Early identification and intervention in children at risk for reading difficulties
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'Wat was alles ook maar weer?' vroeg de zwaan, terwijl hij een scherpe bocht nam en rakelings langs een klaproos scheerde.
'Alles,' zei de vlinder.
'O ja!' zei de zwaan. 'Hoe kon ik dat nou vergeten.'

Uit: Toon Tellegen (2002), 'Misschien wisten zij alles' (p. 225)
Chapter 6

General Discussion

In this general discussion, the significance and limitations of the studies reported in Chapters 2 to 5, and the implications for future directions will be discussed in the broader perspective of early identification and intervention in children at-risk for developing reading difficulties (RD).

6.1 Identification of at-risk children

6.1.1 Early or not so early identification?

At what time point in (pre-)literacy development is it functional to identify children based on known risk factors that signal difficulties with reading acquisition? Functional in the sense that identification includes (most) children who truly are in need of early or additional support? It may be clear that functional identification highly depends on the measures used to verify risk status. As with screening for medical conditions, those measures are required to be sensitive (correctly identify those at-risk) as well as specific (correctly identify those not at-risk).

A quick literature search indicates that the extent to which early identification studies can predict academic outcomes varies considerably across studies and samples. In a research analysis that already dates back 35 years, Mercer, Algozzine and Trifiletti (1979) found that in terms of percentages the identification approaches of the included prediction studies yielded overall hit rates (total number of correctly identified regardless of risk status) between 46% and 92%, with a median overall hit rate of 73% for studies using single instruments as predictors and 79% for studies using multiple-instrument batteries. Differential patterns of maturation and mild impairments as well as identification before the onset of formal academic instruction appeared to contribute to incorrect identification.

In a more recent meta-analysis of 70 available longitudinal studies (with data from 62 independent samples) published between 1985 and 1998, La Paro and Pianta (2000) found that the average correlations between academic/cognitive assessment in kindergarten and academic/cognitive outcome in first and/or second grade ranged from .12 to .78. The overall effect size of .51 of these studies may either be interpreted as reflecting
moderate stability in child performance across early school years or substantial variability. An estimate of this size also indicates that children’s rank order will change over the kindergarten to second grade period, therefore schools should not only monitor the progress of children’s skills, but also take into account background variables and environmental factors - including their own contribution - which may affect development.

The above meta-analytic reviews both illustrate the limitations of early identification by measure-based prediction studies considering various outcomes in the academic domain. In an influential review concerning the prediction of reading acquisition from kindergarten measures, Scarborough (1998) reported similar findings in an examination of 55 studies (61 samples) conducted over a twenty-year period, in which a wide variety of predictors were used. Measures that required print processing (print familiarity, letter naming, and emergent reading skills) yielded higher average correlations between kindergarten predictors and reading skills one or two years later (in the range from .46 and .57), as compared to measures of oral language proficiency (range .22 - .46) and nonverbal abilities (range .16 - .31). Higher multiple correlations were obtained (R = .75, on average) when combining kindergarten measures. Predictive accuracy of reading status for the bottom 12% of the distribution improved considerably, resulting in a percentage of true positives (children accurately assessed as being at high risk) of 89% (McCardle, Scarborough & Catts, 2001). The number of false negatives (children incorrectly predicted as not at-risk) appeared to be lower than the number of false positives (children incorrectly predicted as at-risk).

6.1.2 Early identification in an unselected subsample without familial risk

The samples partaking in the studies included in this thesis can be used to shed light on the issue of early identification and subsequent outcome in relation to background and intervention variables. First, the focus will be on the literacy development of the children who did not receive intervention and were included in the study in Chapter 3 as no family risk controls. In this otherwise unselected subsample normal variation in pre-literacy and other cognitive skills may be expected. To illustrate developmental dynamics, of this group of fifteen children, eleven made significant progresses in pre-literacy skills during the remaining five months in kindergarten. You might say they were ‘ready’ to start learning to read because they picked up letters informally. After having visited Grade 1 for
about half a year, word reading fluency scores indicated that reading development was above the 50th percentile for eight out of those eleven children. However, in Grade 5\(^1\) only five remained in the upper half, as can be seen in Figure 6.1.

**Figure 6.1.** Literacy trajectories for 15 subjects not at family risk (noFR). In the left shaded area scores on receptive letter knowledge (maximum number correct = 32; in percentages) at enrolment in kindergarten, and 5 months later just before the onset of formal reading instruction at the end of kindergarten. In the right shaded area scores on word reading fluency (percentiles) halfway Grade 1 and Grade 5.

For six of the eleven children with good initial letter knowledge at the end of kindergarten, different trajectories were found. Three remained in the upper half with word reading fluency halfway Grade 1 but fell back to the 20-30th percentile in Grade 5, two already showed up in the below average area in Grade 1 and stayed there in Grade 5. One (noFR4) developed into a very poor reader straight away halfway Grade 1, scoring in the bottom 10\% in word reading fluency in Grade 5. Relatively good initial letter knowledge appeared to be a forerunner of good word reading fluency in only five out of eleven cases.

With regard to the four noFR children who could have been labelled at-risk based on poor letter knowledge at the end of kindergarten, just one

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\(^1\) The follow-up assessment halfway Grade 5 is not included in Chapter 3.
(noFR8) performed poorly in Grade 1 and 5. One (noFR13) remained in the 20-30th percentile area. The child with code noFR12 had a slow start in Grade 1 but performed as one of the best in Grade 5, whereas another one (noFR3) made the reverse progress from a score above the 50th towards round about the 20th percentile in Grade 1-5. Three out of the four cases with poor initial knowledge at the end of kindergarten, developed more or less as expected, but one was a very positive exception.

Suppose we had used the cognitive risk profile of the 15 children at enrolment in kindergarten to identify risk, taking as the risk criteria either a score greater than 1 $SD$ below the noFR control group means on phonemic awareness ($\leq 6$) or receptive letter knowledge ($\leq 11$), or greater than 1 $SD$ below the age-standardized means on serial naming speed of objects or colours? How well would the outcome criterion of Grade 5 word reading fluency below 10th percentile be predicted? One noFR child being at-risk (true-positive) would have been correctly identified (noFR8), three incorrectly (false-positives: two of which, apart from late-bloomer noFR12, ended up as relatively poor readers, i.e. noFR3 and noFR13), and another one completely missed (false-negative: noFR4). It would have left ten (two-thirds) of the children correctly identified as true-negatives, which results in a moderate overall hit rate (total number of correctly identified children) of 73%.

When we know that reading difficulties do not run in the family of a particular child, and can only rely on cognitive profiles half a year prior to or at the start of formal instruction, what does this outcome tell us? Should we wait and see which student eventually will fail to read properly before providing additional help? Adherence to such a policy, as in the older days, does not seem to be an attractive option. According to Scarborough (2009) about 65-75% of children identified as reading poorly early in the process of learning to read continue to do so in later years, whereas only 5-10% of children who start out satisfactorily will develop literacy problems after all. The results of our small sample of noFR children corroborate these estimates. If we had provided focused instruction starting halfway the last year of kindergarten, one child was certainly in need of that, another would possibly have started to progress earlier, and two others might have become better readers. The same picture can be drawn when the cognitive profile at the end of kindergarten was used. However, Figure 6.1 also supports the view that monitoring progress in Grade 1 and onwards is necessary to identify both false negatives at a later stage and provide them with focused
intervention, and false positives for who the intervention may be terminated once progress allows that.

6.1.3 Early identification in a selected subsample with cognitive risk

Identification of children as being at-risk just after the start of formal reading instruction was used in the sample that participated in the Chapter 5 intervention. The advantage of such an approach is, that similar to the first stage of the response-to-intervention method (RTI) (Fuchs & Fuchs, 2006), identification is limited to those children who do not immediately show the expected progress when being immersed with reading instruction that very first month in Grade 1. A major difference between this Grade 1 sample and the kindergarten sample of section 6.1.2 is that the reading abilities of the participants’ parents were not assessed upon enrolment. Thus, while the absence of an FR background was the only selection criterion for inclusion in the kindergarten control sample, inclusion in the Grade 1 study was according to a two-step procedure to determine cognitive risk. Children were referred by teachers and required to meet screening criteria for high risk based on emergent literacy skills. Thus, the sample comprised only of children considered to be cognitively at-risk.

As described in 6.1.1, initial letter knowledge, phonological awareness and naming speed of letters are the best predictors of later reading development in Grade 1 or 2. The total explained variance, however, is not higher than 50% (Schatschneider, Fletcher, Francis, Carlson, & Foorman, 2004). For example, recently follow-up results of the Finnish Jyväskyla Longitudinal Study of dyslexia (JLD) consisting of FR and noFR children were reported with respect to outcome in reading in Grade 2 (Eklund, Torppa, & Lyytinen, 2013). Approximately half of the children with a high early cognitive risk profile appeared to have problems with learning to read, the large majority among them were also at family risk. Their findings can be compared to the data of the sample of the Chapter 5 study.

In our Grade 1 intervention study (Chapter 5), after referral by their own teacher as possible at-risk students, participants were screened on letter knowledge and decoding accuracy, as well as on phonological awareness. Poor performance on either one of these measures would lead to inclusion. Included children were randomly assigned to two conditions: intervention and control (no-intervention). Mingled with letter knowledge, phonemic skills were an important target of our intervention. While naming speed was not one of the screening tools, rapid naming of colours and objects was
assessed right before the start of the intervention period. So in retrospection we are able to tell whether early identification of later reading difficulties would have improved, if we had included naming speed.

For the purpose of providing an example in this general discussion, and using only the data of the 66 control children who did not receive focused intervention the initial screening criteria were sharpened as follows. Considered to be low performance in this subsample of referred at-risk students were productive letter knowledge and decoding accuracy scores below 1.3D above the overall mean (values < 20 and < 7 respectively), a phonological awareness score in the bottom 9th percentile (≅ standard score < 7), in addition to rapid naming scores in the bottom 12th percentile (≅ standard score < 7). A risk profile based on two out of three indicators (emergent literacy and low phonological awareness or low rapid naming), and outcome defined as word reading fluency in Grade 3 below threshold 10th percentile, appears to yield the best trade off between sensitivity (78%) and specificity (52%), and an overall hit rate of 59% (see Table 6.1).

Table 6.1
Outcome reading status based on word reading fluency <10th percentile in Grade 3 and cognitive risk profile begin Grade 1 (based on at least two scores below criterion on emergent literacy, phonological awareness and rapid naming) in the at-risk control group enrolled in Grade 1 (overall hit rate is 59%)

<table>
<thead>
<tr>
<th>Risk</th>
<th>Reading Difficulties</th>
<th>No Reading Difficulties</th>
</tr>
</thead>
<tbody>
<tr>
<td>Cognitive</td>
<td>14</td>
<td>37</td>
</tr>
<tr>
<td></td>
<td></td>
<td>23</td>
</tr>
<tr>
<td>No Cognitive</td>
<td>4</td>
<td>29</td>
</tr>
<tr>
<td></td>
<td></td>
<td>25</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td>18 RD</td>
<td>48 No RD</td>
</tr>
<tr>
<td></td>
<td>14 were predicted</td>
<td>25 were predicted</td>
</tr>
<tr>
<td></td>
<td>(sensitivity is 78%)</td>
<td>(specificity is 52%)</td>
</tr>
</tbody>
</table>

In considering the Grade 3 outcome, almost three quarters (73%) did not develop RD based on a threshold <10th percentile on standardized word reading fluency. The rate of false positives in our subsample of at-risk controls (62%) appears to be somewhat higher than in the Eklund et al. (2013) study (52%), in which almost half of the children with cognitive risk profiles indicating high risk were reported to have RD at the end of Grade

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2 Schools were allowed to provide them with additional support.
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(based on a composite of word reading accuracy and fluency measures below the 10th percentile). With respect to false negatives, comparable rates were found (16% in the Eklund sample versus 13% in our subsample).

An explanation for the high rate of false positives may be that according to the reports of the schools about half of the referred control students received school initiated help in addition to regular reading instruction for at least one school year. Among them were 15 (65%) out of the 23 students identified as false positives in Table 6.1. So, although these control children did not practice with the intervention program their reading outcome may have been raised due to the additional support delivered by the school. Furthermore, it would have been informative to make up the cognitive developmental profile of the individual cases identified as false negatives, for whom no problems with reading acquisition were expected given their performance at the beginning of Grade 1. As in the study of Eklund et al. (2013), these poor readers with normal pre-literacy skills may have shown additional risk factors like for example poorer cognitive skills, task avoidance and/or less time spent reading as compared to children who did not develop reading difficulties later on.

Recall that the overall hit rate achieved for the noFR controls classified in kindergarten on the basis of a less stringent risk profile (6.1.2) was 73% versus the overall hit rate of 59% in the Grade 1 controls. An obvious explanation is that the latter group was composed of a more heterogeneous population, including children from families with a home language different from Dutch, and either with or without a familial risk for dyslexia. Apparently, hit rate increases when a familial risk factor is taken into account, and identification of true positives and negatives is not solely based on cognitive risk indicators.

6.1.4 Early identification in a selected subsample with familial risk

The outcome in the FR control group enrolled in kindergarten offers insight in the probable development over time of high-risk children if no early intervention is provided. Of the 26 FR controls, seventeen (65.4%) were found to have a high cognitive risk profile as assessed after enrolment, using the same pre-literacy criteria as determined for the noFR group (6.1.2). Three of those seventeen children with a ‘double’ risk appeared to be reading at age-appropriate levels (scores above 25th percentile on word
reading fluency) halfway Grade 5. Thus, they were doing fine\(^3\) although their cognitive profile in kindergarten in combination with their family background may have suggested otherwise. Four more obtained reading levels between the 10\(^{th}\) and 25\(^{th}\) percentiles. Of the remainder nine FR children whose pre-literacy skills did not suggest a cognitive risk, two belonged to the lowest 25\(^{th}\) in Grade 5, the other seven performed at age-appropriate levels.

Taking into account both familial and cognitive risk factors, when using a RD criterion (10\(^{th}\) percentile on word reading fluency) the rate of sensitivity reaches 100\% - at the expense of specificity (see Table 6.2). Given the chosen risk cut-off levels, it also results in a relatively high rate of false-positives (41\%), i.e., over-identification of children at-risk. The overall hit rate (73\%) is comparable to what was found in the subsample of noFR controls. What is striking is that in this subsample of FR controls a familial risk alone does not result in severe reading difficulties. A child who *combines* a cognitive risk with a familial risk is at great risk of developing into a very poor reader.

<table>
<thead>
<tr>
<th>Risk</th>
<th>Reading Difficulties</th>
<th>No Reading Difficulties</th>
</tr>
</thead>
<tbody>
<tr>
<td>Familial + Cognitive</td>
<td>10</td>
<td>7 false positives</td>
</tr>
<tr>
<td>Familial + No Cognitive</td>
<td>0</td>
<td>9 false negatives</td>
</tr>
<tr>
<td></td>
<td>0</td>
<td>9</td>
</tr>
</tbody>
</table>

In comparing the predictive outcomes for two subsamples from different studies, one selected on the basis of cognitive risk only, the other in which both familial and cognitive risk was considered, it is clear that the rates of false-positives are high (62\% in the Grade 1 at-risk controls versus 41\% in the kindergarten FR controls), rendering specificity quite low (overall 53\%). The question is whether this disadvantage of over-

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\(^3\) It is not known which of the FR controls received additional support from Grade 1 onwards.
identification in pre-readers would have great impact on school policy or on
the children involved. The rate of false negatives resulting in moderate
sensitivity seems of greater concern but relates to the cognitive risk
subsample only, thus when no information is available regarding familial
background.

6.2 Neurobiological indicators of risk

Knowledge of the risk factors in a child’s background and pre-literacy
development may result in identifying all true positives, but leaves much
room for over-identification. A relevant question is whether and to what
extent neurobiological indicators of risk may further contribute to the rate
of specificity in the identification of at-risk pre-readers when a combined
cognitive and familial risk profile has already been recognized.

6.2.1 Habituation in children with and without a familial risk

In the project proposal leading to this thesis, it was pointed out that
investigating pre-attentive learning in children at familial risk would be
scientifically relevant because “the relation between skill automatization at
the cognitive level and more fundamental neurobiological mechanisms has
received very little attention in scientific research but may be crucial in
developing the right diagnostic tools and intervention methods” (p. 3). The
specific research question regarding the role of pre-attentive learning
mechanisms (Chapter 2) was whether there would be differences in
habituation between FR and noFR children under pre-attentive conditions.
The most striking result of this study was that at pre-reading age there were
ERP differences in strength and decrease of responding to a repeated
presentation of a novel stimulus between the three subgroups. The
subgroup of noFR controls who turned out to be normal readers, the FR
subgroup comprising of children with normal reading skills and the
subgroups consisting of FR and noFR children with reading difficulties
differed significantly from each of the other two subgroups in habituation
pattern.

Looking at the individual data in order to evaluate whether our
neurobiological findings may add to early identification, we found that only
a small minority within the subgroups of normal reading noFR and poor
reading children did not fit the subgroup pattern (averaged subgroup
slopes). Among the normal noFR readers, ten out of twelve had a positive
slope indicating habituation (decrease) of the strength of the N1, whereas the negative slope in two noFR readers was indicative of a small increase in N1 amplitude (range normal noFR, 1.59 to -0.28). Of the poor readers ten out of eleven had a negative slope, indicating an increase in N1 strength, and only one showed a small decrease (range poor, 0.23 to -2.12). Although in the normal reading FR subgroup the pattern of averaged slope data indicated that N1 amplitude neither decreased nor increased in negativity, it was found that ten out of fifteen children had a negative slope indicative of habituation, whereas five showed an increase (range normal FR, 1.05 to -1.17).

Full group correlations revealed a significant relation between the individual regression slopes of N1 amplitude obtained in kindergarten and reading fluency of words \((r = .573)\) in Grade 5, see Figure 6.2, but also of pseudowords \((r = .515)\), and word spelling \((r = .492)\). Notably, when correlations were tested for each of the subgroups of readers separately, in the more heterogeneous normal reading FR and noFR subgroups no correlations reached significance. However, in the poor reading subgroup, the N1 slope correlated highly with Grade 5 word reading fluency and spelling \((r = .754, \text{ and } r = .705, \text{ respectively})\), as can also be derived from the narrow spread of the subgroup’s data points in the scatterplot shown in Figure 6.2.

Combined with familial and cognitive risks versus familial risk only, the N1 measure did not impact the 100% sensitivity rate in the FR control sample that participated in the kindergarten intervention study (see Table 6.2), as all eight poor readers at familial and cognitive risk within this sample for whom ERP data were available \((N = 24)\) showed a negative N1 slope. But specificity was raised from 56% to 69% when taking the N1 measure into account due to a small reduction (minus two) in the number of false positives, resulting in an overall hit rate of 79% (was 73%). Computations regarding the complete sample of 38 children \((\text{FR, } N = 24; \text{noFR, } N = 14)\) with subjects classified as having a risk profile based on both familial plus cognitive risk plus negative N1 slope versus a risk profile based on none to two of those risk factors yielded high rates for sensitivity (90%) and specificity (82%), and an overall hit rate of 84%. The one child identified as a false negative (noFR5 in Figure 6.1) was neither at familial nor at cognitive risk, but showed the negative N1 slope. The implication that deficits in (visual) information processing in at-risk pre-readers are involved in later reading failure and may serve as a neural marker in early identification is of
general discussion course quite intriguing. This finding also provides evidence for the theoretical assumption that a deficit in a pre-attentive learning mechanism dealing with the processing of novel information may affect attentive learning in reading acquisition, and thereby automatization of the skill.

![Figure 6.2: Scatterplot of the correlation (r = .573) between the N1 slope and word reading fluency assessed in Grade 5 in three groups of readers. The triangles represent poor reading children (N = 11), the filled black circles noFR normal reading children (N = 12) and the open circles FR normal reading children (N = 15). Vertical lines mark the reading fluency thresholds <10th and <25th percentiles, the horizontal line marks the threshold below which N1 slope is negative. Note that 8 out 11 children identified as poor reader at the end of Grade 2 were still poor readers mid-Grade 5 (thresholds <10th percentile), two poor readers improved over the years and had scores below the 25th percentile, as was the case for the one poor reader showing a small decrease in N1 strength across 14 habituation trials.

6.2.2 Picture-primed word processing in children at familial risk: Differences between low and high responders to early intervention

When investigating the neural correlates of picture-primed word identification in FR kindergartners, all provided with an early reading intervention (Chapter 4), the evidence for between-subgroup differences in neurophysiological characteristics was less distinct. In this second ERP study, the analyses concerned the overall strength of brain activation and
topographic distribution. Without the assistance of a picture prime, during the stage of phonological feature processing, word recognition elicited a difference in topography between the subgroups of low and high responders to intervention, but no difference in neural activation strength. In a later time window, related to the retrieval of lexical and semantic information from memory, the observed topographic divergence (y-axis) in responding to congruent and incongruent trials in the presence of a small change in neuronal activation may implicate a developing congruency effect in the subgroup of high responders only. However, full group correlations yielded no significant associations between later reading(-related) measures and the computed differences between conditions in activation strength and topography for the relevant time windows. As discussed in Chapter 4, small differences in neural activation between the subgroups may have been concealed due to a limited coverage of occipital and temporal channels. But for now, these data do not contribute to identification specificity, as more research is needed to link the possible subtle topographic differences in phonological and congruency processing of picture-primed words to early reading skill.

6.2.3 Application of neurobiological precursors

Our ERP studies required children to pay a visit to the laboratory and undergo an EEG recording. To be able to interpret their brain activity in response to the habituation task, we used single trial analysis, a rather laborious and time-consuming procedure. Apart from practical inconveniences and the high costs that are involved, there might be reluctance on the part of the parents or the child itself about the application of techniques like EEG and neuroimaging in assessing which risk factors for developmental dyslexia apply to this given child. More importantly, akin to the structural and functional neural precursors of dyslexia observed by other researchers (for a review, see van der Leij et al., 2013; see also Hämäläinen et al., 2013; Hämäläinen, Lohvansuu, Ervast, & Leppänen, 2014; Raschle, Chang, & Gaab, 2011; Raschle, Zuk, & Gaab, 2012) our ERP findings have limited precision and predictive utility in the individual case. Notwithstanding the progress in technology, as yet current neurophysiological findings are not sufficiently precise for screening and diagnosis of (later) literacy problems.

However, lately, three studies from the Swiss research group have begun to demonstrate the potential of visual and auditory neurobiological
markers for early prognosis by combining them with behavioural indicators. The most recent findings concerned the development of neural correlates of print processing in poor and normal readers prior to and following an 8-week training in kindergarten in letter-sound correspondences. Brem et al. (2013) found that a pre-training N1 amplitude difference between the conditions words and false fonts measured over the right hemisphere improved the prediction of reading outcomes in Grade 2 when combined with the two behavioural kindergarten measures that yielded significant predictions rapid naming and non-verbal IQ, thereby increasing the explained variance from 27% to 40%. In a related ERP and fMRI study, Bach, Richardson, Brandeis, Martin, and Brem (2013) reported that the post-training recorded differential N1 print sensitivity (words minus symbols), along with the corresponding activation in the visual word form system, and the behavioural measure rapid naming, could discriminate poor Grade 2 readers (threshold < 25\textsuperscript{th} percentile) from normal readers (threshold > 40\textsuperscript{th} percentile), resulting in a correct classification of 94.1% (sensitivity = 100%; specificity = 90.9%). The third study which already dates a few years back involved a late mismatch negativity (LMMN) indicating hemispheric lateralization of automatic phoneme processing (Maurer et al., 2009). This neurophysiological measure obtained just before the onset of reading instruction in both FR and no-FR children appeared to be the only significant predictor for normal reading outcome in Grade 5. In separate analyses of FR subgroup, the LMMN combined with the behavioural measure rhyming failed to predict severe reading problems (threshold < 10\textsuperscript{th} percentile), but these two measures did predict poor reading (threshold < 20\textsuperscript{th} percentile), correctly classifying 81% of the at-risk children (sensitivity = 93.3%, specificity = 50%).

At this point of time there may even be dyslexic dysfunction revealed by neuroimaging (e.g., Landi, Mencl, Frost, Sandak, & Pugh, 2010) that cannot be observed with behavioural measures. Again we have to stress that despite the potential utility, such findings have yet to impact the diagnosis in case of apparent difficulties. In regard to developing the right diagnostic tools for children assumedly at-risk for reading difficulties, there is progress, as reviewed above and given our habituation data. Is it just a question of time before neurobiological markers for dyslexia can be used to make a diagnosis on a case-by-case basis and help us understand the hitherto unexplained causes for reading failure in children recognized as false negatives? Which relates to the 5 to 10 per cent of children mentioned by Scarborough (2009),
like the one child out of 15 (noFR5 in Figure 6.1) detected in our noFR subsample who turned out to be a very poor reader. A negative N1 slope was found in this particular child when still a pre-reader, but he was neither at familial nor at cognitive risk. So there would not have been any cause for further (neurological) assessment before his difficulties with learning to read became apparent in first grade. Also, once useful diagnostic ERP and fMRI precursors have been identified, will a referral to a specialized clinic to find neurobiological confirmation of the manifest symptoms of the disorder become standard practice given the financial and personal costs? Until we have answers to these questions, the best option is to rely on familial risk and behavioural indices that best predict future literacy acquisition - albeit not in all cases.

6.3 Intervention for children at-risk for reading difficulties

6.3.1 When is the right time for ‘early’ intervention?

Since the first influential prevention studies in children at familial and/or cognitive risk, in Denmark (Lundberg, 1994; Borstrøm & Elbro, 1997), England (Hatcher, Hulme, & Ellis, 1994), Germany (Schneider, Küspert, Roth, Visé, & Marx, 1997), the United States of America (Torgesen et al., 1999), and Australia (Byrne, Fielding-Barnsley, & Ashley, 2000), it has been increasingly acknowledged that preventive interventions are very much needed to reduce the number of children with (avoidable) reading failure (Torgesen, 2001). As from then, quite a few other early interventions have been carried out of which several took place in the Netherlands, (for a review, see van der Leij, 2013), one of which is the Chapter 3 study. What has been learned from these studies is that improved pre-literacy skills do not readily transfer to prevent reading difficulties. Also, as observed earlier by Elbro and Scarborough (2004), even in case programs have proven their efficacy, the proportion of children who fail to respond appears to be considerable and poses a great challenge in early intervention research.

Obviously, interventions gain in effectiveness, when the active ingredients of training are identified. However, based on her meta-analysis of kindergarten predictors, Scarborough (2009) noted that although there appears to be much continuity in literacy development over the years, it is still difficult to tell which (pre-)literacy developments are “necessary and sufficient” for reading acquisition to be successful. One of the findings commonly reported in the literature reviewed by Scarborough is that some
children identified as being at-risk in fact develop normal literacy skills (the so-called false-positives), whereas others not identified as at-risk become poor readers (the so-called false-negatives). As already discussed in section 6.1, the results of our intervention studies reported in Chapter 3 and 5 are no exception in this respect.

Another common finding is that despite the successful improvement of their early literacy skills FR children still remain at high risk for developing literacy difficulties. This was very much the case for our kindergarten sample consisting of two FR groups of which only one group received the early intervention during 14 weeks. At the end of kindergarten, the literacy development in the FR intervention group was as far advanced as in the noFR controls, and the FR control group was outperformed on all relevant literacy skills (letter knowledge, decoding accuracy, and phonemic awareness). Important to recall is that many children with initially poorest pre-literacy skills benefitted largely from the intervention. Nonetheless, the head start of the intervention group did not result in a normal reading acquisition in a substantial proportion of the participants. Looking at the outcome in Grade 5, both FR groups with and without early intervention comprised an equally high percentage of children who went on to develop dyslexic problems. Of the trained FR children no less than 38.7 per cent versus 38.5 per cent of the untrained FR children scored in the bottom 10 per cent on word reading fluency.

Apart from the notion that in dealing with subjects at high risk for dyslexia our expectations with respect to the outcome obviously were too optimistic, what else is there to learn from the effects of early intervention being completely washed out within a half year of formal reading instruction? Assuming that the intervention in itself was sufficiently effective, and contained the right ingredients, could it be that the period of intervention was too early to have had sufficient impact? The intervention may have induced a rudimentary level of literacy, such that children did acquire an initial understanding of the alphabetic principles and an enhanced grasp of the phonemic structure of words. However, since the intervention period had no immediate follow-up due to a break of several months (including a summer holiday), it may have been the case that the still relatively unstable gains did not abide and failed to transfer to the formal context of first grade reading instruction.

It has been argued before (O‘Connor, Bocian, Sanchez, & Beach, 2012) that earlier intervention is not always better than later intervention.
But it seems not that likely that the outwardly beneficial effects of the short early intervention in kindergarten were washed away due to a lack of developmental readiness. Seven out of nine participants scored not only low on reading at the first test occasion after the onset of formal instruction (mid-Grade 1) but also at the last (mid-Grade 5). Since they did not benefit from the intervention nor responded appropriately to formal instruction, most of them can be called true non-responders. Also, although at the individual level large differences between the participating FR children were found, just a small minority seemed not quite ready to transfer newly acquired literacy skills to the skill of (automatized) reading. That the intervention nonetheless did not overcome the enhanced susceptibility for reading difficulties in a too great part of the children may rather reflect other limitations, such as the quantity or duration of the delivery.

### 6.3.2 Is sustained early intervention preferable to a short one?

Four different Dutch interventions (van der Leij, 2013) were executed aimed at strengthening phonological awareness and letter-sound associations in kindergartners, each with a different emphasis and approach, three FR samples from different regions of the country, all four intervention groups showed clear post-intervention improvement compared to FR controls, but all four studies shared the worrisome outcome that in a nontrivial proportion of participants the enhancement in pre-literacy skills did not serve as a solid basis for the acquisition of adequate reading fluency. These Dutch interventions all resembled one another in terms of duration: they were delivered during a relatively short period of 14 weeks, summing up to approximately 12 hours of instruction. The conclusion was that the intervention period was obviously too short to install improvements in the longer run, and selection based on FR alone included too many children with relatively low risk of developing RD.

Recently, a British intervention study (Duff et al., 2014) was carried out with children at-risk for dyslexia who were identified in their first or second year at school as having the weakest reading skills. After a 9- or extended 18-week period of intervention at school delivered by trained teaching assistants, the enhanced emergent literacy skills did not result in normal reading skills at group level. This study is particularly interesting because it demonstrates that the reported null results and those in the Dutch

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4 Two studies used a split-sample collection.
interventions were not so much due to mode of delivery (school-based versus school- or home-based) and timing of the intervention (second semester of Grade 1 versus second semester of kindergarten), although according to the authors, the classroom literacy approach may have intervened in inducing larger effects. Rather, in our opinion for Dutch and British interventions alike, delivery was just too short, supporting our conclusions based on the study of Chapter 3 (Regtvoort & van der Leij, 2007).

The absence of long-term transfer in the kindergarten participants indicated that to make full use of the potential of the computerized intervention program, extension beyond the initial stage of learning to read was essential to also include instruction in larger orthographic units both consistent as inconsistent to emphasize multiple orthographic-phonological mappings. Such a sustained design, used in the school-based intervention of the study in Chapter 5, also better suits the educational needs of at-risk children. Foremost they require supplementary as well as repeated instruction (van der Leij, 1981) - afforded by human and computer-assisted tutoring - to enhance the number of successful exposures given their difficulties with the formation of high quality orthographic representations (Ehri & Saltmarsh, 1985; see also, Compton, Miller, Elleman, & Steacy, 2014, who recently arrived at the same conclusions). With additional practice, poor readers may obtain an adequate level of direct word recognition and automatization (Mol & Bus, 2011; Share & Shalev, 2004). As reported in Chapter 5, this approach showed its effectiveness in children identified on the basis of a cognitive risk profile at the onset of reading acquisition - on the condition of intervention completion. At the end of Grade 3, the proportions of good readers (threshold > 75th percentile) and of poor readers (threshold < 10th percentile) within the intervention subgroup that completed the program (N = 40; 56% of intervention subjects) approached the reference group’s proportions. However, the substantial number of participants who failed to complete the program (N = 31) signifies that monitoring adherence to the instructional protocol becomes even more important in case children receive intervention over an extended period. Protocol deviations pose even a greater threat to treatment integrity in sustained interventions relative to short(er) ones. Also, the more

\[ ^5 \text{Low pre-literacy skills were used as the only inclusion criteria, whereas there were no exclusion criteria. So, it concerns a heterogeneous sample of first grade at-risk readers with respect to personal and familial characteristics like age, IQ and second language background. Information about a familial history of RD was not available.} \]
complex the implementation, e.g., involvement of third parties, the lower the expected adherence to the protocol may be (Gresham, Macmillan, Beebe-Frankenberger, & Bocian, 2000).

In a separate study highly related to the one in Chapter 5, children at high cognitive risk had the onset of sustained intervention in kindergarten. While schools were responsible for the implementation, the parents at home were the tutors delivering the program prior to formal instruction. From first grade onwards until halfway second grade, non-professionals at school (i.e., higher grade students, volunteers) assumed the role of tutor. Within this particular kindergarten sample, 38 (62%) of the at-risk children completed the intervention within the designated period of two years, whereas the remainder 23 (38%) children did not. This later percentage matched with the incompletion percentage of 44% in the Chapter 5 subsample that started out in Grade 1 - suggests that involving the parents did not lower treatment integrity in this respect. In comparing the Grade 2 word reading fluency outcome in both kindergarten and Grade 1 subsamples that completed the program, Regtvoort, Zijlstra, and van der Leij (2014) reported that in the subsample that started intervention earlier in literacy development the proportion of poor readers (threshold < 10th percentile) was lower than in the subsample that started half a year later (start kindergarten, 5% versus start Grade 1, 13%), and the proportion of good readers (threshold > 75th percentile) much higher (start kindergarten, 47% versus start Grade 1, 22%).

6.4 Implications

6.4.1 Implications for future research

For early identification of children at risk for poor reading outcomes, past research has consistently shown which behavioural correlates of reading development are the most critical prior to or shortly after the onset of literacy instruction. As has been argued before, with respect to neurophysiological precursors this is not yet the case. Over the last decade a growing body of evidence has suggested that specific neural precursors may contribute to early prognosis. To clarify which functional or structural measures are the most predictive and prognostic - in itself and for complementing behavioural measures - systematic review involving both longitudinal prospective and retrospective studies is strongly recommended. Promising candidates are in particular early ERP components – which have
been reported to differentiate between kindergartners who do and do not develop reading difficulties and can be collected more easily than for instance fMRI indices – both in the auditory (Hämäläinen et al., 2013; 2014; Maurer et al., 2009) and visual modality (Bach et al., 2013; Brem et al., 2013; Regtvoort, van Leeuwen, Stoel, & van der Leij, 2006, see also Chapter 2). It might also be interesting to further investigate the congruency effect in pre-reading children, not only with respect to visual or auditory linguistic processing like integrating letters and speech sounds (cf. Blau et al., 2010), but also to processing of non-linguistic information e.g., integrating natural objects and features, or motion and sounds so as to examine whether at-risk children differ from not-at-risk children in how efficient they access and integrate information from different visual and auditory neural systems and subsystems.

With respect to early intervention, we might say that there is strong consensus among researchers and educators what an appropriate intervention should target. Letter-sound correspondences, phonological awareness, preferably in the context of reading, consistent and sustained practice with a focus on repetition to foster accuracy and automaticity from smaller to larger orthographic units. The question is whether the early interventions that have proven to be successful at the group-level are sufficient flexible to accommodate the instructional needs of the individual poor reader. Heterogeneity in strengths and weaknesses in (pre-)literacy skills, and in learning characteristics implies the need for specific diagnostic tests and differential (modes of) instruction, and continuously monitoring progress. So it would be worthwhile to construct both adaptive testing tools and adaptive interventions to examine their impact on various periods in reading acquisition. While going through the phases of emergent, beginning and advanced decoding, children may differ in offset and the extent to which they should be provided with explicit and additional instruction, as well as in type and intensity of support. For instance, for those with low phonological processing skills, it would be helpful to prolong the practice of using pictures so as not only to stimulate letter knowledge, but also to allow for enhanced access to the phonological codes of novel or lower frequency words. Also, for some at-risk readers an individualized computerized intervention may as well be effective, while others may thrive only with the (additional) support of a human tutor. However, in all cases, continuous monitoring of developmental progress is crucial.
With regard to intervention content, we endorse the view propagated by Compton et al., 2014, that to promote the learning mechanisms of poor readers a well-balanced training corpus of words tuned to the specific features of the Dutch orthography should be created and examined in interventions with proven efficacy. Such a corpus should consist of various sets of words varying systematically with respect to orthographic and phonological correspondences, including both high-frequency complex words, which can be processed as complete word-specific representations, as well as words comprising subword orthographic-phonological connections of multiple sizes (phonemes, diphones and morphemes) that are invariant to position within the word and surrounding letters. The training corpus of words used in our intervention program was formed according to these principles and may serve as a basis for further development.

6.4.2 Practical implications

For educational practitioners this thesis offers three take-‘school’ messages: 1) no quick fix but long-drawn-out for children at-risk for reading difficulties; 2) no magic involved but plain commitment; and 3) at-risk children will not learn reading from implicit learning but need explicit, focused instruction and practice. While these positions may speak for themselves, below we will consider them in more detail.

No quick fix but long-drawn-out

The two intervention studies described in Chapter 3 and 5 together provide evidence that a sustained intervention is to be preferred above a short one. An intervention that is restricted to a limited period of time cannot adequately address the needs of poor and severely poor readers to raise their (pre)literacy skills to satisfactorily levels. Ideally, intervention should cover an extended period, encompassing broad instruction in basic decoding complemented with prolonged practice in accurate and fluent recognition of more complex word structures.

Such an extended intervention period also entails that kindergarten and early grade teachers remain aware of students showing deflected developmental trajectories. Some at-risk students may not have been identified in kindergarten, apparently because their background characteristics and pre-literacy development may not have given rise to concern. For them, the act of reading itself, e.g., integration of letter knowledge with skills of segmenting and blending and/or the necessary transition to direct word recognition may pose the key stumble blocks. In
those cases, actual progression is not in accord with the expected progression. In contrasting cases, children tend to stay behind in kindergarten, yet gain in early proficiency as a result of individualized help, and accordingly might or might not stay on course in the years to come. To accommodate different kind of trajectories, it is therefore advisable for schools to adopt a stepwise procedure in which identification and intervention are entwined and bear on each other as a coherent whole. This procedure should be spread out over several years (from kindergarten through to Grade 4) conform the RTI procedure (Fuchs & Fuchs, 2006).

In the first step, which can already take place prior to the children’s arrival at school, schools gather as much information as possible about the student’s familial and socio-cultural background. In the context of family risk, not only the reading skills of a parent with dyslexia seem to signal increased risk for his or her offspring. Reading skills of both parents should be taken into account since non-dyslexic parents in the families of affected and unaffected FR children are also found to differ on reading ability (van Bergen, de Jong, Maassen, & van der Leij, 2014). Presumptive indications of risk, like for instance having relatives with (severe) reading or spelling difficulties, or a home language different from Dutch, as well as classroom observations by kindergarten teachers prompts schools to screen for identification (step 2) according to a risk profile comprising letter knowledge, phonological awareness and naming speed. In step 3, all children who satisfy the cognitive and FR criteria receive early intervention on individual basis, at home or at school. Just before the onset of formal reading instruction, response to intervention is being assessed in step 4. Of the children with a cognitive risk profile, only those noFR children who do not show the expected progress and all FR children continue intervention (step 5). After about one and a half month of formal instruction, the early decoding skills of all students whether or not receiving intervention are being assessed at step 6, so as to identify in an early stage the children who are falling behind (see e.g., O’Connor, Bocian, Sanchez, & Beach, 2012) and provide them with extra support as well. Mid Grade 1, in step 7, all students are again tested on word reading fluency. By then, it will be quite clear which children have (very) poor decoding skills (thresholds < 10th and < 25th percentiles). They are the ones who continue intervention until they have reached a pre-set level. Across first and second grades, however, it is essential for teachers to periodically monitor (lack of) progress in no-intervention students, and act accordingly.
No magic involved but plain commitment Even the best intervention programme is only as good as its application. In other words, a programme with proven efficacy will result in significant improvement on condition that it is implemented and delivered as intended. This means that the efforts of schools (supervisors, teachers) and tutors matter (considerably). As shown by our intervention study described in Chapter 5, adherence to the protocol (or not) may have long-term consequences for reading outcome.

No implicit learning but explicit, focused instruction and practice According to Ehri (2005), knowledge of orthographic to phonological mappings must be acquired through either explicit learning or implicit learning and practice. In line with our habituation data of Chapter 2, normal reading children have learning mechanisms that allow for the acquisition of knowledge about environmental regularities, which is not driven by conscious strategies (e.g., Reber, 1989), and can be transferred to novel tasks (Pavlidou & Williams, 2014). In poor readers, learning of sequences appears to be impaired, but only in case learning is implicit and not intentional (Jimenez-Fernandez, Vaquero, Jimenez, & Defior, 2011). Given their tendency to process words as whole entities, i.e., they are less sensitive to specific spelling patterns of words, poor readers need more exposures than normal readers to recognize the word’s pattern (Reitsma, 1983). Hence, children who struggle with learning to read fluently need explicit and focused instruction followed by (repeated) practice at all levels of reading fluency (letter-sound conversions, orthographic patterns, whole words) systematically linked to the context of specific words (Compton et al., 2014) to forestall persistent letter-by-letter decoding. In addition, they need multiple exposures to all word-forms to achieve (minimum) mastery at each stage of reading development. In the case of children at-risk for reading difficulties, only early interventions that target the systematic build-up of a well-defined orthographic lexicon allow for adequate reading proficiency.

Reducing variation in reading and spelling ability can be brought about through enhancing the literacy skills of children at early (or not so early) risk for difficulties in learning to read. What is required though, is supplementary, focused instruction provided systematically and extensively by schools along with progress monitored consistently and support adjusted accordingly, challenging at-risk children to practice - a lot -, with carefully selected material in order to develop automaticity through overlearning. So that proficiency in reading will escape no one.