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DEVELOPMENTAL AND ACQUIRED DYSLEXIAS

Christine M. Temple

(Developmental Neuropsychology Unit, Department of Psychology, University of Essex, Wivenhoe, UK)

ABSTRACT

Marshall (1984) highlighted potential parallels between children with developmental disorders of reading and adults who had acquired reading disorders. He advocated the use of a cognitive neuropsychological framework in the investigation of children with developmental abnormalities of cognition, including those with developmental dyslexias.

Developmental phonological dyslexia has been extensively described and is a pervasive disorder. The relationship between reading difficulty and phonological difficulties evident in explicit oral phonological tasks continues to be a focus for debate. Clear cases of developmental deep dyslexia have now been described and the syndrome has also been described as characterising early reading development in Williams syndrome (WS), where there are also semantic errors in other domains, including naming and receptive vocabulary and there may be a generalised difficulty with the activation of fine grain semantic specifications. In the domain of number, highly selective reading disorders characterised by high rates of semantic errors have been documented, indicating that semantic reading errors can be domain-specific. They can occur to number words despite intact ability to read Arabic numbers and they can occur to Arabic numbers and number words despite intact ability to read words in other domains. Current models of reading written words do not allow for such material-specific dissociation. Developmental surface dyslexia has also been described in a range of countries, languages and orthographies. Descriptions of cases for whom there is no phonological impairment in reading have generated contrary evidence for theories suggesting that phonological impairment underlies all developmental dyslexia. As reading develops in Williams Syndrome, phonological reading skills may improve with over-reliance on these leading to surface dyslexia. Surface dyslexia has also been reported in cases of developmental amnesia in which there are semantic memory impairments. Hyperlexia can take several forms including broad hyperdevelopment with elevated phonological reading abilities, lexico-semantic reading abilities and reading comprehension as in Turner’s syndrome (TS). This advantage has early onset in school-starters.

These specific modular effects do not have pervasive impact across systems but demonstrate the limitations of functional plasticity in developmental and genetic disorders. The framework Marshall (1984) outlined has provided a foundation for the development of systematic investigation of developmental disorders.

Key words: dyslexia, reading, amnesia, semantic representation, number reading

In their seminal papers, Marshall and Newcombe (1966, 1973) described acquired deep dyslexia and acquired surface dyslexia in adults who has previously read well but who had sustained gun shot and shrapnel injuries to the brain. The contrast between the skills in the two disorders argued for distinct modular components within the reading system that could be dissociated following injury or disease with selective components of a functional architecture impaired but leaving residual components intact. More specifically, the cases produced strong evidence in favour of distinct reading mechanisms one of which reflected non-lexical phonological processes involved in translating graphemes or letter clusters to phonemes or phonological segments, and blending these to produce an integrated output (Coltheart et al., 2001; Newcombe and Marshall, 1981; Shallice et al., 1983). In contrast, lexico-semantic and direct reading routes enabled recognition of a whole word and thereby activation of its meaning and then pronunciation or its pronunciation directly. Description of phonological dyslexia (Beauvois and Derouesne, 1979) and direct dyslexia (Schwartz et al., 1980) strengthened the evidence in favour of these distinctions.

These clearly delineated acquired reading disorders and their subsequent theoretical interpretation raised issues concerning the development of reading. Specifically, if the adult system included parallel reading routes with modular sub-components how did these become established and were the distinctions specified early in development? If a template for a comparable functional architecture unfolded during development it raised the possibility that within the developmental dyslexias one might see different forms of disorder reflecting disturbance of distinct components of the reading system and that some of these developmental dyslexias might even have a form parallel to that of the acquired disorders. Marshall (1984) discussed a potential taxonomy for the developmental dyslexias based on a rational, psycholinguistic analysis of reading skill utilizing a case study methodology, which would avoid the averaging out of distinctive patterns of performance of theoretical significance. Marshall (1984) discussed four distinct developmental dyslexias: phonological dyslexia, surface dyslexia, deep dyslexia and hyperlexia. The former three were proposed as parallels to the adult dyslexias of the same name and the latter was proposed as a
parallel to direct dyslexia (Schwartz et al., 1980). This paper discusses studies which have developed in the exploration of each of these four different forms of developmental dyslexia. It highlights the contribution which Marshall’s (1984) ideas have made to the emergence of a number of contemporary studies and developments within developmental neuropsychology. The paper draws together results from a range of studies which are relevant both to issues of the extent to which there is modularity within the development of reading systems and also more broadly to issues of modularity with the development of cognitive systems. It addresses the limitations of functional plasticity within cognitive development but highlights the case for residual normality within developmental disorders, such as the developmental dyslexias.

**DEVELOPMENTAL DYSLEXIAS**

*Developmental Phonological Dyslexia*

Of the developmental dyslexias, discussed by Marshall (1984), the one which has received the most detailed subsequent investigation is that of phonological dyslexia. Developmental phonological dyslexia was first described by Temple and Marshall (1983) with subsequent descriptions extensively reported in English and then in a broad range of languages and orthographies (e.g., Masterson et al., 1995; Snowling et al., 1986; Temple, 1984a, 1985, 1988a, 1990a). The syndrome has also been described as an acquired disorder in childhood (Pitchford and Funnell, 1999).

Developmental phonological dyslexia is characterised by disproportionate difficulty in the reading of non-words and new unfamiliar words, interpreted as reflecting impaired development of a phonological reading route. The children have also been found to have disproportionate difficulty with the grammatical markings comprising morphological endings of words, with a tendency like their adult parallels to drop or substitute these endings thereby generating morphological errors, evident in single word reading but also pervasive in distorting the flow of textual reading (e.g., Temple, 1984a, 1988a).

Many recent investigations into this disorder and the developmental dyslexias more broadly have focused on the issue of whether there is an impairment in phonological awareness in purely oral language skills which underlies the impaired development of phonological reading skills (Castles and Coltheart, 2004; Masterson et al., 1995). In support of very early abnormality in phonological awareness in some children who subsequently develop reading disorders, Molfese (2000) has demonstrated that auditory evoked potentials at birth to speech and non-speech syllables discriminated those who 8 years later would become dyslexic. Nevertheless, electrophysiological studies have also provided support for distinctive differences in neurobiological performance in different groups of children with dyslexia. For example, specific EEG abnormalities have been reported, which have greater focus in temporal-parietal areas in some cases and in frontal areas in other cases (e.g., Duffy and McAnulty, 1985).

Cossu et al. (1993) argued that phonological awareness is not a necessary prerequisite for reading development, since children who fail tests of phonological awareness, may nevertheless develop reading skills, though subsequent debate has addressed the normality of these reading skills. There is also dispute as to whether impaired phonological awareness, if relevant, causes or is a consequence of dyslexia. Hatcher et al. (1994) reported that remediation, involving training in phonological awareness, only generalised to a significant improvement in reading when training in reading had also been involved. In an extensive review, Castles and Coltheart (2004) conclude that there is no definitive evidence that phonemic awareness training alone improves reading and that studies arguing that they are able to demonstrate such an effect, all have the potential to be explained in another way. In contrast, the positive benefits on reading of teaching letter-sound correspondences themselves were overwhelming.

The current paper will focus discussion less on phonological dyslexia and more on three other dyslexias and related work which have been studied less intensively but which have each been the focus of recent developments: deep dyslexia; surface dyslexia; and hyperlexia. In each case the nature of the issues, the analyses employed and the question raised are ultimately derivative from the foundations laid down in Marshall (1984).

*Developmental Deep Dyslexia*

Developmental deep dyslexia is characterised by an inability to read non-words, semantic errors in single word presentation, imageability effects, such that more highly imageable words are read more easily than low imageability words, and frequency effects such that words of high frequency are read more easily than those of low frequency (Johnston, 1983; Stuart and Howard, 1995; Temple, 1988b). In all these characteristics, the syndrome parallels that of acquired deep dyslexia. Developmental deep dyslexia differs from acquired deep dyslexia in that function words are often read relatively well in the developmental form of the condition, being amongst the small set of items which the children are able to read correctly (Stuart and Howard, 1995; Temple, 1988b, 2003). In acquired deep dyslexia, function word reading is compromised (Marshall and Newcombe, 1966), an effect which had been
attributed to the necessary involvement of a phonological reading route in function words reading and the reading of morphological endings (Patterson, 1982). The relatively competent reading of function words in developmental deep dyslexia despite severe impairment of the developing phonological reading route, places this interpretation in doubt and is more consistent with the original proposal of Caramazza et al. (1983) that there may be a distinct system dedicated to function word processing which happens to be impaired additionally in deep dyslexia and in many, though not all, cases of phonological dyslexia.

The syndrome of developmental deep dyslexia has been thought to be relatively rare, however, recently, it has been described in the early reading development of children with Williams Syndrome (WS) (Temple, 2003). WS is a genetic disorder in which there is microdeletion on the long arm of chromosome 7 at 7q11.23 which affects one allele of the elastin gene and other contiguous genes (Ewart et al., 1993; Frangiskakis et al., 1996; Tassabehji et al., 1996). The incidence is 1 in 25,000. There is a characteristic facial appearance. The syndrome is characterised by learning difficulties including a spatial disorder. The issue of language development has been a focus for recent discussion with evidence for selective impairment in components of morphological and grammatical function (Clahsen and Almazan, 1998; Clahsen and Temple, 2003; Clahsen et al., 2004; Temple and Clahsen, 2002) with specific problems with activation of lexically based instantiations of specific exemplars but relative ease with generalised rule applications, though others claim that there is no residual normality and the entire language systems is specified differently from normal (Thomas and Karmiloff-Smith, 2002). There have been a limited number of studies of reading development (e.g., Laing, 2002; Laing et al., 2001; Menghini et al., 2004) with the report of deep dyslexia linked to the early stages of development (Temple, 2003).

**Developmental Deep Dyslexia in Williams Syndrome**

The case described by Temple (2003) was that of a girl with WS called Emily. Emily was a 13 year old girl with a mental age of 5;4. She was tested on a series of reading tasks addressing letter reading, non-word reading, single word reading and text reading skills.

Emily had a Schonell reading age of 5;1. She was unable to read any non-words though she could name 18 of the 26 letters of the alphabet and sound out 16 of them. Presented with 361 words to read aloud, she refused 13 items, read 25 correctly and made 320 overt paralexias. The words read aloud correctly included nouns (egg, cat, mummy, people), colour names (red, blue), numbers (two, four) and function words (the, and, is, it, you, we, me, to, for, no). Of the overt paralexias, 51 were semantic paralexias, a rate of (15.9%), classified on the basis of common semantic features (e.g., gentleman → ‘Richard’; blue → ‘red’) or a direct semantic association (e.g., cup → ‘saucer’; chimney → ‘house’; eyes → ‘frog’). Emily also had a tendency to make specific semantic error responses to the written names of living creatures (e.g., fish → ‘people’; dog → ‘people’; frog → ‘people’; mouse → ‘people’; queen → ‘people’; daddy → ‘people’; chicken → ‘mum’; cow → ‘mum’; tiger → ‘mum’). Emily had a visuo-semantic error rate of 5.9% (e.g., sun → ‘fun’; tree → ‘teak’; sheep → ‘people’) and a visual + semantic error rate of 5.3% [e.g., floor → ‘stupid’ (via fool); men → ‘five’ (via ten); glass → ‘everyone’ (via class)].

In order to demonstrate that the semantic error rate was higher than that expected by random pairing, Temple (2003) randomly paired all of the responses which Emily made on the reading task with all of the stimuli which had been presented and then rescored these randomly paired stimuli and responses. The random pairing generated error rates for semantic errors of 5.3%, for visuo-semantic errors of 1.6% and for visual + semantic errors of .6%. Thus, for controls there is a random semantic error rate of 5.3% and a rate for errors with a semantic component of 7.5%, whilst for Emily there is a semantic error rate of 15.9% and a rate for error with a semantic component of 27.2%. Emily makes significantly more semantic errors than are seen in the random pairings ($\chi^2 = 17.91, p < .001$) and also makes significantly more errors with a semantic component than are seen in the random pairings ($\chi^2 = 41.91, p < .001$). Emily also makes significantly more semantic errors than the young normal children reported by Seymour and Elder (1986) who were acquiring reading within an educational regime that emphasized whole word approaches and minimized phonological reading skills and for whom one might thereby expect to see a maximal normal semantic errors rate, however, in Seymour and Elder’s (1986) sample the mean semantic error rate was under 1.85% and the mean rate of visual + semantic errors was less than .52%.

**Conclusions**

For Emily, a case of WS, early reading was characterised by failure to read nonwords and a significantly elevated semantic error rate in reading words aloud. She had developmental deep dyslexia. Temple’s (2003) interpretation for this, as well as other phenomena in WS, was that there was failure within the reading system in the activation of the detailed semantic knowledge about words required for accuracy and failure to activate the fine grain semantic features.
The idea that the reading impairment characterised by semantic errors arises partly because of failure to activate fine grain semantic specification is supported by the naming performance of children with WS reported by Temple et al. (2002). Normal children make naming errors which usually indicate a relationship between the target and the response. Thus, for example, employing the line drawing pictures of Snodgrass and Vanderwart (S&W) (1980) normal young children sometimes might name the picture of an eagle as a “parrot”, the jacket as a “coat”, the light as a “lamp”. In each case, the response is incorrect but shares many semantic features with the target and is in this sense close to the correct response. For the children with WS whilst their naming errors are not random, the relationship between the target and the response in terms of shared semantic features, can be much looser. Emily named the picture of a piper as an “antelope” and the peacock as a “tortoise”. Another child with WS with a slightly higher mental age, named a picture of a lobster as an “ostrich” and a spanner as a “corkscrew”.

Error Analysis for Naming

In order to appraise this more formally, Temple et al. (2002) rated the naming errors of children with WS and normal mental age matched control children for their typicality. Any error that was made by more than one in 20 of the controls or the normal young sample described in detail by Cycowicz et al. (1997) was classified as a typical error. All other errors were classified as atypical. The error responses from the control cases and the WS cases were then all rated individually as being either typical or atypical. The results indicated that the children with WS made significantly more atypical errors than the normal controls. This was true regardless of the semantic class of the stimulus item with items from the categories foods, clothes, animals and indoor objects employed. This naming data supported the theory of inadequate activation of semantic specification in WS.

Semantic Knowledge of Vocabulary in WS

Further evidence comes from receptive vocabulary assessment. On the British Picture Vocabulary Scale (BPVS) (Dunn et al., 1997) and the Peabody Picture Vocabulary Test (Dunn and Dunn, 1997), children with WS have been reported in a number of studies to score more highly than would normally be predicted on the bases of age and IQ (e.g., Bellugi et al., 1990; Tyler et al., 1997). This has contributed to the view that children with WS have greater knowledge of vocabulary than would be expected on the basis of age and IQ. Temple et al. (2002) demonstrated that this is a simplification. Whilst they replicated the finding that children with WS scored above mental age level on the BPVS, they also demonstrated that differing results emerged if the distracters in the task bore a closer semantic relationship to the target than that seen in the BPVS. They constructed grids of 24 items in the same semantic class of which one was the target and the other were distracters. Thus, in order for the children to answer correctly they had to distinguish the target not from 3 pictures some of which were relatively unrelated to the target as in the BPVS but from 23 close semantic neighbours such that detailed semantic knowledge was required. In this task, children with WS performed significantly more poorly than mental age matched controls. Temple et al. (2002) argued that this supported the hypothesis of inadequate activation of fine grain semantic knowledge in WS.

One difficulty with this interpretation was that in the construction of the grids, the relationship of the distracters to the target became much closer semantically but the number of distracters also increased. Thus two variables, semantic relationship and number of distracter items, were confounded. Given the spatial difficulties of those with WS, one possible alternative explanation for the phenomena reported by Temple et al. (2002) was that it was the number of distracter items that was the variable of importance rather than the relationship between the target and the distracters. A further study was therefore conducted within which these two variables were dissociated. The results of this study are reported in Temple and Sherwood (submitted for publication, a).

A new sample of 8 children with WS was employed by Temple and Sherwood (submitted for publication, a). They had a mean mental age of 6.7 and a mental age range of 5.9-7.9. The control sample was comprised of 12 normal children with an average age of 6.5 and an age range of 5.6-7.4. Two sets of grids were constructed. In the first set, the distracters were taken from pictures employed in the BPVS and they had a loose or nil semantic relationship to the target item. In the second set, the distracters were all S&W items and had a close semantic relationship to the target item. The grids sets varied in size. Some had 8 items. Some had 12 items and some had 24 items.

The results of this study are reported in Table I. Given the a priori prediction of the direction of the differences, analyses were one-tailed. With the grids with BPVS distracters, which had loose or nil semantic relationship to the target, the children with WS performed significantly better than their mental age controls. For the grids with S&W distracters, which had a close semantic relationship to the target the children with WS performed significantly more poorly than their mental age controls. As
Temple and Sherwood (submitted for publication, a) highlight this effect cannot be attributed simply to some peculiarity of low mental age, as children with Downs syndrome of similar mental age are impaired in relation to controls on both sets of items and do not demonstrate the material-specific phenomena of the children with WS.

**Conclusions**

Children with WS make semantic errors in reading, make semantic errors in naming and make semantic errors on receptive vocabulary tasks. In each case, detailed semantic knowledge is required for accuracy. There is not a broad lexical deficit but rather failure to access fine-grain semantic feature specifications of lexical items. As Temple and Clahsen (2002; Clahsen and Temple, 2003; Clahsen et al., 2004) have discussed there is also failure to access fine-grain morphological features of lexical items. Thus in WS, deep dyslexia derives in part from an abnormality in the activation of the semantic system. Newcombe and Marshall (1980) speculated that semantic errors in adults with deep dyslexia might arise from an intrinsic instability in the semantic system which is normally blocked by phonological input and explained why semantic errors are essentially absent in phonological dyslexia, in which, although explicit phonological skills are weak, they are not absent. In relation to children, it could be argued that instability of the semantic system is not intrinsic but in WS, difficulty in activating fine grain semantic features leads to a pattern of unstable performance characterised by semantic errors which emerge in reading, naming and receptive vocabulary.

**Semantic Errors in Number Reading**

A second child with WS, Jane, has also been the focus of an investigation into early reading development.

Jane was an 11 year old girl with WS, with a mental age of 5;7. She was tested on a series of reading tasks addressing letter reading, non-word reading, single word reading and text reading skills.

Jane had a reading age of 5;5 on the Schonell single word reading test. She had a reading age on the British Ability Scales of 6;1. She could name 17 of the 26 letters of the alphabet and sound out 2/59 simple nonwords read correctly. Presented with 509 words to read aloud, she read 128 correctly, of which the majority were nouns (e.g., egg, house, mummy, sheep, television) or function words (e.g., the, and, but, it, her, down, of, up, by). There was a low semantic error rate with single words of 4-5% but a striking semantic error rate with number words. Her reading of the number words 1-12 is given in Table II, where it is contrasted with reading of the same numbers presented in Arabic form. The stimuli were presented in random order. Semantic paralexias comprising number words substitutions were made to 9/12 (75%) of the number words. The number word substitutions did not retain the digit length of the number represented, as single digit numbers were substituted for two-digit numbers and vice versa. All stimuli were represented by a single word. All errors were single words. Thus, the size of the lexical item was retained but not the digit length of the number. In some previous studies of number reading disorders, teen numbers have appeared to represent a distinct class within which substitutions occur but for Jane the teen class was violated with teen responses being given to single digit stimuli. In contrast, Arabic number reading of the numbers 1-12 is perfect.

**Conclusions**

Despite the difficulties with reading numbers words, reading of Arabic numbers was very good. Thus, the difficulty related not to the modality of input or output but to the form of the numerical stimulus. It was the written verbal representations of numbers which was problematic. Semantic errors were generated when reading number words

<table>
<thead>
<tr>
<th>Type of Grid</th>
<th>WS</th>
<th>Controls</th>
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<tbody>
<tr>
<td>BPVS 8 item</td>
<td>66.84*</td>
<td>63.10</td>
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<tr>
<td></td>
<td>(12.77)</td>
<td>(4.12)</td>
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<tr>
<td>BPVS 12 item</td>
<td>63.11*</td>
<td>58.74</td>
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<tr>
<td></td>
<td>(14.37)</td>
<td>(4.84)</td>
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<tr>
<td>BPVS 24 item</td>
<td>58.58*</td>
<td>49.06</td>
</tr>
<tr>
<td></td>
<td>(14.77)</td>
<td>(6.15)</td>
</tr>
<tr>
<td>S&amp;W 8 item</td>
<td>89.68**</td>
<td>97.51</td>
</tr>
<tr>
<td></td>
<td>(8.95)</td>
<td>(1.66)</td>
</tr>
<tr>
<td>S&amp;W 12 item</td>
<td>87.33**</td>
<td>96.44</td>
</tr>
<tr>
<td></td>
<td>(10.58)</td>
<td>(2.99)</td>
</tr>
</tbody>
</table>

*Significantly enhanced [Manova, Pillai’s Trace F (3, 20) = 3.60, p < .016].
**Significantly impaired [Manova, Pillai’s trace F (3, 20) = 3.66, p < .015].
but not when reading Arabic numbers. This supports the view that the mechanism underlying the number processing of these two types of stimuli are distinct. This possibility was implied by some of the discussion of Hinchelwood (1917) who noted for example that a patient with alexia who could no longer read nevertheless “read at once the number standing at the top of each paragraph of test type”. He could also read more complex Arabic numbers “rapidly and fluently”. This suggested a distinct representation or mechanism for reading Arabic numbers. The impairment evident in Emily suggests that number words may also have representations distinct from other words, as the high semantic error rate in reading number words is not mirrored in the semantic error rate to other single words, where it is only 4-5% and not therefore definitively higher than chance (Ellis and Marshall, 1978).

A distinction between the processing of most written words and those that represent numbers has been described previously in the case Paul (Temple, 1989), however, in contrast to Jane, Paul had high reading levels for both words and nonwords. Paul was an 11 year old boy of normal intelligence. He was assessed on a battery of reading tasks addressing not only the reading of words but also the reading and writing of numbers in both verbal and Arabic form. Details of the tasks employed are given in Temple (1989).

Paul had both a Schonell reading age and a Neale reading age of 11 years. He had good ability in reading irregular words, for example, he could read *orchestra, physics, choir and colonel* accurately. He thus had a well developed lexico-semantic reading system. He also had good skills in reading aloud long unfamiliar regular words, for example, he could read *hectographic, chitterling* and *intertergal* accurately. He thus had a well developed phonological reading system.

Nevertheless, when asked to read aloud written number words he made semantic errors substituting one number for another. The errors had a characteristic form with the response always preserving the length and syntactic structure of the stimulus but with the substitution of lexical items within the number. Examples of these number reading errors are given in Table III, where they are compared to Paul’s reading of Arabic numbers. For Paul, Arabic number reading is characterised by a similar difficulty to that seen in the reading of number words. The length of the number is preserved in the error but digits are substituted so that inappropriate lexical items are entered into an accurate syntactic frame for the number (McCloskey et al., 1985; Temple, 1989).

For Paul, semantic errors are not made when reading words except where the words represent numbers. Semantic errors are made to both number words and Arabic numbers. For Jane, the number reading difficulty was more specific, affecting the reading of number words but not the reading of Arabic numbers. The semantic errors in number reading which characterise Paul’s skills indicate that number reading appears to be distinct from the reading of other forms of written material as his general reading skills for both irregular and phonological forms are very good. The deficit is manifest in a child who does not otherwise have developmental dyslexia, in the sense that he does not have phonological or surface dyslexia or a reading age which is below expectation. Nor does he have deep dyslexia, and a general difficulty with semantic access. The difficulty is specific to numbers. The evidence from Jane’s case indicates that within this number processing system, the mechanisms underlying the reading of numbers words are distinct from those underlying Arabic number reading as her Arabic number reading is accurate. It is only in the reading of number words that problems arise. Jane is not a good reader of other forms of written material and she does have a dyslexic disorder, though in the context of reduced intelligence it is not markedly out of line with mental age, however, her weak reading skills in other domains are not characterised by high rates of semantic errors. Her semantic errors rate is slightly elevated in relation to mental age but only marginally. In contrast, the semantic error level to the reading of even single digit numbers was 75%. The dissociation between this weak reading skill and the perfect reading of the same numbers when presented in Arabic forms indicates that the reading difficulty is not some kind of output problem occurring after accurate identification of the number, rather it appears to be more centrally based and be a consequence of the nature of the stimulus material rather than its cognitive content.

**Reading Development in Williams Syndrome**

Two cases of WS were discussed above. In one case, Emily, reading ability was very low and reading was characterised by a pattern of deep dyslexia and an elevated rate of semantic errors. In the second case, Jane, reading ability, though also...
very weak was nevertheless above that of Emily and the semantic error rate though slightly higher than normal was much less than for Emily. This raises a question about how reading will develop over time in WS. Temple and Sherwood (submitted for publication, b) have investigated reading skills in a sample of children with WS whose reading age is above that of Emily and Jane.

The eight children with WS had a mean mental age of 6;7 and a mental age range of 5;7-7;9. The control sample was comprised of 12 normal children with an average age of 6;5 and an age range of 5;6-7;4. The average reading age for the children with WS was 86.7 months (SD = 27.2 months). The average reading age for the controls was 85.2 months (SD = 14.7 months).

The children with WS had no impairment in reading age in comparison to mental age. They were not therefore dyslexic. They also had no impairment relative to controls of comparable reading ages in the reading of nonwords. The phonological reading skills of the children with WS are therefore normal in relation to mental age. Nevertheless, there remained a small semantic error rate for the children with WS which was significantly elevated in comparison to controls. The children with WS were also impaired in relation to controls in the reading of irregular words and in the reading of high frequency words.

Conclusions

One possible interpretation of the evident pattern of deep dyslexia at the lowest reading ages in WS and then surface dyslexia in the children with somewhat higher reading ages is that as reading skill improve in WS, phonological skills emerge over time leading to decline in the pattern of deep dyslexia. In relation to this interpretation however, the lexico-semantic reading difficulties which generated marked semantic errors when reading skills were poorer have not disappeared entirely. Although reduced in magnitude, the semantic error rate does remain significantly elevated in relation to reading age. A difficulty with irregular word reading also persists. There is thus over-reliance upon the phonological reading skills which are now developing well. Thus, the very early pattern of deep dyslexia appears to resolve into that more akin to surface dyslexia as phonological reading skills start to unfold.

The proviso to this interpretation is that these comparisons are cross-sectional, involving different children rather than longitudinal studies of the same children. Thus, an alternative interpretation is that the deep dyslexia shown by Emily and Jane may continue with age and the surface dyslexic pattern observed in the children with slightly higher reading ages may represent a different profile in children with WS who can develop sublexical skills. The issue will be resolved with time as longitudinal data becomes available tracking individual trajectories of reading development in WS. The pattern we report for the older children is consistent with that reported in the studies of Laing (Laing, 2002; Laing et al., 2001) who found a reduced influence from the imageability of words in reading and concluded that reading was compromised by weak semantics. Weak semantic skills in reading therefore characterise reading skills in both younger and older children with WS, with at least some older children demonstrating surface dyslexia. As discussed below, developmental surface dyslexia may also be seen in other children for whom there are semantic memory difficulties.

SURFACE DYSLEXIA IN DEVELOPMENTAL AMNESIA

Semantic memory is memory for factual information about the world such as “Paris is the capital of France”. It is also memory for factual information which contributes to knowledge about words, thus a semantic system underpins our knowledge of vocabulary. A semantic system is also a core component of most contemporary reading models. In contrast to semantic memory, episodic memory concerns memory for specific event or episode. In the original use of the term episodic memory, Tulving (1985) considered autobiographical memory for personal events. Nowadays the term episodic memory is generally used more broadly to include memory for recently presented material, though variation in the use of the term continues to confuse some discussions of studies in the field. Here, the term episodic is used to refer to memory for recently presented material and the term autobiographical is used to refer to more personal episodic memories.

Temple and Richardson (2004) have recently discussed a child, C.L., with developmental amnesia, who was detected amongst children attending normal mainstream schools after screening for episodic and semantic memory skills.

The screening measure for semantic memory consisted of the information subtest of the WISC-III which contains a series of factual questions about the world which are of increasing difficulty. The screening measure for episodic memory consisted of a modified version of the Rey (1964), in which a series of twenty words was read aloud for recall of as many as possible. Controls were eleven children matched for age and intelligence who did not demonstrate any abnormality of performance on the screening measures for semantic or episodic memory. In addition to the screening measures, C.L. and the controls were given an extensive battery of tasks addressing components of memory and literacy. Of particular interest to the discussion here, were the memory tasks addressing semantic memory. A new task
addressing children’s factual knowledge about the world had been constructed which contained 75 questions (e.g. “What colour are emeralds?”). These questions were asked of the child but if the child was unable to answer accurately, they were also given a choice of three alternative responses, from which to make a forced choice recognition memory response. Thus, for example, for the question above, they were given the forced choice A Green B Grey C Red. Episodic memory tasks included the Design Memory subtest of the WRAML (Sheslow and Adams, 1990) and the Story Recall subtest of the WRAML (Sheslow and Adams, 1990). Further details of the method are given in Temple and Richardson (2004). C.L.’s reading was assessed on a battery of reading tasks. These included measures of single word reading, nonword reading and the reading of homophones.

C.L. was of normal intelligence, attaining an IQ of 115 on Raven’s (1998) Progressive Matrices. On the screening measure for episodic memory, performance was also normal, however, on the screening measure for semantic memory there was significant impairment. This deficit was explored further, initially with a new semantic memory quiz. Given the a priori expectation of a memory deficit, analyses for C.L. were one-tailed. Performance on the memory quiz was significantly impaired in relation to controls on both free recall of answers and with forced choice recognition memory for answers (see Table IV). In contrast, performance on all the episodic memory tasks was unimpaired (see Table IV). The results of C.L.’s reading are detailed in Table V. Reading of single words was significantly impaired in comparison to controls ($z = 1.77, p < .05$). In contrast, reading of non words on the Graded Nonword Reading Test (Snowling et al., 1996) was unimpaired. There was significantly elevated homophone confusion in relation to controls ($z = 2.02; p < .02$). Irregular word reading was comparable to controls for high frequency words but was significantly impaired in relation to controls for low frequency words. Thus, C.L. demonstrated the classical characteristics of surface dyslexia (Castles and Coltheart, 1996; Coltheart et al., 1983; Marshall and Newcombe, 1973; Temple, 1984b; Wolf and Bowers, 1999).

### Conclusions

C.L., although of normal intelligence and with normal episodic memory had a developmental amnesia which took the form of impaired semantic memory. C.L.’s performance formed a double

### Table IV

<table>
<thead>
<tr>
<th>Semantic memory for facts</th>
<th>C.L.</th>
<th>Controls</th>
<th>Controls</th>
</tr>
</thead>
<tbody>
<tr>
<td>Free recall</td>
<td>20*</td>
<td>33.18 (7.86)</td>
<td>22 45</td>
</tr>
<tr>
<td>Plus forced choice</td>
<td>41**</td>
<td>54.73 (4.84)</td>
<td>47 63</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Episodic memory for designs</th>
<th>C.L.</th>
<th>Controls</th>
<th>Controls</th>
</tr>
</thead>
<tbody>
<tr>
<td>Free recall</td>
<td>41</td>
<td>34.36 (7.00)</td>
<td>24 45</td>
</tr>
<tr>
<td>Recognition</td>
<td>23</td>
<td>23.36 (5.18)</td>
<td>15 30</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Episodic memory for text</th>
<th>C.L.</th>
<th>Controls</th>
<th>Controls</th>
</tr>
</thead>
<tbody>
<tr>
<td>Free recall</td>
<td>26</td>
<td>31.6 (7.1)</td>
<td>17 41</td>
</tr>
<tr>
<td>Recognition</td>
<td>23</td>
<td>24.2 (2.0)</td>
<td>22 28</td>
</tr>
</tbody>
</table>

\*Significantly impaired ($z = 1.67, p < .05$)

\**Significantly impaired ($z = 2.84, p < .002$)

### Table V

<table>
<thead>
<tr>
<th>Reading Scores for C.L. demonstrating surface dyslexia (adapted from Temple and Richardson, 2004)</th>
<th>C.L.</th>
<th>Controls</th>
<th>Controls</th>
</tr>
</thead>
<tbody>
<tr>
<td>Single words</td>
<td>35*</td>
<td>60.55 (14.39)</td>
<td>34 78</td>
</tr>
<tr>
<td>Nonwords</td>
<td>18</td>
<td>21.64 (2.84)</td>
<td>16 25</td>
</tr>
<tr>
<td>Homophonic errors (e.g., sale → sail)</td>
<td>14**</td>
<td>4 (3.46)</td>
<td>0 10</td>
</tr>
</tbody>
</table>

\*Significantly impaired ($z = 1.78, p < .05$).

\**Significantly impaired ($z = 2.89, p < .02$).
dissociation to the performance of the cases of developmental amnesia following birth hypoxia and hippocampal damage reported by Vargha-Khadem and colleagues (Baddeley et al., 2001; Gadian et al., 2000; Vargha-Khadem et al., 1997, 2001, 2002) for whom semantic memory was intact but episodic memory was selectively impaired.

C.L. had a developmental amnesia characterised by impaired semantic memory in the face of intact episodic memory. Reading was characterised by impaired development of the lexico-semantic reading route within which the semantic system is core component, yet there was intact development of the phonological reading system as reflected by normal development of nonword reading skills. The impairment to the reading system was selective and affected only those elements of reading which incorporated a semantic system for accurate performance. Thus, in the differentiation of homophones, phonological information about the stimulus is insufficient. Word specific knowledge of the relationship between the orthographic stimulus and its semantic representation is required for differentiation of homophones. In surface dyslexia, where the lexico-semantic reading route is impaired, this becomes problematic (Coltheart, 1981; Coltheart et al., 2001). Irregular word reading was significantly impaired in relation to controls for lower frequency words. As irregular words violate phonological reading rules an intact lexico-semantic reading system is also required in order to read them accurately. Temple and Richardson (in press) have also discussed a second case of developmental amnesia in a child M.M. Like C.L., M.M. was detected following screening of the normal population as detailed above. M.M. also had a semantic memory impairment, though additionally had verbal episodic memory impairment and displayed memory skills similar to those described in a case by Casalini et al. (1999). M.M. also had surface dyslexia, with an impaired semantic reading system but intact phonological reading skills.

HYPERLEXIA

Hyperlexia is a reading abnormality which has been most commonly described in children with mental retardation for whom reading levels are significantly higher than expectation of the basis of intelligence, however, in many cases the reading skills relate to accuracy or reading aloud rather than superiority in reading comprehension and understanding of the material which has been presented. The form of the superiority in reading aloud has been discussed. Thus, it has been suggested that in hyperlexia the reading skill is like surface dyslexia with good nonword reading and strong phonological reading skills (Aram et al., 1984; Henderson et al., 1993). In contrast, Marshall (1984) suggested that it was like acquired direct dyslexia and Temple (1990b) found support for this view in the study of a child with hyperlexia, for whom accuracy in reading aloud some irregular words was possible despite absence of understanding of the meaning of these words. Irregular words cannot be read with accuracy using the phonological reading system and in the absence of understanding of their meanings, there is also evidence of impoverished semantic knowledge of the words and therefore also an impaired lexico-semantic reading system. This leaves the possible development of a direct reading system, within which words are recognized and abstract representations activate phonological representations for the pronunciation of the whole words, without ever activating meaning. A further alternative, in terms of good orthographic reading has also been discussed (Seymour and Evans, 1992). It is likely that hyperlexia may take several forms and good development of reading skills more broadly, in some cases of hyperlexia, has been indicated.

Good skills in both nonword and word reading are described by Fletcher-Flinn and Thompson (2000). Temple and Carney (1996) have also described hyperlexia in a study of fifteen children, aged 10-12 years, with Turner’s syndrome (TS), a genetic disorder in girls, in which there is normal intelligence but selective impact on components of spatial ability and language development. Temple and Carney (1996) described reading levels which were significantly above the level expected upon the basis of intelligence. Reading skills were also significantly better than controls for both non-word reading and for irregular word reading, indicating elevated development of both the phonological reading system and also the lexico-semantic reading system. Further, the girls were also significantly better than controls in reading comprehension. The hyperlexia was therefore a genuinely advanced skill in all domains of reading.

Recently, a further study has been completed of a sample of much younger girls with TS assessed in their second term at school (Temple et al., in preparation). Participants were ten girls with TS with a mean age of 5;3 (4;9-5;7) and fifteen matched controls with a mean age of 5;1 (4;9-5;9). They were assessed on standardized psychometric measures including the Schonell reading test and the reading subtest of the British Ability Scales (Elliott et al., 1996). They were also assessed on Temple’s (1985) list of words and nonwords. Two further stimulus lists were composed of familiar words which appear frequently in children’s early reading books. The first had 64 items and the second had 124 items. Stuart et al.’s (2003) items were also employed which are suitable for initial stages of reading development with matched sets of items which conform to letter-sound rules, grapheme-phoneme rules or consist of exception words.
On average, the reading age of the controls, was 5;1 on the Schonell. In contrast, on average the reading age of the TS group was 5;9, a difference between the groups of eight months in reading age. This had appeared by the time of testing, which for all children was in the second term of school. Of the other eight reading lists (see Table VI) which were employed with each group, for each stimulus list the TS group attained a higher average score a consistency which was statistically significant.

Conclusions

TS is a genetic disorder in which there is absence of the second X chromosome and also early prenatal gonadal dysgenesis, leading to reduction of the normal hormonal influences upon foetal development. Yet, it is associated with reading skills which are stronger than normal in middle childhood (Temple and Carney, 1996). In the study of school starters with TS, there was also a consistent advantage across tasks in favour of the reading skills of those with TS in comparison to controls. This suggested that the reading advantage seen in middle childhood emerged early as it was evident consistently by the second term at school. TS is therefore a syndrome in which a genetic abnormality is associated not only with normal intelligence but also hyperlexia. Certain other specific language skills may also be significantly elevated in TS, including vocabulary (Temple, 2002) and components of verbal memory (Bishop et al., 2000; Shephard et al., in preparation). The hyperlexia in TS is neither direct route reading, nor like surface dyslexia in character, nor orthographic reading. It represents an advantage in all components of reading skill including comprehension. This advantage is independent of intelligence and occurs despite a range of perceptual and spatial difficulties (e.g., Temple and Carney, 1995).

DISCUSSION

Marshall (1984) highlighted potential parallels between children with developmental disorders of reading and adults who had acquired reading disorders as a consequence of neurological injury or disease. He advocated the use of a cognitive neuropsychological framework in the investigation of children with developmental abnormalities of cognition, including those with developmental dyslexias. Following the description of developmental phonological dyslexia (Temple and Marshall, 1983; Temple, 1984a), the syndrome has been extensively described in many countries and scripts and it is clear that it is a pervasive form of disorder. The relationship between this pattern of reading difficulty and phonological difficulties evident in explicit phonological tasks employing aural and oral language continues to be the focus of considerable debate.

Although much less pervasive than developmental phonological dyslexia, clear cases of developmental deep dyslexia have now also been described (e.g., Stuart and Howard, 1995). The syndrome has also been described as characterising early reading development in Williams syndrome, where there are also semantic errors in other domains, including naming and receptive vocabulary. Although receptive vocabulary performance in Williams syndrome has been described as strong on simple receptive vocabulary tasks like the Peabody and BPVS which have a small number of distracters of which some may not be closely semantically related to the target, when asked to select items on the basis of their spoken names from arrays in which there is a close semantic relationship between the target and the distracters, the children with WS are impaired. In contrast, they are superior to controls if fine grain semantic knowledge is not required for the items and the distracters have nil or no semantic relationship to the targets. Thus, deep dyslexia in WS may be part of a more generalised difficulty with the activation of fine grain semantic specifications despite relatively large vocabulary stores.

Whilst the semantic errors in developmental deep dyslexia are pervasive, reading disorders in children for whom semantic errors are constrained in relation to domain have also been described. Notably in the domain of number, highly selective reading disorders characterised by high rates of semantic errors have been documented. This indicates that semantic reading errors can be domain-specific. They can occur to number words despite intact ability to read Arabic numbers and they can occur to both number words and Arabic numbers despite intact ability to read words in other domains. This argues for distinct systems or subcomponents within the functional architecture of the reading system which extends beyond the discussions of dissociations between lexico-semantic and phonological reading mechanisms upon which much debate has focussed. Reading models must account for how a child can look at the word five and say “three”, yet look at 5 and say “five”. This phenomenon argues strongly for

<table>
<thead>
<tr>
<th>Reading task</th>
<th>TS group (N = 10) Mean (SD)</th>
<th>Control group (N = 15) Mean (SD)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Schonell reading</td>
<td>6.80 (9.61)</td>
<td>3.13 (5.77)</td>
</tr>
<tr>
<td>BAS reading</td>
<td>13.70 (13.57)</td>
<td>9.00 (9.51)</td>
</tr>
<tr>
<td>Graded non-word reading</td>
<td>3.50 (4.88)</td>
<td>2.75 (2.79)</td>
</tr>
<tr>
<td>64 word list</td>
<td>21.90 (20.19)</td>
<td>15.20 (15.50)</td>
</tr>
<tr>
<td>124 word list</td>
<td>37.10 (35.61)</td>
<td>23.00 (26.90)</td>
</tr>
<tr>
<td>Temple’s 3 letter words</td>
<td>7.40 (5.54)</td>
<td>5.67 (4.70)</td>
</tr>
<tr>
<td>Temple’s 3 letter non-words</td>
<td>5.50 (5.25)</td>
<td>3.67 (4.42)</td>
</tr>
<tr>
<td>Stuart letter – sound words</td>
<td>59.20 (23.54)</td>
<td>45.07 (30.33)</td>
</tr>
<tr>
<td>Stuart grapheme – phoneme words</td>
<td>27.60 (29.72)</td>
<td>22.40 (32.14)</td>
</tr>
<tr>
<td>Stuart exception words</td>
<td>36.00 (26.71)</td>
<td>33.63 (29.23)</td>
</tr>
</tbody>
</table>
suggesting that phonological impairment underlies cases have generated contrary evidence for theories phonological impairment in reading and so these developmental surface dyslexia for whom there is no nevertheless not rare and has been well documented. There are now clear case descriptions of cases of than developmental phonological dyslexia it is described in a range of countries, languages and potentially dissociable within development.

The basis of such a distinction within current models is unclear. Further, in the case of Paul, the semantic paralexias in reading number words occur despite excellent lexico-semantic and phonological skills for other forms of material. Yet, these skills do not appear to be accessible when the input is a number word. Current models of reading written words do not allow for such material-specific dissociation. If the phonological skills accessible in other reading domains were accessible in the reading of number words, they would enable Paul to sound out at least the regular number words. Yet the number words appear to access a specific lexico-semantic activation into which these phonological skills do not feed and in which there is an inaccuracy not evident for other types of written word. This segregation must occur beyond the very initial stages of reading, as it would not at that point be evident what was a number word. For number words to be processed distinctly, they must first be recognized as number words. This would be straightforward in terms of some logographic or iconic mechanism for Arabic numbers but cannot occur simply on the basis of orthographic features for number words. There is limited orthographic distinction between "five" which is problematic and "live" which is not. Current models do not account for these focal dissociations within domains in reading. Whether number words are exceptional in this dissociation from other forms of written material is uncertain. They may represent an exceptional class of items with distinct types of semantic representation or they may be one of a number of domains whose processing is distinctive and potentially dissociable within development.

Developmental surface dyslexia has also been described in a range of countries, languages and orthographies. Although suggested as less common than developmental phonological dyslexia it is nevertheless not rare and has been well documented. There are now clear case descriptions of cases of developmental surface dyslexia for whom there is no phonological impairment in reading and so these cases have generated contrary evidence for theories suggesting that phonological impairment underlies all cases of developmental dyslexia. Whether there is some other common deficit in case of developmental surface dyslexia has yet to be determined. Common difficulties with visual recognition of other complex visual stimuli have not been established. At a slightly higher reading age, children with WS are described, for whom phonologal reading skills are better and deep dyslexia is not seen. Instead over-reliance on phonological reading, leads to a pattern akin to surface dyslexia. This may reflect the resolution of deep dyslexia into surface dyslexia with the acquisition of phonological skills or may reflect distinct subtypes of children with WS with distinct reading disorders. Surface dyslexia has also been reported in cases of developmental amnesia in which there are semantic memory impairments. The cases to date are too limited in number to establish whether there is a causal association, such that a developmental memory disorder of the semantic system will always affect the development of the lexico-semantic reading system as well and thereby generate surface dyslexia or whether the co-existence of the two disorders is merely coincidental or attributable to some other common causal influence which affects both memory and reading. The relationship between disorders of memory of both semantic and episodic form and the development of language and knowledge systems will become more evident in the next few years, given that the number of identified cases of developmental memory disorders is now increasing rapidly.

Hyperlexia with reading at a level above expectation for intelligence can, as Marshall (1984) suggested, be like direct dyslexia but it can also be a genuinely hyperdeveloped skill with elevated phonological reading abilities, elevated lexico-semantic reading abilities and elevated reading comprehension as in TS. Moreover, study of school-starters with TS indicate that this advantage is detectable early in life and is not simply an emergent skill in middle childhood.

Thus, Marshall’s (1984) ideas have had impact not only in the field of developmental dyslexia but in the study of vocabulary development, naming skills, memory disorders, number processing, genetic disorders, learning difficulties and exceptional talents. In language, memory and even mathematics, his arguments in favour of a relatively preformed system have received substantial support from childhood studies demonstrating specific modular effects, without pervasive impact across systems. Nevertheless, in the domain of reading, the issue of what aspect of the functional architecture could be preformed remains. Although one cannot prove that early man was not reading, it is unlikely that a system dedicated to a processing written words is entirely preformed as reading is a culturally developed and transmitted system which occurs in response to specific orthographic exposure and instruction. If the system is parasitic
upon more fundamental preformed component skills though, these may specialise very early in relation to orthographic input. Phonological reading skills could be parasitic upon explicit phonological processes originally developed to address complex aspects of oral communication. A preformed component that might underlie word recognition has not been identified though we know that it is not pattern recognition, visual memory or phonological processes as all of these have been reported as intact in some cases of surface dyslexia.

The selectivity of the impairments discussed in the current paper cannot be accounted for by neuroconstructivist theories of cognitive development which argue for intrinsic interdependency of skills. The selectivity of the impairments also demonstrates the limitations of functional plasticity in compensating for these abnormalities. We have suggested that functional plasticity may be less effective in developmental disorders including those with genetic bases, than in disorders where there is an acquired injury which disrupted what was previously normal development (Temple, 1997). It may be that Marshall’s (1984) arguments in relation to preformism are, in fact, at their most relevant in the field of genetic disorders. The framework and issues Marshall (1984) outlined have provided a foundation for the development and elaboration of systematic investigation of not only the developmental dyslexias but developmental and congenital disorders more broadly.

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Christine M. Temple, Developmental Neuropsychology Unit, Department of Psychology, University of Essex, Wivenhoe, Colchester CO7 9UJ, UK. e-mail: tcm@essex.ac.uk

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