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Preschool language profiles of children at family risk of dyslexia: continuities with specific language impairment

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Abstract

Background—Children at family risk of dyslexia have been reported to show phonological deficits as well as broader language delays in the preschool years.

Method—The preschool language skills of 112 children at family risk of dyslexia (FR) at ages 3½ and 4½ were compared with those of children with SLI and typically developing (TD) controls.

Results—Children at FR showed two different profiles: one third of the group resembled the children with SLI and scored poorly across multiple domains of language including phonology. As a group, the remaining children had difficulties on tasks tapping phonological skills at T1 and T2. At the individual level, we confirmed that some FR children had both phonological and broader oral language difficulties (compared with TD controls), some had only phonological difficulties and some appeared to be developing typically.

Conclusions—We have highlighted the early overlap between family risk of dyslexia and SLI. A family history of dyslexia carries an increased risk for SLI and the two disorders both show an increased incidence of phonological deficits which appear to a proximal risk factor for developing a reading impairment.

Keywords

Dyslexia; pre-school; SLI; language

Introduction

For many years, the dominant view of dyslexia has been that it is caused by phonological processing deficits that compromise the development of decoding skills, reading accuracy and fluency (Vellutino, Fletcher, Snowling, & Scanlon, 2004). However, there are similarities between dyslexia and other language disorders, in particular, specific language impairment (SLI; McArthur, Hogben, Edwards, Heath, & Mengler, 2000) and speech sound

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disorder (SSD; Peterson, Pennington, Shriberg, & Boada, 2009) and the relationships between these disorders are not well explained by the phonological deficit theory alone (Pennington & Bishop, 2009). Understanding these relationships has implications for our understanding of the risk factors that predispose children to dyslexia or language impairment and ultimately for the treatment of these disorders. In this paper, we compare the language profiles of preschool children at family risk of dyslexia to children with SLI to elucidate the nature of the comorbidity between these disorders.

In the middle school years, the overlap between SLI and dyslexia is approximately 50% (McArthur et al., 2000); however, evidence from prospective longitudinal studies of children at family risk (FR) of dyslexia suggests that shared risk factors are present much earlier in development. In a seminal study of English-speaking children, Scarborough (1990) found that children at family risk (i.e., from families with a history of dyslexia) who went on to become dyslexic demonstrated poorer articulation, shorter Mean Length of Utterance and poorer vocabulary knowledge than both at-risk children who did not go on to become dyslexic and typically developing children in the preschool years. This finding of early speech and language difficulties has since been replicated. Pennington and Lefly (2001) found that FR children who later became dyslexic had poor phonological awareness at age 5 and, using a broader battery of language measures, Snowling, Gallagher and Frith (2003) found that later affected FR children had poorer receptive vocabulary, expressive language and nonword repetition at age 3. A similar picture emerges in more transparent orthographies. In the Jyvaskyla project in Finland (Torppa, Lyytinen, Erskine, Eklund, & Lyytinen, 2010), vocabulary delays were evident among children at FR of dyslexia from 2 years, deficits in inflectional morphology, phonological sensitivity and letter naming at 3 years and poor comprehension of verbal instructions emerged by age 5. Early deficits in phonological processing have also been found in Dutch children at FR who went on to become dyslexic (e.g., Boets et al., 2011). Importantly, several FR studies (e.g., Pennington & Lefly, 2001; Snowling et al., 2003) have found that phonological difficulties are present in both affected and unaffected individuals from 'at-risk' families, suggesting that a phonological deficit is a putative 'endophenotype' of dyslexia (Skuse, 2001; Snowling, 2008).

In this paper, we focus on the language profiles of preschool children at FR of dyslexia comparing them with children who have SLI, to better understand the risk of reading disorders in children with language learning impairments. It is clear that FR children who go on to develop dyslexia experience language difficulties in the early years, but a key question is whether the nature and/or severity of their language difficulties is similar to those observed in children with a primary language impairment. Catts et al. (2005) proposed three models to explain the overlap between SLI and dyslexia in the school years. First, both SLI and dyslexia are caused by an underlying phonological deficit, the deficit being more severe in SLI (the severity hypothesis). Second, as proposed by Bishop and Snowling (2004), a phonological deficit is at the core of both SLI and dyslexia, but additional areas of language are affected in SLI. Third, the two disorders are caused by different deficits, a phonological deficit in dyslexia and a grammatical deficit in SLI, with overlap due to comorbidity. The present study uses these models as a framework for considering variations in language profile between children at FR of dyslexia and SLI in the preschool years.

According to Model 1, the more severe the phonological deficit, the more likely it will be associated with a broader oral language impairment in addition to a reading impairment. This model predicts that children with SLI and children with dyslexia will have problems on tasks involving phonological processing, but that the problems will be more severe in SLI. We can extend this prediction to children at FR of dyslexia. If the phonological deficit is an endophenotype for dyslexia, it will be present (perhaps varying in severity) in both affected and unaffected children and therefore we hypothesise that children at FR of dyslexia will have difficulty on tasks tapping phonological skills (Bearden & Freimer, 2006). A crucial assumption of this model is that all children with SLI have a phonological deficit. However, SLI is a heterogeneous disorder typically diagnosed based on a child's performance across tests assessing different components of language: phonology (e.g., nonword repetition), semantics (e.g., vocabulary) or grammar (e.g., sentence comprehension). To achieve a diagnosis, the child has to score below average on only a subset of tests. Therefore, some children with SLI may not have a phonological deficit.

Model 2 was initially proposed as a framework for conceptualising how different forms of literacy impairment relate to underlying language difficulties. According to Bishop and Snowling (2004), the model has two dimensions: 'phonological language skills', tapped by tasks such as nonword repetition, and 'nonphonological language skills' (semantics, syntax and discourse, also referred to as broader oral language), best measured by tests of vocabulary and grammatical sensitivity. In this two-dimensional (2D) model, children with dyslexia have poor phonological language skills but average nonphonological language skills, 'poor comprehenders' have average phonological but poor nonphonological language skills and children with SLI have a double deficit. Somewhat problematic for the model is that SLI is both related to the underlying dimensions of language and defined by them. If the quadrants are labelled in terms of literacy outcome then this quadrant might be more accurately labelled 'generally poor readers'. Catts et al. (2005) found that some children with SLI did not have poor phonological skills and consequently had normal decoding. However, they noted that these children are labelled as poor comprehenders in the 2D model, although they might be more accurately described as children with a history of SLI who have problems with reading comprehension in the school years. The predictions of the model with regard to the preschool profiles of the two disorders are that children at FR of dyslexia will have difficulty on phonological tasks while the children with SLI will have phonological and additional nonphonological language difficulties. It does not specify whether the severity of the phonological difficulties will differ between the two groups, as although the two dimensions in the model are continuous, it is assumed that the cut-off for a phonological impairment is the same in both disorders. A potential limitation of the 2D model is the independence of the two dimensions; it does not account for the impact of a phonological deficit on a child's ability to acquire vocabulary knowledge (e.g., Gathercole, 2006) or grammatical skills (e.g., Chiat, 2001; Joanisse & Seidenberg, 1998).

Model 3 argues that the two disorders are caused by different deficits, a phonological deficit in dyslexia and a grammatical deficit in SLI; with overlap due to comorbidity. The comorbidity between SLI and dyslexia exceeds that expected by chance and this could be because the two disorders involve shared risk factors, one disorder creates an increased risk for the other or the comorbid pattern represents a meaningful disorder in itself (Caron &

Rutter, 1991). In the preschool years, this model predicts phonological difficulties in children at FR, cases of children with SLI who have only grammatical difficulties (therefore not at risk of decoding difficulties) and children who have both (due to comorbidity). Catts et al. (2005) concluded that their data were most consistent with this view; in the school years children with just dyslexia, just SLI or both dyslexia and SLI were identified. Furthermore, children with dyslexia had underlying phonological difficulties, children with SLI had semantic and grammatical difficulties and children in the combined group had both.

Only one study has directly compared preschool children at FR of dyslexia with children who have SLI. The Utrecht study found that Dutch FR children fell in between the typically developing and the SLI groups on measures of articulation (Gerrits & de Bree, 2009), nonword repetition (de Bree, Rispens, & Gerrits, 2007), production of grammatical morphemes, mispronunciation detection and rhyme detection (van Alphen et al., 2004). However, there was a large degree of variability within the FR group, suggesting that some of these children may have had SLI.

Carroll and Myers (2010) reported measures from broader language tasks in a comparison of English FR children and children receiving speech and language therapy aged 4–6 years. Data were reported for four groups of children (FR-only, FR plus therapy, therapy-only and typically developing). The FR children who were also receiving therapy (40% of the FR sample), along with the therapy-only children, were impaired on phonological and broader language measures including sentence comprehension, expressive vocabulary and the use of morphological structures. The FR-only children were not impaired on either type of language measure, contrary to the hypothesis that a phonological deficit is an endophenotype for dyslexia.

In the present paper, we report data from the first two phases of a longitudinal study comparing English-speaking children at FR of dyslexia to children with preschool SLI. Our research questions were as follows. First, do some children at family risk of dyslexia meet diagnostic criteria for SLI (compatible with all three models)? Second, do children at family risk and children with SLI share a core phonological deficit (models 1 & 2) and is this deficit more severe in the children with SLI (model 1)? Finally, are broader oral language difficulties seen in those children with a more severe phonological deficit (model 1) or are broader difficulties a separate risk factor (models 2 and 3)?

Method

Design

Data are reported from children at two time-points before the beginning of formal schooling. The first phase (T1) was completed when the children were 3–4 years old and the second a year later at 4–5 years (T2). The time between the two testing occasions was 11 months on average. At T1, we applied diagnostic criteria for SLI to group the children on the basis of whether or not they reached a threshold for a broader oral language impairment. We then examined the performance of the groups concurrently and at T2 on additional measures of language processing drawing differentially on phonological, semantic and grammatical skills.

Participants

Families were recruited to the study via advertisements placed in local newspapers, nurseries and the webpages of support agencies for children with reading and language difficulties and via speech and language therapy services. Of the 242 children recruited, none met our exclusionary criteria (MZ twinning, chronic illness, deafness, English as a 2nd language, care provision by local authority and known neurological disorder such as cerebral palsy, epilepsy, ASD). Ethical clearance for the study was provided by the University of York, Department of Psychology's Ethics Committee and the NHS Research Ethics Committee. Parents provided informed consent for their child to be involved.

Following recruitment, children were classified using a two-stage process: first, we determined whether they were at FR of dyslexia and then diagnostic criteria were used to ascertain whether they had a language impairment (SLI). This led to the classification of children into four groups: FR-only; FR-SLI; SLI-only, Typically Developing (TD) (see Appendix 1 for full details).

Family risk.—Previous FR studies have either used self-report or objective measures to determine risk status. In the current study, we obtained self-report using the Adult Reading Questionnaire (ARQ; Snowling, Dawes, Nash, & Hulme, 2012) and conducted objective testing of parents who consented. To avoid missing children at FR of dyslexia, we employed the following criteria: A child was included in the FR group if (a) a parent self-reported as dyslexic, (b) a parent scored below 90 on a literacy composite of nonword reading and spelling, (c) a parent had a discrepancy between nonverbal ability and the literacy composite of 1.5 standard deviations, with a literacy composite standard score of 96 or below, or (d) a sibling had a diagnosis of dyslexia from an educational psychologist or a specialist teacher. According to these criteria, 120 of the 242 children were considered at FR of dyslexia.

Language impairment.—LI status was determined using three subtests of the Clinical Evaluation of Language Fundamentals—Preschool 2 UK (CELF-Preschool 2 UK, Semel, Wiig, & Secord, 2006); Basic Concepts, Expressive Vocabulary and Sentence Structure and the screener from the Test of Early Grammatical Impairment (TEGI, Rice & Wexler, 2001). These tests assess receptive and expressive language across multiple domains of language (Tomblin et al., 1997). Children were deemed language impaired if they 'failed' 2/4 of these language tests (a fail being a scaled score of 7 or below on the CELF subtests and failure of the TEGI screener). Given the age and low ability of some of the children, there were insufficient data from the diagnostic tests to determine LI status for 22 cases. In these cases, information was used from the separate TEGI screener subtests and the Preschool Repetition test (Seeff-Gabriel, Chiat, & Roy, 2008) at T1 and CELF sentence structure test at T2 to come to a clinical judgement about group membership. Seventeen of the 22 cases were considered language impaired.

Based on our diagnostic criteria, 35/120 children at FR and 31/46 children referred for speech and language difficulties were language impaired. The remaining 15 children whose parents had reported concerns but who did meet our diagnostic criteria were excluded from further analyses. In addition, of the 76 children initially classified as typically developing, 5

met criteria for language impairment. These children were considered to be language impaired for the purposes of this study. Among 1 LI children, 6 had nonverbal IQ (NVIQ) scores below 80 (WPPSI-III; Wechsler, 2003) and a further 3 were not able to complete the tests. Therefore, the majority met criteria for SLI. All children who met LI criteria, regardless of NVIQ were included because research suggests that children in the lower range of NVIQ show the same overall profile of Language Impairment as children in the range of 85 and above (Tomblin & Zhang, 1999). Given that the majority of LI children had NVIQ scores above 80, we will refer to them as SLI. Between T1 and T2, there was a small amount of attrition, 2 TD, 8 FR and 4 SLI children withdrew from the study. Their data are not included here. The following analyses are based on children in four groups, TD (N = 69), FR-only (N = 83), SLI (N = 32) and FR-SLI (N = 29). Of the 61 children (in total) diagnosed with SLI, 74% were in contact with speech and language therapy services at T1. See Table 1 for information about the age, gender, SES status and nonverbal ability of the participants.

The four groups did not differ in age at T1 with averages ranging from 3 years 7 months to 3 years 9 months. There was a nonsignificant trend for more boys than girls to reach criteria for language impairment. The TD group were higher in SES than the two language-impaired groups and the TD group performed significantly better on the NVIQ tasks than the FR group, who in turn performed better than the two language-impaired groups.

Tests and procedures

Each child was administered tests of NVIQ, speech and language. At T1, these were administered in a single one and a half hour session and at T2, across two 1-hour sessions, which were conducted in the child's home with breaks. The tasks were usually administered in a fixed order.

NVIQ (T1).—Each child completed Block Design and object Assembly subtests from the Wechsler Preschool Primary Scale of Intelligence (WPPSI-III; Wechsler, 2003) according to the test manual.

Articulation (T1).—The articulation subtest of the Diagnostic Evaluation of Articulation and Phonology (DEAP, Dodd, Hua, Crosbie, Holm, & Ozanne, 2002) provided a measure of percentage of consonants correctly produced (PCC). The child named 30 pictures (e.g. pig, moon, sheep, five, television) or imitated the name if they could not produce it spontaneously. Arguably, the ability to produce consonants correctly taps underlying phonemic representations, and hence we interpreted this as a phonological language measure.

Word and Nonword repetition (Pre-School Repetition subtest from the Early Repetition Battery, Seeff-Gabriel et al., 2008) (T1, T2).—The child was asked to repeat 18 words (6 one-syllable, 6 two-syllable and 6 three-syllable) and 18 nonwords (6 of each length). The nonwords were created from the words, by altering the vowel in the one-syllable items and by swapping two consonants in the multi-syllabic items (e.g., 'lamb' ->/ lom/, 'machine' ->/shameen/, 'dinosaur' ->/sinodaur/). At T2, the repetition tests were extended to include 6 additional four-syllable words/nonwords to avoid ceiling effects.

Consistent articulation errors were taken into account when scoring the children's responses. Nonword repetition is a measure of phonological language skill.

Expressive vocabulary (CELF-Preschool 2 UK; Wiig, Secord, & Semel, 2006) (T1).—The child was asked to name objects (e.g. carrot, telescope) or to describe what a person is doing (e.g. riding a bike). This test places demands upon both phonological and broader language skills.

Sentence imitation test (SIT-16; Seeff-Gabriel et al., 2008) (T1, T2).—The child repeated 16 sentences increasing in length and complexity (e.g. the cat ate a big mouse). The total number of sentences, content words, function words and grammatical inflections repeated correctly was recorded. Sentence repetition involves semantic and syntactic processing, but also taps phonological memory. In addition, function words and grammatical inflections are less phonologically salient than content words (Chiat, 2001) implying that repetition of these word types may be more heavily dependent on phonological language skills.

Grammatical inflections (TEGI; Rice & Wexler, 2001) (T1, T2).—Two subtests were administered, the third person and past tense probes, which together provide a screening test score that reflects the child's ability to produce grammatical inflections. In the third person singular/s/probe (10 items), the child was introduced to characters and asked to describe what each does. For example, the child was shown a picture of a dentist and asked to say what a dentist does. A response containing the third person singular/s/inflection was scored as correct (e.g., a dentist checks/cleans your teeth). In the past tense probe, the child was shown two pictures, the first picture described by the examiner (e.g., here the boy is brushing his hair). The examiner then pointed to the second picture and said 'now he is done, tell me what he did'. Correctly inflected verbs (e.g., brushed/combed) and overregularisations of irregular verbs were scored as correct. In both subtests, the score was the percentage of items correct (nonresponses and other verb tenses were excluded and only bare stems were scored as incorrect). To correctly inflect the nouns and verbs in these subtests, the child needed to be able to produce/s/,/z/,/t/and/d/. Given the number of children with articulation difficulties at T1, we used data from the articulation test to determine whether each child was able to produce the phonemes required for each grammatical subtest. We present the T1 data for the whole sample and then for those who could consistently produce these phonemes; to determine whether poor performance reflects underlying grammatical difficulties or peripheral speech difficulties. T2 data are presented for the whole sample only. The production of grammatical inflections is a task that involves both phonological and broader language skills. Phonology provides a cue to grammatical morphemes, for example the past tense is marked by phonological variation in form meaning pairs (e.g., walk – walked) (Chiat, 2001) and many morphological rules have a phonological component (e.g., the phonological realisation of the past tense depends on the final phoneme in the verb root) (Joanisse & Seidenberg, 1998).

Sentence structure (CELF-Preschool 2 UK; Wiig et al., 2006) (T1, T2).—The child heard a sentence (e.g., the bear is in the wagon) and had to select from a choice of four, the picture that conveyed its meaning. The sentences included a range of different syntactic structures.

The Sentence Structure and Basic Concepts subtests of the CELF Preschool 2 draw mainly upon broader oral language skills.

Receptive vocabulary (Receptive one word picture vocabulary test (ROWPVT) Brownell, 2000) (T2).—The child heard a word and had to select the corresponding picture, from a choice of four. This test taps semantic memory and draws less on phonological skills than measures of expressive vocabulary.

Results

Applying research criteria for LI at T1 revealed that 35/120 children at FR of dyslexia (29%) had language difficulties severe and pervasive enough to be classified as SLI. We proceeded to investigate how these children compared with those children with SLI who were not at FR (see Appendix 2). The means suggest that differences between groups were small and the absence of group differences was confirmed by t-tests. We therefore pooled data from these two groups to form a single SLI group.

Time 1 (T1; 3 ½ years)

Data from the FR, TD and the pooled SLI group across the language tasks are shown in Table 2 together with the results of between-subjects ANO-VAs and comparisons between groups using the Games Howell post hoc test. The main effect of group was significant for all measures and two different patterns of group differences emerged.

On the tasks considered to have the highest phonological loading (articulation, word and nonword repetition) the SLI group was impaired relative to the controls and so was the FR group, but to a lesser degree (TD>FR>SLI). A similar pattern was observed for tasks that could be considered to draw upon both phonological and nonphonological language skills (the production of grammatical inflections and total sentence and function word repetition). This pattern was not seen for the repetition of content words or inflections. In contrast, on the tests of Sentence Structure and Expressive Vocabulary, the FR group performed like TD controls and only the SLI group were impaired [(TD=FR)>SLI]. Finally, although we had expected to find the latter pattern for Basic Concepts, the data indicated that the FR group was impaired relative to controls, but less severely than the SLI group.

In summary, consistent with the way the groups were classified, the SLI group had impairments in both phonological and nonphonological language skills at T1. However, the FR group (without SLI) was not free of language difficulty and scored below TD controls on

tasks tapping expressive phonology, namely articulation, word and nonword repetition, sentence repetition (particularly function words) and the production of grammatical inflections.

Time 2 (T2; 4 ½ years)

The scores of the three groups on the language measures administered approximately 1 year later are presented in Table 3. For word, nonword and sentence repetition, the pattern was the same as at T1 (TD<FR<SLI). On the tests of Sentence Structure and Receptive Vocabulary, only the SLI group was impaired and in contrast to T1, this was now also the case for the production of grammatical morphemes [(TD=FR)<SLI].

In summary, the pattern of performance at T2 replicated that at T1 with one exception: the FR group's difficulty in producing grammatical inflections appeared to have resolved and performance was now in line with that of TD controls.

Individual differences in phonological and nonphonological language skills

The data presented above suggest that 29% of children at FR of dyslexia had language difficulties similar in nature and severity to those of the children with SLI. In contrast, the difficulties of children in the FR-only group were mainly confined to tasks tapping phonological skills directly (articulation, word and nonword repetition) or indirectly (repeating sentences and producing grammatical inflections).

To explore variations in the relationship between phonological and nonphonological language skills in the sample, we used the framework of the 2D model (Bishop & Snowling, 2004) with nonword repetition as a marker of an individual's position on the phonological dimension and sentence structure (a measure tapping semantic and syntactic skills) to represent the broader language dimension. We created z scores using the mean and SD of the TD group for performance on each dimension and plotted these, adding reference lines at -1 standard deviations on both axes (see Figures 1, 2). Plotting the data in this way highlights the moderate relationship between the two dimensions (T1 r = .37, p < .05; T2 r= .51, p < .05). Given the classification criteria used, we expected most of the SLI group to score more than 1 SD below the TD group mean on both dimensions. Furthermore, we expected around 50% of children at FR of dyslexia to score below the -1 SD cut-off for nonword repetition and fall into the quadrant placing them at risk for dyslexia (based on the estimated risk across FR studies). Conversely, we expected some children in the SLI group to score more than -1 SDs below the TD group on the sentence structure test but higher than -1 SDs on the nonword repetition test, placing them at risk of problems of reading comprehension rather than reading accuracy (the 'poor comprehender' profile).

As expected, at T1 the majority of the cases in the TD group fell in the typically developing quadrant (67%) and the largest percentage of SLI cases were in the SLI quadrant (49%). However, some of the TD group appeared at risk for a specific disorder (13% dyslexia, 14% PC; data were missing from the remaining 6% of TD children). The remaining SLI children were split between being at risk of developing dyslexia (13%) or reading comprehension problems (PC; 16%) although it should be noted that there were 18% missing data for this

group. The FR-only cases were largely split between the TD quadrant (46%) and the risk for dyslexia quadrant (31%), with the remaining 14% in the PC quadrant, 5% in the SLI quadrant and 4% missing data.

At T2 the percentage of TD and FR-only cases in the different quadrants remained similar to T1. The percentage of SLI cases in the SLI quadrant increased from 49% to 66% because of cases moving from the poor comprehender quadrant and also fewer data were missing. The increase in the percentage of SLI children falling in the TD quadrant (3%–10%) suggests some children in this group had resolved their language difficulties.

Discussion

Understanding the overlap between dyslexia and SLI, and how this changes over time, is important for theory and practice. Here, in the first study to separate these groups, we report preschool language profiles of children at family risk (FR) of dyslexia before the onset of formal schooling, comparing them to those of children with SLI and typically developing children.

Perhaps the most important finding of this study is that almost one third of the children at FR for dyslexia met diagnostic criteria for SLI when they were 3 ½ years. As a group, these children (FR-SLI) resembled the children with SLI, who did not have a known family history of dyslexia. The majority of the children fulfilling our diagnostic criteria did indeed fall in the 'SLI quadrant' of the Bishop and Snowling (2004) 2D model, with a double deficit affecting phonological and nonphonological language skills. Furthermore, a moderate correlation between the two dimensions provides some support for the severity hypothesis; that children with the poorest phonology have the broadest language impairments. However, there were children in the sample with a single deficit, placing them either at risk of dyslexia or of a specific reading comprehension impairment, suggesting that these risk factors may be separable as proposed by the 2D model or a separate deficits view. At 4 ½ years, the profile of pervasive language impairment remained and more of the SLI cases had a double deficit (and commensurately fewer were at risk for specific reading comprehension impairment).

The second important finding of the study concerns the language profile of the FR group who did not fulfil criteria for SLI. As a group, these children were impaired relative to the TD group in word, nonword and sentence repetition, particularly of function words. These findings show that this group had relatively circumscribed phonological difficulties, consistent with a core deficit in phonology being shared between SLI and dyslexia. However, there was evidence of 'downstream effects' on aspects of grammar that depend on phonology, namely grammatical morphology and processing of syntactic constructions marked by less salient forms (e.g., function words in sentence repetition) (Chiat, 2001; Joanisse & Seidenberg, 1998). Interestingly, by T2, the problems producing inflections had resolved, suggestive of a developmentally limited impairment in the FR group but one which persists in the groups with broader language deficits (Rice, 2000). We speculate that a difficulty producing grammatical inflections in some children may resolve once the child is able to use phonological cues to morphology. Other children may need more exposure and

time to be able to utilise these cues and/or have additional difficulties that affect their ability to acquire or use grammatical inflections.

Our finding that the FR-only group had significantly poorer phonological skills than controls is in support of an inherited phonological risk that is continuous and therefore that a phonological deficit is an endophenotype for dyslexia. Evidence of milder phonological difficulties in unaffected individuals from at-risk families has been found in previous studies (e.g., Pennington & Lefly, 2001; Snowling et al., 2003) but was not found by Carroll and Myers in their group of FR children who were not also receiving speech and language therapy. A possible explanation of the Carroll and Myers finding concerns the large percentage of FR children who were receiving speech and language therapy (40% compared to 26% diagnosed with SLI in our sample); we would expect the majority of FR children with phonological difficulties to be in the FR plus therapy group and not in the FR-only group, hence the FR-only group appeared to be unimpaired.

Turning to individual differences within the groups, if a phonological deficit is an endophenotype of dyslexia, it should be present to varying degrees in the majority of cases in the FR group (since both affected and unaffected relatives are expected to carry this risk factor). However, within the 2D framework of Bishop and Snowling (2004), at T1 only 44% of the family risk sample (FR and FR-SLI) had a nonword repetition deficit (in absolute terms) and 56% at T2. If these children do go on to develop dyslexia, then this percentage is in line with the estimated risk found across FR studies.

The findings of this study extend those of previous FR studies in the preschool years. When we consider the whole FR sample, the children showed a broad range of language difficulties. However, one third of the FR group fulfilled criteria for SLI and when these were removed from the sample, the FR-only group showed a much more circumscribed pattern of impairment on tasks tapping output phonology. Thus, dyslexia and SLI appear to carry a shared risk of reading difficulties associated with phonological deficits. This finding is in line with all three models of the overlap, as even the third model allows for comorbidity. However, the severity hypothesis is the only model that proposes a relationship between the two domains of language. Given that the phonological deficit was more severe in the SLI children, our data provide some support for this hypothesis. However, only the 2D and separate deficits models allow for a nonphonological language deficit in the absence of a phonological deficit, and we found children with this profile in our sample. Therefore, our data would be most compatible with a version of the 2D model in which the dimensions were not independent.

Given the findings of the present study at T2, which is close to the point of reading instruction, we can make some bold predictions regarding early literacy outcomes guided by the 2D framework. First, the majority of the SLI children (including the FR-SLI group) appear to be at risk for both reading accuracy (decoding) and comprehension difficulties (66%), while the remainder appear at risk only for decoding difficulties (20%) and a small proportion for specific reading comprehension difficulties (3%). To the extent that nonword repetition is a marker of the phonological skills required for decoding, 35% of the FR-only group are at risk for accuracy difficulties. A further 12% are at risk for difficulties with both

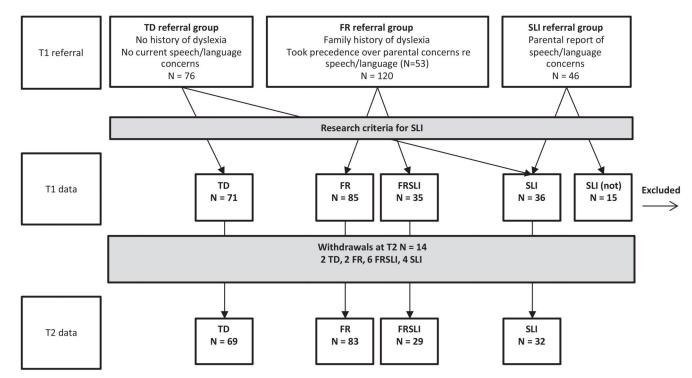
reading accuracy and comprehension and some 6% for comprehension difficulties only. The remaining FR-only children (46%) do not appear to be at risk for developing literacy difficulties of either form. This project is longitudinal and in due course, we will be in a position to test these predictions.

An important clinical implication of this work is that the early speech and language development of children at family risk should be monitored closely, as these children are at increased risk of impairment. In addition, screening on a measure of phonological language (such as nonword repetition) and a measure of broader oral language skills (such as sentence comprehension) would enable practitioners to identify children at risk for different literacy difficulties and put in place suitable support at an early age.

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Appendix 1



Appendix 2

Means (SDs) for the SLI and FR-SLI groups on the language tasks at T1 and T2

					T1							-	T2			
	01	$\mathbf{SLI} \ (N = 32)$	32)	FR	FR-SLI $(N = 29)$: 29)			S	SLI $(N = 32)$	2)	FR	FR-SLI $(N = 29)$	= 29)		
	N	Mean	SD	N	Mean	SD	t	d	N	Mean	SD	N	Mean	SD	t	d
CELF Basic Concepts (/18)	29	10.45	2.86	29	10.79	3.57	41	69.	N/A	N/A	N/A	N/A	N/A	N/A	N/A	N/A
CELF Sentence Structure (/22)	30	7.23	3.28	27	8.15	3.90	96	.34	32	13.19	3.95	29	14.10	3.43	96	.34
CELF Expressive Vocabulary (/40)	28	10.14	6.48	28	9.89	5.67	.15	.88	N/A	N/A	N/A	N/A	N/A	N/A	N/A	N/A
TEGI 3rd person % correct (whole sample)	22	25.43	26.10	25	16.77	28.73	1.08	.29	30	38.02	30.92	28	55.53	39.17	-1.90	.06
TEGI 3rd person % correct (phono probe)	12	30.79	30.45	16	25.50	32.98	.43	.67	N/A	N/A	N/A	N/A	N/A	N/A	N/A	N/A
TEGI past tense % correct (whole sample)	18	39.54	24.28	21	40.46	30.40	10	.92	30	65.28	26.86	26	68.61	26.78	46	.65
TEGI past tense % correct (phono probe)	14	44.27	21.99	15	36.35	30.93	.54	.59	N/A	N/A	N/A	N/A	N/A	N/A	N/A	N/A
Articulation (PCC)	31	62.42	24.24	28	68.92	21.02	-1.10	.28	N/A	N/A	N/A	N/A	N/A	N/A	N/A	N/A
PSrep word repetition (/18 at T1,/24 at T2)	25	10.68	4.01	27	12.07	3.92	-1 27	.21	32	17.59	5.53	29	18.76	4.51	90	.37
PSrep nonword repetition (/18 at T1,/24 at T2)	25	8.36	4.00	27	9.41	3.93	95	.35	31	10.26	3.31	29	10.45	3.57	96	.34
SIT-16 total (/16)	19	0.74	1.28	23	1.87	2.96	-1.55	.13	24	5.96	4.12	25	6.16	4.54	16	.87
SIT-16 content words (/58)	19	36.74	9.52	23	30.30	12.21	1.87	.07	24	47.75	7.18	25	49.36	7.01	79	.43
SIT-16 function words (/58)	19	21.26	11.12	23	20.17	15.18	.26	.80	24	40.29	10.99	25	41.36	13.23	31	.76
SIT-16 inflections (/13)	19	4.37	3.65	23	4.13	2.99	.23	.82	24	9.08	2.64	25	10.24	8.11	67	.51
ROWPVT receptive vocabulary	N/A	N/A	N/A	N/A	N/A	N/A	N/A	N/A	32	49.84	8.94	29	50.62	7.98	-0.36	.72
Only data from children who completed both time-points are included here.	e-points	are inclu	ded here.			-										

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Key Points

- Children at family risk (FR) of dyslexia who go on to develop literacy difficulties experience early language difficulties.
- We investigated the overlap in the early language profiles of children at FR of dyslexia and children with SLI.
- We found an increased prevalence of SLI in children at FR, with almost one third of our sample fulfilling criteria. The remainder of the FR group had milder phonological difficulties; supporting the endophenotype view.
- Approximately 50% of the FR children had a phonological deficit, placing them at risk for decoding difficulties. The majority of the children with SLI had both a phonological and broader language deficit, placing them at risk for pervasive literacy difficulties.
- The early speech and language development FR children should be monitored closely.
- Screening on a phonological measure (e.g., nonword repetition) and a broader language measure (e.g., sentence comprehension) would enable practitioners to identify children at risk for different forms of literacy impairment.

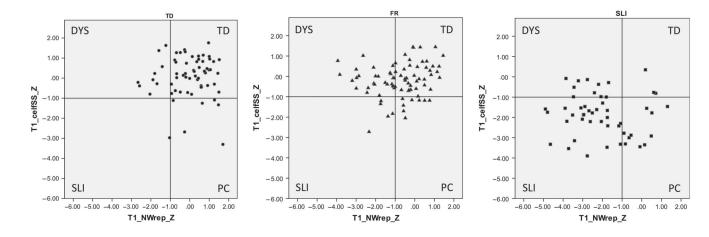


Figure 1.

The distribution of nonword repetition and sentence structure scores in the three groups of children (TD, FR and SLI) at T1. Note: data were missing for 4 TD children, 3 FR children and 11 SLI children

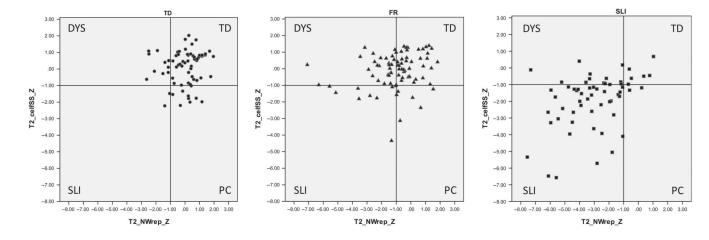


Figure 2.

The distribution of nonword repetition and sentence structure scores in the three groups of children (TD, FR and SLI) at T2. Note: data were missing for 2 children (1 FR child and 1 SLI child)

Table 1

Mean (*SD*) age, SES, nonverbal IQ (NVIQ) and gender for the TD, FR, SLI and FR-SLI groups (including results of between groups analyses)

	TD	FR	SLI	FR-SLI	Significant differences
Ν	69	83	32	29	
T1 Age (months)	44.59 (3.12)	45.17 (3.47)	43.56 (2.09)	45.07 (3.24)	F 2.15, NS
T2 Age (months)	55.46 (3.21)	56.64 (3.85)	55.19 (2.58)	56.76 (3.52)	F 2.56, NS
Gender (% males)	49	58	69	72	χ^2 6.74, NS
SES (postcode% rank 1)	71.97 (27.46)	62.19 (26.67)	52.62 (30.18)	50.84 (30.50)	<i>F</i> 5.57, <i>p</i> < .05 TD > LI & FRLI
NVIQ ²	115.61 (13.93)	107.90 (14.84)	96.07 (13.10)	101.03 (10.15)	<i>F</i> 16.82, <i>p</i> < .05 TD > FR > (LI = FRLI)

¹SES based on postcode in UK, relative rank according to deprivation value; Lower = more deprived (Department of Communities and Local Government, Indices of Multiple Deprivation 2007).

 $^2\mathrm{NVIQ}$ is mean standard score, two WPPSI-III Performance IQ subtests.

Table 2

Means (SDs) for the language tasks at T1 $\,$

		TD (N = 69)	(6)		FR $(N = 83)$	83)		$\mathbf{SLI}\ (N=61)$	(1)				
	N	Mean	SD	N	Mean	SD	N	Mean	SD	F	d	$\eta_{p}{}^2$	Group differences
Articulation (PCC)	69	89.39	7.66	83	81.99	14.82	59	65.50	22.81	37.47	00.	.27	TD > FR > LI
PSrep word repetition (/18)	99	16.70	1.95	81	15.42	2.85	52	11.40	3.99	49.82	00.	.34	TD > FR > LI
PSrep nonword repetition (/18)	65	14.12	2.55	80	12.23	3.21	52	8.90	3.96	37.98	00.	.28	TD > FR > LI
CELF Expressive Vocabulary (/40)	69	20.54	5.22	83	18.58	5.33	56	10.02	6.03	62.91	00.	.38	(TD = FR) > LI
SIT-16 total (/16)	65	9.28	4.16	LL	6.05	4.42	42	1.36	2.40	51.19	00.	.36	TD > FR > LI
SIT-16 content words (/58)	65	51.78	8.32	LL	48.13	10.50	42	33.21	11.41	46.82	00.	.34	(TD = FR) > LI
SIT-16 function words (/58)	65	47.77	9.85	LL	40.95	13.34	42	20.67	13.35	64.93	00.	.42	TD > FR > LI
SIT-16 inflections (/13)	65	10.29	2.59	LL	9.10	3.67	42	4.24	3.27	47.80	00.	.35	(TD = FR) > LI
TEGI 3rd person % correct (whole sample)	65	79.34	24.94	82	62.56	32.61	47	20.82	27.58	57.12	00.	.37	TD > FR > LI
TEGI 3rd person % correct (phono probe)	62	78.82	25.36	71	66.43	29.74	28	27.77	31.45	31.37	00.	.28	TD > FR > LI
TEGI past tense % correct (whole sample)	65	85.16	16.26	82	69.05	30.17	39	40.03	27.39	38.15	00.	.29	TD > FR > LI
TEGI past tense % correct (phono probe)	56	84.72	17.09	73	72.12	27.10	29	40.17	26.82	33.22	00.	.30	TD > FR > LI
CELF Basic Concepts (/18)	69	16.35	1.66	83	15.45	1.91	58	10.62	3.21	113.83	00.	.52	TD > FR > LI
CELF Sentence Structure (/22)	69	14.42	3.31	83	13.63	2.74	57	7.67	3.58	83.22	00.	.45	(TD = FR) > LI

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Table 3

Means (SDs) for language tasks at T2

		TD (N = 69)	(6)		FR $(N = 83)$	83)		SLI $(N = 61)$	61)				
	Ν	Mean	SD	N	Mean	SD	N	Mean	SD	F	d	$\eta_{p}{}^2$	p 11 p ² Group differences
PSrep word repetition (/24)	69	23.51	.85	83	21.87	2.78	61	18.15	5.07	45.73	00.	.30	TD > FR > LI
PSrep nonword repetition (/24)	69	15.17	1.65	83	13.63	3.00	09	12.58	4.59	58.69	00.	.36	TD > FR > LI
SIT-16 total (/16)	68	12.56	2.12	79	11.16	2.99	49	6.06	4.30	65.75	00.	.41	TD > FR > LI
SIT-16 content words (/58)	68	56.65	1.89	79	56.13	3.01	49	48.57	7.07	64.81	00.	.40	(TD = FR) > LI
SIT-16 function words (/58)	68	54.71	2.69	79	52.92	4.67	49	40.84	12.07	65.63	00.	.41	TD > FR > LI
SIT-16 inflections (/13)	68	12.19	1.12	79	11.80	1.49	49	9.67	6.05	9.65	00.	60'	(TD = FR) > LI
TEGI 3rd person % correct	69	91.54	15.33	83	84.38	23.82	58	46.47	35.93	55.55	00.	.35	(TD = FR) > LI
TEGI past tense % correct	69	93.74	11.46	83	89.28	15.90	56	66.83	26.63	37.73	00.	.27	(TD = FR) > LI
CELF Sentence Structure (/22)	69	18.07	2.29	83	17.55	2.43	61	13.62	3.71	48.28	00.	.32	(TD = FR) > LI
ROWPVT receptive vocabulary 68	68	64.60	7.17	83	63.48	63.48 10.16 61	61	50.21	8.44	53.54	00.	.34	(TD = FR) > LI