance for poor readers compared to average readers, specifically in word reading and orthographic knowledge. Moreover, they showed that NL word reading is the strongest predictor of FL word reading performance after controlling for age, vocabulary, and reading group (Morfidi et al., 2007). Accordingly, the LCD hypothesis provides a logic behind the difficulties that students with dyslexia experience in FLs.

However, it is uncertain whether the LCD hypothesis could also explain the differences in literacy level between dyslexic students with and without giftedness, especially when considering their different underlying cognitive profiles (Berninger & Abbot, 2013; van Viersen et al., 2014). The main assumption of the LCD hypothesis is that the association between NL and FL performance is strong because the same underlying factors are involved in the NL and FL (e.g., Sparks, 1995). Indeed, several studies have shown that underlying cognitive skills, such as word decoding-related phonological and orthographic processing, but also vocabulary and grammar, contribute to both NL and FL learning (e.g., Dufva & Voeten, 1999; Kahn-Horwitz, Shimron, & Sparks, 2006; Lindsey, Manis, & Bailey, 2003; Melby-Lervåg & Lervåg, 2011; Morfidi et al., 2007; see Koda, 2005, for an overview). Moreover, difficulties in NL phonology were found to surface in FL phonology (Crombie, 1997; Morfidi et al., 2007), indicating cross-linguistic transfer of underlying deficits associated with dyslexia. Evidence for possible transfer of strengths in cognitive factors is limited though (but see Lindsey et al., 2000; Sparks, Patton, Ganschow, & Humbach, 2012, for some suggestions). It is thus unclear to what extent both underlying risk and protective factors of the NL overlap with factors involved in the FL and whether they are responsible for differences in literacy levels between gifted and averagely intelligent students with dyslexia.

Based on the LCD hypothesis, controlling for NL literacy skills should eliminate differences in FL literacy level between gifted and averagely intelligent students with dyslexia, as these differences are assumed to have their origin in the NL. However, several studies have indicated that (differences in) NL literacy skills do not always translate to FL literacy. Some students with dyslexia were found to read better in an FL than expected based on their NL, especially when the FL is English (e.g., Bekebrede, van der Leij, & Share, 2009; Miller-Guron & Lundberg, 2000; van der Leij & Morfidi, 2006). This better than expected performance is thought to result from a different type of reading process and has been referred to in terms of compensation. For example, higher literacy performance in the FL of students with dyslexia could be the result of an alternative reading strategy, in which the focus is on sight-word storage or larger orthographic units instead of grapheme-phoneme decoding (Bekebrede et al., 2009). Such a reading strategy fosters both reading accuracy and fluency. It is evoked by the greater irregularity of English as an opaque language, in which phonological decoding may be less effective for reading (Seymour, Aro, & Erskine, 2003; Ziegler & Goswami, 2005). Effective use of this alternative strategy, also called orthographic compensation (van der Leij & van Daal, 1999), is thought to be driven by the amount of exposure to print, which is generally larger for English than other foreign languages (see Rack, Snowling, & Olson, 1992; Stanovich & West, 1989). Bekebrede et al. (2009) have shown that secondary school students with dyslexia with higher levels of NL and FL orthographic knowledge show higher English reading performance as well as higher English spelling levels compared to dyslexic students with low orthographic competence. These results suggest that group differences in FL literacy performance would be caused by additional factors, unique to the specific FL, that alter the reading process, resulting in higher literacy levels. Accordingly, differences between gifted and averagely intelligent students with dyslexia may remain present after controlling for NL literacy. However, the results of the above-mentioned studies cannot be directly generalized to other FLs than English.

The influence of NL skills on FL performance may vary between FLs based on language-specific characteristics, such as transparency or exposure (see also Geva & Siegel, 2000). Orthographic transparency refers to (the complexity of) the letter-sound correspondences within a language, which can be consistent (e.g., German and Dutch) or (very) inconsistent (English and French; e.g., Seymour et al., 2003). As outlined above, the high orthographic complexity of English may increase the effect of processes associated with orthographic compensation. Yet, the benefit of advanced orthographic knowledge may be smaller in, for example, French than English or even absent in German. For the latter language, grapheme-phoneme decoding is straightforward and thereby less error prone than in languages that have inconsistent grapheme-phoneme correspondences. In addition, exposure to print (e.g., Brown, Waring, & Donkaewbua, 2008; Mol & Bus, 2011) and to spoken language (e.g., Bisson, van Heuven, Conklin, & Tunney, 2013) have been found to foster FL acquisition, in particular vocabulary. Knowledge of the pronunciation and meaning of words is especially important for orthographic learning in languages with many irregular words (Wang, Nickels, Nation, & Castles, 2013), such as English and French. Considering the dominance of English in today's increasingly multilingual society, both types of exposure may be much more intensive for English than for French and German. Variation in the amount of exposure may thus partly determine the extent to which students have the opportunity to develop compensatory mechanisms based on literacy-related strengths, such as the orthographic compensation described above, or may have to rely on their NL skills. Therefore, differences between FLs in the influence of NL skills on FL literacy can be expected.

CURRENT STUDY

In the present study, we compared four groups of secondary school students with and without giftedness and/or dyslexia, which resulted from the cross-classification of giftedness (yes vs. no) and dyslexia (yes vs. no). Giftedness was defined as academic giftedness or high intelligence (e.g., Winner, 1997), whereas dyslexia was defined as severe and persistent literacy difficulties (e.g., Snowling, 2000). Although the main focus is on the difference in literacy level between both dyslexic groups, typically developing (TD) and gifted groups were also included as a reference for (above) average literacy skills and to provide a more complete overview of group differences in FL performance. First, we examined whether a stepwise pattern of group differences previously found in NL reading and spelling performance (i.e., dyslexia < gifted/dyslexia < TD < gifted) is also attested in FLs. If the pattern is replicated, the second question pertains to whether the group differences, particularly between both dyslexic groups, can be accounted for by the difference in NL literacy level. Controlling for NL skills, thereby also cancelling out NL subskills and compensatory mechanisms, can reveal if differences in literacy levels have their origin in the NL, as suggested by the LCD hypothesis, or may involve factors or processes that are unique to the (specific) FL (e.g., Bekebrede et al., 2009; Morfidi et al., 2007). If NL skills cannot account for the pattern of FL performance, this would indicate that unique factors or processes are responsible for the higher literacy performance of gifted students with dyslexia in the specific FL, providing evidence for the existence of compensation.

In this study, all students had Dutch as their NL. English, French, and German were the FLs under investigation. For all three FLs, we expected to replicate the stepwise pattern previously found in the NL (as described above). Several additional hypotheses were formulated with respect to the different FLs. Specifically, the degree to which this pattern can be accounted for by NL skills was expected to differ between FLs. For English, the language with least orthographic transparency and most exposure, we hypothesized that the stepwise pattern would remain intact after controlling for NL literacy. For French, a less opaque language than English but with generally less exposure, we also hypothesized a stepwise pattern of group differences to remain intact, but possibly less clear than for English. German, however, is a more transparent language than Dutch and comparable to French in terms of exposure. We hypothesized that the great similarity to Dutch would either facilitate or hamper FL performance, which may result in comparable FL levels between both dyslexic groups.

METHOD

PARTICIPANTS

The total sample consisted of 148 Dutch students in secondary education. Parents and students were recruited through calls on the websites of educational magazines, blogs, and contacts with school teachers and clinicians. Written informed consent was obtained from all parents and students. It is important to note that Dutch secondary education is highly tracked (see SBB, 2015). After 6th grade, students continue education at the level befitting their prior development in primary education. A rough distinction can be made between pre-vocational education (vmbo; 4 years), higher pre-vocational education (havo; 5 years), and pre-university education (vwo; 6 years), of which the upper and lower tracks can also be divided into hierarchical sub-tracks. Generally, students start secondary education with two years in a combined track class (e.g., havo/vwo) and then continue with the track that fits best. As the level and amount of instruction may differ between tracks and students came from a variety of (sub-)tracks to cover all four research groups (see Table 1), this needs to be controlled for in the analyses (see below).

The sample was divided into groups based on definitions of giftedness and dyslexia. For giftedness, the cut-off value was set at a full IQ score >120 or a 95% reliability interval around a score of 125 (i.e., 116-131) in case of a short form. For dyslexia, students had to show 1) reading scores below the 10th percentile (standard score ≤ 6), or 2) reading scores below the 15th percentile (standard score ≤ 7) *and* spelling scores below the 10th percentile (stanine ≤ 2), which is in line with Dutch protocols for diagnosing dyslexia (e.g., Kleijnen, Bosman et al., 2008). Students who fulfilled one of these two criteria for dyslexia, but also had an above average score for one or more of the reading and/or spelling tasks, were excluded. Of all students with an official dyslexia diagnosis, 22 gifted students were excluded from further analyses in this study because their reading and/or spelling levels no longer fell below the diagnostic threshold for dyslexia described above. TD students had to show reading and spelling scores above the 25th percentile. This resulted in four research groups covering students with dyslexia (n = 32), gifted students with dyslexia (n = 19), typically developing students (n = 39), and gifted students (n = 36). All students within both dyslexia groups had below average scores (standard score < 8) on at least one out of three measures for phonological processing (i.e., phonological awareness, rapid automatized naming, or verbal short-term memory).

It is important to note that in The Netherlands it is common for children to grow up learning one language during the first years of primary school. FL education (mostly English) has recently been introduced at the end of primary education (Grades 5 and 6). At the time of data collection, it was only a mandatory part of the curriculum from the start of secondary education (Grade 7 onward). All students start with two FLs in 7th grade (i.e., English and either French or German). A third language is added in 8th grade. Therefore, data were collected at two points during secondary education. The first measurement took place when students were in 7th or 8th grade and concerned a broad screening of NL and FL literacy and literacy-related skills. For literacy, the focus was on Dutch and English, because most students had a sufficient basis in both languages. The second measurement comprised an additional assessment of a subsample of 71 students one year later (see Procedure), when they were in 8th or 9th grade. These students were selected based on willingness to participate and kept the group classification of their initial assessment one year earlier. Besides reassessment of Dutch and English literacy, the main focus was on French and German literacy, in which most students received at least several months of formal instruction by this time. Background characteristics of the samples at both occasions are displayed in Table 1.

TABLE 1

Background Characteristics of the Four Groups at the Two Time Points

Measure	T1 D	T1 GD	T1 TD	T1 G	T2 D	T2 GD	T2 TD	T2 G
n	32	19	39	36	16	15	16	24
% Boys	40.6	78.9	43.6	47.2	25.0	73.3	56.3	62.5
% Dyslexia[a]	68.8	100.0	0.0	0.0	81.3	100.0	0.0	0.0
% Gifted[a]	0.0	63.2	0.0	36.1	0.0	53.3	0.0	37.5
Age (months)	154.91_{a}	156.42_{a}	157.10_{a}	154.28_{a}	168.81_{a}	170.13_{a}	170.81_{a}	167.42_{a}
	(7.99)	(7.16)	(7.03)	(9.11)	(8.48)	(7.14)	(6.58)	(8.91)
Level of	5.47_{a}	6.89_{b}	6.31_{ab}	8.11_{c}	5.19_{a}	6.53_{a}	6.06_{a}	8.50_{b}
education[b]	(1.83)	(1.49)	(2.27)	(2.04)	(1.80)	(2.07)	(2.14)	(1.98)
IQ (total)	102.91_{a}	129.63_{b}	106.23_{a}	132.89_{b}	102.00_{a}	130.53_{b}	106.25_{a}	133.75_{b}
	(8.82)	(7.71)	(8.99)	(8.92)	(9.75)	(6.71)	(9.23)	(8.80)
Dutch	5.28_{a}	6.37_{a}	11.67_{b}	12.81_{b}	6.50_{a}	7.80_{a}	11.12_{b}	12.88_{b}
word reading[c]	(3.14)	(3.10)	(2.20)	(2.45)	(4.23)	(3.26)	(1.71)	(2.25)
Dutch	5.36_{a}	5.26_{a}	11.38_{b}	12.56_{c}	-	-	-	-
nonword reading[c]	(2.25)	(2.64)	(2.55)	(9.27)				
Dutch spelling[d]	1.69_{a}	2.68_{b}	5.21_{c}	6.78_{d}	-	-	-	-
	(1.00)	(1.86)	(1.70)	(1.87)				

Note. The group means are given with the standard deviations in parentheses. Means in the same row per time point that do not share subscripts differ at $p < .05$. T = time point; D = dyslexia; GD = gifted + dyslexia; TD = typically developing; G = gifted.

[a] Official diagnosis provided by a professional. [b] On a scale from 1 to 10, lowest track (basic pre-vocational education) representing 1, highest track (bilingual pre-university education) representing 10. [c] Standard score with a mean of 10 and standard deviation of 3. [d] Stanine score on a scale from 1 to 9.

INSTRUMENTS

INTELLIGENCE. The general cognitive abilities of the students were assessed using the Dutch version (Kort et al., 2005) of the *Wechsler Intelligence Scale for Children – Third Edition* (WISC-III; Wechsler, 1991). If recent test results were available (i.e., not older than two years), students were not reassessed. If not, a short form was used, consisting of two verbal subtests (i.e., vocabulary and similarities) and two performance subtests (i.e., block design and picture arrangement). Total IQ scores could be computed based on the sum of the standardized subtest scores (see Kaufman, Kaufman, Balgopal, & McLean, 1996). Reliability and validity quotients of the short form are all greater than .83 (Kaufman et al., 1996).

WORD READING. Timed word reading ability was assessed in four languages. For Dutch, the *Eén Minuut Test* (EMT; Brus & Voeten, 1999) was used. The *TOWRE* sight word efficiency subtest (Torgesen, Wagner, & Rashotte, 1999) was used for English. The French task was part of a screening tool for dyslexia in secondary education (Kleijnen, Steenbeek-Planting, & Verhoeven, 2008). For German, an experimental task was composed from a large list of words that are generally taught in the first two grades of secondary education. Word length increased from one to four syllables on all tasks. The student had one minute to accurately read aloud as many words as possible per task. Maximum scores are 116, 108, 100, and 100, respectively. On all tasks, raw scores were the number of correctly read words within the allotted time. Internal consistency of the Dutch task is .90 (Evers et al., 2009-2012). For the English task it ranged between .93 and .96 (Torgesen et al., 1999).

In Dutch, timed nonword reading was measured using *Klepel* (van den Bos, lutje Spelberg, Scheepstra, & de Vries, 1994). The student had two minutes to accurately read as many nonwords as possible. Here, word length also increased from one to four syllables. Raw scores were the number of correctly read nonwords, with a maximum of 116 words. Internal consistency is .92 (Evers et al., 2009-2012). The Dutch word reading and nonword reading tasks were used for the inclusion criteria of dyslexia, for which raw scores were transformed into norm-referenced standard scores (M = 10, SD = 3).

ORTHOGRAPHIC KNOWLEDGE. Production of orthographic knowledge in Dutch and English was assessed using two spelling measures. For Dutch, a normed sentence dictation from the dyslexia protocol for secondary education (Henneman & Kleijnen, 2005) was used. This spelling test, which is about the Dutch weather, consists of 10 sentences of increasing length and difficulty. The total number of correctly spelled words was the raw score, which was used in the analyses. Raw scores were transformed into norm-referenced stanine scores for application of the criteria for dyslexia. For English, an experimental spelling task was used (S. van Viersen & E. H. de Bree, personal communication, October 2014). The task consisted of three blocks of 10 sentences that provided context to the target words. Children had to write down the target word after the sentence was read. The target words were selected from the listed words in the English word reading task. The raw score was the total number of correctly spelled words.

In English, French, and German, orthographic knowledge was (also) assessed using orthographic choice tasks, which were aimed at recognition instead of production. For French and German this type of task was used as an alternative to a dictation, which was thought to be too difficult at this stage of development for most students. The English orthographic choice task (Olsen, Forsberg, Wise, & Rack, 1994) was used as a model for developing the experimental French and German tasks, for which items were composed by selecting words from the respective word reading tasks. Students received a list of 40 items (one list per language) consisting of correctly spelled meaningful words and incorrectly spelled words with the same pronunciation (e.g., English: wurd - word). Students had to recognize and select the word with the correct spelling. Word length increased from one to four syllables. The raw score was the total number of correctly recognized words, which was used in the analyses. Internal consistency (Cronbach's α) of the English task was .78 (Bekebrede et al., 2009).

PROCEDURE

Assessments were conducted by trained and supervised undergraduate and graduate students. Their training comprised an intensive instructional session about the test battery and a test session with a student, on which they received extensive feedback. Students were first tested in 7th or 8th grade, during one two-to-three-hour session (with ample breaks provided) at home, school, or in a clinic. The first assessment covered intelligence as well as all tasks for Dutch and English language reported in this study. One year later (in 8th or 9th grade), a subsample was assessed again during a 30-to-40-minute session covering also the French and German tasks. During both assessments, tasks were always presented in a fixed order. For the order of the languages, the least familiar languages were assessed first (i.e., T2: French and German) as they might take more effort, followed by the more familiar languages (i.e., T1 and T2: English and Dutch). Languages were always separated by intermediate tasks to prevent interference between languages. Furthermore, within languages the order of the tasks was also fixed (i.e., spelling, orthographic choice, word reading) to minimize carry over effects across tasks targeting the same language.

ANALYSES

Bayesian analyses were used to compare groups on their word reading and orthographic knowledge in the four languages. This approach was favored over more traditional methods for several reasons. First, Bayesian model selection (i.e., the Bayesian alternative to traditional frequentist ANOVA) allows for the testing of equality or inequality constrained informative hypotheses, which can be formulated using prior knowledge (Klugkist, Laudy, & Hoijtink, 2005). It is also possible to compare several competing informative hypotheses that represent opposing views or specific expectations. Second, it is suitable for dealing with non-normal data, since Bayesian analyses are not based on normality or asymptotic assumptions (Gill, 2008). As such, it is also a good alternative when having to work with small samples or unequal group sizes. Third, these specific analyses do not involve traditional *p*-values, thereby avoiding multiple testing problems such as alpha inflation or loss of power when adopting a stricter alpha level (Klugkist, van Wesel, & Bullens, 2011).

In short, the hypothesized differences between group means on word reading and orthographic knowledge were translated into equality and inequality constrained statistical hypotheses (Table 2; see also van Viersen et al., 2014). These informative hypotheses were each compared to the alternative hypothesis (i.e., empty model without constraints). Each comparison results in a Bayes factor (BF), representing the amount of evidence in favor of one hypothesis compared to another (Kass & Raftery, 1995). BFs >1 indicate support for the informative hypothesis, whereas BFs <1 indicate support for the alternative hypothesis. The BF can also be interpreted as a measure of effect size (i.e., BF 1-3 = small, BF 3-10 = medium, BF >10 = large; Kass & Raftery, 1995). Competing hypotheses can be compared based on the posterior model probability (PMP), representing the relative support for a specific hypothesis within a set of hypotheses (Klugkist et al., 2011). In general, a difference between PMPs that is smaller than .05 cannot be interpreted and results in equal support for both models. A more detailed description of Bayesian model selection within this field of research is provided by van Viersen et al. (2015).

TABLE 2

Statistical Hypotheses Bayesian Model Selection

Hypothesis	Model	Statistical notation
Alternative	Model 0	$\mu_D, \mu_{GD}, \mu_{TD}, \mu_G$
Informative 1	Model 1	$\mu_D = \mu_{GD} < \mu_{TD} < \mu_G$
Informative 2	Model 2	$\mu_D < \mu_{GD} < \mu_{TD} < \mu_G$

Note. μ represents the group mean. D = dyslexia; GD = gifted + dyslexia; TD = typically developing; G = gifted.

Several covariates were used in the analyses. Group differences were controlled for two types of exposure, i.e., educational level (track, as a proxy for level of instruction) and language-specific didactical age (quantity, number of months a student has received formal education in a specific FL, with a maximum of 10 months per year), at both time points. Controlling for NL skills in the second part of the analyses was done by including Dutch word reading or spelling as (separate) covariates for word reading and orthographic knowledge in English, French, and German. The analyses were conducted using the BIEMS software package (Mulder, Hoijtink, & Klugkist, 2010; Mulder, Hoijtink, & de Leeuw, 2012; Mulder et al., 2009).

RESULTS

Missing data analysis revealed no missing data points. An outlier analysis using z-scores revealed two outliers (*z*-score < -3.29) on two different dependent variables. These outliers were corrected to the closest raw score that corresponded to a *z*-score > -3.29. Checking the assumptions for ANCOVA revealed that four dependent variables (i.e., Dutch and English spelling at T1 and French and German orthographic knowledge at T2) and all covariates were not normally distributed. Hence, Bayesian model selection was considered an appropriate alternative to compare groups.

NL PROFILE

Patterns of group differences were first investigated in the NL to establish that the stepwise pattern previously found in primary education (van Viersen et al., 2014) is also attested in early secondary education (i.e., T1). Table 3 displays the posterior means and standard deviations of the four groups and the BFs and PMPs for the three models under investigation. The results show that for both word reading and spelling Model 2 received most support from the data (PMPs .54 and .88), with BFs indicating strong effects. The posterior means of the unconstrained model indeed indicate a stepwise pattern, as gifted students with dyslexia scored in between students with dyslexia and TD students, and gifted students outperformed all groups. Dutch word reading patterns of group differences are comparable across both occasions of measurements (see Table 3).

FL PROFILES

The group comparisons on FL reading and orthographic knowledge show considerable differences between languages and literacy skills (see Table 3). The pattern for English is largely the same as for Dutch. The BF of Model 2 indicates that the model received about 23 times more support from the data than the alternative model, indicating strong effects for both word reading and spelling (PMPs .80 and .89). Yet, for orthographic choice Model 1 received most support from the data (PMP = .56), also indicating a large effect. The posterior means confirm that students with dyslexia and gifted students with dyslexia obtained about equal orthographic choice scores. The results also indicate comparable patterns of group differences for English word reading at the two measurement occasions.

For word reading in French Model 1 and 2 received a comparable amount of support from the data. This was about 14 times more than the alternative model, which can be considered a large effect. Based on the PMPs (.47 and .50) neither of the models can be preferred over the other. For French orthographic choice, Model 1 received most support from the data, also about 14 times more than for the alternative model. Overall, the posterior means indicate there are no clearly identifiable differences between students with dyslexia and gifted students with dyslexia in French reading and orthographic knowledge.

The pattern for German word reading is about the same as for French word reading. Model 1 and 2 both received about 13 times more support from the data than the alternative model (PMPs .47 and .49). There is no clear difference between averagely intelligent students with dyslexia and gifted students with dyslexia. However, TD students still obtained higher scores and gifted students outperformed all groups. For orthographic choice, the pattern is not in line with the expectations, as the alternative model received most support from the data (PMP = .66). The posterior means show that the students with dyslexia, the TD students and the gifted students all showed comparable performance, but the gifted students with dyslexia obtained considerably lower scores (Table 3).

Table 3

Posterior Means (PM) and Standard Deviations (PSD) of the Literacy Measures for the Four Languages Adjusted for Educational Level and FL-specific Didactical Age and Bayes Factors (BF) and Posterior Model Probabilities (PMP) of the Three Models under Investigation

	Dyslexia		Gifted + Dyslexia		Typically Developing		Gifted		Model 0		Model 1		Model 2	
Skill	PM	PSD	PM	PSD	PM	PSD	PM	PSD	BF	PMP	BF	PMP	BF	PMP
T1														
Dutch														
Word reading	63.04	4.33	66.23	6.48	88.49	3.28	90.95	3.99	1.00	.03	13.21	.43	**16.54**	**.54**
Spelling	149.91	1.95	154.79	2.96	164.70	1.50	167.29	1.87	1.00	.04	1.95	.08	**21.52**	**.88**
English														
Word reading	51.03	2.66	55.49	3.75	63.16	1.96	68.36	2.38	1.00	.03	4.63	.16	**23.15**	**.80**
Orthographic choice	33.42	0.23	33.65	0.33	35.54	0.17	37.18	0.21	1.00	.03	**19.70**	**.56**	14.58	.41
Spelling	13.31	0.45	15.66	0.63	18.20	0.33	20.45	0.40	1.00	.04	1.84	.07	**22.84**	**.89**
T2														
Dutch														
Word reading	69.87	11.36	74.03	10.56	86.31	10.17	94.20	8.37	1.00	.03	11.72	.37	**19.18**	**.60**
English														
Word reading	58.26	6.11	63.65	5.58	71.03	5.39	78.36	4.43	1.00	.03	5.18	.18	**22.65**	**.79**
French														
Word reading	26.65	10.94	28.74	10.40	37.37	9.85	43.21	8.18	1.00	.03	**13.61**	**.47**	**14.54**	**.50**
Orthographic choice	33.33	0.49	33.22	0.47	34.51	0.45	36.70	0.37	1.00	.04	**14.79**	**.59**	9.35	.37
German														
Word reading	29.61	7.10	31.19	6.92	40.29	6.39	43.75	5.31	1.00	.04	**12.93**	**.47**	**13.46**	**.49**
Orthographic choice	26.88	1.04	23.38	1.04	26.56	0.95	27.18	0.78	**1.00**	**.66**	0.42	.28	0.10	.07

Note. Bold values indicate BFs and PMPs of models that received most support from the data. T = time point.

FL PROFILES WHEN CONTROLLING FOR NL

To gain more insight in the influence of NL skills on FL performance, the group differences in English, French, and German reading, spelling, and orthographic knowledge were controlled for word reading and spelling proficiency in Dutch. Overall, controlling for NL skills resulted in smaller effect sizes and less clear group differences (see Table 4) – indicating an influence of NL on FL performance. Yet, the stepwise pattern for the English measures remained intact (both at T1 and T2), indicating that the higher performance of gifted students with dyslexia is the result of factors/mechanisms that are unique to English as an FL. Model 2 received most support from the data for both word reading and spelling (PMPs .54 and .78). The posterior means indicate that the word reading ability of the gifted students with dyslexia moved more toward that of the TD students (even slightly above), while students with dyslexia still had the lowest and gifted students the highest scores. This shift in pattern is illustrated by the considerably lower, but still medium-sized, BF for word reading. The same trend was visible for group differences in spelling. The difference between the gifted students with dyslexia and TD students has become notably smaller. However, gifted students with dyslexia did not close the gap entirely, which kept the stepwise pattern intact as indicated by the larger effect size for English spelling than for word reading. For English orthographic choice the results were unchanged. Model 1 still received most support from the data (PMP = .64). The posterior means show the same pattern, indicating about equal performance for students with dyslexia and gifted students with dyslexia.

For French and German word reading, controlling for NL word reading ability also resulted in smaller effect sizes. The posterior means indicate that the differences in French and German word reading between students with dyslexia, gifted students with dyslexia, and TD students became notably smaller, but the results are more equivocal. In both cases Model 1 received most support from the data and is now clearly favored over Model 2 (PMPs both .55). Model 1 also still received most support from the data for French orthographic choice (PMP = .64). However, the posterior means indicate that the difference in performance between students with dyslexia and gifted students with dyslexia increased, which is illustrated by the smaller effect size, as students with dyslexia and TD students showed comparable performance. The results for German orthographic choice also largely remained the same. The alternative model continued to receive most support from the data (PMP = .76) and the posterior means confirm the unexpected pattern of gifted students with dyslexia showing lowest performance of all groups.

TABLE 4

Posterior Means (PM) and Standard Deviations (PSD) of the Literacy Measures for the Three Foreign Languages Adjusted for Educational Level, FL-specific Didactical Age, and Dutch Reading or Spelling and Bayes Factors (BF) and Posterior Model Probabilities (PMP) of the Three Models under Investigation

	Dyslexia		Gifted + Dyslexia		Typically Developing		Gifted		Model 0		Model 1		Model 2	
Skill	PM	PSD	PM	PSD	PM	PSD	PM	PSD	BF	PMP	BF	PMP	BF	PMP
T1														
English														
Word reading[a]	57.70	2.65	60.77	3.19	59.50	1.69	63.61	2.08	1.00	.10	3.72	.36	**5.54**	**.54**
Orthographic choice[b]	34.27	0.29	34.16	0.33	35.18	0.17	36.55	0.23	1.00	.04	**16.33**	**.62**	8.83	.34
Spelling[b]	14.80	0.55	16.52	0.62	17.56	0.33	19.35	0.43	1.00	.04	4.38	.18	**19.21**	**.78**
T2														
English														
Word reading[a]	64.27	4.90	67.44	4.18	69.44	3.81	73.04	3.70	1.00	.05	6.22	.29	**14.13**	**.66**
French														
Word reading[a]	32.77	10.84	33.64	9.86	34.63	8.63	37.94	8.31	1.00	.08	**6.82**	**.55**	4.62	.37
Orthographic choice[b]	34.29	0.65	33.60	0.46	34.22	0.44	36.03	0.43	1.00	.09	**7.13**	**.64**	3.09	.28
German														
Word reading[a]	36.30	5.75	36.35	5.31	36.91	4.57	38.24	4.21	1.00	.11	**4.80**	**.55**	2.98	.34
Orthographic choice[b]	27.59	1.48	23.62	1.08	26.30	1.00	26.75	0.95	**1.00**	**.76**	0.26	.20	0.06	.04

Note. Bold values indicate BFs and PMPs of models that received most support from the data. T = time point.
[a] Adjusted for educational level, language-specific didactical age and Dutch word reading. [b] Adjusted for educational level, language-specific didactical age and Dutch spelling.

DISCUSSION

Studies into native language (NL) skills have shown that gifted children with dyslexia outperform averagely intelligent children with dyslexia on reading and spelling tasks (e.g., Berninger & Abbott, 2013; van Viersen et al., 2014). In this study, we examined group differences between secondary school students with and without giftedness/dyslexia in foreign language (FL) reading and spelling. The first aim was to assess whether, as in the NL, the level of FL literacy of gifted students with dyslexia is higher than the literacy level of averagely intelligent students with dyslexia. On the assumption that this pattern was found, the second aim was to determine whether this difference could be accounted for by the difference in literacy level in their NL.

As a start, groups were compared on their NL literacy skills. Research in primary education has shown a stepwise pattern of group differences in NL literacy (i.e., dyslexia < gifted/dyslexia < typically developing (TD) < gifted; van Viersen et al., 2014). Finding this pattern of differences in the NL in secondary education is a precondition for the interpretation of differences in FL literacy levels. As expected, secondary school age gifted students with dyslexia were found to have higher NL word reading and spelling levels than their averagely intelligent peers with dyslexia and a full stepwise pattern of performance for NL word reading and spelling skills was attested. This result is in line with previous findings (van Viersen et al., 2014) and with the generally high stability of word reading fluency and spelling skills over time in transparent languages (e.g., Landerl & Wimmer, 2008).

The findings for FL literacy indicate that the stepwise pattern of group differences generalizes to English word reading and spelling. To test whether these group differences can be accounted for by differences in NL skills, we controlled for NL literacy, aiming to cancel out the effect of NL literacy-related subskills and NL compensatory mechanisms. As a result, effect sizes for patterns of group differences in English word reading and spelling decreased, indicating that NL skills partly influence FL performance. Nonetheless, the stepwise pattern remained present. For both word reading and spelling, the performance level of the gifted students with dyslexia approximated that of the TD students while students with dyslexia showed continuously low performance. Consequently, the higher performance of gifted students with dyslexia compared to their averagely intelligent dyslexic peers results from factors or processes that are specific to English as an FL. These findings provide support for the compensation account and are in line with studies showing that additional, language-specific factors can be responsible for higher FL literacy levels than expected based on the NL (e.g., Bekebrede et al., 2009; Morfidi et al., 2007).

These results for English literacy are interpreted in terms of compensation. The differences in literacy level between gifted and averagely intelligent students with dyslexia cannot result from less severe underlying deficits and/or higher literacy-related skills; controlling for NL literacy (and the influence of related subskills) should have cancelled out underlying group differences in the FL as well. The only way in which the higher literacy levels of the gifted students with dyslexia cannot be explained in terms of compensation is if reading and spelling in English would require an *additional* subskill. This subskill would then have to be generally impaired in students with dyslexia, but less impaired in the gifted/dyslexic students. This is unlikely, however, because multiple studies have shown that all alphabetic languages involve largely the same set of literacy-related subskills (e.g., Caravolas et al., 2012; Melby-Lervåg & Lervåg, 2011; Ziegler et al., 2010). Therefore, the current findings are considered to be in favor of the compensation account.

The stepwise pattern of group differences in NL literacy level did not generalize to French and German literacy. Gifted students with dyslexia and averagely intelligent students with dyslexia showed similar performance on the French and German literacy tasks. This did not change after controlling for NL skills. For orthographic knowledge, the absence of a stepwise pattern in French and German might be due to features of the orthographic choice task. This task did not reveal a stepwise pattern in the performance of the groups in English either. However, gifted and averagely intelligent students with dyslexia showed no differences in performance on the French and German reading tasks, while the former group clearly outperformed the latter in Dutch and English word reading. Generally, the findings for French and German as FLs align with the LCD hypothesis (Sparks & Ganschow, 1991, 1993, 1995), which proposes a strong association between the NL and FL.

There are two possible explanations for the fact that patterns of group differences in NL literacy level only generalized to English as an FL and not to French and German. The first lies in differences between the FLs in orthographic depth. German and Dutch can be considered (semi-)transparent languages whereas French is often assumed to be opaque and English even extremely opaque (Seymour et al., 2003). However, quantification of orthographic depth using entropy measures (Borgwald, Hellwig, & de Groot, 2005) indicates that French (0.46) is actually much closer to Dutch (0.23) in orthographic transparency than to English (0.83;

Ziegler et al., 2010). For gifted students with dyslexia, both German (as hypothesized) and French may thus not be orthographically complex enough to evoke the development of mechanisms, as for example the processing of larger letter clusters, that help to compensate their underlying deficits and improve literacy performance (e.g., see Bekebrede et al. 2009; Morfidi et al., 2007).

A second explanation pertains to differences between the FLs in terms of exposure. In the Netherlands, both German and French are FLs that depend primarily on in-class instruction, whereas English is omnipresent in Dutch society. Although we have no data about the students' amount of exposure to these FLs outside of the classroom, we consider it very likely that secondary school students learn to understand and use English in daily life, in contrast to French and German. This happens, for example, while watching international television shows (with Dutch subtitles), listening to popular music, or playing online computer games (see also van der Leij, Bekebrede, & Kotterink, 2010). As such, students are provided with less opportunities to develop compensatory mechanisms in French and German than in English, whereas for English children can benefit from input through multiple modalities as well as a broader language environment (see also Sparks et al., 2012).

The results of the current study have several implications. First, as it is clear that literacy outcomes may vary depending on the (characteristics of the) specific FL, future research should focus on a wider range of FLs. Also, more research on FLs that vary in the required and/or provided amount of instruction is necessary. Our findings suggest that the development of compensatory mechanisms may (partly) hinge on a combination between the orthographic depth and the amount and type of exposure of an FL. Nevertheless, more research is necessary to determine what general and language-specific factors (e.g., motivation, immersion) may influence success in FL learning. As English as an FL has a clear advantage over French and German in terms of exposure for Dutch children, it is important to consider both factors that lie in the broader language environment and lower-level (e.g., phonological processing) as well as higher-level (e.g., vocabulary, grammar) cognitive factors (Koda, 1992, 2005; Sparks & Miller, 2000). Moreover, gaining more insight in how compensatory mechanisms or strategies may influence (FL) literacy acquisition requires more advanced methods, such as eye-tracking.

Overall, this study has shown that Dutch gifted secondary school age students with dyslexia have higher NL literacy levels than their averagely intelligent dyslexic peers. As in the NL, a stepwise pattern of group differences, i.e., dyslexia < gifted/dyslexia < TD < gifted, was found for English word reading and spelling. This pattern was not found for French and German literacy performance. The higher English literacy level of gifted students with dyslexia as compared to averagely intelligent students with dyslexia results from factors or mechanisms that are unique to English as an FL. The findings suggest that gifted students with dyslexia can partly compensate for their NL dyslexia in English FL learning.

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CHAPTER 7

GENERAL DISCUSSION

In this dissertation several issues were addressed regarding current knowledge about dyslexia, specifically in the area of risk and protective factors and possibilities for compensation. Risk factors were assessed at various stages during development. The main focus was on the role of early oral language and the influence of having a family risk (FR) of dyslexia (Chapter 2 and 3). In addition, the potential impact of protective factors on literacy development was investigated in a population of gifted children with dyslexia (Chapter 4, 5, and 6). Findings are discussed and connected below to add to theories on dyslexia and inform diagnostic practice.

RISK FACTORS

EARLY LANGUAGE

The studies reported in Chapter 2 and 3 focused on early language as a risk factor for dyslexia and the influence of having an FR. In Chapter 2 we investigated the early stages of language acquisition, i.e., very early development of receptive and expressive vocabulary size. Chapter 3 builds on this as early oral language was assessed, though not *very* early oral language (thus picking up where we left in Chapter 2), and its longitudinal effect throughout literacy development. In both studies, children's FR status was considered. In Chapter 2 a further group distinction was made by dividing the FR group in children who do (FR-dyslexic) and do not (FR-nondyslexic) become dyslexic. This allowed us to determine the potential of early language as a possible risk factor as well as its association with FR or later literacy outcomes (i.e., dyslexia).

A novel element of the study in Chapter 2 was that we used the *same* instrument at each point in development to measure receptive and expressive vocabulary during infancy (between 17 and 35 months). Thereby, in contrast to previous studies, we were able to assess differences in growth trajectories of receptive and expressive vocabulary between FR children with and without dyslexia and typically developing (TD) children without an FR.

This set-up showed that, compared to FR-nondyslexic and TD children, FR-dyslexic children had a smaller expressive vocabulary size already from 17 months onward and a smaller receptive vocabulary size from 23 months onward. There were no differences in receptive and expressive vocabulary size between FR-nondyslexic and TD children. The group differences between dyslexic and nondyslexic children persisted over time as FR-dyslexic children showed continuously low performance up to 35 months. However, differences were small. Moreover, the majority of FR-dyslexic children had no clinically relevant vocabulary deficits, but only mild ones.

Modeling growth trajectories revealed that FR-dyslexic children show delayed receptive and expressive vocabulary development compared to both nondyslexic groups. Across groups, receptive vocabulary growth trajectories were curvilinear and characterized by a gradual increase and subsequent deceleration of growth, whereas expressive vocabulary growth trajectories were S-shaped and characterized by an acceleration in growth during the second year of life followed by a deceleration during the third year. For both receptive and expressive vocabulary, FR-dyslexic children showed lower initial growth rates and thus a slower increase in vocabulary size during their second year. Yet, part of this acquired lag seemed to have resolved toward the end of their third year. If this growth curve is assumed to generalize outside of the period of investigation, then the delay would have fully resolved before the start of kindergarten. We do not have data at older ages to support this generalization. However, given the small differences in receptive and expressive vocabulary that remained at 35 months and the likelihood that these differences might even disappear, we conclude that, based on our data, (a lag in) infant vocabulary development is at most an additional but small risk factor for the development of dyslexia.

The parent report measures in Chapter 2 provide a subjective view on children's vocabulary development. Therefore, objective measures of early language (at 42 months) were used as a starting point for the study in Chapter 3. Interestingly, these measures also indicated that FR-dyslexic children have lower early oral language scores than FR-nondyslexic and TD children. These findings align with Chapter 2 as well as previous FR studies (e.g., van Bergen, de Jong, Maassen, Krikhaar, Plakas, & van der Leij, 2014) in that there are no differences between FR-nondyslexic and TD children in early language skills. Yet, they also indicate that differences between FR-dyslexic and nondyslexic groups reappear when using objective measures instead of parent reports. In Chapter 3 we assessed the influence of (differences in) early oral language on pathways into literacy development, with reading comprehension as the ultimate outcome. An important asset of this study was that data from *seven* occasions between the ages of 4 and 12 years were used to map literacy development.

Early language abilities were indeed found to underlie two pathways toward reading comprehension. For the first pathway, the effect was mediated by preliteracy skills and word decoding ability, while in the second pathway, the effect was mediated by later language skills. Early oral language skills had a limited effect on the development of preliteracy skills and there were no independent effects of early language on later word decoding skills. Although preliteracy skills in turn had a large effect on literacy development, the effects of early language on literacy development via the word decoding pathway fade away and are, as in Chapter 2, hard to trace in the long run. Long-term effects of early oral language on reading comprehension are both stronger and easier to trace in the language pathway.

Overall, the findings of the studies reported on in Chapter 2 and 3 suggest that the role of early oral language skills in the emergence of severe and persistent literacy difficulties is limited. These results deviate somewhat from those of previous studies. Studies comparable to the one in Chapter 2 (Scarborough, 1990; Torppa et al., 2010) showed small to moderate infant vocabulary (and grammar) deficits in FR-dyslexic children. These studies did not provide effect sizes (Cohen's *d*), but when these are computed and averaged, they show an increase of deficits with age (i.e., 1.5 years: receptive = -.22, expressive = -.46; 2.5 years: receptive = -.58, expressive = -.32; 3.5 years: receptive = -.78, expressive = -.75). In our study, the deficits were limited and did not increase with age. A recent meta-analysis by Snowling and Melby-Lervåg (2016) provided an overview of FR studies. It included studies that focused on long term effects of early oral language skills and provided the accompanying effect sizes for group differences on language measures at even older ages. The meta-analysis showed that for infant vocabulary knowledge, the average effect size for FR-dyslexic children versus TD control children was moderate (-.55). At kindergarten age, however, this average effect size became large (-.83). Moreover, FR-nondyslexic children also showed a small deficit compared to TD control children (-.34). The same results were found for grammar at preschool age (-.72 vs. -.28). In primary school, however, vocabulary deficits were again limited to FR children who go on to become dyslexic and they were only moderate in size (-.46; Snowling & Melby-Lervåg, 2016). Taken together, these findings suggest that early vocabulary deficits may be relatively small, but not as small as we found them to be.

These differences between our findings and those of previous studies raise the question why effects of early oral language were so small in our sample, and, vice versa, why effects are larger in other studies. There are two possible explanations. First, the early oral language findings so far predominantly stem from English-speaking children, whereas our data concerned Dutch-speaking children. The differences between the orthographic transparency of Dutch and English might affect the pattern of findings. Language skills, and vocabulary in particular, have been found to be more important for learning to read in English due to the high number of irregular words that this language contains (Wang, Castles, Nickels, & Nation, 2011; Wang, Nickels, Nation, & Castles, 2013). In the case of irregular words, it is key to *know* the words, as relying on decoding may lead the reader astray. In more transparent languages, such as Dutch, learning to read requires less vocabulary knowledge as most words are regular, resulting in a weaker relation between vocabulary and literacy acquisition. Poor early language skills, and small vocabulary size in particular, may thus have a stronger impact on later literacy development in English children than in Dutch children.

A second explanation, not incompatible with the first, concerns differences between studies in the composition of FR and/or dyslexic groups. FR of dyslexia is generally determined based on the criterion that children should have a first degree relative (usually a parent) with a history of severe and persistent literacy difficulties. It is good practice to affirm the dyslexia status of the parent(s) with additional (preferably direct) measures of their reading skills, which was also done in the studies mentioned below. However, in several recent studies (based on two different samples; e.g., Duff, Reen, Plunkett, & Nation, 2015; Hulme, Nash, Gooch, Lervåg, & Snowling, 2015; Snowling, Duff, Nash, & Hulme, 2015) children with an FR of dyslexia and children with preschool speech and language difficulties/specific language impairment (SLI), or children with comorbid disabilities, were subsequently collapsed into one FR of dyslexia group. These children differ essentially in the nature of their risk (family risk vs. language risk) as well as the cause of their (possible) reading difficulties. As children with SLI are oversampled and many of these children also become dyslexic, comorbidity rates increase from the outset, resulting in higher numbers of children with language difficulties in FR-dyslexic groups. Mixing groups of language and literacy impaired children in studies assessing the significance and relevance of risk factors for dyslexia could thus lead to an overestimation of the effect of early language deficits on the emergence of severe and persistent literacy difficulties. Stated differently, these studies may say more about the characteristic underlying deficits of children with comorbid language and literacy difficulties than of children with an FR of dyslexia. The findings of Chapter 2 and 3 suggest that language effects may be smaller in samples of children with a 'pure' FR of dyslexia and low comorbidity with speech and language difficulties.

A more theoretical explanation for why early language abilities only had a small impact on early literacy development in our sample is derived from the support we found for the segmentation theory in Chapter 2. With increasing segmentation of speech sounds, phonological representations become more detailed, affecting the development of phonological awareness and facilitating vocabulary growth (Boada & Pennington, 2006). When this process falters and children are unable to deal with the increasing demands on their segmentation skills, children may start to show small deficits in their vocabulary development, but more serious deficits may surface for their phonological awareness (see Chapter 2). At this stage of development, vocabulary deficits may still be small because the poor segmentation skills are thought to be compensated by other factors such as lexical richness and input clues to word meaning (Hoff, 2006). Deficits may be aggravated as vocabulary size increases further with development and places more demands on the impaired segmentation skills (e.g., Rispens & Baker, 2012).

Although segmentation is also required for language development, including receptive and expressive vocabulary (Chapter 2 and 3), as well as language comprehension and expressive syntax (Chapter 3), the demand for segmentation at (this early stage) might be highest/higher in the context of the preliteracy skill PA. In line with this, we suggest that the weak relation between early language and preliteracy skills results from (differences in) the extent to which the involved aspects require underlying segmentation skills. As children's segmentation skills need to undergo significant development changes in order to allow analysis and storage of phonological representations at the phoneme level, failure to make these changes thus results in severe deficits in preliteracy skills. Moreover, as literacy development progresses, the interaction between these language and literacy skills may add to these difficulties (Nation & Hulme, 2011). Following this line of reasoning, we contend that the ability to deal with the *required developmental changes* in segmentation skills, demanded by progressing literacy development, constitutes the additional risk factor for dyslexia; the small early language deficits are merely a weak symptom of impaired speech sound segmentation skills.

FAMILY RISK

One of the merits of the study in Chapter 3 was that the effect of FR on literacy development was modeled explicitly. FR was included as a predictor in the model to examine the exact moments during development at which FR might have an influence. Results showed no direct effects of FR on early or later language skills. This is in line with the findings from Chapter 2, where we only found deficits in FR-dyslexic children, which form the minority of all FR children in the study (approximately 1/3rd). These findings already indicated that early deficits are associated with the later dyslexia status of the children and not with their FR.

FR, however, had a small effect on early oral language skills through the second order general IQ factor, which represents what both verbal and nonverbal abilities have in common, i.e., 'general learning ability'. In other words, controlling for nonverbal abilities while investigating the effect of early oral language abilities on literacy development revealed that FR weakly influenced general learning ability and not early verbal abilities. More importantly, there was a stronger effect of FR on literacy(-related) abilities from the onset of literacy acquisition; it influenced preliteracy skills at age 6 (kindergarten), word reading accuracy as well as fluency at age 7.5 (start Grade 2), and reading comprehension at age 12 (Grade 6). FR thus had an additional effect on skills assessed in every phase of literacy development. This is in line with genetic evidence showing that from preschool to around Grade 2 new genetic effects kick in with the emergence of new literacy skills (Byrne et al., 2009). All of these genes are represented within parents' literacy skills (van Bergen, de Jong, Maassen, & van der Leij, 2014) and are thus captured within this FR.

Although the studies in Chapter 2 and 3 were mainly aimed at investigating risk factors associated with dyslexia, our findings may have revealed a possible protective factor. In an overview of the Dutch Dyslexia Program (van der Leij et al., 2013) several studies were highlighted in which patterns of performance on precursors of literacy development indicated absence of mild deficits in FR-nondyslexic children. In these studies, equal performance of FR-nondyslexic children compared to TD control children on underlying literacy-related skills was explained in terms of protective factors that (to some extent) prevented these children from developing dyslexia. These precursors included speech processing at 2 months (van Zuijen, Plakas, Maassen, Maurits, & van der Leij, 2013), nonverbal IQ at 47 months (van Bergen, de Jong, Maassen, Krikhaar et al., 2014), expressive syntax, verbal short-term memory, and language comprehension at age 53 months (van Bergen, de Jong, Maassen, Krikhaar et al., 2014; Plakas, van Zuijen, van Leeuwen, Thomson, & van der Leij, 2013), letter knowledge at age 6 (van Bergen, de Jong, Maassen, & van der Leij, 2014), and RAN at ages 6 and 8 (van Bergen et al., 2012; van Bergen, de Jong, Maassen, & van der Leij, 2014).

Following this line of reasoning, the receptive and expressive vocabulary performance of FR-nondyslexic children reported in Chapter 2, which was comparable to TD control children, indicates that infant vocabulary levels within the normal range could also be considered a protective factor for children at FR of dyslexia. We acknowledge that these vocabulary levels were based on indirect measures using parent reports, but the pattern of performance was confirmed by direct measures from age 53 months (see also van Bergen, de Jong, Maassen, Krikhaar et al., 2014) and age 9. Moreover, the longitudinal study reported in Chapter 3 showed that there was no independent direct effect of FR on early language or vocabulary measures at any point during development. Combined with other studies showing that early language deficits are specific to FR children who go on to become dyslexic (e.g., Scarborough, 1990; Torppa et al., 2010) we would say there is at least some evidence to suggest that normal progression of (early) vocabulary development could be considered a relative strength for FR children that might keep them from developing severe and persistent literacy difficulties. Further research is necessary to investigate how this might work.

PROTECTIVE FACTORS

GIFTEDNESS

The studies reported in Chapter 4, 5, and 6 focused on dyslexia in gifted children. Assessing the characteristics of this specific population allowed us to test and evaluate contradictory views and underlying assumptions regarding the role of protective factors and possibilities for compensation coming from the fields of twice-exceptionality and dyslexia. In short, the twice-exceptionality view assumes that gifted children with dyslexia can compensate their *dyslexia*-related risk factors with their *giftedness*-related protective factors (Foley Nicpon, Allmon, Sieck, & Stinson, 2011), resulting in higher literacy levels than expected based on their underlying deficits (indicating a certain degree of masking; e.g., Brody & Mills, 1997). In contrast, the core-deficit view by Stanovich (1996) assumes that the underlying risk factors are in line with the literacy level and that skills/strengths related to intelligence do not function as protective factors. Accordingly, if the literacy levels of gifted children with dyslexia are found to be higher than those of averagely intelligent children with dyslexia, this is the result of slightly less severe underlying deficits.

LITERACY PROFILES

We first aimed to establish whether gifted children with dyslexia, although meeting low achievement criteria for dyslexia, indeed have higher literacy levels than their averagely intelligent peers with dyslexia. If this is not the case, compensation (and thus some extent of masking) would not be assumed, which would run counter to the twice-exceptionality view. To the best of our knowledge, the studies reported in Chapter 4 and 6 are the first to provide comprehensive overviews of the literacy profiles of gifted children with dyslexia in various stages of development. Chapter 4 focused on children in Grades 2 to 4 of primary education (age 7 to 10) who were still in the process of learning to read. Chapter 6 involved students in the early grades of secondary education (ages 11 to 14) who were learning to read foreign languages in addition to reading in their native language (Dutch). In both studies children were divided into four groups based on low achievement criteria for dyslexia and high intelligence criteria for giftedness. This resulted in two dyslexia groups, i.e., one with high intelligence and one with average intelligence, and two reference groups without literacy difficulties, i.e., one typically developing (TD) and one with high intelligence.

Results of Chapter 4 showed that, despite the fact that both dyslexic groups fulfilled the same criteria for low achievement, gifted children with dyslexia had higher literacy levels than averagely intelligent children with dyslexia. These findings generalized across literacy skills, including both word reading and nonword reading fluency, as well as text reading fluency and word-level spelling accuracy. In addition, compared to the reference groups, the performance of gifted children with dyslexia was lower than the performance of TD control children on *all* assessed literacy abilities. The gifted control children were found to outperform all groups. Together these findings represent a stepwise pattern of group differences for literacy abilities in primary education, that is, dyslexia < gifted/dyslexia < TD < gifted.

The findings for primary school children were also attested for secondary school students, confirming that the pattern of group differences seems to be stable across development. Chapter 6 showed that gifted students with dyslexia continued to show higher word reading and sentence spelling levels than averagely intelligent students with dyslexia in their native Dutch language. Moreover, the complete stepwise pattern of performance (including both reference groups) persisted up to the first grades of secondary education. In addition, foreign language literacy performance of secondary school students was assessed in Chapter 6, including English, French, and German. Findings showed that the stepwise pattern of group differences found in Dutch generalized to English word reading and (word) spelling ability, but not to French and German literacy skills. For these latter two languages, gifted children with dyslexia did not show higher reading and/or spelling levels than their averagely intelligent peers with dyslexia.

Combined, the findings of Chapter 4 and 6 confirm that gifted children with dyslexia have higher literacy levels than their averagely intelligent peers with dyslexia despite meeting the same cut-off criteria for dyslexia. We found stable differences in patterns of performance that generalize from primary to secondary education as well as from Dutch to English. The question that remains is whether these higher literacy skills of gifted children with dyslexia are simply the result of less severe underlying deficits or may be the result of compensation by protective factors.

UNDERLYING DEFICITS

Next, we aimed to explain the differences in literacy levels between gifted and averagely intelligent children with dyslexia by looking into underlying deficits. This was done in Chapter 4 and 5. The study reported in Chapter 4 focused on mapping performance on underlying risk factors associated with dyslexia in both dyslexic and both reference groups. This approach reveals how groups relate to each other in terms of (severity of) underlying deficits and thus yields important information for diagnostic practice. In Chapter 5, in depth analyses were conducted on both dyslexic groups, while controlling for differences in literacy level, to test the specific assumptions of both theories under investigation.

The results of Chapter 4 indicated that gifted children with dyslexia had higher performance on PA and rapid automatized naming (RAN) tasks than averagely intelligent children with dyslexia. A comparison with the reference groups again resulted in a stepwise pattern of performance on underlying deficits associated with dyslexia. Results for verbal short-term memory (VSTM) were different; although averagely intelligent children with dyslexia indeed showed lowest performance, considerably lower than the TD control group, the gifted children with dyslexia scored in between the TD and gifted group. This finding aligns with previous studies indicating that VSTM is an ambivalent risk factor for dyslexia in terms of predictive strength (Vaessen, Bertrand, Denes, & Blomert, 2010; Ziegler et al., 2010) and suggests that deficits in VSTM are generally not found in gifted children with dyslexia.

In Chapter 5, differences between gifted and averagely intelligent children with dyslexia were controlled for differences in literacy level between both groups. This is necessary to determine whether the higher literacy levels of gifted children with dyslexia indeed result from somewhat less severe underlying deficits. This approach revealed that gifted children with dyslexia still showed higher performance on PA (and VSTM) tasks than the averagely intelligent children with dyslexia. Both dyslexia groups showed comparable performance on the RAN tasks. The results for RAN are in line with the core-deficit view, but the results for PA are not, as controlling for differences in literacy level should have resulted in about equal performance on underlying deficits. We suggest that the higher PA levels of the gifted children with dyslexia can be explained in terms of compensation on task-related skills. All tasks are impure to some extent, requiring, for example, processing speed or vocabulary knowledge in addition to phonological processing skill (e.g., van der Sluis, de Jong, & van der Leij, 2007). As gifted children may possess particular strengths in these ('impure') areas (e.g., Catts, Gillispie, Leonard, Kail, & Miller, 2002; Johnson, Im-Bolter, & Pascual-Leone, 2003), it is likely that they can use these strengths to increase their task performance, without having better phonological skills. Although this is not the type of compensation we are looking for in relation to the twice-exceptionality view, it does have important implications for diagnostic practice (see below).

So far, findings from Chapter 4 and 5 on underlying deficits supported both views. Deficits associated with dyslexia are present, *also* in gifted children with dyslexia, which is important for the twice-exceptionality view. However, when controlling for differences in literacy levels, findings on the size of deficits in both groups are largely in line with the core-deficit view, especially given the interpretation of task-related compensation.

Underlying Strengths

Subsequently, we examined whether children with dyslexia can have giftedness-related strengths or protective factors that are *relevant for literacy development* in the presence of underlying deficits associated with dyslexia. In Chapter 4 performance on protective factors across dyslexic and reference groups was mapped by adopting a diagnostic approach. In Chapter 5, differences in literacy level between both dyslexic groups were controlled for to identify possible resources for compensation in gifted children. The possible protective factors that were included were all giftedness-related. This means that they concerned skills that are considered relevant for literacy development to some extent (e.g., language and working memory) and have been found to be marked strengths in gifted children in previous studies.

The results of Chapter 4 showed that gifted children with dyslexia scored similarly to TD children on verbal WM, scored in between TD and gifted children on visuospatial WM and grammar, and scored equally to the gifted children on vocabulary. Compared to averagely intelligent children with dyslexia, gifted children with dyslexia thus seem to have specific strengths in verbal and visuospatial working memory (WM), as well as in grammar and vocabulary. These may function as protective factors in the development of dyslexia. Nevertheless, it should be noted the verbal and visuospatial WM and grammar performance of the gifted children with dyslexia may at the same time be considered a relative weakness in comparison with the gifted reference group, as the gifted children with dyslexia did not obtain the levels they should be able to given their high intelligence.

In Chapter 5, profiles of protective factors were investigated further while controlling for the difference in literacy level between gifted children with dyslexia and their averagely intelligent peers with dyslexia. The results showed that gifted children with dyslexia still outperformed averagely intelligent children with dyslexia on verbal and visuospatial WM, visuospatial short-term memory, and on vocabulary. There were no clear differences in performance on grammar after controlling for differences in literacy level. Complementing the findings of Chapter 4, Chapter 5 confirms that gifted children with dyslexia can have marked strengths in giftedness-related areas that are considered relevant for literacy in the presence of underlying deficits for dyslexia.

Overall, the findings of Chapter 4 and 5 showed that the underlying cognitive profiles of gifted children with dyslexia consist of dyslexia-related risk factors (PA and RAN) and giftedness-related strengths (verbal and visuospatial WM, visuospatial short-term memory, and vocabulary). Up to this point these characteristics of gifted children with dyslexia are still in line with both the twice-exceptionality view and the core-deficit view. However, a next step is to determine whether giftedness-related strengths can actually function as protective factors and might influence the manifestation of dyslexia in these children. Therefore, both views were further evaluated and tested by looking into possibilities for compensation.

COMPENSATION

To evaluate the twice-exceptionality view in relation to the core-deficit view and put the underlying assumptions of both views to the test, we took three different approaches to assessing compensation. First, the cognitive characteristics of gifted children with *relative* literacy difficulties, so called 'borderline-dyslexic' children, were examined. These children form a special group for whom compensation and masking are thought to prevail, which would explain why they do not meet the cut-off criteria for dyslexia. Second, specific hypotheses regarding the *underlying cognitive profiles* of gifted children with dyslexia were evaluated to test for the existence of direct compensation. Third, possibilities for compensation in a foreign language were assessed. Compensation is hard to test in the native language, but it is easier to do so in foreign language learning. By assessing foreign language performance of gifted children with dyslexia and controlling for differences in native language performance, possible compensation in the native language can be controlled for. This allows for testing the influence of *additional factors*, specific for that foreign language, that might explain the higher literacy levels of gifted students with dyslexia.

In Chapter 5, we compared the cognitive profiles of borderline-dyslexic children to those of gifted children with dyslexia. In contrast to gifted children with dyslexia, borderline-dyslexic children only show a significant discrepancy between their high intelligence and their literacy skills (instead of also *low* achievement), and miss the diagnostic low reading and/or spelling achievement threshold for dyslexia. According to the twice-exceptionality view this missed diagnosis is unjust, because the borderline-dyslexic children's literacy difficulties are thought to be masked as a result of compensation. However, the results of Chapter 5 proved otherwise. Results showed that the borderline-dyslexic children had less severe underlying deficits in PA compared to gifted children with dyslexia and at most equal deficits in RAN. In addition, both groups showed comparably high performance on protective factors across the board (with verbal WM as the only exception).

Supplementary findings resulting from the individual profiles of risk and protective factors of children in both groups revealed that both groups had about equal percentages of children with deficits in PA, but the borderline-dyslexic group had a lower percentage of children with a deficit in RAN. These figures rendered an accumulation of risk factors for the gifted children with dyslexia; children with a double deficit were more common in the gifted/dyslexia group than in the borderline-dyslexic group. Moreover, results on protective factors indicated that both groups contained comparable percentages of children with a strength in language skills (vocabulary and/or grammar). In addition, the borderline group had a somewhat higher percentage of children with a strength in verbal WM, whereas the gifted/dyslexia group contained relatively more children with a strength in visual WM. These numbers translated into comparable percentages of children with an accumulation of strengths in both groups.

In sum, the findings of Chapter 5 regarding the characteristics of borderline-dyslexic children provide no support for the twice-exceptionality view. There was no evidence that borderline-dyslexic children have more pronounced, more relevant (e.g., language skills), or an increased accumulation of protective factors compared to gifted children with dyslexia that would point toward (more efficient) compensation. Findings were more in line with the core-deficit view, as the higher literacy levels of the borderline-dyslexic children seemed to result from a combination of less severe phonological deficits and a reduced accumulation of risk factors. Therefore, it follows that borderline-dyslexic children at this point simply do not qualify for a dyslexia diagnosis.

Chapter 5 also provided a first test of the existence of 'direct compensation'. That is, we assessed whether there was a moderation of the size of a risk factor by an additional or co-occurring protective factor. This was done by evaluating hypotheses regarding differences in underlying cognitive profiles of risk and protective factors between gifted and averagely intelligent children with dyslexia. Hypotheses for compensation can be tested by controlling for differences in literacy level between both dyslexic groups. Compensation is attested when underlying cognitive deficits associated with dyslexia would be *more severe* in gifted children with dyslexia than in averagely intelligent children with dyslexia. Such more severe deficits are a prerequisite for direct compensation, as otherwise there would be no room for protective factors to compensate. As stated above, findings showed that gifted children with dyslexia showed equal performance on RAN and even somewhat *higher* performance on PA tasks than averagely intelligent children with dyslexia. Consequently, the findings provide no support for this part of the twice-exceptionality view. We found no evidence for the existence of direct compensation in terms of a balance between risk and protective factors at a specific point in time, which would cause remediation of a defective underlying skill and resulting in higher literacy levels.

A third way of looking into compensation was reported in Chapter 6. In this study, the focus was on finding evidence for compensation in foreign languages. In foreign language (FL) learning, compensation is expected more to concern mechanisms or strategies that affect the reading process rather than the direct compensation described above. It can be tested whether there are additional factors that could be involved in these mechanisms and may explain the higher FL literacy levels of gifted students with dyslexia compared to their averagely intelligent peers with dyslexia. This can be done by controlling for differences in native language (NL) skills. This controlling for NL literacy does not only cancel out group differences in underlying subskills, but also possible compensatory mechanisms that already exist in the NL, and can thus reveal whether group differences in the FL have their origin in the NL, or result from additional factors, unique to the specific FL. The findings of Chapter 6, again including both dyslexia groups and both reference groups, indicated that the stepwise pattern of group differences found for secondary school students in the NL only generalized to English word reading and spelling, but not to French and German literacy skills. Controlling for NL skills showed that the stepwise pattern indeed persisted for English word reading and spelling skills. In other words, the higher literacy levels of gifted students with dyslexia compared to their averagely intelligent peers with dyslexia could not be accounted for by differences in NL skills. Unique factors or processes, specific to English as an FL, are responsible for the higher word reading and spelling levels of gifted students with dyslexia. These findings provide evidence for the existence of compensation (in English) and thus support the twice-exceptionality view.

Taken together, the findings of Chapter 5 and 6 provided important information about compensation. As a start, there seems to be no evidence for a compensatory influence of protective factors when *severe* underlying deficits are absent, as has been suggested for borderline-dyslexic children. As a consequence, it cannot be stated that borderline-dyslexic children unjustly miss a diagnosis of dyslexia due to compensation; at this point in development, they simply have no dyslexia (yet). Furthermore, we found no evidence for direct compensation in terms of moderation of the severity of a risk factor by a protective factor. However, direct compensation as suggested by Snowling et al. (2003) could still be possible in moderation of the *effect* of a risk factor on literacy development by a protective factor. This aspect of direct compensation is yet to be investigated. Finally, we did find evidence for existence of indirect compensation, though in the limited context of English as a foreign language. This proves that, once in place, compensatory mechanisms could be used to circumvent an underlying deficit or subdue the negative influence of this deficit and thereby improve literacy performance to the extent that was demonstrated in Chapter 6.

In general, these findings indicated that there is, in fact, a role for IQ in literacy development. This conclusion does not disqualify the core-deficit view, but does call for a nuance of its underlying assumptions (see below). Especially in the native language, almost all results were in line with the core-deficit view. The findings provided only some support for the twice-exceptionality view. Although main prerequisites concerning the literacy levels and cognitive characteristics of gifted children with dyslexia were confirmed, direct compensation was ruled out and evidence for possible compensatory mechanisms was very much limited to the specific context of English as a foreign language. Based on the combination of these findings, we contend that the role of IQ in literacy development does deserve further attention, and especially high IQ in relation to dyslexia. Further theoretical implications for both views are discussed below.

THEORETICAL IMPLICATIONS

MULTIPLE DEFICIT MODEL

The assumptions underlying the multiple deficit model (MDM; Pennington, 2006) have proven to be very useful for investigating cognitive risk and protective factors associated with dyslexia, also in this dissertation. The framework itself, however, is merely an empty mold. For each particular disorder, relevant risk and protective factors have to be postulated as well as a description of *how* they might interact. At the same time, the way findings from the literature fill the MDM in the case of dyslexia also affects the evolution of the theory behind the framework. Some risk factors are relatively undisputed (e.g., PA, RAN) and have proven their value across alphabetic languages (Caravolas et al., 2012; Ziegler et al., 2010). However, the role of early oral language was less clear. The findings reported in Chapter 2 and 3 indicated that early language should be considered relevant in relation to the development of dyslexia, but as a risk factor, its influence is smaller and its position less prominent in Dutch than in English. The addition of protective factors to the MDM, which was one of the main focuses of this dissertation, and the way FR is defined or modeled also influences how the MDM shapes our knowledge of dyslexia. Overall, the findings of the studies presented in this dissertation illustrate that the 'content' of the MDM in terms of risk and protective factors may not only vary across developmental disorders but, with respect to dyslexia, can also differ across languages (e.g., English vs. Dutch) and/or per specific population (e.g., gifted vs. averagely intelligent children with dyslexia).

TWICE-EXCEPTIONALITY FRAMEWORK

Some of the studies reported in this dissertation were the first to provide empirical data of the cognitive and behavioral characteristics of gifted children with dyslexia, which allowed us to evaluate the twice-exceptionality view and put its underlying assumptions to the test. This dissertation showed that some assumptions were supported, but some were refuted, and others remain speculative and require further research. The main assumption that the cognitive profiles of gifted children with dyslexia consist of dyslexia-related deficits and giftedness-related strengths was indeed confirmed, but it is still debatable whether these strengths may also function as protective factors in the development of dyslexia. The assumption of higher literacy levels in gifted children with dyslexia compared to averagely intelligent children with dyslexia was also confirmed, indicating some degree of masking. However, whether this masking results from compensation can be disputed, as no evidence for compensation was found in the native language. The fact that we found a first clue that compensatory mechanisms could exist, and could be responsible for higher literacy levels in foreign language learning, suggests that exploring the avenue of compensatory mechanisms or strategies is most promising in this area. In line with this, we contend that the twice-exceptionality framework certainly provides an interesting new view on the role of intelligence in the development and manifestation of learning disabilities. However, the underlying assumptions should not be oversimplified or overgeneralized; a critical stance toward and direct testing of these assumptions is necessary for this framework to be of potential influence on future research.

CORE-DEFICIT VIEW

The main assumptions underlying the core-deficit view were largely supported across studies on the combination between giftedness and dyslexia reported in this dissertation; there were no major contradictions that would dispute its position as one of the most important theories in research on dyslexia over the past few decades. However, as not all of our findings were completely in line with the core-deficit view, statements derived from this theory should be nuanced to some extent. As stated in Chapter 5, findings showed that literacy performance and underlying cognitive deficits associated with dyslexia seem to be *largely* independent of intelligence, *also* in gifted populations with literacy problems. Especially the latter indicates where this dissertation has added to previous work in this area.

Interestingly, it is the counterevidence found in Chapter 5, i.e., the higher PA (and VSTM) scores of the gifted children with dyslexia, as well as the proposed explanation, i.e., task-related compensation, that further underlines the added value of testing the assumptions of the core-deficit view in high IQ populations. Although this study was the first to suggest the existence of task-related compensation in gifted children with dyslexia, and replication is thus essential, this explanation for the higher PA levels illustrates a paradox concerning the theoretical consequences for the core-deficit view. In one way, the gifted children with dyslexia were necessary to demonstrate where the assumptions of the core-deficit view may *not* hold;

which would be when the severity of deficits, as measured on tasks, is influenced by factors associated with intelligence. On the other hand, it is precisely this task-related compensation, which may be most pronounced in gifted children, that could be considered an *extreme outcome* of Stanovich's own line of reasoning; skills that are further away from the core deficits, and thus more related to intelligence, may lead to some differentiation in underlying cognitive profiles of children with dyslexia, but *also for core deficits*. This alternative, which could only be discovered when testing the core-deficit view in a gifted population, would take away the earlier proposed nuance and provide the ultimate (yet unanticipated) confirmation of this theory.

PRACTICAL IMPLICATIONS

The findings of the studies reported in this dissertation speak to current clinical practices surrounding diagnosing dyslexia. We will address several practical implications that may influence evaluation of test information during and decision making following diagnostic assessment.

DIAGNOSTIC ASSESSMENT

EARLY IDENTIFICATION

In terms of early identification, the FR studies of Chapter 2 and 3 have provided more information about the role of early oral language. It can be concluded that, for Dutch children, its significance as a screening measure for dyslexia proved to be fairly limited. Early receptive and expressive vocabulary deficits of children with an FR of dyslexia who later become dyslexic are generally mild and only present in a limited percentage of these children. However, when early deficits emerge in children's receptive as well as expressive language or when early deficits co-occur with other risk factors, impact on later literacy development may increase. As (early) language is not only relevant for word reading ability but also for reading comprehension (Lee, 2011; Muter, Hulme, Snowling, & Stevenson; Ricketts, Nation, & Bishop, 2007), monitoring language development may be beneficial in general (though not for early identification of dyslexia).

COMPENSATION

The task-related compensation that was described in Chapter 5 is of major significance for diagnostic practice. If gifted children with dyslexia are indeed able to artificially increase their performance on (some) phonological tasks by relying more heavily on their higher vocabulary levels (e.g., Catts et al., 2002) or higher processing speed (e.g., Johnson et al., 2003), this means that the presence of underlying deficits is harder to attest during diagnostic assessment. In some protocols for dyslexia (e.g., Blomert, 2006), a demonstrable phonological deficit is a prerequisite for reimbursed treatment. As this additional requirement may lead to unequal treatment of children with severe and persistent literacy difficulties, and especially disadvantages gifted children given what is described above, we contend that it is essential to diagnose dyslexia based on literacy performance *only*, and *not* include literacy-related deficits as additional criteria (e.g., SDN et al., 2016). Mapping these risk factors can provide important information for *underpinning* a diagnosis or inform decision making regarding suitable interventions, but should not be part of the diagnostic criteria.

More in general, the findings of Chapter 4, 5, and 6 illustrate the relevance of also mapping a child's particular underlying strengths during the diagnostic process, as they may form a (partial) explanation for specific findings at the behavioral level (e.g., less severe underlying deficits and/or higher literacy levels) or provide clues for a tailored intervention. This can be done by standard intelligence testing (as a first indicator of the presence of specific strengths) or by testing specific skills (e.g., vocabulary, working memory). Although this dissertation mainly focused on cognitive factors, it is important to also consider environmental factors in this respect. Some studies already suggested environmental factors that may or may not function as protective factors in the development of dyslexia (e.g., van Bergen et al., 2011; Kiuru et al., 2013), but more research in this area is necessary.

Criteria for Diagnosis and Intervention

The studies reported in this dissertation showed that diagnostic criteria for dyslexia work just as well for gifted children as for averagely intelligent children. If literacy difficulties are severe and persistent, both groups of children should receive a diagnosis of dyslexia (Blomert, 2006; Kleijnen et al., 2008; SDN et al., 2016). However, a recurring problem in our gifted samples is that children may not have underlying deficits, nor severe and persistent literacy difficulties, but still experience serious problems with their literacy levels. Refraining from help is extremely unsatisfactory for clinicians and poses serious threats to the educational prospects and psychological well-being of these children. This issue has been previously addressed by Elbro (2010) who differentiated between dyslexia as a disability, i.e., poor ability, and dyslexia as a handicap, i.e., the consequences of poor ability.

A solution for this problem could be the introduction of a so called 'handicap-criterion' during diagnostic assessment. This criterion should involve a quantification of the extent to which a child experiences its literacy difficulties as a handicap. For example, a first step could be to determine the difficulty of texts that are generally read in the first grade of pre-university education (i.e., vwo), or an educational level that is in accordance with the potential of the child. Subsequently, it is important to define the level of text reading fluency that would be sufficient to be able to process these texts. Comparing the required literacy level with the current literacy level of the child would then result in an objective and *meaningful* discrepancy measure (e.g., an 'achievement-requirement' discrepancy), that could inform further decision making concerning appropriate interventions. The essence of this solution, which also addresses the ethical aspect of the issue, is that a diagnostician can comply with a client's legitimate request for help without compromising the dyslexia diagnosis.

Subsequently, what interventions for gifted children with dyslexia and/or borderline-dyslexic children should look like should be the focus of future studies (see Pavey, 2016, for some suggestions). For gifted children with dyslexia, interventions could involve a combination between traditional dyslexia treatment (e.g., systematic phonics training) and addressing strengths to develop compensatory mechanisms, although tailoring would be necessary. As traditional interventions are made for averagely intelligent children, the learning pace or learning style may frustrate gifted children with dyslexia (Fischer, 2002). Fischer (2002) therefore suggests that interventions for shorter periods and at higher levels of cognitive demand could provide most benefit for the alleviation of dyslexia in gifted children. For borderline-dyslexic children, interventions could focus on a combination between the development of compensatory mechanisms in an attempt to attain a higher literacy level and addressing more general educational needs (e.g., in terms of motivation, study skills, or dealing with the experienced handicap). Montgomery (2015) proposes that gifted children with literacy difficulties mostly need higher-order strategies in order to allow them to use their cognitive strengths to gain knowledge that is not acquired naturally. These strategies could involve error analysis, analysis of linguistic structures and language concepts, and the development of methods to address their own errors.

To conclude, it is our view that every child having literacy difficulties or experiencing the *consequences* as problematic should be eligible for remediation or intervention, *irrespective* of a diagnosis. Although the focus of this dissertation was mainly on the question which risk and protective factors may be involved in (a diagnosis of) dyslexia, future questions should be more about how to offer every child the help and support it needs. Evidently, this outlook may demand essential changes in current educational and diagnostic practice. Yet, as researchers and diagnosticians, we owe it to these children to give our best effort to make these changes.

APPENDICES

APPENDIX A	Chapter 2
APPENDIX B	Chapter 2
APPENDIX C	Chapter 3
APPENDIX D	Chapter 5

APPENDIX A (CHAPTER 2)

Parameter Estimates of the Final Latent Growth Curve Model of Receptive Vocabulary

Parameter	Unstandardized	*SE*	Standardized
Predictor means			
Family risk	0.675***	.032	-
Dyslexia	0.241***	.029	-
Gender	0.410***	.034	-
Latent factor means			
IS	-1.335***	.035	-
LS	1.241***	.036	-
QS	-0.159***	.011	-
Predicted means			
Vocabulary T1	-1.335***	.035	-
Vocabulary T2	-0.253***	.039	-
Vocabulary T3	0.511***	.037	-
Vocabulary T4	0.957***	.025	-
Predictor variances and covariances			
Family risk	0.220***	.021	1.000
Dyslexia	0.183***	.018	1.000
Gender	0.242***	.024	1.000
Family risk ← → Dyslexia	0.078***	.015	.391
Family risk ← → Gender	0.006	.016	.027
Dyslexia ← → Gender	0.005	.014	.024
Latent factor residual variances and covariances			
IS	0.237***	.025	.989
LS	0.174***	.027	.959
QS	0.014***	.003	.950
IS ←→ LS	-0.015	.018	-.076
IS ←→ QS	-0.013*	.005	-.227
LS ←→ QS	-0.046***	.008	-.931
Direct effects			
Family risk → IS	0.031	.081	.030
Dyslexia → IS	-0.099	.091	-.086
Gender → IS	0.072	.071	.072
Family risk → LS	0.012	.081	.013
Dyslexia → LS	-0.189*	.091	-.190
Gender → LS	0.077	.071	.089
Family risk → QS	-0.011	.024	-.043
Dyslexia → QS	0.062*	.027	.219
Gender → QS	-0.022	.021	-.088
Error variances			
E_{T1}	0.000	.000	.000
E_{T2}	0.071***	.011	.198
E_{T3}	0.043***	.008	.138
E_{T4}	0.000	.000	.000

Note. Standardized estimates for residual variances and error variances are proportions of unexplained variance. T = time point; IS = Intercept; LS = linear slope; QS = quadratic slope.
$*p < .05$. $** p < .01$. $*** p < .001$.

APPENDIX B (CHAPTER 2)

Parameter Estimates of the Final Latent Growth Curve Model of Expressive Vocabulary

Parameter	Unstandardized	*SE*	Standardized
Predictor means			
Family risk	1.675***	.032	-
Dyslexia	1.241***	.029	-
Gender	1.410***	.034	-
Latent factor means			
AS	1.40***	-	-
IS	-1.23***	-	-
CF	0.23	-	-
Predicted means			
Vocabulary T1	-1.234	-	-
Vocabulary T2	-0.479	-	-
Vocabulary T3	0.515	-	-
Vocabulary T4	1.072	-	-
Predictor variances and covariances			
Family risk	0.220***	.021	1.000
Dyslexia	0.183***	.018	1.000
Gender	0.242***	.024	1.000
Family risk ← → Dyslexia	0.078**	.015	.391
Family risk ← → Gender	0.006	.016	.027
Dyslexia ← → Gender	0.005	.014	.024
Latent factor residual variances and covariances			
AS	0.201***	.035	.996
IS	0.021***	.002	.962
CF	2.059*	.748	.912
AS ←→ IS	-0.018	.012	-.276
AS ←→ CF	-0.213*	.104	-.312
IS ←→ CF	0.147**	.037	.710
Direct effects			
Family risk → AS	0.045	.084	.215
Dyslexia → AS	-0.008	.113	-.044
Gender → AS	0.036	.082	.161
Family risk → IS	-0.027	.024	-.382
Dyslexia → IS	-0.046	.027	-.731
Gender → IS	0.021	.021	.282
Family risk → CF	-0.018	.285	-.025
Dyslexia → CF	-0.854*	.314	-1.330
Gender → CF	0.532*	.246	.720
Error variances			
E_{T1}	0.000	.000	.000
E_{T2}	0.081***	.018	.205
E_{T3}	0.058***	.058	.162
E_{T4}	0.000	.000	.000

Note. Standardized estimates for residual variances and error variances are proportions of unexplained variance.
T = time point; IS = Intercept; CF = change factor; AS = Asymptote.
*p < .05. ** p < .01. *** p < .001.

Appendix C (Chapter 3)

Correlations between Variables for the Complete Sample (Part 1)

Variables	1.	2.	3.	4.	5.	6.	7.	8.	9.	10.	11.	12.	13.	14.	15.	16.	17.	18.
1. T1 Patterns	-																	
2. T1 Block design	.58	-																
3. T1 Object assembly	.43	.49	-															
4. T1 Picture completion	.26	.32	.29	-														
5. T1 Analogies	.27	.37	.23	.24	-													
6. T1 Categories	.37	.40	.38	.39	.41	-												
7. T2 Language comprehension	.35	.36	.23	.26	.39	.37	-											
8. T2 Expressive syntax	.33	.25	.20	.22	.32	.36	.61	-										
9. T2 Expressive vocabulary	.35	.28	.19	.22	.33	.37	.67	.66	-									
10. T2 Verbal STM	.36	.33	.29	.25	.32	.32	.48	.50	.49	-								
11. T3 Active LK	.27	.35	.29	.22	.26	.22	.39	.27	.37	.37	-							
12. T3 Passive LK	.30	.37	.35	.20	.25	.25	.39	.25	.33	.34	.90	-						
13. T3 Phoneme blending	.28	.35	.23	.16	.29	.22	.46	.37	.43	.46	.70	.67	-					
14. T3 Phoneme segmentation	.31	.40	.26	.16	.26	.19	.43	.29	.38	.39	.68	.66	.86	-				
15. T3 RAN colors	.32	.26	.15	*.12*	.17	.16	.28	.26	.24	.29	.43	.42	.38	.40	-			
16. T3 RAN objects	.28	.24	.18	*.13*	*.15*	*.16*	.25	.26	.20	.29	.41	.43	.29	.31	.82	-		
17. T4 Phoneme deletion	.36	.27	.17	*.07*	.20	.20	.30	.37	.21	.35	.42	.46	.47	.46	.37	.32	-	
18. T4 RAN colors	.20	.15	*.11*	*.05*	*.05*	*.11*	*.11*	*.10*	*.03*	*.09*	.20	.21	.16	.19	.65	.64	.25	-

Note. Coefficients in italics are not significant. T1 = kindergarten year 1; T2 = kindergarten year 1; T3 = kindergarten year 2; T4 = start Grade 2; T5 = end Grade 2; T6 = end Grade 3; T7 = Grade 6; STM = short-term memory; LK = letter knowledge; RAN = rapid automatized naming.

Appendix C (Chapter 3)

Correlations between Variables for the Complete Sample (Part 2)

Variables	1.	2.	3.	4.	5.	6.	7.	8.	9.	10.	11.	12.	13.	14.	15.	16.	17.	18.
19. T4 RAN digits	*.09*	*.01*	*-.02*	*-.04*	*-.00*	*-.05*	*.09*	*.10*	*.03*	*.02*	.24	.28	.21	.20	.49	.54	.24	.53
20. T4 Word accuracy	.24	.24	.17	.15	*.10*	.17	.34	.34	.28	.37	.54	.53	.53	.50	.42	.37	.61	.24
21. T4 Nonword accuracy	.30	.26	.17	.15	.14	.18	.30	.29	.21	.31	.49	.47	.50	.53	.28	.21	.59	.24
22. T4 Word reading fluency 1	.24	.25	.25	.14	.15	.23	.30	.34	.26	.28	.61	.63	.50	.53	.45	.45	.50	.25
23. T4 Word reading fluency 2	.25	.25	.23	.15	.18	.22	.30	.36	.27	.29	.60	.61	.49	.51	.44	.42	.48	.22
24. T5 Word accuracy	.24	.24	.17	*.09*	.19	.19	.35	.41	.25	.33	.47	.50	.45	.41	.36	.34	.57	.23
25. T5 Nonword accuracy	.26	.27	.16	*.09*	*.11*	.15	.31	.39	.22	.27	.48	.51	.44	.44	.34	.27	.58	.22
26. T5 Word reading fluency 1	.30	.25	.21	*.10*	.21	.21	.31	.37	.29	.30	.55	.59	.45	.46	.47	.47	.53	.30
27. T5 Word reading fluency 2	.27	.23	.22	*.10*	.17	.19	.31	.40	.29	.28	.54	.59	.45	.47	.46	.48	.52	.28
28. T6 Nonword accuracy	.27	.26	.20	*.04*	.15	.15	.20	.32	.17	.27	.42	.48	.43	.45	.33	.27	.56	.24
29. T6 Word reading fluency	.24	.24	.23	*.13*	.19	.17	.35	.38	.26	.32	.55	.58	.49	.52	.48	.44	.52	.26
30. T6 Nonword reading fluency	.25	.25	.22	*.06*	*.08*	.17	.28	.34	.21	.30	.38	.45	.39	.44	.36	.34	.48	.23
31. T6 Vocabulary	.19	.26	.14	.20	.20	.31	.53	.63	.55	.33	.33	.34	.35	.37	.31	.22	.30	.12
32. T7 Word reading fluency	*.16*	*.10*	*.15*	*.15*	*.00*	*.10*	.32	.34	.19	.27	.44	.45	.35	.32	.46	.52	.42	.34
33. T7 Nonword reading fluency	.21	*.16*	*.11*	*.15*	*-.01*	*.10*	.26	.23	*.12*	.24	.38	.42	.31	.28	.45	.42	.51	.33
34. T7 Vocabulary	.21	.22	*.14*	*.04*	*.03*	.20	.28	.40	.33	.30	.25	.32	.29	.32	.31	.34	.31	.21
35. T7 Reading comprehension	.29	.20	*.06*	*.11*	*.07*	*.14*	.28	.31	.23	.31	*.18*	.25	.30	.30	.35	.29	.33	.22
36. Family risk	*-.11*	*-.06*	*-.07*	*-.02*	*-.07*	*-.10*	-.17	*-.12*	*-.08*	-.22	-.25	-.27	-.32	-.33	-.26	-.31	-.34	-.17

Note. Coefficients in italics are not significant. T1 = kindergarten year 1; T2 = kindergarten year 1; T3 = kindergarten year 2; T4 = start Grade 2; T5 = end Grade 2; T6 = end Grade 3; T7 = Grade 6; STM = short-term memory; LK = letter knowledge; RAN = rapid automatized naming.

APPENDIX C (CHAPTER 3)

Correlations between Variables for the Complete Sample (Part 3)

Variables	19.	20.	21.	22.	23.	24.	25.	26.	27.	28.	29.	30.	31.	32.	33.	34.	35.	36.
19. T4 RAN digits	-																	
20. T4 Word accuracy	.33	-																
21. T4 Nonword accuracy	.21	.71	-															
22. T4 Word reading fluency 1	.36	.75	.71	-														
23. T4 Word reading fluency 2	.34	.72	.72	.95	-													
24. T5 Word accuracy	.32	.75	.61	.70	.66	-												
25. T5 Nonword accuracy	.28	.75	.78	.74	.76	.74	-											
26. T5 Word reading fluency 1	.42	.70	.63	.87	.83	.75	.72	-										
27. T5 Word reading fluency 2	.38	.71	.68	.90	.89	.73	.76	.94	-									
28. T6 Nonword accuracy	.35	.69	.74	.69	.69	.69	.82	.66	.71	-								
29. T6 Word reading fluency	.40	.70	.66	.87	.87	.73	.77	.90	.93	.72	-							
30. T6 Nonword reading fluency	.37	.67	.71	.80	.82	.66	.79	.79	.83	.78	.86	-						
31. T6 Vocabulary	*.11*	.36	.28	.39	.39	.33	.40	.38	.41	.33	.44	.34	-					
32. T7 Word reading fluency	.50	.57	.55	.75	.73	.62	.62	.82	.84	.59	.85	.73	.34	-				
33. T7 Nonword reading fluency	.46	.67	.65	.76	.77	.70	.74	.81	.82	.71	.83	.87	.33	.84	-			
34. T7 Vocabulary	.29	.43	.41	.42	.44	.37	.44	.35	.40	.45	.43	.38	.46	.32	.31	-		
35. T7 Reading comprehension	.32	.37	.35	.39	.41	.43	.42	.45	.47	.36	.42	.39	.33	.40	.39	.63	-	
36. Family risk	-.28	-.43	-.36	-.41	-.42	-.40	-.40	-.39	-.44	-.45	-.47	-.50	-.16	-.39	-.45	*-.17*	-.34	-

Note. Coefficients in italics are not significant. T1 = kindergarten year 1; T2 = kindergarten year 1; T3 = kindergarten year 2; T4 = start Grade 2; T5 = end Grade 2; T6 = end Grade 3; T7 = Grade 6; STM = short-term memory; LK = letter knowledge; RAN = rapid automatized naming.

Appendix D (Chapter 5)

Case Series showing the Cognitive Risk and Protective Factors observed among Averagely Intelligent Children with Dyslexia

	Riskfactors					Protective factors							
Case no.	PA deletion SS < 8	PA spoonerism SS < 8	RAN alpha-numeric SS < 8	RAN non-alphanumeric SS < 8	VSTM SS < 85 SS > 115	VWM SS > 115	VSSTM SS > 115	VSWM SS > 115	Grammar SS > 12	Voca-bulary SS > 12	IQ score	Number of weaknesses	Number of strengths
76	-	-	-	-			+	-		-	91	2	1
153			-	-					+		111	1	1
207			-	-			+	+			112	1	1
95	-		-	-		-	-	-			84	2	0
189	-			-	-		-	-			87	3	0
201	-		-	-	-	-		-		-	88	3	0
116	-							+			110	1	1
77	-		-	-	-	-				-	90	3	0
188	-	-	-		-	-					89	3	0
119	-	-			-	-	-	-		-	100	2	0
123	-		-	-	-	-	-	-		-	92	3	0
190			-	-				-		-	89	1	0
117	-		-	-				-			100	2	0
72	-	-	-	-				-		-	108	2	0
64	-	-			-		+			-	111	2	1
70	-	-	-	-	-		+	+		-	102	3	1
21	-		-	-					+		118	2	1
89	-	-	-	-		-	-	-	+		98	2	1
215	-	-	-	-							95	2	0
135	-	-	-	-			+	-		-	104	2	1
137	-	-	-	-				-		-	71	2	0
209	-	-	-	-			+	+			121	2	1
88	-		-								92	2	0
127	-	-	-	-	-					-	104	3	0
214	-	-	-	-			+	+			105	2	1
71	-	-	-								103	2	0
231	-	-	-	-	-			-		-	86	3	0
232	-	-	-					-			86	2	0
96	-		-	-	-	-		-			102	3	0
147		-						-			97	1	0
172	-	-	-	-	-	-	-	-		-	95	3	0
218			-	-	-			-	+	-	96	2	1
47	-	-					+	+			120	1	1
Total	28	19	27	24	13	0	8	6	4	0	98.70	2.12	0.40

Note. PA = phonological awareness; RAN = rapid automatized naming; VSTM = verbal short-term memory; VWM = verbal working memory; VSSTM = visuospatial short-term memory; VSWM = visuospatial working memory; SS = standard score.

SUMMARY

Dyslexia is a specific learning disability that is generally defined as having severe and persistent difficulties with acquiring reading and/or spelling skills at the word level. In The Netherlands, dyslexia affects approximately 4-7% of the children in primary education and around 12% of the students in secondary education. Recent models of developmental disorders assume that a disability results from a complex interplay between risk and protective factors. Thus despite having the same learning disability/diagnosis, children with dyslexia can greatly differ in their developmental trajectories and the difficulties that they experience. This dissertation contains studies on two specific populations of children, i.e., children with a family risk (FR) of dyslexia (Chapter 2 and 3) and gifted children with dyslexia (Chapter 4, 5, and 6). Combined, these five studies reveal more about risk factors, protective factors, and possibilities for compensation in children with dyslexia.

In Chapter 2, we examined the role of early oral language as a risk factor for dyslexia in children with and without an FR between the ages of 17 and 35 months. All children participated in the Dutch Dyslexia Program (DDP), in which children were followed from the age of 2 months until 12 years. Receptive and expressive vocabulary were assessed using parent reports. Estimation of growth trajectories revealed that FR children that became dyslexic (FR-dyslexic) had lower receptive vocabulary knowledge from 23 months onward and lower expressive vocabulary knowledge from 17 months onward than FR children who did not become dyslexic (FR-nondyslexic). In addition, FR-dyslexic children showed lower initial growth rates, followed by partial recovery. This indicates delayed receptive and expressive vocabulary development for the FR-dyslexic group. Growth trajectories of FR-nondyslexic and typically developing (TD) children did not differ. It was concluded that early deficits in receptive and expressive vocabulary are associated with later literacy outcomes and not with FR. Vocabulary can be considered as a small risk factor for dyslexia.

In Chapter 3, we investigated the influence of early oral language on pathways into literacy development, with reading comprehension as the ultimate outcome, in children with and without an FR of dyslexia between 4 and 12 years. As in Chapter 2, the data came from the DDP sample. Early oral language ability was (directly) assessed with tests. The results

showed that in the first pathway into literacy development, the effect of early oral language ability on reading comprehension was mediated by preliteracy skills and word decoding ability. In the second pathway, this effect was mediated by later language abilities. FR had no effect on (early) oral language abilities, but affected literacy development through its subsequent effects on preliteracy skills, word decoding, and reading comprehension.

Chapter 4, 5, and 6 focused on the combination between giftedness and dyslexia to gain more insight in the role of protective factors in the development of literacy difficulties. Differences between gifted children with dyslexia and averagely intelligent children with dyslexia were examined regarding literacy levels, cognitive risk and protective factors, and possibilities for compensation. Both dyslexic groups met the same low achievement criteria for dyslexia. This approach allowed us to test and evaluate the underlying assumptions of the twice-exceptionality view, assuming that gifted children with dyslexia have higher literacy levels because they can compensate underlying dyslexia-related deficits with giftedness-related strengths, and contrast it with the core-deficit view, assuming that the underlying deficits associated with dyslexia are not influenced by intelligence.

Chapter 4 is the first study to present empirical data on the behavioral and cognitive characteristics of gifted children with dyslexia learning to read in the early to middle grades of primary education. The study followed a diagnostic approach; performance differences between groups were presented as they would occur during diagnostic assessment. Therefore, two nondyslexic reference groups were included (i.e., averagely intelligent TD children and gifted children without literacy difficulties) in addition to both dyslexic groups. Findings showed that gifted children with dyslexia performed in between averagely intelligent children with dyslexia and TD children on all literacy tests, while gifted children outperformed all groups, resulting in a stepwise pattern of performance (i.e., dyslexia < gifted/dyslexia < TD < gifted). The cognitive profile of gifted children with dyslexia showed signs of weaknesses in phonological awareness (PA) and rapid automatized naming (RAN), and strengths in verbal short-term memory (VSTM), verbal and visuospatial working memory (WM), and language skills. Findings indicate that phonology is a risk factor for gifted children with dyslexia, but that it could be moderated by other skills, such as WM, grammar, and vocabulary. These findings suggest that opportunities for compensation of a cognitive deficit and masking of literacy difficulties may exist in gifted children with dyslexia.

Chapter 5 built on the results of Chapter 4 by assessing differences between gifted and averagely intelligent children with dyslexia in dyslexia-related deficits and giftedness-related strengths while controlling for differences in literacy level. In addition, differences in cognitive profiles between gifted children with dyslexia and gifted children with relative reading difficulties (i.e., borderline-dyslexic children) were investigated in terms of severity and accumulation of deficits and strengths. Both comparisons were aimed at gaining more insight in possibilities for compensation. Findings showed that gifted children with dyslexia had about equally severe deficits in RAN as averagely intelligent children with dyslexia. Yet, their deficits in PA and VSTM were seemingly less severe. This latter finding is interpreted to be caused by higher performance on *task-related* aspects that are unrelated to the deficits associated with dyslexia that were under investigation. The higher literacy levels of borderline-dyslexic children compared to gifted children with dyslexia seemed the result of both fewer combinations of risk factors as well as less severe phonological deficits in this group. Borderline-dyslexic children had no specific strengths that are more relevant for literacy development. Overall, findings showed no indication of compensation of dyslexia-related deficits by giftedness-related strengths in gifted children with dyslexia or borderline-dyslexic children.

The study reported in Chapter 6 focused on foreign language literacy profiles of gifted students with dyslexia in the first grades of secondary education. The existence of compensatory mechanisms was tested by controlling for group differences in native language literacy performance. Controlling for native language literacy cancels out underlying native language subskills and possible compensatory mechanisms that might already exist in the native language. Foreign languages under investigation were English, French, and German. Results revealed the same stepwise pattern for *native language* literacy skills as previously attested in primary education, i.e., dyslexia < gifted/dyslexia < TD < gifted. This performance pattern translated to English word reading and spelling, but not to French or German literacy. These patterns persisted after controlling for native language skills. The findings suggest that the higher English word reading and spelling levels of gifted students with dyslexia result from additional factors or mechanisms that are unique to English as a foreign language. Differences in results between foreign languages are discussed in terms of variation in orthographic transparency and exposure.

Although some of the main assumptions underlying the twice-exceptionality view were supported, i.e., presence of higher literacy levels and cognitive risk as well as protective factors in gifted children with dyslexia, evidence for compensation was limited and restricted to compensatory mechanisms in English as a foreign language. Findings in the native language were largely in line with the core-deficit view of dyslexia. Nevertheless, if approached with care, the twice-exceptionality view has the potential to provide an interesting new outlook on the role of intelligence in the development and manifestation of learning disabilities in future studies.

Taken together, the results of the studies reported in this dissertation further specify the role of early language and FR as risk factors for dyslexia and the influence of giftedness as a context for potential protective factors. Concerning risk factors, early oral language was found to form the foundation of literacy development and can be considered an additional but small risk factor for dyslexia in children learning to read in Dutch. Effects of FR do not influence early oral language, but affect literacy development from the early onset of literacy acquisition. With respect to protective factors, gifted children with dyslexia were confirmed to have higher literacy levels than their averagely intelligent peers with dyslexia, both in primary and secondary education. Their underlying cognitive profiles consisted of both dyslexia-related weaknesses and giftedness-related strengths. These strengths may function as protective factors and could be involved in compensation. However, evidence for compensation was limited; the option of direct compensation was eliminated, that is, there is no moderation of the size of a risk factor by a co-occurring protective factor. Instead, findings pointed more toward the development of compensatory mechanisms, especially in specific contexts such as English as a foreign language. Overall, these studies illustrate the importance of mapping both risk factors and protective factors during diagnostic assessment, as outcomes may provide clues for remediation and inform intervention.

SAMENVATTING

(SUMMARY IN DUTCH)

Dyslexie is een specifieke leerstoornis waarbij sprake is van ernstige en hardnekkige moeilijkheden in het aanleren van accuraat en vlot lezen en/of spellen op woordniveau. In Nederland heeft circa 4-7% van de basisschoolkinderen dyslexie en ongeveer 12% van de leerlingen op de middelbare school. Recente modellen van ontwikkelingsproblemen veronderstellen dat stoornissen ontstaan vanuit een complex samenspel tussen risico- en protectieve factoren. Hieruit volgt dat, hoewel kinderen dezelfde diagnose krijgen, grote onderlinge verschillen kunnen bestaan tussen individuele ontwikkelingstrajecten en de moeilijkheden die kinderen ervaren. Deze dissertatie bevat studies die twee specifieke groepen kinderen omvatten, dat wil zeggen, kinderen met een familiair risico (FR) op dyslexie (Hoofdstuk 2 en 3) en hoogbegaafde kinderen met dyslexie (Hoofdstuk 4, 5 en 6). Tezamen verschaffen deze vijf studies meer inzicht in risicofactoren, protectieve factoren en mogelijkheden voor compensatie bij kinderen met dyslexie.

In Hoofdstuk 2 is de rol van de ontwikkeling van vroege verbale taalvaardigheden als risicofactor voor dyslexie onderzocht bij jonge kinderen (17-35 maanden) met en zonder een FR. Alle kinderen in deze studie namen deel aan het Dutch Dyslexia Program (DDP), waarin kinderen werden gevolgd tussen de leeftijd van 2 maanden en 12 jaar. Receptieve en productieve woordenschat werden onderzocht door middel van oudervragenlijsten. Groei-analyses lieten zien dat FR-kinderen die later dyslectisch werden (FR-dyslectisch) vanaf een leeftijd van 23 maanden een kleinere receptieve woordenschat hadden dan FR-kinderen die later niet dyslectisch werden (FR-niet dyslectisch) en een kleinere productieve woordenschat hadden vanaf 17 maanden. Daarnaast maakten FR-dyslectische kinderen een minder sterke groei door aan het begin van hun ontwikkeling, gevolgd door een gedeeltelijk herstel. Dit duidt op een vertraagde ontwikkeling van de receptieve en productieve woordenschat in de FR-dyslectische groep. Er waren geen verschillen tussen de groeitrajecten van FR-niet dyslectische en normaal ontwikkelende kinderen. Hieruit werd geconcludeerd dat vroege tekorten in receptieve en productieve woordenschat gerelateerd zijn aan latere leesuitkomsten en niet aan het hebben van een FR.

In Hoofdstuk 3 is de invloed van vroege verbale taalvaardigheden op leesbegrip via verschillende ontwikkelingspaden en de rol van FR onderzocht. Evenals in Hoofdstuk 2 kwamen deze data van het DDP. Longitudinale data van kinderen tussen de 4 en 12 jaar met en zonder een FR zijn gebruikt voor de analyses. De resultaten lieten zien dat het effect van vroege verbale taalvaardigheid op leesbegrip gemedieerd werd door voorschoolse (*preliteracy*) vaardigheden en decodeervaardigheden in het eerste ontwikkelingspad. In het tweede ontwikkelingspad werd dit effect gemedieerd door latere taalvaardigheden. Er was geen effect van FR op (vroege) verbale taalvaardigheden, maar FR had wel invloed op de leesontwikkeling via achtereenvolgende effecten op voorschoolse vaardigheden, decodeervaardigheden en leesbegrip.

In Hoofdstuk 4, 5 en 6 is gekeken naar de combinatie tussen hoogbegaafdheid en dyslexie om meer zicht te krijgen op de rol van protectieve factoren (voortkomend uit hoge intelligentie) in de ontwikkeling van leesproblemen (dyslexie). Verschillen tussen hoogbegaafde kinderen met dyslexie en gemiddeld intelligente kinderen met dyslexie werden onderzocht op het gebied van lees- en spellingvaardigheden, cognitieve risico- en protectieve factoren en mogelijkheden voor compensatie. Kinderen in beide dyslexiegroepen voldeden aan dezelfde criteria voor dyslexie, namelijk een zwakke prestatie op lezen/spellen (laagste 10%). Deze aanpak bood de mogelijkheid de onderliggende aannames van de '*twice-exceptionality*' benadering te testen en evalueren. Deze visie veronderstelt dat hoogbegaafde kinderen met dyslexie betere lees- en spellingvaardigheden hebben dan gemiddeld intelligente kinderen met dyslexie, omdat ze hun onderliggende dyslexie-gerelateerde tekorten kunnen compenseren met hun hoogbegaafdheid-gerelateerde sterktes. Deze aanname is afgezet tegen de '*core-deficit*' benadering, die veronderstelt dat de onderliggende tekorten die geassocieerd worden met dyslexie niet worden beïnvloed door intelligentie.

De studie in Hoofdstuk 4 is de eerste die empirische data presenteert met betrekking tot de gedrags- en cognitieve kenmerken van hoogbegaafde kinderen met dyslexie die leren lezen in de eerste klassen van het basisonderwijs. De prestaties van vier groepen werden vergeleken; een groep gemiddeld intelligente kinderen met dyslexie, een groep hoogbegaafde kinderen met dyslexie, normaal ontwikkelende kinderen met een gemiddelde intelligentie en een groep hoogbegaafde kinderen zonder leerproblemen. Prestatieverschillen tussen groepen werden weergegeven zoals deze zouden voorkomen tijdens diagnostisch onderzoek. De bevindingen toonden aan dat de prestaties van hoogbegaafde kinderen met dyslexie op alle lees- en spellingtaken tussen die van gemiddeld intelligente kinderen met dyslexie en normaal ontwikkelende kinderen in lagen, terwijl hoogbegaafde kinderen betere uitkomsten hadden dan alle andere groepen. Er was dus sprake van een stapsgewijs patroon (dyslexie < hoogbegaafd/dyslexie < normaal ontwikkelend < hoogbegaafd). De cognitieve profielen van hoogbegaafde kinderen met dyslexie vertoonden aan de ene kant tekenen van tekorten in het fonologisch bewustzijn en de benoemsnelheid en aan de andere kant sterktes in het verbale kortetermijngeheugen, het verbale en visueel-ruimtelijke werkgeheugen en taalvaardigheden. Deze resultaten tonen aan dat een fonologisch probleem een risicofactor vormt voor hoogbegaafde kinderen met dyslexie, maar dat dit tekort gemodereerd kan worden door andere vaardigheden, zoals werkgeheugen, grammatica en woordenschat. Kortom, hoogbegaafde kinderen met dyslexie hebben mogelijk de kans een cognitief tekort te compenseren en hun lees- en/of spellingproblemen te maskeren.

Hoofdstuk 5 bouwde verder op de resultaten van Hoofdstuk 4 door verschillen tussen hoogbegaafde en gemiddeld intelligente kinderen met dyslexie op het gebied van dyslexie-gerelateerde tekorten en hoogbegaafdheid-gerelateerde sterktes te onderzoeken en tegelijkertijd te controleren voor verschillen in lees- en spellingvaardigheid. Daarnaast werd onderzocht of er verschillen waren tussen cognitieve profielen van hoogbegaafde kinderen met dyslexie en hoogbegaafde kinderen met lees- en spellingproblemen die (net) niet ernstig en hardnekkig genoeg waren om voor een dyslexiediagnose in aanmerking te komen (i.e., grens-dyslectische kinderen). Het ging in deze vergelijking om zowel de prestaties op de cognitieve taken als de opeenstapeling van tekorten en sterktes. De vergelijkingen waren erop gericht meer inzicht te verkrijgen in mogelijkheden voor compensatie. De resultaten lieten zien dat de tekorten in de benoemsnelheid van hoogbegaafde kinderen met dyslexie vergelijkbaar waren met die van gemiddeld intelligente kinderen met dyslexie. Echter, hun tekorten in het fonologisch bewustzijn en verbale kortetermijngeheugen leken minder ernstig. De interpretatie die wordt voorgesteld voor deze laatste bevinding is dat de hoogbegaafde kinderen met dyslexie beter zijn in *taak-gerelateerde* aspecten die geen verband houden met de tekorten die geassocieerd worden met dyslexie en daardoor beter presteren. De hogere lees- en spellingniveaus van grens-dyslectische kinderen in vergelijking met hoogbegaafde kinderen met dyslexie leken voort te komen uit zowel minder combinaties van risicofactoren als minder ernstige fonologische tekorten in deze groep. Grens-dyslectische kinderen beschikten niet over specifieke sterktes die meer relevant geacht worden voor de leesontwikkeling. Over het geheel genomen gaven de bevindingen geen indicatie voor compensatie van dyslexie-gerelateerde tekorten door hoogbegaafdheid-gerelateerde sterktes bij hoogbegaafde kinderen met dyslexie of (hoogbegaafde) grens-dyslectische kinderen.

De studie die beschreven is in Hoofdstuk 6 richtte zich op de lees- en spellingprofielen van hoogbegaafde leerlingen met dyslexie in de eerste klassen van het voortgezet onderwijs. Hierbij lag de focus op lezen en spellen in moderne vreemde talen. Het bestaan van compensatiemechanismen werd getest door de prestaties van de vier groepen op moderne vreemde talen te vergelijken, nadat gecontroleerd was voor groepsverschillen op de lees- en spellingprestaties in de moedertaal (Nederlands). Door hiervoor te controleren werden zowel het effect van onderliggende vaardigheden in de moedertaal als mogelijke al aanwezige compensatiemechanismen in de moedertaal teniet gedaan. De vreemde talen die onderzocht werden zijn Engels, Frans en Duits. De resultaten toonden hetzelfde stapsgewijze patroon aan voor lees- en spellingvaardigheid in de *moedertaal* als eerder gevonden in het basisonderwijs (Hoofdstuk 4), namelijk; dyslexie < hoogbegaafd/dyslexie < normaal ontwikkelend < hoogbegaafd. Dit prestatiepatroon vertaalde zich ook naar woordlezen en spellen in het Engels, maar niet naar lees- en/of spellingvaardigheid in het Frans of Duits. De patronen voor woordlezen en spellen in het Engels bleven intact na het controleren voor lees- en spellingvaardigheden in de moedertaal. Betere Engelse woordlees- en spellingvaardigheden van hoogbegaafde leerlingen met dyslexie lijken het resultaat te zijn van additionele factoren of mechanismen die uniek zijn voor het Engels als tweede taal. De afwijkende bevindingen voor de verschillende moderne vreemde talen worden verder besproken in termen van variatie in orthografische transparantie en de hoeveelheid blootstelling.

Uit deze studies komt naar voren dat ondersteuning is gevonden voor een aantal basisaannamen van de *twice-exceptionality* benadering, namelijk de aanwezigheid van hogere lees- en spellingniveaus en zowel cognitieve risico- als protectieve factoren bij hoogbegaafde kinderen met dyslexie. Echter, het bewijs voor compensatie is beperkt en begrensd tot compensatiemechanismen in het Engels als tweede taal. De bevindingen in de moedertaal waren grotendeels in lijn met de *core-deficit* benadering van dyslexie. Mits zorgvuldig toegepast, heeft de *twice-exceptionality* benadering desalniettemin het potentieel om een nieuwe kijk op de rol van intelligentie bij de ontwikkeling en openbaring van leerstoornissen te bewerkstelligen in toekomstig onderzoek.

Samengevat geven de resultaten van de studies in deze dissertatie een verdere specificatie van de rol van vroege taalvaardigheid en FR als risicofactoren voor dyslexie en de invloed van hoogbegaafdheid als context voor potentiële protectieve factoren. Wat betreft risicofactoren bleek vroege verbale taalvaardigheid het fundament te vormen voor de leesontwikkeling. Daarnaast kan vroege taalvaardigheid beschouwd worden als een bijkomende maar kleine risicofactor van dyslexie voor kinderen die leren lezen in het Nederlands. Een FR is niet van invloed op de vroege verbale taalvaardigheid, maar heeft wel een effect op de leesontwikkeling vanaf het begin van ontluikende geletterdheid. Met betrekking tot protectieve factoren werd bevestigd dat hoogbegaafde kinderen met dyslexie hogere lees- en spellingniveaus hebben dan gemiddeld intelligente kinderen met dyslexie, zowel in het basis- als voortgezet onderwijs. Hun onderliggende cognitieve profiel bestaat uit zowel dyslexie-gerelateerde zwaktes als hoogbegaafdheid-gerelateerde sterktes. Deze sterktes functioneren mogelijk als protectieve factoren en zouden betrokken kunnen zijn bij compensatie. Echter, het gevonden bewijs voor compensatie was beperkt; de mogelijkheid van directe compensatie is uitgesloten, dat wil zeggen, er is geen sprake van moderatie van de grootte/sterkte van een risicofactor door een bijkomende protectieve factor. In plaats daarvan wezen de bevindingen meer in de richting van de ontwikkeling van compensatiemechanismen, vooral in de specifieke context van Engels als tweede taal. Tezamen benadrukken deze studies het belang van het in kaart brengen van zowel risico- als protectieve factoren tijdens diagnostisch onderzoek. De uitkomsten hiervan kunnen belangrijke aanwijzingen voor remediëring bevatten en helpen bij het samenstellen van een passende interventie.

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ABOUT THE AUTHOR

Sietske van Viersen was born on 4 November 1989 in Utrecht, The Netherlands. She completed secondary education at the Leidsche Rijn College in Utrecht in 2007. From September 2007 until February 2011 she studied Pedagogical Sciences at Utrecht University, specializing in Special Educational Needs. During this time, Sietske was an avid rower and coach at A.U.S.R. ORCA. In 2013, she completed her research master Educational Sciences: Learning in Interaction at Utrecht University by graduating cum laude. She combined this research master program with a clinical track, including an internship at the Ambulatorium in Utrecht. Her final thesis (supervised by dr. Evelyn Kroesbergen and dr. Elise de Bree) was a project about giftedness and dyslexia she designed from scratch. The thesis was nominated for the Peter G. Swanborn award (Utrecht University) for best research master's thesis.

Sietske worked as a teacher and Junior Researcher on several projects (including her own project about giftedness and dyslexia) at the Department of Cognitive and Motor Disabilities of Utrecht University. In April 2014 she started her PhD project at the Research Institute of Child Development and Education of the University of Amsterdam with prof. dr. Peter de Jong, dr. Elise de Bree, and dr. Evelyn Kroesbergen. For her research project on risk factors, protective factors, and possibilities for compensation in children with dyslexia, she visited the ARC Centre of Excellence in Cognition and its Disorders (CCD) at Macquarie University, Sydney, Australia. After submitting her dissertation in November 2016, she started as a Postdoctoral Researcher at the same department. During this postdoc, Sietske continues to work on the role of protective factors and compensation in dyslexia by assessing reading processes using eye tracking and participation in an international research project about orthographic learning. She also teaches various (clinical) courses in the College and Graduate School of Child Development and Education.

List of Publications

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