

# Annual Research Review: Reading disorders revisited – the critical importance of oral language

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This paper discusses research on reading disorders during the period since their classification within the overarching category of neurodevelopmental disorders (*Journal of Child Psychology and Psychiatry*, 53, 2012, 593). Following a review of the predictors of learning to read across languages, and the role of language skills as critical foundations for literacy, profiles of reading disorders are discussed and putative causal risk factors at the cognitive, biological, and environmental levels of explanation considered. Reading disorders are highly heritable and highly comorbid with disorders of language, attention, and other learning disorders, notably mathematics disorders. The home literacy environment, reflecting gene-environment correlation, is one of several factors that promote reading development and highlight an important target for intervention. The multiple deficit view of dyslexia (*Cognition*, 101, 2006, 385) suggests that risks accumulate to a diagnostic threshold although categorical diagnoses tend to be unstable. Implications for assessment and intervention are discussed. **Keywords:** Dyslexia; reading disorders; reading comprehension; comorbidity; multiple risks.

## Introduction

Learning to read for meaning entails mapping a written language (orthography) on to spoken language. Children with good oral language skills learn to read better than children with oral language difficulties. Snowling and Hulme (2012) situated reading disorders in the context of language difficulties and proposed that reading disorders are the outcome of multiple risk factors that can be construed as liabilities for poor decoding (dyslexia) or poor reading comprehension. In the intervening years, longitudinal studies tracing reading development from its foundations in spoken language through the early years of instruction have burgeoned. We begin by reviewing the findings from these studies as a prelude to discussing the variety of causal risk factors associated with reading problems. We consider the comorbidities between reading and other disorders within the multiple deficit framework (McGrath, Peterson, & Pennington, 2020) and discuss implications for assessment. Finally, we briefly consider interventions and what they can tell us about causal mechanisms.

## Learning to read

In discussing how children learn to read, it is essential to make a clear distinction between the ability to translate printed words into speech (referred to as decoding) and the ability to read for meaning (the ability to understand what has been decoded). These two skills are related; if a child cannot decode a text,

they cannot comprehend it. On the other hand, some children may be able to decode a text but have limited comprehension of it because of language comprehension problems. These ideas are encapsulated in the Simple View of Reading (Gough & Tunmer, 1986), which proposed that reading is the product of decoding and linguistic comprehension. This dimensional view of reading development can be related to categorical descriptions of reading disorders. Thus, children with deficits in decoding but intact language comprehension skills are classified as having reading disorder (RD or dyslexia). Conversely, children with adequate decoding skills but poor language comprehension are classified as having reading comprehension impairment (poor comprehenders). A third group of children, including many with developmental language disorders, have problems with both decoding and language comprehension (e.g., Bishop & Snowling, 2004).

The simple view of reading is an important framework; it emphasizes the need to assess both decoding and comprehension skills, and also implies that literacy instruction should be balanced, and include both the teaching of 'phonics' and reading texts for meaning. However, it raises a separation between the phonological (sound-based) aspects of language critical for decoding and broader semantic aspects (meaning and grammar) needed for comprehension. However, current evidence suggests that in the early school years oral language is a unitary construct (Bornstein, Hahn, Putnick, & Suwalsky, 2014; Tomblin & Zhang, 2006) and provides an essential foundation for literacy development (Lervag, Hulme & Melby-Lervag, 2018).

Hjetland, Lervåg, Lyster, Hagtvet, Hulme, and Melby-Lervåg (2018) studied the predictors of the

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growth of reading comprehension in a 6-year longitudinal study of Norwegian children from age 4 years, well before reading instruction began. This study provided evidence for a unitary language factor influencing reading comprehension, which was highly stable, and a less stable pathway in decoding-related skills [reflecting variations in letter knowledge, the ability to manipulate speech sounds (phoneme awareness), and the ability to name common symbols (such as letters or digits) at speed – referred to as rapid automatized naming (RAN)]. In this study, as in a similar study with a sample of English children at risk of reading difficulties (Hulme, Nash, Gooch, Lervåg, & Snowling, 2015), preschool language skills were strong predictors of both the foundations of decoding skills and later reading comprehension.

Nevertheless, there are inconsistencies in this area, particularly concerning the relative strength of predictive relationships shown in different studies. Although there is broad agreement that language can be considered a unitary factor in preschool (Hjetland, Brinchman, Scheerr, Hulme, and Melby-Lervag, (2020) for a review), some studies suggest it may become multidimensional later in development (Foorman et al., 2015; Tomblin & Zhang, 2006). Still other studies suggest that morphological awareness – the ability to analyze the meaning-based components of word – is an additional predictor of word reading as well as reading comprehension beyond oral language (Kirby et al., 2012). Together, these findings remind us that learning to read depends on a broad range of cognitive skills, and that there are likely to be reciprocal relationships between language skills and the development of reading skills.

### *Becoming a skilled reader*

The desired end point of learning to read is to become a skilled reader. A skilled reader can comprehend the meanings of printed words rapidly and automatically without laborious decoding. According to the Lexical Quality Hypothesis of Perfetti and colleagues (Perfetti, 2010; Perfetti & Hart, 2002), skilled reading depends upon having well-specified orthographic (spelling) representations of words that are closely connected with phonological (sound-based) and semantic/syntactic (meaning-based) representations (see Seidenberg & McClelland, 1989 for a similar conceptualization). Within this view, it is clear that language skills provide the critical foundation for the development of the orthographic representations that serve *both* decoding fluency and reading comprehension, and allow reading to become an automatic process. According to the theory, decoding and reading comprehension skills, in turn, have reciprocal effects on the development of vocabulary and related oral language and metalinguistic abilities. This principle of reciprocity may provide an explanation for multifactorial structure of

language, which has been observed in studies of older children and adults.

### *Reading development in different languages*

A key question for reading research is whether the skills required for developing reading fluency are universal across languages. As noted above, reading involves learning to transcode a written representation of language into speech- and meaning-based linguistic representations. However, languages differ in the types of writing systems used (their orthographies) – and the nature of the mappings between symbols, sounds, and meaning. In alphabetic orthographies, a small number of symbols (letters) represent all the sounds (phonemes) in a language and the mappings are at a fine-grain size. Other languages use intermediate sized units, such as syllables to represent speech, for example, the Japanese Kana, or the akshara of some Indian languages (e.g., Kannada), whereas Chinese (Mandarin and Cantonese) uses a morphosyllabic orthography.

Differences between languages in their orthographies mean that the language skills that are most critical for learning to read differ between languages (Verhoeven & Perfetti, 2017). In alphabetic orthographies (especially English), phonological skills play a dominant role in learning to decode. Conversely, learning to read in Chinese appears to place heavy demands on semantic, morphological, and visual skills in addition to phonological skills (e.g., Hulme, Zhou, Tong, Lervåg, & Burgoyne, 2019; Li, Shu, McBride-Chang, Liu, & Peng, 2012). More generally, the size of the symbol set and the visual complexity of the symbols are determinants of reading across languages (Nag & Snowling, 2012).

Here, we will concentrate on the predictors of decoding development in alphabetic orthographies. The longitudinal studies discussed above reveal three cognitive factors that are important influences on how well children learn to decode in alphabetic orthographies such as English: phoneme awareness, letter-sound knowledge, and RAN. Evidence that phoneme awareness and letter-sound knowledge are causal influences on the development of decoding in English comes from the fact that interventions to improve these skills facilitate reading development (Hulme, Bowyer-Crane, Carroll, Duff, & Snowling, 2012). RAN is another likely causal influence on the development of decoding fluency (Kirby, Georgiou, Martinussen, & Parrila, 2010) and appears to tap into the integrity of brain processes that are recruited to form the basis of the brain's word reading network (Lervåg & Hulme, 2009; Norton & Wolf, 2012).

When considering alphabetic orthographies that are more transparent than English, some have argued that phoneme awareness is a less important predictor of the development of decoding. Landerl,

Freudenthaler, et al. (2019) explored the role of RAN and phoneme awareness as predictors of reading in five languages (English, French, German, Dutch, Greek). They reported that RAN was a consistent longitudinal predictor of reading fluency in all five languages but suggested that ‘the association between phoneme awareness and reading was complex and mostly interactive’. Similar conclusions were drawn from studies which explored the concurrent correlates of dyslexia in five languages (Finnish, Hungarian, German, Dutch, French, English) concluding that phoneme awareness was a weaker correlate of dyslexia in regular orthographies than in English (Landerl, Ramus, et al., 2013). However, both of these studies focus on children with significant reading experience; this complicates the picture since phoneme awareness is known to be a particularly important predictor of reading *early* in development (Lervag, Braten, & Hulme, 2019) and it is well established that reading experience has a reciprocal influence on phoneme awareness (Morais, Bertelson, Cary, & Alegria, 1986; Nation & Hulme, 2011).

A clearer picture emerges from studies that start before children have made any appreciable progress in learning to read. Caravolas et al. (2012) reported a longitudinal study of reading development in four languages differing in orthographic transparency (English, Spanish, Slovak, and Czech). The study began just before, or very soon after, the start of formal literacy instruction in each language. Phoneme awareness, letter-sound knowledge, and RAN measured at the onset of literacy instruction were equally important longitudinal predictors of reading and spelling skills across the four languages. In a follow on study, Caravolas et al. (2013) examined the growth of word reading skills in English, Spanish, and Czech children over a period of 30 months from kindergarten through Grade 2. The growth of reading skills was slower and followed a different trajectory in English than in two more regular orthographies (Spanish and Czech). However, phoneme awareness, letter-sound knowledge, and rapid automatized naming measured at the onset of literacy instruction were similarly important as predictors of variations in the rate in growth across the three languages. Hence, although children learn to read more rapidly in more transparent than in less transparent orthographies, it appears that there are universal influences on learning to read in all alphabetic orthographies.

Possible differences between languages in the factors that influence the development of reading comprehension have also been investigated. It has been suggested that, in orthographies with relatively consistent letter-sound mappings, decoding is a less powerful predictor of reading comprehension than in English (García & Cain, 2014; Salceda et al., 2014). This appears to be because decoding is acquired more quickly in these orthographies, and hence,

language comprehension skills become a more powerful limiting factor. This claim was supported by a meta-analysis (Florit & Cain, 2011) which showed that the association between decoding and reading comprehension in relatively transparent orthographies was weaker than in English. Caravolas, Lervåg, Mikulajová, Defior, Seidlová-Málková, and Hulme (2019) provided further support for this view. In four languages (English, Spanish, Czech, Slovak), variations in decoding skills predicted reading comprehension but language skills were a more important predictor of reading comprehension in the three more transparent orthographies (Spanish, Slovak, and Czech) than in English (where variations in decoding were more important).

### Diagnosis and prevalence of reading disorders

Longitudinal studies point to the fact that oral language weaknesses are a major risk factor for reading disorders. They also highlight the predictors of individual differences in decoding in English: phoneme awareness, letter knowledge, and rapid automatized naming. Although reading skills follow a normal distribution in the population, it remains important to decide when ‘poor reading’ warrants a clinical diagnosis.

DSM-5 (American Association of Psychiatry, 2013) recognizes that some people have a life-long specific learning disorder (SLD) that affects reading, writing and/or arithmetic and, because these disorders frequently co-occur, places them together in this one category. To qualify for diagnosis, the affected skill (reading, writing, or arithmetic) must have been significantly impaired relative to age expectation for at least 6 months despite intervention, and the onset must have been in the school years (though the manifestation may change over time). DSM-5 recommends adding a specifier to the diagnosis of ‘reading disorder’ to indicate whether accuracy, fluency, and/or reading comprehension are affected. Specifically, it is important to differentiate problems affecting word-level reading accuracy and fluency from problems affecting reading comprehension (see Castles, Rastle, & Nation, 2018 for a review).

Similarly, ICD 11 (World Health Organisation, 2018) uses the term ‘developmental learning disorder’ as an overarching category, which includes subcategories with impairment in reading, in written expression, in mathematics or with other specified impairments of learning. Again, for each of these diagnoses, performance in the academic domain must be below expectation, not only in relation to age but also in relation to intellectual ability. ICD-11 therefore retains aspects of the heavily criticized ‘IQ-discrepancy definition’. In neither classification system is a diagnosis of learning disorder appropriate if there is evidence of intellectual impairment or other adverse factors affecting education, including proficiency in the language of instruction.



Moll et al. (2014) investigated the prevalence of specific learning disorders (defined as low performance in one or more domains of learning rather than by a clinical diagnosis) in a large representative sample of German children in 3rd and 4th Grade. In contrast to English, where most individuals with reading disorder (dyslexia) also have spelling deficits, these two disorders dissociate about 50% of the time in German speakers. Moll et al. (2014) reported the prevalence of reading disorder and spelling disorder, both as single disorders and/or in combination with each other and/or with arithmetic disorder. Using a criterion of  $-1.5$  SD to define a disorder, they report prevalence rates for isolated disorders of 3.8% for reading disorder, 5.0% for spelling disorder and overall prevalence rates of 7.4% for reading disorder, and 8.9% for spelling disorder (4.5% for arithmetic disorder). A further finding was that arithmetic disorder was more likely to occur with spelling than with reading disorder, suggesting that they may draw on similar cognitive and brain mechanisms.

### Gender ratio

Epidemiological studies have tended to report more boys than girls with reading disorders and a sex difference in reading development (favouring females) that increases along with the severity of reading difficulties (Arnett et al., 2017; Quinn & Wagner, 2015). In contrast, Moll et al. (2014) reported no gender differences in reading disorder but did identify more boys than girls with a spelling disorder and more girls with an arithmetic disorder. While differences in the reported sex ratio between studies are likely related to measurement issues or to sampling bias, an issue of clinical concern is a lack of agreement between ascertainment based on research criteria and school-based identification procedures: Quinn and Wagner (2015) reported that only about 20% of children identified as having reading problems using objective tests were identified as learning disabled by schools and, in particular, it was females who were less likely to be identified.

### Risk factors for poor reading

Longitudinal studies provide us with important information regarding some of the risk factors for poor reading. Such risk factors can be identified at different levels of explanation: biological, cognitive, and environmental. We know most about the cognitive risk factors and therefore will begin with these; however, before proceeding it is important to acknowledge that methodological differences between studies can influence how we conceptualize disorders. Is it now recognized that neurodevelopmental disorders, including SLDs, are dimensional with no clear cutoff from normal variation (Thapar et al., 2017). One major approach to studying

reading disorder therefore is to assess the longitudinal predictors of reading across the distribution and to identify putative causal risk factors. Studies of children at high risk of reading disorder are useful in this regard. A quite different approach is to study children who have already developed specific learning disorders. Such an approach, particularly if it recruits a clinically diagnosed sample, is likely to identify a wider range of deficits, some of which will be causal, whereas others may be secondary consequences of the disorder and still others may be co-occurring features (comorbidities). Based on these different types of study, we aim to provide a critical overview of the current state of knowledge with regard to reading disorders/dyslexia. We will then turn to consider disorders commonly comorbid with dyslexia and draw together evidence within a multiple deficit framework (McGrath, Peterson, & Pennington, 2020). While acknowledging that different types of reading difficulty are associated with distinct risk factors, our review primarily focuses on dyslexia where most is known; we refer to reading comprehension impairment when discussing language disorders (see Bishop et al., 2009; Snowling, Hayiou-Thomas, Nash, & Hulme, for more details).

### Cognitive risk factors

*Phonological deficits.* While learning disorders appear to reflect multiple risk factors, the predominant theoretical view is that difficulties with reading accuracy and fluency (RD/dyslexia) arise from difficulties with phonological processing, and here, the findings of longitudinal and clinical studies converge (Melby-Lervag, Lyster & Hulme, 2012). Thus, the effect size for differences between groups with reading disorder and typical readers on tasks tapping phoneme awareness is very large relative to the performance of children of the same age ( $d = -1.37$ ) and large relative to children reading at the same level ( $d = -0.57$ ). Group differences in rime awareness and in verbal short-term memory are smaller but still substantial relative to age-controls. Speeded naming of familiar objects and digits (RAN), a skill requiring the rapid retrieval of stored phonological information (the names of familiar items), is also associated with reading skill; interestingly, RAN appears to be a skill that differentiates between children with language disorders who do, and do not, develop dyslexia (e.g., Ramus, Marshall, Rosen, & van der Lely, 2013). In addition, clinical symptoms include deficits in nonword repetition, word finding and in new word learning but, arguably, these may be facets of comorbid language difficulties.

*Auditory and speech processing deficits.* Beyond phonological skills, a number of longitudinal studies suggest that children with a broader range of sensorimotor deficits are more likely to develop reading problems (e.g., Carroll, Solity, & Shapiro, 2016; van

der Leij et al., 2013) though whether or not they have a direct effect on reading remains unclear. A prominent hypothesis is that auditory processing problems predispose a child to problems with speech perception that underpin their phonological processing difficulties. A substantial number of studies show differences between children with dyslexia and controls on auditory processing tasks (Hämäläinen et al., 2013); however, it has been suggested that problems in attention control explain this relationship (Schulte-Körne, 2010). Consistent with this hypothesis, Snowling, Gooch, McArthur, and Hulme (2018) showed that variations in frequency discrimination at age 5½ years were predicted by performance on executive function tasks a year earlier in a large sample of children, including children at risk of dyslexia. Moreover, auditory processing was not a longitudinal predictor of reading.

Similarly, O'Brien, McCloy, Kubota, and Yeatman (2018) reported large group differences between 8- and 10-year-old readers with dyslexia and controls on two phoneme categorization tasks. Importantly, however, lapses of attention were more common in those with dyslexia, especially when performance was measured in a demanding paradigm. Moreover, contrary to the theory, the relationship between speech categorization and reading was not mediated by phonological awareness (see also Hakvoort et al., 2016; Snowling et al., 2018).

In short, despite the large number of studies of auditory and speech processing in dyslexia, evidence for a causal association between these skills and reading development is lacking.

*Visual processing deficits.* Vision is another area of sensory processing that has attracted much attention from dyslexia researchers, specifically the putative roles of deficits in the magnocellular visual system or in visual attention. According to Grainger, Dufau, and Ziegler (2016), visual deficits in dyslexia are most likely to affect the development of orthographic representations. Orthographic processing, they argue, requires the parallel processing of letters within words and typically within sentence frames. It follows that the efficiency of these processes will depend upon visual acuity (which is better closer to fixation), possible crowding among letters (which is less when there are spaces between letters) and visuo-spatial attention (the span of apprehension beyond the fixated word). Any of these processes could be affected in dyslexia (see Boden & Giaschi, 2007 for a review).

Saksida et al. (2016) assessed a large sample of French-speaking children with dyslexia aged 8–13 years, examining their performance on tasks tapping visual attention span, visual stress (the experience of visual distortions when reading text), and phonological skills. The most widespread difficulties were in phonological skills (92.1%); in

contrast, only 5.5% of the group experienced visual stress (which was actually more frequent among controls). Although 28.1% of the children with dyslexia showed reduced visual attention span, all of these children also experienced phonological deficits. Grainger et al. (2016) argue that this co-occurrence of poor phonological skills and limited visual attention is to be expected for two interconnected reasons. First, there is a close association between phonological skills and reading development. Second, spatial attention develops alongside literacy skills in childhood (White, Boynton, & Yeatman, 2019; see also Olulade, Napoliello, & Eden, 2013).

More generally, research on visual factors in dyslexia has been hampered by a dearth of longitudinal studies assessing the possible association between visual factors and individual differences in reading. In one such study, Valdois et al. (2019) measured visual attention span in a large sample of French kindergarten children, along with tests of letter knowledge, early reading skills, phonological awareness (at the level of syllables and rimes), and verbal short-term memory, re-assessing their reading at the end of Grade 1. There was a significant effect of early measured visual attention span on measures of word, nonword, and text reading fluency in Grade 1 when kindergarten measures of letter knowledge and phonological awareness were controlled. However, the absence of any direct effects of phonological awareness in this study is inconsistent with a large body of evidence on this issue and the proposed model accounts for only a small amount of variance in reading outcome. It is plausible that, had tests tapping phonemic skills been included, the findings would have been different. Nonetheless, the study represents an important direction for future research aimed at gaining a better understanding of visual factors which may compromise learning to read.

*Learning deficits.* Learning to read requires the associative learning of mappings between symbols and sounds (letters and phonemes in alphabetic languages). It follows that some children with dyslexia, rather than experiencing modality-specific deficits, may have deficits in associative learning. An influential hypothesis is that a deficit in implicit (procedural or statistical) learning is a causal risk factor for both dyslexia and developmental language disorder (Ullman & Pullman, 2015). However, a recent meta-analysis suggests that the evidence that dyslexia is associated with a statistical learning deficit is subject to publication bias (Witteloostuijn et al., 2017). Further, West, Vadillo, Shanks & Hulme (2018) found that currently used measures of implicit learning have very low reliabilities and do not predict variations in reading skills in the general population; indeed, there are low correlations between different statistical learning tasks and

reading skills and the causal relationships between reading and statistical learning are unproven (Schmalz et al., 2019).

Another learning task that may be critical for learning to read is the ability to map between visual and verbal domains (print to sound). Warmington and Hulme (2012) showed that, in addition to phoneme awareness and RAN, visual-verbal paired association learning was a predictor of word reading and decoding skills in 7- to 11-year-old children. Further Litt, de Jong, van Bergen, and Nation (2013) showed that it was not the cross-modal nature of this learning task that was critical but rather the requirement for verbal output that accounts for its relationship with reading (see also Clayton, Sears, Davis, & Hulme, 2018).

Together, these findings refute the hypothesis that dyslexia primarily affects the learning mechanisms that are required for the development of orthographic relationships – rather they suggest that specific learning deficits stem from broader impairments; while described as ‘verbal’, it seems likely they are in the domain of phonological skills.

*Summary.* At the cognitive level of explanation, there have been various hypotheses regarding the causes of reading disorders. In this area, there are significant problems associated with unraveling the causes of reading disorders from their consequences; this problem is particularly acute when the aim is to differentiate the roles of language versus visual skills as foundations for learning to read, given the established reciprocity between reading and other cognitive skills. Notwithstanding this, there is strong evidence that the most prevalent core deficit in dyslexia is in phonological skills, and phonological deficits predate literacy acquisition.

## Biological risk factors

### *Heritability of reading and language skills*

It is well established that reading skills and the phonological skills that underpin them are highly heritable (Christopher, Hulslander, et al., 2015; Little et al., 2017). Tosto and et al. (2017) used data from the UK Twins Early Development Study (TEDS) to examine the proportion of variance in reading ability attributable to genetic and environmental factors at ages 7, 12 and 16 years. There were common genetic influences affecting reading fluency at all time-points, and also novel genetic factors that came into play from age 12 years. At no point was the variance due to environmental factors large and it was only significant at age 7 years. In the same study, data were available from tests of language and phonological skills. There was substantial shared genetic variance between reading fluency and phonological skills and, to a lesser extent, between reading fluency and language. Importantly, however,

the genetic correlation between language and reading comprehension was high (above 0.8), suggesting the same genetic influences were acting on both of these skills, consistent with the evidence that oral language skills have a direct and long-term influence on reading comprehension.

### *Genetic risk factors*

Heritability estimates are based on group data and tell us little about the risk to an individual of developing a reading disorder. It is possible that advances in molecular genetics will move the field toward more precise predictions of who is likely to be dyslexic. The use of the genome-wide association (GWA) method has provided important leads. In a GWA study, the entire genome is scanned to identify gene loci associated with a range of reading phenotypes (e.g., nonword reading, phonological awareness, RAN) in very large samples of affected (dyslexic) and unaffected individuals. The focus is on a number of DNA variants (single nucleotide polymorphisms or SNPs) to see whether a specific allele at a given gene locus is correlated with an individual's score on a particular trait. This strategy has revealed many possible associations and at least 17 candidate SNPs (Carrion-Castillo et al., 2013; Gialluisi et al., 2019). However, in all but two cases, it has been difficult to replicate these associations in independent samples, and generally, effect sizes are extremely small (Bishop, 2015). The fact that reading is measured in different ways and at different ages in different cohorts, and that there are differences in recruitment and ascertainment criteria in case-control studies, contributes to the lack of consistency across studies.

Despite these failures of replication, some findings are of note. A gene locus on chromosome 15 – DYX1 – was the first to be identified and there has been considerable support through replication in independent samples. At this locus, a candidate gene DYX1C1 with a potential role in brain development (growth and function of cilia; also see Paracchini, Diaz, & Stein, 2016) has attracted much attention. Another linkage on chromosome 6 – DYX2 – has also been consistently associated with dyslexia (Zhou et al., 2012). One of the candidate genes in this area, KIAA0319, is especially interesting because it seems also to be linked with phenotypes of other neurodevelopmental disorders, such as ADHD or developmental language disorder (DLD or SLI; Newbury et al., 2010), suggesting that some of the genetic effects are pleiotropic.

In short, the genetic basis of dyslexia is heterogeneous. Furthermore, a complex trait like reading can be expected to be influenced by many genetic variants, each of small effect, and there are likely to be gene-gene and gene-environment interactions. It follows that aggregating genetic effects across SNPs within an individual should increase predictive

power if the SNPs combined all have some (weak) association with reading traits. Polygenic risk scores can be computed from a weighted average of many different alleles associated with a trait (reading).

The first study to examine polygenic risk scores and reading ability used the number of years of education as a proxy for reading ability (based on the assumption that educational attainment is closely related to reading). Since a measure of years of education is collected as a standard demographic variable in all GWA studies, data can be aggregated across many samples to provide sufficient power to derive a genome-wide polygenic score. Okbay et al. (2016) used DNA from more than 300,000 individuals to derive a polygenic risk score called 'EduYears'; this score accounted for 4% of the variance in years of education. In a related study, Selzam et al. (2017) found that this polygenic risk score accounted for 2.1%–5.1% of variance in reading ability across development and 2.7% of the variance in a general cognitive ability factor. When general cognitive ability and family SES were controlled, the polygenic risk score predicted a mere 0.5%–1% of variance in reading skills.

More recently, Gialluisi et al. (2019) reported data from a genome-wide association study of nine cohorts of European children ( $N > 3,000$ ). This study identified an association between a gene locus on chromosome 18 with rapid automatized naming for letters and a locus with less strong association on chromosome 8. A further analysis investigated the genetic overlap between the 'EduYears' polygenic risk score, dyslexia-related traits, and other commonly associated phenotypes including ADHD, autism-spectrum disorder, depression, and schizophrenia. Genetic overlap was found between the 'EduYears' risk score and ADHD. In addition, variants associated with the 'EduYears' risk score accounted for some 2% of the variance in reading (and also showed associations with spelling, non-word reading, phonological awareness, and digit span).

To summarize, progress is being made in understanding how genes confer the risk of dyslexia. However, polygenic risk scores currently explain very small amounts of variance in reading skills; simple cognitive measures therefore have much more clinical relevance.

### *Brain structure and function*

The ultimate goal of genetic research into dyslexia is to understand the molecular pathways through which genes affect brain development (Mascheretti, et al., 2017), but longitudinal studies are needed to relate variations in brain development to the growth of reading skills.

In an important example of this approach, Preston et al. (2016) found that the co-activation of regions involved in processing speech and print across the

left hemisphere's reading network in beginner readers was a predictor of reading skill two years later when general activation in each region alone and pretest reading scores were controlled.

Similarly, Hoeft et al. (2011) followed children with dyslexia for 2½ years taking measures of phonological skills, reading, and brain structure and function. At the start of the study, children had MRI scans while completing a written rhyme judgment task; the scans were analyzed to identify patterns of brain activation and brain structure in regions of interest. A novel finding was that the children with dyslexia who improved the most over time in reading showed more activation in the right frontal cortex and greater white matter integrity in the right arcuate fasciculus at baseline. It appeared therefore that this structure, which connects the right frontal and parieto-temporal regions, had been recruited to compensate for under-activation in homologous left hemisphere regions.

The next step was to examine the structure of the left and right arcuate fasciculus in children who had been assessed on tests of prereading and cognitive skills in preschool and were reassessed on reading measures in Grade 3 (Myers et al., 2014). In this study, measures of preliteracy and a measure of change in white matter density in the left arcuate fasciculus over time accounted for a substantial proportion of the variability in reading in Grade 3 (56%). In fact, growth of this 'temporal' pathway continued to predict individual differences in reading when prereading in kindergarten, socioeconomic status, and measures of familial factors including the home literacy environment were controlled. A further study by the same group focusing on children at family risk of dyslexia reported that cortical thickness in left brain reading-related areas was lower in children at family risk, while the surface area of right cortical regions was higher.

The differences in brain connectivity associated with the growth of reading skills in the Myers et al. (2014) may have at least two different explanations. On the one hand, variations in brain development may reflect a causal constraint on the development of reading skills; conversely, children who learn to read better may show experience-related changes in brain development (more practice in reading may result in increases in myelination in the neural circuits responsible for reading). Huber et al. (2018) explored these possibilities using diffusion MRI data to monitor changes in brain connectivity in children undergoing an intensive reading intervention lasting 8 weeks. They found large-scale changes in a collection of white matter tracts that paralleled increases in reading skill. However, they also found tracts which were predictive of reading skill that did not change in volume as a result of intervention. This study was very short-term, but it suggests that there may be certain fiber tracts that show structural changes quite rapidly as a result of increased



reading practice and skill, while other structures that may be causally related to reading skill are less malleable as a result of intervention. Further studies of this sort over longer time periods would clearly be very informative.

### Familial risk of dyslexia

A potential way of elucidating how biological risk factors are related to cognitive liabilities is to study children at family risk of dyslexia by virtue of having a first degree affected relative with dyslexia, usually a parent. Such studies also offer a way of studying gene–environment interactions and how these might lead to specific learning disorders. There have now been some 20 family risk studies of dyslexia published. In these studies, children at family risk of dyslexia are followed longitudinally with controls (without familial risk of dyslexia) from preschool until around Grade 3 when children are classified as dyslexic or not. This approach is a powerful way of identifying the precursors of dyslexia.

A meta-analytic review of such studies involving children learning to read in English, Danish, Dutch, Finnish, and Chinese confirmed an elevated risk of dyslexia in the offspring of affected parents, with a mean prevalence rate of 45%, compared with 12% for children not-at-risk (Snowling & Melby-Lervåg, 2016). Further, the review showed that children who go on to reach criteria for dyslexia have significant language difficulties in the preschool years, chiming with the findings of longitudinal studies of representative samples of children. In addition, they show phonological difficulties and have problems acquiring the foundations of decoding (letter knowledge, phonological awareness, and RAN). Importantly, there was no evidence of significant differences in the precursors of dyslexia across the languages studied (see also Moll et al., 2016) for Czech and Slovak children at family risk of dyslexia) although morphological awareness was only measured in Chinese where it was found to be important predictor of outcome (Tong, McBride, Lo, & Shu, 2017).

The incidence of early language difficulties in children who go on to be classified with dyslexia raises the question of the relationship between dyslexia and developmental language disorder (DLD). Snowling, Nash, Gooch, Hayiou-Thomas and Hulme (2019) followed children at family risk of dyslexia, children with preschool language difficulties and controls from age 3½ to 9 years. Dyslexia at age 8 years was defined as  $-1.5$  *SD* relative to the control mean on a composite reading and spelling score: 29% of children at familial risk were classified as having dyslexia and 33% of children with preschool language difficulties. At the same age, developmental language disorder (DLD) was defined as falling as  $1.5$  *SD* below the control mean on a composite measure of receptive and expressive

language skills. This led to the formation of four outcome groups: Dyslexia-only, DLD-only, Dyslexia + DLD, and Typical Control. All three clinical outcome groups showed large deficits in letter knowledge, phoneme awareness, and RAN at age 4½ years. However, from 5½ years onwards, the DLD-only group showed smaller deficits than either of the two groups with dyslexia.

In the same sample, Hayiou-Thomas, Carroll, Leavett, Hulme, and Snowling (2017) examined the outcomes of children with preschool speech difficulties in the absence of language disorder. At age 5½ years, these children showed deficits in phoneme awareness and early spelling. However, the risk of reading impairment was relatively mild and short-lived except among children who showed disordered speech errors or had co-occurring language disorders. However, Burgoyne, Lervag, Malone, and Hulme (2019) in a large, broadly representative sample of children found that speech errors at school entry were correlated with broader oral language difficulties, and were an independent predictor of later reading difficulties. Furthermore, the effects of early speech difficulties on later reading were entirely mediated by difficulties in developing phoneme awareness, and these effects were independent of broader oral language skills. Speech difficulties are easily noticed and children with such difficulties at school entry should be monitored for language and reading difficulties. Together, these findings highlight the important role of shared risk factors between dyslexia and other language learning disorders, an issue to which we return in the section on comorbidities.

## Environmental risk factors

### Family environment

There is a well-established socioeconomic gradient in children's reading skills; however, this is likely to reflect multiple influences including family, home, and school factors (Jerrim, Vignoles, Lingam, & Friend, 2015). From a very early stage, the home literacy environment has a direct effect on the development of vocabulary and pre-reading skills (Sénéchal & LeFevre, 2014) although, even in the early stages of reading, there is evidence that letter knowledge and phonological awareness are moderately heritable skills (Byrne et al., 2005). Chow et al. (2017) examined the relationships between family socioeconomic status, the home literacy environment and reading and language in a sample of Cantonese-speaking twins aged 3–11 years. Both socioeconomic status and home literacy environment mediated environmental influences on reading and language, but genetic influences were not moderated by these factors. However, the sample size was small given the age range and these findings need replication.



It might be expected that children at family risk of dyslexia will experience a different home environment to controls (because adults with dyslexia tend to read less), but evidence on this issue is limited. Hamilton, Hayiou-Thomas, Hulme, and Snowling, (2016) assessed the home literacy environment of 4-year-old children at family risk of dyslexia and controls with no known risk of reading difficulty. Questionnaire measures assessed both informal (storybook exposure) and formal home literacy practices (direct parental instruction in decoding). When socioeconomic status was controlled, there were no group differences in the home literacy environment experienced by children at family risk and controls. In the family risk group, both storybook exposure and direct instruction at age 4½ years predicted phoneme awareness and emergent decoding skills at age 5½ years which, in turn, predicted reading at age 6½ years. In controls, the influence of storybook exposure on phoneme awareness was not significant but otherwise the model was the same: storybook exposure was a predictor of oral language at 5½ years which, together with decoding, predicted reading comprehension a year later.

A critical question is whether the home environment affects child reading outcomes once other heritable influences are taken into account. Van Bergen, Zuijlen, Bishop, and de Jong (2016) conducted a study in which families were assessed for reading fluency and completed a survey assessing the home literacy environment. Several aspects of the home environment were correlated with parental reading skills and habits, throwing doubt on the hypothesis that home literacy environment is a pure environmental measure. In similar vein, Puglisi, Hulme, Hamilton and Snowling (2017) found no direct effect of measures of informal literacy on child outcomes after controlling for maternal language and literacy skills (suggesting that such effects may reflect, at least in part, genetic effects); however, direct instruction did appear to be a true environmental influence on later decoding skills.

Finally, one factor which is widely believed to predict reading fluency is the amount of reading a child does outside of school, perhaps in the home. Van Bergen et al. (2018) extracted data from the Netherlands Twin Register to estimate genetic and environmental influences on reading ability. Using parental ratings of the amount children read and parent and teacher ratings of reading ability they found, as expected, that reading ability was highly heritable, while print exposure was influenced equally by genetic and environmental factors. Direction of causality models suggested that reading ability was the driver of how much children read and not vice versa. A clear implication is that poor readers need to be encouraged to practice reading (at home or elsewhere); otherwise, they are more likely to avoid doing so.

### *School environment*

Classic studies of school effectiveness show wide variations in reading attainment among pupils attending schools varying in quality (Sammons, Nuttall, & Cuttance, 1993). A key question, therefore, is whether school-level environmental factors moderate genetic influences on literacy attainments. A small number of studies have attempted to answer this question using genetically sensitive designs, but findings are inconsistent. Haughbrook, Hart, Schatschneider, and Taylor (2017) compared the etiology of reading skills in twins attending schools that varied in quality, comparing 'A' schools (in which the progress of pupils is generally better) versus 'non-A' schools (with lower pupil attainments). Shared environmental influences between twins were larger in 'non-A' than 'A schools', and conversely, there were greater genetic influences on the reading of those attending 'A' schools. Since children of lower socioeconomic status who have experienced a poorer home literacy environment are more likely to attend a low-quality school, the authors suggest that the poor school environments may have depressed relatively poorer pre-reading skills further.

Similarly, Taylor, Erbeli, Hart, and Johnson (2019) investigated the outcomes in 7th to 10th grade of twins who had been studied during the early years of schooling. A measure of gains in reading fluency for all children in the 1st- and 2nd-grade classrooms in the schools attended by these children was taken as a measure of classroom environment. When the quality of the environment was defined as poor, there was more variability in reading outcomes in adolescence (and variance due to shared environmental influences was high). In contrast, when the early classroom environment was good, outcomes were better and there was less variability in reading skills that were subject to strong genetic influences. However, these arguments are circular (since school classrooms were defined according to pupil attainment) and Grasby, Coventry, Byrne, and Olson (2019) failed to find equivalent effects in an Australian sample of twins.

Notwithstanding this, it would be odd if there were no environmental influences on reading given that it is a learned skill that depends on practice. Indeed, Lervåg Dolean Tincas and Melby-Lervåg (2019) reported a direct effect of socioeconomic status (SES) as well as school absences on the growth of vocabulary and reading comprehension in Roma children being brought up in severe poverty, after controlling for relevant cognitive and linguistic measures. It is likely that the range of environments in this study was wider (with more children living in highly disadvantaged circumstances) than in the studies described above.

In summary, theories of reading disorder need to take account of both the genetic and environmental

influences on learning to read. While our understanding of how these interact over time remains limited, findings serve to remind us that the classroom environment provides a critical foundation for later educational attainment and may be particularly valuable in compensating for poorer home circumstances.

### **Comorbidities of reading disorder**

It is now widely accepted that neurodevelopmental disorders co-occur more often than expected by chance. This co-occurrence may arise because of shared risk factors, because one disorder is the developmental precursor of another or because one disorder confers risk for another disorder. There is good evidence for high rates of comorbidity between dyslexia and mathematics disorder, attention deficit hyperactivity disorder (ADHD), developmental language disorder, speech sound disorder, developmental coordination disorder, as well as with disorders of mental health including anxiety, depression, and conduct disorder. Reported rates of comorbidity are higher than expected by chance and depend on the criteria used to define reading or other disorders (Branum-Martin, Fletcher, & Stuebing, 2013). Thus, the field is moving toward approaches that examine correlations between normally distributed dimensions of reading with other skills in order to identify specific and shared risk and protective factors.

#### *RD and mathematics disorder*

Among specific learning disorders, the co-occurrence of reading disorder and mathematics disorder (MD, dyscalculia) is common (Koponen et al., 2018; Moll, Landerl, Snowling, & Schulte-Körne, 2019). Willcutt et al. (2013) examined a large group of children with RD and/or MD. All groups showed deficits in verbal comprehension, working memory, naming speed, processing speed, inhibition, vigilance, and response variability, which had additive effects, and there were interactions between RD and MD for phoneme awareness, Stroop performance, and set-shifting. These findings suggest that RD and MD share risks in verbal comprehension, working memory, and processing speed, but in addition, there are risk factors specific to each condition (see also Cirino, Fuchs, Elias, Powell, & Schumacher, 2015).

#### *RD and ADHD*

Reading disorder and ADHD show high rates of comorbidity (20%–40%). McGrath and et al. (2011) investigated the specific and shared cognitive deficits associated with these conditions in a sample ranging from 8 to 18 years of age. There were three predictors of reading: phonological awareness, RAN, and processing speed. Phonological awareness and RAN were uniquely associated with reading, and

processing speed accounted for shared variance with ADHD inattentive symptoms. Inhibition was associated with both inattentive and hyperactive/impulsive symptoms but not with reading skills. Thus, processing speed is a predictor of both RD and ADHD dimensions, and may account for the association between reading and inattention. These findings dovetail with those of Wadsworth DeFries, Willcutt Pennington & Olson (2016) who found that genetic influences accounted for the overlap between RD and the inattentive form of ADHD, whereas they were less important in the etiology of the hyperactive/impulsive subtype.

Together, these findings suggest that slow processing speed is a shared risk factor for reading disorder and ADHD and possibly for other disorders as well. Peterson et al. (2017) assessed the risk factors for reading, mathematics, and attention disorders in a large sample of twins aged 8–16 years. Verbal comprehension and nonverbal processing speed predicted both reading and mathematics; in addition, phoneme awareness and verbal processing speed (RAN) predicted reading, while visual working memory predicted mathematics, and inhibition predicted attention. Verbal processing speed partially accounted for the overlap between attention and reading and mathematics, whereas verbal comprehension accounted for the overlap between reading and math (see a meta-analysis by Daucourt et al. (2020) for etiological influences on the overlap between reading, mathematics and attention disorders).

#### *RD and language disorder*

Another disorder that frequently co-occurs with reading disorder is developmental language disorder (DLD). It seems most likely that this co-occurrence is an example of a liability (language disorder) that has downstream effects on the development of both reading and mathematics. It is now well established that poor language is a risk factor for reading disorder; approximately 50% of children with dyslexia in clinical samples have been found to fulfill criteria for DLD and about 50% of children with DLD have significant reading impairments (Bishop et al., 2017) though the risk of children with DLD developing dyslexia is lower in epidemiological samples (Catts, Adlof, Hogan, & Weismer, 2005). A more common outcome for children with DLD is reading comprehension impairment. Poor comprehenders have weak vocabulary knowledge (Henderson, Snowling, & Clarke, 2013) and difficulties with some aspects of morphology and syntactic awareness (Adlof & Catts, 2015; Tong, Deacon, & Cain, 2014) as well as in specific components of comprehension, such as inferencing, comprehension monitoring, and in inhibiting irrelevant information (Borella, Carretti, & Pelegrina, 2010; Pimperton & Nation, 2010).

Nation, Cocksey, Taylor, and Bishop (2010) followed the reading and language development of 242

children from age 5 to 8 years, identifying 15 children who at 8 years exhibited the 'poor comprehender' profile (normal reading fluency but impaired reading comprehension). As a group, the poor comprehenders showed normal phonological skills but mild to moderate impairments in oral language skills. Although not specifically focusing on poor comprehenders, Oakhill and Cain (2012) included measures of comprehension and working memory in a longitudinal study of 100 children from age 7–8 (*t1*) through 8–9 (*t2*) to 10–11 (*t3*) years. Verbal IQ, vocabulary, and knowledge of story structure at *t1* together predicted later reading comprehension outcomes; in the same model, vocabulary at *t1* predicted inference making skills at *t2* which, together with comprehension monitoring ability, accounting for additional variance in reading comprehension. Although performance IQ is a predictor of comprehension monitoring, the skills found to underpin reading comprehension are primarily verbal and differ from the predictors of reading accuracy/dyslexia which include phonological skills. Moreover, it is important to bear in mind that children with language disorders experience a range of other deficits, notably in arithmetic (Cross, Joanisse, & Archibald, 2019; Durkin, Mok, & Conti-Ramsden, 2013) and in the development of executive function and motor skills (Gooch, Hulme, Nash, & Snowling, 2014). Thus, when dyslexia is comorbid with DLD, a range of other problems are likely to be associated.

Together, these findings suggest that preschool language difficulties may be associated with a range of different developmental trajectories. Arguably, language difficulties in some children resolve but leave them at risk of relatively circumscribed phonological problems (this is the 'classic' dyslexia profile), while for other children broader oral language problems persist either without concomitant phonological problems (this is the classic poor comprehender profile) or with concomitant phonological problems (the profile of a generally poor reader, who has both decoding and comprehension difficulties: 'Dyslexia + DLD'). To complicate matters further, some children, especially those at family risk of dyslexia, may have language abilities that are within the normal range in preschool but show late emerging language difficulties during the early school years (Snowling et al., 2016).

### **A multifactorial framework for reading disorders**

For many years, dyslexia was considered a specific disorder arising from a selective phonological deficit and there is strong support for phonological deficits being causally related to reading difficulties. However, it is now recognized that a phonological deficit may be neither necessary nor sufficient to explain the heterogeneity of the condition (e.g., Catts, McIlraith, Bridges, & Nielsen, 2017; Saksida et al.,

2016). While there is little convincing evidence for a direct causal role of auditory processing, speech perception, or visual deficits on reading development, it is important to bear in mind that, at the individual level, such difficulties may exacerbate a reading problem or account for some of the co-occurring features. Indeed, the many risk factors associated with dyslexia and its frequent comorbidity with other disorders suggest that a multifactorial model is needed to explain the condition.

The multiple-risk framework for dyslexia can be traced to Pennington (2006) among others who argued that risks for neurodevelopmental disorders operate probabilistically across biological, cognitive, and environmental levels until they reach a 'diagnostic threshold', determining not only the categories of developmental disorder but also their comorbidities. According to McGrath et al. (2020), the multiple deficit model accounts for some 75%–85% of the variance in the comorbidity of reading and maths but has been less successful in explaining the comorbidity between RD and ADHD (where it is hypothesized that the addition of emotion regulation deficits may improve accuracy of the model). The model makes a further prediction: that risk factors interact with compensatory factors to improve outcomes. At present, there is a paucity of direct evidence for this view.

Exploring the issue of compensation, van Viersen, de Bree, and de Jong (2019) compared children with persisting and resolving dyslexia, matched on IQ, to investigate the comparative strengths of these groups. Groups were comparable on phoneme deletion and nonalphanumeric RAN but those with resolving difficulties showed stronger verbal abilities. However, this was a small-scale study and the interaction between deficits and 'strengths' favouring the resolving group was not tested. More is known about the factors that promote good reading across the population, notably good oral language skills and plenty of practice to develop the high-quality lexical representations that underpin skilled reading comprehension (Perfetti & Hart, 2002).

### *Implications for assessment*

A reasonable implication of the multiple-risk hypothesis of dyslexia is that assessments should tap a broad range of cognitive skills. However, Miciak et al. (2014) have strongly criticized such approaches, not least because there is little evidence of interactions between individuals' strengths and weaknesses. Rather, assessments should focus on the defining symptoms of reading difficulties, the functional impairments, and co-occurring conditions. In fact, brief assessments focusing on reading and proximal skills are effective when a child's response to intervention is monitored (Fuchs & Fuchs, 2017), providing that co-occurring difficulties can be managed.

Research using regression approaches can be useful for identifying optimal assessment batteries.



Thompson et al. (2015) used data from an English sample of children at high risk of reading disorder to show that, in preschool, family risk of dyslexia was the best predictor of a child's dyslexia status at 8 years. At school entry, family risk remained a strong predictor, and poor language was now also significant. From age 6, performance on tests of letter knowledge, phoneme awareness, and RAN produced as good a prediction of outcome as family risk status. These findings dovetail well with those from longitudinal predictive studies of reading development and are consistent with the finding that the risk of reading disorder is reduced among children whose preschool language difficulties resolve by school entry (Snowling, Duff, Nash, & Hulme, 2016). When predicting individual risk of reading comprehension difficulties, poor oral language comprehension and vocabulary are key factors (see above). Second language learners who are required to read in a non-native language are also at risk (Spencer & Wagner, 2017).

Finally, Frijters et al. (2011) examined the predictors of reading outcome among children who showed different levels of response to intervention, defined following growth curve analysis (poor, average, good). The assessment battery included measures of oral language, phonological memory, visuo-motor integration, and IQ as well as a comprehensive range of reading and reading-related tests. In addition to phonological awareness and RAN, which were robust predictors of reading growth, other skills improved the accuracy of classification of good and poor responders by about 18% (depending on the outcome measure). Among these, the most robust predictor was verbal comprehension, with visuo-motor integration, phonological memory, and other IQ components (including freedom from distractibility and processing speed) playing a role in relation to specific outcome measures.

### Outcomes for children with reading disorders

It is widely established that reading difficulties hinder educational attainment. However, there is a dearth of robust evidence concerning longer-term outcomes and very few studies have examined outcomes for children who have received intervention.

#### Reading outcome

Reading ability is highly stable. Moll et al. (2020) examined the one- to two-year stability of deficits in reading and spelling in 167 German-speaking children aged 9 years at the first point of assessment. The correlation between performance at the first time point and one year later was very high for reading fluency ( $r > .9$ ) and high for spelling ( $r = .78$ ). Stability of group membership was moderate in groups with reading disorder but low for a spelling-only disorder group with only 32% remaining impaired

after one year (see Maughan et al., 2009, for contrasting effects in English).

In a similar vein, Elwer, Keenan, Olson, Byrne, and Samuelsson (2013) assessed the longitudinal stability and predictors of poor oral comprehension as well as poor decoding in a large sample of US children over a longer time span (preschool to Grade 4). Children classified as poor decoders in Grade 4 showed weak reading and spelling from the first point of measurement onwards. Reading comprehension profiles were more complex. In the early grades, poor oral language comprehenders actually outperformed poor decoders and it was only in Grade 4 that they showed the 'poor comprehender' profile. This is consistent with the view that in the early years of schooling, reading comprehension depends strongly on decoding skills, whereas later, it is strongly dependent on oral language comprehension.

Finally, from a neuropsychological perspective, there has been interest in whether the cognitive profiles associated with different 'subtypes' of dyslexia, namely phonological dyslexia (in which nonword reading is particularly impaired) and surface dyslexia (in which exception word reading is impaired) are stable over time. Peterson, Pennington, Olson, and Wadsworth (2014) followed adolescents and young adults who had been subtyped in this way when aged 12–22 years, some five years after the first assessment. The phonological subtype was moderately stable but the surface dyslexia profile (rare in this sample) was not; neither subtype was as stable as was dyslexia defined according to psychometrically valid criteria. Furthermore, knowledge of the subtype provided no useful information regarding progress in reading over time. Although there is some evidence that optimal reading interventions take account of child characteristics, cognitive profiles appear to be of limited benefit when planning interventions (Connor et al., 2013).

Together, these findings, and the failure of struggling readers to 'catch-up', mean that the gaps in educational attainment between them and their peers widen over time. This is a *prima facie* argument for reading intervention that starts early and continues (Solis, Miciak, Vaughn, & Fletcher, 2014). Blachman et al. (2014) evaluated the outcomes at ages 19–22 years of 58 children who have received an 8-month intervention in Grades 2 and 3, compared with a control group who had received business as usual. The intervention group remained ahead on standardized measures of basic reading that had been the focus of intervention. Group differences were moderate on these measures ( $d_s = 0.53$ – $0.62$ ) but small to negligible for spelling ( $d = 0.26$ ) and reading comprehension ( $d = 0.06$ ). There was also an indication that the intervention group participated in secondary education longer, and were more likely to go on to complete postschool qualifications. However, the sample size was small and these findings require replication. Arguably, it is

optimistic to think that a relatively short intervention could inoculate children with significant reading difficulties against future failure.

### *Mental health and well-being*

Mental health problems are more common in children with dyslexia (e.g., ADHD, internalizing symptoms, anxiety disorders, depressive disorders, and conduct disorders) and a widely held view is that reading disorders also adversely affect psychosocial adjustment and adult well-being.

Francis et al. (2019) conducted a meta-analysis of studies examining internalizing problems in those with reading disorder. Although the sample size was limited with only 34 studies fitting the criteria, they report moderate associations between reading disorder and internalizing problems as well as a smaller but significant association with depression, confirming findings of earlier reviews. The same group conducted a systematic review of the literature on poor reading and various aspects of self-concept, reporting moderate associations from 13 studies (McArthur et al., 2020). Further, Livingston et al. (2018) following a review of some 100 articles assessing the emotional difficulties associated with dyslexia argued that the effects of dyslexia go beyond the individual to the family and to communities.

It has been suggested from qualitative studies that primary school may be the most difficult period in terms of well-being and self-esteem for poor readers and that a 'diagnosis' can bring a sense of relief and better adjustment (O'Connor, Kadianaki, Maunder & McNicholas, 2018). However, few longitudinal studies have followed children through adolescence into adulthood so evidence is limited. In one follow-up study, Aro et al. (2019) analyzed data from 430 Finnish adults who had been identified clinically as having either Reading and/or Mathematics Disorder. The database used contained records of education, sickness benefits, and other allowances. As expected, fewer of these individuals had attended university than controls, and more had been unemployed; sickness benefits and allowances granted following psychiatric diagnosis were more common, as was the prescription of medications for anxiety and depression. Interestingly, the rate of problems was higher in individuals with MD than RD and the use of medications for anxiety and depression, higher among females than males.

Finally, a cohort study of almost 9,000 Swedish individuals using a national dataset reported that those with reading problems had elevated risks for almost all psychiatric disorders with the exception of anorexia nervosa and criminality, with odds ratios adjusted for IQ ranging from 1.23 for nonviolent criminality, 3.14 for autism and 4.83 for ADHD (Cederlöf et al., 2017). The magnitude of risk was generally higher for developmental disorders with

childhood onset than for disorders with onset in adolescence or adulthood (e.g., bipolar disorder).

In summary, here, as in other areas, the issue of cause versus correlation arises; whereas a classic view of dyslexia was that problems of attention, behavior, and self-esteem were secondary consequences of reading problems, it is now clear that, in some cases, such difficulties reflect separate pre-existing conditions. It is critical to understand the nature and causes of such comorbidities and to gain a better understanding of developmental pathways from poor reading.

### **Educational implications**

Traditionally, different forms of reading problem have been viewed as relatively modular with decoding problems reflecting phonological processing weaknesses and reading comprehension problems reflecting broader oral language deficits, particularly deficits in semantics (vocabulary knowledge) and grammar (Snowling & Hulme, 2012). However, the finding that early, broadly defined, language difficulties are predictive of both later decoding and reading comprehension difficulties strongly suggests a role for early language intervention as a way of preventing both types of reading difficulty. There is now good evidence that language interventions can produce meaningful improvements in children's oral language skills especially when these interventions are well implemented and involve small group teaching (Hulme et al., 2020; Rogde, Hagen, Melby-Lervåg, & Lervåg, 2019). Preschool parent-delivered programmes may also be helpful for ensuring a secure foundation in language skills at school entry (e.g., Burgoyne et al., 2018). An important issue for future studies is to examine the durability of any effects of early language intervention and whether such interventions produce transfer effects to reading accuracy and comprehension.

### *Interventions for reading difficulties*

Space precludes a comprehensive review of reading interventions, however, as might be expected from the theoretical framework presented here, children with dyslexia benefit from teaching that directly targets word-level decoding as well as underlying skills (spelling-sound relationships and phonological skills, see Galuschka et al., 2019; McArthur et al., 2018 for reviews). There is also some evidence that, for children with dyslexia, interventions to improve their decoding problems lead to improvements in reading comprehension (since for these children the principal limiting factor for comprehension is their low level of reading accuracy). Conversely, for poor comprehenders, interventions that improve language skills also improve reading comprehension (Clarke, Snowling, Truelove, & Hulme, 2010; Fricke, Bowyer-Crane, Haley, Hulme, & Snowling, 2013)

although such transfer effects are not always obtained. At a more general level, future research needs to address issues surrounding the implementation of interventions if they are to be successfully embedded in practice (see Foorman, Dombek, & Smith, 2016 for discussion).

### *'Alternative' interventions for reading difficulties*

The approaches to intervention for children's reading difficulties with demonstrated effects grow out of cognitive level theories about the nature and causes of children's reading problems. However, in recent years there has been a large growth in other approaches to intervention for reading and language problems. Among those claimed to improve reading and language skills is working memory training, however, two meta-analyses provide clear evidence against the claim that working memory training can remedy reading problems (Melby-Lervag et al., 2016; Melby-Lervag & Hulme, 2013). Similar negative conclusions have been reached about another form of computerized training called FastForWord, also marketed as a treatment for reading and language difficulties (Strong, Torgerson, Torgerson, & Hulme, 2011). In contrast, it is interventions that tackle 'head on' the proximal cognitive causes of poor reading that are likely to succeed.

### **Conclusions**

The science of reading has flourished during the past decade, and our understanding of the causes of dyslexia and related learning disorders has advanced considerably. A challenge for the field is to convey to education policymakers the implications of this knowledge (Seidenberg, 2013).

It must be acknowledged that, having abandoned the discrepancy definition of 'dyslexia', a broader range of children with persistent reading disorders is being identified. Dyslexia is now recognized as a dimensional disorder, highly comorbid with other learning disorders. Thus, while the methodological tradition of 'controlling' for difficulties in language or attention when selecting samples of children with

dyslexia has provided much important information about its core characteristics, it does not provide a full picture of its etiology. Now our task is to consider how multiple deficits (at the biological and cognitive levels) interact with compensatory resources and educational experiences to produce the profiles of reading disorder that we observe.

A theme which is woven through this review is that oral language skills provide the critical foundation for all aspects of literacy development. We now know that the influence of language skills on learning to decode is mediated via phonological skills. Similarly, there is a division of labor between decoding and language comprehension processes that changes at different rates, according to whether languages are easier or more difficult to learn to decode. Although we know a great deal about how to get poor readers off to a good start in decoding, there is a dearth of evidence regarding how to improve reading fluency and comprehension or spelling, and only limited evidence that early language interventions can improve reading outcomes (Elleman, Lindo, Morphy, & Compton, 2009). Finally, the strong association between oral and written language skills brings to the fore the urgent need to consider the much neglected field of developmental language disorders and the difficulties these children have, not only with reading, writing and mathematics, but with a range of other psychosocial and emotional issues (Bishop et al., 2017). In addition, the timing and frequency of reading interventions for such 'at-risk' groups (prevention vs. remediation) would be a fruitful avenue for future research (Maughan & Barker, 2019; Volkmar, Galushka & Schulte-Korne, 2019).

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### **Key points**

- Language is a critical foundation for learning to read aloud (decode) as well as for reading comprehension.
- Children with language difficulties at school entry are at high risk of reading disorders.
- The etiology of reading disorders is multifactorial.
- Reading disorders are highly comorbid with disorders of mathematics, language, and attention.
- Evidence-based interventions for decoding promote word reading with integrated training in phonological awareness and reading practice using books.
- Evidence-based interventions for reading comprehension are language based, promoting comprehension through vocabulary instruction, work on oral narrative, and reading comprehension strategies.



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