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Who will develop dyslexia? Cognitive precursors in parents and children

van Bergen, E.

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The great tragedy of science – the slaying of a beautiful hypothesis by an ugly fact.

- Thomas Huxley -
Dutch children at family risk of dyslexia: Precursors, reading development, and parental effects

Abstract

The study concerns reading development and its precursors in a transparent orthography. Dutch children differing in family risk for dyslexia were followed from kindergarten through 5th grade. In 5th grade, at-risk dyslexic \( (n = 22) \), at-risk non-dyslexic \( (n = 45) \), and control children \( (n = 12) \) were distinguished. In kindergarten, the at-risk non-dyslexics performed better than the at-risk dyslexics, but worse than the controls on letter knowledge and rapid naming. The groups did not differ on phonological awareness. At-risk dyslexics read less fluently from 1st grade onwards than the other groups. At-risk non-dyslexics’ reading fluency was at an intermediate position between the other groups at the start of reading. By 5th grade they had reached a similar level as the controls on word reading, but still lagged behind on pseudoword reading. Results further showed that the parents of the groups of at-risk children differed in educational level and reading skills. Overall, the groups of at-risk children differed on pre-reading skills as well as on reading development. These differences do not seem to stem from differences in intellectual abilities or literacy environment. Instead, the better reading skills of parents of at-risk non-dyslexics suggest that these children might have a lower genetic liability.
2.1 Introduction

It is well established that reading and spelling problems of dyslexics are accompanied by deficits in phonological processing. Dyslexics’ impairments have been found in phonological awareness, verbal short-term memory, and rapid naming (see for reviews Elbro, 1996; Vellutino, Fletcher, Snowling, & Scanlon, 2004). Most of the evidence is based on studies in which the phonological deficits were examined after reading problems had become manifest. There is, however, a small number of studies that started before reading onset and continued to follow children until an age that reading problems could be established. Such longitudinal studies can provide stronger evidence for a causal relationship between deficits in phonological processing and the development of dyslexia.

Most prospective studies involved children who have a dyslexic parent and, hence, have a family risk of becoming dyslexic. Such studies provide the opportunity to examine the differential development of at-risk children who do and do not become dyslexic (hereafter called dyslexics and at-risk non-dyslexics, respectively), and controls (i.e., not at-risk non-dyslexics). The majority of studies concerned children learning to read in an opaque orthography. The first issue of importance in the present study is that it involved children learning to read in a relatively transparent orthography: Dutch. Several studies suggest that the differential importance of precursors of later reading success or failure is moderated by orthographies’ transparencies (de Jong & van der Leij, 2003; Puolakanaho et al., 2008; Wimmer, Mayringer, & Landerl, 2000). The second issue that is addressed in this study is the impact of parental reading status on their offspring’s reading success or failure. Although differences between the three groups on child characteristics are well documented, differences on parental characteristics have largely been neglected.

Several earlier prospective studies showed that phonological deficits were already present before reading started (Boets, Wouters, van Wieringen, & Ghesquière, 2007; Elbro, Borstrøm, & Petersen, 1998; Pennington & Lefly, 2001; Scarborough, 1990; Snowling, Gallagher, & Frith, 2003). For example, Elbro, Borstrøm, and Petersen (1998) followed the progress of Danish children with and without family risk from kindergarten through the beginning of second grade, when dyslexia was determined. In kindergarten, the at-risk dyslexics showed deficits as compared to the control group in phoneme awareness,
verbal short-term memory, and distinctness of phonological representations, whereas the at-risk non-dyslexics did not. In contrast, the at-risk non-dyslexics did perform as poorly as the at-risk dyslexics on morpheme awareness and articulation, whereas they performed better than the at-risk dyslexics but less well than the controls on letter knowledge.

Snowling and colleagues carried out a similar study with children learning to read in English (Gallagher, Frith, & Snowling, 2000; Snowling, Gallagher, & Frith, 2003; Snowling, Muter, & Carroll, 2007). Children's reading status was determined at the age of 8. At 3 years and 9 months, the at-risk non-dyslexics performed better than the at-risk dyslexics but poorer than the controls on non-word repetition and letter knowledge. The dyslexic, but not the non-dyslexic at-risk readers, exhibited early deficits in vocabulary, narrative skills, and verbal short-term memory. Snowling et al. (2003; 2007) concluded that the children with a family risk that did not become dyslexic had probably compensated for their deficits in grapheme-phoneme knowledge with strong oral language skills.

Pennington and Lefly (2001) also conducted an at-risk study with English-speaking children. Dyslexia was assessed in second grade. At the start of first grade, the at-risk dyslexics had, compared to both groups of future normal readers, impairments on emergent literacy skills and verbal short-term memory. The at-risk dyslexics also performed poorer on phoneme awareness and speech perception than at-risk non-dyslexics, who performed poorer than controls. Interestingly, Pennington and Lefly also assessed rapid naming skills, on which the at-risk dyslexics also scored weakest and the controls highest.

Generally, learning to read in an opaque orthography like Danish or English poses extra challenges to the beginning reader and might affect the various manifestations of dyslexia (Seymour, Aro, & Erskine, 2003; Ziegler et al., 2005). In a study of dyslexics (with and without risk combined), at-risk non-dyslexics, and controls in a relatively transparent orthography (Dutch), Boets et al. (2007; 2010) found that the dyslexic readers performed as kindergartners worse than the not at-risk non-dyslexic readers on letter knowledge, verbal short-term memory, phonological awareness and rapid naming. The performance of the at-risk non-dyslexic group was at an intermediate position between that of the other two groups, but did not differ significantly from either of them. These results are roughly in accordance with studies in opaque orthographies, but the results on phonological awareness are at odds with
findings that kindergarten phonological awareness predicts reading skills less well in transparent orthographies (de Jong & van der Leij, 2003; Puolakanaho et al., 2008; Wimmer, Mayringer, & Landerl, 2000).

Regardless of the subtle differences between orthographies in the cognitive profile of dyslexic readers, the question remains in what respect at-risk children that do not develop dyslexia differ from those that do, and especially, where these differences stem from. One possibility is that at-risk non-dyslexic children are able to compensate their impaired reading potential with other skills, as suggested by Snowling et al. (2003). Another possibility is that this group is at lower genetic risk. Finally, it could be that this group experiences more advantageous environmental factors.

Torppa et al. (2007) assessed the relation between home literacy, reading interest, phonological awareness, vocabulary, and emergent literacy variables. They found a direct effect of home-literacy environment (shared reading) on vocabulary growth, but not on emergent literacy. Similarly, Elbro et al. (1998) did not find differences between dyslexic and non-dyslexic parents, nor between parents of dyslexic and non-dyslexic children in the amount of shared reading in kindergarten. Finally, Snowling et al. (2007) did not find differences between the two at-risk groups in parent reading behavior, family literacy behavior, and socioeconomic background. Overall, there is no strong evidence that children who develop dyslexia grow up in a relatively disadvantageous literacy environment.

Dyslexia is commonly seen as a complex multifactorial disorder with numerous genes involved that interact with one another and with the environment (Bishop, 2009). Each gene affects the probability of the disorder, but is in itself not necessary and sufficient for causing it. The involvement of multiple genes may function as a normally distributed genetic liability (Rutter, 2006; van den Oord, Pickles, & Waldman, 2003): as more genes are affected the probability increases that dyslexia becomes manifest. A multifactorial etiological conceptualization resulting from a normally distributed genetic liability is in accordance with the view from behavioral studies (Snowling, Gallagher, & Frith, 2003) that the family risk of dyslexia is continuous. This model may be useful to speculate about the question why some at-risk children develop dyslexia whereas others do not.

Surprisingly, possible differences in reading ability between the dyslexic parents of the two at-risk groups have been largely neglected. Such
differences might suggest that at-risk dyslexic children have a higher genetic liability than at-risk non-dyslexics. As an exception, Snowling et al. (2007) reported that the parents of at-risk dyslexics did not differ from the parents of at-risk non-dyslexics on spelling and word-reading accuracy. However, it should be noted that in these analyses the pairs of parents were divided according to gender instead of according to reading status. Furthermore, within the at-risk group the literacy levels of the children and their parents were not associated. It is of interest whether similar results will be found for word-reading fluency or rate. Word-reading fluency is seen as the most important feature of reading ability in transparent orthographies (Wimmer, 2006).

In conclusion, studies following children at-risk of dyslexia indicate that pre-readers who become dyslexic show early phonological deficits and broader language difficulties. In addition, at-risk non-dyslexics are not completely unaffected but show a milder phenotype. What is missing, though, are long-term studies in orthographically transparent languages. In addition, attention should be devoted to the reading skills of the parents.

In our study, the literacy development of Dutch children with and without family risk for dyslexia was followed from kindergarten through fifth grade. In fifth grade groups of at-risk dyslexics, at-risk non-dyslexics, and controls were distinguished. Next we considered, retrospectively, differences in the development of (pre-)reading skills among these groups. In addition, differences in parental characteristics and home-literacy environment were examined.

2.2 Method

2.2.1 Participants
Twenty-two children were included in the at-risk dyslexic group and 45 in the at-risk non-dyslexic group. The control group included 12 not at-risk non-dyslexic children (see Table 2.1).

The children were recruited earlier (Regtvoort & van der Leij, 2007; van Otterloo & van der Leij, 2009). The original sample consisted of 98 children (82 at-risk). None of the parents reported (suspicion of) developmental disorders. In addition, the children were recruited from regular education. In the Netherlands children with developmental disorders are usually referred to special education. Fifty-nine at-risk children received intervention before
they received formal reading instruction. The intervention group showed a beneficial effect on phonological awareness and letter knowledge at the end of kindergarten, but this head start did not lead to better literacy levels in first grade. The only measurement occasion with an intervention effect present (i.e., end of kindergarten) was not included in the present study.

<table>
<thead>
<tr>
<th>Characteristic</th>
<th>At-risk dyslexic</th>
<th>At-risk non-dyslexic</th>
<th>Control</th>
<th>F (2, 76)</th>
<th>p</th>
<th>η²_p</th>
</tr>
</thead>
<tbody>
<tr>
<td>Sample size</td>
<td>22</td>
<td>45</td>
<td>12</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>No (%) of boys</td>
<td>16 (73%)</td>
<td>28 (62%)</td>
<td>6 (50%)</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>No (%) of children in intervention</td>
<td>14 (64%)</td>
<td>32 (71%)</td>
<td>0 (0%)</td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

Note: Characteristics are given in raw scores, standard deviations in parentheses. Numbers and means in the same row that do not share subscripts differ at p < .05 on the χ²-test and the Tukey honestly significant difference comparison. No = number; KG = kindergarten; Gr = Grade.

Children were considered at-risk if at least one of the parents reported to be dyslexic and if this was confirmed by tests measuring word and pseudoword-reading fluency (Brus & Voeten, 1972; van den Bos, Luifje Spelberg, Scheepstra, & de Vries, 1994). Also, the subtest similarities of the Wechsler Adult Intelligence Scale was administered to measure verbal reasoning (Wechsler, 1997). For inclusion of a child in the at-risk group, at least one parent had to score below or at the 20th percentile on both reading tests, or below or at the 10th percentile on one of them (with the other ≤ 40th percentile). Children of parents who showed a large discrepancy (≥ 60 percentiles) between verbal reasoning and one of the reading-tests were also included, with the restriction that neither reading-test percentile score exceeded 40. Of not at-risk children both parents had to score ≥ 40th percentile on both reading tests.

1 T-tests (comparing at-risk children with and without kindergarten intervention) on January Grade 1 measures showed no significant differences: for letter-naming fluency, mean difference = 0.008, t(65) = 0.13, p = .896; for word-reading fluency, mean difference = -1.56, t(65) = -0.67, p = .504.

2 There were 14 at-risk children for whose parents only the discrepancy criterion applied. Excluding this group of children did not alter the pattern of results.
One of the 16 not at-risk and 15 of the 82 at-risk children dropped out of the study. Within the at-risk group, children who dropped out had a smaller vocabulary than the others, \( t(80) = 2.21, p = .030 \), but they did not differ in age, \( t(80) = 1.59, p = .146 \). Their parents did not differ on word-reading fluency, \( t(80) = -0.71, p = .481 \), pseudoword-reading fluency, \( t(80) = -0.95, p = .346 \), and verbal reasoning, \( t(80) = -1.05, p = .298 \). The attrition rate in the not at-risk group was too small to do group comparisons. One not at-risk child was excluded from the study because an elder sibling was diagnosed as dyslexic. The child itself also became dyslexic.

Reading status was determined in fifth grade (at a mean age of 10 years and 10 months). Children with a reading score on a word-reading fluency task (WRF2, see below) corresponding to the weakest ten percent in the population were considered dyslexic. The not at-risk dyslexic group consisted of only two children and was excluded in the study. The remaining three groups did not differ on sex, age, vocabulary, and nonverbal IQ (Table 2.1). The proportion of children in the at-risk groups who had received intervention in kindergarten did not differ.

### 2.2.2 Measures

Measures were selected to investigate intelligence, phonological awareness, letter knowledge, rapid naming, and (pseudo)word-reading skills. Test reliabilities ranged from .73 to .97.

**Intelligence**

**Vocabulary.** In the receptive vocabulary test (Verhoeven & Vermeer, 1996) the child had to choose among four alternatives the picture that best matched a given word. The 98 items were of increasing difficulty. Administration was stopped when the child failed six out of the last eight items.

**Nonverbal IQ.** Coloured Progressive Matrices (J. C. Raven, Court, & Raven, 1984) were used to measure nonverbal IQ.

**Home-literacy environment**

Parents were given a short questionnaire when their child attended kindergarten. They were asked to indicate whether their child was a library member, to estimate their shared reading frequency on a scale from 1 (*never*) to 5 (*more than five times a week*) and the number of books they had at home scaled from 1 (*less than 50*) to 5 (*over 200*). Next, the level of education of both parents was ranked on a scale ranging from 1 (*primary school only*) to 7 (*university degree*).
Phonological Awareness

*Alliteration* (Irausquin, 2001). The word with a different onset sound from three words had to be named (‘odd one out’). All words had a consonant-vowel-consonant (CVC) structure. The experimenter named the three words and repeated them when necessary. There were 3 practice items (with feedback) and 10 test items, all of approximately equal difficulty.

*Phoneme blending and segmentation.* In phoneme-blending (Verhoeven, 1993a) the child was required to blend aurally presented phonemes into a word. For example, children listened to the successive phonemes /r/ /u/ /p/ /s/, after which they had to merge this into /rups/ (caterpillar). Phoneme-segmentation (Verhoeven, 1993b) was the reverse of phoneme-blending. Now the child had to segment a given word into its constituent phonemes. Both tasks began with three practice trials (with feedback). Test items consisted of 20 monosyllabic words per test, increasing from two to five phonemes, with four to six items per number of phonemes. The tests were stopped when all items with the same number of phonemes were blended or segmented incorrectly.

Rapid Naming

Serial rapid naming (van den Bos, 2003) was measured for colors (black, yellow, red, green, and blue) and objects (bike, tree, duck, scissors, and chair). Each of the tasks consisted of 50 randomly ordered symbols arranged in five columns of 10 symbols each. Before test administration, the child practiced by naming the last column of symbols. Children were instructed to name the symbols column-wise as quickly as possible. For both tasks, the time to completion was transformed to number of symbols per second to normalize the score distribution.

Letter Knowledge

*Receptive letter-knowledge.* The test (Verhoeven, 2002) required the child to point from six alternative lowercase letters to the letter that matched a given sound. For instance, the child was asked “Where do you see the /m/ of mooi (beautiful)?” The knowledge of 32 graphemes (including digraphs) was tested.

*Letter-naming fluency.* Children were asked to provide the sound of 34 graphemes (including digraphs) correctly as quickly as possible (Verhoeven, 1993a). When the child gave letter names instead, the child was corrected and the test was started once again. The randomly ordered graphemes were printed in lowercase in two columns of 17 items each. Scores were transformed to number of correctly-read letters per second.
Reading Skills

Word-reading fluency (WRF). This was measured as how many disconnected words one can read correctly within a certain time allowed. Three lists were used (Verhoeven, 1995; Verhoeven & van Leeuwe, 2009). The first and second list (WRF1 and WRF2, respectively) consisted of 150 monosyllabic words each. WRF1 included words with a CV, VC, or CVC pattern. All words in WRF2 contained at least one consonant cluster. WRF3 comprised 120 polysyllabic words with various orthographic complexities. The words had to be read correctly as quickly as possible. The score per list was the number of words read correctly within one minute. WRF2 was used to assess dyslexia in Grade 5, because it differentiates best between normal and dyslexic readers.

Pseudoword-reading fluency. The test (van den Bos, Sutje Spelberg, Scheepstra, & de Vries, 1994) consisted of a list of 116 pseudowords of increasing difficulty. The child was asked to correctly read aloud as many pseudowords as possible within two minutes.

2.2.3 Procedure
Tests were administered individually in a fixed order by trained graduate students in a separate room at school, in January of the second kindergarten year, in January and June of Grade 1, June of Grade 2, and January of Grade 5.

2.2.4 Analytic Approach
One-way analyses of variance (ANOVAs) and χ²-tests were used to compare groups. Contrasts were evaluated post-hoc with Tukey’s procedure to correct for multiple testing. When the same ability was measured on several occasions, repeated-measures ANOVAs were performed with Group as between subject-factor and Time as within-subject factor.

2.3 Results

2.3.1 Parent Characteristics and Home-Literacy Environment
The characteristics of the children’s parents and home environment are presented in Table 2.2. In both at-risk groups, almost 75% of the children had a dyslexic father. Regarding the level of education for mothers and fathers, control families had the highest level whereas the lowest level was found in
Table 2.2 Characteristics of the Parents and Home Environment from the Three Groups, with $\chi^2$ and ANOVA Results

<table>
<thead>
<tr>
<th>Characteristic</th>
<th>At-risk dyslexic</th>
<th>At-risk non-dyslexic</th>
<th>Control</th>
<th>$F$</th>
<th>df</th>
<th>$p$</th>
<th>$\eta^2_p$</th>
</tr>
</thead>
<tbody>
<tr>
<td>Parent characteristics</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>No (%) of dyslexic fathers</td>
<td>16 (73%)</td>
<td>32 (71%)</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Level of education of</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Mother</td>
<td>4.32 (1.43)</td>
<td>5.07 (1.57)</td>
<td>5.92 (1.31)</td>
<td>4.58 (2, 76)</td>
<td>.013</td>
<td>.108</td>
<td></td>
</tr>
<tr>
<td>Father</td>
<td>3.86 (1.96)</td>
<td>4.91 (1.85)</td>
<td>6.17 (1.53)</td>
<td>6.27 (2, 76)</td>
<td>.003</td>
<td>.142</td>
<td></td>
</tr>
<tr>
<td>Dyslexic parent</td>
<td>4.00 (1.98)</td>
<td>4.73 (1.83)</td>
<td>2.24 (1, 65)</td>
<td>2.24 (1, 65)</td>
<td>.139</td>
<td>.033</td>
<td></td>
</tr>
<tr>
<td>Non-dyslexic parent</td>
<td>4.18 (1.44)</td>
<td>5.24 (1.55)</td>
<td>7.25 (1, 65)</td>
<td>7.25 (1, 65)</td>
<td>.009</td>
<td>.100</td>
<td></td>
</tr>
<tr>
<td>Verbal reasoning</td>
<td>17.50 (5.20)</td>
<td>16.22 (4.78)</td>
<td>18.42 (4.14)</td>
<td>1.13 (2, 76)</td>
<td>.085</td>
<td>.063</td>
<td></td>
</tr>
<tr>
<td>Word-reading fluency</td>
<td>60.41 (11.96)</td>
<td>69.00 (13.61)</td>
<td>97.83 (11.08)</td>
<td>34.33 (2, 76)</td>
<td>&lt; .001</td>
<td>.475</td>
<td></td>
</tr>
<tr>
<td>Pseudoword-reading fluency</td>
<td>38.77 (15.32)</td>
<td>53.22 (17.85)</td>
<td>101.75 (8.45)</td>
<td>61.55 (2, 76)</td>
<td>&lt; .001</td>
<td>.618</td>
<td></td>
</tr>
<tr>
<td>Home-literacy environment</td>
<td></td>
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<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>No (%) of library members</td>
<td>16 (73%)</td>
<td>32 (71%)</td>
<td>6 (50%)</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Shared reading</td>
<td>4.05 (1.29)</td>
<td>4.39 (0.91)</td>
<td>4.83 (0.39)</td>
<td>2.55 (2, 76)</td>
<td>.085</td>
<td>.063</td>
<td></td>
</tr>
<tr>
<td>Books at home</td>
<td>3.05 (1.53)</td>
<td>3.31 (1.49)</td>
<td>4.00 (1.65)</td>
<td>1.54 (2, 76)</td>
<td>.220</td>
<td>.039</td>
<td></td>
</tr>
</tbody>
</table>

Note. Parental reading fluency and reasoning are scores of the weakest-reading parent. Numbers and means in the same row that do not share subscripts differ at $p < .05$ on the $\chi^2$-test and the Tukey honestly significant difference comparison. No = number.
Table 2.3 Descriptive Statistics and ANOVA Results for the Measures of Phonological Awareness, Rapid Naming, and Letter Knowledge

<table>
<thead>
<tr>
<th>Measure</th>
<th>At-risk dyslexic</th>
<th>At-risk non-dyslexic</th>
<th>Control</th>
<th>F (2, 76)</th>
<th>p</th>
<th>η²_p</th>
</tr>
</thead>
<tbody>
<tr>
<td>Phonological awareness (Jan. KG)</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Alliteration</td>
<td>4.15_s 2.99</td>
<td>4.53_s 3.08</td>
<td>4.83_s 3.10</td>
<td>0.21</td>
<td>.808</td>
<td>.006</td>
</tr>
<tr>
<td>Phoneme blending</td>
<td>6.18_s 6.63</td>
<td>7.27_s 6.15</td>
<td>7.67_s 6.51</td>
<td>0.29</td>
<td>.748</td>
<td>.008</td>
</tr>
<tr>
<td>Phoneme segmentation</td>
<td>4.86_s 4.84</td>
<td>5.22_s 5.35</td>
<td>4.58_s 5.55</td>
<td>0.09</td>
<td>.918</td>
<td>.002</td>
</tr>
<tr>
<td>Rapid naming (Jan. KG)</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Colors</td>
<td>0.48_s 0.15</td>
<td>0.63_b 0.18</td>
<td>0.74_b 0.21</td>
<td>9.85</td>
<td>&lt;.001</td>
<td>.206</td>
</tr>
<tr>
<td>Objects</td>
<td>0.53_s 0.14</td>
<td>0.62_ab 0.17</td>
<td>0.74_b 0.17</td>
<td>6.72</td>
<td>.002</td>
<td>.150</td>
</tr>
<tr>
<td>Letter knowledge</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Receptive (Jan. KG)</td>
<td>10.36_s 4.86</td>
<td>14.42_s 6.70</td>
<td>18.00_s 5.94</td>
<td>6.51</td>
<td>.002</td>
<td>.146</td>
</tr>
<tr>
<td>Naming fluency (Jan. Gr.1)</td>
<td>0.82_s 0.21</td>
<td>0.96_b 0.24</td>
<td>1.18_s 0.23</td>
<td>9.52</td>
<td>&lt;.001</td>
<td>.200</td>
</tr>
<tr>
<td>Naming fluency (June Gr.1)</td>
<td>1.01_s 0.26</td>
<td>1.24_b 0.28</td>
<td>1.24_b 0.29</td>
<td>5.72</td>
<td>.005</td>
<td>.131</td>
</tr>
</tbody>
</table>

Note: Maximum scores are 10, 20, and 20 for the phonological-awareness tasks, and 32 for the receptive letter-knowledge task. Jan. = January. Numbers and means in the same row that do not share subscripts differ at p < .05 on the Tukey honestly significant difference comparison. KG = kindergarten; Gr = Grade.

The families of the at-risk dyslexics. Within at-risk families, we also investigated level of education for dyslexic and non-dyslexic parents. An ANOVA (with Group as between-subject and Parent as within-subject factor) revealed that the parents of the at-risk non-dyslexics had a higher educational level than those of the at-risk dyslexics (main effect of Group: $F(1, 65) = 5.26, p = .025, \eta^2_p = .075$). The main effect of Parent ($F(1, 65) = 2.73, p = .103, \eta^2_p = .040$) and the interaction effect ($F < 1$) were not significant. In contrast to level of education, no differences were found for parental verbal reasoning. Interestingly, reading fluency of the parents of at-risk dyslexics was significantly lower than of at-risk non-dyslexics for both words and pseudowords. This finding was further endorsed by a correlation of $.27 (p = .026)$ between reading fluency scores (a composite of words and pseudowords) of the dyslexic parents and their offspring.

With respect to home-literacy environment in kindergarten (Table 2.2), the proportion of children who were member of the library did not differ significantly among the groups, nor did the groups differ significantly in the amount of shared reading. Finally, the Group effect was not significant for the number of books at home.


2.3.2 Child Characteristics

Pre-Reading Skills

The scores on the measures of phonological awareness, rapid naming, and receptive letter knowledge (Table 2.3) were subjected to a multivariate analysis of variance (MANOVA). The three groups differed significantly: Wilks’ λ = .668, F (12, 142) = 2.65, p = .003, η² = .183. The univariate results revealed that the groups did not differ on phonological-awareness tasks. Conversely, differences were found on rapid naming of colors and objects, and on letter knowledge. The at-risk dyslexic children were slower on rapid naming and knew fewer letters than the controls. The at-risk non-dyslexics also seemed to perform more poorly than controls, but this did not reach significance at the (conservative) Tukey post-hoc comparisons.

Reading Development

Letter-naming fluency (Table 2.3) was measured at the middle and end of first grade. There was a main effect of both Time, F (1, 76) = 21.23, p < .001,

<table>
<thead>
<tr>
<th>Measure</th>
<th>At-risk dyslexic</th>
<th>At-risk non-dyslexic</th>
<th>Control</th>
</tr>
</thead>
<tbody>
<tr>
<td>Words-WRF1</td>
<td>11.14 ± 4.52</td>
<td>20.76 ± 8.62</td>
<td>37.75 ± 15.02</td>
</tr>
<tr>
<td>Words-WRF2</td>
<td>22.68 ± 11.12</td>
<td>41.42 ± 15.48</td>
<td>58.33 ± 15.20</td>
</tr>
<tr>
<td>Words-WRF3</td>
<td>10.68 ± 7.10</td>
<td>26.78 ± 13.17</td>
<td>43.92 ± 17.18</td>
</tr>
<tr>
<td>Pseudowords</td>
<td>6.77 ± 7.10</td>
<td>16.58 ± 9.44</td>
<td>29.67 ± 11.91</td>
</tr>
<tr>
<td>Grade 2, June</td>
<td>11.32 ± 8.91</td>
<td>20.82 ± 9.71</td>
<td>36.58 ± 11.91</td>
</tr>
<tr>
<td>Words-WRF1</td>
<td>42.09 ± 17.10</td>
<td>66.29 ± 15.30</td>
<td>77.58 ± 11.81</td>
</tr>
<tr>
<td>Words-WRF2</td>
<td>30.18 ± 16.57</td>
<td>65.63 ± 16.48</td>
<td>72.50 ± 12.52</td>
</tr>
<tr>
<td>Words-WRF3</td>
<td>19.05 ± 13.34</td>
<td>41.33 ± 16.40</td>
<td>52.17 ± 12.81</td>
</tr>
<tr>
<td>Pseudowords</td>
<td>19.41 ± 11.27</td>
<td>35.64 ± 12.86</td>
<td>47.50 ± 8.93</td>
</tr>
<tr>
<td>Grade 5, January</td>
<td>63.45 ± 15.78</td>
<td>93.04 ± 12.44</td>
<td>95.67 ± 13.21</td>
</tr>
<tr>
<td>Words-WRF1</td>
<td>55.00 ± 16.20</td>
<td>89.76 ± 11.48</td>
<td>93.25 ± 12.68</td>
</tr>
<tr>
<td>Words-WRF2</td>
<td>43.82 ± 16.70</td>
<td>75.71 ± 12.05</td>
<td>80.75 ± 11.15</td>
</tr>
<tr>
<td>Pseudowords</td>
<td>36.55 ± 15.94</td>
<td>61.87 ± 14.35</td>
<td>78.50 ± 11.71</td>
</tr>
</tbody>
</table>

Note. The maximum scores for the word-reading fluency (WRF)-lists are 150, 150, and 120, respectively; for pseudoword-reading 116. Numbers and means in the same row that do not share subscripts differ at p < .05 on the Tukey honestly significant difference comparison.
Figure 2.1. Development of reading. First panel: words (i.e., WRF1); second panel: pseudowords.
Dutch children at family risk of dyslexia

η²ₚ = .218, and Group, F (2, 76) = 9.71, p < .001, η²ₚ = .203. The Time by Group interaction approached significance, F(2, 76) = 2.59, p = .082, η²ₚ = .064. Pairwise comparisons revealed that halfway through Grade 1 the at-risk non-dyslexics were significantly slower than the controls, but significantly faster than the at-risk dyslexics. By the end of the school year, this group had reached the same level as the controls.

The statistics for reading fluency are presented in Table 2.4. In addition, the results for WRF1 and pseudowords are presented graphically in Figure 2.1. On all word and pseudoword reading tests, the main effects of both Group and Time, as well as their interaction were significant (ps < .001, results available on request). On all word and pseudoword reading tests and on all occasions, the at-risk dyslexic group lagged significantly behind the other two. Moreover, the performance on both word and pseudoword reading of the at-risk non-dyslexics was significantly below that of controls in first and second grade. In Grade 5, however, the at-risk non-dyslexics still read pseudowords significantly less fluently than the controls, but performed at the same level on word reading.

2.4 Discussion

The reading development of children born to families with or without a history of dyslexia was followed from kindergarten to fifth grade. In fifth grade reading status (dyslexic or not dyslexic) was determined. Then, the development of early reading-related abilities and reading in the groups differing in family-risk status and reading status were compared.

In kindergarten, we found that the amount of letter-knowledge and rapid naming differed among the groups of at-risk dyslexic, at-risk non-dyslexic, and not at-risk normal-reading children (controls). However, differences among the groups in the development of phonological awareness were not found.

The finding that the level of phonological awareness in kindergarten was similar in dyslexic and normal reading children is in accordance with results reported by de Jong and van der Leij (2003). They also did not find differences between groups of kindergartners that later did or did not develop dyslexia. De Jong and van der Leij attributed this null-result to Dutch kindergartners’ low level of phonological awareness (Wesseling & Reitsma, 1998). Apparently,
as long as instruction pertaining to phonological awareness is sparse, there are no group differences (de Jong & van der Leij, 1999).

However, in a recent prospective study, Boets et al. (2007; 2010) did find deficits in a Dutch speaking dyslexic group (with and without risk combined) relative to controls on phonological awareness assessed in kindergarten. There are a number of reasons that might account for the different findings on phonological awareness between the current study and the study of Boets et al. (2010). To start with, it should be acknowledged that most of the phonological-awareness performance in our study was at floor level, which makes it difficult to detect differences. Another possibility concerns the moment of assessing reading status. Boets et al. assessed reading status at the start of third grade, whereas we assessed it mid-fifth grade. It is plausible that, in general, groups will differ more on a correlate of reading when the time span between the assessment of the correlate and the categorization into reading groups is shorter. Indeed, when we repeated the analyses with the classification based on reading scores at the end of second grade, the group differences on phonological awareness were larger, although still not significant. In addition, the relationship of phonological awareness and reading might change over time. Vaessen and Blomert (2010) showed that the relation between phonological awareness and word-reading fluency decreases as a function of reading experience, so assessing reading status earlier might lead to groups that differ more in kindergarten phonological awareness.

In contrast to phonological awareness, risk status and reading outcome were related to the kindergartners’ knowledge of letters, as well as their ability to rapidly name colors and objects. In line with previous studies (Pennington & Lefly, 2001; Snowling, Gallagher, & Frith, 2003), rapid naming and letter knowledge were impaired in the at-risk children who were later classified as dyslexic, whereas the at-risk children who were later classified as normal readers seemed to show milder impairments. These outcomes support the notion that it is rapid naming rather than phonological awareness that discriminates between reading groups in transparent orthographies, as dyslexics in transparent orthographies are characterized by sufficient reading accuracy but deficient reading fluency (de Jong & van der Leij, 2003; Georgiou, Parrila, & Papadopoulos, 2008; Landerl & Wimmer, 2008). Since the genes contributing to the manifestation of phonological awareness and rapid naming seem to be only partially shared (Naples, Chang, Katz, & Grigorenko,
2009), a consequence of this notion is that the set of genes involved in the etiology of dyslexia might differ somewhat between language environments that differ in their orthography’s transparency. At present, this intriguing idea remains speculative and warrants further investigation.

After half a year of reading instruction, the at-risk dyslexics were slower in the naming of letters compared to the at-risk non-dyslexics, who were slower than the controls. By the end of first grade the future dyslexics still lagged behind, but the at-risk non-dyslexics performed at the level of controls. The development of word-reading fluency showed a similar pattern, although over a longer time period. Throughout the primary-school years, at-risk children later classified as dyslexic read less fluently than the other two groups. The group of at-risk non-dyslexics performed better than the at-risk dyslexics but poorer than the controls in first and second grade, but in fifth grade they had closed the gap and reached the same level of word reading as the controls. Meanwhile, the group differences in pseudoword-reading fluency were stable over the five years studied, with the at-risk dyslexics performing less well and the controls better than the at-risk non-dyslexics at all times.

Taken together, the results of the development of letter knowledge and reading suggest that the at-risk non-dyslexics are genetically affected by their dyslexic parent. Although they become normal readers in the long run, they show subclinical deficits: first in pre-reading skills and later in reading itself. These findings at the behavioral level are in line with an earlier report on the present sample, showing atypical brain functioning during the processing of simple visual stimuli at the age of five in the at-risk non-dyslexics (Regtvoort, van Leeuwen, Stoel, & van der Leij, 2006). Recently, a study of Finnish researchers revealed atypical processing of simple auditory stimuli of at-risk non-dyslexics at birth (Leppänen et al., 2010). Despite these subtle deficits of at-risk non-dyslexics, they catch-up with the controls on letter-naming and word-reading fluency. Together with the finding that this group remains to lag behind in pseudoword reading, these results might be taken to suggest that at-risk non-dyslexics need more exposure to letters and words to reach the same level as the controls. As a related possibility, it could be that this group has a larger orthographic competence, the ability to store associations between written and spoken word forms at the word and subword level (Bekebrede, van der Leij, & Share, 2009), than their at-risk dyslexic peers. At the beginning of learning to read differences between the at-risk dyslexic and non-dyslexic groups are
relatively small, as word reading is highly dependent on phonological recoding. However, as reading development proceeds and orthographic competence becomes more important, at-risk non-dyslexic children outperform dyslexic children and reach, though somewhat later, a similar level of word reading as the controls.

The main focus of our study is to elucidate why some of the at-risk children, about one-third, develop dyslexia whereas others with a similar family background do not. Snowling et al. (2003) suggest that this latter group compensates their phonological deficits with a higher general cognitive ability or with good oral language skills. The current study does not provide support for this explanation, as the groups neither differed in nonverbal IQ nor in vocabulary. Likewise, there was no evidence of differences in nonverbal IQ and vocabulary between the two at-risk groups in the studies of Elbro et al. (1998) and Pennington and Lefly (2001). Still, we do not rule out that mild impairments could be observed when broader oral language skills are assessed.

Similar to previous studies (Elbro, Borstrøm, & Petersen, 1998; Snowling, Muter, & Carroll, 2007), we also did not find evidence for the second possible explanation that the at-risk non-dyslexic children experience a more advantageous literacy environment compared to the at-risk dyslexic children, though admittedly none of the studies (including ours) measured the environment thoroughly (Torppa et al., 2007). However, in the present study the parents of the non-dyslexic at-risk children had a higher level of education compared to those of the dyslexic children, which has not been found before. It is ambiguous whether this indicates a difference in the quality of their offspring’s home literacy environment and/or a difference in their offspring’s genetic potential.

A major finding of the current study is that within the group of dyslexic parents, those whose child developed dyslexia performed worse in both word and pseudoword reading. This might suggest that parents with more severe dyslexia are genetically more affected and have offspring at higher genetic risk, which fits with a continuous view of the family risk of dyslexia. Although individual differences in parental reading skills cannot be attributed to genetic influences alone, we suggest that it could be seen as an indication of their offspring’s genetic liability for reading failure. The present finding needs replication, but it shows that examining the relation between children’s reading outcome and parents’ literacy levels is intriguing in itself.
One of the limitations of our study is that our control group is rather small. However, the focus of this study is the difference between the two at-risk groups which are of substantial size. Moreover, our finding that the control group outperforms the at-risk groups in letter knowledge, rapid naming, and reading skills is consistent with previous longitudinal at-risk studies (Boets, Wouters, van Wieringen, & Ghesquière, 2007; Elbro, Borstrøm, & Petersen, 1998; Pennington & Lefly, 2001; Scarborough, 1990; Snowling, Gallagher, & Frith, 2003). Furthermore, the small effect sizes on differences in phonological-awareness suggest that the non-significant results can hardly be attributed to a lack of power.

It should be noted that about two-thirds of the at-risk children received intervention to promote literacy development during kindergarten, which might hamper interpretation of the data for the current aim. But although there were intervention effects observable directly after the intervention had finished, the differences between the trained and untrained at-risk children vanished when all children started formal reading instruction in first grade (Regtvoort & van der Leij, 2007; van Otterloo & van der Leij, 2009). The absence of long-term intervention effects is also shown in a meta-analysis of Bus and van Uzendoorn (1999). Finally, it is reassuring that the children who had received intervention spread equally over the dyslexic and non-dyslexic groups.

In sum, our data show that about two-third of the children born to families with a history of dyslexia acquire in the end adequate word-reading skills, despite mild impairments in rapid naming and letter knowledge in kindergarten and mild difficulties in pseudoword reading throughout middle childhood. The profile of reading and reading-related skills of the at-risk dyslexic group indicates stable and persistent impairments. Overall, our findings suggest that it is not likely that at-risk children who do and do not develop dyslexia differ in their intellectual abilities or literacy environment. Rather, they might differ in genetic liability.
2.5 References


