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Dyslexia: 1965–2005

Michael Rutter and Barbara Maughan

Institute of Psychiatry, London, UK

Abstract. William Yule's many contributions to the field of reading disabilities over the last 40 years are reviewed and set in the context of recent research evidence. The value of regression methods in the measurement of reading performance remains valid, but spelling, as well as reading, difficulties need to be assessed in relation to the diagnosis of dyslexia. Although categorical approaches to diagnosis are needed for some purposes, it is likely that the genetic liability to dyslexia is dimensional. Overall, Yule's identification of the key features of specific reading retardation have been confirmed by subsequent research, but the concept of general reading backwardness as a diagnosis has proved less meaningful. The identification of the high rate of comorbidity between reading disability and emotional/behavioural disturbance, highlighted by Yule 35 years ago, has been amply confirmed but the causal mechanisms remain ill-understood.

Keywords: Dyslexia, W. Yule, genetics, comorbidity, measurement, epidemiology.

Introduction

In many respects, Bill Yule's work in the field of reading disabilities is what first gained him an international reputation. Building on his involvement in the Isle of Wight epidemiological surveys of the 1960s, Bill undertook a major programme of research on children's reading problems that attracted widespread attention. His approach then - as throughout his career – was comprehensive: he made major contributions to the literature on the measurement of reading difficulties, studied aetiological factors, and highlighted the high rates of comorbid behavioural and emotional problems shown by children with dyslexia. Equally importantly, his over-riding concern that research findings be put to practical use was also evident in this early work: he evaluated screening methods for the early detection of reading difficulties, compared different approaches to the delivery of remedial teaching, and set up an in-service training programme for teachers.

Since the time of those early studies research has done much to elucidate the cognitive underpinnings of reading, and to highlight the factors that put a significant minority of children at risk of reading failure. Many of Bill's insights have been borne out by this more recent work, while a few have proved more controversial. As a collaborator with him in much of his early research on reading difficulties (MR), and a student of his since that time (BM), we are

Reprint requests to Professor Sir Michael Rutter, Box Number P080, Social, Genetic and Developmental Psychiatry Centre, Institute of Psychiatry, De Crespigny Park, Denmark Hill, London SE5 8AF, UK. E-mail: j.wickham@iop.kcl.ac.uk.

delighted to have this opportunity to review his many contributions to the field of reading disabilities, and to set them in the context of more recent findings.

Measurement

Although the first clinical reports of dyslexia appeared over a century ago, the issue of how best to operationalize the concept has long presented challenges. When Bill entered the field, the dominant definition was exclusionary in approach (Critchley, 1970). Thus, dyslexia was conceptualized as a specific and persistent failure to acquire efficient reading skills despite conventional instruction, adequate intelligence and sociocultural opportunity. Surprisingly, that remains so today (Démonet, Taylor and Chaix, 2004). In his research on measurement issues, Bill focused on the criterion of reading performance that was discrepant with a person's general intelligence. All sorts of methods had been used at that time to operationalize this criterion – ranging from an exclusion of subjects with an IQ below some arbitrary threshold to a variety of subtraction techniques based on the gap between the reading score and some measure of IQ. He noted that all of this was psychometrically unsound (Yule, 1967). There is not a perfect correlation between IQ and reading – most estimates placed it around +0.60. Given that situation, the expectation has to be that when a child's IQ is above average, his or her reading score will also be above average, but not as much so as the IQ. Similarly, if the IQ is below average the reading may be expected to be also below average, but not as much so as the IQ. The standard statistical technique for quantifying such discrepancies is a regression equation. That was so in the 1960s and it is still so today (Campbell and Kenny, 1999). Moreover, it has been found to work in practice (see Rutter et al., 2004).

So why has it not been universally adopted?Undoubtedly, part of the answer lies in the reluctance of medics to adopt a psychometric procedure that obviates the need for clinical judgement and individual diagnosis. In addition, however, there have been some measurement concerns. First, there is a floor effect problem. With very young children there is limited scope for IQ-reading discrepancies simply because the mean level of reading skills at, say, age 6 or 7 years is so low. In principle, there is no reason why a regression approach should be inapplicable but, in practice, discrepancies are likely to be less reliable and less valid than in older age groups. Second, there is a high IQ problem. Unless modified, application of a regression method will mean that a child with, say, an IQ of 140, but whose reading is only just above the population mean of 100, would nonetheless be classified as having a specific reading retardation, to use the terminology Bill suggested. It is not self-evident that it makes sense to classify a reading performance that is above average for a child's age group as "retarded" simply because a child's IQ is so very superior. In the Isle of Wight epidemiological studies (Rutter, Tizard and Whitmore, 1970), in which the regression approach was applied on a wide scale for the first time, it was found that the great majority (76 out of 86) of the children classified as showing a specific reading retardation also had a reading score that was well below average without taking IQ into account. Nevertheless, there were a few children for whom that was not the case. As Bill noted soon after, it might have been preferable to exclude this minority group from the overall sample with specific reading retardation (Rutter and Yule, 1976).

The third methodological issue concerns the choice of IQ test. Should the regression be based on verbal IQ, performance IQ or full scale IQ – if using scales, such as those developed by Wechsler, which made such distinctions?The correlation between verbal IQ and reading is

usually substantially higher than that with performance IQ, so it might seem desirable to use verbal IQ. The problem, however, is that poor reading is likely to impede the development of verbal intelligence, just as poor verbal intelligence will impede the growth of reading skills. With group tests, there is the additional problem that the tests of verbal intelligence rely on reading skills to such an extent that the measures are not satisfactorily separable. Accordingly, for practical rather than conceptual reasons, most researchers have used either performance IQ or full scale IQ.

The fourth methodological problem concerns the choice of population used to produce the regression equation. This problem first arose in the comparison of reading performance in children living in inner London and those living on the Isle of Wight (Berger, Yule and Rutter, 1975). It was evident from the outset that the comparison of the two populations gave somewhat different results if, instead of choosing one standard population, the regression equation was produced for each population separately. If the latter approach were adopted, it would automatically result in a diminution in difference. The key question was what the concept of dyslexia implied. The alternatives are most easily envisaged by considering the parallel of sex differences on some trait. Thus, Zoccolillo (1993) argued that conduct disorder in girls was under-diagnosed because the base rate of antisocial behaviour was so much lower in girls than boys. He suggested that a lower symptom cut-off should be used in girls than in boys. If he was correct, it should follow that the characteristics and course of conduct disorder in girls defined using the lower cut-off should be equivalent to that in boys using the higher cut-off. In this case, extensive tests based on data from the Dunedin longitudinal study showed that they were not equivalent, and that the sex-differentiated variation in cut-off was not justified (Moffitt, Caspi, Rutter and Silva, 2001). The conceptual point is whether there is any reason to suppose that mild deficits in one population should be equivalent to severe deficits in another. The empirical need, however, is to put that possibility to the test. Bill argued implicitly that there was no reason to suppose that mild discrepancies in reading skills on the Isle of Wight should be equivalent to severe discrepancies in London and, hence, he adopted the standard population approach. The empirical findings (Berger et al., 1975) tended to support his decision but the definitive comparison has yet to be made. One key complicating factor with respect to the inner London/Isle of Wight comparison would be the finding that family and school factors were more influential in the London sample. In other words, when comparing cut-off points in the two populations, the differences in the pattern of risk factors would have to be taken into account.

The fifth measurement issue is that the concept of dyslexia, as well as the empirical evidence, suggests that as children grow older, they may improve somewhat in their reading performance (although the great majority continue to have reading difficulties right into adult life) but nevertheless show marked spelling difficulties, which are likely to reflect the same underlying liability (Yule, 1973). Accordingly, it follows that it may well be desirable to include specific spelling difficulties in the criteria for dyslexia. If so, there is every reason to suppose that a similar regression equation method is appropriate.

We conclude that Bill's arguments on the need to use regression methods to operationalize the IQ-discrepancy criterion for dyslexia remain valid. Nevertheless, it is the case that it is not a method that is free of difficulties; it is just superior to its competitors. Before turning to other topics, it is necessary to note several misconceptions. First, some critics have assumed that the method implies a belief that IQ is in some sense "basic". Bill was quite explicit in rejecting that assumption (Yule, Rutter, Berger and Thompson, 1974; Yule and Maughan, 1998). As he

pointed out, the regression concept has got nothing whatever to do with any presumption about what is basic; it is no more than a way of dealing with the relationships between two tests (see also Campbell and Kenny, 1999). It would be just as legitimate to measure specific intellectual retardation using reading as the predictor, or to assess reading in relation to mathematical performance, or vice-versa. It just so happens that, for most purposes, it tends to be more useful to focus on specific reading retardation.

The second misconception relates to the application of cut-points in discrepancy scores to identify children with severe reading difficulties. Commentators have argued that the use of a cut-off both presupposes that a categorical distinction is preferable to a dimensional approach and that the particular cut-off proposed is necessarily the "correct" one. That is no part of the regression method. It could be used dimensionally, as well as categorically, and the cut-off needs to be chosen on the basis of what is needed for a particular purpose. We return to this issue when discussing diagnostic considerations.

The third misconception is that a psychometrically-based definition is equivalent to a clinical diagnosis. That was never suggested. The point of the regression method was only to identify children with marked and specific difficulties in reading. The approach was neutral on whether or not this resulted in the delineation of a homogeneous diagnostic group. If other tests can be used to subdivide children with marked reading difficulties into subgroups, then they should certainly be used for this purpose. There is no doubt that there is considerable variation in the extent to which children with specific reading difficulties show phonological problems, impaired visual processing, attentional difficulties and other co-ordination deficits (see review by Démonet et al., 2004). What is less clear is whether this variation can be used to delineate meaningfully different discrete subgroups. Opinions differ on this point and research findings are rather inconclusive (but see below).

Diagnosis

In the 1960s, the main dispute in the realm of diagnosis was between neurologists who viewed dyslexia as a clear-cut medical condition due entirely to some set of biological causal factors, and educational psychologists who denied the validity of any such concept and instead sought to attribute reading difficulties to environmental inadequacies in the family or school, or to the child's overall intellectual level. Bill rejected both these extreme views and subsequent research has amply confirmed that he was right to do so. The original Isle of Wight survey findings indicated important associations between specific reading retardation and language delay, currently poor complexity of language, and a family history of speech retardation and reading difficulties, but no association with social disadvantage (Rutter, Tizard and Whitmore, 1970). Follow-up at age 14½ years showed a strong tendency for specific reading retardation to persist (Yule, 1973; Rutter and Yule, 1975) and the recent follow-up into adult life showed a very high degree of persistence with respect to serious spelling problems and also broader aspects of educational achievement. There is no doubt that specific reading retardation indexes a serious impairment that persists into adult life.

Genetics

Quantitative genetic findings in relation to dyslexia have shown substantial heritability and the family data indicate that the inheritance is likely to be multifactorial and not based on a

single gene transmitted in Mendelian fashion. Molecular genetic findings pointed to a locus on chromosome 15 as long ago as 1983 (Smith, Kimberline, Pennington and Lubs, 1983) and there are replicated findings on both this chromosome and chromosome 6, with pointers to possible loci on other chromosomes. No specific susceptibility gene, however, has been certainly identified so far, although there is one very promising report awaiting replication of a quantitative trait locus haplotype (meaning a set of genes occurring near to one another that tend to be inherited together) on chromosome 6 (Francks et al., 2004). The genetic effect was on a broad range of reading-related cognitive abilities (particularly in the severe range) but there was no impact on overall IQ. With respect to the concept of specific reading retardation, it is relevant that the heritability of reading disability is greater in children of higher IQ compared with those of lower IQ (Wadsworth, Knopik and Defries, 2000) and that the linkage data are also stronger (Knopik et al., 2002). The implication is, as Bill argued, that the aetiology of specific reading retardation is not synonymous with the aetiology of general cognitive impairment. It is noteworthy, as we have indicated, that the loci for susceptibility genes for dyslexia do not serve as loci for genes related to overall IQ, although they do overlap with those for specific language impairment (SLI Consortium, 2004), as would be expected in terms of the original concept of specific reading retardation.

Plomin and Kovas (in press) have argued that there is likely to be a shared genetic liability for general cognitive impairment and specific cognitive disabilities such as dyslexia. Given the lack of confirmed susceptibility genes for either overall IQ or dyslexia, it is not possible to confirm or refute this hypothesis in any definitive fashion at this stage. Nevertheless, it is important to be clear on the issues involved. Given the substantial intercorrelations among specific and general cognitive skills, there is almost bound to be a shared genetic liability in so far as overall IQ, or general cognitive ability, partially underlies reading skills. The key question, however, is whether what is specific about dyslexia involves some different genetic influence or, rather, whether the genetic liability is entirely general with the specific aspects of the skills (or deficits) due to some as yet unspecified environmental factor. The key data needed to answer the question are not yet available but the findings so far favour specific genetic factors.

That is not to say, however, that the genes operate in relation to a specific, extreme diagnostic category or that the genetic liability applies only to reading skills. As already noted, spelling disabilities are strongly associated with dyslexia and it is quite common for the reading impairment to extend more broadly to include other specific cognitive dysfunctions. At first sight, that might seem to undermine the concept of a specific reading retardation but it does not necessarily do so. First, as shown by all studies including the Isle of Wight survey (Rutter et al., 1970), and subsequent investigations (Ramus et al., 2003) children identified on the basis of severe reading difficulties that are discrepant with their overall IQ often have some impairments, of a lesser degree, in other skills. The key comparison is between the findings on specific reading retardation and those on a comparably identified mathematical or motor disability. The relevant findings are sparse, but they show substantial differences in numerous features (Snowling, 2002). There are major meaningful differences between different types of deficit in specific scholastic skills, but there is some overlap. Such overlap could derive from common causal factors or the consequences for the acquisition of other scholastic skills from serious reading difficulties (because reading is basic for so much school learning), or from the effects of any specific disability on how schools provide education – or a mixture of these.

The second type of evidence derives from twin and family studies, in which the findings suggest, first, that the deficit is dimensional rather than categorical and, second, that the deficits in co-twins or first degree relatives include a broader range of cognitive deficits than reading alone, although the reading impairment is the most marked and consistent feature (Snowling, Gallagher and Frith, 2003; Lyytinen et al., 2004). Thus, Snowling et al. (2003) compared 56 children who had a first degree relative with dyslexia and 29 controls, with assessments at 3 years 9 months, 6 years and 8 years. Two-thirds of the familial risk group showed literacy problems compared with 1 in 6 controls. The key precursor of dyslexia was a pattern of general language delay, followed by difficulties in phonological awareness at age 6 years. The children in the familial risk group without dyslexia at 8 nevertheless scored below controls on tests of grapheme-phoneme knowledge. It is this last finding that suggests that dyslexia is dimensional rather than categorical.

The results of the Jyväskylä Longitudinal Study (JLS) give rise to the same conclusion (Lyytinen et al., 2004). One hundred and seven children at familial risk for dyslexia were compared with 93 controls, with a follow-up from birth to age 9 years (with intermediate assessments at 1, 2, 3 and 5 years). The children without dyslexia in the familial risk group nevertheless had decoding skills that were worse than those of controls, suggesting a dimensional deficit. It is also notable, however, that the language and phonological measures in the familial risk group accounted for a much greater proportion of the initial reading skills than did the same measures in the control group. The implication is that the phonological and language measures were identifying risk factors for dyslexia that were associated with familiality (and hence probably with genetic risk), but that the risks were nevertheless dimensional rather than categorical.

The pattern of findings in the Snowling et al. (2003) study also suggested that strong language skills may serve to protect against phonological difficulties. The findings, in many respects, mirror those found in individuals with specific reading retardation. The key point, however, is that the findings refer to the relatives of an individual with dyslexia. Given that familial liability, the implication is that the deficit is only moderately specific. What the findings do not mean is that the same would apply in the general population or to the families identified on the bases of a member with, say, an arithmetical deficit or mild general retardation. Unfortunately, the much needed direct comparative studies have yet to be undertaken (see Snowling, 2002).

The parallels with specific language impairment are close. Thus, Bishop and colleagues (Bishop, North and Donlan, 1995; Bishop, 2001) found that the monozygotic (MZ) co-twins of children with a severe specific developmental language disorder had a much greater concordance for this disorder than did dizygotic (DZ) co-twins (indicating a substantial heritability), but the concordance with a broader range of specific and general language impairment was much greater (again more so in MZ than DZ pairs). The genetic liability was clearly concerned with language but it extended more widely and did not conform to the traditional diagnostic categories. Much the same has been found with autism (Folstein and Rutter, 1977; Bailey, Palferman, Heavey and Le Couteur, 1998). It seems that it is likely that this may be a general feature of genetic influences, at least with respect to neurodevelopmental disorders.

Specific reading retardation and general reading backwardness

The Isle of Wight study findings indicated several possibly important differences between specific reading retardation (i.e. defined in terms of a discrepancy with overall IQ) and general

reading backwardness (i.e. defined in terms of a deficit in absolute terms without reference to IQ). Thus, general reading backwardness after excluding those who also showed specific reading retardation showed a lesser male excess, was more likely to be associated with an overt, neurological disorder, and was usually accompanied by a wider range of neurodevelopmental impairments (Rutter and Yule, 1975). It was also more often associated with large family size and social disadvantage. Rispens (1998) brought together the findings of more recent comparisons of general reading backwardness and specific reading retardation defined in this way. He concluded that, on the whole, the findings show that the specific reading retardation group has more serious reading problems but that the groups do not differ in either their pattern of use of reading strategies or in terms of the presence of deficits in phonology. The follow-up of the Isle of Wight sample in adolescence showed that children with general reading backwardness had better spelling and worse arithmetic than those with specific reading retardation. They also made slightly better progress in reading. A parallel follow-up of the inner London sample (who were much more socially disadvantaged) however, did not find the same group difference in educational progress (Maughan, Hagell, Rutter and Yule, 1994). The two groups, nevertheless, have tended to differ in sex ratio and in the presence of neurological disorder, as found in the Isle of Wight study. As Rispens (1998) pointed out, most of the attempted replications suffered from a failure to exclude the overlap group. From the outset, Bill was explicit that the general reading backwardness group was both heterogeneous and of limited interest in relation to understanding causes of reading failure. Thus, the original Isle of Wight report (Rutter et al., 1970) noted that the group of backward readers had no distinctive characteristics of its own. For the most part, those backward readers who did not meet the criteria for intellectual retardation or specific reading retardation had narrowly missed being included in one or other of those categories; that is their IQ or reading level discrepancy was only just above the cut-off points used.

The findings with respect to specific reading retardation have generally been confirmed by other research. For example, despite a claim to the contrary by Shaywitz, Shaywitz, Fletcher and Escobar (1990), the male preponderance has been evident in all subsequent major epidemiological studies (Rutter et al., 2004). Also, the importance of language-related deficits has been a very consistent finding. The most satisfactory study was that undertaken by Ramus et al. (2003). They focused on university students with dyslexia in order to minimize the likelihood that the individuals involved would have other handicaps. Sixteen students with dyslexia were systematically compared with 16 controls on a range of cognitive tests. All 16 dyslexics showed a phonological deficit, 10 had some kind of auditory processing deficit (albeit not all of the same kind), 4 had a motor deficit, and only 2 had a visual magnocellular deficit. The detailed findings suggested that the phonological deficit was the basic causal factor, although it was not clear just why some individuals also had other deficits.

It may be concluded that the key features of specific reading retardation highlighted by Bill in the Isle of Wight study have been confirmed by much subsequent research. As Bill emphasized at the time, the general reading backwardness group was both more heterogeneous and less meaningful in diagnostic terms. It constitutes a varying mixture of features that characterize overall intellectual impairment and those more characteristic of specific reading retardation. Children with reading problems of this kind undoubtedly need educational attention but the group is not particularly useful for the study of aetiology.

The supposed “hump" in the distribution

One of the findings from both the Isle of Wight and inner London surveys was the apparent “hump" at the bottom of the distribution of specific discrepancies between reading and IQ (Yule et al., 1974). This was used to argue for a separateness between specific reading retardation and the broad continuity in the spread of reading skills in the general population. Something similar was found in two other studies (Dobbins and Tafa, 1992; Stevenson, 1988) but bimodality was not found in several other surveys (see Rispens, 1998). Critics pointed out that the hump could be an artefact deriving from ceiling effects in the tests used (Van der Wissel and Zegers, 1985). Clearly, that is possible, although it is still not clear whether this fully accounted for the “hump" found. A more fundamental concern, however, is that for a variety of statistical reasons it is very hazardous to infer heterogeneity or homogeneity on the basis of unimodality or bimodality in curves (Everitt, 1981). We conclude that major uncertainties remain on the reality of any kind of “hump" at the bottom of the curve but that its presence or absence is not particularly informative on the validity of the dyslexia concept.

Neurobiological findings

It is obvious that neurobiological studies should be helpful in testing the validity of the diagnostic concept of dyslexia. A few post-mortem studies have shown apparently distinctive features in the brains of individuals thought to have had dyslexia (see review by Démonet et al., 2004). Unfortunately, it has to be said that, for a variety of methodological reasons, the findings are inconsistent and inconclusive. The findings are compatible with notions of abnormal brain connectivity in dyslexia, but they do not unequivocally show this. There is a much broader range of brain imaging data with both children and adults, using event-related potential (ERP) studies, as well as positron emission tomography (PET) and both structural and functional magnetic resonance imaging (MRI). Most studies have shown differences between individuals with dyslexia and controls, but there are too many contradictions among studies for any firm conclusions. Discrepant findings are clearly due in part to age differences and the development of compensatory strategies, as well as differences in the task demands in different studies. Overall, the conclusion would seem to be that the ways in which individuals with dyslexia process reading-related stimuli differ from controls but the meaning of these differences in terms of the possible neural basis of dyslexia remains to be elucidated. Taken as a whole, the findings are supportive of the diagnostic concept, but not decisively so.

An important study by Paulesu et al. (2001) used a combination of cognitive tests and brain imaging to determine whether the findings were similar in the case of individuals with dyslexia in languages with differing orthographies. By comparison with English, for example, where there are numerous irregularities in the associations between sounds and written language, languages like Italian have highly regular letter-sound correspondences. It was found that dyslexia was about half as common in Italian speakers compared with French or English speakers, but that both the phonological deficit and the imaging findings were strikingly similar. The findings generally point to a comparable deficit in dyslexia even in languages whose simpler orthography makes the condition less commonly manifest in the form of serious persisting reading difficulties.

An important advance has been the use of functional brain imaging to examine the effects of remedial intervention (see review by Démonet et al., 2004). The findings have shown

significant brain changes in response to remediation but, at least so far, the results are not of a kind that are directly helpful with respect to the diagnostic concept.

School and family influences

One of the key reasons that Bill rejected the narrow determinative neurological view of dyslexia was the evidence from the inner London and Isle of Wight studies that both family and school environmental risk factors were associated with differences in the rate of specific reading retardation (Berger et al., 1975; Rutter et al., 1974; Rutter and Quinton, 1977). Thus, for example, specific reading retardation was twice as common in inner London as on the Isle of Wight and this difference did not appear to be a consequence of either differences in overall IQ or differential in- and out-migration. Of course, the design could not take account of possible genetic mediation but it does not seem likely that that could account for the school effects. The later study of secondary schools in inner London (Rutter, Maughan, Mortimore and Ouston, 1979) confirmed that schools differed in the rate of reading and other scholastic difficulties even after taking into account variations in the intake to the schools. There has been surprisingly little systematic study in recent times into the possible school and family influences on reading performance in individuals with dyslexia, apart from the important study by Lyytinen et al. (2004). In their prospective longitudinal study they found that the familial risk and control groups did not differ in their exposure to shared reading at home or their access to reading materials, but (not surprisingly) the parents in the familial risk group were less active readers themselves than the control group parents. Preliminary analyses suggest that children's higher interest in reading and more frequent parent-child shared reading were protective factors in the familial risk group but were less influential in the control group

Recent data suggest a marked social gradient in IQ-discrepant reading problems (Carroll, Maughan, Goodman and Meltzer, 2005), and school experiences can undoubtedly be influential. Classroom studies have shown, for example, that low and high ability reading groups often provide very different contexts for learning: skilled readers benefit from each others' successes in oral reading, while their less skilled counterparts often face disruptions in lessons (so reducing the time spent on reading), and are more likely to be exposed to reading errors by their peers. This is just one example of what have been described as "Matthew effects" in the classroom - rich-get-richer and poor-get-poorer mechanisms whereby the difficulties of struggling readers, and the successes of their more skilled peers, are amplified by classroom experiences. Bill's view that dyslexia was likely to prove to be a multifactorial disorder with probable genetic influences but also susceptibility to environmental factors has not been investigated adequately up to now but it may be anticipated that it will prove to be correct.

Obstetric factors

The Isle of Wight study findings (Rutter et al., 1970) showed only a nonsignificant tendency for children with specific reading retardation to have a low birth weight or a reduced period of gestation (and no tendency for increased birth complications). By contrast, the children with intellectual impairment were much more likely than controls to be small for dates or have a birthweight below 5½ pounds. More recent research following up children of very low birth weight prospectively has confirmed the frequency of both neurological and intellectual disability (Marlow, 2004; Marlow et al., 2005). Specific reading retardation was not studied as

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such but it was noted that most of the learning difficulties seemed to be related to overall low IQ. The research evidence as a whole indicates that it is unlikely that pregnancy complications of any kind play a major causal role in the genesis of specific reading retardation.

Conclusions on diagnostic validity

The weight of evidence clearly supports Bill's view that specific reading retardation (dyslexia) is a meaningfully distinct syndrome but, equally, it supports his concept that it is a multifactorial disorder with both genetic and environmental factors influential in aetiology.

Remediation

Throughout his career, Bill has been committed to the need to ensure that research findings improve policy and practice, but equally to the need to evaluate interventions in a systematic fashion. His work on the Isle of Wight exemplified both. The epidemiological findings emphasized both the high frequency of specific reading retardation and its high persistence over time. The challenge was what ordinary schools could do to provide remedies. The local education authority on the Isle of Wight responded to the survey findings by appointing one remedial teacher. Of course, that constituted a very limited resource but Bill used this limitation to advantage by setting up a pilot study (Yule and Rigley, 1968; Rutter et al., 1970). The eight island secondary schools were divided into four groups. In two schools, the remedial teacher himself taught the children with specific reading retardation in small groups of 3 or 4; in two he advised their regular teachers; in two the children were placed in remedial streams; and in two no special provision was made. The rate of reading progress over one year for these 13 to 14-year-old children was slightly better in the group given personal teaching by the remedial teacher and was worst in the group where the children's regular teachers had been advised. Bill went on to set up a short in-service training course for teachers of 8-year-old children. This was offered only to those teaching in one part of the Island and the reading progress of the children they taught was compared with that of children in control classes, the groups being matched on the basis of their reading scores at the start of the experiment. At the end of the year, the children of the in-service trained teachers were reading at a level nearly 2 months ahead of those in control classes.

In writing up these two small pilot studies, Bill emphasized that replication was needed, and that studies of more specific interventions would also be required. In the meanwhile, the findings offered some modest encouragement that benefits were possible but suggested that something more than remedial classes was likely to be necessary. The model that Bill provided for the application of epidemiology to practice and for the use of experimental methods in ordinary schools remains as valid today as it was 40 years ago. There is now rather more evidence on the value of interventions for children with reading difficulties (see Snowling, 2002; Troia, 1999) but there are still relatively few randomized controlled trials on epidemiological samples. The weight of evidence indicates the value of phonological methods (Ehri, Nunes, Stahl and Willows, 2001; Hatcher, Hulme and Snowling, 2004) but we still lack knowledge on the degree to which remedial methods need to be based on each child's specific pattern of cognitive strengths and deficits (see Xue and Meisels, 2004 for interesting findings from a very large scale study). Also, there is limited evidence on the extent to which short-term gains following treatment, particularly for those children with severe specific reading

retardation, translate into longer-term gains in real life reading skills (Blachman et al., 2004; Yule, 1976; Yule and Rutter, 1985).

Reading difficulties and emotional and behavioural disturbance

The Isle of Wight study emphasized the high frequency with which reading difficulties were associated with antisocial behaviour in childhood (Rutter et al., 1970). As always, Bill was insistent on the need to determine the causal mechanisms involved. The finding that the children with *both* reading difficulties and antisocial behaviour were more like those with "pure" reading difficulties than those with "pure" conduct problems suggested that the antisocial problems may have arisen, in part, as a result of educational failure. Rather than conclude that this was necessarily the complete explanation, Bill remained alert to the necessity of considering alternative mechanisms and other research evidence (Yule and Rutter, 1985). He concluded that no single explanation was satisfactory. School failure probably did have some causal role in the genesis of antisocial behaviour but, in part, temperamental characteristics such as overactivity and certain family influences were likely to predispose to both. Subsequent findings have amply borne this out. Numerous studies have now shown, for example, that developmental reading problems have strong overlaps with inattentiveness (Hinshaw, 1992). In part, these associations are genetically mediated, and may form one pathway to conduct problems in childhood; in adolescence, disaffection from school may add to poor readers' risks of involvement in antisocial behaviour. Adult follow-up of the inner London sample, however, showed no excess of criminality or aggressive behaviour among men with childhood histories of reading problems (Maughan, Pickles, Rutter, Hagell and Yule, 1996), suggesting that positive engagement in adult roles can mitigate earlier risks. By contrast with the extensive literatures on reading difficulties and disruptive behaviour problems, much less attention has been given to possible overlaps with emotional difficulties. Yet it is widely recognized that children with reading problems are vulnerable to low academic self-esteem, and Bill argued that this should be tackled simultaneously with the teaching of reading skills (Yule and Maughan, 1994). More recent evidence suggests that children and adolescents with reading difficulties may be at increased risk of low mood (Maughan, Rowe, Loeber and Stouthamer-Loeber, 2003), and that for small groups of poor readers anxiety disorders may also form part of the clinical picture (Carroll et al., 2005). As Bill stressed, whenever children present with reading difficulties it is crucial to undertake a comprehensive evaluation of all aspects of their functioning, so that associated difficulties of this kind can be detected and treated appropriately.

Overview

As this brief overview suggests, Bill's work on reading disabilities laid the foundations for much recent research in the field. Then, as now, his concern with measurement issues remains fundamental, and his view that developmental reading problems reflect multifactorial causation is consonant with the ever-expanding volume of aetiological research. But as much as the specifics of his findings, his research on reading is important in exemplifying his characteristic approach to the study of children's difficulties. Throughout his career, Bill's work has been marked by a blend of methodological rigour and practical application, and by a concern for

science in the service of children's needs. It has been our pleasure to collaborate with him in this enterprise.

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