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Phonology and Literacy? Follow-up Results of the Utrecht Dyslexia and SLI Project

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Abstract

This chapter assesses the relationship between phonology and literacy by comparing children at family risk of dyslexia, children with SLI, and typically developing children on preschool phonological measures and literacy and phonological skills at eight years of age. As expected, 37% of the family-risk group and half of the SLI group show literacy difficulties. Surprisingly, there are no clear connections between the preschool phonological measures and literacy outcomes. The SLI group generally obtained the lowest scores on the phonological measures, followed by the family-risk group, but these were not necessarily linked to literacy outcome. In contrast, non-word repetition and rapid serial naming at 8 years of age do show a relationship with literacy outcome, as well as group. The study thus 1) shows similarities and differences between the family-risk and SLI groups, 2) confirms that phonology is a multidimensional skill that can affect literacy in different ways, 3) points to compensatory domains across time and thus 4) can be accommodated within multi-risk models of dyslexia and SLI.

INTRODUCTION

Dyslexia is a complex neurodevelopmental disorder that has received investigation at many levels spanning genes, brain, and behaviour. The present chapter presents an investigation of the cognitive deficits of dyslexia based on speech-related data.

There is now good evidence that aspects of early language development can reveal difficulties that lead to later literacy problems. Primarily these difficulties include deficits in phonological awareness (PA), phonological (or verbal) short-term memory (vSTM), and rapid naming (RSN; Wagner & Torgesen, 1987) and are indicative of a core phonological deficit, i.e. difficulties with encoding, storing, or retrieving speech. Importantly, studies of children with a familial risk of dyslexia (i.e., children with at least one dyslexic parent) have demonstrated that phonological difficulties *precede* the onset of literacy instruction and therefore suggest that poor phonological skills might be precursors of dyslexia (e.g. Boets et al., 2010, de Bree, Rispens & Gerrits, 2007, Locke et al., 1997, Lyytinen et al., 2004, Scarborough, 1990, Snowling, Gallagher & Frith, 2003). Examination of the cognitive profiles of both family-risk (FR) children with, and without, literacy difficulties (e.g. Boets et al., 2010, de Bree, Wijnen & Gerrits, 2010, Elbro, Borstrøm & Peterson, 1998, Pennington & Lefly, 2001, Snowling, Muter & Carroll, 2007) shows that this core phonological deficit affects both poor readers and typical readers in FR children (indicative of a genetic liability), and also that this deficit can either be aggravated or compensated by other (linguistic) skills.

Coming from a different perspective, researchers exploring the underlying causes of specific language impairment (SLI) are incorporating literacy development in the frameworks proposed for this order. SLI is defined as an impairment in acquiring language despite normal hearing and non-verbal abilities, and adequate exposure (Leonard, 1998). Interestingly, many children with SLI go on to develop literacy difficulties (e.g. Bishop et al., 2009, Vandewalle et al., 2010, van Weerdenburg et al., 2009). One key issue that has received attention recently is whether phonological deficits in SLI are causally related to literacy difficulties, or whether such deficits can exist without any concomitant

reading problems. More specifically, there is debate as to whether children with SLI display phonological processing difficulties, or whether only those SLI children with additional literacy difficulties do so (e.g. Bishop & Snowling, 2004, de Bree et al., 2010, Catts et al., 2005, Rispens & Parigger, 2010, Scheltinga, van der Leij & Beinum, 2003, Vandewalle et al., 2010).

A direct comparison between the spoken language and literacy abilities of children with dyslexia and children with SLI is thus warranted. In fact, there has been a shift towards the investigation of the comorbidity of dyslexia and SLI, rather than regarding the disorders as unidimensional and separate (e.g., Bishop & Snowling, 2004, Catts et al., 2005, Pennington, 2006, Tallal et al., 1999). Indeed, research on children with pure disorders (dyslexia-only, and SLI-only) comparing them with children with co-morbid difficulties (dyslexia+SLI) is burgeoning (e.g. Bishop et al., 2009, Fraser, Goswami & Conti-Ramsden, 2010, , Marshall & van der Lely, 2009, Robertson et al., 2009).

Dyslexia and SLI: A Longitudinal Approach

A key issue pertaining to the relationship between dyslexia and SLI is the nature of the linguistic abilities of these two groups prior to the onset of literacy instruction. It is important for research to be directed toward this issue in order to map the developmental trajectory of language and literacy abilities, as well as the mutual impact the two abilities can have on each other.

In the Utrecht longitudinal study, language abilities of children with a familial risk of dyslexia were compared with those of children with SLI, as well as typically developing children without literacy difficulties in their family. These three groups of children were seen four times between their third and fifth birthday, and were presented with tasks probing speech perception and production (de Bree, 2007, de Bree et al., 2007, 2010, Gerrits, 2003 Gerrits & de Bree, 2009), morpho-phonology (de Bree & Kerkhoff, 2010), grammatical sensitivity (van Alphen et al., 2004, Wilsenach, 2006) and vocabulary. The preschool findings reveal similarities between the FR and SLI groups in terms of

shared areas of difficulty, with respect to non-word repetition, sentence repetition, speech production, speech sound categorisation, and grammatical sensitivity. However, it is important to know if and how these preschool language abilities relate to subsequent literacy skills, and whether this pattern is the same for affected and unaffected children in family-risk and SLI groups.

Follow-Up Data A first analysis of the follow-up data was presented in de Bree et al. (2010). The non-word repetition (NWR) abilities of 4-year-old children at-risk of dyslexia and children with specific language impairment (SLI) were related to their reading abilities at age eight: the SLI group obtained the lowest NWR score, and the family-risk group performed in-between the typically developing (TD) and SLI group. Approximately half of each of the family-risk and SLI groups showed reading difficulties; we will refer to these as Family risk poor readers (FR-PR) and SLI poor readers (SLI-PR) to distinguish them from those in each group with normal reading (NR). Whereas pre-school NWR abilities correlated with literacy outcomes in the FR group, this was not the case for the SLI group. Rather, for children with SLI, phonological processing was poor regardless of literacy outcome. These findings thus lend support to the notion that dyslexia and SLI are disorders with different trajectories to literacy disorder.

The non-word repetition data prove valuable for understanding the relationship between dyslexia and SLI. It is important, however, to also assess the performance on the related measures of speech perception, production, and processing in order to understand the phonological deficit in the two disorders. The questions to be addressed here are whether difficulties arise on the same tasks and to the same extent, and whether they relate to literacy outcomes. The tasks that will be discussed here include: 1) categorical speech perception, which has shown conflicting results for presence or absence in dyslexia and SLI (see e.g. Robertson, 2009); speech production, which only indirectly relates to literacy outcome but does mark language delay (e.g. Carroll et al., 2003, Raitano et al., 2004); 3) mispronunciation detection as a measure of phonological representations (e.g. Carroll & Snowling, 2004); and 4) rhyme oddity task as a measure of phonological awareness (e.g.

Goswami & Bryant, 1990). Furthermore, it is important to consider the outcome, not only for literacy but also for non-word repetition at the age of 8 years. In fact, there is already a suggestion for longitudinal analysis (de Bree et al., 2010) that NWR at 4 age does not predict literacy outcome. Arguably, this might be because of the reciprocal relationship between NWR and reading development (e.g., Nation, Cocksey, Taylor & Bishop, 2010, Rispens & Parigger, 2010), such that NWR performance is bootstrapped by reading development. In this light, it was predicted that at 8 years, only SLI children with literacy difficulties (SLI-PR) would show poor NWR at age 8, and also that a correlation between NWR and literacy would be found at this age.

A final and related issue is the role of rapid serial naming (RSN) in literacy outcome. This measure contributes to literacy (e.g. de Jong & van der Leij, 2003). Moreover, Bishop et al. (2009) found that rapid serial naming was predictive of the presence or absence of literacy difficulties in a sample of nine-to-ten- year-old children with SLI who had poor non-word repetition. Thus it was SLI children with both NWR and RSN difficulties who presented with reading difficulties, and not those with deficits only in NWR. Vandewalle et al. (2010) extended these findings to a younger age, and reported that RSN skills of Flemish children with SLI at kindergarten age predicted their literacy outcome. RSN abilities thus also need to be addressed in the Utrecht family-risk and SLI children, to assess whether RSN is a protective factor for literacy in these groups.

In sum, in order to gain a better understanding of the relationship between phonology and literacy and between dyslexia and SLI, the present study assessed whether: 1) phonological skills tapped between three and five years of age are early predictors of later literacy disorders; 2) NWR difficulties are persistent across age; and 3) rapid naming is a protective or aggravating factor for literacy.

METHOD

Participants

Three groups of children participated in this study, children with a familial risk of dyslexia (FR, n=60), children with SLI (n=30), and a typically developing group (TD, n=30). The FR children were children of whom at least one of the parents had literacy difficulties as measured by standardised reading and phonological processing scores (including a non-word spelling task, a timed word and timed non-word reading task, a NWR task, and rapid naming). In order for a child to be included in the at-risk group, the parent had to show poor performance on all the tasks, except on an additional verbal competence task, as this is often a relative strength for highly-educated dyslexics, in contrast with their reduced reading and spelling abilities. Specifically, performance on the timed word reading or timed non-word reading task had to be on the 10th percentile, or on the 25th percentile on both timed reading tasks, and a discrepancy of at least 60% between verbal competence and performance on the timed reading tasks (based on criteria from the Dutch Dyslexia Programme, see Koster et al., 2005). The FR children did not include children whose parents or siblings had a history of language impairment.

Children with SLI were recruited through speech therapists and schools for children with severe speech and language difficulties. Prior to the start of the project, they had been classified as SLI after extensive assessment of speech and language abilities by certified speech pathologists. All children had to have normal hearing, absence of neurological difficulties, and normal non-verbal IQ (>75). We only know of one child in this group who had a dyslexic parent. The TD children were all children without literacy and language difficulties in their family and without language or behavioural difficulties of their own.

The children were all presented with the same test battery, but, as testing was free of pressure, not all children completed all tasks in one testing phase. This means that different numbers are present for each task (see de Bree, 2007 for more information on the samples of children).

Measures

Preschool Speech Measures The preschool phonological processing tasks included categorical speech perception, speech production, mispronunciation detection, and rhyme oddity. These will be briefly described below (see also Table 1).

Categorical perception was tested between 3;6 and 4 years with the traditional two-alternative forced choice categorisation task. The stop-consonant acoustic continuum ranged from the word /pɔp/ to /kɔp/ ('doll' and 'cup'). These words were natural utterances produced by a male speaker of standard Dutch. Stimuli in the continuum between the two utterances were obtained by interpolation between the relative amplitudes of the spectral envelopes of the words. Stimulus generation resulted in a continuum of seven stimuli that sounded completely natural and convincingly like utterances of the original speaker. Categorisation behaviour was reflected in categorisation slope, with a steeper slope reflecting better categorisation (see also Gerrits and de Bree, 2009).

A picture naming and matching task as measure of *speech production* was also tapped between 3;6 and 4 years. Targets included mono-, bi- , tri- , and quadrosyllabic words checked for age of acquisition. Percentages consonants correct are reported. More data is assessed here than in Gerrits and de Bree (2009).

A *mispronunciation detection* task was presented to the children at five years of age. The proportion correct was calculated of the 12 maximal (place of articulation, manner, and voicing, e.g. zebra as pebra) and 12 minimal mispronunciations (place of articulation, e.g. zebra as vebra) in the onset (see van Alphen et al., 2004).

Phonological awareness was assessed through a *rhyme oddity* task at five years of age. Children had to select the odd one out of 20 series (e.g. jas-glas-tas-trui) and the proportion correct was calculated (see de Bree, 2007).

Insert Table 1 here

Measures at 8 years of age Four different literacy-related tasks were presented to the children, two timed-reading tasks (a one minute word reading task (Brus & Voeten, 1972), and a two minute non-word reading task (Van den Bos, Spelberg, Scheepstra & de Vries, 1994), and two spelling tasks (spelling dictation (van den Bosch et al. 1993) and spelling selection (Horsley 2005)).

Non-word Repetition. The non-word repetition task of De Jong (1998) was used, containing 48 items that lead to a raw score.

Rapid serial naming. The rapid digit naming task by Van den Bos et al. (2002) was used for RSN wherein 50 digits had to be named as quickly and correctly as possible. The RSN-score is calculated as the number of symbols named per second. The higher the RSN-score, the faster the naming speed.

RESULTS

Literacy Outcomes of Children at Family-Risk of Dyslexia, Children with SLI, and Normally Developing Children at 8 years

Data of children who completed the literacy battery as well as the NWR and RSN tasks at 8 years of age are 24 typically developing children, 44 family-risk children, and 17 children with SLI. A MANOVA on the literacy data in Table 2 (phonological data will be discussed in the next section) shows a main effect of the phonological tasks, as well as significant differences between the groups on all tasks with the exception of the timed non-word reading task ($p = .1$), despite clearly visible differences between the groups (but note the small effect size). On all measures, the family-risk and SLI group performed similarly to each other. For spelling selection, the TD group performed better than both the FR group and the SLI group. For spelling dictation, the pattern was similar but the difference between the TD group and the SLI group was not statistically significant ($p = .09$). Finally, for word

reading, the TD group scored higher than the SLI group. These findings are consistent with the literature, in that both FR and SLI groups will do more poorly in terms of literacy outcomes.

Insert Table 2 here

A composite literacy score was computed by summing the z-scores for the four literacy tasks. A literacy deficit was defined as performance at or below one standard deviation of the mean literacy composite score for the TD group. On this criterion, as expected, the majority of TD children read well (19/24=79%), whereas fewer children in the family-risk (24/38=63%) and SLI groups did so (7/14 = 50%). Again, these findings resemble those found in previous family-risk studies, where between 30 and 60% of FR children become diagnosed with dyslexia, as well as studies of SLI, in which a substantial percentage of children with SLI show literacy difficulties.

Preschool Phonological Skills and Literacy

Table 3 shows performance on the four tasks presented at preschool age for the three intake groups (TD, Family-risk, and SLI). Significant group differences emerged for categorisation slope for speech discrimination, percentages consonants correct for speech production and correct responses for mispronunciation detection, and there was a trend for rhyme oddity correct to differ. The pattern of performance was generally; TD > Family-risk > SLI. However, in contrast to the NWR results (de Bree et al., 2010), this pattern was not borne out statistically in the tasks reported here. Specifically, on the categorisation and mispronunciation detection tasks, the FR and SLI groups did not differ from each other. The FR group differed from the TD group on both tasks, whereas the SLI group differs from the TD group on categorisation (and marginally so on the mispronunciation detection task, p=.052). On the speech production task, the TD and FR group both perform better than the SLI group,

matching the intake criteria of diagnosed language difficulties of the latter group. Effect sizes are small to medium for the preschool tasks.

Insert Table 3 here

In order to ascertain whether children in the FR and SLI groups carry a similar 'risk' of reading difficulties which may be compensated or aggravated during development, we next analysed performance on the phonological measures across 6 subgroups classified in terms of 8-year-old outcome (TD normal readers (TD-NR), TD poor readers (TD-PR), Family-risk normal readers (FR-NR), Family-risk poor readers (FR-PR), SLI normal readers (SLI-NR), and SLI poor readers (SLI-PR), see Table 4). It should be borne in mind that the numbers of children in each subgroup differs across the phonological tasks because of missing data. Visual inspection establishes that the SLI-RP group always performed most poorly and that there was a gradual decline in scores from TD > FR > SLI on the categorical perception and rhyme oddity task.

Insert Table 4 here

Consistent with the findings for NWR reported by de Bree et al. (2010) there were significant group effects on ANOVAs with the task results as dependent variables and group (6 groups) as between-subjects variables for the categorisation slope, mispronunciation, and picture naming score; differences in rhyme oddity were not significant despite discernible differences between the groups. No sub-group differences were found for the mispronunciation data, probably caused by the ceiling scores for the TD groups and the substantial variation in the SLI groups. For speech categorisation, the SLI-PR group and the FR-PR group scored less well than the TD group. For speech production, the

SLI-PR group scored less well than the FR-PR and TD groups. Outcomes remain the same when the TD-PR data are excluded from the analyses. Effect sizes are medium and small.

When these analyses are run on score for the task as dependent variable and group (TD, FR and SLI) and literacy (good and poor) as between-subjects factors, the pattern of findings is the same, with main effects of group for all tasks except the rhyme oddity task, but no effect of literacy outcome. The only task that shows a marginal effect of literacy outcome is NWR (p=.086). There are no interactions between group (TD, FR, SLI) and literacy outcome (good or poor) on any of the tasks. The speech tasks, excepting the rhyme oddity task, thus show differences between the preschool intake groups, but these are not related to literacy.

These analyses do not support a uniform connection between the preschool phonological tasks and literacy outcome at 8 years of age. Furthermore, there were no strong correlations among the measures either within or across groups. Together, these findings suggest that there is no consistent link between the preschool phonological measures and subsequent literacy abilities.

NWR and RSN at 8 Years and Literacy

The previous section assessed whether phonological difficulties can be regarded as precursors of dyslexia that exist prior to literacy instruction. Here, we assess concurrent relationships between literacy and both NWR and RSN at 8 years of age (Tables 2 and 5).

First, an important question is whether NWR performance at preschool age is correlated with NWR at 8 years of age. A high positive correlation for the overall data (r=.777, p<.001) as well a moderate correlation for the TD (r=.505), family-risk (r=.663), and SLI groups (r=.658) separately (all with p<0,001) attests to the stability of this measure over time.

Turning to the group differences, as expected, these were significant for both NWR and RSN. For RSN, the FR-PR group performed worse than the TD groups. For NWR, The SLI-PR group obtained lower scores than both FR and TD groups. An ANOVA with mean score on RSN as dependent variable

and group (TD, FR, and SLI) and literacy outcome (good and poor) as between-subjects variables shows a main effect both of group (RSN fastest for TD > SLI > FR) as well as literacy outcome (good literacy faster RSN that poor literacy). For NWR at 8 years, a main effect of group is also found, with TD > FR > SLI, as well as an effect of literacy (good > poor). There are no interactions between group and literacy outcome on either task. The phonological measures at 8 years of age thus show a relationship to group, as well as to literacy outcome.

Insert Table 5 here

A second analysis ran correlation analyses between the NWR and RSN and the composite z-score of the literacy tasks. A moderate overall positive correlation was found for NWR and RSN coupled with literacy (see Table 6). RSN was only correlated significantly for the FR group separately, while NWR was correlated to literacy for both the FR and TD groups.

Insert Table 6 here

DISCUSSION

The main aim of this study was to assess whether preschool phonological difficulties in children with a familial risk of dyslexia, and children with SLI could predict later literacy difficulties. Additional questions included whether the areas and severity of the difficulties were the same for both groups, whether phonological processing difficulties would also be present at school-age, and whether rapid naming might also be a protective factor for literacy.

The literacy outcomes of the family-risk and SLI children agree with those found in the literature, since from the total sample 37% of the FR and 50% of the SLI group could be labelled poor readers. Furthermore, the FR and SLI groups performed less well than the TD group both on the

preschool and school-age measures of phonology, indicating underlying phonological difficulties in both groups across ages. The finding of a strong correlation between preschool and school-age NWR further supports this assumption.

However, no straightforward pattern emerges connecting the phonological measures to literacy outcome at 8. The generalisation that can be made is that the SLI-PR group performs most poorly on the phonological tasks at preschool and school-age, in line with findings of other studies where dyslexia+SLI groups perform more poorly than poor readers or SLI-only groups (e.g. Bishop et al., 2009, Marshall & van der Lely, 2009). Furthermore, the pattern of performance was such that the FR good and poor readers obtain lowered scores than the TD group, and the SLI good and poor readers obtained even lower scores.

NWR at 8 years of age was also correlated with literacy outcomes for the TD and FR groups but not for the SLI group, though it should be borne in mind that there were missing data in this set. The findings of NWR at 8 years suggest a change in relationship to literacy compared to those found at 4;6 (de Bree et al., 2010). Specifically, at 4;6 the FR-PR, SLI-PR, and SLI-NR performed more poorly than the TD group. At 8 years however, the SLI-NR group performed like TD children on this task, suggesting evidence of 'catch up'. Arguably, these findings can also be accommodated within other lines of research that report an impact of literacy on NWR, such as studies with illiterate subjects (Castro-Caldas et al., 1998) and normally developing children (Nation et al., 2010). They can also be aligned with cognitive neuroscience models that assume a cascade of phonological processing difficulties to processing of visual words (e.g. McCandliss & Noble, 2003).

It could further be proposed that NWR in the children with SLI is determined by language abilities at preschool age (as proposed by Bishop et al., 2009 and Vandewalle et al., 2010, for example) and then shifts towards a reliance on literacy. At 8, the good and poorly reading SLI groups did not differ significantly from each other on NWR. The expectation is that the gap between the two groups will widen as the interaction between orthography and phonological processing becomes

more dominant in acquisition. A similar increase in difference on NWR performance between the good and poor FR readers was expected to occur. However, future analyses will have to assess whether the poorer NWR scores of the FR group at preschool age were also determined by language development or other skills.

Similar to other recent findings (Bishop et al., 2009), RSN seems to be a protective measure for literacy difficulties. Specifically, the FR-PR and SLI-PR groups showed poorer performance than the TD groups and the FR poor readers were outperformed by the good SLI group. These findings sit well with those of de Jong & van der Leij (2003), for example, who found that dyslexic children have poorer RSN than normal readers. The SLI-NR group showed the highest numerical scores on RSN, lending support to the idea that they can compensate for their language difficulties with RSN skills. Unfortunately, no RSN data are available when the children were younger, but the present findings are consistent with those of Vandewalle et al. (2010) that in Dutch, RSN is a protective factor for literacy. An alternative interpretation, however, also deserves further investigation: it could be argued that whereas poor NWR is characteristic of SLI, RSN is unrelated to this disorder. In fact, the SLI-RP did not show significantly lower RSN performance than the TD-NR or SLI-NR groups. This finding and interpretation is presented in the study by Bishop et al (2009) who also found that the SLI-NR did not differ from the TD-NR on RSN. To disentangle these two options, further longitudinal studies are required with larger sample sizes. Additionally, the mitigating influence of orthography, predicted by letter knowledge, deserves further attention (e.g. see Boets et al., 2010).

Another issue that needs to be addressed is the literacy score. The present study used a composite of reading fluency, timed word and non-word reading, as well as spelling dictation and spelling selection. However, it could be argued that the reading fluency and spelling need to be separated at this age (e.g. de Jong & van der Leij, 1999, Landerl & Wimmer, 2008). Analyses with a cut-off determined by the summed fluency tasks showed a similar pattern to that of the spelling and

fluency literacy score for the preschool data and literacy, as well as correlations between NWR, RSN, and literacy. Nevertheless, this is an issue that warrants further attention.

Taken together, these findings lend support to a number of interpretations on language and literacy. First, the results show that phonology is a complex and multidimensional construct. Different skills have a different impact on literacy (and language) and performance develops over time, as the NWR data show. A second issue is that poor phonology is not sufficient for rendering poor literacy outcome (e.g. Bishop et al., 2009, Snowling, 2008, Wolf & Bowers, 1999), as phonology was poorer in the FR and SLI groups but poor phonology did not always lead to poor literacy skills at 8 years in these groups. RSN, arguably a measure of phonological processing speed containing cross-modal matching of visual symbols and phonological codes (e.g. Vaessen, Gerretsen & Blomert, 2009) proved to be one important concomitant risk factor in this study. These results are in line with proposals of dyslexia as a multifactorial deficit (e.g. Bishop & Snowling, 2004; see Pernet, this volume). They stress the need for longitudinal studies into phonology and other linguistic skills and its underlying mechanisms (e.g. Flax et al., 2009; McArthur et al., 2009), as well as research into potential domain-general skills required for successful literacy (e.g. Flax et al., 2009, Maassen et al, this volume; Wijnen, 2006). These results also emphasize the need to look at individual differences and trajectories (see e.g. Leonard, this volume, Snowling et al., 2003) and connect the data with findings on neuroprognosis studies (Black & Hoeft, this volume, Gaab; this volume; Hoeft et al., 2007).

In sum, these findings show similarities and differences between children at familial risk of dyslexia and children with SLI on phonological measures and literacy outcome. These findings can be interpreted within multi-dimensional models of phonology, decoding literacy, and language that also take the role of development (age, orthographical skills) into account. They further indicate that longitudinal studies into phonology, oral language, decoding, and spelling, are badly needed to gain a

full picture of the potential developmental trajectories of preschool early linguistic markers and literacy.

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