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The Only Way is Up - Risk Factors, Protective Factors and Compensation in Dyslexia.

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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.