

Combined auditory and articulatory training improves phonological deficit in children with dyslexia

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Abstract

19 children with dyslexia aged 7;2 to 10;9 were selected from a clinical sample and tested using a large neuropsychological battery in order to specify the severity and subtype of dyslexia as well as the presence of comorbid conditions. Thereafter, they received a standardized training of 6 weeks of daily auditory exercises aimed at reinforcing explicit and implicit phonological awareness. 10 participants also received a specific training of the sensory-motor aspects of articulatory production of individual phonemes during the first 3 weeks of auditory training, whereas the remaining received the same during the last 3 weeks of training. Repetition, phonological awareness, reading and spelling were assessed before the first session, between the two sessions and after the second session. Results confirm the overall efficiency of intensive phonological training, even with exclusively auditory material. The main outcome of this study is a significant improvement of phonology and non-word reading specifically during the periods where the two methods were associated, suggesting a significant contribution of articulatory training to the observed improvement. Finally performance to a motor tapping task proved to be one of the best predictors of training efficiency while comorbid coordination or attention deficit did not interfere. Results are interpreted in reference to current theories about mechanisms underlying dyslexia.

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Remediating dyslexia has become a challenge for both neuroscientists and clinicians during these last ten years for several reasons. First, dyslexia, the commonest form of learning disability, is a widespread condition affecting about 5-7 % of school-age children with major impact on their academic and ultimate social achievement (Habib, 2000; Démonet et al., 2004). Moreover, scientifically proven efficient training is often taken as evidence in favour of the mechanism postulated to be causal to the deficit, thus contributing to expanding knowledge about learning disorders and their determinants. In this connection, the mechanism most often invoked at the origin of dyslexia is that of poor phonological coding due to deficient phonological representation (Swan & Goswami, 1997; Snowling, 2001; Ramus et al., 2003c). This phonological deficit usually shows up as a reduced ability to discriminate and manipulate speech-sounds in spoken words, so-called phonological awareness. Such deficit, which can be traced back to early childhood, eventually results in a profound incapacity to learn associations between written and oral codes (grapheme-to-phoneme mapping), typically an important stage toward expert reading (Stanovitch, 1988; Rack et al., 1992). Accordingly, studies reporting successful phonological training in dyslexia are abundant and their results largely consistent across studies, to such an extent that this constitutes the universal basis for dyslexia therapy (Ehri et al., 2001; Torgesen et al., 2001; Wise et al., 1999; Vellutino et al., 2004; McCandliss et al., 2003; Ortiz-Gonzalez et al., 2002) as well as for interventions carried out in at risk children to prevent subsequent reading difficulties (Foorman et al., 1998; Alexander & Slinger-Constant, 2004; Elbro & Petersen, 2004). In addition to strictly phonological training, there is also recent evidence that training children to simple auditory discrimination of phonemes may result in significant improvement on phonological as well as on reading tasks (Moore et al., 2005). Finally, such improvement in reading-related tasks has been related to changes in brain activation patterns, as demonstrated with various brain imaging techniques (Simos et al., 2002, Aylward et al., 2003, Temple et al., 2003; Kujala et al., 2001, Shaywitz et al., 2004).

However, all these results suffer from two important weaknesses which may hinder their validity and reproducibility : first, recruitments are generally made from classroom populations, and thus probably include children with low reading ability who would otherwise not meet strict criteria for dyslexia, also likely gathering indistinctly dyslexics of various subtypes. Another– and probably more problematic– concern with these

studies is that, for the sake of efficacy, researchers have generally used remediation tools that were already available at the moment of the study, usually covering wide spectra of underlying mechanisms of presumed impairment instead of focusing on one or few processes. As a consequence, all these studies, though otherwise valuable in showing the potential for children to improve, are barely suitable for a fine-grained analysis of the actual mechanisms underlying improvement. For instance, most of these tools are claimed to provide 'phonic' interventions, a term which encompasses a large range of processes including phonology (the sound structure of speech), phoneme discrimination, and acquisition of connections between these and letters (phonological decoding).

One of these is the method proposed by Tallal and co-workers, known as "FastForward®" (FFW: Tallal, et al., 1996; Merzenich et al., 1996). On the basis of observations suggesting that the dyslexic brain would suffer from a specific inability to process brief and rapidly presented stimuli (the "temporal-processing deficit theory of dyslexia", Tallal, 1980), these authors have implemented a series of video-games for which the auditory component includes temporally modified speech in order to facilitate and progressively restore brain temporal processes. In this and other similar complex and multifaceted, interactive audio-video games (Kujala et al., 2001; Agnew et al., 2004; Hayes et al., 2003b; Magnan et al., 2004), it could be difficult to separate the effects of auditory and visual linguistic stimulation, as well as to disentangle attentional from motivational factors. For example, in a study summarizing five separate trials of FFW training program, Gillam et al. (2001b) reached the following conclusion : "*...It is possible that similar improvements ... may be obtained from a variety of interventions that are presented on an intensive schedule, that focus the child's auditory and visual attention, that present multiple trials..., and that reward progress*".

In a series of preliminary studies, and in an effort to avoid such important biases, Habib et al. (1999, 2002) devised a specific protocol of phonic intervention exclusively comprising auditory exercises tackling the phonological system, in order to reduce the possible intervention of confounding factors. Children received daily exercises where they only had to indicate manually, by pointing to one of three labels, which one out of 3 words or non-words successively heard into headphones was phonetically different from the other two. In addition, in order to limit interference with other (including spontaneous) factors of recovery, the training sessions were compacted over a relatively

short period (5 to 6 weeks). Note that with this protocol, which was initially elaborated to test Tallal's temporal deficit theory by comparing intensive training with and without temporally modified speech, authors only found a modest advantage of training children with slowed speech (Habib et al., 1999). In contrast, they showed that, notwithstanding the actual speech form (with or without temporal modification of the signal), children markedly improved their performance on phonological and spelling tasks, with, however, only partial generalization to reading (Habib et al., 2002). The first aim of the present study was then to confirm this preliminary observation in showing that children with dyslexia do benefit from such daily exercises, exclusively involving auditory spoken materials, during a relatively short period, and even under unmodified form. Since such training is virtually devoid of any visual component, we anticipate that the resulting gain in reading will concern "phonological reading" (reading non-words) rather than "sight reading" (reading irregular and exception words).

Another objective for the present study derives from previous evidence that articulatory exercises may be of some benefit to children with dyslexia, an issue first raised by Alexander et al. (1991). These authors gave a series of preliminary tests to 10 children with dyslexia, and found a marked phonological deficit, as expected. Then, they trained the children individually with a program designed to enhance their awareness of articulatory movements during speech production. This program sought to integrate tactile and somatosensory information from articulators, visual information provided by a mirror placed in front of the child in such a way to allow him/her to see his/her own mouth during speech production, and finally auditory feedback resulting from his/her own sound production. This training program included tutoring participants with schematic drawings representing the form of vocal apparatus for each trained phoneme. Although involving a limited number of participants, this study yielded encouraging results. However, subsequent work by Wise et al. (1999), on a much larger group (122 subjects), failed to find significant differences in phonological reading (non-word decoding) between those poor readers similarly trained for articulatory sensory-motor integration, and those only trained with phonological manipulation during the same period.. Indirect support in favour of the utility of such articulatory training comes from the proposal by Nicolson and Fawcett (1999) of a cerebellar origin of dyslexia. More

specifically, these authors suggested that two frequent aspects of dyslexic children deficits, the incidence of which has been underestimated, i.e. motor coordination and automation deficits, may be ascribed to cerebellar dysfunction. This possibility is supported by clinical (Stoodley et al., 2005), anatomical (Finch et al., 2001) and brain imaging (Nicolson et al., 1999) evidence. The proponents of this theory reasoned that while reading and spelling ability would be mainly affected by the deficit in automaticity, the phonological deficit would emerge through poor articulatory ability. However, this motor theory is currently severely criticized by several groups (Raberger & Wimmer, 2003, Ramus et al., 2003a,b), suspecting that the observed occurrence of cerebellar symptoms would be actually an artefact resulting from the comorbidity of dyslexia with other conditions such as dyspraxia (or coordination disorder) and attention deficit.

In the present work, we therefore sought to evaluate the potential benefit of training sensory-motor aspects of articulatory function in addition to auditory phonological training, and the clinical usefulness of combining auditory and articulatory stimulation, in a group of children with dyslexia in whom possible comorbid coordination or attentional disorders were assessed. Thus, careful selection of participants, together with optimized separation of factors of potential improvement, were expected to maximize the reliability and thus practical validity of the use of such methods in clinical settings.

Material and methods

PARTICIPANTS

Nineteen children, aged 7;2 to 10;9, were selected on the basis of (a) a clinical diagnosis of dyslexia (see below), (b) current enrolment in speech-language therapy (usually 1 or 2 weekly sessions, in accordance with usual practice in France, , and (c) acceptance to participate in a research program.

Selection criteria

The diagnosis of dyslexia was made by the speech-language therapist, and confirmed by the research team, based on (1) a report of significant difficulty in beginning to read

(mostly absence of mastering of grapheme-phoneme mapping after one year of conventional teaching); (2) the reading difficulty being non explainable by lack of intelligence or insufficient schooling; and (3) performance on at least one reading task more than one s.d. below that expected for age.

Once thus selected, all children received an evaluation of non-verbal IQ with Raven's Progressive Matrices and several subtests (similarities, block design, picture completion and digit span) of the Wechsler Intelligence Scale for Children, Third Edition.

Children were excluded if they scored below 25th percentile at the PM 47 and/or below 8/19 on the WISC similarities subtest.

For increasing the homogeneity of the group, we chose to exclude children with expressive and/or receptive oral language problems at the time of inclusion, as evidenced by normal or near-normal performance on classical batteries used by speech-language therapists¹. Especially, as expected with a main diagnosis of dyslexia, none of the 19 participants had apparent articulatory difficulty (which does not rule out more subtle, subclinical deviances; see Lalain et al., 2003).

Neuropsychological characteristics

Dyslexia severity was measured using the classical French "test de l'Alouette" (Lefavrais et al., 1965), providing a reading age in months. Besides tasks included in selection assessment, children were also assessed for usual comorbidities of dyslexia : mainly attention deficit and coordination deficit (here not considered exclusion criteria), as well as general executive dysfunction.

Oral fluency (Controlled Oral Word Association test) was carried out under two forms (Chevrie-Muller et al., 1997) : phonetic and semantic, asking children to provide in one minute as many words as possible starting with letter P and letter F (phonetic) and as many words as possible related to a given category (sport, professions, vacation).

Response inhibition was tested with a modified version of the Stroop task ("animals" Stroop task, Jacot-Descombes & Assal (1986) which consists in 3 printed plates representing either a series of 50 drawings of 5 animals horizontally arranged in semi-

¹ Note that, in the French Health system, speech-language therapists are traditionally in charge of diagnosis and remediation of reading/spelling pathologies, which is not the case in other countries.

random order (plate 1), or printed word of the same animals (plate 2), or finally, a superposition of drawings with the name of another animal (plate 3). Time needed for naming all items on plate 3 weighed against that needed to name items on plate 1 serves to calculate an “interference index”, reflecting the individual’s capacity to inhibit spontaneous tendency to read the superposed word. Plate 1 also served as a measure of rapid naming. Z-scores were calculated by reference to age-matched controls.

Children were also tested specifically as to the possible diagnosis of comorbid Attention Deficit -Hyperactivity Disorder (ADHD), according to results to Conners’ questionnaire for parents (Conners, 1989), which also served to rule out other psychiatric conditions such as anxiety traits or behavioural deviances which could interfere with children’s compliance to treatment. Only two participants scored 15/30 or more on the Conners’ questionnaire, thus possibly diagnosed as ADHD.

Visuo-spatial abilities were tested using the classical Rey’s figure copy (Rey, 1959), as well as part of the Signoret’s “Batterie 84” for visuo-spatial memory (Signoret, 1991). Short-term memory was selectively explored by classical span tasks, in the auditory-verbal and visuo-spatial modalities. A coordination deficit was suspected when parents report included instances of delayed motor development, and indirectly measured by the “Stambak test”, a classical tapping task involving the reproduction of rhythmic successions of sounds given under patterns of increasing complexity (Stambak, 1951). Finally, although some participants may still have some subtle motor difficulties, none of them qualified for the main diagnosis of dyspraxia or developmental coordination disorder (Visser, 2003). Nonetheless, a history of slightly delayed motor milestones and/or evidence of dysgraphia, diagnosed both from parents report and direct observation of graphomotor productions, was present in six participants. Copy of the Rey figure was also often awkward or slightly disorganised.

In sum, all 19 participants to this research had a classical picture of moderate to severe dyslexia, without persistent production or reception speech/language pathology, but with usual comorbidity, mainly moderate attentional and/or motor coordination and slight spatial difficulties.

OUTCOME MEASURES

Each of the following tasks was administered 3 times at E1, E2 and E3 (see below, design section).

Articulatory efficiency

Children were required to repeat a series of simple (CV, e.g.: /ba, da, ga/), complex (CCV, e.g. /gra, dra/) syllables, semivowels (e.g. : /oui/) and nasal vowels (e.g. : /ã, õ/), simple and complex words. Productions are rated according to the proximity or distance to each specific target, as judged by a trained speech therapist. A score is given for each list, and a global score calculated as the average score of all lists. These tasks are supposed to explore not only articulatory precision and agility but also, to a lesser degree, auditory short-term memory. This latter aspect was more specifically targeted by a separate sentence (4 items) and non-word repetition task (two lists of 20 CV or CCV, 1 to 5-syllable pseudo-words of increasing difficulty).

Phonological awareness (P.A.)

P.A. tasks were taken from a frequently used battery in French (BELEC, Mousty et al., 1994), exploring segmenting, discrimination and short-term memory abilities. All tasks were played by a tape-recorder to insure homogeneity of testing conditions. These included tasks looking at various levels of P.A. : syllable inversion (10 bisyllabic CVCV pseudowords, e.g.: /radi/ → /dira/)) phoneme inversion (10 CV and 10 VC monosyllabic items; e.g: /bi/ → /ib/ and /ol/ → /lo/), syllable deletion (16 items, e.g. : /fango/ → /go/), simple (10) and complex (10) phoneme deletion (e.g.: /gal/ → /al/; /grõ/ → /rõ/), auditory acronyms (16 word pairs : form a new word with the initial sound of each of the two words).

In addition, a rhyme judgment task of the odd-one-out type, similar to those used in the training exercises (see below) was used : it comprised 10 bisyllabic word triplets, the subject being asked to indicate the 2 rhyming ones (ex: ballon-cochon-robot = rhyming target : /õ/).

Finally, a “phonological awareness score” was calculated by averaging performance from these different tasks.

Reading tasks

Due to limited controlled materials being available in French, reading tasks were different for younger (grades 1&2, i.e. 6-8 y-old) and older (grades 3-4, i.e. 8-10 y-old) children. For grades 1-2, we used a locally standardized battery (Hénin & Dulac, 1985) comprising an extensive, progressive exploration of all the difficulties of the French orthographic system (simple graphemes – syllables – multiple consonants, e.g.: [olp] or [spli] - complex graphemes, e.g.: [ain] read / $\tilde{\epsilon}$ / or [ien] read /j $\tilde{\epsilon}$ / - ambiguous forms, e.g. [payé] read /pəjə/) and, finally, a simple word reading task (24 items). There was no irregular words list for this younger group, due to their relative rarity in French, so that “sight” reading, at this age, could only be tested by reading regular words. For grades 3-4, children were asked to read aloud 3 lists (pseudowords : n=20; regular words, n=10; irregular words n=10) taken from the French L2MA battery (Chevrie-Muller et al., 1997).

All recorded performances were transformed into a percentage of correct responses in order to equate measures across children of different age ranges. A “phonological decoding score” was calculated from the performance to all non-word items, and a “word reading score” from performance to all lexical items from the two age groups.

Spelling tasks

For the same reasons as above, spelling tasks were different for grade 1-2 (transcription to dictation of 20 words and 4 simple sentences), and grades 3-4 (transcription of a short story « Le Petit Poucet » (Hénin & Dulac, 1985).

For all these spelling tasks, errors were classified according to the suspected underlying mechanism (phonological, grammatical, or usage errors). However, for sake of simplicity, we only used a global spelling score in terms of error rate.

A separate non-word spelling task was also given, consisting of eight simple (mainly CVCV) and 15 complex (CCV) pseudo-words

INSERT HERE FIGURE 1

TRAINING PROCEDURE

Experimental design (figure 1)

Our experimental protocol followed an A-B vs B-A design, where each group is its own control in order to avoid 'carry-over effects' due to the risk that the treatment used in the first period affects the outcome of the second period. According to this protocol, 10 children were randomly assigned to group 1 (G1), receiving during 3 weeks (period A) both auditory-phonological training and articulatory training, then only auditory training during the next 3 weeks (period B), whereas the remaining 9 children, assigned to group 2 (G2), received the same total amount of training, but in the opposite order, i.e. in period A, only auditory training, followed by auditory + articulatory training, so as to counterbalance a possible order effect. Note that this design has the advantage that there is no control group which could have its own effect (see Wise et al., 1999), but the inconvenience that periods A and B are not equivalent in terms of overall amount of therapy during the 3-week period.

Auditory phonological training

The method involves daily listening by children with dyslexia of series of exercises, recorded on audio-CDs and mainly containing triplets of words the patient had to compare phonologically to disclose similarity between 2 of the 3 stimuli, either at the beginning, the end, or at the middle of words ("odd-one-out" task). The only indication the children had was about the locus of the expected change in the word and whether they must focus on syllables (weeks 1-3) or sounds (weeks 4-6), each specific instruction being recorded and played before each exercise. Each daily session, corresponding to one track of the CD, lasted 15-25 mn, and was administered under the control of one parent, asked to report the child's response on a pre-printed response sheet. Participants and their parents were informed that the study will involve two parts, evaluation and training, and parents were informed that they will have to participate actively in the latter. Indeed, such participation of parents was crucial to the project. We particularly insisted on the need to avoid any feedback, since parents could have the spontaneous tendency to give the correct answer. Moreover, there was no visual component, and no oral production was needed since participants only had to point to a specific label. Once a week, information on the unfolding of the training procedure was obtained either by a visit at home or by telephone, and the response sheets were asked back to measure the level of compliance and make sure that the treatment is correctly carried out.

The training program has been designed under the principle of non-adaptive progression from simpler to harder phonological tasks. During the first 3 weeks, exercises only involve syllable segmentation, with increasing difficulty due to the closeness of the distractor to the target words. For example, at week 3, children have to discriminate between syllables containing phonemes of the same voiced-unvoiced pair to “find the 2 words with the same syllable in the middle” (“député/ carburant/ débutant”), with the additional difficulty of a common syllable between the distractor and one target word (/de/ in first syllable). During weeks 3 to 6, solving tasks requires detecting the common sound in two words, so that there is a progression from syllabic to phonemic segmentation requirements. At week 4, distractors are unrelated phonologically, whereas at week 6 they are (example : “palais/panda/bandeau” “find the two words beginning with the same sound”, where the rest of the syllable and middle phoneme are common between distractor and one of the target words). Through such manipulation of target-distractor closeness, of course unknown to participants, we wished to tackle not only explicit (metaphonological) but also implicit phonology (Boada & Pennington, 2006).

Articulatory training

Articulatory training involved the combination of two complementary approaches, both used in the same session, one aiming at a reinforcement of the participants’ articulatory awareness, the other one using possibilities offered by a computerized training tool. It was carried out 3 times a week, in a one-to-one setting, each session being monitored by a speech therapist or a neuropsychologist.

a) Articulatory awareness exercises.

This part of the protocol is based on evidence in dyslexics of low awareness of their articulatory system (Griffiths and Frith, 2002), leading to the idea that even in the absence of objective motor or articulation problem, the sensory-motor speech system may usefully be reinforced by specifically becoming aware of each individual phoneme with the support of proprioceptive, tactile and visual information.

Exercises were derived from those described in Alexander et al. (1991), putting emphasis on phonetic oppositions between voiced (/b/, /d/, /g/) and voiceless stop consonants (/p/, /t/, /k/).

The first three sessions were devoted respectively to : p/b ; t/d and k/g contrasts, then all the pairs were worked on again during the subsequent sessions. Each session included the same exercises in the same order :

1- Practice of voiceless phoneme (e.g; /p/) : The child was asked to produce the phoneme in front of a mirror in order for him/her to become aware of the position of lips during the aural production. Likewise, he /she will be taught how to recognize with finger palpation of his/her neck the sensation associated with unvoiced production.

2- Similar exercises with the voiced phoneme (e.g.: /b/).

3-Presentation and explanation of the anatomical sketch associated to this specific pair of phonemes : the child was trained to associate each stop consonant to the specific corresponding scheme of speech organs, a series of drawings inspired from a classical French aphasiology treatise (Lanteri, 1995), each representing the position of lips, tongue, teeth and palate during the production of a specific phoneme. Presence or absence of voicing was figured by a coloured horizontal stroke, a green, straight one for voiceless, and a red, wavy one for voiced consonants.

4- Matching to sketch : after the examiner had pronounced one given word, the child was asked to extract the initial phoneme of the word and to point to the corresponding sketch.

The specific training words have been chosen as to be « paronymes », which means that they are bisyllabic only differing by one phoneme situated in one of the syllables (palais/balai; touche/douche) and varying from one session to another. Each list contains six word-pairs.

b) Phoneme production with computerized visual feedback

This part of the training method takes advantage of facilities included in IBM Speech-viewer ® III (SPV3) program, a specialized software for professionals in speech pathology. Children had to pronounce a chosen phoneme through a microphone, while the program analysed the acoustic quality of the production. The utterance is then transformed into a visual index, for example a coloured part of the picture which changes from green to red, or a slope which is being climbed by a character. The following exercises have been chosen for practice of voicing : « Voice Presence », which develops an awareness of voicing (a clown changes colour, a man's lips move) ,« Voice

Onset » supposed to increase the awareness of voicing onset and control over voicing, and « Voice Timing », which improves coordination of respiration and voicing. For each exercise, the participant was asked to utter specific phonemes or groups of phonemes as accurately as possible, with the support of the visual index provided on the screen, specific to each game.

During the first 3 sessions, these games are being used in succession for practice of voiced/ voiceless contrast with pairs of stop consonants, with in addition for each session one pair of constrictive consonants with similar voicing opposition (f/v, ch/j, s/z).

In addition, other parts of the program were used to complete the above effects : « Phoneme Accuracy », which improves the accuracy of phoneme production (using a fun game format) ; « Multi-phoneme Chains » developing skills in pronouncing a sequence of different phonemes, and « Two-Phoneme Contrast » and « Four-Phoneme Contrast », both designed to improve accuracy in contrasting phonemes.

For instance, for the two-phoneme contrast, the aim of the game is to lead a vehicle through a series of obstacles by pronouncing the selected phoneme.

A different contrast is associated to each specific vehicle (picture of a car : f/v; picture of a bike : ch/j, picture of a Jeep : s/z)

The speed of movement of the vehicle and the threshold of recognition of each phoneme can be modified so as to provide an adaptive control to each child's performance.

Finally, each participant received :

- daily 20-25-mn sessions of auditory phonological training, made up of auditory exercises, without feedback and non adaptive
- 35-40-mn sessions of articulatory training, combining both approaches (articulatory awareness and Speech-Viewer games), 3 times a week, with explicit feedback and adaptive progression of the game content.

Overall, pre-testing began in October, training began in January and extended over a period of 3 months, all participants having received their 6 weeks of training by the end of April. On average, each child had received 14 hours of auditory training and 12 hours of articulatory exercises.

STATISTICAL ANALYSES

The 3 evaluation sessions (E1, E2, E3) respectively took place during the week preceding the beginning of training, after the first 3 weeks of training and after completion of the 6 weeks of training.

A 2-way repeated-measure ANOVA, considering time as the within subject factor, was used to test for different effects of session by group, i.e. one group's improvement follows a temporal course which is significantly different from the other group. Our a-priori hypotheses are (1) : there will be a general effect of time, meaning overall improvement for both groups; (2) improvement will involve primarily tasks dependent on phonological processes; (3) for these tasks, the two periods of combined auditory and articulatory treatment (period A for group 1 and period B for group 2) should result in better improvement. Note that with such an experimental design, one would not expect to find a significant group x time interaction, suggestive of an order effect. More probably, there will be an absence of interaction, reflecting similar cumulative improvement during the first and the second periods of training. Therefore, post-hoc pair-wise comparisons will be necessary to measure the direction and amplitude of each individual effect.

Finally, we calculated correlation coefficients for several measures at E1 to test for their predictive value on training efficacy, measured as an improvement index ($E3 - E1 / E3 + E1$). In line with the general context of this research, we more specially focused on tasks having a motor component (repetition and motor tapping tasks) and tasks exploring rapid word recognition, rapid naming, and response inhibition. Improvement index was calculated for phonology, reading, spelling, and repetition. Correlations at E1 were also computed between all these variables.

Ethical considerations

Parents were met as a group and individually and given detailed explanation of the training methods as well as about their role in this research. All children and parents were informed of the aims and principles of the study, and both parents signed informed consent. The study was authorized by the local Committee of Ethics (CCPPRB).

INSERT HERE TABLES 1&2

Results

Comparisons between the characteristics of the two groups are presented on tables 1 and 2. The two groups did not differ significantly in chronological age, reading age, and overall severity of dyslexia (table 1). There was a non-significant tendency for group 2 toward a better verbal (WISC similarities) and non verbal (Raven's PM47) performance than group 1. The main features are representative of a population of dyslexics of moderate severity, with 10-20 months difference between reading and chronological ages, and deficient phonetic fluency. Reading delay was larger in group 2, but this was not significant and did not affect intra-group comparisons (see below), as was the case for the marginal difference in non-verbal intelligence.

Table 2 summarizes participants' performance on some specific neuropsychological tasks. Rapid automatized naming as well as rapid word recognition were both moderately, but significantly, below expectations, without significant difference between the two groups. Likewise, response inhibition as measured by the naming time on the Stroop condition tended to be poorer than normal, without significant difference between groups. As concerns the rhythm reproduction task, performances also tended to be poorer than in controls of the same age, but here again remained within the low average range. Overall, the two groups did not differ as regards motor coordination (Stambak task), response inhibition (Stroop-like task) or hyperactivity rating using the Conners' score. There was no correlation between the Stroop task and any other task.

INSERT HERE TABLES 3&4

The main performances on phonological tasks are reported on table 3 . Apart from syllabic segmentation, all measures were lower than expected for age, although often marginally, except for repetition tasks where scores in percentiles were more clearly in the pathological range. . It is not surprising to find individual phonological scores falling within the normal range, since most of them had already received phonological training prior to this study. There were no significant differences between groups 1 and 2.

Table 4 and figure 2 summarize the mean and standard deviations obtained at E1, E2 and E3 for composite scores calculated from repetition tasks, spelling tasks, phonological awareness and reading tasks. Simple effects of time (session) were largely significant for all variables, suggesting overall improvement for all domains explored.

No group effect was obtained on either variable, and no group x session interaction was present. In particular, despite a larger reading delay in group 1, the two groups did not differ in overall reading improvement.

Statistics relevant to E1 vs E2 and E2 vs E3 comparisons lead us to consider three different patterns of results :

- typical additive effect : for phonological awareness and phonological decoding scores, the pattern of evolution over time during the training period was strongly suggestive of an additive effect of articulatory on phonological training, with significant improvement during the first training period for G1 and the second training period for G2. As shown in figure 2a and b, although improvement was progressive from E1 to E3 for both groups, it seems to plateau between E2 and E3 for group 1 and between E1 and E2 for group 2, i.e. during the “only phonology” periods. A similar tendency was observed for word and text spelling tasks.

INSERT HERE FIGURE 2

- for repetition and non-word spelling tasks , the only significant improvement occurred between E1 and E2, for both groups, suggesting that articulatory training has had no cumulative effect on phonology training : training phonology, with or without articulatory awareness component, significantly improved repetition and non-word spelling during the first three weeks, but this tendency was short-living and vanished later on. Alternatively, this could reflect a ceiling effect, especially for the repetition task where the performance on E2 is already near maximal.

- Finally, a third pattern, here represented by regular and, for the older participants, irregular word reading tasks (thus exploring the lexical route of reading), is that of delayed improvement (only significant between E2 and E3), without difference between the two groups, meaning that reading words from sight takes longer to benefit from phonological exercises, whether or not accompanied with articulatory training (figure 2c)². This pattern seems to reveal potential long term effect of combined treatment on reading. However, in the absence of follow-up measures, it is hazardous to speculate about possible subsequent benefit.

² Note that progression between E1-E2 and E2-E3 may be significantly different even though the slopes do not seem different.

Figure 3 reports correlation analyses carried out between neuropsychological variables and some pre-test measures on the one hand, and between these same variables and pre/post-training improvement of composite scores on the other hand.

The most significant results are those obtained between pre-test repetition scores, and phonological efficiency, as measured by pre-test phonological awareness scores ($r=0.535$ $p=0.0182$). In other terms, there is statistical evidence of a link between production efficiency and phonological awareness, but one can hardly speculate further on the exact nature of this link.

The last issue to be considered here is that of the predicting value of some pre-test measures on training efficacy, which we chose to address through simple correlation analyses : one of the best predictors of pre-post training improvement was performance on the “Stambak task”, which significantly predicted reading ($r=0.566$; $p=0.0115$) and non-word spelling ($r=0.656$, $p=0.0023$) improvement. Rapid naming at E1 also predicted improvement on phonological awareness tasks ($r=0.643$; $p=0.003$), whereas word recognition was significantly correlated with reading improvement ($r= 0.631$; $p=0.0038$) as well as phonological awareness improvement ($r=0.661$; $p=0.0021$). Finally, the mean initial performance on repetition tasks, mainly assessing fluidity and accurateness of speech utterance, was found correlated with the rate of improvement on phonological awareness tasks ($r=0.536$; $p=0.0218$).

Interestingly, the Stroop interference condition was not found to be correlated with any measure of improvement, suggesting that effects obtained with the naming and reading conditions are independent of attentional influence.

Finally, several of E1 measurements were found inter-correlated, not shown in detail here. One interesting result, however, the correlation between phonological awareness and non-word repetition, is illustrated in figure 3a.

INSERT HERE FIGURE 3

To summarize, poor rapid picture naming and word recognition were predictive of good response to phonological treatment, with an additional motor coordination predictor for reading improvement, suggesting a possible link between the motor automaticity component involved in the Stambak rhythm reproduction task and participants’ responsiveness to phonological/ articulatory treatment. Finally, of the 6

participants showing evidence of relative weakness in the area of motor coordination with associated dysgraphia, none appeared to react somehow distinctly to treatment, since they did not differ from the rest of group in terms of rate of improvement (but the sample, here, is too small to draw firm conclusions).

Discussion

As expected, all children in this study showed significant improvement in most domains of their reading-related cognitive abilities : reading, spelling, phonological awareness, but also a less often explored domain, that of speech production, here measured through several repetition tasks designed to assess speech accuracy and agility. In keeping with our expectations, derived from previous work with similar daily phonological exercises, this general improvement may be ascribed to the effect of the auditory component of this intervention, confirming that, even without using temporally modified stimuli, as was the case in previous reports (Habib et al., 1999; 2002), 6 weeks of training is sufficient to yield consistent gain in phonology, reading, spelling and speech production. Moreover, and contrary to most similar work in the literature, daily phonology exercises exclusively used auditory stimulation, a particularity which will be discussed below.

Our decision of choosing to use unmodified speech in training exercises follows our concern of minimising the number of different potentially active components present within the same remediation. This choice does not mean however that effects would not have been stronger if temporally-modified speech had been used instead, but our study does not allow to discuss further this otherwise still debated issue (Gillam et al., 2001a, b; Hook et al., 2001; Troia & Whitney, 2003).

Likewise, although providing feedback is usually considered of valuable support in remediation practice, it may involve additional mechanisms (see Tricomi et al., 2006) which one would prefer to avoid for experimental purpose. Arguably, if motivation is an important factor for training efficacy in the “ecologic” context of remediation, it could be important to minimize its role in experimental perspective, when trying to obtain scientific evidence of efficacy of a specific intervention.

The main result of this study was that combining articulatory and auditory phonological training adds significant advantage to auditory training alone, at least as concerns those measures depending on phonological processes, i.e. phonological awareness and non-word reading (phonological decoding). This combined effect is remarkable because, although modest, it allows to draw valuable indications for dyslexia therapy, as well as interesting speculations as to some theoretical issues.

Training phonology with purely auditory exercises.

As pointed out earlier, most of the previously reported successful interventions for phonological deficit in dyslexics have dealt with more or less complex “phonics” methods, indistinctly providing auditory, visual, verbal and non verbal stimulation, so that it remained unclear whether positive changes may occur in dyslexics from phonology training only using the auditory modality. Moreover, contrary to most similar work taking advantage of the motivational content of video-games, with systematic reinforcement and valorisation of performance, we chose to minimize this aspect to get rid of any additional factor of uncontrolled variability.

Although there is strong evidence that dyslexics as a group suffer from some kind of auditory perceptual deficit (see for example Banai & Ahissar, 2006 for a recent account), there is no consensus about the causal relationship between this deficit and impairment in reading-related processes in dyslexics (Rosen, 2003; Ramus et al., 2003). Except for rare studies having tried to train normal (Moore et al., 2005) or dyslexic children (Schaffler et al., 2004) with auditory stimulation, evidence is lacking of a specific effect of phonological awareness training through exclusively auditory exercises. In a recent meta-analysis of the literature relevant to interventions on dyslexics, Alexander and Slinger-Constant (2004) point out the fact that the two most famous treatments targeting auditory processing, FastForword®(FFW) and Earobics® (Pokorni et al., 2004), are not really specific to the auditory modality: although presented through the auditory channel, FFW also trains syntactic and semantic comprehension, and Earobics incorporates graphemes and written words into the program.

The phonological exercises included in the present study typically draw on those cognitive processes referred to under the term of “phonological awareness” (Goswami & Bryant, 1990), covering a wide range of processes from phoneme discrimination to

explicit metaphonology, including implicit phoneme sensitivity, auditory attention, and some forms of verbal working memory. Our results thus lend support to the widely admitted notion that training phonological awareness is a helpful instrument in order for dyslexics to successfully learn to read, even though, as often pointed out, such training efficacy is strongly enhanced when associated with explicit teaching of grapheme-phoneme correspondence (McCandliss et al., 2003; see Castles and Coltheart, 2004 for a discussion of this point).

Of particular interest, in this context, is the finding in the present study of a correlation between rapid naming on pre-tests and overall gain in phonological awareness, with slower naming performance being associated to larger overall pre-post phonological improvement. Specifically, poor pre-training rapid naming is a good predictor for phonological improvement, confirming that the two processes are closely related (Vellutino et al., 2004).

Finally, our results with the rhythm reproduction task (Stambak, 1951) are remarkable, not only because they confirm that children with dyslexia are often impaired on such tasks (here scoring on average below 25th percentile), but also because they demonstrate, through the outcome of correlation analyses, that this purely auditory-motor task is strongly correlated with improvement on reading-related measures, arguing for non-linguistic determinants to reading impairment, such as postulated in the cerebellar theory (see below).

Usefulness of training articulation

The main result of this study was thus the demonstration of statistically significant benefit of adjoining articulatory training to more classical phonological exercises. This was mostly obvious on tasks relying on phonological processing, i.e. phonological awareness and phonological decoding. For these two tasks, the advantage of combining auditory and articulatory training was clearly demonstrated through the A-B/B-A design, which revealed a reversed effect of group. Since both tasks require access to phonological — especially phoneme — representation, this strongly suggests that we have thus been acting at the level of phoneme representation. In other words, simultaneous training of auditory and articulatory phonology appears an efficient way to improve and/or reinforce poor representation of phonemes, a deficit considered both

common to most dyslexic readers and central to their reading difficulties (Swan & Goswami, 1977). Therefore, it should be important for clinicians and other specialists involved in remediation to possess such a potentially useful additional tool among their range of possible interventions.

More specifically, our correlation study showed that training was more efficient when initial impairment was more severe on a non-word repetition task, especially for progression in phonological processes. We also looked at this correlation considering separately the first and second training periods for each group, and found that this correlation was even stronger for periods of combined articulatory and auditory training. In addition, the correlation between non-word repetition and phonological awareness at E1 confirms that the two processes share common mechanisms.

Taken together, these results are indicative of a close link between auditory and articulatory mechanisms in phonology, even though articulatory treatment did not seem to add much to background auditory training on repetition tasks.

From a more theoretical point of view, it seems appropriate to recall here that the idea of a link between articulation and phonology during early stages of development is not a new one. In their “motor theory of speech perception”, Liberman and Mattingly (1985; 1989) postulate the existence of a specific module within which “speech perception processes the acoustic signal so as to recover the coarticulated gestures that produced it. These gestures are the primitives that the mechanisms of speech production translate into actual articulator movements”.

It is conceivable that some children with dyslexia have suffered from early disruption of that system and that simultaneously training its auditory and motor components can help restore harmonious interplay between parts of the system. Accordingly, the classical McGurk effect has been found defective in children with learning disabilities compared to normal learning controls (Hayes et al., 2003a), suggesting a specific inability for dyslexics to properly achieve sensory motor integration between an auditory message containing phonemic information and a visual stimulus conveying bucco-facial features of the same phoneme.

A new look at the cerebellum theory of dyslexia

Fawcett and Nicolson (1999) have pointed out that many children with dyslexia also experience a series of coordination and balance problems, which could suggest

cerebellar involvement. Moreover, dyslexics are also generally slow to acquire procedures, which these authors have referred to as a deficit in automatization. In their formulation of the theory, Nicolson et al. (2001) suggest that a general motor coordination deficit would impact on the development of phonology through the suspected links between articulation and phonological awareness, a possibility consistent with later evidence (Lalain et al., 2003) of systematic subtle deviances in phoneme production in dyslexics. Finally, non-motor functions of the cerebellum are more and more often emphasized in the cognitive literature, describing the cerebellum together as a motor organ, a cognitive organ, and an organ of learning (Ito, 2005). The correlation found in our study between motor tasks (such as rhythm reproduction and non-word repetition) and cognitive performances and their improvement, would fairly well take place in such a framework. Although the rhythm reproduction task we have used also likely involves auditory and short-term memory processes, it basically represents a measure of the quality of output realisation and this motor component makes it a plausible candidate as an indicator of cerebellar dysfunction (Chen et al., 2005).

In our study, the 6 patients with (moderate) coordination impairment (mainly clumsiness, delay in motor development, associated dysgraphia), and the 2 with suspicion of associated hyperactivity did not seem to behave differently on pre-test as well as pre-post training improvement than the rest of the group. Moreover, they were not especially poor at the rhythm reproduction task, which was one of the best predictors of improvement. Taken together, these results suggest that although possibly related to a sensory-motor component, improvement on reading-related tasks occurred independently of the presence or absence of comorbid coordination deficits, as suspected by Raberger and Wimmer (2003) as well as Ramus et al (2003a). Likewise, the absence of statistical link between the Stroop interference score and rate of progression across training sessions, does not support any contribution of associated attentional or executive deficit. While recognizing that this constitutes only an indirect and speculative case for a cerebellar origin of dyslexia, we believe it opens interesting perspectives for future research.

Limitations of the present study

A first methodological issue, also raised in previous studies (Wise et al., 1999), is that of comparing training periods with different amounts of input (auditory vs auditory + articulatory periods), which could weaken the main result of this study, that of a larger effect of the combination of the two methods. However, it must be said that this superiority of the dual treatment appears more clearly on those tasks where it was most expected to occur, and not in most other tasks.

Also, it is important to note that, in this study, like others whose experimental designs involve repeated measurements at relatively short intervals (here 3 weeks), some apparent improvement may in fact only reflect practice effects. Although this could be the case for all tasks using meaningful materials, it is much less likely to have occurred with non-lexical materials such as non-word reading and spelling, or purely phonological tasks. Note that our assessment phonological tasks were totally different from those used during training. Moreover, our results on phonology and non-word reading are particularly robust, since the advantage of auditory / articulatory training emerged just as much whether it occurred during the first (E1-E2) or the second (E2-E3) session, allowing us to rule out a learning effect.

One important limitation of the present study is that we have no follow-up data, so that the improvements observed could possibly prove to be only transient. Moreover, the effects observed are rather small, so one can wonder about the possibility of generalizing such results. Of course, this would be of considerable importance in order for clinicians to consider using these data in their practice, but one must keep in mind that with the kind of experimental design we used here, we did not expect larger effects to occur. On the contrary, in the context of such theory-based, fine-grained interventions, even a small effect, and even on a short period of time, is an indication that the underlying theory may apply and that the small effect detected is apt to be amplified if associated to other tools within a wider intervention, (as is generally the case in clinical —and even more in pedagogical— practice).

In conclusion, we would certainly recommend using auditory and articulatory treatments in association (or temporal succession) with other techniques favouring grapheme-phoneme mapping, with the objective of maximizing the effect by acting at different levels of the deficit. However, for scientific purpose, we would favour

proceeding step by step, testing each component as exclusively as possible, at the expense of effects magnitude.

Finally, now that there is no doubt anymore about the crucial role, in dyslexia, of training phonology, further studies should concentrate on how to take into account all the associated symptoms and/or pathologies which usually accompany the reading disorder. Even if the gains are of moderate, even modest, magnitude, it is of theoretical importance to try to disentangle potential factors of improvement in order, as was tentatively shown in this article, to progressively improve the pertinence of our remediation protocols, and, ultimately, the efficacy of our interventions in learning disordered children.

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Table 1

Population characteristics : sample size, mean (standard deviation), and significance of group differences.

	Group 1 (n=10)	Group 2 (n=9)	difference
Age (mth)	109.8 (11.3)	102 (13.7)	p=0.1907
Reading age (mth)	88.4 (8.2)	93.3 (10.2)	p=0.2203
WISC simil	9.5 (2.1)	11.2 (2)	p=0.0863
WISC pict completion	12	13.1	P= 0.10
WISC blocks	8.9 (3.4)	11.1 (3)	p= 0.1763
WISC digit memory	10.5 (2.8)	10 (3.1)	p=0.7154
PM 47 percentile	36.5 (16.3)	53.9 (20.4)	p=0.0551
Phonetic fluency	12 (4.8)	9.6 (3.8)	p=0.266
percentile	18	13	
Semantic fluency	17.8 (4)	17.4 (7.5)	p=0.898
percentile	45	51	

Table 2 : Main neuropsychological characteristics : mean (standard deviation), and significance of group differences.

	Group 1 (n=10)	Group 2 (n=9)	difference
Memory (BEM84)			
Raw score	60.4 (8.7)	59.3 (11.5)	p=0.813
z-score	-2.2 (0.6)	-2.2 (0.8)	
Animals naming (time)			
Raw score	49.8 (17)	55.8 (4.3)	
z-score	0.1 (1.3)	1.3 (1)	p=0.4034
Anim. Reading (time)			
Raw score	47.8 (23.1)	38.1 (12.7)	
z-score	2.6 (2.6)	1.5 (1.4)	p=0.2811
Anim. Stroop (time)			
Raw score	85.1 (19)	81.4 (17.5)	
z-score	1.8 (1.2)	1.6 (1.1)	p=0.67
Rhythmic reproduction (Stambak)/ 21			
Raw scores	13.8 (3.5)	13.7 (3.3)	
z-score	-1.075 (1.5)	-1.135 (1.5)	p=0.933

Table 3 : Performance on phonological awareness pre-training evaluation

	<i>Group 1 (n=10)</i>	<i>Group 2 (n=9)</i>	<i>difference</i>
Repetition CV/20	14.5 (4.4)	14 (5)	p=0.82
percentile	37.7	33.8	
Repetition CCV/20	6.5 (2.4)	7.4 (2.0)	p=0.3755
percentile	16.5	17.7	
Non-word repet./20	10.5 (2.4)	10.7 (3.2)	P=0.8677
percentile	35.3	36.5	
Syllable inversion/10	9.1 (1.2)	8.3 (2.3)	p=0.390
percentile	64	53	
Phoneme inversion/10	7.7 (2)	7.3 (2.3)	p=0.717
percentile	37.6	35.3	
Phoneme delet./16	7.3 (3.1)	6.5 (4.0)	p=0.656
percentile	43.8	38.8	
Acronyms /16	11.5 (4.7)	11.1 (2.08)	p=0.822
percentile	42.5	37.4	

Table 4 : Main measures obtained before training (E1), after 3 weeks of training (E2) and after 6 weeks training (E3). Group 1 (G1) received phonological and articulatory treatment first (E1 – E2) and then only phonological treatment (E2-E3) . Group 2 (G2) received the same in the reversed order. For each series of measures, post-hoc comparisons are reported for each group and each interval. ** : $p < 0.05$;

	E1 Mean (s.d.)	E2 Mean (s.d.)	E3 Mean (s.d.)	E1/E2 (Fischer's LSD)	E2/E3 (Fischer's LSD)
Mean repetition score (%)					
G1	89.3 (0.068)	94.4 (0.039)	96.1 (0.047)	$p = 0,00027^{**}$	$p = 0,175$
G2	88 (0.095)	92.2 (0.097)	93,7 (0.083)	$p = 0,00152^{**}$	$p = 0,251$
Mean non-word spelling (%)					
G1	21.3 (0.134)	26.3 (0.128)	29.3 (0.101)	$p = 0,0258^{**}$	$p = 0,165$
G2	20.8 (0.094)	27.5 (0.103)	30.2 (0.101)	$p = 0,0051^{**}$	$p = 0,248$
Mean word and text spelling (errors)					
G1	5,30 (3.26)	4,40 (2.911)	4,06 (2.68)	$p = 0,0001^{**}$	$p = 0,101$
G2	3,98 (2.85)	3,65 (2.678)	3,25 (2.48)	$p = 0,132$	$p = 0,076$
Mean phonol awareness (/ 14)					
G1	9.94 (2.586)	11.071 (1.945)	11,37 (1.926)	$p = 0,0097^{**}$	$p = 0,471$
G2	9.508 (2.033)	10.238 (2.027)	11,58 (0.816)	$p = 0,101$	$p = 0,0038^{**}$
Mean reading words (%)					
G1	76,4 (15.39)	79,52 (13.09)	83,23 (13.73)	$p = 0,126$	$p = 0,070$
G2	80,76 (13.73)	82,63 (12.39)	87,72 (10.11)	$p = 0,378$	$p = 0,020^{**}$
Mean phonological decoding (%)					
G1	70,69 (14.98)	76,69 (13.36)	77,87 (13.47)	$p = 0,035^{**}$	$p = 0,671$
G2	82,25 (12.11)	79,01 (12.33)	88,61 (7.92)	$p = 0,271$	$p = 0,0021^{**}$

Figures captions

Figure 1 :

Diagram of the sequence of training for each group : each participant received two 3-week periods of training, separated by a break of 2 weeks. Both groups receive daily auditory phonological training during periods A and B, with additional articulatory training during period A for group 1 , and during period B for group 2. Outcome measurements are carried out at E1, E2, E3.

Figure 2 :

Mean performance of 19 children with dyslexia at 3 successive evaluations : E1=before training, E2=after 3 weeks of training, E3 = after 6 weeks of training. Both groups received auditory daily training all along the 6 weeks. Group 1 received additional articulatory training between E1 and E2, group 2 between E2 and E3.

a : progression on a composite score of phonological awareness (/14)

b : progression on phonological decoding tasks (non-word reading)

c : progression on word reading tasks

** : $p < 0.05$. Note that, for each of these three diagrams, scales are not derived from standard scores but averaging scores to several independent tasks.

Figure 3 :

Correlation plots computed between : pre-test phonological awareness and non-word repetition scores (a); rhythm reproduction task and pre/post-training progression of a reading score (b); rapid naming and pre/post-training progression of phonological awareness score (c).