Who will develop dyslexia? Cognitive precursors in parents and children

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Citation for published version (APA):
I wish you to gasp not only at what you read
But at the miracle of its being readable.

- *Vladimir Nabokov* -
Child and parental literacy levels within families with a history of dyslexia

Abstract

Background: The present study concerns literacy and its underlying cognitive skills in Dutch children who differ in familial risk (FR) for dyslexia. Previous studies with FR-children were inconclusive regarding the performance of FR-children without dyslexia as compared to the controls. Moreover, van Bergen et al. (2011) recently showed that FR-children with and without dyslexia differed in parental reading skills, suggesting that those who go on to develop dyslexia have a higher liability. The current study concerned 1) the comparison of three groups of children at the end of 2nd grade and 2) the intergenerational transfer of reading and its underlying cognitive skills from parent to child.

Method: Three groups of children were studied at the end of 2nd grade: FR-dyslexia (n = 42), FR-no-dyslexia (n = 99), and control children (n = 66). Parents and children were measured on naming, phonology, spelling, and word and pseudoword reading.

Results: The FR-dyslexia children were severely impaired across all tasks. The FR-no-dyslexia children performed better than the FR-dyslexia children, but still below the level of the controls on all tasks; the only exception was RAN, on which they were as fast as the controls. Focusing on the FR sub-sample, parental reading and RAN were related to their offspring’s reading status.

Conclusions: We replicated and extended van Bergen et al’s study in showing that the FR-children who develop dyslexia are likely to have a higher liability. Both the group comparisons and the parent-child relations highlight the importance of good RAN skills for reading acquisition.
Literacy Levels within Families with a History of Dyslexia

3.1 Introduction

There is a fair amount of evidence to support the observation that dyslexia tends to run in families. In studies with children at familial risk (FR) of dyslexia, i.e. children with at least one dyslexic parent, approximately 33% to 66% have been reported to become dyslexic (Boets et al., 2010; Pennington & Lefly, 2001; Scarborough, 1990; Snowling, Gallagher, & Frith, 2003; Torppa, Lyytinen, Erskine, Eklund, & Lyytinen, 2010; van Bergen et al., 2011). In contrast, the percentage of dyslexic children of parents without dyslexia has been found to range from 6% to 16%. In combination with the results from quantitative genetic studies (e.g., Olson, Byrne, & Samuelsson, 2009; Pennington & Olson, 2005), such results suggest a strong hereditary basis of dyslexia.

Reading ability and disability is the end product of the effects of many genes in interaction with the environment. From this multifactorial etiology it follows that the underlying liability (see Falconer, 1965) for reading failure is continuously distributed. Plomin, DeFries, McClearn and McGuffin (2008, p. 33) show that the involvement of only a few genes already lead to continuous variation at the phenotypic level. From a liability continuum two propositions can be deduced. Firstly, children with a FR of dyslexia but without dyslexia themselves (FR non-dyslexics) have a higher position on the liability continuum than control children. That is, they inherit and/or experience more risk factors and therefore are expected to show weaknesses in literacy and its underlying cognitive skills. Secondly, within the group of FR children there might be a difference in liability to dyslexia between children with and without dyslexia. Insight into this liability might be gained by the skills of their parents. The current study explores both of these propositions.

3.1.1 FR Children Without Dyslexia

Several FR-studies demonstrated that not only FR children who went on to become dyslexic were impaired on precursors of reading. Those who became normal readers performed in general better than the FR-dyslexia children, yet poorer than the controls. This is in accordance with a continuum of liability to dyslexia, in which FR-no-dyslexia children have a higher liability than controls. However, there are only a few studies in which groups of FR-dyslexia, FR-no-dyslexia, and control children were compared on reading and its underlying cognitive skills after a few years of reading instruction. The first focus of the
current study was on the differences among these three groups when the children were 8 years of age, that is, after two years of formal reading instruction. Specifically, the emphasis was on the status of the FR-no-dyslexia children as compared to the controls.

Previous studies have provided a fairly clear picture of the performance of the groups regarding literacy skills, but nevertheless, they have been inconclusive regarding the cognitive skills underlying reading. Underlying skills are of interest because they represent endophenotypes, that is, the layer between the genotype and the phenotype of the reading behavior itself (Snowling, 2008). The most important cognitive skills underlying reading are assumed to be rapid naming (RAN) and phonological awareness (PA) (Snowling & Hulme, 2000). RAN is the rapid naming of highly familiar visual symbols and PA refers to the ability to identify and manipulate the sounds in spoken words. Verbal short-term memory is sometimes seen as a third underlying cognitive skill (Boets et al., 2010), but in literate individuals it tends to be closely tied to PA (de Jong & van der Leij, 1999), which suggests that both measures tap phonological processing.

Comparing at age 8 (i.e., after two years of reading instruction) FR-children classified as dyslexic with controls, studies report pronounced difficulties in reading, spelling, PA, verbal short-term memory, and RAN (Boets et al., 2010; de Bree, Wijnen, & Gerrits, 2010; Pennington & Lefly, 2001; Snowling, Muter, & Carroll, 2007). These studies did not agree, however, on whether the two groups of normal readers differ from each other. It is particularly intriguing that the FR-children classified as non-dyslexic showed mild impairments relative to the controls on reading and spelling, while the findings regarding the difference between these two groups on the underlying skills are inconsistent. In the study of Pennington and Lefly (2001) the FR-no-dyslexia children were comparable to the controls on PA, but showed difficulties in verbal short-term memory, RAN, and all literacy measures. Snowling et al. (2003) also found that FR-no-dyslexia children and controls differed on the literacy tasks. In addition, there was a trend for the FR-no-dyslexia children to perform somewhat weaker on PA. However, they performed equally well on the repetition of nonwords, a phonological task that measures verbal short-term memory. More recently, Boets et al. (2010) reported mild impairments in the FR-no-dyslexia group relative to controls on two out of the six literacy measures, although it seemed that larger groups would yield a significant difference on the other literacy
measures as well. The FR-no-dyslexia children were comparable to the controls on RAN and verbal short-term memory, whereas there was a trend, as in the study of Snowling et al., for PA to be lower. Taken together, it seems that difficulties of FR-children without dyslexia are subtle and may not be detected in studies with small samples.

3.1.2 Intergenerational Transfer
Since the etiology of dyslexia is multifactorial, involving both genetic and environmental influences, individual differences between children in dyslexic families must be associated at least in part with variations in their parents’ reading and cognitive skills. As an extension to earlier FR-studies, the second focus of our study was the relation between the skills of the children and those of their parents. In particular, we examined the relation between child and parental literacy achievement and their underlying cognitive skills.

FR-studies start from the premise that children with and without a FR of dyslexia differ in their liability of dyslexia. Implicitly, the assumption has been that within the FR-subsample, children do not differ in their FR. Since the reading skills of dyslexic parents all fall in the lower tail of the distribution, differences in parental reading skills between FR-children with and without dyslexia are not expected. To our knowledge, only two studies have tested this assumption. Snowling et al. (2007) looked at the relation between parents’ and children’s reading skills when the children were 12-13 year old. Although reading skills of parents and children were related within the control subsample, they were not within the FR-subsample. Based on these findings, Snowling et al. conclude that “it does not seem to be the case that more severely affected parents have more severely affected offspring”. This conclusion was contradicted by the study of van Bergen et al. (2011). Despite the fact that the dyslexic parents performed on average close to the fifth percentile, they found that the dyslexic parents whose child manifested dyslexia at age 11 were more severely impaired on word and nonword reading than those whose child acquired reading skills within the normal range. This finding suggests that the two groups of FR-children are not equal in their FR, i.e. the FR-children who become dyslexic have a higher liability of developing dyslexia. Differences in parental reading skills raise the question as to whether the FR-groups also differ in parents’ cognitive skills underlying reading.

In the present study we compared groups of children differing in FR-
status and reading outcome on literacy and its underlying skills. In addition, we assessed literacy and its underlying skills in their parents, which allowed us to investigate intergenerational transfer of skills from parent to child. Given a liability continuum and the high heritability of both reading and its underpinnings, we expected the FR-dyslexia children to show deficits across all tasks and the FR-no-dyslexia children to display mild weaknesses. Based on the findings of van Bergen et al. (2011), we predicted the FR-dyslexia children to have more severely affected dyslexic parents than the FR-no-dyslexia children.

3.2 Method

3.2.1 Participants
Three groups of children were involved in this study. The FR-dyslexia group consisted of 42 dyslexic children with a FR for dyslexia. The FR-no-dyslexia group comprised 99 children with a FR for dyslexia but without dyslexia. Finally, the control group included 66 children without a FR for dyslexia and without dyslexia.

All children participated in the Dutch Dyslexia Programme (DDP): a longitudinal multi-center study including the universities of Amsterdam, Nijmegen, and Groningen. Couples with and without a history of dyslexia who expected a baby were recruited between 1999 and 2002. They were recruited via advertisements in newspapers and leaflets at GPs and midwives. Only healthy, full-term babies were admitted to the program. The program was approved by the ethical commission and all parents gave informed consent for participation. About two-thirds of the sample consisted of families with a history of dyslexia.

Children were considered to have a FR for dyslexia if at least one of the parents and a close relative reported to have dyslexia. The dyslexia of the parent had to be confirmed by tests measuring word and nonword-reading fluency (see below). Moreover, the subtest Similarities of the Wechsler Adult Intelligence Scale was administered to measure verbal reasoning (Wechsler, 1997). For inclusion of a child in the FR-group, the parent with dyslexia had to fulfill at least one out of the following criteria: 1) scores on both reading tests ≤ 20th percentile, 2) one reading score ≤ 10th percentile and the other reading score ≤ 40th percentile, or 3) a discrepancy of ≥ 60 percentiles between the
verbatim score and one of the reading scores, with the restriction that neither reading-test percentile score exceeded 40. On average, the dyslexic parents of the FR-children scored on the reading tests close to the 5th percentile. For inclusion in the no-FR-group both parents had to score ≥ 40th percentile on both reading tests.

Dyslexia in children was assessed at the end of second grade. Children were considered dyslexic when their reading score on a word-reading fluency task (WRF2, see below) corresponded to the weakest ten percent in the population (see Boets et al., 2010 for a similar criterion). In the no-FR-group only two children turned out to be dyslexic. These children were excluded from the study. The characteristics of the three groups are presented in Table 3.1. Percentages of boys were similar across groups. However, compared to the other two groups, the FR-dyslexia group had a lower nonverbal IQ (SON-R, Tellegen, Winkel, Wijnberg-Williams, & Laros, 1998) at 48 months of age. Furthermore, the FR-dyslexia group was slightly older than the controls.

Table 3.1 Children’s Characteristics by Group

<table>
<thead>
<tr>
<th>FR</th>
<th>Characteristic</th>
<th>Dyslexia</th>
<th>No-dyslexia</th>
<th>Control</th>
<th>F(2, 204)</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Sample size</td>
<td>42</td>
<td>99</td>
<td>66</td>
<td></td>
</tr>
<tr>
<td></td>
<td>No (% of boys)</td>
<td>25a (60%)</td>
<td>56a (57%)</td>
<td>39a (59%)</td>
<td></td>
</tr>
<tr>
<td></td>
<td>Nonverbal IQ (at 48 months)</td>
<td>105.90 (15.27)</td>
<td>113.70 (14.31)</td>
<td>114.14 (15.38)</td>
<td>4.75</td>
</tr>
<tr>
<td></td>
<td>At assessment end Grade 2</td>
<td>98.60 (5.52)</td>
<td>97.67 ab (4.47)</td>
<td>96.41 c (4.34)</td>
<td>3.02</td>
</tr>
<tr>
<td></td>
<td>No of months reading instruction</td>
<td>18.69a (0.68)</td>
<td>18.87a (0.74)</td>
<td>18.80a (0.66)</td>
<td>0.96</td>
</tr>
</tbody>
</table>

Note. The group means are given with standard deviations in parentheses. Numbers and means in the same row that do not share subscripts differ at p < .05 on the χ²-test or Tukey’s test. No = number.

3.2.2 Measures

Children

The children were measured at the end of second grade on reading accuracy and fluency, spelling, PA, and RAN.

Word and nonword-reading accuracy. In these tests (de Jong & Wolters, 2002) the child was required to read a list of (non)words without time pressure. Each test consisted of 40 items increasing in difficulty from one to four
syllables. Testing was discontinued when six out of the last eight items were read incorrectly.

**Word-reading fluency (WRF).** This was assessed using three lists (Three-Minutes-Test, Verhoeven, 1995). The first and second (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 score per list was the number of words read correctly within one minute. WRF2 was used to assess dyslexia, as it has a high reliability (.96 at the end of Grade 2, Moelands, Kamphuis, & Verhoeven, 2003) and differentiates well between children at this age.

**Nonword-reading fluency.** This test (Klepel: van den Bos, lutje Spelberg, Scheepstra, & de Vries, 1994) consisted of a list of 116 pseudowords of increasing difficulty. The child was asked to read as many pseudowords as possible within two minutes, with the score being the number correctly read.

**Spelling.** The child had to write down (without time pressure) mono- and disyllabic words presented in a sentence (van den Bosch, Gillijns, Krom, & Moelands, 1993). The test had two versions, an easier and a more difficult one, consisting of 38 and 36 items. The easier version was given at two centers, the difficult version at the third. According to the test manual the scores of the tests could be transformed to one scale (based on Rasch analyses). The scores on this scale ranged from 80 to 144.

**PA.** PA was measured using a phoneme-deletion test (de Jong & van der Leij, 2003). On each item of the test a phoneme, always a consonant, had to be deleted from a nonword resulting in another nonword. The test consisted of two parts. The first part comprised nine monosyllabic and nine disyllabic nonwords. The second part consisted of nine disyllabic nonwords, with the phoneme that had to be deleted occurring twice. Testing was discontinued when six consecutive items were answered incorrectly in the first part or three in the second part. Each attempt was scored as either correct or incorrect. Both parts of the test started with two items for practice.

**RAN.** Serial RAN (RAN: van den Bos, 2003) was measured for numbers (2, 4, 5, 8, and 9) and colors (black, yellow, red, green, blue). Each of the tasks consisted of fifty randomly ordered symbols arranged in five columns of 10 symbols (numbers or colors) 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. The time to completion was transformed to number of symbols per second to normalize the score distribution.

Parents
The dyslexic parent of the FR-children and both parents of the control children were assessed on reading fluency, spelling, nonword repetition, and RAN. The data presented are the data of the weakest-reading parent. In addition, the level of education of both parents was ranked on a scale ranging from 1 (primary school only) to 5 (university degree).

Word-reading fluency. The test (One-Minute-Test: Brus & Voeten, 1972) consisted of a list of 116 words of increasing difficulty. The parent was asked to correctly read as many words as possible within one minute.

Nonword-reading fluency. The same test as for the children was used (see above).

Spelling. The ability to apply phonological analysis and spelling rules was tested with a test that required writing nonwords to dictation. The 26 nonwords ranged from one to three syllables.

Nonword repetition. The test (de Jong & van der Leij, 1999) consisted of 48 randomly ordered nonwords ranging from one to four syllables. The nonwords were presented twice on audiotape. After the nonword was presented the parent had to repeat back the nonword. The task started with three practice items. Each attempt was scored as either correct or incorrect.

RAN. Three serial RAN tasks were developed for the present study: one for upper case letters (all, except I, Q, and Y), one for numbers (2 up to and including 11), and one for colors (green, red, yellow, and blue). Each of the tasks consisted of 50 randomly ordered symbols arranged in five columns of 10 symbols each. Before test administration, the parent practiced by naming the last column of symbols. Parents were instructed to name the symbols column-wise as quickly as possible. For all three tasks, the time to completion was transformed to number of symbols per second to normalize the score distribution.

3.2.3. Procedure
Parents and children were tested individually by trained graduate students. The parents were tested in a quiet room at home or at the hospital around the birth of their child. The children were tested in a quiet room at the university between May and July of Grade 2.
3.3 Results

3.3.1 Data Screening
One dyslexic parent and their dyslexic child were removed from the analyses because of missing scores and scores on literacy measures below 3.3 SD's of the mean of their group. Data were missing for just a couple of the parents' tests (1-2%) and educational levels (4%). The analyses of these variables therefore included somewhat fewer cases. All score distributions were approximately normal. The one exception was for word-reading accuracy, which was too easy for the two groups of normal readers. These scores were logarithmically transformed before analysis.

3.3.2 Children
Table 3.2 displays the performance of the three groups of children (FR-dyslexia, FR-no-dyslexia, and controls) on the literacy and cognitive measures. One-way ANOVAs followed up with Tukey's test were used to investigate group differences. Effect sizes (Cohen's $d$'s) were computed using the SD's of the control group.

All literacy measures showed the same pattern: the FR-no-dyslexia group scored considerably higher than the FR-dyslexia group (with effect sizes between 1.22 and 2.41), but was still impaired relative to the control group (with effect sizes between 0.37 and 0.74). On average, the children with dyslexia could only read the mono- and disyllabic words and monosyllabic nonwords of the accuracy tests.

Similar group differences were observed for PA, but not for RAN. The FR-dyslexia group was significantly impaired on both RAN tasks; however, the FR-no-dyslexia group performed similarly to the control group. Finally, some extra analyses were performed to investigate whether the observed group differences on literacy(-related) skills were genuine rather than due to group differences on children's non-verbal IQ (Table 3.1) or parental educational level (Table 3.5). Nonverbal IQ correlated between .24 and .32 with the reading measures, .37 with spelling, .28 with PA and .07 and .20 with RAN digits and colors. Controlling for IQ in a series of one-way ANCOVAs yielded virtually the same results as the reported ANOVAs (Table 3.2), as did ANOVAs on a subset of data with groups matched for IQ. The level of education of the weakest-reading parent only correlated significantly with the outcome.
### Table 3.2: Children’s Performance on Literacy and Cognitive Measures by Group

<table>
<thead>
<tr>
<th>Measure</th>
<th>FRD vs. C</th>
<th>FRD vs. FRND</th>
<th>FRND vs. C</th>
<th>Effect size (Cohen’s d)</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Reading accuracy</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Words</td>
<td>40, 10.52</td>
<td>40, 8.19</td>
<td>40, 6.22</td>
<td>40, 5.73</td>
</tr>
<tr>
<td>Nonwords</td>
<td>40, 19.32</td>
<td>150, 11.83</td>
<td>150, 7.19</td>
<td>150, 6.85</td>
</tr>
<tr>
<td>Reading fluency</td>
<td>40, 11.93</td>
<td>150, 11.85</td>
<td>150, 11.32</td>
<td>150, 11.28</td>
</tr>
<tr>
<td>Words - WRF1</td>
<td>110, 19.85</td>
<td>110, 19.68</td>
<td>110, 21.57</td>
<td>110, 21.54</td>
</tr>
<tr>
<td>Words - WRF2</td>
<td>110, 19.85</td>
<td>110, 21.57</td>
<td>110, 21.54</td>
<td>110, 21.54</td>
</tr>
<tr>
<td>Words - WRF3</td>
<td>110, 19.85</td>
<td>110, 21.57</td>
<td>110, 21.54</td>
<td>110, 21.54</td>
</tr>
<tr>
<td>Nonwords</td>
<td>110, 21.57</td>
<td>110, 21.54</td>
<td>110, 21.54</td>
<td>110, 21.54</td>
</tr>
<tr>
<td><strong>Spelling</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Words</td>
<td>144, 6.38</td>
<td>144, 4.80</td>
<td>144, 4.26</td>
<td>144, 4.13</td>
</tr>
<tr>
<td>Nonwords</td>
<td>144, 14.47</td>
<td>144, 14.47</td>
<td>144, 14.47</td>
<td>144, 14.47</td>
</tr>
<tr>
<td>PA</td>
<td>27, 9.26</td>
<td>27, 9.26</td>
<td>27, 9.26</td>
<td>27, 9.26</td>
</tr>
<tr>
<td><strong>RAN</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Digits</td>
<td>122, 1.22</td>
<td>122, 1.32</td>
<td>122, 1.22</td>
<td>122, 1.22</td>
</tr>
<tr>
<td>Colors</td>
<td>198, 0.83</td>
<td>198, 0.84</td>
<td>198, 0.84</td>
<td>198, 0.84</td>
</tr>
</tbody>
</table>

Note: Children’s skills were assessed at the end of Grade 2. Max. = maximum score; WRF = word-reading fluency. Numbers and means in the same row that do not share subscripts differ at p < .05 on Tukey’s post-hoc test. Cohen’s d is calculated using the SDs of the controls.

FR = FR-dyslexia, FRND = FR-no dyslexia, C = control.
variable PA: \( r = .16 \). Adjusting for parental education in ANCOVAs did not alter the pattern of results. Although equating for group differences can be disputed (Miller & Chapman, 2001), the fact that the ANCOVAs yield the same picture as the ANOVAs strengthen our findings.

### 3.3.3 Intergenerational Transfer

Tables 3.3 and 3.4 display correlations between children's and parents' measures of literacy and their underlying skills. As an illustration, the relation between nonword-reading fluency in children and parents is shown graphically in Figure 3.1. This measure was chosen since it was assessed with the same test in children and parents. Due to our strict selection criteria (i.e., percentile scores of the dyslexic parents ≤ 40 and those of the control parents ≥ 40), correlations within the FR and control subsamples suffered from restriction of range in the parental measures, which suppressed correlations. The correlations presented are therefore of the FR and control subsamples combined. In the control sample only the data of the weakest-reading control parent was selected. Correlations are given separately for the weakest-reading mothers and their children and the weakest-reading fathers and their children. The correlations show that reading skills of parents and children were moderately correlated. As expected, mothers' nonword repetition correlated strongest with children's PA. Mothers' RAN, however, correlated stronger with children's reading than with children's RAN. No such clear pattern was observed for father-child correlations. Children's reading fluency seemed to correlate higher with the performance of mothers than of fathers. The significance of this difference was tested using Mx (Neale, 2004). It appeared that an unconstrained two-group model fitted the data better than one with the set of eight correlations (i.e., two reading-fluency measures times four parental measures) constrained to be equal across mothers and fathers, \( \Delta \chi^2(8, N=205) = 15.55, p = .049 \). Children's PA and RAN, on the other hand, did not correlate differently with test scores of mother and fathers, \( \Delta \chi^2(8, N=205) = 7.86, p = .447 \). The proportion of dyslexic parents was virtually equal in the two groups, as was the variability in test scores. Hence, these factors could not explain the differences between maternal and paternal correlations.
Table 3.3 Correlations among skills of mothers and their children

<table>
<thead>
<tr>
<th>Children</th>
<th>Reading fluency</th>
<th>Mother</th>
<th></th>
<th></th>
<th></th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td></td>
<td>Words</td>
<td>Nonwords</td>
<td>Nonword repetition</td>
<td>RAN digits</td>
</tr>
<tr>
<td>Words</td>
<td>.43</td>
<td>.47</td>
<td>.23</td>
<td>.38</td>
<td></td>
</tr>
<tr>
<td>Nonwords</td>
<td>.51</td>
<td>.56</td>
<td>.21</td>
<td>.49</td>
<td></td>
</tr>
<tr>
<td>Phonological awareness</td>
<td>.54</td>
<td>.50</td>
<td>.34</td>
<td>.31</td>
<td></td>
</tr>
<tr>
<td>RAN digits</td>
<td>.13</td>
<td>.12</td>
<td>.10</td>
<td>.19</td>
<td></td>
</tr>
</tbody>
</table>

Note. Word-reading fluency of children is WRF2. n’s [83, 87]. For n = 85: p < .05 for r ≥ .21 and p < .001 for r ≥ .35.

Table 3.4 Correlations among skills of fathers and their children

<table>
<thead>
<tr>
<th>Children</th>
<th>Reading fluency</th>
<th>Father</th>
<th></th>
<th></th>
<th></th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td></td>
<td>Words</td>
<td>Nonwords</td>
<td>Nonword repetition</td>
<td>RAN digits</td>
</tr>
<tr>
<td>Words</td>
<td>.39</td>
<td>.38</td>
<td>.22</td>
<td>.27</td>
<td></td>
</tr>
<tr>
<td>Nonwords</td>
<td>.30</td>
<td>.32</td>
<td>.15</td>
<td>.15</td>
<td></td>
</tr>
<tr>
<td>Phonological awareness</td>
<td>.22</td>
<td>.29</td>
<td>.22</td>
<td>.16</td>
<td></td>
</tr>
<tr>
<td>RAN digits</td>
<td>.28</td>
<td>.26</td>
<td>.17</td>
<td>.19</td>
<td></td>
</tr>
</tbody>
</table>

Note. Word-reading fluency of children is WRF2. n’s [119, 120]. For n = 119: p < .05 for r ≥ .18 and p < .001 for r ≥ .30.
Figure 3.1. Scatterplots (with regression lines) illustrating the relation for nonword-reading fluency between children and mothers (top panel, \( r = .56 \)) and children and fathers (lower panel, \( r = .32 \)).
Differences among the FR-dyslexia, FR-no-dyslexia, and control children were also investigated for parental characteristics and skills. There was no difference between the two FR-groups in gender of the dyslexic parent: The dyslexic parent was the father in 55% of the cases in the FR-dyslexia group compared to 62% in the FR-no-dyslexia group ($\chi^2 < 1$). The other parental characteristics and test scores by group are shown in Table 3.5. The effect sizes of the comparisons with the control group were large; the comparisons of the two FR-groups yielded small effect sizes, except for word-reading fluency and alphanumerical RAN with medium effect sizes. Regarding background characteristics, the mothers and fathers of both FR-groups had a lower educational level compared to the control group. The two FR-groups differed in educational level of the non-dyslexic parent, but not in that of the dyslexic parent. The general Dutch population has an average level of education of 3.20 (with SD = 0.94, Statline, 2010), which indicates that the parents in the FR and control samples were slightly (Cohen’s $d$ = .31) and considerably (Cohen’s $d$ = 1.01) above average, respectively.

The parental skills investigated included verbal reasoning, literacy, and literacy correlates. The parents in the control group obtained the highest scores on verbal reasoning, followed by the FR-dyslexia and FR-no-dyslexia groups, respectively. By definition, the dyslexic parents in the FR-groups were weaker readers than the parents in the control group, but they scored also significantly lower than the controls on spelling. Since we hypothesized that the parents of the FR-dyslexia children would score lower than those of the FR-no-dyslexia children on literacy(-related) tasks, we performed planned $t$-tests. As predicted, the dyslexic parents of the FR-dyslexia group scored significantly lower than those of the FR-no-dyslexia group on word-reading fluency ($t(139) = 1.69, p = .047$, one-tailed). However, the FR-groups did not differ in nonword-reading fluency and spelling. The reading measures correlated similarly with educational level (within FR-sample: word reading: $r = .38$; nonword reading: $r = .42$).

Concerning the correlates of reading, the parents of the controls were better than those of the FR-children on nonword repetition and RAN. The two FR-groups did not differ significantly on nonword repetition and RAN of colors, but they did on alpha-numeric RAN: the dyslexic parents of the FR-no-dyslexia children were significantly better on both letters ($t(137) = 2.16, p = .016$, one-tailed) and digits ($t(137) = 2.52, p = .006$, one-tailed). Parental RAN hardly correlated with their educational level (within FR-sample: .14, .19, and .10 for letters, digits, and colors, respectively).
Table 3.5 Parents’ Characteristics and Performance on Literacy and Cognitive Measures by Group

<table>
<thead>
<tr>
<th>Characteristic</th>
<th>Max.</th>
<th>M</th>
<th>SD</th>
<th>M</th>
<th>SD</th>
<th>M</th>
<th>SD</th>
<th>F</th>
<th>df</th>
<th>Effect size (Cohen’s (d))</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>FR</td>
<td>Dyslexia</td>
<td>No-dyslexia</td>
<td>Control</td>
<td>F</td>
<td>df</td>
<td>FRD vs. FRND</td>
<td>FRD vs. C</td>
<td>FRND vs. C</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>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Mother</td>
<td>5</td>
<td>3.40</td>
<td>0.87</td>
<td>3.56</td>
<td>0.89</td>
<td>4.14</td>
<td>0.72</td>
<td>12.69</td>
<td>(2,196)</td>
<td>0.22</td>
</tr>
<tr>
<td>Father</td>
<td>5</td>
<td>3.34</td>
<td>0.88</td>
<td>3.51</td>
<td>0.91</td>
<td>4.16</td>
<td>0.88</td>
<td>13.66</td>
<td>(2,196)</td>
<td>0.19</td>
</tr>
<tr>
<td>Dyslexic parent</td>
<td>5</td>
<td>3.41</td>
<td>0.87</td>
<td>3.41</td>
<td>0.90</td>
<td>&lt; 1</td>
<td></td>
<td></td>
<td></td>
<td>0.00</td>
</tr>
<tr>
<td>Non-dyslexic parent</td>
<td>5</td>
<td>3.33</td>
<td>0.89</td>
<td>3.67</td>
<td>0.89</td>
<td>4.26</td>
<td></td>
<td></td>
<td></td>
<td>0.38</td>
</tr>
<tr>
<td>Verbal reasoning</td>
<td>26</td>
<td>16.98</td>
<td>3.04</td>
<td>16.43</td>
<td>3.79</td>
<td>18.43</td>
<td>3.16</td>
<td>6.63</td>
<td>(2,202)</td>
<td>-0.17</td>
</tr>
<tr>
<td>Reading fluency</td>
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<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Words</td>
<td>116</td>
<td>61.81</td>
<td>12.19</td>
<td>65.86</td>
<td>13.35</td>
<td>96.61</td>
<td>8.48</td>
<td>167.62</td>
<td>(2,204)</td>
<td>0.48</td>
</tr>
<tr>
<td>Nonwords</td>
<td>116</td>
<td>46.17</td>
<td>17.94</td>
<td>47.73</td>
<td>15.00</td>
<td>100.95</td>
<td>9.84</td>
<td>318.42</td>
<td>(2,204)</td>
<td>0.16</td>
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<tr>
<td>Spelling</td>
<td>26</td>
<td>12.98</td>
<td>4.79</td>
<td>13.47</td>
<td>3.96</td>
<td>18.66</td>
<td>3.12</td>
<td>41.54</td>
<td>(2,200)</td>
<td>0.16</td>
</tr>
<tr>
<td>Nonword repetition</td>
<td>48</td>
<td>36.50</td>
<td>5.99</td>
<td>37.24</td>
<td>5.67</td>
<td>42.83</td>
<td>3.50</td>
<td>28.07</td>
<td>(2,200)</td>
<td>0.21</td>
</tr>
<tr>
<td>RAN</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Letters</td>
<td>48</td>
<td>1.91</td>
<td>0.45</td>
<td>2.10</td>
<td>0.50</td>
<td>2.79</td>
<td>0.41</td>
<td>60.37</td>
<td>(2,200)</td>
<td>0.46</td>
</tr>
<tr>
<td>Digits</td>
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<td>1.91</td>
<td>0.47</td>
<td>2.14</td>
<td>0.53</td>
<td>2.72</td>
<td>0.44</td>
<td>41.23</td>
<td>(2,200)</td>
<td>0.52</td>
</tr>
<tr>
<td>Colors</td>
<td>48</td>
<td>1.41</td>
<td>0.27</td>
<td>1.45</td>
<td>0.29</td>
<td>1.87</td>
<td>0.29</td>
<td>50.48</td>
<td>(2,199)</td>
<td>0.14</td>
</tr>
</tbody>
</table>

Note. Test scores belong to the weakest-reading parent. Numbers and means in the same row that do not share subscripts differ at \(p < .05\) on Tukey’s post-hoc test. Cohen’s \(d\) is calculated using the SDs of the controls. Max. = maximum score, FRD = FR-dyslexia, FRND = FR no-dyslexia, C = control.
Finally, it was investigated whether the effect of parental characteristics on children's literacy outcome was fully mediated by children's characteristics. Therefore, a set of hierarchical regression analyses focusing on the FR-group was performed. In predicting children's literacy outcome (i.e., WRF2), first child predictors were entered (nonverbal IQ, PA, and RAN) and then one of the parental characteristic. It was found that parental word-reading fluency ($\beta = .136, p = .031$) and alphanumeric RAN (letters: $\beta = .149, p = .018$; digits: $\beta = .123, p = .054$) added further variance to the model, indicating that they affected children's literacy outcome (partly) independent from children's cognitive skills. Parental characteristics that did not improve the model were nonword-reading fluency ($\beta = .096, p = .128$), RAN colors, nonword repetition, and level of education of the dyslexic and non-dyslexic parent (all $p$s $> .40$). The predictive value of parental RAN on children's literacy outcome is nicely illustrated when comparing groups of dyslexic parents with and without a RAN deficit (using the scores marking the bottom 10% in the control group as cut-offs). In the group without a RAN deficit in letters or digits 14% and 16%, respectively, manifested dyslexia, while these proportions where 39% and 41%, respectively, for the parents with a RAN deficit.

3.4 Discussion

After two years of reading instruction, 30% of the children with a family history of dyslexia had developed dyslexia, compared to only 3% of the children without such a history. These figures agree with other studies using a similarly (strict) criterion to identify dyslexia.

The FR-dyslexia children were significantly impaired relative to the FR-no-dyslexia children on PA, RAN, spelling, and reading accuracy and fluency of both words and nonwords. This is in line with previous longitudinal FR-studies contrasting groups of 8-year-olds (Boets et al., 2010; Pennington & Lefly, 2001; Snowling et al., 2003; Torppa et al., 2010; van Bergen et al., 2011). In contrast to several earlier studies in transparent orthographies (e.g., de Jong & van der Leij, 2003; Wimmer, 1993), but in accordance with Boets et al. (2010), reading accuracy differentiated dyslexic from normal readers and even FR-no-dyslexia children from controls. This shows that differences in accuracy are observable until at least second grade, provided that the task is not restricted to one- or two-syllable items.
In accordance with previous studies (Boets et al., 2010; Snowling et al., 2003), the FR-no-dyslexia group performed better than the FR-dyslexia group, but weaker than the controls on PA and literacy, although their performance was well within the normal range. In fact, compared to the national norm scores for the reading-fluency and spelling tests, the performance of the FR-no-dyslexia group was just below average, while the performance of the controls was just above.

Regarding PA, this study is the first study that shows reliable differences between FR-no-dyslexia children and controls. This confirms the earlier reported trends observed by Snowling et al. (2003) and Boets et al. (2010). Strikingly, the FR-no-dyslexia children were unimpaired on RAN. Of the two other FR-studies that also included RAN, our result resembles that of Boets et al. (2010), but contrasts with that of Pennington and Lefly (2001), who found the FR-no-dyslexia group to perform slower. Unlike Boets et al. and our study, Pennington and Lefly used reading accuracy to diagnose dyslexia. However, reanalyzing RAN differences with reading status based on word-reading accuracy still yielded equivalent performances of our two non-dyslexic groups.

The poor performance on RAN of the FR-children with dyslexia does not seem to be a consequence of their poor reading. The current study showed that the FR-children without dyslexia were faster on RAN than their relatively weak reading fluency suggested. Furthermore, longitudinal studies have demonstrated that FR-children who later manifested dyslexia were already slow at RAN before they came to the task of learning to read (Boets, Wouters, van Wieringen, & Ghesquière, 2007; Pennington & Lefly, 2001; van Bergen et al., 2011). Finally, Lervåg and Hulme (2009) found RAN to be a predictor of reading development, but failed to find support for a reciprocal influence of reading on RAN. These findings argue against RAN performance as a consequence of reading performance.

Besides the skills of the children in the three groups, we aimed to investigate the relation between child and parental skills. Child and parental reading skills were moderately correlated: ~.35 for fathers and ~.50 for mothers. These correlations are higher than the correlation of .28 reported by Bynner and Parsons (2006), although this may well be explained by the fact that parental literacy skills in their cohort study were self-reported instead of tested. Interestingly, children’s reading fluency, but not their PA and RAN, correlated higher with mothers’ than with fathers’ skills. This is suggestive of an
environmental influence: it could be that mothers are in general more involved in parenting and that this has an effect on their offspring’s reading skill but not on its underlying cognitive skills. On the other hand, we cannot rule out the possibility that some of the fathers in our sample are not the biological fathers. This would suppress father-child correlations. Moreover, the finding should be interpreted with caution since it involves only data from the weakest-reading parent. Replication with complete parent data is therefore warranted.

Importantly, we found that the dyslexic parents whose child manifested dyslexia were even more impaired on word-reading fluency than those whose child had an age-appropriate reading level. However, the difference was smaller (i.e., a medium effect size of $d = .48$) than the difference observed by van Bergen et al. (i.e., a large effect size of $d = .78$). Moreover, the current study failed to replicate a difference in nonword-reading fluency, as reported by van Bergen et al. Since reading ability is not yet fully stabilized after two years of reading instruction, the moment of assessing reading status could partially explain the differences between the studies. Indeed, when we reanalyzed van Bergen et al.’s data with reading status based on children’s reading skills at the end of second grade (instead of fifth grade, as in the original study), the difference in parental reading ability between the two FR-groups decreased, although it remained larger than in the current sample. Furthermore, we wondered whether the presence of a word-reading difference and the absence of a nonword-reading difference in the current sample could be subscribed to a difference in the amount the two groups of dyslexic parents read. However, the two reading measures correlated similarly with educational level, which argues against this notion.

As expected, the dyslexic parents were impaired on all underlying cognitive skills. Although nonword repetition (as a proxy for PA) discriminated well between parents with and without dyslexia, within the FR-subsample parental nonword repetition appeared to be unrelated to children’s reading outcome. Interestingly, the dyslexic parents of the FR-dyslexia children were somewhat slower on RAN than those of the FR-no-dyslexia children. Parental differences in reading and RAN might be related to differences in liability of their offspring. Compared with parental reading, parental RAN might be a cleaner indicator of their genetic potential, because it is less influenced by reading experience (as shown by the correlations with educational level).

Another indicator of children’s liability is that of the parents’ level of
education. As also observed by van Bergen et al. (2011), the FR-no-dyslexia children have non-dyslexic parents with a higher educational level compared to FR-dyslexia children. This might be an additional factor which affects the outcome of children, operating via inheritance and the home-literacy environment.

Subsequently, we examined possible intergenerational transfer by examining whether cognitive skills of the dyslexic parents were related to children's reading outcome. It appeared that differences in parental word-reading fluency and alphanumeric RAN were predictive of differences in children's reading ability, beyond the effect of children's cognitive skills. The predictive value of parental RAN is also shown by the fact that of the children of a dyslexic parent without a RAN deficit just 15% developed dyslexia, compared to as many as 40% of those with a RAN deficit. This agrees with the finding that the two groups of FR-children differed in parental RAN. Both underline that RAN deficits in dyslexic parents added to their offspring's risk for dyslexia, over and above their reading status. Furthermore, parental RAN correlated stronger with children's reading than with children's RAN. Apparently, RAN of parents and RAN of children somehow measure different aspects of children's reading ability.

The mild deficits in PA and literacy displayed by the FR no-dyslexia group corroborate a liability continuum. Additionally, both children's data and parent-child relations highlight that – at least within transparent orthographies – RAN plays a more important role in reading acquisition than PA. A parallel can be drawn between the present findings and those of Bishop, McDonald, Bird, and Hayiou-Thomas (2009), who studied another group at-risk for dyslexia: children with language impairment. They also found that those who achieved adequate reading skills despite their risk showed remarkable good RAN skills. They suggest that what RAN taps might protect against reading failure. Our data support this hypothesis, although longitudinal studies are required to elucidate whether and how the skills underlying RAN performance might protect children at FR for dyslexia.
3.5 Key Points

- Prospective studies with children at FR of dyslexia demonstrate the multifactorial etiology of dyslexia.
- The current study shows that FR-children who do not meet criteria for dyslexia are mildly impaired on literacy and PA, but not on RAN.
- Dyslexic parents of dyslexic children tended to be more severely impaired on word-reading and RAN skills than dyslexic parents of non-dyslexic children.
- Parental characteristics which affect the reading outcome of their offspring are reading, RAN, and level of education. These factors can be used to improve a child's risk assessment for developing dyslexia.
3.6 References


Literacy Levels within Families with a History of Dyslexia


