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Failure to learn a new spatial format in children with developmental dyslexia

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A general problem in studying children with developmental dyslexia is how to separate inefficiency in learning on the one hand from exposure to written texts on the other. To evaluate dyslexic children's learning abilities with graphemic materials, we tested their improvement in a condition that minimized previous experience with words (i.e., "novel words") and with the standard, horizontal spatial letter array (i.e., a non-canonical "zigzag" format). We selected five pairs of children with dyslexia and (younger) typically developing readers matched for reading speed and accuracy in these specific conditions. Reading performance on novel words in the zigzag format was measured in 23 sessions; learning curves were fitted by power functions. Similar to typically developing readers, children with dyslexia improved their reading of novel words presented in the new format; however, their rate of learning was slower than that of typically developing readers. Furthermore, their learning to read in the new format did not generalize to novel untrained items, whereas significant generalization was present in typically developing readers. As the failure to generalize learning of the spatial format could not be attributed to reduced experience, it indicates a genuine disability and points to impaired perceptual learning as a factor in developmental dyslexia.

Children with developmental dyslexia fail to learn to read effectively. This learning disability prevents the development of accurate and fluent reading despite adequate school attendance, normal intelligence and absence of gross neurological disorders, and has widespread effects on their lives, with important negative consequences on academic achievement and self-esteem (e.g.¹). A general problem in studying these children is to discriminate between inefficiency in learning on the one hand and exposure to written texts on the other; in fact, because of their pervasive reading disturbance, children with dyslexia are much less willing to spend time reading than normal readers². Consequently, they have less exposure to written texts than their peers.

In the present study, we aimed to evaluate dyslexic children's learning abilities in a condition that minimized their previous experience with texts by considering two aspects of this experience: *i*) the reading materials and *ii*) the spatial array of letters in reading. As for reading materials, we used novel words (i.e., legal strings of letters without any corresponding entry in the lexicon), so that neither children with dyslexia nor typically developing readers had previous experience with them. As for spatial array, we used a non-canonical zigzag format which was unfamiliar to both groups (see an example in the Methods section). This letter arrangement should minimize the difference between groups in one of the perceptual abilities supporting reading, that is extracting visual information from a horizontal string of letters.

In framing our study we refer to a recent influential model, the Local Combination Detector (LCD) model³, which has already being used to describe the effect of word degradation⁴. According to this model, written words are encoded by a hierarchy of detectors tuned to increasingly larger and more complex word fragments (visual features, single letters, bigrams, quadrigrams and, possibly, words). At the neural level, bilateral information from letter features and single letter converges to the left occipital-temporal region called visual word form area (VWFA); here, a posterior-to-anterior gradient is present with a progression in selectivity to increasingly word-like stimuli⁴⁻⁶. Thus, over years of practice, frequent combinations of letters are selected to be represented by dedicated neurons⁴, and the VWFA becomes attuned to the regularities of the writing system, yielding fast parallel processing in reading^{4,6}. Notably for the present study, this perceptual learning is selective for standard print, *i.e.* words aligned horizontally and printed at high contrast in a standard font. Studies in typically developing readers have shown that, when presentation deviates greatly from this hyper-learned format, reading speed decreases and word length effects emerge, indicating a shift from parallel to effortful serial processing. This occurs



Table 1 | Description of participants. Some characteristics of dyslexic and typically developing readers are reported. The children with dyslexia were below their age norms for reading speed and accuracy as assessed by a standard Italian reading test^{39,40}. The control readers performed near average (i.e., approximately 0 in terms of z score depending on individual grade level) on the same test and were younger than the children with dyslexia by about three and a half years. All children demonstrated normal intelligence on Raven's Coloured Matrices

Children with dyslexia		Age	Sex	Grade	Raven test	Reading speed	Reading accuracy
1 d	MS	13.5	F	7	26	-0.32	-3.41
2 d	DB	13.2	M	7	32	-0.88	-2.39
3 d	AD	13.5	M	7	26	-2.26	-3.41
4 d	VG	13.3	F	7	29	-2.13	-7.61
5 d	MA	14.0	M	8	31	-4.11	-6.90
Average		13.5			28.8	-1.94	-4.74
SD		0.3			2.8	1.47	2.35
Typically developing readers		Age	Sex	Grade	Raven test	Reading speed	Reading accuracy
1c	AR	10.0	F	4	28	1.05	0.53
2c	MA	9.5	M	4	27	0.66	-0.10
3c	GI	10.8	M	5	28	0.49	0.03
4c	IA	10.7	M	5	27	0.20	0.19
5c	LI	8.5	F	3	27	0.07	0.02
Average		9.9			27.4	0.50	0.13
SD		0.9			0.5	0.39	0.24

with low-contrast⁷, rotated⁸, or highly spaced^{4,9} words as well as with words presented in case-alternation^{10,11}, zigzag or vertical mode¹². Results of neuroimaging, ERP and MEG studies^{4,13,14} indicate that reading degraded words activates dorsal posterior parietal areas, pointing to the role of attentional areas in reading tasks. In proficient readers, this effortful serial processing emerges most clearly when the automatic and parallel processing supported by the VWFA cannot deal with stimuli, as in the case of degraded words, since these stimuli are out of the limits of the perceptual expertise of the system⁴.

Notably, slow, serial processing dominate the reading of children with dyslexia also with standard texts (as indicated by the presence of large length effects¹⁵) as if they were unable to optimize the functioning of their VWFA. Indeed, there is now considerable evidence indicating that children with dyslexia show very poor activation of the VWFA in various reading tasks (e.g., for a review¹⁶). However, it is unclear whether this is the cause or the effect of their learning disturbance. As mentioned above it is difficult to separate inefficiency of learning on the one hand from exposure to written texts on the other. In the present learning study, we controlled experience with orthographic material using novel words rather than real words. We also controlled experience with format using a zigzag display of letters rather than a standard horizontal array. The zigzag format was selected because it proved quite disruptive in preliminary testing and decreased reading speed more than other format manipulations, such as vertical presentation or case alternation. According to the LCD model³, the zigzag presentation should leave intact the visual analysis of single letters performed bilaterally, and the first stage of VWFA processing, but should impact on later stages, starting from the level where bigrams are computed. In fact, zigzag presentation activates non-aligned detectors, and this spatial arrangement is out of perceptual expertise of VWFA⁴. Thus, the contribution of effortful serial processing, with attentional selection of space-distributed letters, should be important in carrying out the task.

A case series of children with dyslexia and typically developing readers participated in the present study (see Table 1). We selected pairs of dyslexic and (younger) typically developing readers matched for reading speed (vocal reaction times, RT) and accuracy on our experimental task (i.e., reading zigzag novel words) before training. Starting the training from these comparable baselines allows addressing two key issues in dyslexia. First, are dyslexic children able to learn to read novel words in a new zigzag format at the same rate as typically developing readers? To test this, we examined the perform-

ance of the children's pairs through a sufficiently large number of sessions to reliably estimate their curve of learning (based on the law of practice hypothesis that learning occurs following a power function¹⁷). Second, if children with dyslexia are able to learn through training, is this learning specific to the trained items or does it extend to new untrained items? Generalization was tested by presenting "new", untrained novel words in the same zigzag format at the end of the training. The performance on these items was compared to that on the original set of stimuli at the pre- and post-training. Some decay is expected when comparing performance on these new items with the items subjected to training. However, better performance on the new novel words with respect to pre-training (when zigzag novel words were presented for the first time) would indicate that children learnt to read in this non-canonical format (i.e., generalization of format). By contrast, a large performance decay with the new untrained items (with respect to post-training) equaling the performance at the pre-training, would indicate specific learning for trained items and no generalization of format.

Results

Learning curves based on RTs are presented in Fig. 1, separately for the two groups of readers (individual curves are presented in Supplementary Information files). Due to the selection criterion, children with dyslexia and typically developing readers had closely comparable baselines in the pre-training session 1 (see details in Table 2 top and bottom for RT and accuracy data, respectively). To allow for a full appreciation of the effect of training, on the right of Fig. 1 (bar plot), the performance on the new set of untrained novel words at the post-training session is also presented.

During training, typically developing readers improved at a fast rate, closely following a power function of practice, both in terms of RTs (Fig. 1) and errors (Fig. 2)¹⁸; details of the fitting functions are presented in the captions of the two figures. Dyslexics' RTs and accuracy also improved progressively with training (again well fit by a power function). However, their rate of learning was slower than that of typically developing readers (as indicated by a lower power function coefficient). Thus, by the end of training they were slower and more inaccurate than typically developing readers (see Table 2, POST: trained NW). At the post-training session with the new set of novel words (black bar in Fig. 1), the performance of children with dyslexia was very similar to that at the first session; by contrast, typically developing readers (white bar in Fig. 1) showed

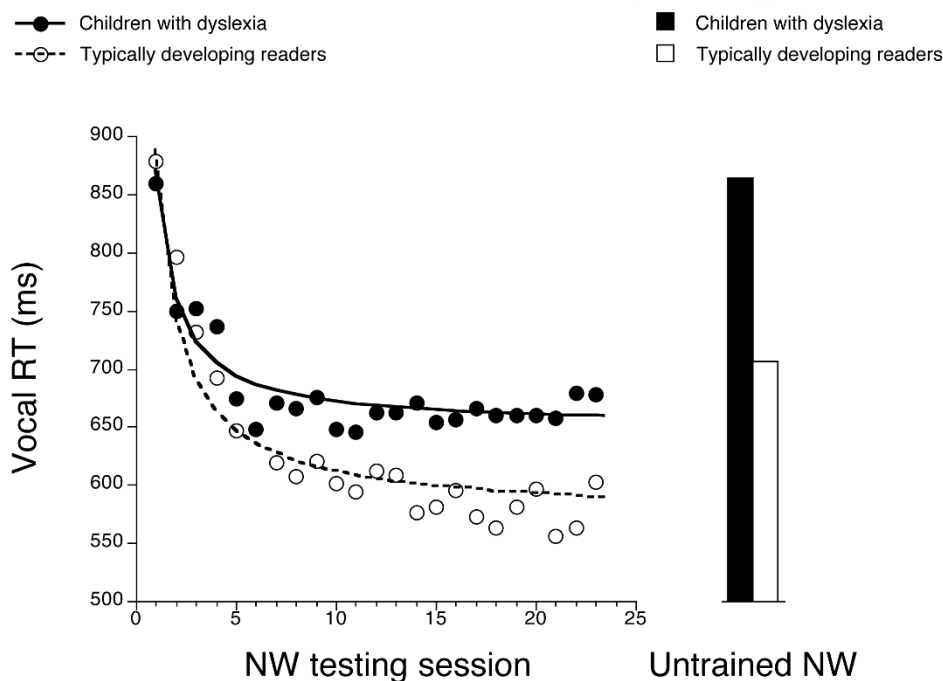


Figure 1 | RTs for zigzag novel words (NW) as a function of the number of testing sessions. Open symbols and white bars report the averages for controls; filled symbols and dark bars report data for dyslexic children. RT data were fit by power functions separately for the groups of dyslexic children (continuous lines) and controls (dotted lines). The learning curves were well fit by a power function for both groups of children ($R^2 = 0.92$ and $R^2 = 0.88$ for control and dyslexic children, respectively). The equations describing the learning curves were $y = 220x^{-1.00} + 650$ (2) and $y = 318x^{-.88} + 570$ (3), for dyslexic children and controls, respectively. The bars represent RTs for untrained novel words at the post-training session.

an improvement in performance with respect to the beginning of training.

The RT data were formally analyzed by linear mixed effect model analyses (summarized in Table 3; for description of the models see the Methods section).

The effect of “training” was examined in the two groups by comparing pre vs post RTs in reading zigzag novel words (model A, see Linear mixed effect model analyses section). The analysis showed the significance of the training effect ($F(1, 813.7) = 298.5, P < 0.0001$) as well as the group \times training effect ($F(1, 805.6) = 20.2, P < 0.0001$). Both groups of children showed a significant training effect (children with dyslexia: pre = 859.2 ms, post = 677.2; $P < 0.001$; typically developing readers: pre = 878.6 ms; post = 603.0 ms; $P < 0.001$), but the effect was numerically larger in typically developing readers (diff. 275.6 ms) with respect to children with dyslexia (diff. 182.0 ms) (see Tables 2 and 3). Due to the pairing selection, groups did not differ in the pre-training condition but differed in the post-training condition ($P < 0.001$). With regard to the random effects, the effect of item was significant ($Wald Z = 2.7, P < 0.01$) but the effect of participant was not ($Wald Z = 1.4, n.s.$). This indicates that individual differences in the two case series of children did not appreciably contribute to generating the observed effects.

The effect of “session” was examined by comparing RTs in each of the 23 sessions with the same novel words (model B, Linear mixed effect model analyses section). The analysis showed the significance of the effect of group ($F(1, 9925.8) = 456.4, P < 0.0001$), session ($F(22, 9925.3) = 61.5, P < 0.0001$) and group \times session ($F(22, 9925.2) = 9.4, P < 0.0001$). An exploration of the interaction shows that the groups differed significantly from the third session onwards; no difference was present for the first presentation of the zigzag novel words and a tendency ($P = 0.067$) was present for the second presentation. Again, the random effect of item was significant ($Wald Z = 4.7; P < 0.0001$), but the effect of participant was not ($Wald Z = 1.4, n.s.$).

The effect of “stimulus material” was examined by comparing RTs to trained novel words at post-training with RTs to “new” untrained novel words (see model C, Linear mixed effect model analyses section). The analysis showed the significance of the effect of group ($F(1, 784.9) = 108.9, P < 0.0001$), stimulus material ($F(1, 786.0) = 171.7, P < 0.0001$) and group \times stimulus material ($F(1, 785.1) = 7.8, P < 0.01$). The interaction showed that both groups of children were slower on untrained than trained targets (children with dyslexia: untrained = 863.2 ms, trained = 677.2; $P < 0.0001$; typically developing readers: untrained = 706.2 ms; trained = 603.0 ms; $P < 0.0001$); however, the interaction showed that the difference in reading trained with respect to untrained novel words was larger for children with dyslexia than typically developing readers (children with dyslexia: diff. = 186.0 ms; typically developing readers: diff. = 103.2 ms; see Table 2). Also in this case, the effect of item was significant ($Wald Z = 3.1, P < 0.01$), but the random effect of participants was not ($Wald Z = 1.4, n.s.$).

The “generalization of format” to untrained novel words was examined comparing RTs in pre-training to RTs to untrained novel words after training (model D, Linear mixed effect model analyses section). The analysis showed the significance of the effect of group ($F(1, 707.6) = 11.0, P < 0.001$), generalization of format ($F(1, 717.4) = 22.3, P < 0.0001$) and group \times generalization of format ($F(1, 702.4) = 31.7, P < 0.0001$). The interaction indicates that typically developing readers showed faster RTs to untrained novel words in the post-training compared to the pre-training condition ($P < 0.001$; in Fig. 1, compare the white bar with the open circle in session 1; see also individual and group data in Table 2 top), whereas children with dyslexia did not (in Fig. 1, compare the dark bar with the filled circle in session 1; see also Table 2 top). The random effect of item was significant ($Wald Z = 2.3, P < 0.05$), but the effect of participant was not ($Wald Z = 1.4, n.s.$).

In Table 4, we detail (at both individual and group levels) how much of the improvement with training could be ascribed to item-



Table 2 | Median vocal RTs (top) and accuracy (bottom) for zigzag novel words (NW). Each child with dyslexia was individually matched with a typically developing reader on the basis of his/her performance on both vocal RTs and percentage of errors in reading zigzag novel words at the pre-training session (*baseline*). Individual dyslexic-control pairs of readers are presented in each row. The “*PRE*” column represents the baseline performance in the first session with the 50 zigzag novel words. The “*POST: trained NW*” column reports performance after 22 sessions with the same list. The “*POST: untrained NW*” column reports performance on a new list of 50 novel words (matched for bigram frequency and initial phoneme with the trained list). Individual and averaged group data are reported separately for children with dyslexia and typically developing readers

Children with dyslexia	PRE: Baseline	POST: trained NW	POST: untrained NW	Typically developing readers	PRE: baseline	POST: trained NW	POST: untrained NW
Vocal RTs (ms)							
MS	661	591	875	AR	715	520	600
DB	722	567	674	MA	715	590	672
AD	840	751	865	GI	741	653	788
VG	901	641	712	IA	1097	499	540
MA	1172	836	1190	LI	1125	753	931
Average	859.2	677.2	863.2	Average	878.6	603.0	706.2
SD	198.9	113.5	203.4	SD	212.8	103.5	156.1
Accuracy (percentage of errors)							
MS	22	0	24	AR	14	0	12
DB	12	4	32	MA	22	2	20
AD	14	2	12	GI	16	2	8
VG	16	4	32	IA	6	0	26
MA	24	4	20	LI	20	4	30
Average	17.6	2.8	24.0	Average	15.6	1.6	19.2
SD	5.2	1.8	8.5	SD	6.2	1.7	9.2

specific learning vs generalization of format. In controls, improvement on trained items (275.6 ms) was due to learning the format (172.4 ms) and the specific items (103.2 ms). By contrast, there was no improvement in the ability of dyslexic children to generalize learning of the zigzag format to untrained novel words; that is, the improvement after training (182.0 ms pre-post training difference)

was entirely due (100%) to learning specific items in the zigzag format (186.0 ms), not the format *per se* (−4.0 ms).

Discussion

Through repeated presentation of novel words in a novel format, both typically developing readers and children with dyslexia progres-

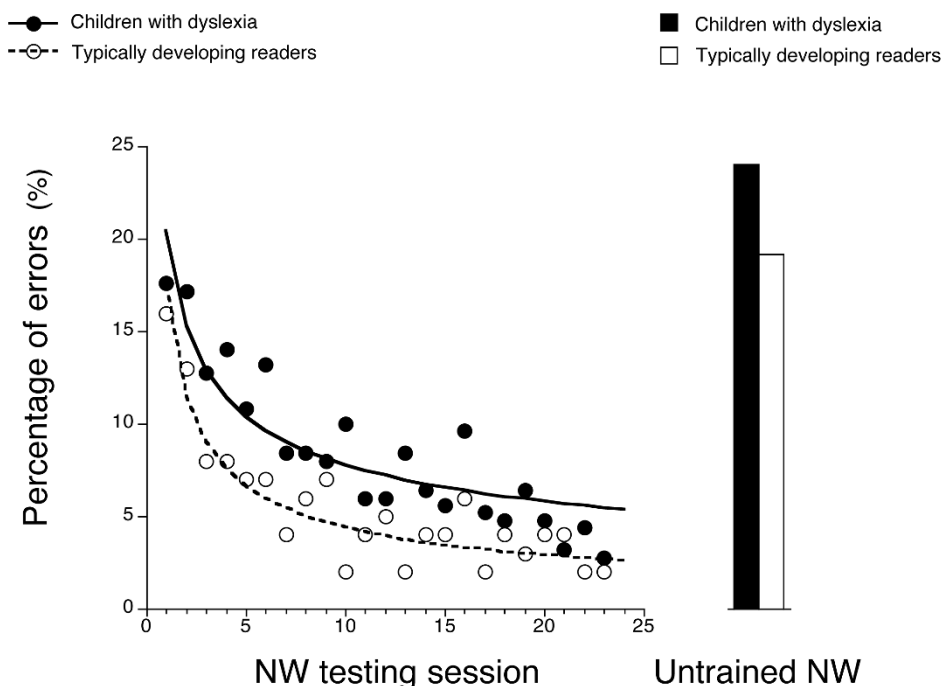


Figure 2 | Accuracy data (percentage of errors) for zigzag novel words (NW) as a function of testing sessions, separately for the two groups of children. Filled circles: dyslexic children; open circles: controls. Accuracy data were fit by power functions separately for the groups of dyslexic children (continuous lines) and controls (dotted lines). The equations describing the learning curves were $y = 20.5x^{-0.42} + 0$ (4) and $y = 17.5x^{-0.60} + 0$ (5), for dyslexic children and controls, respectively. The bars represent accuracy for untrained novel words at the post-training session. Note that also for accuracy learning by training was higher in controls than dyslexic children; however, generalization to untrained items was low for both groups.



Table 3 | Results of the Linear mixed effect model analyses (see Data analysis for details). Note that, in all analyses, the random effect of item was significant, while the random effect of participant was not

MODEL	EFFECT	Degrees of freedom	F	Probability level
A) Training: pre vs post RTs in reading zigzag trained novel words	Group	1, 806.1	3.08	0.080
	Training	1, 813.7	298.48	0.000
	Group × Training	1, 805.6	20.18	0.000
B) Session: RTs at each of the 23 presentations with the trained non-words	Group	1, 9925.8	456.40	0.000
	Session	22, 9925.3	61.50	0.000
	Group × Session	22, 9925.2	9.42	0.000
C) Stimulus material: post-training RTs on trained novel words vs RTs on new untrained novel words	Group	1, 784.9	108.87	0.000
	Stimulus material	1, 786.0	171.68	0.000
	Group × Stimulus material	1, 785.1	7.77	0.005
D) Generalization of format: RTs in pre-training vs RTs to untrained novel words after training	Group	1, 707.6	11.04	0.001
	Generalization of format	1, 717.4	22.27	0.000
	Group × Generalization of format	1, 702.4	31.68	0.000

sively improved their reading performance. Nevertheless, children with dyslexia did so to a lesser extent than controls. Furthermore (and critically for the present analysis), the children with dyslexia failed to transfer learning of a new format to new untrained items, indicating that their improvement during training was entirely due to the learning of specific novel words. As experience with both format and stimulus materials was minimized, the failure of children with dyslexia to attune to a new graphic format cannot be easily attributed to reduced exposure to print and indicates a genuine perceptual disability to learn a new format (despite partially spared ability to learn specific items). Namely, children with dyslexia were unable to build-up an efficient, fast, parallel processing of multiple letters displayed in a new array. We suggest that their performance in this experiment mimics (in a comparatively short time window) their failure to learn to read at school in the standard horizontal array. The reduced ability to cope with zigzag format of children with dyslexia contrasts with the ability of typically developing readers to adapt to this new perceptual condition and transfer learning to new items.

The present findings are in keeping with the idea that VWFA hypo-activation in children with dyslexia¹⁶ is not a by-product of limited reading experience but indicates selective inability to build up an efficient hierarchy of detectors tuned to larger and more complex word fragments. The VWFA provides a very flexible perceptual task-specific mechanism that allows reading regardless of size, font, cursive or capital letters, and even when atypical features¹⁹ or other sensory modalities²⁰ define the shape of the letters. In good readers, the VWFA ensures tolerance for a certain degree of deviation from the standard horizontal format (small rotation of letters, size etc.⁴). The present data gathered in typically developing children, particularly their fast learning and transfer of learning to untrained items, suggests growing tolerance for misalignment of the pre-existing VWFA mechanism and confirms its high plasticity throughout the lifespan (for adult subjects, see²⁰). Thus, the VWFA is also able to learn destructive manipulations of the standard (e.g., zigzag format) possibly through plasticity of binding between detectors starting from the bigram stage³.

The slower learning rate and absence of transfer suggest that this *flexible perceptual* task-specific mechanism is poorly developed in children with dyslexia. This is consistent with fMRI results indicating impaired tuning of VWFA²¹ and focal disruption of functional connectivity between VWFA and left inferior frontal and inferior parietal areas, two major components of the language network²². The slow, serial, stiff, reading, likely driven by attentional control of dorsal parietal areas¹⁴, is used by typically developing readers in the case of degraded stimuli⁴ and, likely, also when the zigzag format was administered before learning (present study). More generally, it is used by all beginner readers when they initially learn to read at school in the standard format. However, children with dyslexia are unable to shift toward a fast parallel mode, and after years of reading experi-

ence still show strong dependency on stimulus length, a serial processing cue that has been shown in both languages with regular^{23,24} and irregular orthography²⁵, and which is dominant only in the early stages of learning in typically developing readers²⁶.

Although both dyslexic and typically developing readers improved their performance in training novel words, the representations of the children with dyslexia were item-specific. In the LCD model³, the inability to learn a new format indicates a deficit of the mechanisms that allow perceptual learning through local combination detectors; this deficit could be the base of developmental dyslexia. According to the physiological literature, perceptual learning is mediated by sprouting and pruning of connections, with effects that modify the functional properties of neurons even at the V1 level²⁷. One may speculate that plasticity processes taking place through reading experience in VWFA are less efficient in children with dyslexia and thus unable to shape efficient and flexible connections within the VWFA, and possibly also towards other cortical areas relevant in the reading process.

An alternative explanation of present results may be framed within the attentional deficit hypothesis of dyslexia (for reviews, see^{28,29}). According to this view, the ability to dynamically reallocate attention is reduced in dyslexia^{30,31}. Thus, the inability to learn the zigzag format may express the difficulty of isolating the appropriate chunk within the target and shifting the locus of attention as the zigzag word form is parsed. Although the presence of an attentional deficit is debated^{32,33}, the role of dorsal attention network in reading is supported by increased parietal activity for words presentation in unusual formats^{4,11,13,14}. Furthermore, it has been recently reported that the VWFA shows maximal resting-state correlation with the dorsal attention network³⁴. Thus, if an attentional deficit were present in dyslexia, one might expect the VWFA to be inefficient because of a poor input. Within the attentional hypothesis, the total lack of transfer to new untrained items would indicate that spatial attention does not improve through repetition. Possibly, the partial improvement present for trained novel words could be seen as due to the partial functioning of the VWFA.

Overall, the present results indicate that children with dyslexia failed to learn a new graphic format despite partially spared ability to learn specific items. By minimizing experience with both format and stimulus materials we showed that this deficit is not due to reduced exposure to print and indicates a genuine learning perceptual disability. With notable exceptions³⁵ research on developmental dyslexia (and more generally on word recognition) has largely relied on single-session experiments and only in the last few years learning designs are starting to be used more extensively (e.g.³⁶). For example, it has been recently found that non-word learning occurred more slowly in college students with a diagnosis of dyslexia than in typical readers (and the length effect persisted for longer periods of time³⁷). These results are generally consistent with the present findings and



$$y = ax^{(b)} + c \quad (1)$$

where y is the reaction time, a is the difference between initial and asymptotic performance, x is the session number (i.e., the amount of practice), b is the exponent that determines the shape of the function, and c is the asymptote indicating the limit of performance.

Linear mixed effect model analyses. Pre-post RT training effects were analyzed with linear mixed effect model analyses^{46,47}. This is a robust analysis that allows controlling for the variability of items and subjects; it prevents the potential lack of power of by-subject and by-item analyses and limits the loss of information due to prior averaging of by-item and by-subject analyses.

Four different analyses were carried out. In each analysis, the dependent variable was the individual child's vocal RT on each item in each experimental condition. Participant (i.e., the "super-subject" made by each dyslexic child and their paired control) and item were entered in each analysis as random factors and group (children with dyslexia vs typically developing readers) as a fixed factor. Furthermore, depending on the objectives of each analysis, the following were entered as fixed factors: A) training (comparing pre vs post RTs in reading zigzag novel words object of training); B) session (comparing performance on each of the 23 presentations with the novel words of the trained list); C) stimulus material (comparing post-training RTs on trained novel words to RTs to "new" untrained novel words); and D) generalization of format (comparing RTs in pre-training to RTs to untrained novel words after training).

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Author contributions

M.P. designed and performed the experiment and analyzed data. M.D.L. designed and implemented the computerized experiment and carried out part of statistical analyses. A.W.E. designed the project and wrote the manuscript. C.V.M. carried out part of statistical analyses, D.S. and P.Z. supervised the project and wrote the manuscript.

Additional information

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